

## Concise report

## Developing and validating an index for measuring health in patients with ankylosing spondylitis

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## Abstract

**Objectives.** The impact of disease on functioning is the essential information for clinicians when reporting on health problems of individuals. The International Classification of Functioning, Disability and Health (ICF) is a comprehensive and universally accepted model to classify and describe functioning, disability and health in a systematic way. The objective of this article is to outline the development and validation of a health index for patients with AS based on the ICF as a use case.

**Methods.** The project is a combined effort of the Assessment of SpondyloArthritis International Society, the ICF Research Branch of the World Health Organization (WHO) Collaboration Centre of the Family of International Classifications and the WHO. There are five steps in the development and validation of the health index for patients with AS: (i) development of an item pool; (ii) identification of candidate items; (iii) item selection; (iv) item reduction; and (v) creation of a final version. Consensus about items that have to be part will be reached in a final consensus conference.

**Results.** During a meeting in February 2009, we coordinated the development process of the health index for patients with AS. The results of this investigation will be the health index for patients with AS.

**Conclusion.** The goal of developing a health index for patients with AS based on the ICF is very much in line with the broader goal of the WHO to define health indices to ensure the comparability of them within the framework of the ICF.

**Key words:** Ankylosing spondylitis, Functioning, Health status, Health status measures, International Classification of Functioning, Disability and Health.

## Introduction

AS, the main subgroup of the spondyloarthritides, is a chronic inflammatory disease characterized by inflammation and ankylosis of the axial skeleton [1]. Since AS

usually starts in early adulthood, the lifetime impact of AS on patients can be considerable. Apart from symptoms such as pain, stiffness and fatigue, patients are limited in activities and restricted in participation in society.

Functioning is increasingly being taken into account in assessing the impact of chronic diseases on the individual, as well as the effectiveness of treatments. Functioning is often referred to as function. However, it is important to realize that function is often limited to physical function ignoring the complexity of global functioning. With the approval of the International Classification of Functioning, Disability and Health (ICF) by the World Health Organization (WHO) there is now a universally accepted model to classify and describe functioning, disability and health in persons with all kind of diseases or conditions in a more systematic way [2]. The ICF allows clinicians to comprehensively describe and categorize functioning and disability of their patients in a way that can be understood by all health professionals. The

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ICF model identifies three components of the dimension of functioning, namely body functions, body structures, as well as activity and participation. To apply the ICF in practice, ICF Core Sets, which comprise specific categories relevant for a particular condition, have been developed for various diseases. Both a Brief ICF Core Set and a Comprehensive ICF Core Set have been established for patients with AS [3].

Clinicians often rely on summary scores that integrate different aspects of functioning and which are usually constructed by adding up the responses to different items. Summary scores allow clinicians to estimate the overall level of functioning of patients, to monitor disease and rehabilitation management, and to follow patients along the continuum of care over the lifespan.

The objective of this article is to outline the development and validation of a health index for patients with AS based on the categorical profile built on the ICF Core Set for AS as a use case.

## Materials and methods

The development and validation of the health index for patients with AS is a combined effort of the Assessment of SpondyloArthritis International Society (ASAS), the ICF Research Branch of the WHO Collaboration Centre of the Family of International Classifications and the WHO. This effort was coordinated in a meeting held in Zurich, Switzerland on 26 February 2009. The following members from the above outlined partners were involved: A.B., J.B., U.K., D.vdH. (all ASAS), A.C., G.S. (ICF Research Branch) and B.Ü. (WHO). Starting points that were agreed upon during this meeting and issues that have to be resolved at a later stage are described in this article. All aspects of the project will be conducted in conformity with the ethical principles of the Declaration of Helsinki. The appropriate study-related documents will be presented to the corresponding ethics committees for review and approval.

### Methodology

There will be five steps in the development and validation of the health index for patients with AS. A summary of the methodology of the preliminary surveys and of the consensus meeting is shown in Table 1.

#### Methodology of the development

##### *Preparatory phase (Phase I)*

*Selection of instruments.* As a starting point we will create an item pool covering the categories of the

Comprehensive and Brief ICF Core Set for AS [3]. The item pool will be collected from various instruments currently available for use in patients with AS, which focus on symptoms, functioning and composed measures such as questionnaires about quality of life (QoL). Questionnaires used should assesses different aspects of patients with AS. The following instruments have already been agreed upon during the meeting in Zurich: ASQoL Questionnaire; BASFI; Dougados Functional Index; EuroQoL-five dimensions (EQ5D); Functional Assessment of Chronic Illness Therapy-fatigue (FACIT-fatigue); HAQ modified for SpAs (HAQ-S); Health Utilities Index-Mark 3 (HUI-3); Hospital Anxiety and Depression Scale (HADS); Jenkins Sleep Disorder; Nottingham Health Profile; Psoriatic Arthritis QoL (PsAQoL) Questionnaire; RA-specific QoL Instrument; Revised Leeds Disability Questionnaire; Short-form 36; WHO Disability Assessment Schedule II; Work Productivity; and Activity Impairment Questionnaire- General Health [4-19]. These questionnaires may not cover the whole spectrum of the ICF Core Set for AS. In this case, we want to screen additional instruments that are not commonly used in patients with AS, but are already linked to the ICF (e.g. Daily Stress Inventory) [20].

*Linkage of instruments to the ICF.* Some of these instruments have been already linked and the results have been published elsewhere [21-25]. Items out of the following instruments will be linked explicitly for this project: FACIT fatigue, HUI-3, HADS, Jenkins Sleep Disorder and PsAQoL. The items will be linked separately by two trained health professionals (U.K. and A.B.) according to established linking rules [26]. Consensus between the two health professionals has to be reached. To resolve disagreements between them concerning the selected categories, a third person trained in the linking rules will be consulted (A.C.).

*Selection of items.* Items of these instruments will be selected for inclusion if they contain only one concept and if the sentences are short and simple. A preliminary estimate assumes that the item pool will consist of ~150 items.

*Adaptation of items.* Each item will be adapted to the following criteria: the items should be expressed in the first person and in the present tense. As far as possible, potential items will be direct quotations from the original instruments.

**TABLE 1** Summary of the methodology of the preliminary surveys and of the consensus meeting

Phase	Objectives	Methods
I. Preparatory phase	Development of an item pool	Linkage of various assessment tools to ICF
II. First postal survey	Identification of candidate items	Rasch analysis
III. Expert survey	Item selection	Nominal consensus process
IV. Second postal survey	Validation of the draft version; Item reduction	Testing psychometric properties; Rasch analysis
V. Consensus meeting	Creation a final version	Nominal consensus process

## Methodology of validation

### *First postal survey (Phase II)*

The objective of this step is to identify items that differentiate between persons with different levels of functioning. The item pool will be studied in a multicentre, cross-sectional study. The item pool will be split into three different sets, which will be designed for ~300 patients. Within the sets, there will be items that are similar in each set. These repeated items are considered to reflect basic ideas such as pain or impaired mobility, and therefore serve as comparators in the selection process.

To identify candidate items the data set will be subjected to Rasch analysis to assess the overall fit of the model, individual item fit, the response scale used and the presence of differential item functioning (DIF). The fit of the data to the model is a prerequisite to construct measures. DIF assesses the effect of external factors, such as gender or age, on individual item response. It is important to demonstrate that items will not be valued differently by subgroups. Thus, a reduced item pool will be evolved by removing items not fitting the model expectations.

### *Expert survey (Phase III)*

To confirm the content validity of the items selected in Phase II, an expert survey will be performed. The steering committee together with additional invited international experts in the field of spondyloarthritis from most WHO regions will produce a draft version by means of item selection with a nominal consensus technique. In addition, we will decide on the response format on the basis of the results of the first postal survey and the results of the Rasch analysis.

### *Second postal survey (Phase IV)*

The objective of this step is to test scaling properties, reliability, internal consistency and construct validity of the draft version. In a multicentre, cross-sectional study 250 AS patients will receive the draft version together with a demographic questionnaire and additional comparator measures (BASFI and HAQ-S). This package will be sent twice, 2 weeks apart, short enough to minimize change in condition over time, but long enough to minimize memory of responses. Patients will be asked if there has been a change in disease status between the first and the second questionnaire. Data from this stage will be fitted to the Rasch model to confirm that the items form a unidimensional scale, that they represent hierarchical ordering, and to confirm the absence of DIF. After performance of Rasch analysis, a revised version of the health index for patients with AS will be available.

### *Consensus meeting (Phase V)*

The objective of this step is to create a final version of the health index for patients with AS. In a consensus meeting, we will discuss controversial items. Agreement will be achieved by a nominal consensus process.

## Results

The results of this investigation will be the health index for patients with AS.

## Discussion

The increasing recognition of functioning and health has led to an impressive effort in research to develop instruments to measure them. The ICF as a common framework for functioning is being increasingly applied in clinical research and practice. A methodological exploration has shown that it is possible to principally construct clinical measures for patients with AS based on the ICF [27]. However, for the development of health indices basic requirements have to be fulfilled. The members of the WHO Functioning and Disability Reference Group are currently discussing the components: (i) comparability, (ii) metric equivalence and (iii) assessment of QoL.

- (i) Comparability of diseases is requested for ranking different diseases. Therefore, instruments should contain generic items not specific to AS, which will allow comparability between other diseases than AS.
- (ii) Metric equivalence is another prerequisite to be able to compare different instruments. Instruments with metric properties that provide summary scores across a number of ICF categories would provide clinicians with an understanding of patients' overall level of functioning.
- (iii) Assessing QoL is important especially for patients with chronic disabling conditions. In the absence of a cure for the disease, information about QoL provide evidence that therapy really makes people feel better.

As shown in content comparison of health-related QoL (HRQoL) instruments different aspects are scarcely represented within the instruments and many generic tools lack the precision required for effective health care decision making [23]. Based on this, ASAS started to develop an index for measurement of health in patients with AS. The aim of the ASAS Health Index is to cover all concepts of functioning relevant to patients with AS. It has been shown that only 50% of concepts important for functioning of patients with AS are covered by instruments included in the core sets of outcomes for AS or other disease-specific instruments [22]. Especially, aspects of contextual factors (such as support, attitudes of family, medication and financial assets) are not adequately covered by standard measures. But the contribution of contextual factors to functioning is important. Goordeev *et al.* [28] showed that contextual factors explained 37 and 47% of the variance in EQ5D and ASQoL, respectively, which is in line with the finding by Dagfinrud *et al.* [29] that impairment variables (disease activity and pain) explained only one-third of activity and participation restrictions.

As in any scientific endeavour, there are uncertainties that have to be resolved during the development process. As mentioned, inconsistencies in linking ICF items have

been shown in former procedures and are likely to occur within the development process of the health index.

The fact that the concepts, health status, functional status, QoL and HRQoL, are often applied interchangeably in the literature is less irritating when the measures for which those concepts are applied are linked to the ICF. Since HRQoL and ICF represent two different perspectives from which to look at functioning and health, it is expected that both will often be used concurrently.

The goal of developing a health index for a specific condition based on the ICF and its Core sets is very much in line with the broader goal of the WHO to define health indices with the above outlined basic requirements of instruments to ensure the comparability of them within the framework of the ICF.

#### Rheumatology key messages

- Functioning is an important aspect in patients with chronic diseases.
- Valid disease-specific instruments are needed to describe functioning based on the ICF.
- We described the process of instrument selection and adaptation of items.

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