

Letter to the Editor

Patient involvement in the development of asthma-specific patient-reported outcome measures: A systematic review

To the Editor:

Patient-reported outcome measures (PROMs) are increasingly being used as indicators of health from the patient's perspective, and have the potential to improve the quality of health services and treatment outcomes. PROMs are usually standardized self-completed questionnaires measuring health status (symptoms or function), health-related quality of life, or general health perceptions.¹ For their full potential to be realized, the outcomes measured must be relevant to patients,² which suggests that patients should be involved in the development cycle.³ Three recent structured reviews⁴⁻⁶ of PROMs for asthma have focused on psychometric properties as directed by published lists of attributes and criteria, but neither reviewed the extent of patient involvement in their development. However, Apfelbacher et al⁴ observed that user involvement appeared to decrease as the PROM development process progressed, with psychometric analysis displacing patient involvement.

The desirability of patient involvement in the development of PROMs has 2 main components. The first, which applies to health services research in general, is that as consumers (and often funders) of research, patients should have some input into the research process, the assumption being that this will lead to higher-quality research that is more relevant to the needs of patients.⁷ The second, which is specific to the development of PROMs, is that the face validity, acceptability, interpretability, and relevance of the outcomes measured will be improved by developing the PROM in consultation with patients.⁸ There is very little systematic research into the degree to which patient involvement improves the quality of research; however, there is some evidence that patient involvement can improve the quality of PROMs with respect to acceptability,⁹ readability and interpretability,¹⁰ and appropriateness/relevance.¹¹ We therefore aimed to determine the extent of patient involvement in the development of existing patient-reported outcome measures for asthma.

We obtained the references cited in the systematic review by Gibbons and Fitzpatrick⁵ of 22 asthma-specific PROMs plus additional modified versions of 2 of these PROMs. Two reviewers summarized the degree of patient involvement at each stage in the PROM's development cycle: (A) concept generation, (B) item generation, (C) item reduction, and (D) content validation. Patient involvement was scored by using the scheme of Haywood et al⁹ devised from a UK advisory group's definition of involvement in research¹²:

Level 1 (Consultation): Patients are consulted for their views (eg, via survey, focus group, or other methodology), but these views may not necessarily be adopted.

Level 2 (Collaboration): This involves active, ongoing partnership between researchers and patients in the development of the PROM.

Level 3 (User-led): Patients control, direct, and manage research.

This definition of patient involvement requires that patients at the very least be consulted about the process at hand and not merely be participants in a study; hence, studies in which patients provided only data as participants were rated 0, reflecting the lack of consultation.

TABLE I. Level of patient involvement in the development of questionnaires

PROM	Year of publication	Development phase				
		A	B	C	D	All
Living with Asthma Questionnaire (LWAQ)	1990	1	1	0	1	3
Asthma Quality of Life Questionnaire (AQLQ)	1992	0	1	0	1	2
Mini AQLQ	1999	0	0	0	1	1
Standardized AQLQ (AQLQ(S))	1999	0	0	0	1	1
Marks Asthma Quality of Life Questionnaire (MAQLQ)	1991	0	1	0	0	1
Asthma Therapy Assessment Questionnaire	1999	0	0	0	1	1
Acute AQLQ	2004	0	0	0	1	1
Asthma Self-efficacy Scale (ASES)	1987	0	1	0	0	1
Asthma Symptom Utility Index (ASUI)	1998	0	1	0	0	1
St George's Respiratory Questionnaire (SGRQ)	1991	0	0	0	1	1
Asthma Quality of Life Utility Index (AQL-5D)	2007	0	0	0	0	0
Modified Marks Asthma Quality of Life Questionnaire (m-MAQLQ)	2000	0	0	0	0	0
Perceived Control of Asthma Questionnaire (PCAQ)	1997	0	0	0	0	0
Asthma Control Diary (ACD)	2000	0	0	0	0	0
Asthma Control Questionnaire (ACQ)	1999	0	0	0	0	0
Asthma Control Scoring System (ACSS)	2002	0	0	0	0	0
Modified & Shortened LWAQ (ms-LWAQ)	1999	0	0	0	0	0
Lara Asthma Symptom Scale	2000	0	0	0	0	0
Asthma Control Test (ACT)	2004	0	0	0	0	0
Asthma Impact Survey (AIS-6)	2007	0	0	0	0	0
Royal College of Physicians 3 Questions	1999	0	0	0	0	0
Work Productivity & Activity Impairment: Asthma (WPAI)	2008	0	0	0	0	0
Airways Questionnaire 20 (AQ-20)	2008	0	0	0	0	0
Asthma Outcomes Monitoring System	2000	0	0	0	0	0

A, Concept generation; B, item generation; C, item reduction; D, content validation.

Hence, the extent of patient involvement at each stage in the development of PROM could range from 0 (no patient involvement) to 3 (maximal patient involvement). In addition, a summary total was computed as the sum of all stages of questionnaire development in which any patient involvement was recorded. The summary totals could range from 0 (no patient involvement at any stage) to 12 (highest level of patient involvement at all stages). Ratings were completed independently and reviewer agreement was 100%.

Asthma PROMs varied considerably in the degree of patient involvement in their development: 13 questionnaires (54%) were developed with no patient involvement according to Haywood et al's definition, and in no case was patient involvement beyond consultation (level 1; see Table I). The concepts (or domains) measured by the questionnaire were developed with meaningful patient involvement in only 1 case (Living with Asthma Questionnaire [LWAQ]). Five questionnaires (LWAQ, Asthma Quality of Life Questionnaire [AQLQ], Marks AQLQ, Asthma Self-efficacy Scale, and Asthma Self-efficacy Scale) involved patients in the generation of items and 7 in the content validation (LWAQ, AQLQ, mini AQLQ, Asthma Therapy Assessment

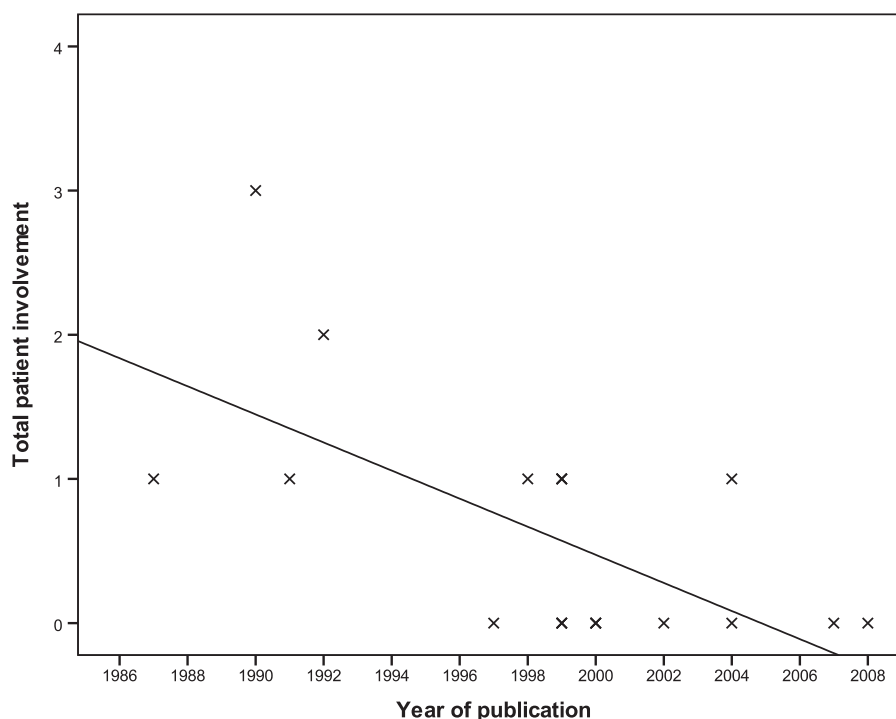


FIG 1. Patient involvement over time (1987 to 2008).

Questionnaire, Acute AQLQ, Standardized AQLQ, and St George's Respiratory Questionnaire). No questionnaire involved patients in the item reduction phase. Of all the instruments we reviewed, the LWAQ scored the highest for patient involvement, with meaningful patient involvement in 3 of the 4 phases of development. There was a significant negative correlation between the total extent of patient involvement and the date of publication ($r = -0.65$; $P = .001$; $\rho = -0.67$; $P = .001$), indicating that patient involvement in questionnaire development has declined over time (Fig 1).

This is the first systematic review focusing on the extent of patient involvement in the development of asthma-specific quality-of-life measures. It shows that there has been substantial variation in the role of patient involvement in the development of PROMs for asthma. Clearly, the extent to which patients may be involved, and in what capacity, will vary over the development cycle: it may not be possible, for example, for content development to be completely user-led. Despite this, neglecting patient involvement may have an effect on the extent to which different instruments reflect the patients' perspectives of health status and quality of life. Indeed, the extent to which the questionnaires measure meaningful outcomes for patients is largely determined by the concept generation phase, yet only 1 questionnaire (LWAQ) systematically included the patient perspective as to what should be measured in the first place. In this context, the subsequent development phases of item selection, item reduction, and content validation are meaningful only to the extent that they support the themes and perspectives derived from patient experience: yet in only 1 case were these processes applied to material derived from patients.

The complete absence of patient involvement in the item reduction phase appears to be driven by the largely statistical nature of this process: typically items are deleted from the questionnaire following principal components analysis or factor

analysis. While this may form a defensible basis for excluding items, it allows for items important to patients to be dropped from the final version of the questionnaire. It is therefore possible that the material generated by patients in the earlier phases is excluded in the item reduction phase, "losing" important indices of quality of life and health status.

Of greater concern is the apparent decline in patients' involvement in the development of more recent PROMs for asthma. This may reflect a consensus that earlier development work had already identified the areas of health status of importance to patients with asthma, but our review challenges the solidity of these foundations.

Our review did not examine the specific nature of the patient involvement: hence, it is possible that the patient involvement we have recorded was itself inappropriate or suboptimal. For example, involving only a self-selected and interested group of patients would not be expected to generate representative material. It is also possible that the patient involvement was biased by the nature of the interaction with the researchers. It is therefore unsafe to assume that the instances of patient involvement identified in this review were all equally valid and necessarily resulted in an improved PROM. However, this underscores our conclusion that future developers of asthma-specific PROMs should be attentive to appropriate patient involvement at all stages of the development cycle, and to documenting the nature of this involvement, if PROMs are to reflect health outcomes relevant to the patient.

Edward Frew, BA^a
Matthew Hankins, PhD^b
Helen E. Smith, DM^c

From ^aNorwich Medical School, University of East Anglia, Brighton; ^bthe Faculty of Health Sciences, University of Southampton, Southampton; and ^cthe Division of

Primary Care and Public Health, Brighton and Sussex Medical School, Brighton, United Kingdom. E-mail: m.hankins@soton.ac.uk.

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