Aspects relevant for functioning in patients with ankylosing spondylitis according to the health professionals: a Delphi study with the ICF as reference

Annelies Boonen1,2, Monique van Berkel3, Inge Kirchberger4,5, Alarcos Cieza4,5, Gerald Stucki4,5 and Désirée van der Heijde1,2,*

Objective. In AS there is no agreed definition of which aspects are important when describing functioning. This limits the possibility to classify, evaluate and investigate the consequences of the disease. This study aimed to achieve consensus among health professionals on which aspects of functioning are typical and relevant for AS patients using the International Classification of Functioning, Disability and Health (ICF) as reference.

Methods. An international Delphi study through e-mail was performed among different health professions. Answers to open questions on areas relevant for functioning in the first round were linked to ICF categories and analysed in the two following two rounds for the degree of consensus.

Results. Of the 267 experts invited, 126 agreed to participate and 74 participated in all rounds; 28 were rheumatologists, 6 rheumatology nurses, 24 physiotherapists, 2 occupational therapists, 4 psychologists, 8 rehabilitation physicians and 2 social workers. More than 80% agreement was reached on 141 ICF categories, of which 30 (21%) were part of Body functions; 27 (19%) of Body structures; 56 (40%) of Activities and Participation; and 28 (20%) of Environmental factors. In addition, two Personal factors—illness knowledge and coping—were agreed upon.

Conclusion. 141 ICF categories and two personal factors represent the reference of functioning in AS from the perspective of health professionals. The largest number of categories concerned restrictions in activities. Also, the impact of AS on participation in life situations and the role of environmental factors were underscored. This broadens the view on functioning in AS and has implications for future research into functioning.

Key words: Ankylosing spondylitis, Disability evaluation, Quality of life, Attitude of health professionals, Outcome measures.

Introduction

AS is considered to be the most typical form of spondylo-arthritis with a prevalence in Caucasians ranging from 0.1 to 1.4 [1–3]. AS is diagnosed three times more often in men. The disease predominantly affects the axial skeleton with SI involvement as the hallmark of the disease. Pain and stiffness on one hand and spinal ankylosis on the other hand can decrease spinal mobility. In addition to the spinal manifestations, extra-skeletal comorbidity, comprising peripheral arthritis, enthesitis and uveitis or AS-related diseases such as psoriasis and IBD, can add to the burden of the disease. It is well known that the physical disability results in productivity loss for paid work and in substantial use of healthcare resources [4, 5].

Existing instruments that assess the impact on functioning in AS encompass a limited part of the full spectrum of consequences of AS on global functioning [6]. Based on the International Classification of Functioning, Disability and Health (ICF), which was endorsed by the World Health Assembly in May 2001, it is possible to systematically define a prototypical spectrum of functioning and health domains by using a globally agreed-upon language of functioning and health [7]. The components that are addressed in the ICF framework include Body functions and Body structures, Activities and Participation and Environmental factors. The ICF classification offers a universally accepted detailed list of ICF categories that can be necessary to describe functioning across conditions. Each ICF category is assigned a unique number and an agreed definition. The total ICF classification contains 1545 categories at the second, third and fourth levels. In the hierarchy of the system, the third- and fourth-level categories are specifications of the less specific higher level (i.e. second level). The definition of the prototypical spectrum of domains diminishes the risk of missing any important aspects of functioning and health of a patient with a specific health condition as well as any important aspect of the environment that may influence functioning. To be able to apply the ICF classification in medicine, disease-specific classifications are necessary. The categories that are considered relevant will not only differ according to the condition studied, but also according to the perspective taken. From the patients’ point of view other categories might be relevant than from the point of view of their different healthcare providers, their caregivers or healthcare decision makers [8, 9]. The present study aims to identify the most typical and relevant aspects of functioning for patients with AS according to the view of health professionals, using the ICF framework and classification as universal model and language of functioning.

Methods

A consensus-building, three-round, e-mail survey among medical experts using the Delphi technique was conducted. The Delphi technique is a qualitative research method, which takes its name from the Delphic oracle’s skills of interpretation and consenting on foresight, and has been used widely in health research within the fields of technology assessment, education and training, and in developing nursing and clinical practice [10]. It is a structured
communication process with four key characteristics: anonymity, iteration with controlled feedback, statistical group response and expert input [10]. It enables a large group of experts to be contacted cheaply, by mail or e-mail with a self-administered questionnaire, with little geographical limitation of the sample.

Recruitment of participants

The aim was to include experts from different health professions from all over the world. Within this context, experts are persons with interest and self-reported experience in AS. The type of health professionals that were considered were rheumatologists, rehabilitation physicians, physiotherapists, occupational therapists, social workers, rheumatology nurses and psychologists.

In order to compose the e-mail list of candidate rheumatologists, the mailing list of the full members of the Assessment in Ankylosing Spondylitis working group (now Assessment in SpondyloArthritis international Society; ASAS) was used. ASAS is an international group of experts comprising exclusively rheumatologists among the full members. To compose the e-mail list of other health professionals, presidents of national or international societies of rheumatology were contacted in order to learn whether e-mail lists per type of health profession could be made available and, if not, to name persons within their organization who could help in composing such lists. Also, a selection of rheumatologists from different world regions of the ASAS were invited to suggest names of non-rheumatologist health professionals with expertise in AS. Finally, the ICF Research Branch at the Ludwig-Maximilian University was asked to provide the e-mail list of non-rheumatologist health professionals that was used in similar Delphi exercises, performed in other musculoskeletal diseases.

Data collection and measures

Possible participants received in the first round of the Delphi study an invitation by e-mail. The invitation explained the aim and procedure of the study, and introduced the ICF. Those who were not willing to participate were asked to return the reason for non-participation which could be: no expertise, no time and no interest in research.

Those who were willing to participate were asked to complete the first part of the Delphi study, which was included in the e-mail. First, participants had to document their age, gender, professional background (specialty), the country where they work, and duration and level of professional expertise with AS. Level of self-reported expertise ranged from 1 (low) to 5 (excellent). Secondly, participants were asked to list areas they considered relevant and/or typical for functioning in AS for each of the ICF components: (i) Body functions, (ii) Body structures, (iii) Activities and Participation and (iv) Environmental factors.

To help participants understand these components, the WHO definitions of Body functions, Body structures, Activities and Participation and Environmental factors were added. To clarify the kind of information even better, an example containing a list of relevant and/or typical Body functions, Body structures, Activities and Participation and Environmental factors for patients with Parkinson’s disease was provided. Participants had to respond within 2 weeks, and a reminder was sent out ~3 days before the deadline. The experts were not informed about the names of other participants.

For all returned questionnaires, relevant areas reported by each individual expert were identified as ‘meaningful concepts’ and listed. Next, these concepts were linked to the closest related ICF categories [11]. Each of these steps was done by one investigator (M.vB.) and a random selection of 50% of the participants was checked blindly by a second investigator (I.K.). In case of disagreement, a third investigator was consulted (A.C.). All three persons were trained in rules to link concepts to ICF categories [11, 12]. The concepts recognized as personal factors cannot yet be linked to ICF categories, since personal factors are as yet not specified in categories. Therefore, these concepts were listed separately in the second round questionnaire as a fifth list ‘Personal factors’.

In the second round of the Delphi exercise, the list with all linked ICF categories (number of the category and definition) for each of the components Body functions, Body structures, Activities and Participation, Environmental factors as well as the list of personal factors was sent to the participants, together with an instruction on how to complete the second round. The participants were asked to consider whether each named ICF category and each personal factor is relevant and/or typical for patients with AS (yes/no). All experts who answered the first Delphi round were included. The participants had 2 weeks to respond, and reminders were sent out 3 days before the deadline.

In the third round, the list with all relevant ICF categories of the second round was sent to those who answered the first and second Delphi round, together with corresponding instructions for the third round. The questionnaire of the third round was constructed similarly to the questionnaire of the second round. It included all the ICF categories in the different components as well as the personal factors from the second round which were considered relevant and/or typical at least by one participant. In addition, information was provided from the previous round (i) as to whether the individual participant himself had considered this ICF category or personal factor as relevant and/or typical for patients with AS and (ii) as to which percentage of all participants had considered this ICF category as relevant and/or typical. The time schedule for response was identical to the previous rounds.

Analysis

Descriptive statistics were used to analyse participation rates and the characteristics of the participants. For each of the identified ICF categories and personal factors, the proportion of experts agreeing about the relevance of an ICF category was calculated. Consensus threshold was set at 80% of the participants indicating the relevance of the category after the third round. The results of the third round were further summarized on the second-level categories of the ICF by scrolling up third- or fourth-level category to the overlying second-level category. For each agreed upon second-level category after the third round, it was explored whether the agreement was consistent across professional subgroups and continents.

Results

Response and participation rates

Of all 267 experts who were invited, 189 (71%) responded initially that they were willing to participate. However, 63 declined before completing the first round, and the main reasons were: (i) lack of time for rheumatologists and (ii) lack of experience in AS for the other disciplines. In the second round, 89 (71%) of the responders in the first round and 47% of all completed the second Delphi round. Seven of the 37 non-participants in this round could not be reached by e-mail (the e-mail was returned repeatedly because of a full mail-box) and nine specified that they declined because they experienced difficulties to understand the ICF language in English. Finally, 74 of the 89 responders in the second round (83% of the responders in the second and 59% of the responders in the first round) completed the third round. Seventy-nine percent of participants were European or North American. Other participants came from Middle and South America (n = 5), Africa (n = 7) (mainly Morocco), Asia (n = 2) and Australia (n = 1). Table 1 shows the distribution of participants over the professions and the characteristics per profession. Those who had consented to participate but declined in the course of the
Delphi exercise were more often males (60% as opposed to 52% females), more often social workers or occupational therapists, had longer experience with AS (19 years as opposed to 12 years) and had a lower level of self-reported expertise (31% as opposed to 19% scored 1–3) when compared with those who completed all rounds.

Relevant and/or typical ICF categories

Consensus process. The development towards consensus during the Delphi rounds is summarized in Table 2. In the first round, the participants named 296 different ICF categories and seven personal factors. In the second and third rounds, all these categories were reported as relevant and/or typical at least by one participant. After the third round, >80% agreed on the importance of 141 categories, of which 30 (21%) were part of Body functions; 29 (21%) of Body structures; 55 (39%) of Activities and Participation; and 27 (19%) of Environmental factors. In addition, there was agreement on two Personal factors: illness knowledge and coping. Of the 141 categories, 50 were second-level categories, 82 were third-level and 9 were fourth-level categories. The largest number of third-level specifications was found in the component Activities and Participation. The 141 categories could be scrolled up onto 57 categories of the second level (taking into account that agreement on a third or fourth-level category indicates by definition agreement on the second level). As shown in Tables 3–6, 13 of these categories (23%) were part of the component Body functions; 10 (15%) of Body structures; 21 (37%) of Activities and Participation; and 14 (25%) of Environmental factors. In addition, there was >80% agreement on two Personal factors: illness knowledge and coping. The Appendix (available as supplementary data at Rheumatology Online) presents the percentage agreement on categories at each specific level.

Of the 57 finally agreed upon categories at the second level, full agreement across professional subgroups was found for 38 (66%) of the categories and full agreement across all continents for 46 (81%) categories. For the finally agreed upon personal factors,
there was 100% agreement across continents and professions. Subgroups not agreeing are indicated in the Tables 3–5. It is of note that in case of disagreement (which means <80%) across subgroups, the percentage agreement within that group was in most cases still >70%.

**Discussion**

In this Delphi exercise, consensus was reached on 141 ICF categories (57 at the second level) that are relevant to describe global functioning in patients with AS according to 74 experts from different professional backgrounds. In the component Body functions and Body structures, many categories were consistent with the inflammatory process, resulting in pain (b280) and stiffness (b780) and affecting functions and structures related to movement (Chapters 4, 7 and 27). In addition, exercise tolerance (b455), functions of sleep (b134), sexual functions (b640), recreation functions (b660) and experience of self (body image) (b180) were agreed as impairments relevant for functioning in AS. Also, the relevance of respiratory problems in case of involvement of the thoracic cage and of structures of the eye, in case of uveitis, was recognized. The largest number of categories agreed upon was part of the component Activities and Participation, and most categories pertained to mobility (Chapter 4: Changing and maintaining body position) and self-care (Chapter 5). In addition, >80% agreement was reached on the importance of restrictions in participation, including household work (b640), caring for household objects (d650), remunerative employment (b850), acquiring, maintaining and terminating a job (b845) and recreation and leisure (b920). In the component Environmental factors, categories concerning products for consumption (referring to medication), health professionals and societal attitude were perceived as most relevant and/or typical. Experts are also aware of the importance of personal factors for functioning. Illness knowledge and coping were perceived as relevant and typical.
Table 6. The second-level ICF categories in the component Environmental factors for which >80% agreement was reached, and the proportion of the participants (n=74) who agreed on the relevance

<table>
<thead>
<tr>
<th>Chapters and second-level categories</th>
<th>Experts, %</th>
<th>Professional subgroups not agreeing &gt;80%, % agreeing</th>
<th>Continental subgroups not agreeing &gt;80%, % agreeing</th>
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</thead>
<tbody>
<tr>
<td>Chapter 1: Products and technology</td>
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<tr>
<td>e110: For personal consumption</td>
<td>91</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>e115: For personal use in daily living</td>
<td>86</td>
<td>–</td>
<td>N-Am (75)</td>
</tr>
<tr>
<td>e120: For personal in- and outdoor mobility and transportation</td>
<td>85</td>
<td>PT (76)</td>
<td>N-Am (75)</td>
</tr>
<tr>
<td>e135: Products and technology for employment</td>
<td>88</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Chapter 3: Support and relationships</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>e310: Immediate family</td>
<td>81</td>
<td>PT (76)</td>
<td>N-Am (75)</td>
</tr>
<tr>
<td>e355 Health professionals</td>
<td>91</td>
<td>–</td>
<td>–</td>
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<tr>
<td>Chapter 4: Attitudes</td>
<td></td>
<td></td>
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<tr>
<td>e410: Individual attitudes of immediate family members</td>
<td>85</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>e450: Individual attitudes of health professionals</td>
<td>85</td>
<td>RN (71)</td>
<td>A (71)</td>
</tr>
<tr>
<td>e460: Societal attitudes</td>
<td>92</td>
<td>PT (60)</td>
<td>N (71) and S-Am (75)</td>
</tr>
<tr>
<td>Chapter 5: Services, systems and policies</td>
<td>100</td>
<td>PT (70); RP (78)</td>
<td>E (74)</td>
</tr>
<tr>
<td>e570: Social security, services, systems and policies</td>
<td>100</td>
<td>RN (71); RP (78)</td>
<td>A (71)</td>
</tr>
<tr>
<td>e575: General social support services, systems and policies</td>
<td>100</td>
<td>RN (71); RP (78)</td>
<td>A (71)</td>
</tr>
<tr>
<td>e580: Health services, systems and policies</td>
<td>100</td>
<td>RN (57); RP (67)</td>
<td>A (71)</td>
</tr>
<tr>
<td>e590: Labour and employment services, systems and policies</td>
<td>94</td>
<td>–</td>
<td>–</td>
</tr>
</tbody>
</table>

C/s: agreement for the second level as well as one or more third- or fourth-level categories; s: agreement for one or more third- or fourth-level categories; c: agreement for the second-level category and third or fourth level identified but not agreed; RN: rheumatology specialist nurse; PT: physiotherapist; RP: rehabilitation physician; N-Am: North Americans; A: African; E: Europeans.

The identified categories reflect the clinical experience of the experts with the most frequent limitations and restrictions encountered by patients but also their knowledge about manifestations of the disease that are less frequent, but might have important impact on functioning. For example, sleep (b134), pain (b280), structures of the pelvis (s740), maintaining body position (d415) likely are relevant categories for functional limitations for the majority of patients while respiration (b440) or structures of the eye (s220) can have important functional consequences but affect only a minority subgroup of patients.

Although the study was not designed to study differences across healthcare providers, for the large majority of second-level categories for which there was agreement, this was consistent across professions. Notwithstanding, interesting differences were seen. Psychologists did not agree that body image (b180) was impaired and likely feel the change in posture is experienced as real and not distorted. Rheumatologist emphasized the importance of possible impairments in structures of the eyeball and cardiovascular system, while this was not shared by most other professionals. Surprisingly, rehabilitation physicians agreed less on the participation restrictions that were identified by the other professionals. It could be that rehabilitation physicians are used to treating patients with conditions that result in more important restrictions in participation. The largest differences between subgroups and continents were seen for the environmental factors; the opinion of rheumatologists and Europeans was not shared in many cases by other health professionals or continents. This might indicate true difference in opinions, but also limited experience with assessing such factors in clinical practice.

When trying to compare the perspective of experts with the perspective of patients with AS, data from empiric interviews with the ICF checklist (165 categories) among 111 patients are available. Patients had to mention whether the stated category was relevant for them or not [13]. Within the limitations of comparability of both studies, a difference between the perspectives was noticed. Energy and drive (b130) was identified by 81% of the patients as impaired but agreed upon as relevant according to 69% of the experts only. Categories that were agreed upon by >80% of physicians to be relevant but were less frequently impaired or restricted by patients were respiration (b440; limited in 58% of the patients), sexual functions (b640; 30% of the patients), muscle power (b730; 58% of the patients), cardiovascular system (s440; 2% of the patients), respiratory system (s430; 58% of the patients), moving around using equipment (d465; 28% of patients), driving (d475; 75% of patients), washing oneself (d520; 70% of the patients) and remunerative employment (d830; 58% of the patients). This emphasizes that experts pay attention to aspects that might be rare but which can cause a large degree of disability. In addition, experts agreed upon the relevance of many more Environmental factors than were identified as facilitators or barriers by the patients. For the patients, only availability of drugs (e110), support of immediate family (e310) and health professionals (e355) were relevant in >80%. Experts also agreed on the importance of assistive products (e115, e120 and e135), transportation services, societal attitudes of family (e410) health professionals (e450) and society (e460) and services and employment policies on health, social security and employment and policies (e570, e580 and e590).

Some issues of this study need consideration. Despite efforts to recruit experts from different continents and cultures, the majority of responding experts came from Western Europe. This may reduce the generalizability of our results to other cultures. Furthermore, it was difficult to retrieve e-mail addresses of the health professionals. Partly, this is due to the fact that in most countries the non-rheumatologist health professionals are not yet organized as a separate and recognizable group and e-mail lists of societies for rheumatology or allied health professionals in rheumatology do not provide subgroups per profession. Also, of all 189 non-rheumatologist health professionals that were invited, only 80 (42%) participated and the main reason for not participating was no expertise in AS or difficulties to understand the language of the ICF in English. For rheumatologists, one of the main reasons to decline was lack of time. Notwithstanding, attrition rates in our study is higher than reported in most Delphi studies [14, 15]. Participants were included based on self-reported expertise and this was not verified externally. It was noticed that in the course of the Delphi rounds those with the lowest experience had the highest drop-out.

The completion of each Delphi round was quite time-consuming. However, none of the questionnaires returned were incomplete, suggesting responder fatigue did not play a major role.

The 80% cut-off point to express the consensus of the group of participants is based on the recognition that a higher cut-off point (e.g. 90%) would generate very few items, while lower cut-off point (e.g. 60%) would generate many items. However, it continues being arbitrary and can therefore be criticized.

In summary, 74 health professionals reached >80% agreement on 141 ICF categories and two personal factors that are relevant
for global functioning of patients with AS. The majority of categories pertain to restrictions in activities, but experts underscore the impact of AS on participation in several life situations and the role of several environmental factors in functioning and health. The findings will also be used in a further step to define the ICF Core Set for AS, which proposes a new standard to classify functioning by integrating the different perspectives (including the health professional’s perspective) on aspects important and typical for functioning and health in patients with AS [16].

Rheumatology key messages
- Health professionals agree on 141 ICF categories relevant for functioning AS.
- The importance of the impact of AS on Participation and role of Environmental factors is highlighted.
- The ICF Core Set for AS will integrate the perspective of this study with those of others.

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Supplementary data

Supplementary data are available at Rheumatology Online.

References