

ARTICLE

The Cognitive and Behavioural Responses to Symptoms Questionnaire (CBRQ): Development, reliability and validity across several long-term conditions

Federica Picariello¹  | Joseph Chilcot¹  | Trudie Chalder²  |
David Herdman^{1,3}  | Rona Moss-Morris¹ 

¹Health Psychology Section, Department of Psychology, Institute of Psychiatry, Psychology and Neuroscience King's College London, London, UK

²Psychological Medicine, Institute of Psychiatry, Psychology and Neuroscience King's College London, London, UK

³St George's University Hospitals NHS Foundation Trust, London, UK

Correspondence

Rona Moss-Morris, Health Psychology Section, Institute of Psychiatry, Psychology and Neuroscience, 5th Floor Bermondsey Wing, Guy's Campus, King's College London, UK.
Email: rona.moss-morris@kcl.ac.uk

Abstract

Objectives: Cognitive and behavioural responses to symptoms can worsen or maintain the severity of symptoms across long-term conditions (LTCs). Although the Cognitive and Behavioural Responses Questionnaire (CBRQ) has been used in research, its original development and psychometric properties as a transdiagnostic measure have not been reported. Our aim was to evaluate the psychometric properties of the CBRQ and a recently proposed short version, across different LTCs.

Design: Psychometric validation study.

Methods: Confirmatory factor analysis (CFA) tested the factor structure of the CBRQ in two datasets from the CBRQ's original development; (chronic fatigue syndrome, $N = 230$; and multiple sclerosis, $N = 221$) and in additional groups: haemodialysis ($N = 174$), inflammatory bowel disease ($N = 182$) and chronic dizziness ($N = 185$). Scale reliability and construct validity were assessed. The factor structure of the shortened CBRQ (CBRQ-SF) was also assessed.

Results: CFA revealed that a 7- or 8-factor structure had generally appropriate fit supporting the originally proposed 7 factors (Fear avoidance, Damage beliefs, Catastrophising, Embarrassment avoidance, Symptom focusing, All-or-nothing behaviour and Avoidance/Resting behaviour). Omega coefficients indicated satisfactory internal reliability. Correlations with related constructs suggested construct validity. The scale appeared sensitive to change.

Federica Picariello and Joseph Chilcot are joint first authors.

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The CBRQ-SF also displayed good psychometric quality, with a better model fit than the CBRQ.

Conclusions: The CBRQ and the shortened version were shown to be reliable and valid at assessing a range of cognitive and behavioural responses to symptoms, highlighting the multi-symptom, transdiagnostic properties of this questionnaire. Further research is necessary to determine the test–retest reliability and sensitivity to change of the CBRQ and CBRQ-SF and a thorough evaluation of the content validity of the items.

KEYWORDS

CBRQ, cognitive, factor analysis, psychometrics, reliability, symptoms, transdiagnostic, validity

Statement of Contribution

What is already known on this subject?

- Cognitive and behavioural responses to symptoms can worsen or maintain the severity of symptoms across long-term conditions (LTCs).
- The Cognitive and Behavioural Responses Questionnaire (CBRQ) has been used in research, however its original development, and psychometric properties as a transdiagnostic measure have not been reported.

What this study adds?

- The CBRQ and a shortened version (CBRQ-SF) were shown to be reliable and valid at assessing a range of cognitive and behavioural responses to symptoms, highlighting the multi-symptom, transdiagnostic properties of these measures.
- Whilst both versions capture clinically important cognitive and behavioural constructs, we recommend using the CBRQ-SF given its stronger factor structure and brevity.
- Further research is necessary to determine the content validity, test–retest reliability and sensitivity to change of the CBRQ and CBRQ-SF.

INTRODUCTION

According to the 2018 Health Survey for England, 43% of adults have at least one long-term medical condition (LTC) (NHS Digital, 2018). Seventy per cent of the total health and care budget is spent on LTCs (Department of Health, 2012). Many people with LTCs experience a range of unpleasant and often debilitating symptoms, with 50% reporting pain, breathlessness and fatigue (Solano et al., 2006). In a recent Danish population-based study ($N = 47,452$), respondents over a 4-week period experienced an average of 4.8–7.4 symptoms which were associated with the level of morbidity (Willadsen et al., 2021). Furthermore, higher levels of symptoms are associated with poorer quality-of-life and functioning, lower mood and increased healthcare utilization (Cleeland, 2007; Eckerblad et al., 2015; Katon et al., 2007).

The severity of symptoms and the degree to which these impact on people's lives cannot be purely explained from a biomedical viewpoint. One framework that has been broadly used to explain individual differences in symptom experience and disability across LTCs is the Common-Sense Model of Self-Regulation (CSM-SR; Hagger et al., 2017; Leventhal et al., 1984, 1997, 1998). The CSM-SR postulates

that illness representations, the idiosyncratic beliefs and perceptions people have about their illness, guide behavioural coping and emotional responses to illness (Leventhal et al., 1984, 1998).

The Illness Perception Questionnaire (IPQ) was developed to provide a quantitative assessment of the five components that constitute an illness representation in Leventhal's CSM-SR – including identity (the symptoms associated with the illness), and beliefs about the consequences, timeline, control/cure and cause of the illness (Weinman et al., 1996). A revision to the IPQ (IPQ-R) added subscales around coherence and emotional representations (Moss-Morris et al., 2002). The development of these measures was a catalyst to research investigating the role of patients' cognitive representations of illness, which have been shown to impact a range of outcomes across LTCs (Hagger et al., 2017: for a review). Whilst these overarching illness beliefs are clearly important in understanding individual variations in the impact of an illness, qualitative studies suggest that day-to-day interpretations of symptoms also appear to be particularly important in determining coping behaviours which may enhance the experience of the symptoms (Harrison et al., 2015; Picariello et al., 2017; Taylor et al., 2018).

Other frameworks, largely focused on conditions where underlining pathophysiology is less clearly defined, such as primary chronic pain, have defined specific cognitive behavioural responses to symptoms to explain variation in extent and impact of symptoms. For example, according to the fear-avoidance model of chronic pain (Asmundson et al., 2004), a common emotional response to pain is fear. Fear of pain or the view that pain signals tissue damage often leads to avoidance behaviour or fear avoidance. Catastrophising, defined as negative and inflated beliefs in anticipation of pain/fatigue (Jensen et al., 2011; Leeuw et al., 2007), has also been identified as a key unhelpful cognitive interpretation driving increased distress and avoidance behaviours in both pain (Asmundson et al., 2004) and fatigue (Lukkahatai & Saligan, 2013). Changing these beliefs and related behaviours have been shown to be important mechanisms of change in reducing the impact of pain and fatigue (Burns et al., 2012; Chalder et al., 2015; Wertli et al., 2014).

Current published psychometric measures of catastrophising and fear avoidance are symptom-specific, such as the Fatigue Catastrophising Scale (FCS; Jacobsen et al., 1999) or the Pain Catastrophising Scale (PCS; Sullivan et al., 1995) which limits the exploration of cognitive and behavioural responses to a broad range of symptoms, particularly in the context of LTCs. Avoidance behaviour can also be driven by embarrassment of symptoms, a recurrent theme across qualitative studies of both pain and fatigue (Crowe et al., 2017; Whitehead et al., 2016 – for meta-syntheses). So far, there is no measure capturing avoidance behaviour due to embarrassment of symptoms. In addition to behavioural avoidance, another recurrent behavioural response to symptoms is all-or-nothing behaviours (boom or bust), where people push themselves to get things done when symptoms allow and then crash because of overdoing things (Moss-Morris, 2005; Spence et al., 2005). Avoidance and excessive resting behaviours and all-or-nothing behaviours are likely related to cognitions with different affective components, namely fear avoidance (i.e. activity is harmful for symptoms) or embarrassment (my symptoms might flare if I go out and will embarrass me). In addition to measuring how people interpret symptoms, how much they focus on the symptoms may also be important. Greater attention to symptoms has been shown to exacerbate symptoms and reducing symptom focus has been associated with reductions in symptoms after treatment (Barends et al., 2020; Cella et al., 2011; Moss-Morris et al., 2005). A broader transdiagnostic measure that can capture cognitive and behavioural responses to a wide range of symptoms across different LTCs is particularly valuable for the identification of *shared* and *idiosyncratic* nuances between symptoms and conditions, meaning that interventions can be more readily and efficiently adapted based on this knowledge.

The Cognitive and Behavioural Responses to Symptoms Questionnaire (CBRQ) was developed to capture these broader symptom interpretations, symptom focusing and associated behaviours in general across LTCs (details of the seven subscales presented in the methods). The original development of the CBRQ scale was based on data collected from two distinct patient groups, those with multiple sclerosis (MS) and those with chronic fatigue syndrome (CFS) and has never been published (Moss-Morris & Chalder, 2003), so these data are presented here alongside a broader psychometric analysis of the questionnaire across a wider group of patients.

The psychometric properties of the CBRQ have recently been further evaluated in a larger CFS cohort (Ryan et al., 2018) with the subscales having good fit with the data, although suggested an eight

rather than seven subscale structure whereby the behavioural subscale, avoiding activity and resting to symptoms in response to symptoms was divided into two subscales. The subscales also showed satisfactory internal consistency (Cronbach's $\alpha \geq .76$; Ryan et al., 2018). The authors also constructed a shortened 18-item version of the larger CBRQ (CBRQ-SF) with acceptable reliability (Cronbach's $\alpha .67-.88$; Ryan et al., 2018).

To reiterate, despite the untapped potential of the CBRQ at better defining processes of change following intervention, the original development of the questionnaire has not been documented nor have the measurement priorities been evaluated across different patient populations. Therefore, the overarching aim of this paper is to present the details of the original questionnaire development process and more importantly based on the COSMIN taxonomy (Mokkink et al., 2010) to evaluate the factor structure and estimate measurement properties, including internal consistency, responsiveness and construct validity of this questionnaire transdiagnostically, with the following objectives:

1. To corroborate the original structure of the scale using exploratory factor analysis in MS and CFS patients.
2. To test the factor structure of the full CBRQ across five LTC patient populations, MS, CFS, haemodialysis, inflammatory bowel disease and chronic dizziness
3. To test the factor structure of the CBRQ-SF across five LTC patient populations.
4. To estimate the reliability of the CBRQ using the omega index across five patient populations.
5. To assess the construct validity between subscales of the CBRQ and conceptually similar (yet different) constructs, specifically symptom severity, depression, anxiety and functional impairment.
6. To explore CBRQ's sensitivity to change using existing trial data.

METHODS

The psychometric evaluation of the CBRQ is reported here according to the COSMIN guidelines (Gagnier et al., 2021).

Participants

Multiple datasets drawn from a range of existing studies which included CBRQ data alongside measures of symptom severity, level of symptom-related impairment and/or mood were used. Although the questionnaire has been widely used, the original development and validation in chronic fatigue syndrome (CFS) and multiple sclerosis (MS) have never been published. The confirmatory factor analysis (CFA), the focus of the current study, was conducted with the CFS and MS samples and additional longitudinal studies of (1) fatigue in haemodialysis (HD), (2) fatigue in inflammatory bowel disease (IBD) and (3) chronic dizziness. Baseline data of the latter three datasets were used in the CFA analysis. See Table 1 for information on the datasets including participant characteristics.

Measures

The scale was originally conceptualized as four cognitive subscales (fear avoidance beliefs, embarrassment avoidance beliefs, symptom focusing and catastrophising) and two behavioural subscales (resting or avoidance of activity and all-or-nothing behaviour; see File S1 for details of the preliminary development).

The cognitive subscales items are scored from 0 (Strongly disagree) to 4 (Strongly agree), while the behavioural subscales are scored from 0 (Never) to 4 (All the time). Item scores are summed together to obtain a total score for each subscale (File S2). Higher scores indicate more negative cognitive responses to a symptom on the cognitive subscales. Higher scores on the behavioural subscales indicate greater

TABLE 1 Characteristics of the datasets.

Dataset	Study design	Focus of study	Population	Sample size	Ethical approval reference	CBRQ missingness	Symptom severity ^a	Functional impairment ^a	Mood ^a
CFS	Cross-sectional study	Questionnaire development	Adults with CFS based on either CDC or Oxford criteria recruited from the CFS research and treatment service at King's College London the Seacroft	230	185/02	The proportion of item-level missing data on the CBRQ fluctuated between 0% and 4.8% (item AL5).			
MS (Skerritt & Moss-Morris, 2006, van Kessel et al., 2008)	Cross-sectional study Randomized-controlled trial	To identify predictors of fatigue and impairment To assess efficacy of CBT for fatigue in MS	Adults with a confirmed diagnosis of MS recruited from around New Zealand in 2003 Adults with a confirmed diagnosis of MS recruited from Auckland Hospital MS service and the Auckland MS Society between July 2004 and August 2005	149 72 at baseline	AKY/03/03/070 AKX/03/03/084	The proportion of item-level missing data on the CBRQ fluctuated between 0% and 1.8% (item AL5).			

(Continues)

TABLE 1 (Continued)

Dataset	Study design	Focus of study	Population	Sample size	Ethical approval reference	CBRQ missingness	Symptom severity ^a	Functional impairment ^b	Mood ^c
HD (Chicot et al., 2016, Picardillo et al., 2016, 2018, 2020).	Longitudinal study (3-year follow-up)	To identify predictors and consequences of fatigue	Adults with kidney failure receiving thrice-weekly in-centre HD for 90 days or longer recruited between April and August 2014 from a renal outpatient unit in England	174 at baseline	14/EM/0037	At baseline, the proportion of item-level missing data on the CBRQ fluctuated between 2.9% (item FA1) and 10.9% (item C6).	Fatigue severity: CFQ (Chalder et al., 1993)	Fatigue-related functional impairment: WSAS (Mundt et al., 2002)	Depression and anxiety: HADS (Zigmond & Snaith, 1983)
IBD (Aritom et al., 2017)	Longitudinal study (2-year follow-up)	To identify predictors of fatigue	Adults with IBD recruited between September 2015 and March 2016 from IBD services (out-patient clinics and biologic infusion units) at three tertiary referral hospitals in England	182 at baseline	15/LO/1081	At baseline, the proportion of item-level missing data on the CBRQ fluctuated between 1.6% and 3.3% (items FA14, FA15, FA17, C1 and L1).	Fatigue: MFI (Smets et al., 1995)		Depression and anxiety: HADS (Zigmond & Snaith, 1983)
Chronic Dizziness (Herdman et al., 2020b, 2020b)	Longitudinal study (3-month follow-up after initial consultation)	To identify predictors of chronic dizziness	Adults with chronic dizziness recruited consecutively between March and December 2018 from a waiting-list to attend a tertiary dizziness service	185 at baseline	16/NI/0256	There was no item-level missing data on the CBRQ.	Frequency of dizziness, vertigo and unsteadiness symptoms: VSS-VIER (Yardley et al., 1992, 1998),	Self-reported dizziness interference and disability: DHI (Jacobson & Newman, 1990)	Depression: PHQ-9 (Kroenke & Spitzer, 2002) Anxiety: GAD-7 (Spitzer et al., 2006).

Abbreviations: CFQ, Chalder Fatigue Questionnaire; MFI, Multidimensional Fatigue Inventory; CFS, Chronic Fatigue Syndrome; DHI, Dizziness Handicap Inventory; GAD-7, 7-item Generalized Anxiety Disorder Scale; HADS, Hospital Anxiety and Depression Scale; HD, Haemodialysis; IBD, Inflammatory Bowel Disease; MS, Multiple Sclerosis; PHQ-9, 9-item Patient Health Questionnaire; VSS-VIER, short form Vertigo Symptom Scale; WSAS, Work and Social Adjustment Scale.

^aQuestionnaire scores were computed using prorated to account for missing items within scales, with a threshold of at most one-third of items missing (where scale includes six items, at least four items must have been completed otherwise the scale score was set to missing for that individual). This was deemed appropriate here based on Graham (2009)'s recommendations.

use of avoidance and/or all-or-nothing behaviours in response to a symptom. The instructions can be tailored directing participants to provide their views in relation to a symptom specifically, such as pain, fatigue or breathlessness or symptoms of their illness in general.

When the original version was developed, principal component analysis (PCA) was conducted on the combined CFS and MS datasets to test the factor structure and select best-fit items. PCA was conducted separately for items pertaining to cognitive responses to symptoms and those pertaining to behavioural responses, due to differences in the wording of the response options. Preliminary analysis indicated that fear avoidance appeared to split into two subscales, one pertaining to the affective interpretation of symptoms (fear avoidance beliefs), and another capturing an interpretation of symptoms as signally damage to the body. PCA produced a self-report instrument consisting of 40 items, loading onto five cognitive subscales and two behavioural subscales measured on a 5-point Likert scale (see File S3, for the 7-factor model).

Subsequent Confirmatory Factor Analysis (CFA) conducted by Ryan et al. (2018) further refined the original CBRQ suggesting separating avoidance and resting behaviours into two separate factors (referred to later as the 8-factor model).

In addition to the CBRQ, participants completed measures of symptom severity, distress and impairment in functioning relevant to each study. These measures were used to explore the construct validity of the scale (Objective 5). Different measures to capture these constructs were used in the datasets and are described in Table 1.

STATISTICAL ANALYSES

Although PCA was used originally as part of scale development, this method has been since criticized for only accounting for the variance of measured variables and not distinguishing between common and unique variance of the items in the resultant factors, while exploratory factor analysis (EFA) is a method for determining latent structure, not just item reduction, that derives latent factors from common variance alone (Howard, 2016). We therefore reanalysed the original CBRQ here based on the combined CFS and MS datasets, given the large pool of items, using EFA to determine consistency with the factor structure proposed through PCA. Details of EFA methodology and results are presented in File S4. The focus of this paper is on the CFA of the CBRQ across different LTCs.

The factor structure was evaluated based on fit indices across the five samples separately (CFS, MS, HD, IBD, chronic dizziness) in Mplus using Weighted Least-Squares with Mean and Variance (WLSMV) adjustment estimation as previously recommended across different models, varying in sample size, complexity and normality (Beauducel & Herzberg, 2006; Flora & Curran, 2004). The latent factors were approximately normally distributed (Finney & DiStefano, 2006; skew and kurtosis values and normality plots available in File S5). One-factor, 7-factor (as originally proposed) and 8-factor models, based on Ryan et al. (2018),¹ were also tested. A statistical comparison between the 7- and 8-factor models using chi-square difference testing was not possible because the 7-factor model is not fully nested within the 8-factor model. The CBRQ-SF proposed by Ryan et al. (2018) was also evaluated in the five samples here using CFA. Separate analyses were conducted in each sample to (1) establish how the CBRQ performs in different samples, and (2) due to slight variation in wording of the questionnaire stem between the datasets, in particular focus on different symptoms and framing of CBRQ in relation to the symptom ('Views about your symptoms in general' versus 'Views about your fatigue or dizziness specifically').

Assessment of goodness-of-fit was based on standard model fit criteria (Kline, 2005). Given that χ^2 is sensitive to sample size, relative χ^2 value (χ^2/df , where values close to 2 indicate a good fit) was used (Hoelter, 1983). Additional fit indices were examined, including the Root Mean-Squared Error of Approximation (RMSEA) (Steiger, 1990) with a recommended cut-off value close to $<.06$ ($<.08$ considered adequate fit; Hu & Bentler, 1999); Comparative Fit Index (CFI) (Bentler, 1990), and Tucker–Lewis Index (TLI) (Tucker & Lewis, 1973) with cut-off of $\geq.95$ indicating good for the latter two fit criteria

(Hu & Bentler, 1999). These fit criteria are less sample-size dependent and the RMSEA and TLI penalize models for lack of parsimony.

Reliability of the subscale scores was assessed using the omega index (Objective 4; Zinbarg et al., 2005), to circumvent the limitations of Cronbach's α (Dunn et al., 2014; Trizano-Hermosilla & Alvarado, 2016). Construct validity (Objective 5) was evaluated using Pearson's correlations between subscales of the CBRQ and conceptually similar constructs, specifically symptom severity, depression, anxiety and functional impairment using HD, IBD and chronic dizziness samples, as currently no other instruments measure cognitive and behavioural responses to symptoms, as also highlighted by Ryan et al. (2018).

Sensitivity to change (Objective 6) was estimated based on standardized mean differences and standardized response means using findings from two randomized-controlled trials of CBT. One used CBT designed to treat IBS, including changing IBS unhelpful cognitive interpretations about symptoms and associated avoidance behaviours as well as reducing symptom focusing and all-or-nothing behaviour if present (Moss-Morris et al., 2010). The other used a specifically designed CBT for fatigue in MS protocol which also included identifying and challenging unhelpful cognitive interpretations about symptoms, reducing all-or-nothing behaviour and using graded activity to reduce rest and avoidance of activity if present (van Kessel et al., 2008). Standardized mean difference was calculated as difference in mean change between groups divided by pooled standard deviation of mean change and standardized response mean was calculated as mean change divided by the standard deviation of mean change in each group. Standard deviation was estimated from 95% confidence intervals. Missing data were not imputed. Analyses were conducted in STATA 16 and Mplus 8.8 (Muthén & Muthén, 2017).

RESULTS

Sample characteristics

Table 2 presents the sample characteristics across the five datasets. Sociodemographic characteristics varied considerably by patient population. Except for the HD sample, there were more female than male patients. Gender proportion differed significantly across the datasets ($\chi^2[5, N = 991] = 74.59, p < .001$). Age differed significantly across the samples, according to a one-way ANOVA ($F(4, 987) = 69.97, p < .001$). A Games–Howell post hoc test revealed that all samples differed significantly in age, except for the comparison between CFS and IBD samples. Ethnicity was only available in the HD and chronic dizziness datasets. There were significantly more patients from minority ethnic groups in the HD sample compared to the chronic dizziness sample ($\chi^2[2, N = 359] = 58.84, p < .001$). Marital status and employment status were available in the HD, IBD and chronic dizziness datasets, with significant differences in proportions across these variables ($\chi^2[3, N = 527] = 13.23, p = .001$; $\chi^2[3, N = 503] = 116.47, p < .001$, respectively). More HD patients were not married/living with a partner and either retired or unemployed compared to IBD and chronic dizziness patients.

Confirmatory factor analysis results

Across the five samples, the 7- and 8-factor models performed comparably. Fit of the 8-factor model was only very slightly better than the 7-factor model in the CFS sample (see Table 3). The fit indices demonstrated generally adequate fit of the 7- and 8-factor models based on the relative χ^2 and RMSEA; however, the CFI and TLI parameters were marginally below the recommended cut-off of $\geq .95$. Small to moderate significant correlations were evident between most latent factors across the samples (see File S6). Not surprisingly, there were very large significant correlations between the subscales of Avoidance and Resting with the combined Avoidance/Resting subscale. Interestingly, correlations were moderate to large between Avoidance and Resting subscales (range .46–.69) and weaker between these subscales and all-or-nothing behaviour (range .16–.56). In the MS sample, Resting was not significantly correlated with

TABLE 2 Sample characteristics of each dataset.

Variable	CFS (N = 230)	MS (N = 221)	HD (N = 174)	IBD (N = 182)	Chronic dizziness (N = 185)
Female N (%)	169 (73.8%)	143 (64.7%)	64 (36.8%)	104 (57.1%)	137 (74.1%)
Age M (SD), range	39.57 (11.77), 16–79	44.70 (9.32), 22–67	58.96 (15.17), 25–92	41.05 (15.04), 20–83	53.57 (17.39), 18–90
Ethnicity N (%)					
Caucasian	187 (81.3%)	136 (91.3%)	75 (43.1%)	Not known	152 (82.2%)
Black			81 (46.6%)		17 (9.2%)
Asian			15 (8.6%)		8 (4.3%)
Marital status N (%)					
Married/Living with partner	144 (62.6%)	102 (68.5%) ^a	78 (44.8%)	112 (63.6%)	97 (52.4%)
Divorced/separated/never married/single/single parent/widowed	86 (37.4%)	47 (31.5%) ^a	92 (52.9%)	60 (33.0%)	88 (47.6%)
Employment status N (%)					
Working full-time/working part-time/housekeeping/self-employed			29 (16.7%)	132 (74.6%)	87 (47.0%)
Retired			59 (33.9%)	18 (10.2%)	58 (31.4%)
Unemployed			67 (38.5%)	27 (15.3%)	26 (14.1%)
Illness duration in months median (interquartile range), range	47.40 (73.20), 1.2–480	84.0 (111.0), .96–420 ^a	Dialysis vintage 34.50 (52), 3–304	144.0 (168), 12–600	24.00 (36), 1–384

Abbreviations: CFS, Chronic Fatigue Syndrome; HD, Haemodialysis; IBD, Inflammatory Bowel Disease; M, Mean; MS, Multiple Sclerosis; SD, Standard Deviation.

^aBased on cross-sectional dataset only (N = 149).

Damage beliefs, Embarrassment avoidance and Symptom focusing. As such, the model fit indices and correlations between latent factors do not provide definitive evidence on the superiority of the 7- or 8-factor models.

Descriptive statistics using the 7- and 8-factor models in each dataset are available in Table 4. Box plots for each of the subscales across the samples are available in File S7. As indicated by one-way ANOVAs, there were significant differences in scores on subscales of the CBRQ across the samples (see File S7).

Reliability

Omega coefficients of reliability for the 7- and 8-factor models across each sample are available in File S8. Subscales of the 7- and 8-factor models displayed satisfactory reliability across the samples ($\omega \geq .700$; except for Avoidance and Resting subscales in the MS dataset). The total score reliability fluctuated between .81 and .85 across the samples.

TABLE 3 Summary of CFA models in each sample.

Model	Description	Relative chi-square (χ^2)	CFI	TLI	RMSEA (90% CI)
Chronic Fatigue Syndrome					
1	1-factor	4.64	.639	.620	.126 (.122, .130)
2	7-factor	2.07	.898	.889	.068 (.063, .073)
3	8-factor	1.95	.909	.900	.064 (.059, .069)
Multiple Sclerosis					
1	1-factor	3.75	.642	.623	.112 (.107, .116)
2	7-factor	1.86	.891	.882	.062 (.057, .068)
3	8-factor	1.93	.883	.872	.065 (.060, .070)
Haemodialysis					
1	1-factor	3.29	.801	.791	.116 (.111, .121)
2	7-factor	1.60	.950	.946	.059 (.053, .066)
3	8-factor	1.80	.933	.927	.069 (.063, .075)
Chronic Dizziness					
1	1-factor	4.17	.743	.729	.131 (.126, .136)
2	7-factor	1.73	.943	.938	.063 (.057, .068)
3	8-factor	1.86	.933	.927	.068 (.062, .074)
Inflammatory Bowel Disease					
1	1-factor	3.29	.797	.786	.112 (.107, .117)
2	7-factor	1.62	.946	.942	.059 (.052, .065)
3	8-factor	1.69	.941	.936	.062 (.056, .068)

Note. 1-factor model $df = 740$; 7-factor model $df = 719$; 8-factor model $df = 712$.

Abbreviations: CFI, confirmatory fit index; RMSEA, Root mean-squared error of approximation; TLI, Tucker–Lewis Index.

Construct validity

The correlations between subscales of the CBRQ and related constructs, specifically: symptom severity, impairment, depression and anxiety are presented in Table 5. All CBRQ subscales were significantly associated with the full range of symptom severity, functional impairment and mood measures across conditions. Most of the correlations were moderate in size with some smaller relationship between some of the cognitive subscales and fatigue severity, depression and anxiety. Correlations did not exceed $r = .65$.

Sensitivity to change

In the sensitivity to change analysis of the CBRQ pre- and post-interventions from two RCTs: (1) An RCT of CBT designed specifically to treat MS fatigue, which included techniques to change unhelpful symptom interpretations and behaviours compared to a therapist time matched, relaxation therapy (Table 6), and (2) an RCT of CBT for IBS which addressed IBS-related symptom interpretations and behaviours compared to Treatment-As-Usual (TAU; Table 7). Standardized mean differences between groups were consistently in favour of CBT (Tables 6 and 7) with the CBT group ($SMR \geq -.9$) compared to relaxation training ($SMR \geq -.1$) and treatment as usual (TAU; SMR between $-.15$ and $.05$) in both the MS and the IBS datasets. In the MS dataset, 95% confidence intervals for the mean change across all subscales of the CBRQ (except for fear avoidance as not included in the study) in the CBT group did not contain zero, indicating a significant effect. A smaller improvement was evident in the MS relaxation

TABLE 4 Descriptive statistics based on 7-factor and 8-factor structure in each dataset.

Subscale of CBRQ (possible range)	CFS (N = 229–230) mean (SD), range	MS (N = 221) mean (SD), range	HD (N = 163–169) mean (SD), range	IBD mean (SD), range	Chronic dizziness (N = 185) mean (SD), range	Comparison
Fear avoidance (0–24)	13.52 (4.72), 2–24	11.36 (4.34), 0–22	11.03 (4.50), 0–23	8.69 (4.86), 0–24	12.94 (4.80), 0–24	ANOVA $F(4, 978) = 32.06, p < .001$
Catastrophising (0–16)	7.97 (3.55), 0–16	9.15 (3.54), 0–16	7.03 (3.91), 0–16	4.78 (3.75), 0–16	7.63 (3.87), 0–16	ANOVA $F(4, 976) = 36.49, p < .001$
Damage beliefs (0–20)	10.74 (3.85), 1–20	11.29 (3.25), 0–19	10.11 (3.60), 0–20	9.89 (3.95), 0–20	10.46 (4.01), 0–20	ANOVA $F(4, 977) = 4.35, p = .0017$
Embarrassment avoidance (0–24)	12.03 (5.82), 0–24	10.79 (5.82), 0–24	8.39 (5.80), 0–24	9.37 (6.23), 0–24	10.19 (6.33), 0–24	ANOVA $F(4, 971) = 10.33, p < .001$
Symptom focusing (0–24)	12.60 (5.03), 0–24	12.66 (4.83), 0–24	11.40 (5.66), 0–24	11.47 (5.42), 0–24	13.84 (5.72), 0–24	ANOVA $F(4, 972) = 6.46, p < .001$
All-or-nothing behaviour (0–20)	9.89 (4.53), 0–20	9.37 (4.37), 0–20	6.16 (4.71), 0–20	6.99 (5.18), 0–20	7.02 (5.18), 0–20	ANOVA $F(4, 976) = 23.60, p < .001$
Avoidance (0–16)	7.49 (3.50), 0–16	6.14 (3.05), 0–16	6.24 (4.04), 0–16	4.55 (3.45), 0–14	6.54 (3.60), 0–16	ANOVA $F(4, 976) = 16.50, p < .001$
Resting (0–16)	6.89 (3.52), 0–16	5.17 (2.86), 0–15	6.04 (3.72), 0–16	4.29 (2.97), 0–16	4.86 (3.60), 0–16	ANOVA $F(4, 975) = 19.13, p < .001$
Avoidance/resting (0–32)	14.38 (6.04), 2–32	11.31 (5.04), 0–29	12.28 (7.12), 0–32	8.81 (5.85), 0–30	11.40 (7.19), 0–30	ANOVA $F(4, 976) = 20.95, p < .001$

training group across subscales of the CBRQ, but only the within-group change in damage beliefs was significant. These within-group patterns were not observed in the TAU group in the CBT for IBS RCT. In the TAU group, there was an increase in fear avoidance beliefs and all-or-nothing behaviour, albeit not significant based on the 95% confidence intervals.

Factor structure and reliability of the CBRQ-SF

Ryan et al. (2018) proposed a shortened 6-factor version of the CBRQ, consisting of 18 items (displayed in File S9). This version was developed by removing lowest loading items in each factor and items that cross-loaded onto other factors. The catastrophising subscale was removed, as the items were found to be problematic, with low factor loadings and cross-loading. According to Ryan et al. (2018), CBRQ-SF (18 items) explained 67% of variance in their dataset, compared to 60% of variance explained by the full CBRQ (40 items).

The factor structure of the CBRQ-SF proposed by Ryan et al. (2018) across the five samples utilized here for analysis is presented in Table 8. Overall, the CBRQ-SF displayed good fit based on the model fit indices across the five samples, meeting recommended thresholds. Descriptive statistics for each subscale across the samples are available in File S10. Like the full version of the CBRQ, there were significant differences in subscales scores between the five samples (File S10). Although model fit of the

TABLE 5 Pearson correlations between subscales of the CBRQ and related constructs.

Sample	Variable	Fear avoidance	Catastrophising	Damage beliefs	Embarrassment avoidance	Symptom focusing	All-or-nothing behaviour	Avoidance / resting	Resting	Avoidance
Haemodialysis	Fatigue severity (CFQ)	.29 ($p = .002$)	.39 ($p < .001$)	.34 ($p < .001$)	.46 ($p < .001$)	.41 ($p < .001$)	.51 ($p < .001$)	.57 ($p < .001$)	.54 ($p < .001$)	.51 ($p < .001$)
	Fatigue-related functional impairment (WSAS)	.40 ($p < .001$)	.47 ($p < .001$)	.51 ($p < .001$)	.52 ($p < .001$)	.46 ($p < .001$)	.42 ($p < .001$)	.63 ($p < .001$)	.56 ($p < .001$)	.60 ($p < .001$)
	Depression (HADS D)	.41 ($p < .001$)	.51 ($p < .001$)	.50 ($p < .001$)	.59 ($p < .001$)	.64 ($p < .001$)	.42 ($p < .001$)	.59 ($p < .001$)	.53 ($p < .001$)	.55 ($p < .001$)
	Anxiety (HADS A)	.35 ($p < .001$)	.53 ($p < .001$)	.51 ($p < .001$)	.63 ($p < .001$)	.65 ($p < .001$)	.51 ($p < .001$)	.52 ($p < .001$)	.45 ($p < .001$)	.50 ($p < .001$)
Chronic Dizziness	Dizziness severity (NSS)	.46 ($p < .001$)	.41 ($p < .001$)	.23 ($p = .002$)	.44 ($p < .001$)	.28 ($p = .00001$)	.27 ($p = .00002$)	.43 ($p < .001$)	.37 ($p < .001$)	.41 ($p < .001$)
	Impact of dizziness on daily life (DHI)	.43 ($p < .001$)	.54 ($p < .001$)	.39 ($p < .001$)	.59 ($p < .001$)	.42 ($p < .001$)	.44 ($p < .001$)	.63 ($p < .001$)	.53 ($p < .001$)	.61 ($p < .001$)
	Depression (PHQ-9)	.35 ($p < .001$)	.49 ($p < .001$)	.35 ($p < .001$)	.51 ($p < .001$)	.42 ($p < .001$)	.46 ($p < .001$)	.59 ($p < .001$)	.55 ($p < .001$)	.52 ($p < .001$)
	Anxiety (GAD-7)	.25 ($p = 0.0006$)	.46 ($p < .001$)	.30 ($p < .001$)	.42 ($p < .001$)	.45 ($p < .001$)	.41 ($p < .001$)	.42 ($p < .001$)	.40 ($p < .001$)	.37 ($p < .001$)
Inflammatory Bowel Disease	Fatigue severity (MFI-20)	.55 ($p < .001$)	.54 ($p < .001$)	.37 ($p < .001$)	.57 ($p < .001$)	.54 ($p < .001$)	.49 ($p < .001$)	.66 ($p < .001$)	.67 ($p < .001$)	.51 ($p < .001$)
	Depression (HADS D)	0.43 ($p < .001$)	.47 ($p < .001$)	.32 ($p < .001$)	.45 ($p < .001$)	.45 ($p < .001$)	.49 ($p < .001$)	.51 ($p < .001$)	.39 ($p < .001$)	.53 ($p < .001$)
	Anxiety (HADS A)	.53 ($p < .001$)	.40 ($p < .001$)	.37 ($p < .001$)	.42 ($p < .001$)	.49 ($p < .001$)	.44 ($p < .001$)	.41 ($p < .001$)	.28 ($p = .00002$)	.45 ($p < .001$)

TABLE 6 Change scores across subscales of the CBRQ from baseline to post-treatment following receipt of CBT versus relaxation training in MS (Knoop et al., 2012; van Kessel et al., 2008).

Subscales of the CBRQ	CBT (<i>N</i> = 35)		Relaxation training (<i>N</i> = 35)		
	Mean change from baseline to post-treatment (95% CI)	SMR	Mean change from baseline to post-treatment (95% CI)	SMR	SMD
Catastrophising	-2.8 (-3.8 to -1.8)	-.96	-.5 (-1.4 to .3)	-.20	-.85
Damage beliefs	-4.0 (-5.5 to -2.5)	-.92	-1.1 (-2.2 to -.1)	-.36	-.77
Embarrassment avoidance	-2.9 (-4.1 to -1.8)	-.87	-.1 (-1.4 to 1.2)	-.03	-.78
Symptom focusing	-3.3 (-4.4 to -2.3)	-1.08	-.7 (-1.6 to .2)	-.27	-.91
All-or-nothing behaviour	-2.8 (-4.5 to -1.0)	-.55	-.1 (-1.0 to .9)	-.04	-.66
Avoidance/Resting	-2.9 (-4.6 to -1.2)	-.59	-.6 (-1.6 to .4)	-.21	-.57

Note: Negative SMRs indicate improvement (post-treatment-baseline); negative SMDs indicate effect in favour of CBT.

Abbreviations: CBT, cognitive behavioural therapy; CI, confidence interval; SMD, standardized mean difference between groups; SMR, standardized mean response within group.

TABLE 7 Change scores across subscales of the CBRQ from baseline to post-treatment following receipt of CBT versus TAU in IBS (Chilcot & Moss-Morris, 2013; Moss-Morris et al., 2010).

Subscales of the CBRQ	CBT (<i>N</i> = 31)		TAU (<i>N</i> = 33)		
	Mean change from baseline to post-treatment (95% CI)	SMR	Mean change from baseline to post-treatment (95% CI)	SMR	SMD
Fear avoidance	-1.2 (-2.0, -.3)	-.52	.20 (-1.1, 1.6)	.05	-.44
Catastrophising	-1.8 (-2.9, -.8)	-.63	-.3 (-1.2, .6)	-.12	-.56
Damage beliefs	-4.8 (-6.4, -3.3)	-1.13	-.4 (-1.6, .9)	-.11	-1.13
Embarrassment avoidance	-1.2 (-2.9, .4)	-.27	-.6 (-2.1, .8)	-.15	-.14
Symptom focusing	-2.4 (-4.1, -.6)	-.50	-.4 (-1.6, .8)	-.12	-.49
All-or-nothing behaviour	-.9 (-2.2, .5)	-.24	.07 (-.9, 1.0)	.03	-.05
Avoidance/Resting	-1.1 (-2.31, .1)	-.33	-.04 (-1.0, 1.0)	-.01	-.35

Note: Negative SMRs indicate improvement (post-treatment minus baseline); negative SMDs indicate effect in favour of CBT.

Abbreviations: CBT, cognitive behavioural therapy; CI, confidence interval; SMD, standardized mean difference between groups; SMR, standardized mean response within group; TAU, treatment as usual.

CBRQ-SF was good across the samples, scale reliability was somewhat reduced given the item reduction, fluctuating between .59 and .90 (File S11). Lower reliability was particularly evident for the Fear Avoidance subscale.

DISCUSSION

The aim of this paper was to evaluate the psychometric quality of the Cognitive Behavioural Responses to Symptoms Questionnaire (CBRQ) in five different LTC patient populations, positioning this scale as a transdiagnostic measure of cognitive interpretations of symptoms, and related behaviours which are associated with the experience of more severe and disabling symptoms, low mood and anxiety. For completeness, an overview of the original development of the CBRQ was also reported.

TABLE 8 Summary of model fit of the CBRQ-SF across the 5 samples.

Sample	Relative chi-square (χ^2)	CFI	TLI	RMSEA (90% CI)
CFS	1.99	.967	.958	.065 (.053, .078)
MS	1.61	.970	.962	.052 (.038, .066)
HD	2.22	.959	.947	.08 (.071, .099)
Chronic Dizziness	2.02	.973	.965	.075 (.061, .088)
IBD	1.49	.983	.979	.052 (.035, .067)

Note: $df = 120$.

Abbreviations: CFI, confirmatory fit index; RMSEA, Root mean-squared error of approximation; TLI, Tucker–Lewis Index.

Summary of findings

The preliminary pool of 62 items was originally reduced using PCA into a 40-item scale consisting of 7 latent factors (5 pertaining to cognitive interpretations of symptoms, namely Fear avoidance, Catastrophising, Damage beliefs, Embarrassment avoidance, and Symptom focusing; and 2 to behavioural responses to symptoms, namely All-or-nothing behaviour and Avoidance/Resting behaviour). According to CFA results, the fit indices of the originally proposed 7-factor structure and the 8-factor structure (where the avoidance/resting behavioural subscales divide into an Avoidance subscale and a Resting subscale); were generally comparable and displayed adequate fit in all five patient populations. The fit indices and correlations between latent factors do not provide definitive evidence in favour of the 7-factor or 8-factor models. Unlike Ryan et al. (2018), we, therefore; do not propose the use of one structure over the other. Instead, we strongly believe that further work is needed to expand on the behavioural items of this scale to determine how much overlap there is between avoidance and resting behaviours and as such whether they do indeed need to be considered as distinct behavioural responses. It is also important to note that while resting is a discrete behaviour that is likely to maintain its meaning across LTCs, the meaning of avoidance may differ across patient populations and may be harder to capture fully transdiagnostically. For instance, in response to dizziness as a symptom, an individual may avoid physical exercise/activity due to fear of falling, while in response to bowel symptoms, avoidance may entail avoiding going out unless an individual knows where the toilets are (Windgassen et al., 2017, 2019). Furthermore, based on the findings, we suggest that the use of subscale scores is most appropriate and do not advocate for the use of a total sum score on this measure. Omega reliability coefficients indicated satisfactory to excellent reliability of the subscales across the samples.

There were significant small to large positive correlations between subscales of the CBRQ and measures of symptom severity, impairment, depression and anxiety in HD, chronic dizziness and IBD samples, in line with what would be conceptually anticipated, indicating that the CBRQ has good construct validity and can reasonably well discriminate against related constructs as correlations did not exceed $r = .65$. Additionally, based on published trial data (Moss-Morris et al., 2010; van Kessel et al., 2008), CBRQ displayed good sensitivity to change in capturing changes in the CBRQ cognitions and behaviours following CBT when compared to relaxation training or TAU. It is important to note that whilst the CBT in these trials utilized different protocols, one specific to IBS cognitions and behaviours and the other to fatigue in MS, they both targeted similar processes to improve symptoms in these conditions.

Development of the CBRQ revealed further complexity to the construct of fear avoidance, encompassing an affective interpretation of symptoms related to fear of consequences of activity, and a cognitive interpretation of symptoms as a signal of damage happening in the body. It is therefore the only current measure capturing both dimensions. It is also the only measure that captures embarrassment avoidance and all-or-nothing behaviours related to chronic symptoms. These have emerged as important themes in qualitative research. Avoidance can be driven by embarrassment of symptoms, and not just fear (Crowe et al., 2017; Whitehead et al., 2016). To date, research has predominantly focused on avoidance behaviours, but an inconsistent pattern of activity and rest contingent on symptom severity is important based

on patients' narratives (e.g. Hewlett et al., 2005; Picariello et al., 2017; Scott et al., 2011) and can exert a negative influence on physiological processes over time, like the sleep–wake cycle (Moss-Morris, 2005). Therefore, in addition to the widely researched constructs of catastrophising (Lukkahatai & Saligan, 2013; Quartana et al., 2009), symptom focusing (Barends et al., 2020; Cella et al., 2011; Moss-Morris et al., 2005; Sarter et al., 2021), and avoidance behaviours (Hagger et al., 2017), the CBRQ enables a more nuanced understanding of processes related to the experience of symptoms. The findings here indicate that CBRQ is appropriate to be used across LTCs, both with populations who experience persistent physical symptoms without a currently defined pathophysiology and those who experience symptoms in the context of conditions with known biomedical aetiology.

The CBRQ-SF displayed good fit across the five samples, superior to the fit of the 40-item questionnaire; with a slight attenuation in reliability of the subscales likely as a result of the dramatic reduction in items, in line with the findings of Ryan et al. (2018). The internal consistency of the CBRQ-SF was also supported in an adolescent CFS sample (Loades et al., 2020). The original CBRQ is lengthy and may be burdensome for patients; therefore, the CBRQ-SF may be a good option to minimize patient burden and possibly missing data. It is important to note that the CBRQ-SF excludes the Catastrophising subscale. Further work is needed across different patient populations and symptoms to gauge how the CBRQ performs. Think-aloud methods could be valuable to address weaknesses of items related to catastrophising, particularly as recently observed with regards to content validity (Crombez et al., 2020), and avoidance behaviours and to build the robustness of these subscales.

Previous research and clinical implications

Even though, psychometric evidence was not available for this questionnaire until recently (Loades et al., 2020; Ryan et al., 2018), studies have utilized the CBRQ to identify correlates of symptoms across different patient populations (Ali et al., 2017; Artom et al., 2017; Chilcot et al., 2016; Herdman et al., 2020a, 2020b; Skerrett & Moss-Morris, 2006) and to explore mechanisms of change following CBT (Chalder et al., 2015; Chilcot & Moss-Morris, 2013; Stahl et al., 2014). Given the satisfactory psychometrics of this questionnaire, the CBRQ can introduce consistency in the measurement of cognitive and behavioural responses to symptoms across different conditions which can facilitate evidence synthesis. A recent meta-analysis of factors associated with CBT treatment outcome in populations living with persistent physical symptoms accentuated the sporadic and mixed nature of the current evidence base on processes of change (Sarter et al., 2021). Additionally, it may not only capture important changes that lead to improved outcomes following CBT, but it can also be utilized as part of assessment, for example, the focus and content of therapeutic techniques may vary depending on the coping procedures employed by a client (avoidance/resting versus all-or-nothing behaviour); or developing lower-intensity interventions while retaining key therapeutic techniques in line with stepped-care treatment models.

Limitations and future directions

Limitations of this paper should be considered. Firstly, sample sizes across conditions were relatively small which may have influenced model fit estimates for the CBRQ. The psychometric evaluation was restricted to the analysis of factor structure, internal reliability and construct validity. Although, there is some indication of the questionnaire's ability to capture change based on mediation analyses of trial data (Chalder et al., 2015; Chilcot & Moss-Morris, 2013; Stahl et al., 2014) and a preliminary evaluation of sensitivity to change here; test–retest reliability, responsiveness and sensitivity to change need to be evaluated in other conditions if this questionnaire is to be used in trials of psychological interventions, like CBT, as a key mediator of change. As mentioned earlier, concurrent validity cannot be evaluated given the unique nature of this measure; however, more work is required to evaluate the content validity of items, across patient populations, but also in different cultural groups and contexts, utilizing in-depth cognitive

interviewing for instance (Patrick et al., 2011). In light of the complexity of cognitive and behavioural constructs, establishing content validity is fundamental and more recently appropriate attention to validity has been advocated (Dixon & Johnston, 2019). For example, in a recent survey focused on examining the content validity of pain-catastrophising measures, considerable overlap was evident with pain-related worry and pain-related distress constructs (Crombez et al., 2020).

CONCLUSION

In conclusion, across five different samples, the CBRQ and its short version were reliable at capturing cognitive and behavioural responses to symptoms with evidence of construct validity, highlighting the multi-symptom and transdiagnostic properties of this questionnaire. The CBRQ-SF may be a suitable alternative for use in research and clinical practice. Further research is necessary to determine the test-retest reliability and sensitivity to change of the CBRQ and CBRQ-SF, but more importantly a more thorough evaluation of the content validity of the items, particularly across different patient populations.

AUTHOR CONTRIBUTIONS

Federica Picariello: Conceptualization; data curation; formal analysis; methodology; writing – original draft. **Joseph Chilcot:** Conceptualization; formal analysis; writing – original draft; writing – review and editing. **Trudie Chalder:** Conceptualization; data curation; methodology; writing – original draft; writing – review and editing. **David Herdman:** Data curation; writing – original draft; writing – review and editing. **Rona Moss-Morris:** Conceptualization; data curation; formal analysis; writing – original draft; writing – review and editing.

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CONFLICT OF INTEREST

TC reports grants from UK NIHR, UKRI and Guy's and St Thomas' Charity. She has delivered workshops on persistent physical symptoms including fatigue in the context of long-term conditions, during the conduct of the study for which she has received payment. She is the author of self-help books on fatigue.

DATA AVAILABILITY STATEMENT

Data are available on request from the corresponding author. The data were drawn from a number of studies all of which received ethical approval but were not pre-registered.

ETHICS STATEMENT

The authors assert that all procedures contributing to this work comply with the ethical standards of the relevant national and institutional committees on human experimentation and with the Helsinki Declaration of 1975, as revised in 2008.

ORCID

Federica Picariello  <https://orcid.org/0000-0002-2532-3290>

Joseph Chilcot  <https://orcid.org/0000-0001-6427-4690>

Trudie Chalder  <https://orcid.org/0000-0003-0775-1045>

David Herdman  <https://orcid.org/0000-0003-2122-5922>

Rona Moss-Morris  <https://orcid.org/0000-0002-2927-3446>

ENDNOTE

¹ Unlike Ryan et al. (2018), item EA4 did not load on the Avoidance factor in the MS and CFS datasets; therefore, it was left in the Embarrassment Avoidance latent factor.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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