

TITLE: Development and initial validation of a disease-specific bowel continence questionnaire for inflammatory bowel disease patients: the ICIQ-IBD

RUNNING HEAD: IBD bowel continence questionnaire: ICIQ-IBD

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ABSTRACT 246 words

Background and aims: Faecal incontinence (FI) related to inflammatory bowel disease (IBD) affects up to 74% of patients and is often under-reported in clinical encounters. A previous study found that several important bowel function concerns of patients with IBD are not addressed by existing FI questionnaires, especially differences between symptoms in relapse and remission. We have therefore adapted an existing FI assessment questionnaire specifically for patients with IBD.

Methods: 190 people participated. Phase 1 (development): the initial draft of the new questionnaire was developed from previously collected data and from results of a modified Delphi survey of IBD clinicians; questions were refined through six rounds of cognitive interviewing (n=24). Phase 2 (validation): the final version was tested (n=166), and retested (n=143) three to four weeks later.

Results: Missing data were minimal (1- 4%). Weighted kappa analysis showed moderate to good agreement of test-retest data. Factor rotational analysis revealed the relationship of questions to each other. The new questionnaire has two domains: 'Symptoms' and 'Quality of Life,' recording remission and relapse scores with simple summary scores for each. An additional 10 stand-alone questions address issues of specific concern to patients with IBD. The questionnaire demonstrates ability to capture changing symptoms and concerns between remission and relapse.

Conclusion: The new questionnaire has good content validity and is stable and reliable. Further testing to establish sensitivity to change is needed. The scale can be used by patients, researchers and practitioners to assess severity, fluctuation and impact of IBD-related FI.

Key words: continence, assessment, inflammatory bowel disease

INTRODUCTION

Inflammatory bowel disease-related faecal incontinence (FI) is a troublesome symptom which receives little attention clinically and is under-reported. Despite best-practice guidelines for active case-finding in people at high risk of FI,[1,2] clinicians may not ask [3] and patients may not report their bowel control problems either due to embarrassment, or not knowing how to raise the subject [4]. Concerns about loss of bowel control [5,6] and fear of incontinence even if it has never actually been experienced [7] amongst IBD patients are well-documented. The European “Impact of IBD” survey has highlighted the availability of toilets as a major concern [8] and a priority-setting exercise has put continence in the “top ten” research priorities of IBD patients [9] .

The International Consultation on Incontinence Questionnaire – Bowels (ICIQ-B) [10] is an example of a validated self-report tool for assessing FI and associated quality of life (QoL) in the general population. It was not known if it was suitable for assessing these issues in people with IBD. As part of a previous study [11], we asked participants to complete the ICIQ-B. Following completion, participants provided free text written comments on the suitability of the ICIQ-B for assessing their situation, and whether they could accurately report their incontinence symptoms, difficulties and concerns. Analysis revealed that the original questionnaire was only partially suitable for IBD, mostly because it did not allow reporting of the fluctuation in symptoms between remission and relapse and some other issues of importance to people with IBD, such as noise and social disruption caused by bowel activity, were not adequately addressed. Since the disease activity status of IBD directly affects psychosocial consequences and quality of life [12], the true impact of the unpredictable nature of IBD and related bowel symptoms cannot be captured using existing FI questionnaires, none of which enable reporting of symptoms in active and quiescent phases of disease.

To enable patients to self-report their symptoms, and researchers and clinicians to accurately assess IBD-related FI, we aimed to develop a new, IBD-specific FI assessment tool following the International Consultation on Incontinence Modular Questionnaire (ICIQ) methodology [13]. The ICIQ is an international project developing a series of fully validated questionnaires for both research and clinical use. All questionnaires are developed with a similar methodology [14] putting the patient voice at the heart of the content by asking people what is important to them, and using this information alongside clinician / expert opinion when designing questionnaires. These questionnaires, currently available for urinary and faecal incontinence and pelvic organ prolapse, are seen as the international gold standard for incontinence research and clinical practice, and are being widely adopted [13].

METHODS

The study had two phases. The questionnaire was developed in phase one, and underwent initial validation (test-retest) in phase two.

Study sample

Participants were recruited from our database of people with IBD willing to take part in research. All contacts (n=935) were emailed and invited to visit a secure webpage which provided information about the study and the inclusion criteria: over 18 years old, self-reported diagnosis of Crohn's disease (CD) ulcerative colitis (UC), or other form of IBD (including indeterminate IBD, and proctitis), no current stoma, living anywhere within the UK. 149 people agreed to participate in either the cognitive interviews, or completion of paper questionnaires during the validation phase. Further recruitment via the newsletter, facebook® and Twitter® pages of a major UK IBD charity attracted an additional 64 potential participants (total=213). Participants contributed once in Phase 1 or in Phase 2. Twenty five respondents were purposefully selected for Phase 1 (development) to represent a broad

spread of gender, age, diagnosis and geographic location; one person later withdrew due to family illness. The remaining 188 were invited to participate in Phase 2.

Sample size

Sample size for Phase 1 was not calculated, since the process of item development requires cognitive interviews to continue until no further changes are required. Our previous experience, supported in other work [10,14] is that this is usually achieved over four or five rounds of interviews, with between 24 and 30 participants.

Sample size for Phase 2 was guided by published recommendations for a minimum sample size of 30 – 40 participants for questionnaire development [15], with participant numbers increasing accordingly where there is an increase in the number of questionnaire items being tested [16]. Without prior knowledge of the likely distribution of responses within the new questionnaire, the ability to determine the width of the confidence interval was affected, making an *a priori* calculation impossible.

Data collection tools

Demographic data (age, gender and IBD diagnosis) were collected from all participants either at interview, or during completion of the paper questionnaire. In Phase 2, disease activity was assessed using the Harvey Bradshaw Crohn's Disease Activity Index (CDAI)[17] for those with CD, and the Walmsley et al. Simple Clinical Colitis Activity Index (SCCAI) [18] for those with UC. A disease activity score of 4 or below was taken to indicate remission; a score of 5 or above indicated active IBD.

Phase one (questionnaire development)

Item generation

We elected to amend the original ICIQ-B, since participants in a previous study [11] had reported that it performed reasonably well despite several IBD-related omissions from, or

recommended amendments to, the questionnaire. It is usual practice to begin development of this type of patient-reported measure by interviewing people with the condition of interest to identify potential content [19], but the existing feedback [11] replaced this stage. We extracted responses of 650 previous participants (20% of a total dataset of 3264) to the question: *Please comment on whether the ICIQ-B is relevant to your needs, and whether it reflects your situation and concerns. Does it ask about the issues that are important to you?* A simple thematic analysis, continuing until nothing new emerged, identified several recurring issues [Table 1].

A modified Delphi survey of a convenience sample of 10 clinicians (four IBD nurse specialists, three gastroenterologists, two dieticians and one specialist physiotherapist) was conducted by compiling a checklist of all symptoms and concerns identified by patients, plus the items in the original ICIQ-B, and inviting the clinicians to indicate whether each item was *Not clinically relevant*, *Moderately clinically relevant*, or *Essential to know*. Consensus was achieved at the first attempt, and all items marked as *Essential to know* were included in the first draft questionnaire. Notably, many of the psychosocial issues identified by patients as important were not marked as such by clinicians, but these were retained so that the new questionnaire included a combination of clinician and patient priorities, enhancing content validity. The preliminary version of the International Consultation on Incontinence Questionnaire-Inflammatory Bowel Disease (ICIQ-IBD) retained 13 original questions from the ICIQ-B, amended the wording of a further seven original questions, and included 19 new questions based on patient and clinician views. It also included capacity to record a 'Usual / remission', and an 'At worst / relapse' response to each question. Bother scores for each question (recorded on a scale of 0 (no bother at all) to 10 (very bothersome) were retained from the original ICIQ-B. Although not included in scoring, bother scores enable patients to indicate the impact that symptoms have on them and can help focus clinical intervention.

Data collection and concurrent analysis

Data were collected via cognitive interviews to establish face and content validity of the draft questionnaire¹⁷. Face validity refers to transparency of purpose which enables the completing person to readily understand the purpose of the questions. Content validity refers to the ability of the questionnaire to address the most important issues relevant to the topic. Face and content validity of the draft ICIQ-IBD were tested through a series of six cognitive interview rounds. Using 'think aloud' and verbal probing techniques, a total of 24 participants were interviewed individually. 'Think aloud' encourages the participant to explain verbally what they think of when they first read the question, and when considering their response [20]. This enables the identification of questions which may be ambiguous or open to misinterpretation, and confirms the ability of the person to give the response they wish. Early interviews (rounds 1 – 4) were digitally audio-recorded and professionally transcribed. Later interviews (rounds 5-6) were not recorded as the questionnaire was nearing its finalised form and few new issues were being raised. Written notes were instead made by the interviewer (LD) after the interviewees had completed the draft questionnaire. The questionnaire was revised after each round of interviews following simple thematic analysis of transcripts and notes, and discussion between two of the authors (LD, CN). Grammar, presentation, layout, content, ease of use and item completion rate with a researcher present, and absent, were tested. After six rounds of interviews and modifications, there were no further amendments, resulting in a 39-item questionnaire ready for initial validation.

Phase 2 (initial validation)

The purpose of initial validation is to determine how well the new questionnaire does what it sets out to do, by identifying whether the questions are completed by the majority of respondents (completeness), whether the tool can discriminate between respondents by detecting better or worse disease status (sensitivity), and whether it performs the same or similarly on more than one occasion (test-retest reliability).

Data collection and analysis

Participants completed paper copies of the ICIQ-IBD and either the CDAI[17], or the SCCAI [18] on two occasions, approximately three to four weeks apart, returning these by post. One email reminder was sent to participants. Of 188 people who agreed to take part, 166 returned the test questionnaire (88%); of these, 143 returned the retest questionnaire (86%). Data from the questionnaires were input into Excel manually, checked for errors, and transferred to SPSS for analysis.

The first set of analyses examined the completeness of each item of the questionnaire. The number and percentage of responses with either missing or not applicable data was calculated for each item. Individual questions were summarised by the number and percentage of responses in each category. In addition to providing descriptive information, the spread of responses to each question was examined to ensure that there were a range of responses, indicating that the question was useful in discriminating between patients.

The reliability of the questionnaire items was examined by assessing the agreement between the test and retest questionnaires in people with stable disease. Due to the ordinal nature of each question, the agreement between the questionnaires was assessed using the weighted kappa method. Kappa values of less than 0.20 were considered as poor agreement, between 0.2 and 0.4 as fair agreement, between 0.6 and 0.8 as good agreement, and between 0.8 and 1.0 as very good agreement.

Ethical Considerations

Ethical approval for the study was given by the Research Ethics Committee at King's College London (Reference PNM/12/13-23). Written informed consent was secured from Phase 1 interviewees immediately before the interview took place; completion and return of completed test and retest questionnaires was taken as implied consent for Phase 2.

RESULTS

Participant details for both phases are presented in Table 2. Phase 1 results were incorporated iteratively into the questionnaire as it developed, and are not discussed further here. Phase 2 results are presented below.

Item reduction

Two questions, addressing rectal pain with or without fistulising Crohn's disease were relevant to only a very few participants and have been removed as retaining these would weaken the validity of the tool. All remaining questions with intermediate 'Not applicable' scores were retained, since the team agreed this information was important in accurately assessing the severity of FI. Two questions (Bristol Stool Chart and a free-text question) are formatted differently from the other questions. Although not reported further here, these are retained in the final questionnaire. The analysis of the remaining 35 questions is reported here.

Completeness

Levels of missing data were low: 1- 4% of respondents failed to respond to all or part of a question, mostly apparently caused by turning two pages over together and missing the same block of four questions. 1-2% of people missed sporadic responses, usually in the 'at worst' section of a question. Levels of 'Not applicable' data were moderate (8 - 34%) for five questions referring to having solid stool, mucus, bowel accidents, having a sexual partner, and disruption to working life. The majority of questions were applicable to the majority of respondents.

Sensitivity

The ability of a questionnaire to discriminate between respondents is demonstrated when there is a good spread of responses across all available categories, rather than the majority of responses clustering at one end of the range or the other. Results of the test data (n=166)

indicated that the majority of responses were spread across the available categories, demonstrating that the questionnaire can discriminate between patients with a range of symptoms and concerns.

Four questions related to having bowels open at night, being incontinent at night, ability to reach the toilet in time, and taking constipating medication showed 'usual / in remission' responses clustered towards the lower scores (less of a problem). This is probably explained by these issues generally being less of a problem to most participants in remission. 'At worst / relapse' responses to having bowels open at night, being incontinent at night, and ability to reach the toilet in time questions were spread across all score categories showing varying degrees of problem caused by relapse. Three questions related to taking constipating medication, experiencing constipation, and straining to open bowels, showed 'at worst / relapse' responses clustered towards the lower scores (less of a problem).

Stability (test-retest reliability)

There were a total of 143 patients with both test and retest data. Of these, 22 patients changed from remission to relapse or vice-versa between the two questionnaires, and a further 11 did not have sufficient SCCAI or CDAI data at both time-points to indicate if the disease had changed. These 33 patients were excluded, resulting in 110 paired sets of data suitable for inclusion in the test-retest analysis.

The agreement between the test and retest measurements was examined for both the 'usual' and 'at worst' scores. In the 'usual' scores, 24 of 35 questions showed good agreement (kappa score 0.6-0.8), and 11 of 35 showed moderate agreement (kappa score 0.4-0.6). None showed very good, fair or poor agreement. In the 'at worst' scores, 23 of 35 questions showed good agreement, 11 showed moderate agreement, and one, asking if bowel accidents or leakages are predictable, showed fair agreement.

Factor analysis

The associations between the component items of the questionnaire were examined using a factor analysis. A scree plot was used to help select the number of factors to examine further, and a varimax rotation aided the interpretation of the results. For each identified factor, a score (or factor loading) for each question on that component, was obtained. Where participants could select a 'not applicable' option, a score of 0 was assigned. No imputations were performed for missing values. An initial unrestricted exploratory factor analysis identified only one factor containing twenty five items, with ten items remaining that did not load onto the factor. Further solutions were sought to explore whether there were identifiable domains within the single factor, by separating symptom and quality of life items for further analyses. Within the symptom item dataset a single factor was identified with an eigenvalue greater than 1 and factor loadings that ranged from 0.4132-0.7901. These 11 items were termed the *IBD-FI Symptoms* domain. Within the quality of life items dataset a further factor was identified, and factor loadings ranged from 0.5504-0.8607. These 14 items were termed the *IBD-FI Quality of Life (QoL)* domain. The originally identified 10 items which did not load onto either of the two main domains were retained as stand-alone items under the heading *Other Concerns* to enable assessment of other issues considered to be important from the patient or clinical perspective [Table 3].

Development of a scoring system

A simple summative score for each domain is indicated with stand-alone items being unscored. Logistic regression is normally used to identify the items which should be included in questionnaires and to include some weighting for retained variables when there is a definitive outcome being predicted. Where there is no hard endpoint, as in this case, this approach is not appropriate. Scores for both domains can be calculated to indicate severity of FI symptoms and FI-related QoL in remission, and in relapse. The *IBD-FI Symptoms* domain is scored from 0 to 46, and the *IBD-FI Quality of Life* domain is scored from 0 to 56

for both remission and relapse. A higher score for each domain indicates more severe symptoms and greater impact on quality of life.

Other concerns are not scored and should be evaluated on a per item basis. The Bristol Stool Chart is additionally included in this section to enable indication and comparison of stool type, and a free-text option is provided to enable reporting of related issues of concern not addressed in the previous questions.

Overall, there are 37 items in this new questionnaire for assessing IBD FI symptoms and QoL in remission and relapse.

Participants' questionnaire responses

The test results of all participants with stable disease who provided test-retest data (n=110) was aggregated and the mean overall scores were analysed descriptively. Bother scores, indicated in the ICIQ-IBD on a scale of 0 (no bother) to 10 (greatly bothersome) are, for ease of reporting here, clustered into 3 groups: 0 – 3 = low, 4 – 6 = medium, and 7 – 10 = high bother.

The IBD-FI Symptoms domain

In remission, the majority have low bother scores (0-3) on most items, but report high bother scores (7-10) for having to rush to the toilet (n=50; 45.5%) and feeling that they produce foul-smelling flatus (n=56; 51%). In relapse, these, plus the time between feeling the call to stool and defecating are the most bothersome symptoms for 79% (n=87), 73.6% (n=81) and 81.8% (n=90) respectively.

The IBD-FI Quality of Life Domain

In remission, the majority have low bother scores (0-3) for 9 out of 14 QoL items, but report high bother scores (7-10) for worrying about using a toilet away from home (n=60; 54.5%),

feeling embarrassed about bowel control problems (n=49; 44.5%), being concerned about others' perceptions of them (n=47; 42.7%), making sure they know where a toilet is and worrying about reaching the toilet in time (both n=50; 45.5%). Worrying about the possibility of having a bowel action drew similar low and high bother scores (n=48; 43.6%, and n=47; 42.7% respectively). In relapse, the majority of participants (n=72-93; 65.5% - 84.5%) report high bother scores (7-10) for all 14 questions. For some items, the numbers reporting high bother more than doubled between remission and relapse, for example, Q.18 (worrying about getting to a toilet on time).

Other Concerns

During remission, most respondents reported low (0-3) bother scores on all items except feeling less attractive to their partner, which had equal numbers for low (0-3) and high (7-10) bother (both n=36; 32.7%). Respondents were bothered by noisy flatus than not, in both remission and relapse (n=56; 50.9%, and n=81; 73.6%). In relapse, they were most bothered by noisy flatus, the frequency of bowel actions (n=88; 80%), and stool type (n=74; 67.3%).

DISCUSSION

The ICIQ-IBD complements a range of other patient reported outcome measures applicable to IBD, including those addressing patient perception of disease control [21], fatigue [22] and separate CD and UC outcome measures [23,24]. On initial testing, the ICIQ-IBD scale has been found to have good face and content validity, and reasonable stability (reliability). Clinical monitoring relies on dependable findings and validity is affected if the instrument is unreliable [25]. In the ICIQ-IBD, many questions show a clustering of responses at the 'never' or 'rarely' end of the scale in remission, and at the 'most of the time' or 'always' end of the scale in relapse; this aligns with expected changes in symptoms between remission and relapse, indicating that the ICIQ-IBD is able to detect the difference between these two disease states. Some questions, particularly in the QoL domain, cluster responses at the 'most of the time' or 'always' end of the scale in both relapse and remission, indicating the

persistent impact that bowel control concerns can have on patients regardless of disease status.

At this stage of validation, the tool scores 10 positive ratings out of 18 recommended quality assessment items for evaluation of health status questionnaires [13]. The remaining eight quality assessment items will be addressed in later full psychometric validation. The robustness of the questionnaire is confirmed through the diligent and rigorous developmental process, with participants with IBD contributing throughout, ensuring that the language of the ICIQ-IBD is straight-forward, using lay terms which increase readability and enable accurate interpretation.

FI in IBD is a troublesome aspect of disease. As we have demonstrated previously, poor control leading to FI is a complex experience [7] and help-seeking for the symptom can be impaired by difficulties verbalising this taboo and embarrassing symptom [4]. The UK National Institute for Clinical Excellence (NICE) recommends active case-finding amongst patient populations where FI is likely to be a problem, including those with IBD [1,26]. The Nurses-European Crohn's & Colitis Organisation (N-ECCO) consensus statement specifically recommends that nurses assess IBD patients for bowel control difficulties [2]. The ICIQ-IBD provides a means of enabling people to accurately report their symptoms, the impact on their QoL, and other contextual influences without having to verbalise these. It can be completed face to face with the clinician present, or independently by the patient alone. As such, it can create a communication bridge and promote discussion and assessment of patient need. It also identifies changing concerns and priorities that arise for patients between remission and relapse, information which can guide individualised patient advice and support.

Overall, there is reasonable agreement between test and retest scores, suggesting that the questionnaire is stable and reliable. The value of including response options for remission

and relapse in the ICIQ-IBD is demonstrated by variation in spread of responses across the five response options of *Never*, *Rarely*, *Some of the time*, *Most of the time* and *Always*. In remission, most results were clustered in one or two response options, whilst in relapse similar numbers of responses are often spread across response options. This, and the reporting of high bother scores for 10 out of 11 symptom questions in relapse, versus two out of 11 in remission, indicates that patients experience troublesome and bothersome symptoms in relapse. The ability to report response options and bother scores in remission and relapse enables reporting of the fluctuating impact of continence-related issues on QoL in IBD. Bother scores for QoL issues were as high as for symptoms, indicating patients are bothered by impact as well as symptoms.

Clinical utility

With initial validation demonstrated, the ICIQ-IBD provides a means of undertaking an assessment of patients' FI symptoms, such as that required by specialist IBD and continence nurses, or for research purposes. The questionnaire takes approximately 15 minutes to complete. A PDF file of the questionnaire is available on request from the lead author and the ICIQ office: www.iciq.net. Lengthy questionnaires are likely to be less applicable in day to day clinical practice because of time constraints [27] and so a short form is in development. Sensitivity to change will be tested during an intervention study taking place between 2015 and 2017.

The final full version of the ICIQ-IBD for patient self-completion consists of 37 questions, and includes 'bother' scores. Patients select a score per item for how they are in remission and for relapse, and indicate the degree to which the particular item bothers them. Bother scores are not included in the scoring but enable reporting of the impact of symptoms. Some symptoms and quality of life issues might be reported as more bothersome in remission than in relapse. For example, a patient might indicate a bother score of 4 for ability to control loss

of loose stool in relapse, but score this as 10 when in remission suggesting that the symptom is tolerated during disease flare-up, but is disruptive during remission.

Between 46 (41.8%) and 87 (79%) participants report high (7-10) bother scores per question in relapse. The discrepancy between those reporting higher bother scores, and the distribution of scores across items indicates that a symptom does not need to be at its worst for it to cause a great deal of bother. Details can be easily identified when assessing each patient's scores, providing information to assist clinical decision-making and to identify priorities for treatment from the individual's perspective.

LIMITATIONS

Participants were drawn from an existing database of research-keen individuals, all members of a UK IBD charity. These were all self-selected, and may have more severe FI symptoms than the wider IBD community which may affect representativeness of the sample. However, in questionnaire development the intention is not to have a representative sample of the whole population, but rather a rich sample of those with experience of the issue of interest.

Although the demographics of phase one and phase two participants are similar, the majority of participants were female (72%) compared with the female membership (64%) of the charity from which they were recruited, and the wider IBD population in which only slightly more than 50% of patients are female.

CONCLUSION

The ICIQ-IBD is shown preliminary validity and reliability instrument for assessing IBD-related FI in clinical and research settings. Future development of an electronic version with inbuilt scoring function, and a shorter format will facilitate use. The questionnaire provides a

means of assessing and addressing a potentially embarrassing problem which patients and clinicians find difficult to broach. Further testing is required to determine whether the new ICIQ-IBD correlates with existing bowel symptom and QoL assessment scores, such as the IBD-Q [28], and to assess sensitivity to change.

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