

Which Aspects of Functioning Are Relevant for Patients with Ankylosing Spondylitis: Results of Focus Group Interviews

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ABSTRACT. *Objective.* To investigate whether concepts important to patients with ankylosing spondylitis (AS) are covered by disease-specific self-report health status instruments.

Methods. A qualitative focus group study was conducted with AS patients on problems in daily functioning. Group sessions with 4 to 5 patients each were organized up to the point that no new information was brought forward. Group sessions were tape-recorded, transcribed, and divided into meaning units. Concepts contained in the meaning units were extracted. Self-report instruments on health status specific for AS were identified in a literature search. Using the International Classification of Functioning, Disability and Health (ICF) as a common reference, it was determined whether the concepts identified in the focus groups were covered by the instruments.

Results. Nineteen patients participated in 4 focus group interviews. In total, 332 unique meaning units were linked to 90 second-level ICF categories, of which 25 referred to body functions, 10 to body structures, 35 to activities and participation and 30 to environmental factors. In addition, several concepts relating to personal factors were identified. Only 47 categories were also covered by one of the self-report instruments in AS. Only a minority of concepts addressed by the AS-specific questionnaires were not revealed as relevant in the interviews.

Conclusion. Relevant aspects of the influence of AS are not covered by the classic disease-specific instruments. In particular, the influence of AS on socializing and leisure and the relevance of environmental and personal factors are not adequately assessed by available instruments. (First Release Oct 15 2009; J Rheumatol 2009;36:2501–11; doi:10.3899/jrheum.090156)

Key Indexing Terms:

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Ankylosing spondylitis (AS) is prototypical for the group of inflammatory rheumatologic diseases referred to as the spondyloarthropathies. The prevalence in Caucasians ranges from 0.1 to 1.4 and the disease is diagnosed 3 times more often in men^{1,2}. AS predominantly affects the axial skeleton, with inflammation of the sacroiliac joints and spine as the hallmark of the disease. In addition to the spinal inflammation, peripheral arthritis and enthesitis, uveitis, psoriasis, and inflammatory bowel disease can add to the burden of AS². The impairments and limitations as a consequence of the disease manifestations are reflected in decreased health-related quality of life (HRQOL) including restrictions in participation in life situations³.

Several outcome domains and corresponding instruments have been proposed to evaluate the health status of patients with this disease for trials as well as clinical practice. Following an expert-driven consensus-building process and based on existing evidence from outcome research, the ASAS/OMERACT (ASsessments in Ankylosing Spondylitis/ Outcome Measures in Rheumatology) working group

selected a core set of domains and instruments to assess the influence of AS along intervention trials with physiotherapy or symptom-modifying drugs, with disease-modifying drugs, or for clinical record-keeping⁴. These core sets typically represent a minimal set of domains that are relevant to be assessed in these settings. In a broader perspective of functioning and health, other health domains are likely to be important. In particular, health domains that are relevant for patients have received little attention. It is essential to include the perspective of the patient because relevance for outcomes varies between and among patients and professionals⁵⁻⁸. Data allowing judgment of which aspects of functioning are important to patients with AS remain scarce.

In order to incorporate a comprehensive understanding of functioning into the assessment and management of chronic diseases, the International Classification of Functioning, Disability and Health (ICF) was endorsed by the World Health Assembly in order to enhance a universal approach to understand and classify functioning and health. The ICF framework adheres to the bio-psycho-social model of disease and recognizes that functioning and health result from a complex interplay of the health components body functions, body structures, activities and participation, and contextual factors, such as environmental and personal factors. In addition to the framework, the ICF also offers a universal and hierarchical classification of the elements (called categories) within each component that are necessary to classify functioning^{9,10}. The framework and classification can be applied to all perspectives, including the different types of health professionals, researchers, decision makers, and also the patients.

Our aim was, first, to determine in a qualitative methodology the concepts important for functioning to patients with AS. Second, it was checked whether concepts important for patients are covered by the instruments included in the core set of outcomes of the ASAS/OMERACT working group or other disease-specific instruments. The ICF framework was used as the starting point to perform semistructured interviews with patients. The ICF universal classification was used as the interface when performing the content comparison of concepts important to patients and represented in disease-specific questionnaires.

MATERIALS AND METHODS

A qualitative study was performed based on focus group interviews of patients with AS.

Participants, sample size, and number of focus groups. A list of 187 patients with AS according to the New York criteria¹¹ and registered in the rheumatology outpatient clinic of the Maastricht University Medical Center in 2006 was circulated among the rheumatologists. The treating rheumatologist confirmed whether his/her patient fulfilled modified New York criteria for AS and identified 5 patients he/she considered able to contribute in the focus group interviews. From the resulting list of 30 patients, preliminary groups of 5 patients were composed, assuring in each group diversity based on characteristics such as sex, age, disease duration, education level [primary school, secondary school, vocational (lower, middle or higher) or

university], work status (employed, work-disabled, home worker), profession (mainly manual opposed to mental workload), and AS related comorbidity. A diverse range of participants was needed to obtain the required level of rich and meaningful data¹². The patients were contacted as a group and informed about the study procedures. Any patient not willing to participate was replaced by a similar patient from another group and groups were reconsidered again for diversity before patients were contacted for a new group. The number of new focus groups conducted was determined by saturation, i.e., the point at which an investigator obtained sufficient information from the field¹³. Saturation was defined as the point during data collection when the new focus groups revealed no additional concepts that were not obtained before.

Interviews. All focus groups were chaired by the same moderator (AB) with one assistant responsible for observation of the group and recording of data. Each focus group meeting was tape-recorded and transcribed verbatim by the observer. A short introduction of the purpose of the study was given in lay terms to all patients at the beginning of each focus group. Five open-ended questions were formulated around functioning in daily life based on the components of the ICF model. Patients were asked (1) which AS-related problems in the functioning of their body they were experiencing; (2) which body structures were involved; (3) which limitations of activities and restrictions in participation were significant to them; and (4) which environmental factors and (5) which personal factors were barriers or facilitators.

Qualitative data analysis. Qualitative data analysis followed the method of "meaning condensation"¹⁴: in the first step, the transcribed interviews were read by the first author for an overview of the collected data. Second, the data were divided into meaning units. A meaning unit was defined as a specific unit of text, either a few words or a few sentences with a common theme. Third, the concepts contained in the meaning units were identified. A meaning unit could contain more than one concept. A concept was defined as a meaningful entity distinct from other concepts. An example is the meaning unit, "due to the pain in my neck, I avoid driving long with the car," in which we identified the concepts "pain in the neck" and "driving a car." Meaning condensation was performed by AB, and to ensure validity the result was checked by a second author (MvB).

Instruments. Current disease-specific self-report instruments assessing functioning in AS were identified through a Medline search (1967-2006). Part of the literature search had been done in a previous study on content comparison of self-reported instruments on physical function in patients with AS¹⁵, but concentrated on "function," "physical function," or "functioning." An additional search was done with the key words "health status" or "health state" or "quality of life" in AS. Descriptive, evaluative, and psychometric studies in AS were selected. Case reports, economic evaluations, primary prevention studies, and reviews were excluded. Articles were selected that describe the use of self-report instruments, were published in a peer-reviewed journal, existed in an English-language version, and were feasible in a clinical setting. Finally, the ASAS/OMERACT publication on the Core Outcome Set for AS^{4,15} was checked for additional outcome instruments. Self-report disease-specific instruments were chosen because this study focused on the perspective of patients, which is likely specific to a disease.

Procedure to verify whether a concept was covered by an instrument. For every concept obtained from the patient focus groups, we examined whether it was covered by the instruments. To perform this step, the ICF was used as external reference. Each concept obtained in the focus group was linked to the appropriate ICF category according to published linking rules^{16,17} that have been used in qualitative studies^{8,18,19}. According to these linking rules, every concept is linked to the most precise ICF category. If a concept was not contained in the ICF classification, it was assigned the category "not covered" (NC), such as the concept "exercising is important" in our study. In the hierarchical system of the ICF, there are categories of the second, third, and fourth levels. Third and fourth level categories are specifications (if applicable) of the second level categories. If several sec-

ond level categories cover a common theme, they are grouped into ICF chapters (first level of the classification) or subchapters. The personal factors are not yet classified in ICF categories and for personal factors the concepts identified in the meaning units are provided without linkage to an ICF category.

In order to determine whether a concept from the interviews was covered by any of the available instruments, the linked categories from the focus groups were matched to the linked categories in the instruments. Linking of concepts in the questionnaires had been done for the majority of selected instruments [Bath AS Functional Index (BASFI), Dougados Functional Index (DFI), Health Assessment Questionnaire-Spondyloarthropathies (HAQ-S), revised Leeds Disability Questionnaire (RLDQ)]¹⁵ and was done for this study for the remaining questionnaires [AS Quality of life instrument (ASQoL), patient globals]. Thus, it was possible to distinguish the concepts covered and not covered in the questionnaires. An example is the concept “difficulty reaching” from the focus groups, which was linked to the ICF category d4452 and was also contained in items from the BASFI, HAQ-S, and RLDQ.

In order to ensure the validity of linking to the ICF of the focus interviews, a sample of 50% of concepts linked by MvB was independently linked by a second trained ICF specialist. The sample was not chosen randomly but was based on concepts considered difficult to link by the first linker. In case of disagreement this was discussed with a third trained ICF specialist (AC) and leader of the project (AB), and a decision was made based on consensus.

RESULTS

Participants and focus groups. After the fourth focus group, interview saturation was reached. Characteristics of the 19 participating patients are presented in Table 1. Fourteen (74%) patients were male; mean age was 54 years (SD 11.5, range 31–69), mean disease duration 18.7 years (SD 10.0, range 4–36). Ten patients (53%) had achieved a higher vocational or university degree. Fourteen patients were of working age, of which 3 (21%) were work-disabled because

of AS. Of 10 patients with paid employment, 3 (30%) had a mainly manual profession. Extraspinal articular disease was present in 6 (32%) and AS-related comorbidity (inflammatory bowel disease, psoriasis, or uveitis) was present in 6 (32%). Twelve patients (63%) were treated with a tumor necrosis factor- α (TNF- α) inhibitor.

Literature search and instruments. Four questionnaires were identified in the previous literature search on physical function in AS: the BASFI²⁰, the DFI²¹, the RLDQ²², and the HAQ-S²³. Three additional self-report instruments to assess health were derived from the ASAS/OMERACT Core Set of outcome measures²⁴; the Bath AS Disease Activity Index (BASDAI)²⁵, the patient global assessment of disease activity, and the patient global pain assessment (pain at night and pain in the spine). Further search of the literature revealed the ASQoL²⁶.

Categories derived from the focus groups. In the transcripts, 322 meaning units were identified and overall a total of 519 concepts. A sample of 260 concepts were selected for quality control and these included all the concepts considered “difficult to link.” Forty-two of these 220 concepts (16%) were selected by the second (blinded) linker for further discussion by the project group. The majority of these concepts (n = 38) were already identified as difficult to link by the first observer, and for the 4 other concepts there was a discrepancy in the level of specification of the category. In all cases, a consensus solution was found in the group discussion.

The concepts were linked to a total of 148 ICF categories, representing 90 categories of the second level categories. Thirty-nine categories (25 at the second level)

Table 1. Characteristics of the 19 patients participating in the focus groups.

| Patient | Sex, Age, yrs | Disease Duration, yrs (diagnosis) | Highest Education | Employment | Peripheral Arthritis | AS-related Comorbidity | TNF Inhibitor |
|---------|---------------|-----------------------------------|-------------------|---------------------------------------|----------------------|------------------------|---------------|
| 1 | F 60 | 11 | Primary school | Housewife | Yes | No | No |
| 2 | M 38 | 17 | Middle vocational | Fulltime self-employed (salesman) | No | No | Yes |
| 3 | F 31 | 8 | Higher vocational | Fulltime (waitress) | No | Yes | Yes |
| 4 | M 36 | 4 | Higher vocational | Fulltime (clerk) | No | No | Yes |
| 5 | M 54 | 34 | University | Work disabled (self-employed dentist) | No | No | Yes |
| 6 | M 52 | 8 | Middle vocational | Fulltime self-employed (manager) | No | No | No |
| 7 | M 68 | 30 | Higher vocational | Retired (miner) | Yes | Yes | No |
| 8 | M 65 | 36 | Higher vocational | Retired (manager) | No | Yes | Yes |
| 9 | F 52 | 12 | Higher vocational | Part-time self-employed (beautician) | Yes | No | Yes |
| 10 | F 48 | 15 | Secondary school | Part-time employed (clerk) | No | Yes | Yes |
| 11 | M 56 | 18 | Middle vocational | Self-employed (café owner) | No | No | Yes |
| 12 | M 64 | 32 | Higher vocational | Retired age (chemist) | No | No | No |
| 13 | F 69 | 4 | Higher vocational | Housewife | Yes | No | Yes |
| 14 | M 68 | 18 | Lower vocational | Work disabled/retired | No | No | Yes |
| 15 | M 49 | 18 | Lower vocational | Fulltime employed (farmer) | No | Yes | Yes |
| 16 | M 49 | 16 | Lower vocational | Work disabled | Yes | No | No |
| 17 | M 59 | 25 | Middle vocational | Self-employed (farmer) | Yes | No | No |
| 18 | M 69 | 20 | Higher vocational | Early retired because of AS (banker) | No | No | No |
| 19 | M 46 | 30 | University | Fulltime employed (medical doctor) | No | Yes | Yes |

TNF: tumor necrosis factor therapy.

belonged to body functions (Table 3), 18 categories (10 at the second level) to body structures (Table 4), 66 categories (35 at the second level) to activities and participation (Table 5), and 25 categories (20 at the second level) to environmental factors (Table 6).

Several concepts were related to the ICF component “personal factors,” which is not yet further specified in categories. In this component concepts related to accepting the disease and adapting to the disease and its wide range of consequences was a recurring theme in all groups. Overall, accepting and adapting were considered positive and helpful strategies. However, patients who currently experienced the beneficial effects of TNF inhibitors also recognized that accepting might have a negative consequence, since it could make you ignore the severity of disease and underestimate the possible benefits of more intensive treatments. Accepting and adapting are strategies patients develop when confronted with the need to adjust to the consequences of disease and specifically in these interviews included a series of positive aspects such as creating new purposes in life, adopting a healthier lifestyle, and creating time to do exercises and sports. Other strategies were efforts to keep the disease at a distance, taking regular rest, remaining optimistic, acquiring knowledge and self management skills, educating people in the direct environment about the disease, and concealing the disease from others. As well, trust in health professionals was repeatedly mentioned as important. It was also noted that those with better cognition and

those with greater financial resources had a position of advantage to deal with the consequences of the disease.

Some other concepts contained in the meaning units could not be linked to any of the ICF components (Table 2). These concepts differed from concepts that can unambiguously be linked to an ICF category because (1) they are really not covered by a specific ICF category (for example, posture) or (2) refers to a positive attitude to deal with disease (exercising is important).

Comparison with the instruments. Tables 4 to 6 present for each component of the ICF classification whether concepts identified in the interviews were also represented in each of the self-report instruments that assess disease-specific health status in AS. ICF categories are given in italic when the same category is represented by one of the questionnaires. When grouped at the second level, only 6 of 25 (24%) of body functions, 18 of 35 (51%) of activities and participation, and 6 of 20 (30%) of environmental factors were covered by self-report instruments. Only a limited number of second level categories were represented in the questionnaires and not mentioned in the interviews. The concepts “do you need help for eating” (d450: eating) from the HAQ-S, “can you cough or sneeze” (b450: additional respiratory functions) from the DFI, “can you turn in bed” and “can you roll over in bed” (d4201: transferring when lying) from the DFI and RLDQ, respectively, “AS limits places I can go” (d460: moving around in different locations) and “I am unable to join in activities with my

Table 2. Concepts not covered or no agreement on whether they were covered or not specified.

| Concept | Best Option of Specification | Discussion |
|--|------------------------------|--|
| The change in my body posture | nc-s, nd | Change in body posture is the result of changes in different parts of joints and bones (vertebral column and possibly hips and knees) |
| Delay in diagnosis had an important influence on my life | nc-e | Points to health service delivery and medical knowledge and its implementation across professionals |
| Worrying about hereditary aspects | nc-a&p | “Worry” refers to an emotional function (b 152) and the total concept to d760: family relationships (creating and maintaining kinship relations) |
| Losing time because of disease | nc-a&p | Relates to chapter d2: general tasks and demands |
| Reduce the speed of activities | nc-b, nd-a&p, nc-pf | Relates to energy (b130), to chapter for general tasks and demands, but also the coping strategy (personal factor) |
| I need to take rest during the day | nc-b; nc-pf | Relates to energy level (b130) but also to a coping strategy (personal factor) |
| Resting provides new energy | nc-a&p; nc-pf | Relates to energy level but also to a coping strategy (formulated in a positive way) |
| Acquiring information is important | nc-pf; nc-e | Relates most closely to a coping strategy (personal factor). Relates also to health services (chapter e5) |
| Providing information to others is important | nc-pf; nc-e; nc-a&p | Relates most closely to a coping strategy (personal factor). Relates also to health services (Chapter e5) and relationships (Chapter d7) |
| Try to hide my disease from others | nc-pf; nc-a&p | Relates most closely to a coping strategy (personal factor). Points also to relationships (Chapter d7) |
| Exercising is important | nc-pf; nc-a&p | Relates most closely to a coping strategy (personal factor). Points also to health services (e580) |
| AS forces me to have a healthy lifestyle | nc-pf; nc-e | Relates most closely to a coping strategy (personal factor). Points also to health services (e580) |
| Feel that one misses a lot | nc-b; nc | Relates to emotional function (b152). Also points to several other concepts (energy, participation, etc.) |

nc-s: not covered, structural factor; nc-e: not covered, environmental factor; nc-b: not covered, body function; nc-a&p: not covered, activity and participation. nc-pf: not covered: personal factor; nd: not defined.

Table 3. Categories in the component “Body Functions” identified after linking meaningful units reported during the focus interviews and their coverage (categories shown in *italic*) by AS-specific health status questionnaires.

| ICF Chapters or Subchapters | Concepts | ICF Categories | DFI | BASFI | RLDQ | HAQ-S | ASQoL | BASDAI | Patient Global for Pain |
|---|--|--|------------|------------|------------|-------|-------|--------|-------------------------|
| b110–b139: Global mental functions | Optimism | b1265 | | | | | | | |
| | Energy | b1300, b1302 | | | | | X | X | |
| | Sleep | <i>b134</i> , b1342 | X | | X | | X | | |
| b140–b189: Specific mental functions | Sustaining attention | b1400 | | | | | | | |
| | Emotions | <i>b152</i> | | | | | X | | |
| | Judgment and problem-solving | b1645, b1646 | | | | | | | |
| b210–b229: Seeing and related functions | Seeing and sensations; eyes | b210, 2153, b220 | | | | | | | |
| b230–b249: Hearing functions | Hearing sounds too sharp | b230 | | | | | | | |
| b280–b289: Pain | Pain in neck, back, chest, limb, joints, abdomen | <i>b28010</i> , b28011, b28012, b28013, b28014, b28015, b28016 | | X (= b289) | | X | X | X | X |
| b430–b439: Functions of the hematological and immunological systems | Vulnerable to infections and allergies | b43501, b4351 | | | | | | | |
| b440–b449: Functions of the respiratory system | Respiration | <i>b4402</i> | X (& b450) | | X (= b450) | | | | |
| | Exercise tolerance, fatigability | b455, b4552 | | | | | | | |
| b510–b539: Functions related to digestive system | Diarrhea, weight loss, abdominal cramps | b525, b5256, b530, b5352 | | | | | | | |
| b540–b559: Functions related to metabolism and endocrine system | Feeling too cold or too warm | b550, b5500 | | | | | | | |
| b640–b680: Genital and reproductive functions | Sexual interest and performance | b640 | | | | | | | |
| b710–b729: Functions of joints and bones | Mobility of joints and bones | b7100, b7101, b720 | | | | | | | |
| b750–b789: Movement functions | Muscle cramps and muscle weakness | b755, <i>b760</i> | | X | | | | | |
| | Gait pattern | b770 | | | | | | | |
| | Stiffness | <i>b780</i> | | | | X | | X | |

ICF: International Classification of Functioning, Disability and Health; DFI: Dougados Functional Index; BASFI: Bath AS Functional Index; RLDQ: revised Leeds Disability Questionnaire; HAQ-S: Health Assessment Questionnaire-Spondyloarthropathies; ASQoL: AS Quality of life instrument; BASDAI: Bath AS Disability Index.

Table 4. Categories of the component “Body Structures” identified after linking the meaningful units reported during the focus interviews.

| ICF Chapter | Concepts | ICF Categories |
|--|---------------|------------------------------|
| s2: Structures of the eye, ear, and related structures | Eyeball | s220 |
| s5: Structures of digestive, metabolic, and endocrine system | Intestine | |
| s7: Structures related to movement | Neck | s710, s7103 |
| | Upper limb | s720, s7302, s73021 |
| | Pelvic region | s740, s7401 |
| | Lower limb | s7500, s75011, s7502, s75021 |
| | Trunk | s760, s7600, s76002, s7701 |
| s8: Skin and related structures | Skin | s810 |

friends/family” (d7500: informal relationships), both from the ASQoL, were not specifically identified during the inter-

views. In general it was striking that the HAQ-S contained several categories not identified in the interviews and that

Table 5. Categories in the component “Activities and Participation” identified after linking meaningful units reported during the focus interviews and their coverage (categories shown in *italics*) by AS-specific health status questionnaires.

| ICF Chapters or Subchapters | Concepts linked to ICF Categories | ICF Categories Linked to the Concepts of Previous Column | DFI | BASFI | RLDQ | HAQ-S | ASQoL | BASDAI | Patient Global for Pain |
|--|--|--|----------------|----------------|-----------------------|------------------------|----------------|--------|-------------------------|
| d130–d159: Basic learning | Acquiring skills | d134 | | | | | | | |
| d160–d179: Applying knowledge | Reading, solving problems, making decisions | d166, d175, d177 | | | | | | | |
| d2: General tasks and demands | Carrying out routine | d23001 | | | | | | | |
| | Managing one’s activity level, handling stress | d2303, d2401 | | | | | | | |
| d410–429: Changing and maintaining body position | Sitting, standing, bending, standing up, prolonged standing or sitting | <i>d4100, d4101, d4103, d4104, d4105, d4106, d4150, d4153, d4154</i> | X (+ d4201) | X | X (+ d4109, d4201) | X | | | |
| d430–d449: Carrying, moving and handling objects | Lifting, pushing, reaching, fine hand use | <i>d4300, d4350, d440, d445, d4452, d4453</i> | X | X | X | X | | | |
| d450–d469: Walking and moving | Walking, running, swimming | <i>d450, d4501, d4552, d4554</i> | X (= d4551) | X (= d4551) | X | X (= d4502 & d4551) | X (= d460) | | |
| d470–d489: Moving around using transportation | Using private and public transport | d470, d4701, d4702 | | | | | | | |
| | Driving a car | d475, d4750, d4751 | | | | X | | | |
| d498–d499 | Mobility other, unspecified | <i>d498</i> | X | | X | X | | | |
| d5: self-care | Washing oneself | <i>d510</i> | | | | | | | |
| | Caring for body parts (washing hair, caring for toenails) | d5202, d5204 | | | X | X | X | | |
| | Toileting, dressing, drinking | <i>d530, d540, d5401, d5402, d5403, d560</i> | X (= d5400) | X | X (+ d5404) | X | X | | |
| | Looking after one’s health | <i>d570</i> | | X (= d5709) | | | | | |
| 610–d629: Acquisition of necessities | Shopping | <i>d6200</i> | | | | X | | | |
| d630–d649: Household tasks | Doing housework, cleaning | <i>d640, d6402</i> | X | X | | X | X (= d6409) | | |
| d650–d669: Caring for household objects and assisting others | Maintaining objects, furnishing, taking care of animals | <i>d650, d6501, d6505, d6506</i> | | X (= d6509) | | X (= d6509) | | | |
| | Assisting others | d6601 | | | | | | | |
| d710–d729: General interpersonal interactions | Appreciation in relationships | d7101, d7102, d7104 | | | | | X (= d710) | | |
| d730–d779: Particular interpersonal relationships | Relationships with peers, partner, children, grandchildren | d7600, d7601, d7603 | | | | | X (+ d7500) | | |
| d840–d895: Work and employment | Seeking and maintaining employment | d8450, d850, d8501 | X | X | | | | | |
| d9: Community, social, and civic life | Recreation and hobby | d9100, d920, d9200, d9201, d9204 | | X (= d9209) | | | | | |
| | Socializing | d9205 | | | | | | | |

For abbreviations, see Table 3.

refer to impairments in the function of the hands including “fine hand uses,” “twisting,” and “picking up.” A third level specification that was included in the BASFI, DFI, and

HAQ-S but that was not raised in the interviews was related to “climbing stairs” (d4551: climbing).

It is notable that in the ASQol two concepts were identi-

Table 6. Categories in the component “Environmental Factor” identified after linking meaningful units reported during the focus interviews and their coverage (categories shown in *italic*) by AS-specific health status questionnaires.

| ICF Chapter | Concepts | ICF Categories | DFI | BASFI | RLDQ | HAQ-S | ASQoL | BASDAI | Patient Global Pain |
|---|---|---|-----|--------|------|----------|-------|--------|---------------------|
| e1: Products and technologies | Drugs | <i>e1101</i> | | | | X | | | |
| | Aids and adaptations (home, work, transportation) | <i>e1151, e1201, e1351</i> | X | X | | X | | | |
| | Accessibility of buildings | <i>e150, e155</i> | | | | (+ e135) | | | |
| | Financial assets | <i>e165, e1650</i> | | | | | | | |
| e2: Natural and human made changes to the environment | Cold and humidity | <i>e225, e2250, e2251</i> | | | | | | | |
| e3: Support and relationships | From family, friends, colleagues, employer, health professionals, and society | <i>(e3), e310, e320, e325, e330, e335, e340, e355</i> | X | X (e3) | | X | | | |
| e4: Attitude | From family, friends, colleagues, employer, health professionals, and society | <i>e410, e425, e460</i> | | | | | | | |
| e5: Services, systems, and policies | Health services, employment systems, and services | <i>e5800, e5801, e590, e5901</i> | | | | | | | |

For abbreviations, see Table 3.

fied that could not be linked to any ICG category (not covered; NC) and these are not included in any of the Tables 3 to 6 — “I have to keep stopping what I’m doing to rest,” and “I feel I miss out a lot.” Both concepts were also mentioned by the patients during the interviews.

DISCUSSION

This focus group study is the first qualitative attempt to comprehensively determine the perspective of patients with AS. The present view on outcome is largely driven by evidence-based knowledge, which usually represents the perspective of clinical researchers. Increasingly, the importance of the patient perspective in health outcomes research is recognized, and differs in several aspects from the perspective of clinicians and researchers^{7,27}. In this quantitative research, patients came up with a large number of concepts, supporting the relevance of the ICF framework for the patients’ perspective. We also found that some concepts important to people with AS are not adequately covered by standard self-report instruments that are developed to assess health in AS. The majority of disease-specific instruments for outcome assessment in AS have definitely proven their value, but we should realize they concentrate on limited aspects of functioning and health, such as physical functioning in the BASFI, DFI, HAQ-S, and RLDQ, or pain, stiffness, and fatigue in the BASDAI and patient globals. The ASQoL instrument has the broadest view on functioning and health, but still misses some areas that were relevant to patients, such as limitations in sustaining attention or solving problems and restrictions in recreation or socializing. Patients repeatedly highlighted the role of personal factors to adjust to the disease, and optimism, accepting, adapting (i.e., finding creative solutions), gaining insight into the dis-

ease, and trust in health professionals were frequently mentioned. These areas are not considered in usual questionnaires. Although it was primarily our aim to evaluate whether aspects relevant for functioning were covered by disease-specific instruments, we also compared the categories from the focus interview by other health-related quality of life instruments (Appendix). Including these questionnaires would slightly raise the coverage of patient perspective with regard to participation issues, but at the expense of limitations in body functions and activities specific for the disease.

Although qualitative research has evident advantages, there are also limitations. When we prepared the focus group interviews, care was taken to cover the entire spectrum of patients with AS, by selecting patients with a broad range of demographic and disease characteristics. Some concepts that were revealed were the individual experience of an individual patient. For example, “hearing sounds too sharp” was mentioned by one patient, who experienced this phenomenon systematically when experiencing fatigue in episodes of flare. Other concepts that were mentioned were clearly linked to typical AS-related comorbidities such as difficulties because of bowel problems, visual functions, and structures of the skin in patients with inflammatory bowel disease, uveitis, or psoriasis, respectively. It is recognized that these comorbidities occur only in subgroups of patients, but these should not be neglected as they can add considerably to the impact of the disease on functioning. Also, all patients were recruited from one center, which would challenge the generalizability and especially the transferability of findings across cultures. Specifically for participation, differences of environment and personal factors across cultures can be expected. In future research on

patients' perspectives, transferability across cultures should receive more attention. An ICF classification that is specific for a disease does not imply that it is necessary to assess all

categories in every patient. Therefore, in addition to the qualitative study, more quantitative data are needed. Also, new techniques such as item response theory (IRT) and

APPENDIX. Categories identified during the interviews and coverage by generic health-related quality of life instruments.
Appendix Ia. Categories of the components *Body Functions* identified in the focus group interviews and coverage by generic health-related quality of life instruments.

| ICF chapters or subchapters | Concepts identified in the interviews | ICF categories linked to the concepts | SF-36 | EQ-5D | NHP | WHO-DASII |
|--|--|--|-------|-------|-----------|-----------|
| b110-b139: global mental functions | Optimism | b1265 | | | | |
| | Energy | b1300, b1302 | X | | X | |
| | Sleep | b134, b1342 | | | X | |
| b140-b189: specific mental functions | Sustaining attention | b1400 | | | | x |
| | Emotions | b152 | X | X | X | X |
| | Judgement and problem solving | b1645, b1646 | | | | |
| b210-b229: seeing and related functions | Seeing and sensations eyes | b210, 2153, b220 | | | | |
| b230-b249: hearing functions | Hearing sounds too sharp | b230 | | | | |
| b280-b289: pain | Pain in neck, back, chest, limb, joints, abdomen | b28010, b28011, b28012, b28013, b28014, b28015, b28016 | X | X | X | |
| b430-b439: functions of the haematological and immunological systems | Vulnerable to infections and allergies | b43501, b4351 | | | | |
| b440-b449: functions of the respiratory system | Respiration | b4402 | | | X (b450*) | |
| | Exercise tolerance, fatiguability | b455, b4552 | | | | |
| b510-b539 : functions related to the digestive sytem | Diarrhea, weight loss, abdominal cramps | b525, b5256, b530, b5352 | | | | |
| b540-b559: functions related to metabolism and the endocrine system | Feeling too cold or too warm | b550, b5500 | | | | |
| b640-b680: genital and reproductive functions | Sexual interest and performance | b640 | | | | |
| b710-b729: functions of the joints and bones | Mobility of joints and bones | b7100, b7101, b720 | | | | |
| b750-b789: movement functions | Muscle cramps and muscle weakness | b755, b760 | | | | |
| | Gait pattern | b770 | | | | |
| | Stiffness | b780 | | | | |

SF-36: Short Form 36 items; EQ-5D: EuroQol 5 dimensions; NHP: Nottingham Health Profile; WHO-DAS: WHO Disability Assessment Schedule; *b450; additional respiratory functions.

Appendix Ib. Categories of the components *Activities and Participation* identified after linking the meaningful units reported during the focus interviews and inclusion in other self-reported generic health related quality of life instruments.

| ICF chapter or subchapters | Concepts identified in the interviews | ICF categories linked to the concepts | SF-36 | EQ-5D | NHP | WHO-DASII |
|--|--|---|-------|---------------|-----|-----------|
| d130-d159: basic learning | Acquiring skills | d134 | | | | X |
| d160-d179: applying knowledge | Reading, solving problems, making decisions | d166, d175, d177 | | | | X |
| d2: general tasks and demands | Carrying out routine, | d23001 | X | X | | X |
| | managing one's activity level, handling stress | d2303, d2401 | | | | |
| d410-429: changing and maintaining body position | Sitting, standing, bending, standing up, prolonged standing or sitting | d4100, d4101, d4103, d4104, d4105, d4106, d4150, d4153, d4154 | X | d4 (mobility) | x | |
| d430-d449: carrying, moving and handling objects | Lifting, pushing, reaching, fine hand use | d4300, d4350, d440, d445, d4452, d4453 | X | | X | |

| | | | | | | |
|--|--|---------------------------------------|---|-----------------|----|---|
| d450-d469: walking and moving | Walking, running, swimming | d450, d4501, d4552, d4554 | X | X | X | |
| d470-d489: moving around using transportation | Using private and public transport, | d470, d4701, d4702 | | | | |
| | driving a car | d475, d4750, d4751 | | | | |
| d498 – d499 | Mobility other-unspecified | d498 | | | | |
| d5: self-care | Washing oneself | d510 | | x | | |
| | Caring for body parts (washing hair, caring for toenails) | d5202, d5204, | | | | |
| | Toileting, dressing, drinking | d530, d540, d5401, d5402, d5403, d560 | X | | X | X |
| | Looking after one's health | d570 | | | | |
| d610-d629: acquisition of necessities | Shopping | d6200 | | | | |
| d630-d649: household tasks | Doing housework, cleaning | d640, d6402 | X | X (unspecified) | X | X |
| d650-d669: caring for household objects and assisting others | Maintaining objects, furnishing, taking care of animals | d650, d6501, d6505, d6506, | | | X | |
| | Assisting others | d6601 | | | | |
| d710-d729: general interpersonal interactions | Appreciation in relationships | d7101, d7102, d7104 | | | | |
| d730-d779: particular interpersonal relationships | Relationships with peers, partner, children, grandchildren | d7600, d7601, d7603 | | | d7 | X |
| d840-d895: work and employment | Seeking and maintaining employment | d8450, d850, d8501 | X | X (unspecified) | X | X |
| d9: community social and civic life | Recreation and hobby | d9100, d920, d9200, d9201, d9204 | X | X (unspecified) | X | X |
| | Socializing | d9205 | X | | | |

SF-36: Short Form 36 items; EQ-5D: EuroQol 5 dimensions; NHP: Nottingham Health Profile; WHO-DAS: WHO disability Assessment Schedule; *b450; additional respiratory functions.

Appendix Ic. Categories of the components *environmental factor* identified after linking the meaningful units reported during the focus interviews and inclusion in other self-reported generic health related quality of life instruments.

| ICF chapter | Concepts identified in the interviews | ICF categories linked to the concepts | SF-36 | EQ-5D | NHP | WHO-DASII |
|--|--|--|-------|-------|-----|-----------|
| e1: products and technologies | Drugs | e1101 | | | X | |
| | Aids and adaptations (home, work, transportation) | e1151, e1201, e1351 | | | | |
| | Accessibility of buildings | e150, e155 | | | | |
| | financial assets | e165, e1650 | | | | |
| e2 natural and human made changes to the environment | Cold and humidity | e225, e2250, e2251 | | | | |
| e3 support and relationships | From family, friends, colleagues, employer, health professionals and society | (e3), e310, e320, e325, e330, e335, e340, e355 | | | X | |
| e4 attitude | From family, friends, colleagues, employer, health professionals and society | e410, e425, e460 | | | | X |
| e5 Services, systems and policies | Health services, employment systems and services | e5800, e5801, e590, e5901 | | | | |

SF-36: Short Form 36 items; EQ-5D: EuroQol 5 dimensions; NHP: Nottingham Health Profile; WHO-DAS: WHO Disability Assessment Schedule; *b450; additional respiratory functions.

computer adaptive testing (CAT) can help to identify categories that are redundant, give no additional information, or

are only relevant in some patient groups, and can help to identify a more limited number of ICF categories relevant

for particular patients²⁸. Preliminary data on IRT in interviews with the preliminary ICF checklist in AS showed that only 3 items showed a negative misfit — d560: drinking, d6200: shopping, and d830: higher education. This means that these categories give no additional information (local dependency or redundancy) when measuring the level of difficulty on the measure “functioning”²⁸. However, it does not mean these items cannot have importance in individual patients and does not exclude that remaining items are inter-related or dependent on each other. Graphically represented statistics offer possibilities to see how items/categories are related. It is clear that many concepts, i.e., those referring to participation and environmental and personal factors, are not unique for patients with AS. Comparison in the future of the concepts/categories that are common across musculoskeletal diseases can allow development of common instruments to assess these aspects and common strategies to improve them. This kind of approach could at the same time explore possible differences across cultures.

It must be emphasized that this qualitative research is intended to identify “what is important” for patients, and does not aim to create a new instrument. The ICF classification was found to be fairly complete in covering the concepts raised by the patients. The notable concepts that could not be linked to the ICF were the impairments as a consequence of postural changes and the influence of late diagnosis on the course of the disease, which could not be covered at that level of specification^{8,19,29}.

The findings of our study are not only of interest for clinical researchers that develop and improve instruments to assess functioning, but also for clinicians when assessing patients in clinical practice: it is important to recognize and discuss all limitations and restrictions experienced by the individual patient. Physicians likely tend to concentrate on their own perspective and ignore problems important to patients, particularly the effects of AS on socializing and leisure and the relevance of environmental and personal factors. Therefore, the list of ICF categories important for patients might help to broaden the scope of problems in functioning with AS for researchers and for physicians. The main challenge is to determine which aspects revealed in this study and not covered in AS-specific or general-use HRQOL instruments — including which subgroups of patients (sex-specific, working age, stage of disease) and which settings (clinical practice, educational programs, observational studies, non-drug intervention trials) — should now receive attention.

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REFERENCES

1. Khan MA. Ankylosing spondylitis: clinical features. London: Mosby; 1998.
2. Khan MA. Update on spondyloarthropathies. *Ann Intern Med* 2002;136:896-907.
3. Boonen A, de Vet H, van der Heijde D, van der Linden S. Work status and its determinants among patients with ankylosing spondylitis. A systematic literature review. *J Rheumatol* 2001;28:1056-62.
4. van der Heijde D, Calin A, Dougados M, Khan MA, van der Linden S, Bellamy N. Selection of instruments in the core set for DC-ART, SMARD, physical therapy, and clinical record keeping in ankylosing spondylitis. Progress report of the ASAS Working Group. Assessments in Ankylosing Spondylitis. *J Rheumatol* 1999;26:951-4.
5. Carr A, Hewlett S, Hughes R, Mitchell H, Ryan S, Carr M, et al. Rheumatology outcomes: the patient's perspective. *J Rheumatol* 2003;30:880-3.
6. Hewlett S, Wit M, Richards P, Quest E, Hughes R, Heiberg T, et al. Patients and professionals as research partners: challenges, practicalities, and benefits. *Arthritis Rheum* 2006;55:676-80.
7. Hewlett SA. Patients and clinicians have different perspectives on outcomes in arthritis. *J Rheumatol* 2003;30:877-9.
8. Stamm TA, Cieza A, Coenen M, Machold KP, Nell VP, Smolen JS, et al. Validating the International Classification of Functioning, Disability and Health Comprehensive Core Set for Rheumatoid Arthritis from the patient perspective: a qualitative study. *Arthritis Rheum* 2005;53:431-9.
9. Stucki G. International Classification of Functioning, Disability, and Health (ICF): a promising framework and classification for rehabilitation medicine. *Am J Phys Med Rehabil* 2005;84:733-40.
10. Boonen A, Rasker JJ, Stucki G. The International Classification for Functioning, Disability and Health: A challenge and a need for rheumatology. *Clin Rheumatol* 2007;26:1803-08.
11. van der Linden S, Valkenburg HA, Cats A. Evaluation of diagnostic criteria for ankylosing spondylitis. A proposal for modification of the New York Criteria. *Arthritis Rheum* 1984;27:361-8.
12. Jones K. The turn to narrative knowing of persons: one method explored. *Nursing Times Res* 2000;8:60-71.
13. Depoy E, Gitlin LN. Introduction to research. St Louis: Mosby; 1998.
14. Kvale S. Interviews — An introduction to qualitative research interviewing. Thousand Oaks, CA: Sage; 1996.
15. Sigl T, Cieza A, van der Heijde D, Stucki G. ICF based comparison of disease specific instruments measuring physical functional ability in ankylosing spondylitis. *Ann Rheum Dis* 2005;64:1576-81.
16. Cieza A, Brockow T, Ewert T, Amman E, Kollerits B, Chatterji S, et al. Linking health-status measurements to the International Classification of Functioning, Disability and Health. *J Rehabil Med* 2002;34:205-10.
17. Cieza A, Geyh S, Chatterji S, Kostanjsek N, Ustun B, Stucki G. ICF linking rules: an update based on lessons learned. *J Rehabil Med* 2005;37:212-8.
18. Coenen M, Cieza A, Stamm TA, Amann E, Kollerits B, Stucki G. Validation of the International Classification of Functioning, Disability and Health (ICF) Core Set for rheumatoid arthritis from the patient perspective using focus groups. *Arthritis Res Ther* 2006;8:R84.
19. Stamm TA, Nell V, Mathis M, Coenen M, Aletaha D, Cieza A, et al. Concepts important to patients with psoriatic arthritis are not adequately covered by standard measures of functioning. *Arthritis Rheum* 2007;57:487-94.
20. Calin A, Garrett S, Whitelock H, Kennedy LG, O'Hea J, Mallorie P,

- et al. A new approach to defining functional ability in ankylosing spondylitis: the development of the Bath Ankylosing Spondylitis Functional Index. *J Rheumatol* 1994;21:2281-5.
21. Dougados M, Gueguen A, Nakache JP, Nguyen M, Mery C, Amor B. Evaluation of a functional index and an articular index in ankylosing spondylitis. *J Rheumatol* 1988;15:302-7.
 22. Haywood KL, Garratt AM, Dziedzic K, Dawes PT. Generic measures of health-related quality of life in ankylosing spondylitis: reliability, validity and responsiveness. *Rheumatology* 2002;41:1380-7.
 23. Daltroy LH, Larson MG, Roberts NW, Liang MH. A modification of the Health Assessment Questionnaire for the spondyloarthropathies. *J Rheumatol* 1990;17:946-50.
 24. van der Heijde D, Bellamy N, Calin A, Dougados M, Khan MA, van der Linden S. Preliminary core sets for endpoints in ankylosing spondylitis. Assessments in Ankylosing Spondylitis Working Group. *J Rheumatol* 1997;24:2225-9.
 25. Calin A, Nakache JP, Gueguen A, Zeidler H, Mielants H, Dougados M. Defining disease activity in ankylosing spondylitis: is a combination of variables (Bath Ankylosing Spondylitis Disease Activity Index) an appropriate instrument? *Rheumatology* 1999;38:878-82.
 26. Doward LC, Spoorenberg A, Cook SA, Whalley D, Helliwell PS, Kay LJ, et al. Development of the ASQoL: a quality of life instrument specific to ankylosing spondylitis. *Ann Rheum Dis* 2003;62:20-6.
 27. Hewlett S, Smith AP, Kirwan JR. Values for function in rheumatoid arthritis: patients, professionals, and public. *Ann Rheum Dis* 2001;60:928-33.
 28. Cieza A, Hilfiker R, Boonen A, van der Heijde D, Braun J, Stucki G. Towards an ICF-based clinical measure of functioning in people with ankylosing spondylitis: A methodological exploration. *Disabil Rehabil* 2009;31:528-37.
 29. Schwarzkopf SR, Ewert T, Dreinhofer KE, Cieza A, Stucki G. Towards an ICF Core Set for chronic musculoskeletal conditions: commonalities across ICF Core Sets for osteoarthritis, rheumatoid arthritis, osteoporosis, low back pain and chronic widespread pain. *Clin Rheumatol* 2008;11:1355-61.