

The impact of ankylosing spondylitis on the life of patients

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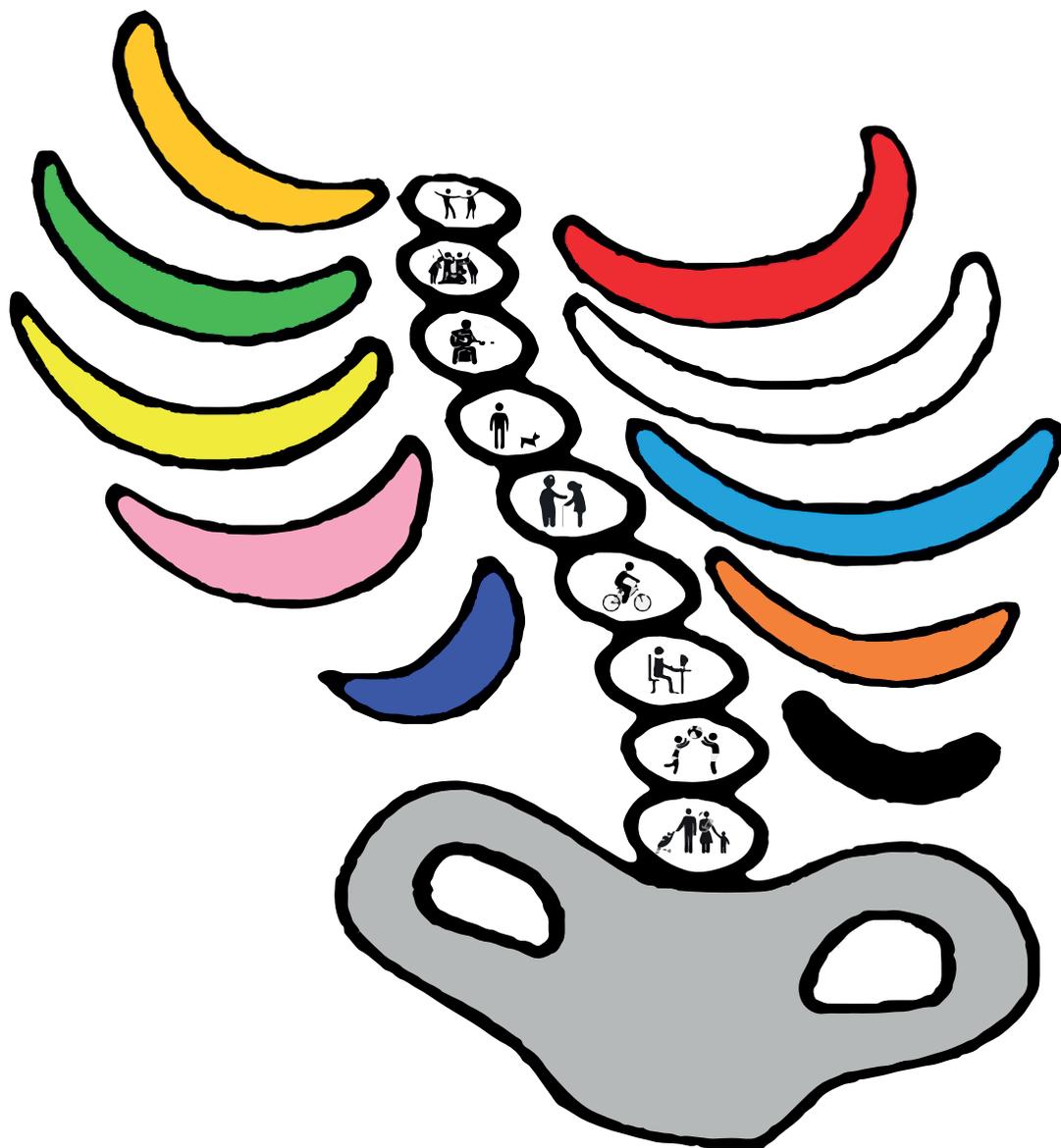
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The impact of ankylosing spondylitis on the life of patients; social role participation and physical activity



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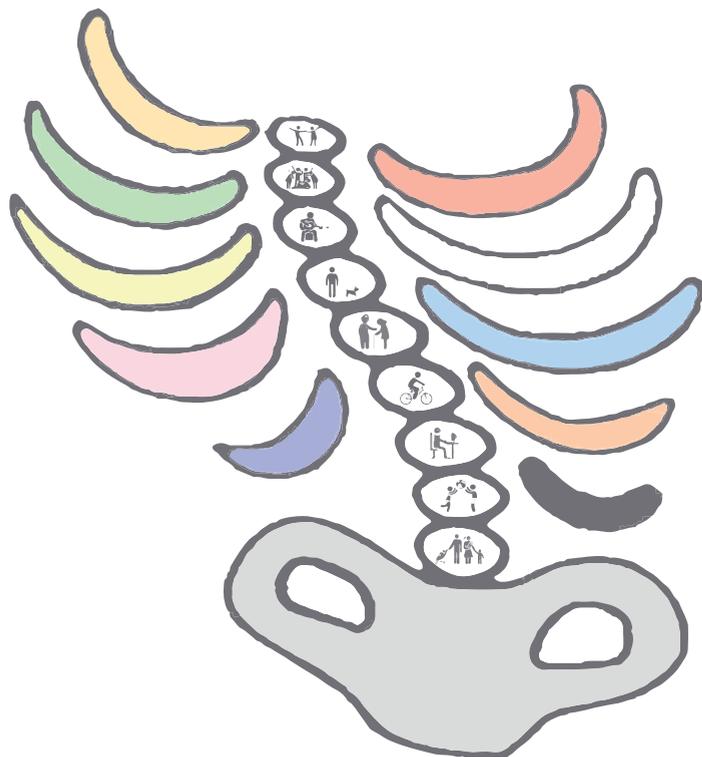
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General introduction



Ankylosing spondylitis (AS) is chronic inflammatory rheumatic disorder characterized by inflammation of the sacroiliac joints and spine, and in a subgroup of patients also of the peripheral joints (1). With a pooled prevalence of 0.18% (95% CI 0.15-0.23) worldwide and 0.25% (95% CI 0.18-0.33) in Europe, AS is not uncommon. Within the inflammatory rheumatic diseases, AS can be classified among the axial Spondyloarthritis (axSpA) type of diseases, which are all characterized by sacroiliac inflammation but lack the radiographic damage that is typical for AS (2). An important risk factor for AS is the presence of the human leukocyte antigen (HLA) B27: while the population prevalence is approximately 8%, 90% of Caucasian patients with AS have a positive HLA-B27 status (3, 4). Males are diagnosed with AS about two to three times more than females, but it has been shown that this may be prompted by deficits in the diagnosis of AS in females (5-7). The pro-inflammatory cytokines tumor necrosis factor alpha (TNF α) and interleukin 17 (IL17), play a central role in the pathobiology of AS. The inflammation causes pain, (morning) stiffness, reduced mobility and fatigue, and these are also the symptoms that have the largest impact on patients' lives (3, 8). The course of the disease usually follows a limited number of patterns, that mainly differ in the level of disease activity (9). Within these distinguished trajectories, disease activity can fluctuate over time with episodes of improvement and worsening (10). After diagnosis, patients undergo both pharmaceutical and non-pharmaceutical treatment (11). Pharmacological treatments aim to suppress disease activity, and have shown to modify importantly the course of inflammation (9, 12). Non-pharmacological treatment consist of patient education but mainly of exercise and physical exercise. The latter proven effects in maintaining and improving mobility and physical function (13).

Core outcomes to assess the impact of AS include pain, spinal stiffness, spinal mobility, physical function, fatigue, patient overall assessment, acute phase reactants, peripheral arthritis and radiographic damage. These outcomes and their relationships have been studied extensively (14). Although not all outcomes are necessarily core outcomes, two aspects relevant for patients' lives have not received sufficient attention. First, in view of the onset of the disease in the third decade of life, a period in which individuals are committed to various social roles like family life, professional career, but also social and leisure time, patients face the challenge how to cope best with these roles in life. Limitations in social role participation as a consequence of AS have received little attention in research and clinical care. Second, in both active as well as stable AS, engagement in physical activity (PA) to enhance daily physical functioning is important (12, 15). Patients with AS are known to experience difficulties in their daily tasks and activities, but it is unclear whether their actual (objective) performance of PA is reduced. A prerequisite for being physically active is the presence of appropriate muscle mass also known as fat free mass (FFM). Reduced FFM might contribute to actual performance, but also to experienced difficulty and fatigue when the same physical activity requires higher energy expenditure. Literature is available that report the risk of involuntary loss of FFM and about possibly associated risk factors.

An in-depth review of the literature would help to better understand whether loss of FFM occurs in patients with AS and other related subgroups of patients.

In this thesis we aimed to improve our understanding of social role participation in patients with AS and investigate the relation of AS and objectively assessed physical activity and fat free body mass. This introductory chapter will provide background on both topics, and will specify the research objectives and outline of the thesis.

The impact of AS on the life of a patient; social role participation

Defining participation

The word participation dwells from the Latin *pars* and *capere* (“part” and “to take”) (16). The International Classification of Functioning, Disability and Health (ICF) of the World Health Organization (WHO) terms participation as “an involvement in a life situation”, including being autonomous to some extent or being able to control your own life. Participation is the aspect of functioning beyond impairments in body functions and activities. That is, activities are goal- directed tasks, outside the setting of social life (17, 18).

Notwithstanding, in research on participation the jury is still not out on the demarcation between ‘activities’ and ‘participation’. The ICF describes the separate constructs ‘(limitations in) activities’ and ‘(restrictions in) participation’, but does not provide a straightforward distinction between the two when classifying the various ‘units of health’ into the single component ‘activities & participation’. Instead, the ICF manual offers the user four options predominantly based on the particular selection of domains to separate the constructs from each other (17), which in turn has led to debate (18-24). There seems to be agreement that there are mutual relationships between activity limitations and participation restrictions, which will result in correlations between the two (18, 21, 22). In addition, attempting to provide a distinction, data driven analyses showed no separation between activities (23) and participation (21, 22, 25). Notwithstanding, Badley et al. (2008), emphasized on engagements in social roles as one possible alteration of the ICF definition of participation, which is line with other studies (19, 23, 26, 27) and which is the approach we take in this thesis.

Measuring social role participation

Joining the debate regarding the concept, operationalizing participation into a measurement instrument is not straightforward. A great number of instruments are available, of which some have been validated in patients with rheumatic and musculoskeletal diseases (20, 24). All instruments for application in adults are patient reported questionnaires. In line with the discussion of the definition of participation, instruments vary in the *selection of social roles* they address. In a review of instruments, relationships with family and friends, role at home, social (including political and religious) life, and work/education

were consistently included in participation instruments. Inconsistencies were seen in the inclusion of sexual life, hobbies or travel/transport in the questionnaires (20).

Available instruments also differ in the way they assess different *aspects or dimensions* of social role participation. Classically in healthcare, patient reported outcomes address restrictions or difficulties in the performance of social roles, such as the World Health Organization Disease Assessment Score II (WHODASII) and the Short Form Health Survey 36 (SF-36) (28, 29). Yet, from a patient-centered perspective, other dimensions of social role participation seem to be relevant, as only the level of difficulty might not provide a complete representation of a person's participation. The level of 'satisfaction with the performance' or 'satisfaction with the time spent in participating in a social role' can be of interest, as it provides additional information on the personal appraisal of role performance or time spent independent of the difficulty or social participation (30). Moreover, 'role importance' seems highly relevant, as it includes the value the individual attaches to this role and can provide meaningful information that can be used as an additional qualifier for participation scores (18). Prior to the start of this thesis, evidence on social role participation in AS, was mainly limited to restrictions in worker participation and only two small study addressed restrictions in various social roles (31, 32). Moreover, no data from the Netherlands were available. This is important as participation is likely influenced by the socio-economic environment and culture. However, only one instrument was validated in patients with AS, but no Dutch translation was available (33). Hence, research on participation among Dutch patients would require first translation, cultural adaptation and clinimetric validation. To develop a further understanding of the construct and its specific value for patients with AS, the availability of a validated instrument to measure social role participation was needed. Availability of an instrument that assesses roles across relevant dimensions, would allow to understand the relative importance of different roles to patients and the experienced satisfaction with the performance or with the time spent participating. As availability of a benchmark would improve interpretation of the challenges patients experience when engaging in social roles, comparison of social role participation of persons without chronic disease would be insightful. Finally, although participation in social life can be a meaningful goal on itself, it is also important that it can contribute to a patient's overall life satisfaction. Life satisfaction as an outcome of healthcare receives increasing attention, as disease is incompatible with health, but not incompatible with satisfaction with life (34). No data on life satisfaction in AS are available and the association with social role participation is as yet unknown.

The relevance of social role participation

Regardless of a person's health state, participation in different roles in life has high individual, but also communal, economic and in the Netherlands even political value through the participation law. Why should an individual participate in social roles? In our society, participation is related with social status, financial welfare and autonomy (35, 36). In ad-

dition, participation might contribute to well-being, satisfaction with life or happiness (37, 38). Having entered an era in which person-centeredness is eminent, the goal of medical care now goes beyond the mere absence of disease and strives for more than 'optimal health'. Presumably, persons with chronic disease define their goals in life by selecting social roles they wish to fulfill or accomplish. Gaining insights into the importance of roles and restrictions in participation seems a first step when aiming to support persons in individual life-goals.

The impact of AS on the life of a patient; physical activity and body composition

Physical activity and body composition

Given the importance for AS patients to perform daily activities, 'physical function', has been selected as a core outcome domain that should be measured in trials as well as in clinical record keeping (3, 39, 40). Following the recommendations of the Assessment in AS International Society (ASAS), physical function in AS is usually measured by the Bath Ankylosing Spondylitis Functional Index (BASFI) (41). This is a self-reported questionnaire that asks patients to rate the *level of difficulties* on a visual analogue scale or 0-10 numeric rating scale the patients experience when executing 10 different tasks and activities. As such, scores are a personal appraisal of efforts required to perform tasks (42). Physical activity (PA) addresses the performance of movements, independent of an appraisal or experience and refers to the main underlying process underlying physical function which can be defined as "any bodily movement produced by skeletal muscles that results in energy expenditure" (43). Self-reported measures of PA outcomes are practical for use and usually ask about the *time spent* in tasks or activities with different levels of intensity (44). However, such measures are prone to recall-bias and overestimation (45). As opposed to questionnaire-based instruments an accelerometer or activity monitor offers the opportunity to objectively measure PA. Accelerometers are considered to be the most valid technique to assess PA with information on activity patterns in terms of frequency, duration and intensity. Activities of daily life (ADL) such as dressing, eating and walking all require light to moderate intensity PA (46). PA in moderate and vigorous intensities (MVPA) have well-known beneficial health effects and is associated with a longer life expectancy (47). In AS, twelve studies assessed PA of which six studies applied a self-reported questionnaire (48-53), four studies used an accelerometer (54-57) and two studies used a combination of both (58, 59). Results of these studies reveal varying data on total amount and of time spent in different intensities of PA within patients, on the extent in which patients differ in PA when compared to controls, or whether patients comply with the WHO recommendations. Also, there is lack of insight whether experienced *difficulties* to perform tasks have a different relation with *amount of PA* in patients compared to controls.

As clarified above, PA is any bodily movement caused by skeletal muscles that in turn requires energy. Basically, without skeletal muscles there is no PA and hence, any condition that affects the musculoskeletal system may have a direct impact on PA. Skeletal muscles constitute the largest component of fat-free mass (FFM), i.e. about 44%, other components being internal organs, bone and extracellular tissue (60). Hence, skeletal muscle also comprises the largest protein storage of the human body. In chronic inflammatory rheumatic diseases such as rheumatoid arthritis, an accelerated loss of muscle mass has been observed, also referred to as rheumatoid cachexia. Cachexia is the involuntary loss of muscle mass, or fat-free mass in general, and is also observed in patients with other chronic diseases like chronic obstructive pulmonary disease (COPD) and cardiovascular disease (CVD) (61, 62). Cachexia is a serious condition and can cause (further) functional impairment and even mortality (63). In patients with AS, studies investigating FFM are scarce. Given the importance of cytokines like TNF- α in the development of cachexia and its involvement in the pathogenesis of AS, it is plausible to assume that these patients are also at risk for accelerated muscle loss. In addition, the fact that patients with AS may be hampered in their PA by pain and limitations in body movement poses an added risk for muscle loss. Lower muscle mass in itself may in turn negatively affect PA (figure 1). In short, chronic inflammation in patients with AS does not only causes spinal pain and spinal mobility limitation, but may also increase protein catabolism and decrease protein anabolism, all leading to a decrease in muscle mass which in turn may hamper physical activity even further. Literature on body composition is still scarce and direct comparisons with population controls are still needed. Moreover, available literature on the impact of AS on fat-free mass and possible associated factors and interventions that may affect the loss of FFM in patients has not yet been appraised and summarized systematically.

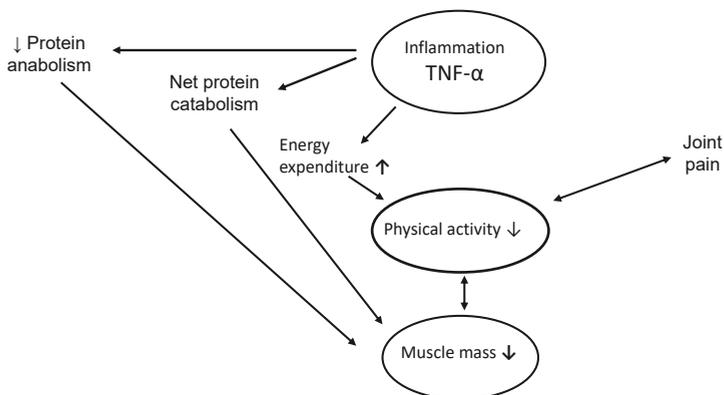


Figure 1. Chronic inflammation in patients with AS not only causes joint destruction with consequent pain and mobility limitations of the joints, but can also increase protein catabolism and decrease protein anabolism, all leading to a decrease in muscle mass which in turn may hamper physical activity even further.

Importance

Sufficient PA is important for the general health and physical inactivity over a longer period can lead to serious conditions like cardiovascular disease (CVD), Diabetes Mellitus II (DMII) and cancer (64). The WHO recommends 150 min of PA a week in moderate intensity, 60–75 min in vigorous intensity, or an equivalent combination of both (64). According to the WHO, more than 80% of the world's adolescent population is insufficiently physically active (65). In comparison with the general population, patients with AS also need to be active to manage the pain and stiffness in their joints. Insight into amounts and patterns of PA in AS patients will help to define actions to further support healthy PA.

In addition, in inflammatory rheumatological disorders, there is still insufficient insight into the magnitude of muscle loss as well as into factors contributing to the problem. Despite pathophysiological plausibility, it remains unresolved whether cachexia is common in all inflammatory musculoskeletal diseases.

Main aim of this thesis

The main aim of this thesis is to better understand the impact of AS on patient's lives by examining two underexplored areas: social role participation and PA. For each of these areas several questions were addressed at the start of this thesis.

In PART I we aim to explore social role participation across different roles and dimensions and want to understand whether social role participation is important for patients' satisfaction with life

More specifically we addressed the following research questions

1. Can we translate the English 'Social Role Participation Questionnaire' into a valid questionnaire to assess social role participation among Dutch patients?
2. What is the social role participation in patients with AS and what are differences compared with population controls?
3. What is the relation of social role participation and satisfaction with life in patients with AS and is this relation different than in population controls?

In PART II we examine physical activity and the literature on the body composition of patients with AS compared to controls

We addressed herein the following research questions:

4. Do patients with AS and healthy controls differ to a similar extent in the difficulties they experience with tasks and activities as in the total amount of physical activity?
5. Do patients with AS and population controls differ regarding their objectively measured activity patterns and which factors explain potential differences?
6. What is the evidence in the existing literature on fat free mass in patients with SpA or RA compared with controls, and on factors associated with FFM in these patient groups?

Sources of data

For the studies described in PART I, a collaboration with an official translation office was established, providing sessions with a translation and separate lay-panel. For the clinimetric validation, the comparison of social role participation and the analyses of the relation with life satisfaction 246 patients with AS and 510 controls were recruited and constituted the Social Participation in Ankylosing Spondylitis Study (SPASS). This regarded patients from six hospitals in the Netherlands of at least 18 years, registered with AS according to Dutch 'diagnosis related groups' or similar patient-lists and in whom the modified New York criteria was confirmed by the treating rheumatologist. The N=510 control subjects were recruited from an open national online panel of the research institute Ipsos (Ipsos, Amsterdam, the Netherlands).

For the studies in PART II, first a smaller patient control study is described, comprising of 24 patients with AS and 24 healthy adults. Additionally, a sub-sample of 135 patients with AS and 99 controls of the SPASS study were examined. In the systematic review of the literature a total of 48 articles were appraised and summarized.

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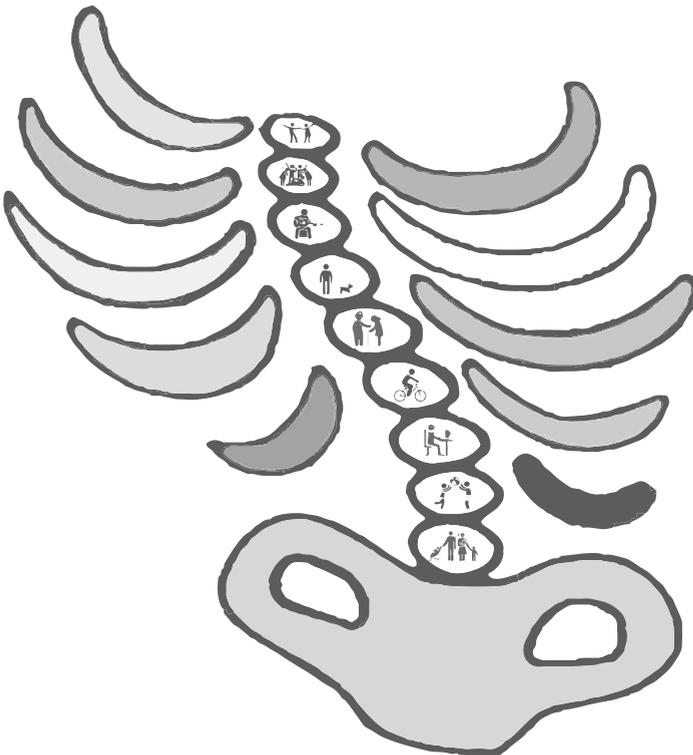
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Part 1

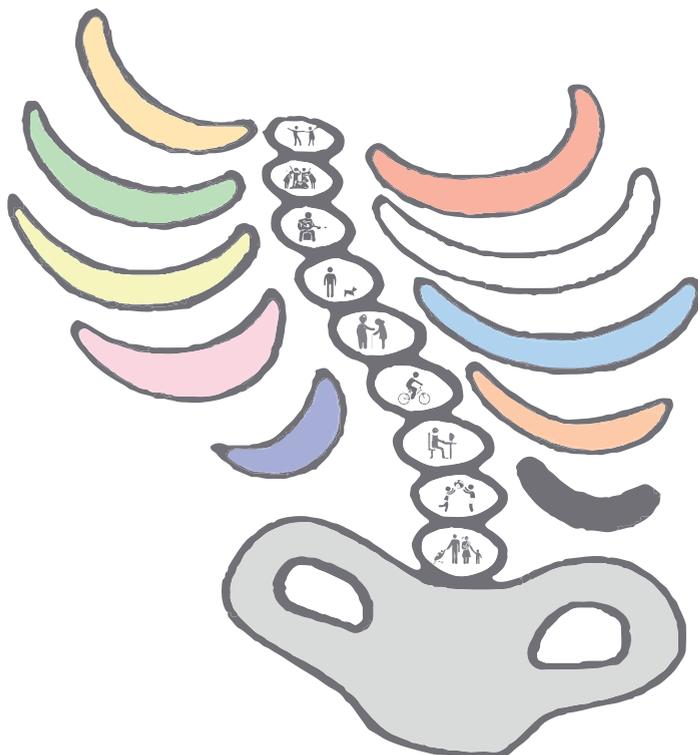
Social role participation



2

The Social Role Participation Questionnaire for patients with ankylosing spondylitis: translation into Dutch, reliability and construct validity

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Abstract

Objective: The social role participation questionnaire (SRPQ) assesses the influence of health on participation in 11 specific and one general participation role across 4 participation dimensions; 'importance', 'satisfaction with time', 'satisfaction with performance' and 'physical difficulty'. This study aimed to translate the SRPQ into Dutch and assess clinimetric properties and aspects of validity among patients with ankylosing spondylitis (AS).

Methods: Translation was performed using the dual panel approach. For each participation dimension, internal consistency, test-retest reliability (n=31), and construct validity were assessed in 246 AS patients.

Results: The translation required only minor adaptations. Cronbach α 's were $\alpha \geq 0.7$. A strong correlation was present between satisfaction with 'time' and 'performance' ($r = 0.85$). Test-retest reliability was satisfactory (Kappa=0.79-0.95). Correlations with participation domains of the Short-Form Health Survey 36, the World Health Organization Disease Assessment Score II, and generic as well as disease specific health outcomes (Physical and Mental component scale of the SF-36, satisfaction with life scale, BASDAI, BASFI) were at least moderate ($r = 0.41$ to 0.75) for all dimensions except for 'role importance' where correlations were weak ($r \leq 0.40$). Discriminative ability across 5 self-reported health states was good for all dimensions ($p < 0.01$). The 'general participation' role showed similar reliability and validity for each dimension, as the average of the all 11 roles.

Conclusion: The Dutch version of the SRPQ is available to help understand social role participation in patients with AS. The dimension 'role importance' measures a distinct aspect of participation. The general participation item was a good global measure of participation.

Introduction

Participation in social roles is increasingly recognised by healthcare professionals, as well as by policymakers, to be an important outcome of healthcare and healthcare services. Ankylosing spondylitis (AS) is a chronic inflammatory rheumatic disease with a usual onset in the third decade of life, when persons are committed to various social roles that adults fulfil (1). While a large amount of literature is available on restrictions on worker participation, the number of studies and the knowledge about restrictions in the full spectrum of adult social roles for patients with AS is limited (2, 3). Research is hampered by continuing discussions on the exact definition of participation and challenges to operationalizing the concept (4, 5).

With regard to the definition, the International Classification of Functioning, Disability and Health (ICF) defines participation as ‘an involvement in a life situation’, and classifies the concept as the aspect of functioning beyond impairments in body functions and activities, which are typical and relevant in the larger setting of someone’s life (4, 5).

When operationalising the concept, existing instruments first vary according to the participation roles they include. In a review of instruments, relationships with family and friends, role at home, social (including political and religious life) life and work/education were recognised as unmistakable participation roles. On the other hand, discussion remains whether sexual life, hobbies or travel/transport reflect social role participation (6). Second, instruments differ with respect to which aspects or dimensions of social roles they measure. While the majority of instruments address restrictions or difficulty in performance of social roles, 4 other aspects/dimensions of social role participation seem to be relevant with regard to health and/or the experience of health. From a patient-centered perspective, the level of ‘satisfaction’ can be of interest as it provides additional information on the personal appraisal of role performance independent of the difficulty or level of social participation (7) Moreover, ‘role importance’ seems highly relevant, as it includes the value the individual attaches to this role and can provide meaningful information that can be used as an additional qualifier for participation scores.⁴

The Social Role Participation Questionnaire (SRPQ) is a promising instrument as it assesses a broad range of roles, including one ‘general participation role’, across various dimensions.^{8 9} The English version of the SRPQ has proven to be reliable and valid for use in patients with AS and osteoarthritis in Canada.^{8 10} Since no validated Dutch patient-reported measure exists to assess participation in Dutch patients with AS, the present study aimed to translate the English version of the SRPQ into Dutch, assess reliability, and further explore the construct validity for Dutch patients with AS.

No major problems for translation and cultural adaptation were expected. With regard to internal consistency and construct validity, it was hypothesised that the ‘importance’ dimension of the SRPQ would provide dissimilar results because this dimension conceptually differs from other dimensions. Finally, we expected that the ‘general participation’ item would have a similar construct validity compared with average scores of all 11 included roles in the SRPQ.

Materials and methods

SRPQ, translation and cultural adaptation

The initial version of the SRPQ assesses the influence of health on 11 specific social roles and 1 ‘general participation’ item across 3 participation dimensions: ‘role importance’, ‘satisfaction with time spent in roles’ and ‘satisfaction with the role performance’ (9). As subsequent validation showed a high correlation between the two ‘satisfaction’ dimensions and indicated that a dimension on difficulty with participation was missing, a modification was proposed to exclude ‘satisfaction with time’ and add ‘physical difficulty’ (8, 10). Given that this suggested modification for SRPQ was never validated, we decided to include all four dimensions in the current study. All roles of the SRPQ can be scored in each dimension on a five-point Likert scale (1: not at all important/not at all satisfied/unable to do to 5: extremely important/ extremely satisfied/no difficulty). For the roles employment, education, intimate relationships and relationship with children/step-children/grandchildren, the patients can indicate that this role is not applicable and consequently, the ‘difficulty’ and ‘satisfaction dimensions’ will not be completed (figure 1).

For each dimension, a summary score can be calculated for presenting the average of all roles. To deal with non-applicable roles, for the dimension ‘physical difficulty’ it is assumed that no restrictions are experienced if the role is not applicable; however, for the satisfac-

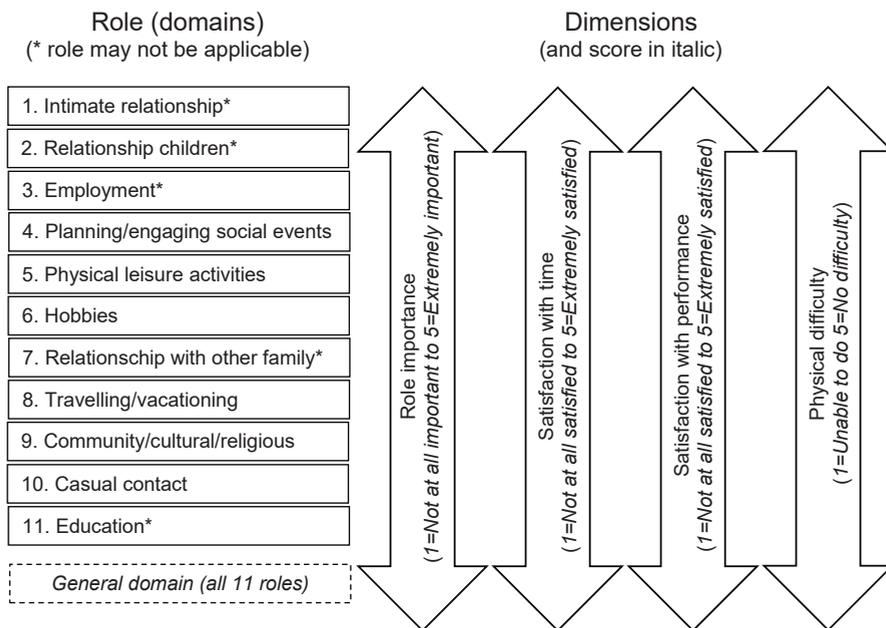


Figure 1. Graphical representation of the content of the Social Role Participation Questionnaire (SRPQ), which assesses several dimensions of role participation across 11 specific roles and general role participation item

tion dimensions the average scores are only calculated if participants participate in at least 9 of the 11 roles (8).

Translation of the SRPQ was performed following the dual panel approach (11). First, four bilingual translators (native Dutch speakers) and 2 native (English) speakers worked together to produce a preliminary Dutch translation. Second, a healthy Dutch lay panel, consisting of three women and three men varying in age (range 25– 64 years) and level of education, discussed the wording and comprehensiveness of the translation under the supervision of the project leader who was part of the first step. Finally, cognitive debriefing interviews with the adjusted version were conducted among five patients with AS. The final version was back-translated into English and the developer's approval was sought.

Clinimetric properties and validation

The COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) guidelines and checklist manual were used to ensure methodological quality of the current study (12,13)

Participants

Patients were recruited as part of a multicentre cross-sectional study, the Social Role Participation in Ankylosing Spondylitis Study (SPASS). Patients from six hospitals in the Netherlands who were at least 18 years of age, registered with AS according to Dutch 'diagnosis related groups' or similar patient lists, and in whom the diagnosis, according to the modified New York criteria, was confirmed by the treating rheumatologist were invited to participate by a letter. Patients were excluded if they indicated they had no access to the internet or were not familiar with the Dutch language. A random subsample of patients was invited to complete the SRPQ again after 2 weeks to evaluate test-retest reliability. The ethics committee of the academic hospital Maastricht and Maastricht University approved the study, and informed consent was obtained from all participating patients.

Assessments

Patients completed an online survey. Questions on socio-economic background comprised age, gender, highest completed educational degree and work status. Social participation was assessed using the SRPQ, and by social role domains of the WHO Disease Assessment Score II (WHODASII) and the Short Form Health Survey 36 (SF-36). The WHODASII contains 36 items across six subdomains assessing difficulties in understanding and communicating, getting around, self-care, getting along with people, life activities, and participation in society. The latter three subdomains are considered to represent participation. Domain scores range from 0 to 100, lower scores reflecting fewer limitations (14). The SF-36 contains 36 items assessing difficulties due to mental or physical health across eight domains: social function, physical function, bodily pain, role-physical, general health, vitality, role-emotional and mental health. The domains social functioning, role-physical and role-

emotional represent participation roles. Domain scores range from 0 to 100 (higher scores reflecting less difficulties/health problems). In addition, two summary scores, the Physical Component Summary (PCS) and Mental Component Summary (MCS) scores (range 0–100) can be calculated to present overall health-related quality of life (HR-QoL) (15). To assess disease-specific aspects of health, patients indicated their disease duration, current use of tumor necrosis factor- α inhibitors, and completed the Bath Ankylosing Spondylitis Disease Index (BASDAI) and the Bath Ankylosing Spondylitis Functioning Index (BASFI). The BASDAI is a composite score of six questions assessing different aspects of disease activity with AS. Scores for each item and the total index range from 0 to 10 (10 reflecting highest disease activity) (16). The BASFI determines the level of normal daily functioning of people with AS across 10 items. Scores for each item and total index range from 0 to 10 (10 reflecting most impairments in functioning) (17). Finally, the Satisfaction With Life Scale (SWLS) was added to assess the overall satisfaction with the respondent's life. The questionnaire has five items that can be rated on a Likert scale (1 not at all agree to 7 totally agree). Total scores range from 5 to 35, higher scores indicating more life satisfaction (18, 19).

Statistical analyses

Statistical analyses were performed using PASW Statistics V.20 (SPSS, Chicago, Illinois, USA). Analyses are performed for each dimension of the SRPQ by first using the averaged 11 specific role score and next the score on the single 'general participation' item.

Internal consistency was assessed by Cronbach's α coefficients (adequate: ≥ 0.70) (20). Item deletion analyses were performed to reveal any outstanding items influencing the Cronbach's α . Spearman correlations between dimensions were computed (correlation weak: ≤ 0.40 ; moderate: > 0.41 but ≤ 0.75 ; strong: > 0.75) (21).

Floor and ceiling effects were defined as $> 15\%$ of patients scoring not at all important/not at all satisfied/unable to do (floor effect) or $> 15\%$ of patients scoring very important/very satisfied/no difficulty (ceiling effect), in the dimensions score or single item score (22).

Test re-test reliability as assessed assuming stable health over the 2 weeks' assessment interval, first by weighted κ s with quadratic weights (agreement slight (≤ 0.2), fair (> 0.2 but < 0.4), moderate (≥ 0.41 but < 0.6), substantial (≥ 0.6 but < 0.8) and almost perfect (≥ 0.81)) (23) and second by the smallest detectable difference (SDD).

Construct validity with other participation measures was tested by Spearman correlations with the participation domains of the WHODASII (6) and the SF-36. Construct validity with other health outcomes was tested using Spearman correlations with PCS and MCS of the SF-36, BASDAI, BASFI and SWLS.

Finally, to assess whether the SRPQ is discriminative between groups that differ in overall health (the five health states of the SF-36 general health item (1=poor to 5=excellent health)), differences between provided scores within each dimension were calculated by using Kruskal-Wallis test.

Results

Translation and cultural adaptation

The translator and lay panel identified issues concerning the wording of the roles representing work and education. The original English version asks about the type of paid work or education that ‘you are able to have’ and not ‘you are able to do’ as for all other roles. The panels considered this difference as liable to misinterpretation. Moreover, in the Dutch situation, ‘able to have’ would be less applicable and ambiguous, as society focuses on maintaining work and less on considering other types of work. Hence, formulation was changed in accordance with the questioning in the other roles. Results of the cognitive debriefing showed that the translated version of the questionnaire was correctly understood, and no roles or dimensions were missing or unimportant. The average time to complete the SRPQ was 12±7 min. The back-translated final version was approved by the developer.

Clinimetric properties of the SRPQ

Samples

Of the 740 patients invited, 296 (40%) agreed to participate of which 246 (83%) completed the questionnaire. Of these patients, 31 participated in the test-retest reliability sub-study. The characteristics of the study population are given in table 1. Most patients were male (62%), average age 51±12 years and diagnosis duration of 17±12 years. For roles that might not be applicable, the table also presents the number and proportions of persons for whom roles were applicable. This resulted in n=235 (96%) patients who had participated in at least nine roles (and therefore contribute to the satisfaction dimension).

Internal consistency

Cronbach’s α was adequate for all dimensions but, as hypothesised, this was lower for ‘role importance’ ($\alpha=0.74$) compared with the ‘satisfaction with time’ ($\alpha=0.83$), ‘satisfaction with performance’ ($\alpha=0.89$) and ‘physical difficulty’ ($\alpha=0.86$). Item deletion did not indicate any outstanding items influencing Cronbach’s α . The correlations between the dimension ‘role importance’ and each other dimensions were weak (‘satisfaction with time’ ($r=0.20$), ‘satisfaction with performance’ ($r=0.21$) and ‘physical difficulty’ ($r=0.10$)). Strong correlations were observed between both ‘satisfaction’ dimensions ($r=0.85$), and moderate correlations between ‘physical difficulty’ and either ‘satisfaction with performance’ ($r=0.69$) or ‘satisfaction with time’ ($r=0.52$). The correlations between the single general participation item and the averages role scores were moderate for the dimension ‘role importance’ ($r=0.59$), and strong for the other dimensions ($r\geq 0.80$).

Floor and ceiling effects

In none of the dimensions were floor or ceiling effects found. The general participation item showed a ceiling effect in 43% of patients in the dimension ‘role importance’ (participation very important).

Table 1. Characteristics of 246 patients with ankylosing spondylitis

	n, % or Mean (SD) [min - max]
Gender (Males)	153 (62%)
Age (years)	51 (12) [24 - 79]
Diagnosis duration (years)	16.8 (11.8) [5 - 44]
BASDAI	4.4 (2.3) [0- 10]
BASFI	4.2 (2.5) [0- 10]
Current use of TNF-alpha inhibitors (n, %)	123 (50%)
Higher professional education or university (n, %)	81 (33%)
SF-36 PCS	38.7 (10.1) [5.2 - 61.9]
SF-36 MCS	49.2 (12.8) [10.9 - 70.5]
SWLS	22.2 (7.2) [5 - 35]
<i>SRPQ roles that are optional:</i>	
Has a partner, intimate relationship (n, %)	187 (79%)
Has (step/grand) children	184 (75%)
Has other family	239 (98%)
Employment (n, %)	140 (57%)
Work disabled (n, %)	59 (24%)
Following some form of education (n, %)	40 (16%)

BASDAI, Bath Ankylosing Spondylitis Disease Index; BASFI, Bath Ankylosing Spondylitis Functioning Index; SF-36, 36 item Short Form health survey Physical Component Score and Mental Component Score; SWLS, Satisfaction With Life Scale

Test-retest reliability

The weighted κ was substantial in the ‘role importance’ dimension ($\kappa=0.79$), and almost perfect for the dimensions ‘satisfaction with time’ ($\kappa=0.84$), ‘satisfaction with performance’ ($\kappa=0.85$) and ‘physical difficulties’ ($\kappa=0.95$). The reliability of the general participation item was lower, with a κ that was fair for ‘role importance’ ($\kappa=0.30$) and substantial ($\kappa\geq 0.7$) for the other dimensions. The SDDs for averaged scores and general participation score [in square brackets] were 0.74 [1.60], 0.69 [1.23], 0.83 [1.35] and 0.38 [1.29] for the dimensions ‘role importance’, ‘satisfaction time’, ‘performance’ and ‘physical difficulty’, respectively.

Construct validity

As expected, the dimension ‘role importance’ correlated weakly with the six participation domains of WHODAS or SF-36 (table 2). ‘Satisfaction’ with time or performance correlated

Table 2. Spearman correlation coefficients of the SRPQ dimensions and other (domains) of questionnaire assessing participation.

	SF-36 Social functioning	SF-36 Role emotional	SF-36 Role physical	WHODASII Getting along with people	WHODASII Life activities	WHODASII Overall participation
<i>SRPQ dimension (average of all roles):</i>						
Role importance	0.063	0.003	0.255	-0.181	-0.009	-0.058
Satisfaction time*	0.586	0.378	0.335	-0.399	-0.435	-0.562
Satisfaction performance*	0.720	0.390	0.560	-0.461	-0.546	-0.646
Physical difficulty	0.735	0.384	0.713	-0.471	-0.714	-0.665
<i>SRPQ General Participation Item:</i>						
Role importance	0.075	0.018	0.164	-0.094	0.133	-0.040
Satisfaction time	0.603	0.377	0.392	-0.412	-0.444	-0.566
Satisfaction performance	0.582	0.323	0.464	-0.402	-0.453	-0.575
Physical difficulty	0.693	0.390	0.611	-0.461	-0.640	-0.625

*Average scores of the 'satisfaction' dimensions could only be calculated for 235 of the 246 patients. SF-36, 36 item Short Form health survey; WHODASII, World Health Organization Disease Assessment Score II

moderately with three or five external measures of participation, respectively. ‘Physical difficulty’ correlated moderately with five participation measures. Correlations of the general participation item with external measures were comparable to the correlations based on the averages of specific roles. The weak correlations for the satisfaction and difficulty dimensions were always seen with the ‘emotional role’ of the SF-36.

The dimension ‘role importance’ correlated weakly with all five measures representing aspects of HR-QoL (table 3). ‘Satisfaction with time’ correlated moderately with two, and weakly with three HR-QoL measures. ‘Satisfaction with role performance’ and ‘physical difficulty’ correlated moderately with all available HR-QoL measures, except for a weak correlation between the dimension ‘physical difficulty’ and the SF-36 MCS. Finally, the general participation item showed moderate correlations for all dimensions with the exception of ‘role importance’, which showed a weak correlation. Overall, the best correlations were seen between the ‘satisfaction’ dimensions and SWLS, and between the ‘physical difficulty’ dimension and SF-36 physical role, BASDAI and BASFI.

Discriminative ability

Finally, all dimensions as well as the general participation item were discriminative between the five health states of the general health question in the SF-36 (table 4), except for the dimension ‘role importance’ in the general participation item.

Table 3. Spearman correlation coefficients of the SRPQ and other instruments assessing generic or disease specific (aspects of) health in AS

	SF-36 PCS	SF-36 MCS	SWLS	BASDAI	BASFI
<i>SRPQ dimension (average all roles):</i>					
Role importance	0.146	0.004	0.166	-0.040	-0.260
Satisfaction time*	0.335	0.528	0.656	-0.339	-0.359
Satisfaction performance*	0.504	0.490	0.653	-0.483	-0.546
Physical difficulty	0.691	0.350	0.468	-0.636	-0.723
<i>SRPQ General participation Item:</i>					
Role importance	0.108	-0.004	0.168	0.002	-0.157
Satisfaction time	0.383	0.503	0.649	-0.370	-0.380
Satisfaction performance	0.439	0.422	0.652	-0.415	-0.426
Physical difficulty	0.634	0.379	0.457	-0.583	-0.585

*Average scores of the ‘satisfaction’ dimensions could only be calculated for 235 of the 246 patients. BASDAI, Bath Ankylosing Spondylitis Disease Index; BASFI, Bath Ankylosing Spondylitis Functioning Index; SF-36, 36 item Short Form health survey Physical Component Score and Mental Component Score; SWLS, Satisfaction With Life Scale

Table 4. Average SRPQ scores across SF-36 rated health states for 246 patients with AS

SF-36 current health state	Poor N=20 8.1%	Fair N=109 44.3%	Good N=93 37.8%	Very good N=22 8.9%	Excellent N=2 0.8%	<i>p</i>
<i>SRPQ dimensions (average all roles)</i>						
Importance (1 - 5)	3.59	3.65	3.85	3.96	4.15	0.024
Satisfaction time* (1 - 5)	2.47	2.99	3.47	3.82	3.90	<0.001
Satisfaction performance* (1 - 5)	2.36	2.85	3.54	4.04	4.01	<0.001
Physical difficulty (1 - 5)	3.45	3.82	4.33	4.67	5.00	<0.001
<i>SRPQ General Participation Item:</i>						
Importance	4.25	4.18	4.25	4.64	5.00	0.062
Satisfaction time	2.45	2.97	3.55	4.18	4.00	<0.001
Satisfaction performance	2.15	2.59	3.41	4.14	3.50	<0.001
Physical difficulty	2.45	3.18	3.85	4.36	5.00	<0.001

*Average scores of the 'satisfaction' dimensions could only be calculated for 235 of the 246 patients. SRPQ dimension scores range from (1: not at all important/not at all satisfied/unable to do to 5: extremely important/extremely satisfied/no difficulty).

Discussion

This study translated and culturally adapted the SRPQ into Dutch, and further tested several aspects of its validity among patients with AS. The translated questionnaire was found to be relevant and comprehensible by Dutch patients with AS; it showed overall good clinimetric characteristics and confirmed the hypothesis on construct validity.

In rheumatology, translation followed by back translation and cognitive debriefing among patients as the last step is the most common approach to translate and culturally validate questionnaires. Our study confirms that the dual panel approach provides greater efficacy due to more intense involvement of possible end users (11, 24-28).

By using the dual panel approach, we identified at an early stage (before the cognitive debriefing) the need to change the wording for the roles education and work in the dimension 'satisfaction with performance' as patients found the initially proposed translation difficult to interpret, liable to make mistakes and less applicable in Dutch society. The proposed adaptations were subsequently tested in the cognitive debriefing round and were not considered to be ambiguous anymore.

As hypothesised, the dimension 'role importance' showed lower internal consistency, weak correlations with the other dimensions of the SRPQ, and weak correlation with other instruments assessing either participation or an aspect of health. Clearly, the assessment of 'role importance' provided different information than other dimensions and can, therefore, be useful in the interpretation of the relevance of scores of the remaining

dimensions. Further, it was confirmed that both dimensions for ‘satisfaction’ correlated strongly. As ‘satisfaction with performance’ had somewhat better construct validity for all hypotheses tested (correlation with other participation measures, correlation with HR-QoL and discriminative ability across groups with different health states), ‘satisfaction with time’ seems less informative and redundant. On the other hand, the dimension ‘physical difficulty’, that was added later to the SRPQ, was found to have good correlations with external measures of participation and with HR-QoL, and discriminated particularly well across different health states. The dimension ‘satisfaction with performance’, as opposed to ‘physical difficulty’, was not found redundant as it correlated better with MCS and SWLS.

In AS, only one other study reported on participation in social roles. Davis et al (10), previously reported on the validity of the original SRPQ in Canada and included 44 patients with AS. It is to be noted that in this version the dimension ‘physical difficulty’ was not tested. The authors found comparable Cronbach’s α , acceptable test-retest reliability and confirmed redundancy for both satisfaction scales. Regarding comparison with other instruments measuring participation, it should be mentioned that Davis included different measures, and reported also moderate correlations of the SRPQ satisfaction scales with the Keele Assessment of Participation, which measures participation in roles ‘as and when you want it’, (29) and the Late Life Disability Instrument evaluating the frequency and limitations in performing life tasks (30). This adds to the validity of the SRPQ as in our study it needs to be recognised that the instruments to assess construct validity participation (SF-36 role domains and WHODASII participation dimensions) addressed only the dimension physical restrictions of participating, therefore somewhat limiting the validation of the ‘satisfaction’ dimensions.

By analysing and presenting the results of the dimensions of the SRPQ as averages of all 11 roles, information on clinimetrics and validity of the specific roles could be missed. Additional analyses showed that clinimetrics and the validity of these items did not differ overall from results based on average scores, and no clear pattern with regard to a specific role could be seen. As a first step to increase the feasibility of the SRPQ, the validity of the summary role ‘general participation’ was explored, and this showed that this single item had a high floor effect for role importance (very important) and somewhat lower (yet with acceptable reliability) similar correlations with external instruments, and appropriate discriminative ability across self-reported health states. Although ‘global’ constructs are informative as secondary end points, there is also a risk to loose information. Additional research, such as item response theory or factor analyses, could help in reducing the number of roles.

Some limitations need to be considered. First, the sample of patients unexpectedly included more females and the average age was higher than in most of the cross-sectional samples of patients with AS (1). Although it is known that AS is increasingly recognised in females (31), it cannot be excluded that some patients might in fact have non-radiographic axial spondyloarthritis (nr- axSpA) (which is also more frequent in females). Second, when

items in the SRPQ are not applicable there are theoretical different approaches to account for in the calculation of the dimension scores. In this study, it was chosen to adhere to the instruction of the original articles to allow for better comparison with the other publications on the validity of the SRPQ.

Third, when evaluating test-retest reliability, erroneously no external measure to evaluate stability of health was added. Overall, it is considered that a 2-week period in a chronic disease is considered an appropriate time interval to avoid a change in health, on the one hand, and avoid a recall bias on the other hand (20). More importantly, some aspects of the measurement properties have not yet been addressed. A priority is to define thresholds to enhance interpretability of the scores with regard to clinically important differences and patient acceptable participation state, and to study sensitivity to change. Finally, although the 'importance dimension' is considered relevant, a feasible approach to include this dimension in a global interpretation of social role participation is needed.

The relevance of this study is clear. The availability of a validated version of a Dutch version of the SRPQ will help researchers to gain more insight on participation as a health outcome, and the role of participation to directly or indirectly predict long-term satisfaction with life (happiness), vitality and healthy ageing, as well as resource utilisation and even mortality. Although relevant for patients, limitations in participation should not be a reason to change the pharmacological treatment, but should rather be a reason to consider non-pharmacological interventions comprising of lifestyle advice, education on coping strategies or the organisation of help from caretakers, friends or colleagues.

In summary, the Dutch SRPQ was understood well by the current sample of patients with AS. The version addressing the dimensions 'role importance', 'satisfaction with performance' and 'physical difficulty' showed acceptable reliability and validity. The general participation item of the SRPQ seems to be a good substitute for the 11 other roles. It can, therefore, be considered to be a more feasible way to measure social role participation.

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Appendix

Short Social Role Participation Questionnaire (sSRPQ) [verkorte vragenlijst deelname sociale rollen]

2017

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Opstellers van de verkorte vragenlijst: Oude Voshaar M., van Onna M., van Genderen S, van de Laar M., van der Heijde D., Heuf L., Spoorenberg A., Luime J., Gignac M., en Boonen A

Doel van de meting: Het beoordelen van het *belang* van deelname in diverse sociale rollen, de ervaren *moeilijkheden* bij deelname en de *tevredenheid* over de deelname in sociale rollen. De vragen zijn ontworpen om te worden beantwoord door individuele personen die hun gezondheid in het algemeen óf meer specifieke gezondheidsaandoeningen (bijv. gewrichtsproblemen gerelateerd aan artritis) in beschouwing nemen. Het woord ‘gezondheid’ kan worden vervangen door andere aandoeningen (bijv. gewrichtsproblemen’ of ‘multiple sclerose’).

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Score short form: Er worden afzonderlijk gemiddelde scores berekend voor de drie sociale roldimensies: 1) het belang van de rol; 2) moeite met iedere rol (d.w.z. beperkingen door gezondheid om de rol uit te voeren); 3) tevredenheid over het vermogen om deel te nemen in iedere rol zoals men zou willen.

Mensen brengen hun tijd op verschillende manieren door. Bij de volgende vragen noemen we verschillende sociale aspecten van het leven en vragen u hoe *belangrijk* ieder aspect voor u is, in hoeverre u door gezondheidsproblemen *moeilijkheden* ervaart en of u *tevreden bent over de manier waarop u deze rol uitvoert*.

U kunt antwoorden door bij de antwoordmogelijkheden aan te geven wat voor u van toepassing is.

1- sociale activiteiten

Belang In het algemeen	Helemaal niet be- langrijk	Enigszins	Redelijk	Erg	Heel erg belangrijk
1a. Hoe <u>belangrijk</u> is het voor u om sociale activiteiten te kunnen plannen of bijwonen (bijv. vrienden/familie op bezoek krijgen of uit eten/koffie drinken met vrienden, uitstapjes zoals een film bezoeken)?	1	2	3	4	5
Moeilijkheden Denkend aan uw gezondheid:	Helemaal geen moei- lijkheden	Enige moeilijk- heid	Redelijk veel moei- lijkheden	Erg veel moeilijk- heden	Niet moge- lijk
b. Hoeveel <u>moeilijkheden</u> ervaart u bij het plannen of bijwonen van sociale activiteiten (bijv. vrienden/familie op bezoek krijgen of uit eten/koffie drinken gaan met vrienden, uitstapjes zoals naar de film gaan)?	1	2	3	4	5
Tevredenheid	Helemaal niet tevreden	Enigszins	Redelijk	Erg	Heel erg tevreden
c. Hoe <u>tevreden</u> bent u met uw vermogen het soort sociale activiteiten dat u <u>zou willen</u> , te plannen of bij te wonen?	1	2	3	4	5

2- reizen of vakanties

Belang In het algemeen	Helemaal niet be- langrijk	Enigszins	Redelijk	Erg	Heel erg belangrijk
2a. Hoe <u>belangrijk</u> is het voor u om te kunnen reizen of op vakantie te kunnen gaan (bijv. vrienden/familie bezoeken, nieuwe plaatsen bezoeken, reizen per auto, vliegtuig, trein, bus etc.)?	1	2	3	4	5
Moeilijkheden Denkend aan uw gezondheid:	Helemaal geen moei- lijkheden	Enige moeilijk- heid	Redelijk veel moei- lijkheden	Erg veel moeilijk- heden	Niet moge- lijk
b. Hoeveel <u>moeilijkheden</u> ervaart u bij het reizen of op vakantie gaan	1	2	3	4	5
Tevredenheid	Helemaal niet tevreden	Enigszins	Redelijk	Erg	Heel erg tevreden
c. Hoe tevreden bent u met uw mogelijkheden om te reizen of het soort vakantie te hebben dat u zou willen?	1	2	3	4	5

3- betaald werk

Belang In het algemeen	Helemaal niet be- langrijk	Enigszins	Redelijk	Erg	Heel erg belangrijk
3a ₁ . Hoe <u>belangrijk</u> is het momenteel voor u om betaald werk te hebben?	1	2	3	4	5
3a ₂ . Heeft u op dit moment betaald werk	<input type="checkbox"/> ₁ Nee (ga naar vraag 4)		<input type="checkbox"/> ₂ Ja (ga naar vraag 3b)		
Moelijkheden Denkend aan uw gezondheid:	Helemaal geen moei- lijkheden	Enige moeilijk- heid	Redelijk veel moei- lijkheden	Erg veel moeilijk- heden	Niet moge- lijk
b. Hoeveel <u>moelijkheden</u> ervaart u met het uitvoeren van betaald werk?	1	2	3	4	5
Tevredenheid	Helemaal niet tevreden	Enigszins	Redelijk	Erg	Heel erg tevreden
c. Hoe <u>tevreden</u> bent u met de mogelijkheden om het <u>betaald werk</u> te verrichten <u>zoals u het zou willen</u> ? (bijv. een functie waarin u uw opleiding/vaardigheden kunt gebruiken)?	1	2	3	4	5

4- vervolgopleiding en of een cursus

Belang In het algemeen	Helemaal niet be- langrijk	Enigszins	Redelijk	Erg	Heel erg belangrijk
4a ₁ . Hoe <u>belangrijk</u> is het voor u om naar school te gaan, een vervolgopleiding en of een cursus te volgen? (bijv. full- of parttime, werkvaardigheden bijscholen, studeren voor een diploma of academische titel)?	1	2	3	4	5
4a ₂ . Gaat u op dit moment naar school of volgt een opleiding, cursus	<input type="checkbox"/> ₁ Nee (ga naar vraag 5)		<input type="checkbox"/> ₂ Ja (ga naar vraag 4b)		
Moelijkheden Denkend aan uw gezondheid:	Helemaal geen moei- lijkheden	Enige moeilijk- heid	Redelijk veel moei- lijkheden	Erg veel moeilijk- heden	Niet moge- lijk
b. Hoeveel lichamelijke <u>moelijkheden</u> ervaart heeft u bij het bezoeken van een school, vervolgopleiding of cursus?	1	2	3	4	5
Tevredenheid	Helemaal niet tevreden	Enigszins	Redelijk	Erg	Heel erg tevreden
c. Hoe <u>tevreden</u> bent u met uw vermogen om deel te nemen aan uw <u>studie, opleiding of cursus</u> op de manier die u zou willen?	1	2	3	4	5

5- intieme relaties

Belang In het algemeen	Helemaal niet belangrijk	Enigszins	Redelijk	Erg	Heel erg belangrijk		
5a ₁ . Hoe <u>belangrijk</u> is het voor u om een <u>intieme relatie/liefdesrelatie</u> te hebben?	1	2	3	4	5		
5a ₂ . Heeft u op dit moment een intieme relatie/liefdesrelatie met iemand (bijv. partner, echtgeno(o)t(e), geliefde)?	<input type="checkbox"/> ₁ Nee (ga naar vraag 6)		<input type="checkbox"/> ₂ Ja (ga naar vraag 5bd)				
Moelijkheden Denkend aan uw gezondheid:	Helemaal geen moei- lijkheden	Enige moei- lijkhed	Redelijk veel moei- lijkheden	Erg veel moeilijkhe- den	Niet moge- lijk	⁸⁸ Wil niet antwoor- den	
b. Hoeveel lichamelijke <u>moeilijkheden</u> ervaart u binnen uw intieme relatie/liefdesrelatie met uw partner, echtgeno(o)t(e) of geliefde?	1	2	3	4	5	88	
Tevredenheid	Helemaal niet tevre- den	Enigszins	Redelijk	Erg	Heel erg tevreden	Wil niet antwoor- den	Geen partner/ echt- genoot
c. Hoe <u>tevreden</u> bent u met uw mogelijkheden om met uw partner/ echtgeno(o)t(e) het <u>soort relatie</u> te hebben dat u <u>zou willen</u> ?	1	2	3	4	5	88	99

6- relatie met andere familieleden

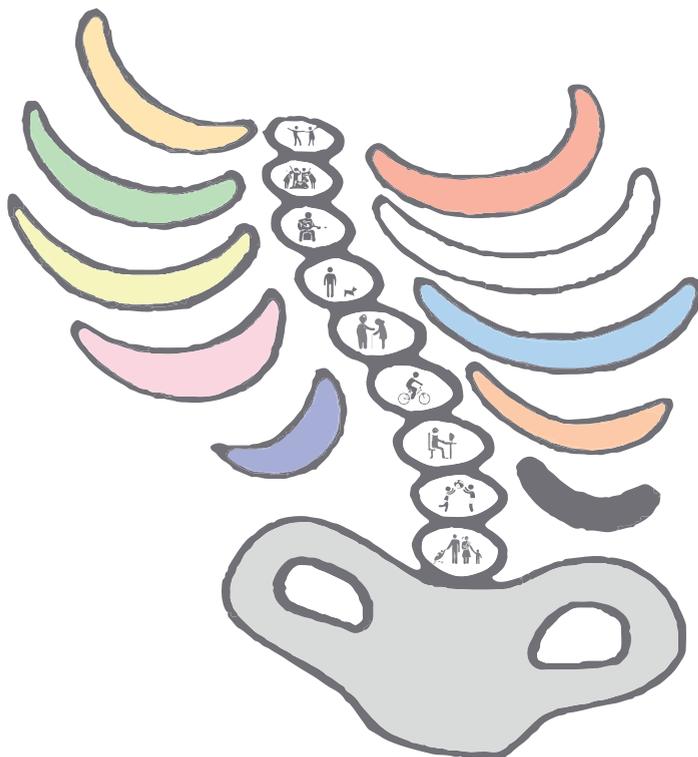
Belang In het algemeen	Helemaal niet be- langrijk	Enigszins	Redelijk	Erg	Heel erg belang- rijk	⁸⁸ Wil niet ant- woorden	Geen familie
6a. Hoe <u>belangrijk</u> is het voor u om een <u>relatie met andere familieleden</u> te hebben?	1	2	3	4	5	88	99
Moelijkheden Denkend aan uw gezondheid:	Helemaal geen moei- lijkheden	Enige moei- lijkheid	Redelijk veel moeilijkhe- den	Erg veel moeilijkhe- den	Niet moge- lijk		
b. Hoeveel <u>moeilijkheden</u> ervaart u bij het deelnemen aan activiteiten met andere familieleden?	1	2	3	4	5		
Tevredenheid	Helemaal niet tevreden	Enigszins	Redelijk	Erg	Heel erg tevreden		
c. Hoe <u>tevreden</u> bent u met uw mogelijkheden om met uw familieleden het <u>soort relatie te hebben dat u zou willen</u> ?	1	2	3	4	5		

Dank voor het invullen van deze vragenlijst

3

Social role participation in patients with ankylosing spondylitis. A cross-sectional comparison with population controls

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Abstract

Objective: Participation in social roles for persons with chronic disease is important for their quality of life, but interpretation of the data on participation is difficult in the absence of a benchmark. This study aimed to compare social role participation in patients with ankylosing spondylitis (AS) to population controls using the Social Role Participation Questionnaire (SRPQ).

Methods: There were 246 AS patients and 510 population controls who completed the SRPQ, which assesses participation in 11 roles (with scores ranging 1–5) across 4 dimensions (importance, satisfaction with performance, satisfaction with time, and physical difficulty), and additionally ranked their 3 most important roles. The ranking of role importance, the SRPQ dimension scores, and the gap between importance and satisfaction with performance of roles were compared between patients and controls.

Results: Patients (62% male; 51±12 years) and controls (70% male; 42±15 years) ranked intimate relationships, relationships with children/stepchildren/grandchildren, and employment as the most important roles. Compared to controls, patients gave higher scores on the SRPQ to importance (3.75 versus 3.43), but reported lower satisfaction with performance (3.19 versus 3.58) and greater physical difficulty (3.87 versus 4.67) ($P \leq 0.05$ for all). The largest differences in gaps between importance and satisfaction with performance for patients compared to controls were seen in the physical leisure, hobbies, and traveling and vacation categories, in which patients assigned higher importance but reported especially low satisfaction.

Conclusion: As society places increasing emphasis on individual responsibility to participate fully in social roles, the current data suggest that health care providers should pay more attention to participation restrictions experienced by patients with AS.

Introduction

Participation in social roles is essential for individuals in all stages of their lives. As a result, social role participation is increasingly considered a relevant outcome in observational studies and in intervention programs that intend to improve the overall functioning and health of patients with chronic diseases (1-3). Available literature on patients with ankylosing spondylitis (AS) and the impact of the disease on the participation in several social roles provides substantial evidence that patients experience restrictions in work (1), as well as more limited evidence of restrictions in intimate relationships, hobbies, leisure, and community life (3-5). However, research examining the impact of AS on a broad range of roles at the same time, and addressing the relative value or importance of different roles in relation to experienced difficulties, is missing. Part of the challenge in participation research is figuring out how to conceive of social role participation and how to measure it. Participation covers multiple roles, and each role can be assessed across several dimensions, such as satisfaction, difficulty, time spent, and role performance, which makes it a complex construct to measure and to report (2, 6).

The Social Role Participation Questionnaire (SRPQ) is a promising instrument, as it assesses participation in 11 roles across multiple dimensions: importance, satisfaction with time, and satisfaction with performance (5, 7). In a subsequent study, it was suggested that “satisfaction with time” be removed, because alongside “satisfaction with performance,” it was redundant (6). Finally, including “physical difficulty” as an additional and relevant dimension was proposed (8). In Canadian patients with AS (n 544), the original SRPQ was found to be valid and reliable and showed that patients highly valued participation in diverse social roles, but were not satisfied with their performance, nor with the time they spent in roles (5).

To provide a better understanding of the interpretation of participation by patients with AS, comparison with the general population is needed. Without such a reference, it is difficult to conclude whether problems in participation are attributable to AS, or are similarly experienced by population controls. The aim of the present study was to examine and compare the level of social role participation in AS patients with population controls. We hypothesized that patients and controls would rate “importance” and “satisfaction with time” similarly, but that patients would rate “physical difficulty” and “dissatisfaction with performance” higher, resulting in a greater importance–satisfaction gap than control subjects.

Subjects and methods

Subjects

Patients were recruited as part of a multicenter cross-sectional study, the Social Participation in Ankylosing Spondylitis Study. Patients from 6 hospitals in The Netherlands, who were at least 18 years old, had registered with AS according to Dutch diagnosis-related groups or similar patient lists, and in whom the modified New York criteria was confirmed by the

treating rheumatologist, were invited by letter (9). Control subjects were recruited from an open national online panel of the research institute Ipsos. Based on knowledge of cross-sectional samples in AS, with recruitment we aimed to achieve a control sample of adults with an average age of 42 years and a male:female distribution of 3:1 (10). Control subjects were excluded if they were indicated to have any musculoskeletal disorders or were not familiar with the Dutch language (11). The study was approved by the ethics committee of Maastricht University Medical Center, and patients provided written informed consent.

Assessments

All subjects were asked to complete the online survey. Sociodemographic background questions addressed age, sex, and education level, and general health was assessed by the 36-item Medical Outcomes Study Short Form 36 (SF-36), which has Physical Component Summary (PCS) and Mental Component Summary (MCS) scores (range 0–100, with higher scores reflecting better health) (12).

Social role participation was assessed using the validated Dutch version of the SRPQ, which assesses the influence of health on 11 roles and a “general participation” role for each of the 3 initial participation dimensions (importance and satisfaction dimensions, as well as the physical difficulty dimension, which was added later by the developers) (8, 13). Each role was rated on a 5-point Likert scale: the dimension importance ranged from 1 (not at all important) to 5 (extremely important), and for both satisfaction dimensions from 1 (not at all satisfied) to 5 (extremely satisfied), and for the physical difficulty dimension from 1 (unable to do) to 5 (no difficulty) (8). Five roles could be rated as not applicable (having a partner/intimate relationship, having children/stepchildren/grandchildren, other family, employment, and current schooling or education), and in such cases, only the dimension importance was rated. Ratings on the individual roles can be averaged into dimension scores. To deal with non-applicable roles, for the physical difficulty dimension, it was assumed that there were no restrictions experienced if the role was not applicable, while for the satisfaction dimensions, scores were averaged over the number of applicable roles, when subjects participated in at least 9 of 11 roles (8). Finally, the SRPQ allows for calculation of a participation gap score, which is the difference between importance and satisfaction with role performance, in those for whom the role was applicable. After completion of the SRPQ, patients were asked to rank which role was most, second, and third most important to them.

Statistical analysis

Statistical analysis was performed using PASW Statistics 20 (SPSS) and Stata, version 12. After we checked for normality of the data, comparisons between patients and controls were performed with independent *t*-tests (age, SF-36 PCS), Mann-Whitney U test (SF-36 MCS), or chi-square test (sex, applicability of role). Before making comparisons regarding the SRPQ and ranks, data of controls were first standardized. Based on the age (in 10-year increments) and sex distribution of patients, indirect standardization (for ranks) was performed with age and

sex strata, and direct standardization as well, by using a standardized weight (for the SRPQ). Subsequently, 95% confidence intervals around the difference of means were calculated. *P* values less than or equal to 0.05 were considered statistically significant.

Results

Of 740 invited patients with AS, 296 (40%) agreed to participate, of which 246 (83%) completed the questionnaire. Of 2767 invited controls, 784 (28%) agreed to participate, of which 510 (65%) completed the questionnaire. Controls were more often male (70% versus 62%) and younger (mean age $42 \pm \text{SD } 15\text{y}$) versus ($51 \pm \text{SD } 12\text{y}$) (appendices table 1). Further, controls more often had a higher professional education or university degree (49% versus 33%), were more often employed (78% versus 57%), had children/stepchildren/grandchildren less often (55% versus 75%), and were more often following an educational track (23% versus 16%) ($P \leq 0.05$ for all). Finally, controls scored better on the PCS (mean $53.5 \pm \text{SD } 8.7$) versus ($38.7 \text{ SD } \pm 10.1$), but similar to patients on the MCS (mean $49.6 \pm \text{SD } 11.8$) versus ($49.2 \pm \text{SD } 12.8$).

Most important roles

When ranking roles according to importance, the first, second, and third most important roles for both patients and controls were intimate relationships, relationships with children/stepchildren/grandchildren, and employment (Table 1). Patients more frequently reported physical leisure and travel/vacation among their 3 most important roles, whereas controls more often ranked social events in their top 3.

Social role participation in patients with AS versus population controls

Table 2 shows the scores of each role as well as of the average of all 11 specific roles of the SRPQ, across the different dimensions, between patients and controls. Based on the averages of 11 roles, patients reported a higher importance, were less satisfied with their performance, and experienced more physical difficulties, but did not differ from controls in satisfaction with time. The largest difference between patients and controls was seen in the dimension physical difficulty, where patients indicated more physical difficulties. When investigating individual roles instead of averages of all roles, the similar patterns of higher importance but more physical difficulty and less satisfaction with performance among patients compared to controls were observed. The role “physical leisure” was much more important for patients than for controls. Exceptions for a significantly higher importance for patients were observed in the roles intimate relationships and employment. However, when limiting the analyses of importance to those for whom the role was applicable, the mean \pm SD employment score became much higher in patients (4.32 ± 0.69) and significantly different from controls (3.93 ± 1.95) (appendices table 2). The largest differences in the dimension ‘physical difficulty’ between patients and controls were observed for physi-

cal leisure, employment, and travel and vacation. The general participation item of the SRPQ showed the same patterns of differences between patients and controls for each dimension, as the scores were based on the average of 11 roles.

Table 1. Frequency that a role was the most important or part of a patient's top 3 of most important roles

Roles	Ankylosing spondylitis (N=246)		Controls (N=510)	
	Most important	In top 3	Most important	In top 3
<i>Intimate relationships*</i>	45.3%	63.8%	42.7%	67.3%
<i>Relationship (grand/step) children*</i>	21.0%	54.5%	20.8%	43.1%
<i>Employment*</i>	11.1%	38.6%	16.6%	43.1%
<i>Social events</i>	8.2%	32.9%	10.9%	37.5%
<i>Physical leisure activities</i>	5.3%	36.6%	3.1%	26.5%
<i>Travel or vacation</i>	3.3%	30.1%	1.8%	26.7%
<i>Hobbies</i>	3.7%	18.3%	2.8%	21.2%
<i>Relationship other family*</i>	0%	8.9%	0%	13.3%
<i>Community, religious, cultural</i>	1.2%	9.8%	0.7%	11.6%
<i>Casual contact with others</i>	0%	4.9%	0%	4.3%
<i>Education*</i>	0.4%	1.6%	0.2%	4.7%

Ranks for controls are standardized for age and gender difference compared to patients.

* Intimate relationships (79.9% of patients/ 75.4% of controls has a partner), Relationships (grand/step-) children (74.8% of patients/ 55.3% of controls has grand/step-) children), Relationship other family (97.6% of patients/ 97.5% of controls has other family), Education (16.3% of patients/ 22.9% of controls follows a form of education), Employment (56.9% of patients/ 77.7% of controls is employed)

The gaps between importance and satisfaction with performance in all roles are represented graphically for patients and for controls separately (Figure 1). Within patients, the 5 largest importance–satisfaction gaps were found in the roles traveling or vacationing, physical leisure, work, relationships with children/stepchildren/ grandchildren, and in planning/attending social events. Compared to controls, patients had significantly greater gaps for all roles. The 5 largest differences in the importance-satisfaction gaps between patients and controls were observed in the roles physical leisure, hobbies, traveling or vacationing, planning/attending social events, and casual contacts.

Table 2. Score on SRPQ (mean, SD) for the dimension scores (11 roles) and the individual social roles separately for patients (n=246) and population controls (n=510)

SRPQ Roles	Role Importance		Satisfaction time		Satisfaction time		Satisfaction performance		Physical difficulty	
	AS patients [#]	Controls [#]	AS patients	Controls	AS patients	Controls	AS patients	Controls	AS patients	Controls
Average 11 roles	3.75 (0.52)	3.43 (0.61)*	3.22 (0.71)	3.32 (0.78)	3.19 (0.75)	3.58 (0.85)*	3.87 (0.66)	4.67 (0.61)*		
General item	4.26 (0.73)	3.98 (1.01)*	3.26 (0.91)	3.51 (1.03)*	3.01 (0.99)	3.53 (1.19)*	3.50 (0.86)	4.56 (0.86)*		
Intimate relationship	4.11 (0.96)	3.97 (1.41)	3.68 (0.96)	3.76 (1.20)	3.64 (1.03)	3.87 (1.15)*	3.96 (0.86)	4.68 (0.73)*		
Relationship children	3.97 (1.37)	3.76 (1.74)*	3.46 (0.98)	3.54 (1.08)	3.55 (1.04)	3.87 (1.06)*	3.68 (0.86)	4.43 (1.20)*		
Employment	3.48 (1.31)	3.42 (1.62)	3.19 (1.04)	3.70 (1.46)*	3.24 (0.91)	3.85 (1.41)*	3.50 (0.81)	4.68 (1.02)*		
Social events	4.11 (0.84)	3.71 (1.08)*	3.16 (0.97)	3.22 (1.19)	3.15 (1.04)	3.57 (1.14)*	3.99 (0.86)	4.75 (0.69)*		
Physical leisure	3.78 (0.98)	3.20 (1.37)*	2.89 (1.02)	2.85 (1.23)	2.71 (1.03)	3.14 (1.34)*	3.29 (0.89)	4.33 (0.98)*		
Hobbies	3.87 (0.92)	3.43 (1.21)*	3.01 (0.95)	3.21 (1.08)*	2.95 (1.00)	3.55 (1.21)*	3.92 (0.81)	4.81 (0.58)*		
Other family	3.90 (0.96)	3.59 (1.19)*	3.42 (0.86)	3.37 (1.05)	3.41 (0.81)	3.56 (1.09)*	4.05 (0.83)	4.80 (0.69)*		
Travel or vacation	4.13 (0.93)	3.86 (1.13)*	2.94 (1.10)	3.18 (1.19)*	2.91 (1.09)	3.45 (1.32)*	3.55 (0.98)	4.69 (0.87)*		
Community	3.00 (1.23)	2.74 (1.64)*	3.06 (1.03)	3.32 (1.23)*	3.02 (1.04)	3.62 (1.23)*	3.94 (0.90)	4.70 (0.89)*		
Casual contact	3.80 (0.90)	3.44 (1.16)*	3.41 (0.88)	3.38 (1.03)	3.45 (0.92)	3.69 (1.10)*	4.59 (0.66)	4.88 (0.57)*		
Education	2.36 (1.20)	1.93 (1.33)*	3.08 (0.95)	3.24 (1.33)	3.08 (0.91)	3.67 (1.69)*	4.02 (0.67)	4.94 (0.26)*		

Scores range from 1 (not at all important/not at all satisfied/unable to do) to 5 (extremely important/extremely satisfied/no difficulty); scores of controls adjusted for differences in age and gender distribution compared to patients; * Significantly different compared with patients (P≤0.05) # Scores of the dimension 'role importance' are provided for all subjects, regardless if the role was applicable

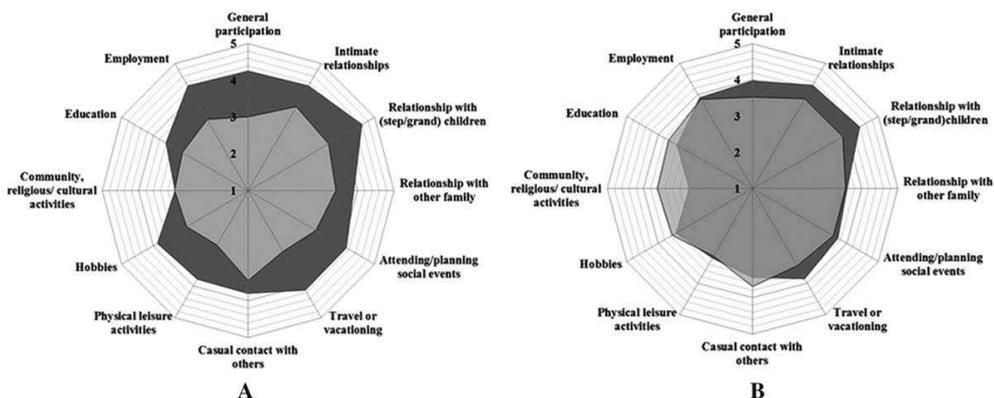


Figure 1. A, Importance of role domain (darkly shaded) in patients with ankylosing spondylitis (AS), where 1= not at all important and 5= extremely important, and satisfaction with role performance (lightly shaded) in patients with AS, where 1= not at all satisfied and 5= extremely satisfied. B, Importance of role domain (darkly shaded) in controls, where 1= not at all important and 5= extremely important, and satisfaction with role performance (lightly shaded) in controls, where 1= not at all satisfied and 5= extremely satisfied.

Discussion

This study is the first that directly compared participation in a broad range of social roles in patients with AS and controls. Patients and controls ranked the same roles as most important. Yet patients provided higher importance scores in almost all roles while reporting more physical difficulties and a lower satisfaction with performance, resulting in larger gaps between importance and satisfaction with performance in several social roles when compared to population controls.

Intimate relationships, relationships with children/stepchildren/grandchildren, and employment were identified as the most important social roles by patients as well as controls, indicating that regarding the current selection of measured roles, patients and healthy subjects value the same types of roles as most important in their lives. Moreover, while patients ranked physical leisure more often in their top 3 of most important roles, controls valued participation in social events more. In contrast to what we hypothesized, patients reported higher importance for almost all roles, compared to controls. Response shift is a likely explanation for this observation. Confrontation with health problems and resulting restrictions in social roles apparently cause patients to reevaluate the importance of social role participation in life (14). While this was in general a positive valuation, patients may also devalue or minimize roles in which they no longer participate, as could be seen for the employment category, when this was no longer applicable. The largest difference in importance between patients and controls was seen in the role physical leisure. This is less surprising, as patients likely experience the benefits of physical activity and exercise on pain and stiffness in the affected joints, explaining the shift toward higher importance

for physical leisure. Our findings in AS are different from those of a previous study of 177 middle and older-aged patients (≥ 40 years) with osteoarthritis (OA), who rated their roles at a similar level of importance as healthy controls (8). While this could be an effect due to age, the majority of the 197 patients in the present study (80.1%) were also ≥ 40 years old. Explaining the differences in observation in AS and OA is complex and likely the result of an interplay between disease burden, contextual factors in the environment, and the personalities of the patients, including reevaluation. Future research on patterns for differential evolution of the importance of roles in life between healthy persons and persons with different chronic diseases might be interesting.

More in line with our expectations and the literature (8), patients were less satisfied with performance and experienced more physical difficulty than controls. Patients experienced the most physical difficulty in the roles physical leisure, employment, and traveling and vacationing, where the SRPQ scores were lowest. Interestingly, for the satisfaction with time dimension, differences with controls were less apparent, and equal satisfaction (or dissatisfaction) was observed in various social roles. Although population controls are also less satisfied with the time spent in several roles, the dissatisfaction is likely caused by different reasons than those in patients. The lower ability of satisfaction with time to discriminate between patients and controls adds to the validity of the previous decision (5, 13) to remove “satisfaction with time” from the SRPQ, as it does not provide additional information.

The higher importance and lower satisfaction with the performance of roles in patients resulted in a greater participation gap for the former group compared to healthy individuals. Within patients, these gaps were largest for roles that were most important to them (relationships with children/grandchildren/stepchildren and employment), but also for traveling and vacationing, physical leisure, and planning/attending social events. Compared to controls, overall the type of roles with gaps observed between patients and controls were largely the same as those observed within the patients-only group. However, the gap difference in the roles that were most important, compared to controls, had a slightly different order, with the largest gap difference seen for physical leisure and hobbies, followed by travel/vacation, planning/attending social events, and in casual contacts. Studying participation gaps is a novel approach to examining participation and provides insight into what patients miss or desire in their lives. This might be particularly helpful to clinicians in treating patients, as well as in designing interventions that target individual patient needs. While society puts increasing emphasis on the individual’s responsibility to participate fully in social roles, the current data suggest that health care providers should pay more attention to participation restrictions experienced by patients with AS.

Some limitations of the study need to be considered. Participants were required to have internet access. An informal survey in our department already some years ago found that almost 90% of AS patients had internet access, and the percentage is likely similar in the same age group in the general population. Also, response rates were low, but likely not

different from those of many other survey studies. Further, the sample of patients unexpectedly included more females and was older than we expected based on our knowledge of other cross-sectional samples (10). Some research suggests that AS is increasingly being diagnosed among women, but the possibility that a sub-group of patients had non-radiologic axial spondyloarthritis cannot be excluded (15). This might influence external validity. However, as we adjusted for the small differences in age and sex, it will not have affected the internal validity of this study. While in patients the importance–satisfaction gap was quite large for several roles, no formal data are available as yet about interpretability and clinical relevance. Future research on important difference and acceptable state could allow research on participation to move forward. While some mechanisms to explain differences in participation between patients and controls have been proposed earlier in the discussion, the cross-sectional design of the study warrants caution with regard to drawing conclusions on causality.

In summary, patients with AS and controls rank the same roles as most important. However, patients value the importance of almost all roles higher while experiencing more physical difficulties and less satisfaction with their performance. These findings suggest that clinicians and researchers should pay more attention to social role participation.

Acknowledgements

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Appendices

Table 1. Characteristics of the patients with AS and healthy control subjects

	Patient with AS (N=246) N (%) or mean (SD) [min - max]	Control (N=510) N (%) or mean (SD) [min - max]	<i>p</i>
Sex (Males)	153 (62%)	355 (70%)	0.04
Age (years)	51 (12) [24 - 79]	42 (15) [18 - 85]	<0.001
Higher professional education or university (n (%))	81 (33%)	248 (49%)	<0.001
SF-36; physical component score	38.7 (10.1) [5.2 - 61.9]	53.5 (8.7) [18.1 - 73.5]	<0.001
SF-36; mental component score	49.2 (12.8) [10.9 - 70.5]	49.6 (11.8) [0.5 - 68.5]	0.49
<i>SRPQ roles that are optional;</i>	<i>N (%)</i>	<i>N (%)</i>	<i>N (%)</i>
Has intimate relationship	187 (79%)	371 (73%)	0.11
Has (grand/step-) children	184 (75%)	279 (55%)	<0.001
Has other family	239 (98%)	497 (98%)	0.81
Employed	140 (57%)	396 (78%)	<0.001
Following education	40 (16%)	117 (23%)	0.04

Table 2. Score on the SRPQ dimensions role importance (mean, SD) of 5 roles that were optional for AS patients (n=246) and population controls (n=510) (scores of controls adjusted for differences in age and gender distribution compared to patients)

<i>SRPQ Roles</i>	AS patients (when role applies)	AS patients (when role not applies)	Controls (when role applies)	Controls (when role not applies)
Intimate relationship	4.33 (0.68)	3.44 (1.23)	4.31 (0.96)	2.76 (1.89)*
Relationship (grand/step-) children	4.55 (0.68)	1.85 (1.02)	4.38 (0.84)*	1.69 (1.82)
Employment	4.32 (0.69)	2.46 (1.65)	3.93 (1.95)*	2.28 (0.92)
Relationship with other family	3.94 (0.93)	1.98 (0.66)	3.63 (1.11)	2.02 (0.11)
Education	3.63 (0.89)	1.67 (0.72)	3.40 (1.30)	2.08 (1.39)*

Scores range from 1 (not at all important) to 5 (extremely important)

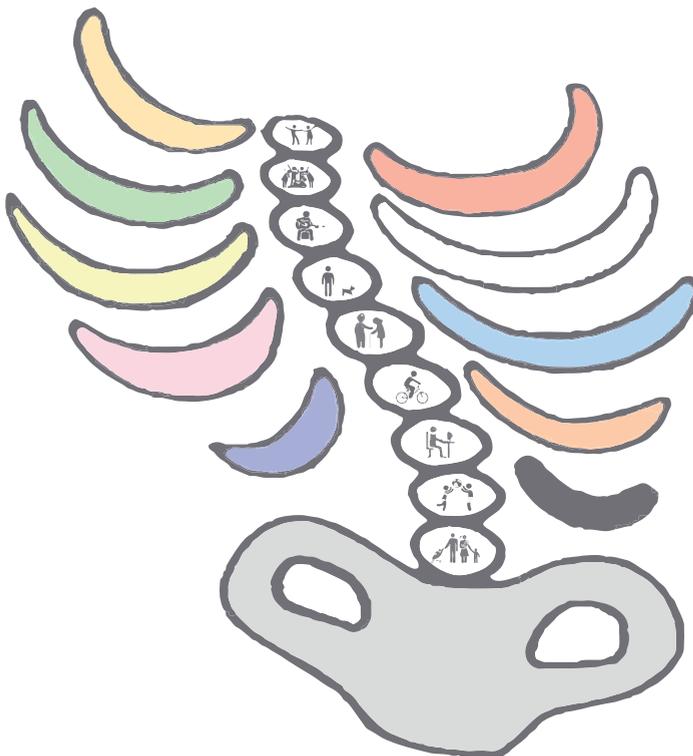
* Significantly different compared with patients within the same category ($P \leq 0.05$)

Intimate relationships (79.9% of patients and 75.4% of controls has a partner); Relationships (grand/step-) children (74.8% of patients and 55.3% of controls has (grand or step) children); Relationship other family (97.6% of patients and 97.5% of controls has other family); Education (16.3% of patients and 22.9% of controls follows a form of education); Employment (56.9% of patients and 77.7% of controls is employed)

4

Social role participation and satisfaction with life: a study among patients with ankylosing spondylitis and population controls

van Genderen, S., Plasqui, G., van der Heijde, D., van Gaalen, F., Heuft, L., Luime, J., Spoorenberg, A., Arends, S., Lacaille, D., Gignac, M., Landewé, R., Boonen, A. Social role participation and satisfaction with life: A study among patients with ankylosing spondylitis and population controls. *Arthritis Care & Research*. 2018;70(4):600-607



Abstract

Objective: Participation in society of persons with chronic diseases receives increasing attention. However, little is known which components of participation are most relevant to life satisfaction. This study examines the association between several aspects of social role participation and satisfaction with life (SWL) in patients with ankylosing spondylitis (AS) compared to population controls.

Methods: In a cross-sectional study, participants completed the Social Role Participation Questionnaire (SRPQ) and SWL scale. The SRPQ assesses several dimensions of participation (importance, satisfaction with performance, satisfaction with time and physical difficulty) in 11 roles representing three domains (interpersonal relations, leisure and work). For individuals with AS and controls, the association between role-domains and SWL were examined using linear regression for each participation dimension separately, in the total and the employed population, adjusting for age, gender, education and income.

Results: 246 AS patients (age: 51±12 years; 62% males; disease duration: 17±12 years) and 510 controls (age: 42±15 years; 70% males) were included. Patients were more frequently (extremely) dissatisfied with life (17.9% vs 8.6%; $p<0.05$). In the total and the employed population, less physical difficulty and higher satisfaction with *interpersonal relations* and *leisure* were associated with higher SWL and this was somewhat stronger in patients than in controls ($p<0.1$). In employed controls but not in employed patients, satisfaction with *work* was independently associated with SWL.

Conclusion These findings highlight the importance to support persons with AS in ameliorating social role participation, particularly in areas like close relationships and leisure activities, which are typically ignored in treating AS.

Introduction

In an era where patient centered care is being implemented in healthcare, social role participation of individuals with chronic diseases receives increasing attention from health professionals. The relevance of social role participation is supported by studies revealing that patients identified several social roles as ‘valued life activities’ (17-20, 66). However, measuring and operationalizing participation is challenging given that participation in social roles covers multiple domains and can be assessed across various dimensions such as role importance, satisfaction or, more typical in health-outcome research, difficulty with roles (18, 20).

The Social Role Participation Questionnaire (SRPQ), asks respondents to rate their participation in 11 specific (e.g., employment, intimate relationships, physical leisure) and one general participation role, across four dimensions: ‘importance’, ‘satisfaction with time’, ‘satisfaction with performance’ and ‘physical difficulty’ (33, 67, 68). Previous studies in individuals with Ankylosing Spondylitis (AS) found that patients rated most social roles as important, and showed that role salience (i.e., importance) was often higher compared to population controls. In addition, patients with AS reported to experience more physical difficulties when participating in roles and were less satisfied with their ability to perform in roles compared to population controls (69). Clear associations between the SRPQ and health outcomes such as disease severity, depression and even healthcare utilization have been reported (33, 70). However, before recommending participation in social roles is a relevant outcome in AS, it is also important to understand the influence of social roles on patient’s overall satisfaction with life.

From the perspective of several philosophies, an individual strives to achieve happiness or satisfaction with life (SWL). The Satisfaction With Life Scale (SWLS) was developed with the specific aim “to assess satisfaction with the respondent’s life as a whole” (71, 72). The questionnaire contains five items and has been applied in several populations of healthy and chronically diseased people (38, 73). As such, the SWLS offers the opportunity to evaluate the contribution of social role participation to life satisfaction, and results could support to the validity of evaluating participation in AS in research and clinical practice. When social role participation is valuable for SWL of healthy persons (69), it would be additionally insightful to understand whether social role participation is differently associated with SWL in patients compared to persons without chronic diseases. While an earlier study indicated that patients attached greater importance to participation in social roles but experienced less satisfaction and more difficulties, it could well be that participation is even more important for patients than controls when examining overall life satisfaction. Evidence on the (differential) impact of social role participation on SWL would further support efforts to pay more attention to social role participation in the daily care of the patients.

The aim of this study was therefore to investigate the relation between social role participation and SWL in patients with AS, and to understand whether this relation is different

when compared to population controls. It was hypothesized that specifically satisfaction with performance for each role domain would be associated with SWL, and that these associations would be stronger in patients compared to controls.

Methods

Subjects

Patients were recruited as part of a multicenter cross-sectional questionnaire study, the Social Role participation in Ankylosing Spondylitis Study (SPASS). Patients older than 18 years that were registered with AS in the 'diagnosis related codes' or other diagnoses list in six hospitals in the Netherlands, whose diagnosis was confirmed by the treating rheumatologist, and who were able to understand Dutch were invited by letter. Control subjects were recruited, parallel to the patient recruitment, from a national online open panel of the research institute Ipsos (Ipsos, Amsterdam, the Netherlands). To ensure comparability between groups and based upon the knowledge of patient characteristics of previous published observational studies in AS, the control group was requested to be older than 18 years with a group-average of 42 years, and have a male/female distribution of 3:1 (1). Control subjects were excluded if they indicated to have any musculoskeletal disorder or were not familiar with the Dutch language (74). The study protocol was approved by the ethics committee of Maastricht University Medical Center (MEC azM/UM) and written informed consent was obtained from all patients.

Assessments

The online survey contained first questions on demographic background including age, gender, highest achieved educational level (five levels, dichotomized into having achieved a higher professional education or university degree, or not) and yearly income (eight categories ranging from ≤ 20.000 to ≥ 140.000 ; with an increase of $\text{€}20.000$). Social role participation was assessed using the validated extensive Dutch version of the *SRPQ* which assess the influence of health on four participation dimensions being; 'role importance', 'satisfaction with time', 'satisfaction with performance' and 'physical difficulty', across 11 social roles and one general participation role. For the latter item, the respondent is asked to provide scores 'while considering all previous social roles'. Each role in each dimension can be scored on a fivepoint Likert scale (1: not at all important/not at all satisfied/unable to do to 5: extremely important/extremely satisfied/no difficulty) (67). For five roles, subjects can indicate the role is not applicable: having a partner/intimate relationship, having (step/grand) children, having relationships with other family, employment and current educational status (i.e. following an educational course of professional relevance).

For further analyses, the SRPQ roles were classified into three domains; *interpersonal relations* (intimate relationships, (grand/step) children, other family, informal contacts,

social activities and religious, cultural and community activities), *leisure* (physical leisure activities, hobbies, travelling and or vacationing) and *work* (employment). Separate scores for each dimension were calculated for the three role-domains based on averages of applicable roles. For *interpersonal relations* and *leisure* the domain scores were only calculated if subjects indicated to participate in at least 2 roles.

To assess general health, the *MOS Short Form Health Survey 36* (SF-36) was included. The SF-36 contains 36 items and has two summary scores, the Physical Component Summary (PCS) and Mental Component Summary (MCS) scores. Both domain and summary scores range from zero to 100, higher scores reflecting less difficulties/health problems (29).

The SWLS asks respondent to assess their agreement with 5 general statements related to feelings about life in general e.g. (“In most ways my life is close to my ideal.”) (“I am satisfied with my life”). Each item which can be rated on a Likert scale (1 not at all agree to 7 totally agree). The total score ranges from 5 to 35, higher scores indicating more satisfaction with life (71, 75). Validated thresholds have been computed by the developers, which classify scores ≤ 9 as ‘extreme dissatisfaction’, from 10 to 14 as ‘dissatisfaction’, from 15 to 19 ‘slight dissatisfaction’, 20 as ‘neutral life satisfaction’, from 21 to 25 as ‘slight satisfaction’, from 26 to 30 ‘satisfaction’, and from 31 to 35 as ‘extreme satisfaction with life’ (76).

Patients additionally reported the date of diagnosis, whether they were currently treated with a biological (anti-TNF α), and completed the Bath Ankylosing Spondylitis Disease Index (BASDAI) and the Bath Ankylosing Spondylitis Functioning Index (BASFI). The BASDAI contains six questions assessing different aspects of disease activity in AS (total score ranging from zero to 10; higher values indicating higher disease activity) (77). The BASFI determines the level of normal daily physical functioning of people with AS across ten items, each representing different activities (total score ranging between zero to 10; higher values indicating more disabilities) (41).

Statistical Analyses

Characteristics of the study sample were described using means (\pm SD) or proportions. Characteristics between patients and controls were compared by independent t-tests (SWLS, age, SF-36, PCS), Mann-Whitney U (SF-36 MCS) or Chi-square (gender, being involved in specific social roles, being (extremely) (dis)satisfied with life) depending on the nature of the data.

Linear regression analyses were performed for patients and controls separately, using the scores on the SWLS as outcome and participation in the three social role domains as the main explanatory variables. As the different participation dimensions represent partially overlapping constructs, with correlations between dimensions ranging from moderate (Spearman rho ≥ 0.4) to high (Spearman rho ≥ 0.75), the influence of each participation dimension was explored in a separate model. Within each dimension, the 11 social roles were combined into three larger social role domains, representing the average of the interpersonal roles, the average of the leisure roles, and employment. The three participa-

tion domains were added to a basic regression model containing age, gender, education and income, as these were considered relevant determinants and and/or potential confounders. Since a substantial proportion of subjects were not employed, and thus had a missing score for the domain 'participation in work', analyses were performed first in the total sample while ignoring the work domain, and secondly in the employed subsample by considering all three domains. To understand whether the general participation role would provide the same information as the three distinguished domains of social role participation, the models were repeated for each dimension of the general participation role instead of the domains (interpersonal relations, leisure and work) in the total populations of patients and controls. The fit of the models with and without participation domains and between patients and controls were compared based on the explained variance (R^2). To examine whether the changes in R^2 were statistically significant ($P = \leq 0.05$) when entering the participation domains or the general participation role to the model, the F-test was used. To test statistically whether social roles were differentially associated with SWL in patients and controls, the interaction terms 'group (AS vs controls)*social role domain' were tested. An interaction was considered significant if $p < 0.1$. Statistical analyses were performed using PASW Statistics 20 (SPSS, Chicago, IL, USA).

Results

Samples. Of the 740 patients invited, 296 (40%) agreed to participate and 246 (83%) completed the questionnaire. Of the 2767 controls invited, 784 (28%) agreed to participate and 510 (65%) completed the questionnaire. Characteristics of both patients and controls are given in table 1. Controls were more often male and were younger. In addition, a larger proportion had achieved a higher educational or university degree, was employed and had a yearly income $\geq \text{€}40,000$. Moreover, a lower proportion of controls had step- or grandchildren. Patients reported an average BASDAI of 4.4 (± 2.3) and BASFI of 4.2 (± 2.5), and 50% was treated with a TNF- α inhibitor. Compared to controls, patients reported lower physical health but similar mental health.

On average, patients scored significantly lower on SWL compared to controls (22.2 (± 7.7) vs. 24.7 (± 6.6)). A larger proportion of patients were (extremely) dissatisfied with life (17.9% vs 8.6%), while controls were more frequently (extremely) satisfied (52.8% vs 37.4%) (Figure 1) (76).

Table 1. Characteristics of the patients with ankylosing spondylitis and population controls

	<i>Patients with AS (N=246)</i> mean ± SD [min - max]	<i>Controls (N=510)</i> mean ± SD [min - max]	<i>p</i>
Gender (Males)	153 (62%)	355 (70%)	0.04
Age (years)	51 ± 12 [24 - 79]	42 ± 15 [18-85]	<0.001
Education level (n, %) (higher professional or university)	81 (33%)	248 (49%)	<0.001
Income ≤ 40.000 € year (n, %)	150 (61%)	184 (43%)*	<0.001
Diagnosis duration (years)	16.8 ± 11.8 [5 - 44]	-	-
SF-36 PCS	38.7 ± 10.1 [5.2 - 61.9]	53.5 ± 8.7 [18.1-73.5]	<0.001
SF-36 MCS	49.2 ± 12.8 [10.9 - 70.5]	49.6 ± 11.8 [0.5 - 68.5]	0.49
SWLS	22.2 ± 7.7	24.7 ± 6.6	<0.001
<i>SRPQ roles potentially not applicable</i>			
Has intimate relationship (n, %)	187 (79%)	371 (73%)	0.11
Has (step/grand) children (n, %)	184 (75%)	279 (55%)	<0.001
Has other family (n, %)	239 (98%)	497 (98%)	0.81
Employed (n, %)	140 (57%)	396 (78%)	<0.001
Following education (n, %)	40 (16%)	117 (23%)	0.04

* missing data with 425 complete cases

SF-36, 36 item Short Form health survey physical component score and mental component score; SRPQ, Social Role Participation Questionnaire; SWLS, Satisfaction With Life Scale

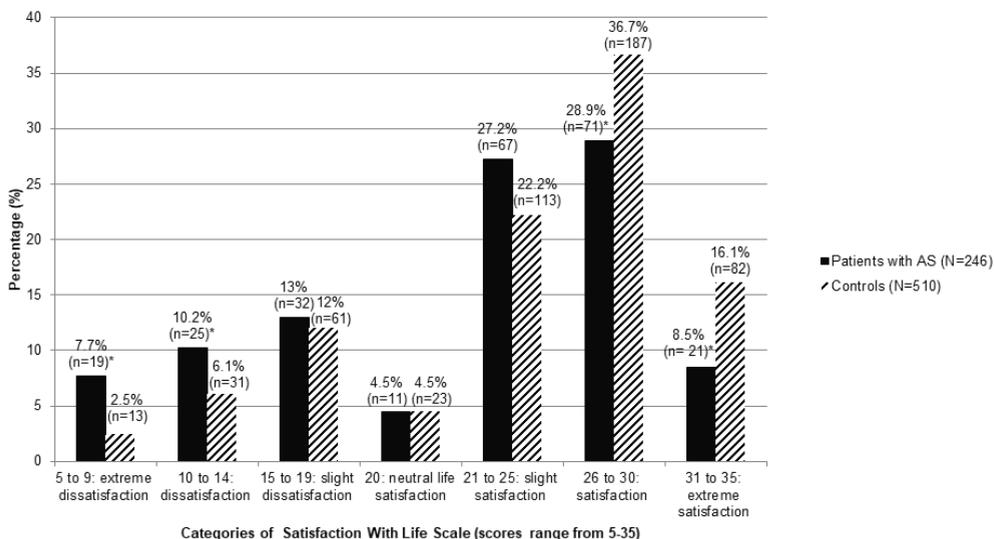


Figure 1. Percentage and absolute number of patients and controls in each category of the Satisfaction With Life Scale. * Indicates a difference between patients and controls within the category (P ≤ 0.05)

Participation in role domains and satisfaction with life in the total group of patients and controls

The explained variance of the basic model (age, gender, education and income) was $R^2 = 0.05$ in patients and $R^2 = 0.06$ in controls (appendices table 1). Only the yearly income was related with SWL in patients ($\beta: 0.9$; $P < 0.001$) and controls ($\beta: 1.06$; $P < 0.001$).

When adding the participation domains *interpersonal relations* and *leisure*, the overall fit of all models (R^2) improved in both patients and controls (table 2). In general, somewhat stronger improvements in model fit (R^2) were seen in patients compared to controls. The largest improvements in explained variance were seen in the models addressing the dimensions 'satisfaction with role performance' (model $R^2 = 0.43$) in patients or 'satisfaction with time' (model $R^2 = 0.24$) in controls. Notably, in persons with AS, the relative contribution of yearly income became negligible, when adding the social roles to models addressing the dimensions 'satisfaction with time or with performance'. The interaction term group*participation was significant for the domain interpersonal relations in the dimension 'satisfaction with performance'. This indicates that satisfaction with performance in interpersonal relations has a stronger association with SWL in patients compared to controls.

Participation in role domains and life satisfaction amongst employed subjects

In the employed sample, the total explained variance of the model with sociodemographic characteristics (basic model) provided an R^2 of 0.07 for patients and of 0.06 for controls (appendices table 2). Similar to as in the total sample, only yearly income was related to SWL in both employed patients ($\beta: 0.87$; $P=0.003$) and controls ($\beta: 0.89$; $P < 0.001$).

When adding the participation domains *interpersonal relations*, *leisure*, and *work* (table 3), again R^2 improved significantly compared with the basic model. The largest improvements were seen in the models addressing 'satisfaction with time' (total $R^2 = 0.46$ in patients and total $R^2 = 0.27$ in controls), but these improvements were very comparable when satisfaction with performance was added. While 'satisfaction with performance and with time' spent in *work* were significantly associated with SWL in controls, such association were not seen in patients. Of note, no association between difficulty with *work* and SWL in patients was observed. The associations between income and SWL lost significance in patients, in the models addressing the dimensions 'physical difficulty', 'satisfaction with time' or 'satisfaction with performance'. The interaction terms group*participation domains were not significant, indicating the relative contribution of participation domains to SWL was not statistically different between employed patients compared to employed controls.

Table 2. Association between the participation domains and satisfaction with life (SWL) among patients with AS and controls for each of the participation dimensions separately.

	Patients with AS (N=246)				Controls (N=510)			
	b	SE	p	Partial R	b	SE	p	Partial R
<i>A: Importance</i>	<i>R= 0.30 (R²= 0.09)</i>				<i>R= 0.34 (R²= 0.12)</i>			
Constant	9.44	4.04	0.020		13.38	2.09	<0.001	
Age	0.03	0.04	0.368	0.06	0.01	0.02	0.702	0.02
Gender (Male)	-0.27	0.94	0.777	-0.02	0.57	0.66	0.390	0.04
University/higher education	-0.15	1.00	0.881	-0.01	0.04	0.62	0.954	0.00
Income (per €20.000)	0.81	0.26	0.002	0.20	0.94	0.22	<0.001	0.20
Interpersonal relations	2.16	0.78	0.006	0.18	2.26	0.51	<0.001	0.21
Leisure	0.22	0.71	0.756	0.02	0.05	0.49	0.921	0.01
<i>B: physical difficulty</i>	<i>R= 0.46 (R²= 0.21)</i>				<i>R= 0.40 (R²= 0.16)</i>			
Constant	1.24	3.57	0.729		-4.58	3.90	0.241	
Age	0.05	0.04	0.137	0.10	0.04	0.02	0.027	0.11
Gender (Male)	-0.67	0.87	0.443	-0.05	-0.31	0.63	0.627	-0.02
University/higher education	-0.95	0.94	0.311	-0.07	0.09	0.60	0.886	0.01
Income (per €20.000)	0.52	0.24	0.033	0.14	0.92	0.21	<0.001	0.21
Interpersonal relations	2.59	1.12	0.021	0.15	3.25	1.11	0.003	0.14
Leisure	1.94	0.88	0.028	0.14	2.04	0.86	0.018	0.12
<i>C: Satisfaction with performance</i>	<i>R= 0.65 (R²= 0.43)</i>				<i>R= 0.44 (R²= 0.19)</i>			
Constant	1.14	2.31	0.622		8.55	1.88	<0.001	
Age	0.04	0.03	0.211	0.08	0.05	0.02	0.014	0.12
Gender (Male)	-0.25	0.74	0.738	-0.02	-0.32	0.62	0.601	-0.03
University/higher education	-1.20	0.80	0.134	-0.10	-0.16	0.59	0.789	-0.01
Income (per €20.000)	0.34	0.21	0.104	0.11	0.86	0.21	<0.001	0.20
Interpersonal relations	3.63	0.73	<0.001	0.31	1.64	0.55	0.003	0.14
Leisure	2.36	0.60	<0.001	0.25	1.73	0.51	0.001	0.17
<i>D: Satisfaction with time</i>	<i>R= 0.65 (R²= 0.42)</i>				<i>R= 0.49 (R²= 0.24)</i>			
Constant	0.68	2.34	0.772		6.22	1.87	0.001	
Age	0.01	0.03	0.822	0.02	0.02	0.02	0.323	0.05
Gender (Male)	0.58	0.75	0.439	0.05	-0.05	0.60	0.938	0.00
University/higher education	-0.57	0.80	0.474	-0.05	0.76	0.57	0.182	0.07
Income (per €20.000)	0.37	0.21	0.078	0.11	0.98	0.20	<0.001	0.23
Interpersonal relations	3.71	0.73	<0.001	0.31	2.44	0.51	<0.001	0.23
Leisure	2.60	0.59	<0.001	0.27	2.12	0.47	<0.001	0.22

Participation domain scores range from 1 (not at all important/not at all satisfied/unable to do) to 5 (extremely important/extremely satisfied/no difficulty). Satisfaction with life scores range from 5 (extreme dissatisfaction) to 35 (extreme satisfaction)

Table 3. Association between the participation domains including work and satisfaction with life (SWL) among employed patients with AS and controls.

	Employed patients (n= 140, 56.9% of total)				Employed controls (n= 396, 77.7% of total)			
	b	SE	p	Partial R	b	SE	p	Partial R
<i>A: Importance</i>	R= 0.30 (R ² = 0.09)				R= 0.34 (R ² = 0.11)			
Constant	9.13	4.50	0.043		13.84	2.35	<0.001	
Age	0.04	0.05	0.411	0.05	0.01	0.02	0.756	0.02
Gender (Male)	-0.28	0.95	0.765	-0.02	0.65	0.66	0.323	0.05
University/higher education	-0.15	1.00	0.883	-0.01	0.17	0.62	0.783	0.01
Income (per €20.000)	0.80	0.26	0.002	0.20	0.94	0.23	<0.001	0.20
Interpersonal relations	2.14	0.78	0.007	0.17	2.17	0.51	<0.001	0.20
Leisure	0.22	0.71	0.759	0.02	0.05	0.49	0.926	0.01
Work	0.06	0.35	0.876	0.01	-0.07	0.25	0.785	-0.01
<i>B: physical difficulty</i>	R= 0.49 (R ² = 0.24)				R= 0.42 (R ² = 0.18)			
Constant	1.72	4.87	0.725		-5.39	4.67	0.249	
Age	0.06	0.05	0.296	0.09	0.00	0.03	0.868	0.01
Gender (Male)	-0.94	1.07	0.383	-0.08	-0.55	0.67	0.409	-0.05
University/higher education	-1.08	1.14	0.345	-0.08	0.33	0.63	0.596	0.03
Income (per €20.000)	0.38	0.28	0.183	0.12	0.87	0.22	<0.001	0.21
Interpersonal relations	2.10	1.47	0.157	0.12	2.40	1.30	0.067	0.10
Leisure	2.63	1.19	0.029	0.19	2.98	1.02	0.004	0.16
Work	0.04	0.82	0.966	0.00	0.45	0.68	0.503	0.04
<i>C: Satisfaction with performance</i>	R= 0.66 (R ² = 0.44)				R= 0.48 (R ² = 0.23)			
Constant	2.53	3.22	0.433		10.68	2.03	<0.001	
Age	0.07	0.05	0.150	0.13	0.01	0.03	0.612	0.03
Gender (Male)	-0.93	0.92	0.316	-0.09	-0.76	0.64	0.241	-0.07
University/higher education	-1.18	0.99	0.236	-0.10	-0.02	0.61	0.980	0.00
Income (per €20.000)	0.27	0.24	0.266	0.10	0.57	0.22	0.01	0.14
Interpersonal relations	2.42	0.97	0.014	0.21	0.78	0.59	0.186	0.07
Leisure	2.39	0.78	0.003	0.26	0.87	0.54	0.111	0.09
Work	0.71	0.52	0.177	0.12	1.78	0.37	<0.001	0.26
<i>D: Satisfaction with time</i>	R= 0.68 (R ² = 0.46)				R= 0.52 (R ² = 0.27)			
Constant	1.69	3.05	0.580		7.17	2.06	0.001	
Age	0.06	0.05	0.190	0.11	0.01	0.02	0.810	0.01
Gender (Male)	0.05	0.92	0.955	0.01	-0.29	0.62	0.646	-0.03
University/higher education	0.11	0.97	0.911	0.01	1.25	0.59	0.034	0.12
Income (per €20.000)	0.20	0.24	0.394	0.07	0.73	0.21	0.001	0.19
Interpersonal relations	2.30	0.90	0.011	0.22	1.31	0.55	0.018	0.13
Leisure	2.67	0.73	<0.001	0.30	1.85	0.50	<0.001	0.20
Work	0.72	0.48	0.136	0.13	1.38	0.31	<0.001	0.24

Participation domain scores range from 1 (not at all important/not at all satisfied/unable to do) to 5 (extremely important/extremely satisfied/no difficulty). Satisfaction with life scores range from 5 (extreme dissatisfaction) to 35 (extreme satisfaction)

General participation role and satisfaction with life

When adding the score on the *general participation role items* of the SRPQ instead of the separate role domains to the basic model, the model fit improved again significantly across all dimensions in patients as well as controls (appendices table 3), except for the dimension ‘importance’ in patients. Notwithstanding, the changes of R^2 when adding the *general participation role* were somewhat smaller than in models with the separate role domains. Among patients, the influence of yearly income on SWL became smaller, but remained a significant covariate.

Discussion

This study aimed to understand whether participation in social roles in patients with AS is associated with overall life satisfaction, and to gain insight whether patients differ in this respect to controls without a chronic disease. First, it was found that more patients with AS experienced (extreme) dissatisfaction with life, and fewer reported (extreme) satisfaction when compared to population controls. Further, in patients and controls, participation in social roles was associated with higher SWL, and this relation was stronger in patients, although the test for interaction was not always significant. Within the participation domains, *interpersonal relations* contributed most to SWL in patients, while in controls *interpersonal relations* and *leisure* related almost equally to the variance in SWL. Of interest, while in controls satisfaction with performance and time spent in *work* was associated with SWL independent of the other participation domains, none of the dimensions that characterize participation in *work* contributed independently to SWL in employed persons with AS.

The lower SWL of patients compared to controls is in line with the study hypothesis and has been found in a number of chronic diseases. Lamu & Olsen (2016), showed that the presence of a chronic disease such as arthritis, diabetes, heart disease or diabetes had a negative relation with SWL (38). Also the dominant role of the domain *interpersonal relations* is in line with findings of their study, showing that SWL was strongly related with predominantly *social relationships*, and to a lesser extent by income and health outcomes (EQ 5D, SF-6D and or VAS_{health}) (38). In concordance with findings of the study mentioned above, the role of income became negligible in patients, when considering difficulties and satisfaction with social role participation. In contrast, the role of income remained important for controls, independent of social roles. Response shift and re-appraisal of the construct *life satisfaction* when being confronted with a chronic disease is a likely explanation for the differential effect of income on life satisfaction; apparently income becomes less relevant than social roles (30, 42).

In contrast to income, none of the other covariates, i.e. age, gender or education were associated with SWL. Especially for education this was surprising, as a study into the construct validity of the SWLS in a general Dutch population (n=3402) revealed that persons

with only a primary school education reported a lower life satisfaction (24). In view of the smaller sample size of our study, we had only few person with only a lower educational achievement and therefore contrasted lower and middle educational achievement with higher education or university degree. Doing so, we might have missed lower satisfaction with life in the lowest educated persons. On the other hand, similar as in the previous study, our findings confirm age and gender did not influence SWL.

While *work* is rated among the three most important social roles by patients and population subjects (69), current data suggest that for patients *work* has no independent value for SWL additionally to *interpersonal relations* and *leisure*. It should be noted that only subjects that remained in the workforce contributed data to the analyses with work. It would be interesting to evaluate prospectively how loss of work affects life satisfaction.

The dimension 'role importance' had only a low contribution to the variance in SWL.

Although role importance is of interest because it signals potential differences in the areas of life that matter to individuals, it seems less relevant by helping to understand whether someone is satisfied with their life (70). Additional multivariable models in patients, in which scores for 'role importance' were used to weigh scores for the 'satisfaction with time', 'satisfaction with performance' or 'physical difficulty' dimensions (3), did not meaningfully influence the strength of association between role domains and SWL, nor did it improve to model fit (data not shown). Despite the conceptual relevance of the importance dimension, it remains unclear how to deal with it in the light of exploring its value in the full construct 'social role participation' (70). On this line it should be acknowledged that it is not surprising that the 'satisfaction with time' and 'satisfaction with performance' dimensions were stronger associated with SWL than the other dimensions, given the similarities with the construct it was associated with, being life satisfaction. Therefore, some circular reasoning or deterministic modelling was likely present. However, we should not ignore that also 'physical difficulty' with role performance was strongly associated with life satisfaction.

Some limitations need to be recognized. Firstly, due to cross-sectional design no causality can be claimed. Secondly, the SRPQ contains 11 roles but our previous studies, including a confirmatory analysis, indicated large correlations between several roles (70, 78). Guided by literature rather than statistical approaches (for example factor analyses), we grouped the 11 roles in three domains *interpersonal relations*, *leisure* and *work*. Of interest, when repeating the analyses with the *general participation* role instead of domains, the results were overall comparable. In case details about specific role (domains) are less relevant, a single participation item can still provide valid and meaningful information (17, 18, 78). Finally, the models explained up to 43% of the adjusted variance of SWL in patients and 24% in controls. Especially in patients these are quite good model fits. Notwithstanding, other variables that are likely associated with SWL, such as level of health impairments, experience social inequality, personality traits, independence, anxiety for the future likely contribute. Although it was not the purpose of our study to understand the construct 'life

satisfaction' it would be interesting to understand SWL in persons with chronic diseases (79).

Our study provides innovative and relevant findings. While the ultimate aim of care in rheumatology is to improve SWL, the current data suggest we should not ignore the contribution of social role participation. Health care professionals could therefore address social roles when discussing care and treatment plans with patients. Further research should clarify which patients require help in improving physical performance and satisfaction with one or more roles and which support programs might be effective in reaching this goal.

In conclusion, this study shows that patients with AS indicated a lower SWL than population controls. More physical difficulties and lower satisfaction with performance and time with *interpersonal relations* were related with a lower life satisfaction. Satisfaction regarding the performance and time spent with *work* was independently associated with SWL in employed controls but not in employed patients.

Acknowledgments and affiliations

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Appendices

Table 1. Influence of age, gender, educational level and income on satisfaction with life among patients with AS and controls.

	Patients with AS (N=246)				Controls (N=510)			
	b	SE	p	Partial R	b	SE	p	Partial R
<i>Basic model (without participation):</i>	R= 0.23 (R ² = 0.05)				R= 0.25 (R ² = 0.06)			
Constant	19.26	2.08	<.001		21.22	1.16	<.001	
Age	0.02	0.04	0.611	0.03	0.01	0.02	0.536	0.03
Gender (Male)	-0.49	0.95	0.606	-0.03	-0.17	0.66	0.802	-0.01
University/higher education	-0.34	1.01	0.739	-0.02	0.31	0.63	0.618	0.02
Income (increase per €20.000)	0.91	0.26	0.001	0.22	1.07	0.23	<0.001	0.23

Participation domain scores range from 1 (not at all important/not at all satisfied/unable to do) to 5 (extremely important/extremely satisfied/no difficulty). Satisfaction with life scores range from 5 (extreme dissatisfaction) to 35 (extreme satisfaction)

Table 2. Influence of age, gender, educational level and income on satisfaction with life among employed patients with AS and controls

	Employed patients with AS (N=140, 56.9% of total)				Controls (N=396, 77.7% of total)			
	b	SE	p	Partial R	b	SE	p	Partial R
<i>Basic model (without participation):</i>	R= 0.28 (R ² = 0.07)				R= 0.24 (R ² = 0.06)			
Constant	19.98	2.85	<.001		22.74	1.32	<.001	
Age	0.04	0.06	0.506	0.06	-0.01	0.03	0.798	-0.01
Gender (Male)	-0.67	1.16	0.564	-0.05	-0.65	0.71	0.363	-0.05
University/higher education	-0.89	1.24	0.475	-0.06	0.59	0.67	0.374	0.05
Income (increase per €20.000)	0.87	0.29	0.003	0.25	0.89	0.24	<0.001	0.20

Participation domain scores range from 1 (not at all important/not at all satisfied/unable to do) to 5 (extremely important/extremely satisfied/no difficulty). Satisfaction with life scores range from 5 (extreme dissatisfaction) to 35 (extreme satisfaction)

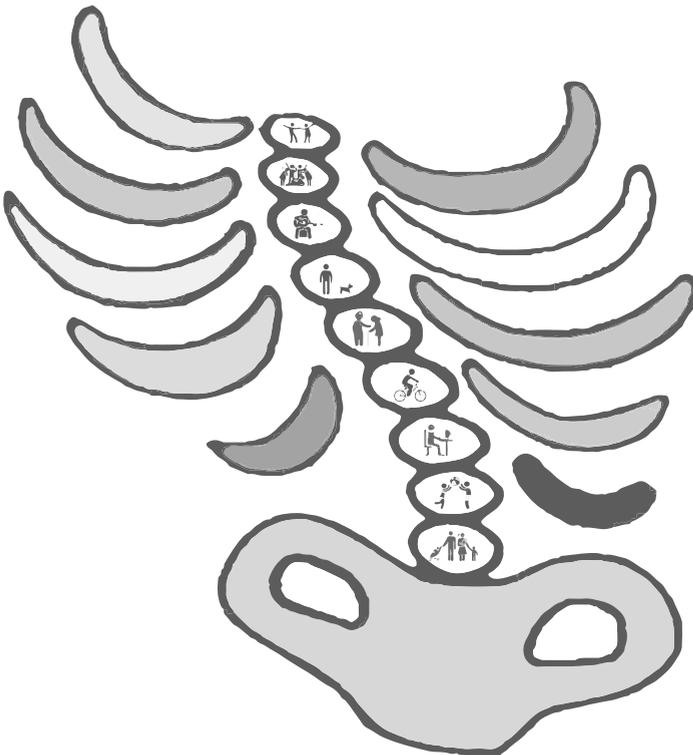
Table 3. The association between overall social role participation and satisfaction with life in patients with AS and controls for each of the participation dimensions separately.

	Patients with AS (N=246)				Controls (N=510)			
	b	SE	p	Partial R	b	SE	p	Partial R
<i>A: Importance</i>	R= 0.26 (R ² = 0.07)				R= 0.31 (R ² = 0.10)			
Constant	13.38	3.73	<0.001		15.48	1.81	<0.001	
Age	0.04	0.04	0.334	0.06	0.02	0.02	0.357	0.05
Gender (Male)	-0.16	0.96	0.871	-0.01	0.20	0.66	0.763	0.02
University/higher education	-0.51	1.01	0.618	-0.03	0.13	0.62	0.839	0.01
Income (per €20.000)	0.86	0.26	0.001	0.21	0.99	0.22	<0.001	0.21
General participation	1.15	0.61	0.060	0.12	1.40	0.34	<0.001	0.20
<i>B: physical difficulty</i>	R= 0.46 (R ² = 0.21)				R= 0.40 (R ² = 0.16)			
Constant	8.26	2.39	0.001		6.49	2.37	0.006	
Age	0.01	0.03	0.684	0.03	0.04	0.02	0.064	0.09
Gender (Male)	-0.76	0.86	0.379	-0.06	-0.21	0.63	0.738	-0.02
University/higher education	-1.20	0.92	0.195	-0.08	0.06	0.60	0.914	0.01
Income (per €20.000)	0.61	0.24	0.011	0.16	0.95	0.21	<0.001	0.21
General participation	3.58	0.48	<0.001	0.43	3.08	0.44	<0.001	0.32
<i>C: Satisfaction with performance</i>	R= 0.67 (R ² = 0.44)				R= 0.56 (R ² = 0.32)			
Constant	7.21	1.85	<0.001		9.05	1.38	<0.001	
Age	0.01	0.03	0.647	0.03	0.04	0.02	0.042	0.10
Gender (Male)	-0.55	0.73	0.449	-0.05	-0.21	0.56	0.713	-0.02
University/higher education	-1.59	0.78	0.043	-0.13	0.24	0.54	0.659	0.02
Income (per €20.000)	0.44	0.20	0.030	0.14	0.59	0.20	0.002	0.15
General participation	4.67	0.36	<0.001	0.64	3.52	0.28	<0.001	0.52
<i>D: Satisfaction with time</i>	R= 0.66 (R ² = 0.44)				R= 0.50 (R ² = 0.25)			
Constant	5.31	5.31	0.007		9.54	1.53	<0.001	
Age	-0.01	-0.01	0.734	-0.02	0.01	0.02	0.507	0.03
Gender (Male)	0.55	0.55	0.456	0.05	0.01	0.59	0.993	0.00
University/higher education	-0.53	-0.53	0.499	-0.04	0.33	0.56	0.556	0.03
Income (per €20.000)	0.46	0.46	0.023	0.15	0.83	0.20	<0.001	0.20
General participation	4.91	4.91	<0.001	0.64	3.50	0.34	<0.001	0.45

Participation domain scores range from 1 (not at all important/not at all satisfied/unable to do) to 5 (extremely important/extremely satisfied/no difficulty). Satisfaction with life scores range from 5 (extreme dissatisfaction) to 35 (extreme satisfaction).

Part 2

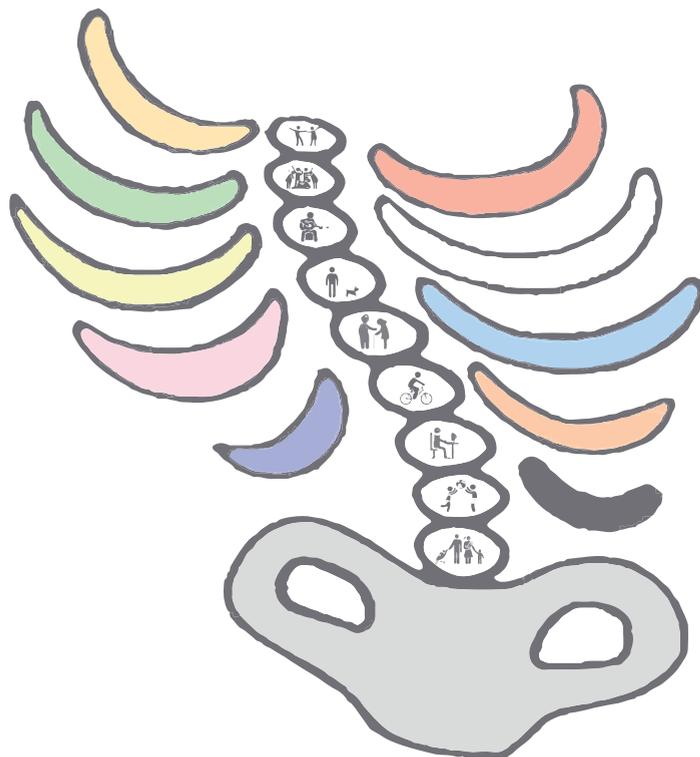
Physical activity and body composition



5

Physical functioning in patients with ankylosing spondylitis: comparing approaches of experienced ability with self-reported and objectively measured physical activity

van Genderen, S., van den Borne, C., Geusens, P., van der Linden, S., Boonen, A., & Plasqui, G. Physical functioning in patients with ankylosing spondylitis: comparing approaches of experienced ability with self-reported and objectively measured physical activity. *JCR: Journal of Clinical Rheumatology*. 2014;20(3):133-137.



Abstract

Background: Physical functioning can be assessed by different approaches that are characterized by increasing levels of individual appraisal. There is insufficient insight into which approach is the most informative in patients with ankylosing spondylitis (AS) compared with control subjects.

Objective: The objective of this study was to compare patients with AS and control subjects regarding 3 approaches of functioning: experienced ability to perform activities (Bath Ankylosing Spondylitis Functional Index [BASFI]), self-reported amount of physical activity (PA) (Baecke questionnaire), and the objectively measured amount of PA (triaxial accelerometer).

Methods: This case-control study included 24 AS patients and 24 control subjects (matched for age, gender, and body mass index). Subjects completed the BASFI and Baecke questionnaire and wore a triaxial accelerometer. Subjects also completed other self-reported measures on disease activity (Bath AS Disease Activity Index), fatigue (Multidimensional Fatigue Inventory), and overall health (EuroQol visual analog scale).

Results: Both groups included 14 men (58%), and the mean age was 48 years. Patients scored significantly worse on the BASFI (3.9 vs 0.2) than their healthy peers, whereas PA assessed by Baecke and the accelerometer did not differ between groups. Correlations between approaches of physical functioning were low to moderate. Bath Ankylosing Spondylitis Functional Index was associated with disease activity ($r = 0.49$) and physical fatigue (0.73) and Baecke with physical and activity related fatigue ($r = 0.54$ and $r = 0.54$), but total PA assessed by accelerometer was not associated with any of these experience-based health outcomes.

Conclusions: Different approaches of the concept physical functioning in patients with AS provide different information. Compared with matched control subjects, patients with AS report more difficulties but report and objectively perform the same amount of PA.

Introduction

Ankylosing spondylitis (AS) is a chronic inflammatory rheumatic disease, characterized by inflammation of the sacroiliac joints and spine and, to lesser extent, of the peripheral joints (1). Important symptoms comprise pain and (morning) stiffness, fatigue, and reduced mobility, which cause limitations in the physical function of patients. Physical function has shown to be one of the most important health variables associated with different aspects of health-related quality of life in AS, including social roles such as work participation (2, 3).

As a consequence, the Assessment in Ankylosing Spondylitis International Society included the domain “physical function” in the core outcomes domains that should be measured in all trials as well as in clinical record keeping (4, 5).

To measure physical function, the Bath Ankylosing Spondylitis Functional Index (BASFI) is proposed (6). The BASFI was selected as the preferred measure out of several self-reported questionnaires asking the respondent to estimate the experienced ability or difficulty to perform a series of tasks and activities. Next to the BASFI, there are various other instruments available to qualify or measure performed activities in daily life (7). When trying to understand the differences in instruments to assess a specific construct or domain, for example, that of physical functioning, it is important to understand the level of individual appraisal that is involved in the assessment (Figure 1). The level of appraisal not only depends on the measure or test itself but also differs across people and over time, and this can greatly affect how people answer, given questions or items (8). On this line, the BASFI possesses a high level of appraisal, as every respondent rates the items to his/her individual perspective on his/her ability to perform the task or activity. When interpreting the general briefing of the BASFI, “indicate your level of ability with each of the following activities during the past week;” this will be a highly subjective evaluation influenced not only by adaptation but also by needs. Other instruments, like the Baecke questionnaire for the measurement of habitual physical activity (PA), has a more fixed construct and asks respondents to give an indication of the perceived amount of PA they perform in different life areas (9).

Finally, an accelerometer or activity monitor offers the opportunity to objectively measure performed PA. It is considered to be the most valid technique to assess PA with information on activity patterns in terms of frequency, duration, and intensity (10).

Although it has never been studied as such, one might postulate that the 3 approaches to measure a same domain (physical functioning) highlight different aspects and provide different but likely additional information. While experienced ability to perform activities is probably more related to different aspects of self-reported health-related quality of life, PA in patients with AS is important as they already have an increased risk for cardiovascular comorbidities, and possible physical inactivity may pose an extra risk (11, 12).

In a previous published study, we observed no differences between AS and healthy subjects for PA while using accelerometers (13). This prorogated the question whether patients and control subjects would differ in approaches requiring different levels of ap-

praisal to assess physical functioning, what the correlation between approaches is, and to what extent differences between approaches could be explained. Hence, in this study, we compared patients with AS and control subjects with regard to approaches for physical functioning (performed PA, perceived PA, and experienced ability) and, second, explored possible differences within patients regarding the approaches and their relation with demographic and health-related variables.

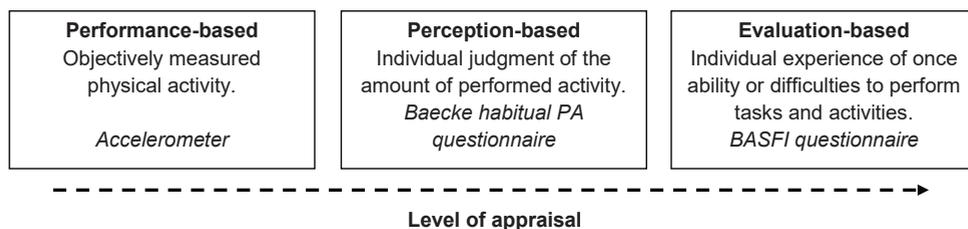


Figure 1. Different methods to assess the construct “physical functioning” and their relation to the level of appraisal.

Materials and methods

This study reports on additional data and analyses of a cross-sectional case-control study that explored body composition and PA in patients with AS (13)

Patients and Control Subjects

Twenty-four AS patients (14 males), with an average age of 48 (SD \pm 11) years, symptom duration of at least 5 years, and diagnosed according to the modified New York criteria, were recruited through the rheumatology departments of Maastricht University Medical Centre+, Maxima Medisch Centrum, Eindhoven, and Atrium Medisch Centrum, Heerlen, the Netherlands. Patients were excluded when they were on anti-tumor necrosis’s factor α therapy or had comorbidities that might affect energy balance, such as diabetes, inflammatory bowel disease, or malignancies. Patients were compared with 24 healthy adults, matched for age, gender, and body mass index (BMI). For 14 of 24 patients, the matched control was a healthy first-degree relative. Because this complicated inclusion substantially, the remaining control subjects were recruited from the general population using poster advertisements around the university. The study was approved by the medical ethics committee of Maastricht University Medical Centre +.

Measures of physical functioning

Accelerometer. Objectively measured PA was assessed using the triaxial accelerometer for movement registration (Tracmor; Philips Research, Eindhoven, the Netherlands). The Tracmor contains 3 uniaxial piezo-electric accelerometers, measures 7.2 x 2.6 x 0.7 cm, and weighs 22g (battery included). It is attached to the lower back of the subject by means of

an elastic belt (10). It measures minute-by-minute accelerations (expressed as kilocounts [kcounts]) in the anteroposterior, mediolateral, and longitudinal axis of the trunk. Subjects were instructed to wear the Tracmor for 7 consecutive days, with a minimum of 10 h/d, during waking hours, except during water activities. First, the wearing time (which is the time in minutes subjects wore the accelerometer) was assessed. Second, the total activity per day was calculated by dividing the total amount of kcounts by the number of days the accelerometer registered at least 10 hours' activity. To measure the intensity of the PA of patients, the total amount of kcounts was divided by the total wearing time, which provided kcounts per minute.

Baecke Questionnaire. This questionnaire was used to assess the self-reported level of perceived habitual PA (9). The questionnaire consists of 16 questions, addressing 3 domains of PA, which are PA at work, sports during leisure time, and PA during leisure excluding sports. The questions in each section are scored on a 5-point scale displaying (a) broad categories of frequency (from “never” to “always” or for other items “very often”) or (b) time engaged (per week, month or year). Total sum scores for each domain range from 1 to 5, and the sum of these 3 domains combined provides an indication of the total level of PA.

Bath Ankylosing Spondylitis Functional Index. The BASFI asks about the level of experienced ability or difficulties to perform 10 activities during the last week. The total score is an average of the 10 items and ranges from 0 to 10 (higher score indicating worse functioning) (6).

Demographic and Health-Related Variables

Age, gender, and height were noted when patients received the accelerometer. Weight was measured in the morning, after an overnight fast to the nearest 0.1 kg. Subjects filled out several questionnaires during the week they wore the accelerometer. The Bath Ankylosing Spondylitis Disease Activity Index (BASDAI) was included to assess disease activity. It is based on 6 questions that give a total score ranging from 0 (indicating no disease activity) to 10 (higher score indicating higher disease activity) (14).

Fatigue was measured by the Multidimensional Fatigue Inventory (MFI), which consists of 20 theses that can be scored to produce 5 dimensions: general fatigue, physical fatigue, mental fatigue, reduced motivation, and reduced activity (15). The scores of each subscale range from 0 (totally agree) to 5 (totally disagree) (higher score indicating more fatigue). General health was assessed by the EuroQol visual analog scale (EQ-VAS), which asks to indicate today's health on a 0 (death) to 100 (full health) vertical rating scale (16).

Statistical Analyses

Comparisons between patients and control subjects were performed using the Chi-square test for the categorical variable gender and the independent Kruskal-Wallis tests for the

variables age, total activity counts, time of PA and intensity of PA, Baecke questionnaire, BASDAI, BASFI, MFI, and EQ-VAS. As all measures of PA were slightly skewed to the right (a few patients and control subjects having higher physical activities), normality of the data could not be assumed. Within patients, first Spearman correlations (2-tailed) were performed between the approaches: PA (total activity and intensity of activity), Baecke questionnaire (all domains), and BASFI. Next, Spearman correlations were computed between BASFI, Baecke questionnaire, and PA measured with an accelerometer with demographic and health-related variables (age, BMI, diagnose duration, BASDAI, MFI general fatigue, and EQ-VAS) (Table 1). For all correlations, coefficients until 0.40 were qualified as weak, 0.41 and 0.75 as moderate, and more than 0.75 as strong (17). All analyses were performed in SPSS version 19.0, SPSS, Chicago, IL.

Table 1. Spearman's correlations between physical functioning, total physical activity measured with the Baecke questionnaire, different dimensions of physical activity measured with accelerometers and (disease) characteristics for 24 patients with AS

	BASFI	BAECKE (total PA)	Total activity a day	Intensity of activity
Age	0.24	0.51*	0.11	0.07
BMI	0.46*	0.04	-0.20	-0.23
Disease duration	0.07	-0.02	-0.21	-0.22
BASDAI	0.49*	-0.22	-0.10	-0.02
MFI-20-Pf	0.73*	-0.54*	-0.33	-0.23
MFI-20-Mf	0.40	-0.48*	-0.12	-0.10
MFI-20-Rm	0.48*	-0.34	-0.17	-0.14
MFI-20-Ra	0.53*	-0.54*	-0.32	-0.40
MFI-20-G	0.35	-0.25	-0.08	-0.06
EQ-VAS	-0.39	0.30	0.30	0.17

*Correlation is significant at the 0.05 level (2-tailed) BMI, Body Mass Index; BASDAI, Bath AS Disease Activity Index; BASFI, Bath AS Functional Index; MFI-20-G, Ph, Mf, Rm, Ra, Multidimensional Fatigue Inventory, General fatigue, Physical fatigue, Mental fatigue, Reduced motivation, Reduced activity ; EQ-VAS, EuroQol Visual Analogue scale

Results

In total, 48 subjects (24 patients and 24 matched control subjects) were included. Table 2 shows that patients and control subjects did not differ with regard to age, gender, BMI, aspects of PA (total PA a day and intensity of activity), and PA measured with the Baecke questionnaire. In addition, no statistical differences between patients and control subjects were found regarding the wearing time of the accelerometer. However, patients scored

significantly worse on the BASDAI, all dimensions of the MFI except mental fatigue, overall health (EQ-VAS), and the BASFI.

Correlations Between Accelerometer, Baecke Questionnaire, and BASFI

Total activity per day assessed by the accelerometer correlated significantly with intensity in patients ($r = 0.90$) ($P < 0.05$). Furthermore, within patients, total activity per day as well as intensity (accelerometer) correlated moderately with Baecke (total PA) ($r = 0.45$ and $r = 0.41$, respectively) and BASFI ($r = 0.47$ and $r = 0.41$ respectively). Between Baecke (total PA) and BASFI, the correlation was weak ($r = 0.39$).

Table 2. Characteristics of patient and control subjects

	Patient (AS (N=24)) Median (ICR) min - max	Control (N=24) Median (ICR) min - max	P
Gender (M:F)	14:10	14:10	NS
Age (y)	47 (15.5) 23 - 62	49 (17.5) 18 - 65	NS
Diagnosis duration (y)	20.5 (22.0) 5 - 44	N/A	N/A
BMI (kg/m ²)	26.0 (4.6) 18.1 - 39.6	25.5 (1.6) 20.5 - 31.5	NS
<i>Accelerometer:</i>			
Wearing time (hours per day)	14.9 (1.4) 11.23 - 16.96	14.6 (0.97) 8.6 - 16.57	NS
Total activity a day (kcounts/day)	277 (105) 156 - 567	295 (75) 165 - 474	NS
Intensity of activity: (kcounts/min)	0.30 (0.09) 0.18 - 0.64	0.33 (0.09) 0.22 - 0.52	NS
Baecke (Work)	2.6 (1.3) 0 - 4.1	2.6 (1.1) 1.5 - 4	NS
Baecke (Sport)	2.6 (1.3) 1 - 4.8	3.0 (1.4) 1.0 - 4.5	NS
Baecke (leisure)	3.0 (1.0) 1.8 - 4	3.3 (0.6) 2.5 - 4.5	NS
Baecke (total PA)	8.4 (1.4) 4.5 - 12.4	8.4 (1.6) 7.5 - 11	NS
BASFI	3.85 (2.1) 0.4 - 7.6	0.2 (0.4) 0 - 0.9	<0.001
BASDAI	4.1 (3.7) 1.1 - 7.3	0.4 (1.1) 0 - 2.6	<0.001
MFI-20 (Physical fatigue)	11.0 (7.8) 4 - 20	5.0 (3.3) 4 - 10	<0.001
MFI-20 (Mental fatigue)	8.0 (9.5) 4 - 16	4.5 (4.3) 4 - 16	NS
MFI-20 (Reduced motivation)	7.0 (6.8) 4 - 16	4.0 (1.3) 4 - 8	0.001
MFI-20 (Reduced activity)	8.0 (7.5) 4 - 17	4.0 (1.0) 4 - 7	<0.001
MFI-20 (General fatigue)	14.0 (5.3) 4 - 20	5.5 (5.3) 4 - 14	<0.001
EQ-VAS (General health)	79.0 (23.8) 35 - 100	95.0 (17.3) 70 - 100	0.003

BMI, Body Mass Index; Total activity in kilocounts per day; Intensity of activity in kilocounts per minute; Baecke physical activity questionnaire; BASDAI, Bath AS Disease Activity Index; BASFI, Bath AS Functional Index; MFI-20, Multidimensional Fatigue Inventory; EQ-VAS; EuroQol Visual Analogue scale; N/A, not applicable; NS, not significant; ICR, Inter Quartile Range

Correlations Between Measures for Physical Functioning and Other Health Outcomes

Although there were no significant correlations between accelerometer-assessed PA (total activity per day and intensity) with any demographic or self-reported health variables, the BASFI and Baecke (total PA) showed moderate to strong correlations with several aspects of fatigue. In addition, the BASFI correlated moderately with BASDAI and BMI and Baecke (total PA) moderately with age.

Discussion

Despite more limitations in experienced ability to perform tasks and activities (BASFI), patients with AS compared with control subjects showed similar amounts of self-reported (Baecke questionnaire) as well as objectively assessed PA (accelerometer). Within patients, objectively measured total PA showed only a moderate correlation with Baecke (total PA) and the BASFI. This indicates that the 3 approaches measure different aspects of physical functioning. Objectively measured PA was not related to any health- or disease-related measure, whereas Baecke (total PA) and BASFI showed moderate associations with different aspects of fatigue (MFI) or disease activity (BASDAI). Although the 3 approaches that we have chosen to address physical functioning differ in the “level of appraisal,” they also differ in the “areas of functioning” they address. While the 3-axis accelerometer addresses almost all activities and participation roles performed, the BAECKE concentrates more on time spent in social roles (sports, work, leisure) and the BASFI on difficulties with specific physical tasks (reaching, standing, walking,...) (18). This becomes clear when we link the content of the BASFI and BAECKE to the International Classification of Functioning, Disability and Health (ICF) and indicate which of these categories are registered by the accelerometer (appendix table 1). As can be seen, BASFI and BAECKE address quite different aspects of functioning. The accelerometer, on the other hand, does not address all ICF categories that are scored in BASFI or BAECKE and likely registers a number of activities not specified in any of the questionnaires. This likely also contributes to the observed moderate correlations between the measures.

An explanation of the equal PA levels might be found in the guideline for physiotherapy of AS patients, which in general urges patients to engage in a form of physical exercise, as the latter temporarily diminishes pain and stiffness symptoms (19). On this account, it seems that AS patients give a good adherence to the guideline since it has recently been shown that 7 of 10 patients with spondyloarthritis met the recommendations of PA and health, which have been stated by the World Health Organization, that is, 150 minutes of moderate-intensity or 75 minutes of vigorous-intensity PA per week (20). Although the correlations between objectively assessed PA, self-reported PA, and experienced ability to perform activities were moderate in patients, our study is the first to compare different approaches of measuring physical functioning between patients with AS and control subjects. In patients with rheumatoid arthritis, a recent report including 171 patients also revealed

modest but statistically significant correlations between a self-reported questionnaire (Yale Physical Activity Survey) and uniaxial accelerometer measures (21). However, no experience-based measure was included. The selection of a triaxial accelerometer over other commercially available motion sensors such as pedometers and uniaxial accelerometers is based on the knowledge that subjects can perform in more types of activities than walking/running alone. Hence, a triaxial accelerometer can provide more information and also shows a better relationship with energy expenditure caused by PA than uniaxial accelerometers (22). However, for use in clinical practice, uniaxial accelerometers or pedometers might also give relevant information; this issue would need further study.

The triaxial accelerometer allowed to compare total activity a day and the intensity of activity between patients and control subjects but revealed no differences in these dimensions. Moreover, high correlations between total activity and intensity were observed, indicating that both dimensions more or less measured the same. When further exploring differences between the approaches to assess physical functioning within patients, no relation was found with health- or disease-related variables and objectively measured PA, whereas both Baecke total PA and BASFI correlated with aspects of fatigue (MFI) and the BASFI also with the BASDAI. Although it might not be surprising that experience-based health measures, such as BASDAI and MVI, are more strongly associated with the experience-based than with the objective functioning outcomes, this finding further supports the importance to distinguish the different levels of measurement. The influence of pain and stiffness (BASDAI) on self-reported ability for tasks and activities (BASFI) is well recognized (23, 24); however, the role of fatigue in physical function had not yet been emphasized. It can be expected that the energy cost to perform PA is higher in patients with AS compared with control subjects. This was previously shown among patients with rheumatoid arthritis who spent more energy on similar activities such as treadmill walking compared with control subjects (25). It is obvious that the present case-control study cannot conclude whether fatigue is a consequence or cause of experienced difficulties with abilities. In this line, recent studies also pointed to the role of psychological factors such as perceived helplessness and coping in reported limitations in functioning (26, 27).

Some limitations need to be considered when interpreting the present results. Most importantly, the sample size was small, and as a result, little differences in PA between patients and control subjects might be missed, and it limited a full exploration of all relationships and interactions. In addition, as the participants partly included family matched control subjects, the performed PA might have been influenced by the PA behavior of one's relative. The possible influential role of relatives on PA-related behavior has been recognized in various populations (28).

However, Rhodes and Blanchard (29), found that PA behavior and the intention to engage in PA especially in patients with arthritis compared with a non-diseased population were not so much influenced by perceived approval from persons in their environment but more by the level of self-efficacy or level of control. One could argue that the BASFI has

not been validated in a healthy population; however, it includes essential daily tasks that require PA that are likely equally relevant for healthy persons. Here, it was surprising that patients, despite having more limitations in activities that are common for functioning in daily life, did not report and perform less PA than did the control subjects.

Also, when considering the prognostic relevance of the 3 approaches with regard to relevant outcomes, such as social participation, falls and fractures, cardiovascular disease, or mortality, we lack long-term data to reveal possible differences.

In conclusion, regarding physical functioning in patients with AS, different approaches to measure experienced abilities or actual performed activity provide different information, but their exact role with regard to long-term mental or physical outcome, including cardiovascular disease and mortality, needs to be further explored. However, despite a decreased experienced ability to perform activities and worse reported disease-related outcomes, patients with AS do not differ in their self-reported or objectively assessed PA compared with control subjects.

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Appendix

Table 1. Linking of the Bath Ankylosing Spondylitis Functional Index (BASFI), the Baecke questionnaire of habitual physical activity to the ICF and an indication if the accelerometer is able to register these activities.

ICF component/chapter	ICF Category	BASFI	Baecke	Accelerometer
Component: Body Functions				
<i>Chapter 1:</i> <i>Global mental functions</i>	b 130; Energy and drive	-	1X <i>Context:</i> d 850; paid work	-
<i>Chapter 8:</i> <i>Functions of the skin and related structures</i>	b 830; Other functions of the skin (sweating)	-	2X <i>Context:</i> (1) d 850; paid work (2) d 920; leisure	-
Component: activities and participation				
<i>Chapter 4:</i> <i>Mobility</i>	d 498; Mobility other specified	1X <i>Specification:</i> (Looking over your shoulder without turning your body) not included in ICF	-	-
	d 4100; Lying down	1X <i>Specification:</i> (e3 support and relationships)	-	X
	d 4103; Sitting	1X <i>Specification:</i> (1) b 7603; Supportive functions of arm or leg (2) (e3 support and relationships)	1X <i>Context:</i> d 850; paid work	X
	d 4105; Bending	1X	-	X

Table 1. Linking of the Bath Ankylosing Spondylitis Functional Index (BASFI), the Baecke questionnaire of habitual physical activity to the ICF and an indication if the accelerometer is able to register these activities. (continued)

ICF component/chapter	ICF Category	BASFI	Baecke	Accelerometer	
Chapter 4: Mobility	d 4154; Maintaining a standing position	IX <i>Specification:</i> b 289; Sensation of pain, other specified and unspecified	IX <i>Context:</i> d 850; paid work	X	
	d 4300; Lifting and carrying objects	-	IX <i>Context:</i> d 850; paid work	X	
	d 4400; Picking up	Ix <i>Specification:</i> (e3 support and relationships)	-	X	
	d 4451; Climbing	IX <i>Specification:</i> (e1201 assistive products and technology for personal indoor and outdoor mobility and transportation)	-	X	
	d 4452; Reaching	IX <i>Specification:</i> (e3 support and relationships)	-	X	
	d 4500; Walking short distances	-	IX <i>Context:</i> d 850; paid work	X	
	d 4501; Walking long distances	-	IX <i>Context:</i> d 920; recreation and leisure	X	

Table 1. Linking of the Bath Ankylosing Spondylitis Functional Index (BASFI), the Baecke questionnaire of habitual physical activity to the ICF and an indication if the accelerometer is able to register these activities. (continued)

ICF component/chapter	ICF Category	BASFI	Baecke	Accelerometer
	d 4558; Moving around, other specified	-	2X <i>Context:</i> d 920; recreation and leisure	X
<i>Chapter 5: Self-care</i>	d 5402; Putting on footwear	1X <i>Specification:</i> (1) (e3 support and relationships) (2) (e1151 assistive products and technology for personal use in daily living)	-	X
	d 5709; Looking after one's health, unspecified	1X	-	X
<i>Chapter 6: Domestic life</i>	d 640; Doing housework	1X	-	-
	d 6509; Caring for household tasks	1X	-	X
<i>Chapter 8: Major life areas</i>	d 850; Remunerative employment	1X	-	X
<i>Chapter 9: community, social and civic life</i>	d 920; Recreation and leisure	1X <i>Specification:</i> (d 9209; Recreation and leisure, unspecified)	4X <i>Context:</i> (d 9202; Sports)	X

Abstract

Objective. To compare the total amount of physical activity (TPA) and time spent in various activity intensities of patients with ankylosing spondylitis (AS) and population controls, and to explore factors related to physical activity (PA).

Methods. Subjects were asked to wear a triaxial accelerometer for 7 days and to complete a series of questionnaires. Multivariable regressions were used to assess generic determinants of TPA in patients and controls, and in patients to explore demographic and disease-specific determinants of various PA intensities.

Results. One hundred and thirty-five patients [51 ± 13 years, 60% men, body mass index (BMI) 26.0 ± 4.3 kg/m²] and 99 controls (45 ± 12 years, 67% men, BMI 25.1 ± 4.3 kg/m²) were included. Patients did not differ from controls regarding TPA (589 vs 608 vector count/min, $p = 0.98$), minutes/day spent in sedentary (524 vs 541, $p = 0.17$), and light PA (290 vs 290 $p = 0.95$), but spent fewer minutes/day in moderate to vigorous PA (MVPA; 23 vs 30 min/day, $p = 0.006$). Perceived functional ability (physical component summary of the Medical Outcomes Study Short Form-36) and BMI were associated with TPA independent of having AS (p interaction = 0.21 and 0.94, respectively). Additional analyses in patients showed that time spent in MVPA was negatively influenced by BMI, physical function (Bath AS Functional Index), and disease duration. In patients ≥ 52 years old, a higher Bath AS Disease Activity Index was associated with less time spent in sedentary and more time spent in light activities.

Conclusion. Compared with controls, patients with AS had similar TPA, but may avoid engagement in higher intensities of PA. Lower levels of functional ability and higher BMI were associated with lower TPA in both patients and controls.

Introduction

Regular physical activity (PA) has well-known beneficial health effects. Moderate and vigorous intensities of PA (MVPA) are especially considered vital to enhance health (1, 2).

Research in the overall population has shown clear positive effects of MVPA on health. A recent population study in 217755 middle-aged adults indicated that time spent in vigorous intensity of PA, independent of the total amount of PA, was associated with a reduced risk of mortality (3). Patients with ankylosing spondylitis (AS) can be hampered in their daily habitual activities because of pain, stiffness, and limitations in mobility (4, 5). It might therefore not be straightforward for patients to engage in sufficient PA to maintain or improve health. In AS, the literature on the total amount of physical activity (TPA), representing all measured movement of the body produced by skeletal muscle (6), as well as time spent in different intensities of movement, is scarce and contrasting.

When using an objective measure to assess PA, evidence of 2 studies examining a small group of patients with AS (7) or axial spondyloarthritis (axSpA) (8). shows that patients have a similar or slightly lower level of TPA than healthy controls, respectively. Regarding time spent in different levels of intensity, patients with axSpA were shown to spend less time in MVPA in comparison with controls (8). However, in a study based on a validated recall questionnaire to assess PA, it was reported that patients with SpA, including AS, more often than healthy controls met the recommendations of the World Health Organization (WHO) (9), i.e., 150 min of PA a week in moderate intensity, 60–75 min in vigorous intensity, or an equivalent combination of both (2, 10). Additional data are needed to elucidate these contradicting findings and to see whether findings such as meeting the WHO recommendations remain once PA is objectively measured.

When aiming at optimizing PA in patients with AS, it is important to understand factors contributing to PA and to explore whether such factors would be different in patients compared with healthy individuals. This is relevant because it might reveal that patients require different approaches to optimize PA and no such data are available. Among patients, the limited cross-sectional evidence suggests that higher levels of PA are associated with lower disease activity and better spinal mobility, physical function, and health-related quality of life, as well as lower levels of C-reactive protein (CRP) and erythrocyte sedimentation rate (ESR) (7, 9, 11, 12, 13). On the other hand, data have been reported that heavy loading jobs can accelerate spinal radiographic damage (14). A first step to elucidate these relevant issues is to explore PA and its patterns in patients with AS.

Therefore, given the previously mentioned gaps in knowledge, the primary aim of our current study was to compare TPA as well as time spent in different intensities of PA in patients with AS and population controls using validated accelerometers. Secondary aims were first to study whether known determinants of PA such as difficulties in functional ability and a higher body mass index (BMI) have a similar influence on the TPA in patients and controls, and next to investigate the association between disease-specific determinants and time spent in various intensities of PA in patients with AS. It was hypothesized that

patients would not differ from controls regarding total performed PA, but would spend less time in MVPA.

Materials and methods

The Social Participation in AS Study (SPASS) was a multicenter cross-sectional study that first aimed to assess social role participation in an online questionnaire survey and second to investigate objectively assessed PA in patients with AS compared with population controls in a random subsample for whom an accelerometer was available. The study protocol was approved by the ethics committee of the academic hospital Maastricht and Maastricht University.

Subjects

Patients. Patients of at least 18 years, registered with a diagnosis of AS in each of the 6 participating rheumatology departments and for whom the rheumatologist confirmed they fulfilled the modified New York criteria for AS, were invited by a letter. Patients were excluded if they had no access to the Internet or were not familiar with the Dutch language. Figure 1 illustrates that 246 of a total of 740 patients with AS (33%) consented to participate in the SPASS study. Of a random sample, 154 also participated in our current substudy on PA. All patients provided written informed consent.

Controls. Control subjects were recruited from a national open online panel of the research institute Ipsos (Amsterdam, the Netherlands). Based upon our expectations of a cross-sectional sample in AS, the population controls were sampled to have an average age of 42 years and a male:female distribution of 3:1 (15). Subjects were excluded if they had any musculoskeletal disorders or if they were not familiar with the Dutch language (16). Figure 1 shows that of the 510 of a total of 2767 controls (18%) who participated in the online questionnaire survey, a randomly selected subgroup ($n = 109$) agreed to wear the triaxial accelerometer.

Assessments

PA was measured using a triaxial accelerometer (Actigraph GT3X, Actigraph LLC Pensacola), which had shown good validity when using energy expenditure measured by doubly labeled water as the gold standard (17). It was attached at the lower back of the subject by means of an elastic belt and measured minute by minute accelerations (expressed as counts) in the anteroposterior, mediolateral, and longitudinal axes of the trunk. Subjects received the accelerometer by mail and were instructed to wear the device for 7 consecutive days during waking hours, except during water activities. Data were considered to be complete if subjects wore the accelerometer for at least 3 days and 10 h per day. In addition, whenever the device measured consecutive zeroes over a period of ≥ 60 min, this was classified as a non-wear period. From the output of the 3 axes, the vector magnitude counts (VM3) could

be calculated [$VM3 = \sqrt{(\text{total counts axis } 1^2 + \text{total counts axis } 2^2 + \text{total counts axis } 3^2)}$], and subsequently counts per day ($CPD = VM3 \div \text{total wear time in calendar days}$) and counts per minute ($CPM = VM3 \div \text{total wear time in minutes}$). For categorizing accelerometer data into different intensities of PA, Troiano cutoff values were used to calculate the number of minutes spent in sedentary (0–99 CPM), light (100–2019 CPM), moderate (2020–5998 CPM), or vigorous PA (≥ 5999 CPM) (18). As a result, average minutes spent per day in each intensity could be calculated as follows: total minutes (intensity) \div total wear time in calendar days. Using the average minutes spent in MVPA per day, subjects who met the WHO recommendations could be identified.

Questionnaire and measures

Questions on socioeconomic background and lifestyle consisted of information about sex, age, and highest finished educational degree, as well as weight and height. To assess generic health, the Medical Outcomes Study Short Form-36 (SF-36) was included. The SF-36 assesses difficulties attributable to mental or physical health across 8 domains: social function, physical function, bodily pain, role physical, role emotional, general health, vitality, and mental health. From the domains, 2 summary scores can be calculated: the physical component summary (PCS) and the mental component summary (MCS), ranging from 0 to 100 (higher scores reflecting better health) (19, 20).

To assess aspects of AS-related health, patients indicated their diagnosis duration (time since diagnosis, in years) and completed the Bath AS Disease Activity Index (BASDAI) and the Bath AS Functional Index (BASFI) 21,22. Finally, information about the current use of medication, i.e., use of tumor necrosis factor- α blocking therapy, was obtained.

Statistics

All analyses were performed with PASW Statistics 20 (SPSS). After checking normality of the data, comparisons between patients and controls were performed with independent Student t tests or Kruskal-Wallis test (SF-36 PCS, MCS, calendar days the accelerometer was worn, and time active in MVPA). To adjust the comparisons of the different accelerometer outputs between patients and controls for multiple comparisons, the Benjamini-Hochberg procedure was performed using a false discovery rate of 20% (23). Given the relatively low amount of time spent in vigorous PA, these minutes were combined with the time spent in moderate PA for further analyses. When exploring the differences between patients and controls in TPA (CPM) or the time spent in various intensities of PA, linear regressions were used to adjust for possible differences in age, sex, and BMI between both groups.

Multivariable regression was also used in an explanatory model to assess whether the effect of generic functional abilities (SF-36 PCS) and BMI on TPA was independent of age, sex, and having a diagnosis of AS. Linearity was checked as well as possible interactions of groups with BMI, PCS, age, and sex.

Finally, to understand the influence of disease-related variables on minutes spent in sedentary, light, and (square root transformed) MVPA in patients with AS, multivariable regression analysis was performed with age, sex, BMI, diagnosis duration, BASDAI, and BASFI as potential determinants. Because minutes spent in MVPA were slightly skewed, the variable was square root transformed. P values ≤ 0.05 were considered statistically significant.

Results

Samples

The final sample consisted of 135 patients with AS and 99 population controls (figure 1). The characteristics of both patients and controls are presented in table 1. Patients were older and had a worse self-reported physical health (SF-36) in comparison with population controls.

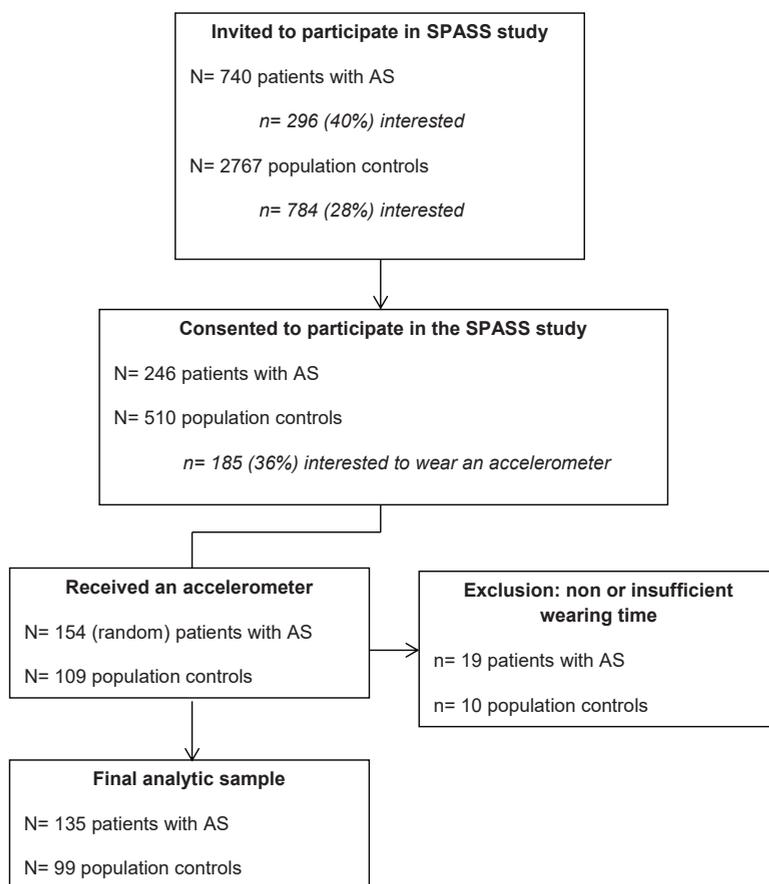


Figure 1. Subjects flow

PA at different intensities in patients with AS and population controls. The median (interquartile range) of the number of calendar days patients wore the accelerometer was 7 (6-7) days and for controls 7 (7-8) days. Patients with AS did not differ from controls regarding the TPA expressed in CPM (table 2), not even after adjusting for age, sex, and BMI. Regarding the sum of time spent in all different intensities, which was also equivalent to the total wearing time in minutes, the population controls registered on average 25 min/day more activity than patients ($p = 0.01$). This difference could be attributed mainly to more time (17 min/day) in sedentary activity by controls, although this was not statistically significant. The time spent in light activities was the same in both groups. Patients spent 7 fewer minutes a day in MVPA ($p = 0.006$) compared with controls, which resulted in a lower percentage of patients meeting the norm of 150 min of at least moderate PA a week (46.7% vs 59.6% in controls, $p = 0.05$). In addition, the findings between groups remained significant after correcting for the false discovery rate and did not change after controlling for age, sex, and BMI.

Table 1. Demographic and health characteristics of the patients with ankylosing spondylitis and population controls

	<i>Patients with AS (N=135)</i> mean (SD) [min - max]	<i>Controls (N=99)</i> mean (SD) [min - max]	p
Age (years)	51 (13) [24 - 79]	45 (12) [23 - 84]	<0.001
Gender (Males)	81 (60%)	66 (67%)	0.34
Education (at least 12y)	118 (87%)	86 (87%)	0.90
Employed n (%)	78 (58%)	84 (85%)	<0.001
BMI (kg/m ²)	26.0 (4.3) [17.0 - 41.8]	25.1 (4.3) [16.8 - 47.7]	0.09
Diagnosis duration (years)	16.5 (12.1) [0 - 61]	-	-
BASDAI (0-10)	4.3 (2.2) [0- 9.5]	-	-
BASFI (0-10)	4.1 (2.6) [0- 10]	-	-
Medication biological n (%)	69 (51%)	-	-
SF-36 PCS (0-100)	38.8 (10.7) [9.9 - 61.9]	55.7 (6.0) [24.1 - 69.5]	<0.001
SF-36 MCS (0-100)	50.2 (12.7) [10.9 - 70.5]	52.2 (8.7) [20.1 - 63.2]	0.99

BMI, Body Mass Index; BASDAI, Bath Ankylosing Spondylitis Disease Index; BASFI, Bath Ankylosing Spondylitis Functioning Index; SF-36, 36 item Short Form health survey physical component score and mental component score

Table 2. Results of total physical activity and time spent in different intensities of physical activity as assessed by the 7-days accelerometer registration, separately for patients with ankylosing spondylitis and population controls

	<i>Patient with AS (N=135)</i>	<i>Controls (N=99)</i>	<i>p</i>	<i>p*</i>
	mean (SD) [min - max]	mean (SD) [min - max]		
VM3 counts/minute	589 (202) [157 - 1158]	608 (219) [150 - 1465]	0.98	0.86
VM3 counts/day	492400 (173511) [116773 - 932839]	521847 (178955) [126538 - 1025867]	0.21	0.245
Time active min/day	837 (77) [646 - 1058]	862 (68) [647 - 993]	0.01	0.04
Sedentary min/day	524 (99) [279 - 783]	541 (94) [247 - 751]	0.17	0.238
Light min/day	290 (86) [40 - 520]	290 (74) [80 - 462]	0.95	0.86
Moderate min/day	22 (18) [0 - 122]	28 (20) [4 - 109]	0.02	0.05
Vigorous min/day[#]	30 (22%), 1 (4) [0 - 22]	2 (5) [0 - 32]	<0.001	0.01
At least 150 min of MVPA a week n (%)	63 (46.7%)	59 (59.6%)	0.05	-

VM3, Vector Magnitude Counts= $\sqrt{\text{total counts axis 1}^2 + \text{total counts axis 2}^2 + \text{total counts axis 3}^2}$; MVPA, moderate to vigorous physical activity. p* is the Benjamini-Hochberg adjusted P-value for multiple comparisons. [#] amount (n, %) of subjects who engaged in vigorous PA are provided.

Association between total PA and possible determinants in patients with AS and controls

In the multivariable analyses on TPA (CPM), the age, male sex, and group (having AS) were not significant contributors. Better self-reported functional ability was associated with more TPA (β 4.3, $p=0.005$). For every point the PCS of the SF-36 increased, TPA increased 4.3 CPM. In addition, a higher BMI was associated with less TPA (β -7.9, $p=0.01$), indicating that every point increase in the BMI was associated with a decrease in TPA of 7.9 CPM. There were no significant interactions between group and any of the other variables.

Determinants of various intensities of PA in patients with AS

In the multivariable models with minutes spent in sedentary PA and light PA as dependent variables, a significant interaction effect of age and BASDAI was found and consequently the regression models were stratified for age (at the median of 52 years). In patients aged under 52 years ($n=66$, 56% men), a higher BMI was associated with more time per week spent in sedentary PA (β 8.6, $p=0.007$; table 3). In contrast, patients 52 years or older ($n=69$, 64% men), being male (β 45.6, $p=0.04$), and having a higher BMI (β 5.6, $p=0.02$) were associated with more time spent in sedentary PA. In addition, the same analyses showed that a lower BASDAI was associated with less time spent in sedentary PA (β -20.7, $p=0.006$). Regarding the analyses with time spent in light PA (table 3), none of the entered explanatory variables were contributory in the group of patients of < 52 years. In the group of patients \geq 52 years, a significant association with higher BASDAI and more time in light PA was found

(β 14.61, $p=0.04$). Of note, the sum of total time spent within all activity intensities did not differ between the strata of patients older and younger than 52 years ($p=0.80$).

Table 3. Results of the multivariable regressions investigating the association of several explanatory factors and minutes spent in (a) sedentary, (b) light and (c) moderate to vigorous PA per day in patients with ankylosing spondylitis.

Explanatory factors	age <52y: n=66 (of which 56% males)				age \geq 52y: n=69 (of which 64% males)			
	b	SE	p	Part R	b	SE	p	Part R
<i>a: Sedentary PA</i>								
Constant	318.6	77.5	<0.001	-	383.9	69.8	0.000	-
Gender (male)	-27.1	27.9	0.34	-0.13	45.6	22.1	0.04	0.25
BMI	8.6	3.0	0.007	0.35	5.6	2.4	0.02	0.28
Diagnose duration (y)	1.9	1.4	0.19	0.17	0.9	0.8	0.30	0.13
BASDAI (0-10)	-11.8	8.3	0.16	-0.18	-20.7	7.3	0.006	-0.34
BASFI (0-10)	6.9	6.8	0.32	0.13	8.8	6.2	0.16	0.18
	R= 0.45 (R ² = 0.20)				R= 0.55 (R ² = 0.31)			
<i>b: Light PA</i>								
Constant	389.8	70.4	<0.001	-	374.9	68.0	<0.001	-
Gender (male)	11.4	25.3	0.65	0.06	-5.6	21.5	0.79	-0.03
BMI	-2.3	2.8	0.40	-0.11	-4.1	2.4	0.09	-0.21
Diagnose duration (y)	-1.0	1.3	0.43	-0.11	-1.0	0.8	0.23	-0.15
BASDAI (0-10)	0.5	7.5	0.95	0.01	14.6	7.2	0.04	0.25
BASFI (0-10)	-9.4	6.2	0.14	-0.20	-4.9	6.0	0.42	-0.10
	R= 0.35 (R ² = 0.12)				R= 0.386 (R ² = 0.15)			
<i>c: Moderate to vigorous PA (in patients across all age categories)</i>								
Constant	7.3	1.2	0.000	-				
Age	0.01	0.01	0.40	0.08				
Gender (male)	0.32	0.35	0.36	0.08				
BMI	-0.1	0.04	0.007	-0.24				
Diagnosis duration (y)	-0.04	0.02	0.02	-0.21				
BASDAI (0-10)	0.1	0.11	0.30	0.09				
BASFI (0-10)	-0.2	0.09	0.03	-0.19				
	R= 0.43 (R ² = 0.19)							

BMI, body mass index; BASDAI, Bath Ankylosing Spondylitis Disease Index; BASFI, Bath Ankylosing Spondylitis Functioning Index

Finally, no interactions were observed when exploring the time spent in the square rooted MVPA. In the model (table 3), negative associations with higher BMI (β -0.01, $p=0.007$), longer diagnosis duration (β -0.04, $p=0.02$), and higher BASFI (β -0.2, $p=0.03$) were observed.

Discussion

The findings of our study confirmed our hypothesis that patients with AS did not differ from population controls in the total amount of objectively measured PA, but spent less time in MVPA. Further explorations revealed that the negative influence of impaired functional ability and BMI on TPA was similar in patients as in controls. Among patients, increased BMI, diagnosis duration, and BASFI, but not reported disease activity, reduced the time spent in MVPA. Remarkably, in older patients, a higher disease activity was associated with less time spent in sedentary but more time spent in light intensities of PA.

When interpreting the current findings from the perspective of existing literature, it should be noted that different assessment tools are used and, in comparison with objective accelerometers, retrospective self-reports are hampered by the possible overestimation of PA because of recall bias (24).

Our findings on TPA assessed with an accelerometer are in agreement with Plasqui, *et al*, who also found no differences in a much smaller group of 48 patients with AS and controls (13).

In contrast, a study by Swinnen, *et al* ($n = 80$) concluded that, in total, activity of patients with axSpA was lower compared with controls. However, in that study, both patients [median PA level (PAL) of 1.45] and controls (median PAL of 1.54) had a remarkably low PAL in comparison with reference values found in the general population (mean PAL ~ 1.7), indicating that care should be taken when interpreting these results (8, 25). For referencing, the study by Plasqui, *et al* found a mean PAL of 1.73 in patients with AS and 1.74 in controls, results in line with reference values as measured in the general population (13).

As for time spent in different intensities of PA, patients in our study spent less time in more vigorous intensities of PA, which was in line with the additional results of the study by Swinnen, *et al* (8). When considering the proportion of patients complying to the current WHO recommendations as assessed by a validated recall questionnaire, Haglund, *et al* (9) reported that 68% of 2126 patients with SpA complied with these recommendations (reflecting at least moderate PA) (2), which was slightly better than the proportion reported from the general population⁹. Using an accelerometer, we found that only 47% of the patients and 60% of the directly compared controls reached this level. Validated recall questionnaires are regarded as more practical for clinical use, yet the selection of commercially available accelerometers and activity monitors is rapidly increasing (26), which might provide opportunities for a more extensive use of these devices in future research.

Regarding factors contributing to the TPA in patients and controls, the negative effect of BMI was apparently not amplified in patients. In the general population, it is known that overweight ($BMI \geq 25 \text{ kg/m}^2$) is associated with lower PA (27). Moreover, in AS it was shown that overweight relates to poorer perceptions about the benefits of exercise and provides a greater awareness of the barriers to exercise (28). Overweight and/or perceptions of overweight may hamper PA engagement (29, 30), and seemingly this effect is the same in controls as in patients.

Among similar lines, no group differences were observed with regard to functional ability measured by the SF-36 PCS, which was associated with less TPA. Although the interaction between the group variable with SF-36 PCS was not significant, the SF-36 PCS in the control group was distributed in a clustered pattern (i.e., controls did not experience functional problems), whereas a broader distribution over the range of functional ability was observed in patients. For this reason, the absence of a significant group effect should be interpreted with some caution.

Within patients, BMI, limitations in physical function (BASFI), and an increasing diagnosis duration were associated with reduced MVPA. The reduced time in MVPA within patients was previously confirmed by Prince, et al, who interviewed 52 patients with AS and confirmed that patients reported a decline in both time and intensity in sports and recreational PA participation after diagnosis (31). The independent influence of diagnosis duration might be relevant to accumulate radiographic damage in AS and likely independently affects physical function and thus engagement in PA (5, 32). However, the BASDAI was not independently relevant for time spent in MVPA. We found that in older patients, but not younger patients, an increase in the BASDAI was significantly associated with less time spent in sedentary PA and also with more time spent in light intensity PA. The availability of more leisure time to be physically active is not a likely explanation because in both age groups, a similar number of patients had paid work. Apparently, older patients have experienced more than younger patients that light intensities of PA, such as walking and mild stretching, have beneficial effects on their functioning and well-being, despite somewhat more pain and stiffness (33). Contrasting results can be found in a questionnaire study in which 78 patients with AS with a higher disease activity (mean age 51.4 years) reported a lower amount of TPA and spent less time in walking and vigorous intensity activities than patients with a lower disease activity (mean age 46.9 years) (12). In our study, patients with a BASDAI ≥ 4 did not differ regarding the total PA or intensities of PA compared with patients with a BASDAI < 4 .

Regular exercise is proposed by the Assessment of SpondyloArthritis international Society and European League Against Rheumatism recommendations as part of the nonpharmacological treatment of AS (34). Activities of a higher intensity as some forms of exercise have known beneficial health effects, including cardiovascular risk factors (35). Since our results revealed that patients spent less time in more vigorous intensities of PA than controls, more emphasis should be given to exercise programs that do not concentrate only on stretching and postural correction, but also on higher intensities of PA, especially because novel evidence in another study indicated that in healthy middle-aged adults, PA of vigorous intensity was related to declined risk of mortality (3). While we call for more engagement in MVPA in patients with AS, we also need to admit that more research is needed on whether such activity would be beneficial for all patients in all stages of their disease. Another recent study found that in patients who were blue-collar workers, a surrogate for more PA, radiographic progression of spinal damage was significantly higher

compared with white-collar workers (14). However, it is unclear whether this depended on the type of vigorous physical activities; the type of overall health benefits is also uncertain. Yet differences in radiographic progression were small and likely positive effects of PA will outweigh these small differences, but more research is needed on this aspect.

Some limitations need to be addressed. Because of the cross-sectional design of our study, statements about causality of relationships cannot be drawn.

Further, we did not examine inflammation markers such as CRP and ESR, and therefore were unable to investigate the role of objective disease activity on the PA of patients. While the inclusion of the control group was a strong point of our study, we were not able to fully match the control population to the age and sex of the AS group, but none of these variables contributed to the measured PA outcomes and were controlled for in all further analyses. Moreover, the recruitment of both patients and controls to wear an accelerometer was voluntary, which could have led to selection bias based on motivation to wear an accelerometer. However, this applied to both patients and controls so that any bias likely affected both groups, preserving internal validity.

Patients with AS performed the same TPA as population controls, but spent less time in MVPA. Higher BMI, impaired physical function, and a longer diagnosis duration are the main factors hampering engagements in MVPA in patients having AS.

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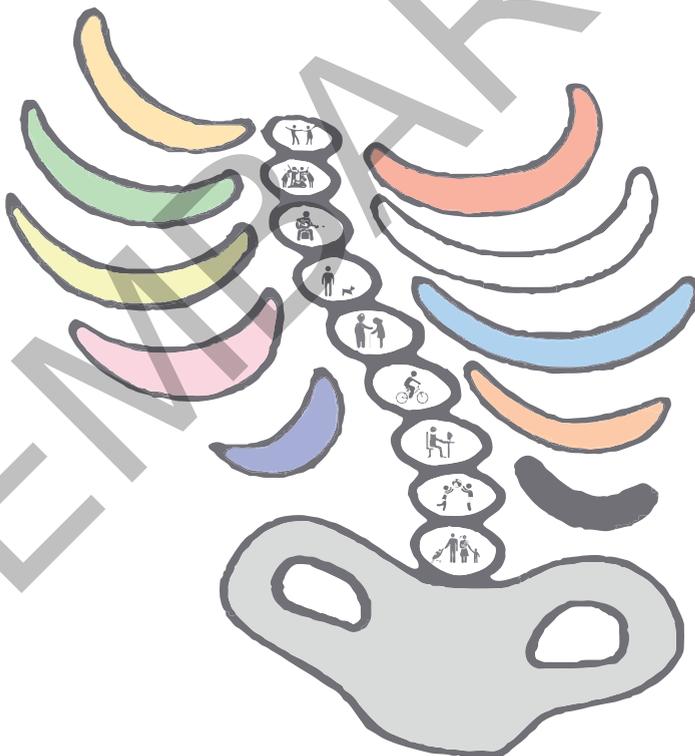
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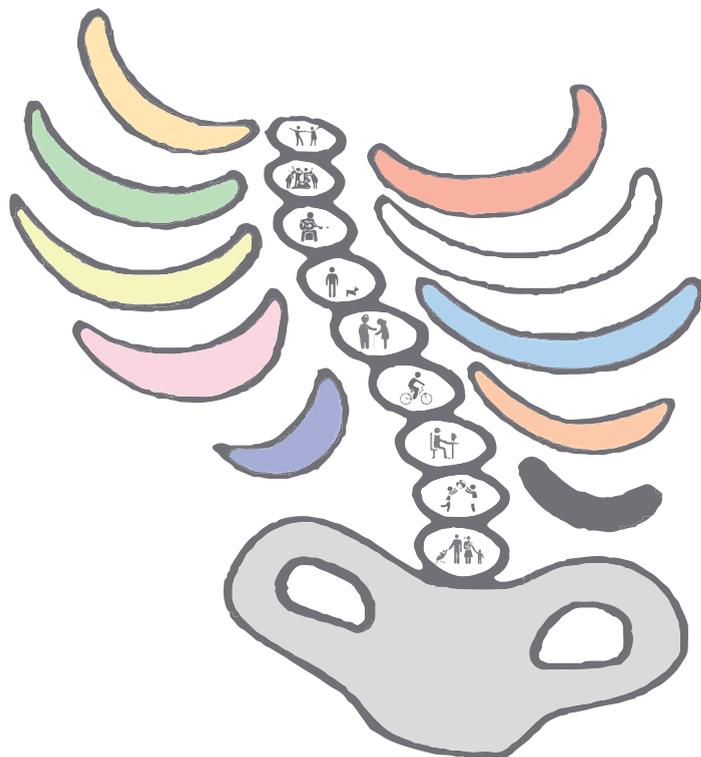
Fat free mass and its determinants in patients with rheumatoid arthritis or spondyloarthritis; a systematic overview of the literature

van Genderen, S., Stolwijk, C., Boonen, A., Plasqui, G. Fat free mass and its determinants in patients with rheumatoid arthritis or spondyloarthritis; a systematic overview of the literature. In preparation for submission



8

Summary of the chapters in this thesis



In order to provide interested persons a quick overview of the thesis, we summarize below the studies that are part of this thesis.

Part 1 social role participation

Although participation in social roles is essential to the lives of persons, participation of persons with chronic diseases in social life has received relatively little attention. However, at the start of this thesis, no validated Dutch questionnaire was available to assess the impact of ankylosing spondylitis (AS) on social role participation. In **chapter 2** we describe the translation and cultural adaptation of the (English) Social Role Participation Questionnaire (SRPQ) into Dutch, and present the validity and clinimetric properties as assessed in a study among 246 patients with AS. The SRPQ addresses participation in eleven roles and one general item (all scores range 1-5) across four dimensions (importance, satisfaction with performance, satisfaction with time, and difficulty). The translation was performed using the dual panel approach and required only minor adaptations. Cronbach's alphas were 0.7 or higher for all dimensions. A strong correlation was confirmed between the dimensions satisfaction with 'time' and 'performance' ($r = 0.85$). Test-retest reliability was tested in 31 patients and was found to be satisfactory ($\text{Kappa} = 0.79-0.95$). Correlations with participation domains of the Short-Form Health Survey 36 (SF-36), the World Health Organization Disease Assessment Score II, and generic- as well as disease specific health outcomes, Physical (PCS) and Mental component (MCS) of the SF-36, satisfaction with life scale (SWL), Bath Ankylosing Spondylitis Disease Index (BASDAI) and the Bath Ankylosing Spondylitis Functioning Index (BASFI), were as expected at least moderate ($r = 0.41$ to 0.75) for the 'role satisfaction' and 'role difficulty' dimensions, but weak for 'role importance' ($r \leq 0.40$). Discrimination across 5 overall self-reported health states (based on the SF-36 overall health item) was good for all dimensions ($p < 0.01$). Of interest, the 'general participation' role, the average of all 11 roles for each of the 4 dimensions, showed similar reliability and validity for each dimension. We concluded, the Dutch version of the SRPQ is available to better understand social role participation in patients with AS. The dimension 'role importance' measures a distinct aspect of participation and it should be retained. The dimension satisfaction with time seems redundant and, as suggested in an earlier publication, can be removed. The general participation item is a good overall measure of participation for each of the dimensions of interest.

In **chapter 3** we aimed to compare the social role participation in 246 patients with AS with 510 population controls using the SRPQ. In addition to completing the SRPQ, all subjects ranked their three most important roles. The ranking of role-importance, the average SRPQ dimension scores, and the gap between 'importance' and 'satisfaction with performance' of roles were compared between patients and controls. Patients and controls ranked 'intimate relationships', 'relationship with (grand/step-) children' and 'employment' as most important roles. Compared to controls, patients provided higher scores on the SRPQ to

‘importance’ (3.75 vs 3.43), but reported lower on ‘satisfaction with performance’ (3.19 vs 3.58) and greater ‘physical difficulty’ (3.87 vs 4.67) (all $p \leq 0.05$). The largest gaps between ‘importance’ and ‘satisfaction with performance’ for patients compared to controls were seen for physical leisure, hobbies, travelling and vacation; patients conferred a higher importance but especially lower satisfaction. As individuals and society place increasing focus on an individual’s responsibility to participate fully in social roles, the current data suggest that healthcare providers should pay more attention to participation-restrictions experienced by patients with AS.

To increase insight into the importance of social role participation for the lives of patients, in **chapter 4** we examined the association between several aspects of social role participation and satisfaction with life in 246 patients with AS compared to 510 population controls. For individuals with AS and controls, the association between SRPQ role-domains and the SWL scale were examined using linear regression for each participation dimension separately, in the total and the employed population, adjusting for relevant confounders including age, gender, education and income. Patients were more frequently (extremely) dissatisfied with life (17.9% vs 8.6%; $p < 0.05$). In the total and the employed population, less physical difficulty and higher satisfaction with interpersonal relations and leisure were associated with higher SWL and this was somewhat stronger in patients than in controls ($p < 0.1$). In employed controls but not in employed patients, satisfaction with work was independently associated with SWL. Findings of this study highlight the importance of social roles and suggest it might be worthwhile to educate patients about the importance of satisfactory relationships and leisure activities in order to improve their satisfaction with life.

Part II Physical activity and body composition

In the second part of the thesis we explored physical activity (PA) and body composition in patients with AS, with an emphasis on possible differences with population controls. In **chapter 5** patients with AS and controls were compared regarding three approaches of functioning: experienced ability to perform activities (BASFI), self-reported amount of physical activity (Baecke questionnaire) and the objectively measured amount of PA (tri-axial accelerometer). In this case-control study 24 AS patients and 24 healthy controls (matched for age, gender and BMI) were recruited. Subjects completed the BASFI and Baecke questionnaire and wore a tri-axial accelerometer. Subjects also completed other self-reported measures on disease activity (BASDAI), fatigue (MFI) and overall health (EQ-VAS).

Both groups included 14 (58%) males and the mean age was 48 years. Patients scored significantly worse on the BASFI (3.9 vs. 0.2) than their healthy peers, while PA assessed by Baecke and the accelerometer did not differ between groups. Correlations between approaches of physical functioning were low to moderate. BASFI was associated with disease activity ($r=0.49$) and physical fatigue (0.73), Baecke with physical and activity related fatigue

($r=0.54$ and $r=0.54$), but total PA assessed by accelerometer was not associated with any of these experience based health outcomes. We concluded that different approaches of the concept physical functioning in patients with AS provide different information. Compared to matched controls, patients with AS report more difficulties and fatigue but report and objectively perform the same amount of physical activity.

In **chapter 6** the total amount of physical activity (TPA) and time spent in various activity intensities of patients with AS and population controls were compared and factors related to PA were explored. Subjects were asked to wear a tri-axial accelerometer for 7 days and completed a series of questionnaires. Multivariable regressions were used to assess generic determinants of TPA in patients and controls, in order to explore demographic and disease specific determinants of various PA intensities.

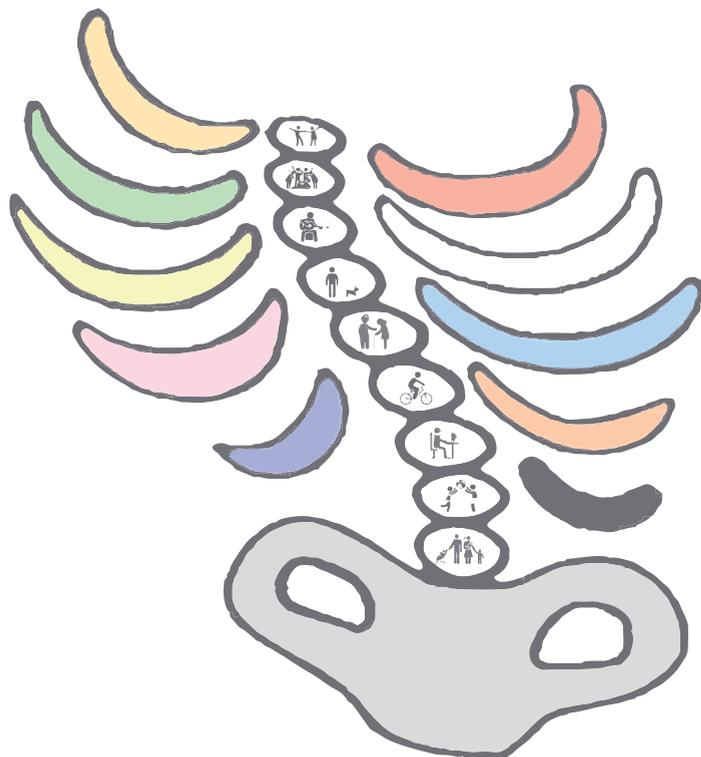
A total of 135 patients (51 ± 13 y; 60% males; BMI 26.0 ± 4.3 kg/m²) and 99 controls (45 ± 12 y; 67% males; BMI 25.1 ± 4.3 kg/m²) were included. Patients and controls did not differ regarding TPA (589 vs. 608 vectorcounts/min, $p=0.98$), min/day spent in sedentary (524 vs. 541, $p=0.17$) and light PA (290 vs. 290 $p=0.95$), but patients spent less min/day in moderate to vigorous PA (MVPA) (23 vs. 30, $p=0.006$). Perceived functional ability (PCS of the SF-36) and BMI were associated with TPA regardless of being patient or control (p interaction= 0.21 and $=0.94$ respectively). Time spent in MVPA was negatively influenced by BMI, physical function (Bath AS functional Index; BASFI) and disease duration. In patients equal to -or older than- 52 years of age, a higher disease activity (BASDAI) was associated with less time spent in sedentary but more time spent in light activities. Compared with controls, patients with AS had similar TPA but may avoid engagement in higher intensities of PA. Lower levels of functional ability and higher BMI were associated with lower TPA in both patients and controls.

In **chapter 7** a systematic overview of the literature on body composition among rheumatoid arthritis (RA) or spondyloarthritis (SpA) when compared to population controls is provided, as well as on factors that are associated with (a change in) fat free mass (FFM). A systematic search in MEDLINE/PubMed database was performed and two independent reviewers assessed risk of bias. Evidence was presented as narrative summaries per disease (RA or SpA), study type (observational or experimental) and study objective. In RA, 17 of 23 studies reported a lower FFM in comparison with controls, but in SpA this was found in only one of the 5 studies. Observational studies in RA revealed that age, female gender and disease activity marker C-Reactive Protein (CRP), disease activity score (DAS 28), disease duration and severity, and disability measured with the health assessment questionnaire (HAQ) were all independently associated with a lower FFM. The conducted experimental studies suggested that progressive resistance training resulted in an increase of FFM. In none of the limited amount of studies, examining the use of disease-modifying anti-rheumatic drugs or dietary interventions reported an effect on FFM.

In conclusion, the selected studies indicated that the FFM of patients with RA was mostly lower than that of controls. In patients with SpA, no such abnormalities were reported but the amount of evidence was more scarce. In RA age, gender, inflammation and disability were independently associated with lower FFM. In addition, progressive resistance training seemed effective to increase FFM over time.

9

General discussion



In this thesis, several gaps in knowledge in relation to social role participation and physical activity in patients with ankylosing spondylitis (AS) were addressed.

First, aspects of social role participation were studied. As no Dutch questionnaire was available to assess social role participation in AS, we were interested in the clinimetric properties and aspects of validity of the translated and culturally adapted Dutch version of the English social role participation questionnaire (SRPQ). Next, the roles and dimensions of social role participation were compared between patients with AS and population controls. Furthermore, the relevance of social role participation for satisfaction with life (SWL) was investigated.

Second, multiple approaches to measure physical activity (PA) were studied in patients with AS. Objectively measured habitual PA and patterns of PA were tested and compared with population controls. Factors explaining differences between patients and controls were explored. In addition, literature on body composition, in particular fat free mass (FFM), in patients with AS and rheumatoid arthritis (RA) was systematically collected and findings regarding possible differences with controls, associated variables and possible strategies to intervene on FFM in patients were sampled from the selected articles. The main findings are summarized below.

- The SRPQ was found to be relevant and comprehensible by patients with AS and showed acceptable internal consistency and test-retest reliability. The dimensions ‘satisfaction with time spent’ and ‘satisfaction with performance’ correlated strongly. ‘Satisfaction with performance’ had better construct validity and discriminated less between patients and controls, suggesting redundancy of the dimension ‘satisfaction with time’. As expected, the dimension ‘role importance’ provides different but likely additional information. The ‘general participation’ role had adequate reliability, construct validity and can be considered as a more feasible approach to measure social role participation.
- The most important social roles for both patients with AS and controls were ‘intimate relationships’, ‘relationship with (grand/step-) children’ and ‘employment’. Patients attached a somewhat higher importance to social roles than controls, but were substantially less satisfied with their performance and experienced more physical difficulty. The greatest difference in the gaps between importance and satisfaction with performance between patients compared to controls were observed for physical leisure, hobbies, travelling and vacationing.
- A higher proportion of patients with AS report (extreme) dissatisfaction with life compared to population controls. Social role participation, and especially interpersonal relations and leisure activities, are associated with greater life satisfaction, and this is somewhat stronger in patients than in controls. In employed controls, but not in employed patients, work participation additionally contributes to life satisfaction.

- Different approaches that capture the concept of physical activity, provide different information. Patients with AS perform a similar amount of objectively assessed total amount of PA as population controls. However, patients with AS spent less time in moderate to vigorous activity intensities. Within patients, a higher body mass index (BMI), worse physical function and longer disease duration were associated with less time spent in moderate to vigorous activity intensities.
- A systematic literature review indicated that the FFM in studies with patients with RA was mostly lower than that of controls. In patients with SpA, no such abnormalities were reported but the amount of evidence was more scarce. In RA age, gender, inflammation and disability were independently associated with lower FFM. In addition, progressive resistance training seemed effective to increase FFM over time.

To enhance interpretation of the results within the methodological choices and limitations, and to contextualize results within available evidence, the paragraphs below provide some more in depth discussions of the overall findings.

The impact of AS on the life of a patient; social role participation

In AS, research into social role participation has mainly focused on restrictions in work participation, and only to a lesser extent on other social roles and other dimensions of participation. To study social role participation in Dutch patients with AS, a valid instrument encompassing all relevant roles and addressing relevant dimensions was mandatory.

This thesis described the development of the Dutch SRPQ and provides evidence the Dutch SRPQ is a valid and reliable self-reported questionnaire to measure multiple social roles across different participation dimensions, and therefore is a worthwhile tool to consider in clinical research. When starting this thesis, several instruments to assess participation were available (1). Two frequently used health related quality of life instruments, the Short-Form Health Survey 36 (SF-36) and the World Health Organization Disease Assessment Score II (WHODASII), incorporate items addressing perceived difficulties in 'social role participation'. However, they do not distinguish between different types of roles and address only the dimension 'difficulties' (2, 3). In addition, several self-reported instruments specifically address social role participation in patients with musculoskeletal disease comprise the Impact on Participation and Autonomy (IPA) (4), Keele Assessment of Participation (KAP) (5), Participation Measure for Post-Acute Care (PM-PAC) (6), Participation Objective, Participation Subjective (POPS) (7), Rating of Perceived Participation (ROPP) (8), and The Participation Scale (9). A review of these instruments showed no advantage of any of the instruments above, although unfortunately the SRPQ was (not yet) included in the overview (10). Content-wise, the SRPQ shows similarities with the IPA, KAP and the POPS as they all address a large number of social roles across one or more subjective dimensions of participation i.e. satisfaction.

The POPS, however, additionally contains items on ‘frequencies’ of participation, as a more objective approach. Information on the amount of time or frequencies spent in social roles is interesting when comparing lifestyle or economy-related roles such as PA or work. However, for other social roles, such as relationships with family, the amount of time persons participate does not have an immediate personal and societal value, and in the absence of a benchmark, this would make interpretation of results difficult. For this reason, satisfaction with time was considered a more relevant dimension to be addressed. Notwithstanding, as shown in chapter 2 and in a previous validation study, the ‘satisfaction with time’ was not providing additional information to ‘satisfaction with performance’ and was considered redundant (11). Since we showed that ‘satisfaction with time’ discriminates less well between controls than expected, we support previous suggestions to remove the ‘satisfaction with time’ dimension of the SRPQ (11).

Although the SRPQ in its current form provides the most complete view of patient’s social role participation in terms of number of roles and number of dimensions, its lengthiness may hamper application in trials and studies, even after removing the ‘satisfaction with time’ dimension. Additional research using item responds theory (IRT) suggested that a shorter SRPQ with six social roles and two dimensions also had satisfactory validity and reliability properties (12). Alternatively, the general participation domains had comparable clinimetric properties and may be valuable when there is no need to distinguish between different role domains.

After publication of validation and application of the Dutch SRPQ, a translated Chinese version was developed and applied in AS, and was shown to perform similarly (13). As such, this adds evidence that dimensions included in the SRPQ represent universal constructs. Furthermore, items in the SRPQ were non-disease specific and could therefore be tested in other disease groups as well, enhancing comparison between diseases. Of note, the SRPQ still warrants extra validation in the form of sensitivity to change for use in longitudinal or intervention studies.

Further findings in this thesis include a confirmation of the importance of work participation for the lives of patients, as well as the observation that relationships with (grand or step)-children and partner or spouse ranked even higher on the role importance, and received a higher average importance score. Comparable preferences were revealed by 44 Canadian patients with AS (11). In addition, patients also ranked engaging in physical leisure, social activities and travelling high on the importance-ladder. Our study was the first to compare role participation in AS with general population controls. Overall, patients did not differ importantly in ranking role importance when compared to population controls. Notwithstanding, it was remarkable that ‘work’ was less frequently the most important role and less frequently among the three most important roles for patients compared to controls. This could mainly be explained by the larger number of patients who had no em-

ployment and for whom work was clearly less important (also when compared to controls without work). Further differences between patients and controls became more apparent when averaging the scores of all roles for the different dimensions. Patients scored higher on the importance for all social roles, but experienced more difficulties and were less satisfied with their ability and (somewhat less pronounced) with time available to participate in social roles. To interpret the relation between relevance of (dis)satisfaction with the performance or difficulty in social roles in perspective to the role importance, we calculated 'for importance weighted' scores for the 'satisfaction' and the 'difficulty' dimensions. Simple presentation of a gap between role importance and satisfaction would not account for the absolute level of importance. This approach resembles the method used in the POPS and aims to optimize the descriptive representation of the finding that patients indicate a higher importance but also experience less satisfaction and more difficulty. Nevertheless, compared to the non-weighted dimensions, no improvements were observed regarding construct validity or in another study increasing the model fit of SWL in linear regression analysis. Therefore, despite the descriptive value of the importance dimension, weighted dimension scores warrant further validation in particular regarding possible associations with related constructs of participation if they should be considered as a separate outcome.

While reduced satisfaction with performance of social roles in patients with AS compared to controls is not surprising, the higher importance merits some reflections. Response shift may explain why patients value the importance of social roles higher. One can imagine that the real value of a role only becomes clear when it is not there anymore or when it takes a different form, which could be the case with social roles once impacted by disease ('perception of loss') (14). A similar observation was done by Gignac (2013), in a study comparing social role participation (SRPQ) between middle-aged (40-59y) and older (>60y) patients with osteoarthritis. The middle-aged group reported higher importance and lower satisfaction with performance and more health care utilization (15). Adaptation, one of the mechanisms underlying response shift, could be one of the explanatory factors. Generically, it would be insightful to further explore both qualitatively and quantitatively the underlying mechanisms of attributing importance and satisfaction to social roles in different stages of a life course, following a diagnosis of a chronic disease. Such information could provide health care professionals to further aware patients on the future impact of the disease on their lives but also suggest a role for life-tested coping strategies.

In this thesis SWL in patients with AS was studied for the first time, and it was found that compared with population controls, more patients were (extremely) dissatisfied with their lives in general and fewer were extremely satisfied. The literature already revealed that presence of a chronic disease such as arthritis, diabetes, heart disease or diabetes was negatively related with SWL (16). Our study suggests that dissatisfaction with social role participation contributes to lower life satisfaction. However, that was more specifically for roles relating to relationships and leisure activities and less to the work role. This was

unexpected as the importance of work compared to other social roles was ranked relatively high. Given that work has been related with SWL in other studies (17, 18), it would be interesting to evaluate prospectively how loss of work affects life satisfaction.

The impact of AS on the life of a patient; physical activity and body composition

In outcome research, the Bath Ankylosing Spondylitis Functioning Index (BASFI) is the most commonly used instrument to assess the patient's ability to perform daily tasks and activities. Since performing tasks and activities require a certain physical effort, instruments that measure PA may measure overlapping dimensions of the construct of physical function. This thesis showed that the BASFI correlates only moderately with PA assessed with a self-reported measure (Baecke questionnaire) and an objective instrument (tri-axial accelerometer), which are both validated measures of PA (19, 20). Similarly, only moderate associations were found between the self-reported and objectively performed PA, indicating that the three measures assess different dimensions of physical function. That is, the three-axial accelerometer addresses measures *minute by minute accelerations (expressed as counts)* of PA in almost all performed activities and social roles, the BAECKE concentrates more on the *time spent in social roles* (sports, work, leisure) and the BASFI on *difficulties* with specific physical tasks. For future research, care should be taken by selecting the proper instrument that best matches the aim of the study to assess PA or physical function.

In this thesis, two independent studies showed that patients with AS perform the same total amount of objectively measured PA compared to population controls. In recent literature, comparable results were reported in one other study in which accelerometers were used to measure the PA of patients with AS and controls (21). In contrast, the findings of Swinnen et al (2014) showed that the physical activity level (PAL) in patients with axSpA was lower compared to controls. However, in that study both patients (median PAL of 1.45) and healthy controls (median PAL of 1.54) had a remarkably low PAL in comparison to reference values found in the general population (mean PAL ~1.7), indicating that care should be taken when interpreting these results (22, 23).

In addition to performing 150 min of moderate to vigorous PA (MVPA) a week, the World Health Organization also points out the importance of performing more vigorous activities for additional health gain (24). This is in line with recently proposed Dutch guidelines for PA, which also recommends less sedentary (sitting) activities (25). The findings in this thesis on time spent in different intensities correspond with the literature. That is, patients with AS compared to controls perform a similar amount of sedentary and light PA (21, 23). These lower intensities, on average, represented the majority of all daily performed PA in both patients and controls. In contrast, but also in line with findings of other studies, patients with AS spent less time in moderate-to-vigorous activities (21, 23). In this thesis, the differences in minutes of MVPA a day between patients (23 minutes) and controls (30 minutes) was on average 7 minutes, which seemed small. However, over a period of a week this

accounted for an additional 13% (47% vs. 60%) of controls performing at least 150 minutes of MVPA a week. These results differ somewhat from the results of other studies in which PA was assessed with a tri-axial accelerometer. In the study of O'Dwyer (2015) patients with AS performed 59 minutes/day of MVPA on average and only 38,9% of the patients with AS managed to comply with 150 minutes of MVPA a week (21). Furthermore, Swinnen et al (2014), reported that patients with axial SpA performed an average of 98 minutes a day of moderate PA (23). These diverse results call for additional research, using uniform measures of objectively assessed PA.

Regarding associated clinical outcomes, this thesis shows that more time spent in MVPA -and not light intensities of PA- was associated with less impairments measured with the BASFI.

Other negatively related factors of the MVPA of patients with AS were a higher BMI and a longer disease duration.

We emphasize that more research is needed on whether activities of at least moderate intensity (equivalent to 3-6 Metabolic Equivalent Tasks) in the form of exercise or PA would be beneficial regarding clinical outcomes of all patients in all stages of the disease. Physiologic responses to exercise are determined by the mode (frequency, intensity, and duration) of exercise (26). Various systematic literature reviews on the effectiveness of exercise programs in patients with AS have been conducted so far (27-29). Exercise programs have proven beneficial effects on spinal mobility and physical function in patients with AS (27) and additional aerobic components to flexibility programs cause improvements of cardiorespiratory outcomes (28). In contrast, a study in patients that were blue-collar workers, a surrogate for manual labor, radiographic progression of spinal damage was significantly higher compared with white-collar workers. It is unclear, however, whether this effect depends on the type of vigorous physical activities or that confounding plays a role. In addition, differences in radiographic progression were small and positive effects of PA may outweigh these small (if any) negative effects.

In light of this, more attention should be given to stimulate and help patients with AS to perform extra MVPA, such as progressive resistance training, which is also beneficial for the enhancement in muscle and bone mass. This is also in line with the recently proposed adapted guidelines of PA by the Dutch Health Council (25). The effectiveness of programs to enhance PA of patients with AS has only sparsely been studied. However, a recent study of O'Dwyer et al provided interesting results about the adherence to PA guidelines within patients with various rheumatic disorders, amongst others SpA.

They showed that the adherence to these guidelines was also predicted by a lack of knowledge about possible benefits (30). Individually tailored weekly semi-structured consultations with a physiotherapist, aimed to motivate and support patients with AS, have shown to improve and sustain health enhancing MVPA to an extent of 3 months (31). More attention should be paid by health care professionals who are in the position to inform and positively influence patients regarding MVPA. According to O'Dwyer (2014), based upon existing litera-

ture, about half of the adults attending rheumatology clinics in the UK and USA reported that PA had never been discussed with them (30, 32-34). In addition, health care professionals in rheumatology may benefit from further training in exercise advice and PA promotion, as it is was reported that there is a need for improving skills and confidence in this area (33, 35).

Concluding and recommendations for future research

Regarding part I, we have shown that SRPQ is a valid and useful tool to measure social role participation. Similar as controls, patients with AS rank relationships with (grand/step-) children, intimate relationships and employment as the three most important roles. Notwithstanding, patients attached a somewhat higher importance to social role participation than controls. At the same time patients were less satisfied with their performance and experienced more physical difficulty, resulting in an important importance-satisfaction or importance-ability gap. When exploring the relation between of social role participation and life satisfaction, it was striking that a higher proportion of patients with AS report (extreme) dissatisfaction with life compared to population controls. Of interest, social role participation and especially interpersonal relations and leisure activities, were associated with greater life satisfaction, and this was somewhat stronger in patients than in controls.

Future studies should address longitudinal discrimination including sensitivity to change in intervention studies. In addition, a difficult discussion is whether social role participation, beyond control of disease activity, should be a target in clinical care. While clinical attention for work participation and physical leisure (exercise) is increasingly implemented, this is less obvious when it comes to personal relationships or leisure. Given the reduced life satisfaction and the importance of relationships and social life for life satisfaction, our data endorse the inclusion of these aspects in educational programs in order to enhance patient's self-management.

Regarding part II, we have shown that patients with AS perform the same total amount of objectively measured PA as compared to population controls but at a less vigorous level. More research is needed on whether MVPA would be beneficial regarding clinical outcomes of all patients in all stages of the disease. Due to the cross-sectional design of the studies conducted in this thesis, statements about causality of relationships could not be drawn. Future studies should focus on prospective evidence in patients of various age groups and in different stages of their disease. Using this design it would also be interesting to further explore the role of medication on PA, as in this thesis, the sample of patients was older and only a weak univariate relation with the use of biologicals was found. The systematic literature review suggested that the fat free mass of patients with RA is lower than that of controls but still reversible with resistance training. However, evidence in patients with SpA was scarce and did not indicate the presence of cachexia. More clinical studies on the body composition and related factors in patients with SpA are recommended.

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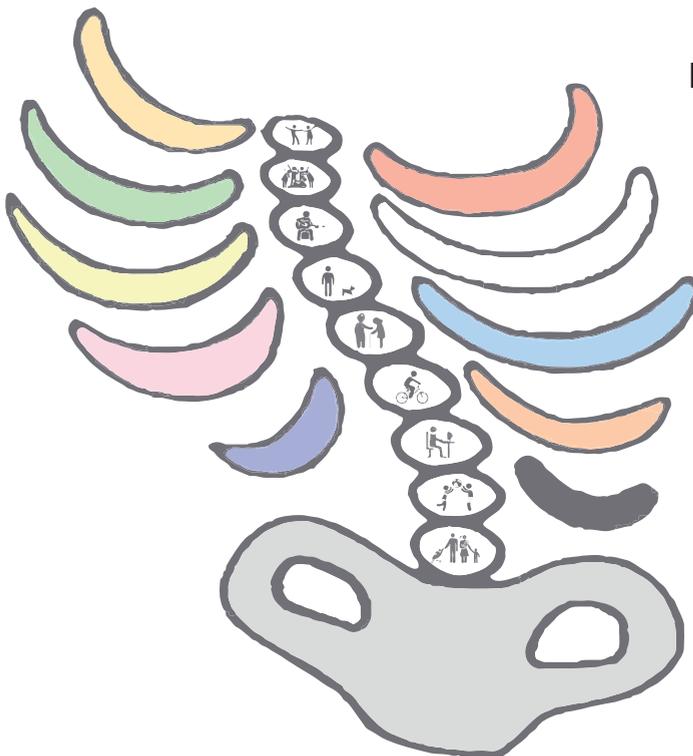
Nederlandse samenvatting

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Nederlandse samenvatting

Ankyloserende Spondylitis (AS), ook de wel ziekte van Bechterew genoemd, is een chronische inflammatoire reumatische aandoening die wordt gekenmerkt door een ontsteking van de sacro-iliacale gewrichten, de wervelkolom en, in een subgroep van patiënten, ook van de perifere gewrichten. De ontsteking veroorzaakt pijn, (ochtend) stijfheid, verminderde mobiliteit en vermoeidheid. Deze symptomen hebben een grote impact op het dagelijkse en sociale leven van patiënten. Het verloop van de ziekte volgt meestal een beperkt aantal trajecten of patronen, die vooral verschillen in de mate van ziekteactiviteit en impact op gezondheids-gerelateerde kwaliteit van leven. Binnen deze patiënt-trajecten kan de ziekteactiviteit en de kwaliteit van leven fluctueren met episoden van verbetering en verslechtering. Bij het grootste deel van de patiënten met AS ontstaat, deels ten gevolge van de ontsteking, ook schade aan de wervelkolom en de gewrichten, en de mate van cumulatieve schade in de wervelkolom of gewrichten draagt bij aan beperkingen in mobiliteit.

Na de diagnose komen patiënten in aanmerking voor zowel niet-medicamenteuze als medicamenteuze behandeling. Medicamenteuze behandelingen zijn erop gericht de klachten van pijn en stijfheid te onderdrukken. Van een specifiek type van deze medicaties werd aangetoond dat ze ook de lange termijn beloop van de ziekte modificeren en ook de gewrichtsschade lijken te remmen. De niet-medicamenteuze behandeling bestaat uit patiëntenvoorlichting, maar vooral ook uit oefeningen en lichaamsbeweging. Laatstgenoemde heeft bewezen effecten bij het handhaven en verbeteren van mobiliteit en het fysiek functioneren.

De belangrijkste uitkomst-domeinen om de impact van AS en behandeling ervan te bepalen betreffen de pijn, stijfheid en verminderde mobiliteit in de wervelkolom, verlies van het fysieke functioneren, vermoeidheid, algemene ziekteactiviteit door de patiënt, ontstekingswaarden in het bloed, artritis van de perifere gewrichten en (radiologische) schade van wervelkolom en gewrichten. Deze uitkomst-domeinen kunnen goed gemeten worden en ook hun relaties zijn uitgebreid bestudeerd. Echter, naast deze zogenoemde 'kern-domeinen', zijn er nog meerdere uitkomsten van de ziekte die van waarde zijn voor de patiënten. Enkele daarvan kregen tot op heden onvoldoende aandacht, en worden besproken in dit proefschrift.

Ten eerste, gezien de klachten van AS vaak ontstaan in het derde decennium van het leven, een periode waarin individuen zich inzetten binnen verschillende sociale rollen, zoals het gezinsleven, professionele carrière, maar ook in andere sociale rollen, zoals vrijetijdsbesteding, staan patiënten voor de uitdaging hoe ze deze rollen kunnen vervullen in het licht van hun aandoening. Beperkingen in sociale participatie als gevolg van AS hebben weinig aandacht gekregen binnen onderzoek en de klinische zorg.

Ten tweede, lichaamsbeweging is essentieel voor het dagelijks fysiek functioneren. Bij patiënten met AS is bekend dat zij moeilijkheden ondervinden in het uitvoeren van hun dagelijkse taken en activiteiten, maar het is minder duidelijk of hun feitelijk gemeten (objec-

tieve) lichaamsbeweging ook is verminderd. Een voorwaarde om fysiek actief te zijn, is de aanwezigheid van spiermassa, ook bekend als vetvrije massa. Verminderde vetvrije massa zou negatief kunnen bijdragen aan het vermogen om te bewegen, maar ook op ervaren moeilijkheden en vermoeidheid. Dit laatste mogelijk omdat dezelfde fysieke activiteit een hoger energieverbruik vereist. Er is literatuur beschikbaar die het risico van onvrijwillig verlies van vetvrije massa en mogelijke risicofactoren rapporteren. Een diepgaande beoordeling van de literatuur zou helpen om beter te begrijpen of verlies van vetvrije massa optreedt bij patiënten met AS en andere gerelateerde subgroepen van patiënten.

In dit proefschrift is getracht de sociale participatie bij patiënten met AS beter te leren begrijpen, is er onderzoek verricht naar objectief gemeten lichaamsbeweging van patiënten met AS en wordt er een overzicht gegeven van de literatuur over de vetvrije lichaamsmassa van patiënten.

Deel I Participatie binnen sociale rollen

Hoewel deelname aan sociale rollen essentieel is voor het leven van personen, heeft de participatie van personen met een chronische ziekte in het sociale leven nog relatief weinig aandacht gekregen. Omdat er nog weinig bekend was welke meetinstrumenten wetenschappelijk voldoende kwaliteit hebben om participatie te evalueren bij patiënten met AS, wat de impact is van AS op sociale participatie zelf, en of sociale participatie belangrijk is voor de ervaren levenstevredenheid werden diverse studies verricht (beschreven in hoofdstukken 2 t/m 4), die deze informatie aanvullen.

In **hoofdstuk 2** beschrijven we de vertaling en culturele aanpassing van de (Engelse) Social Role Participation Questionnaire (SRPQ) naar het Nederlands. Tevens presenteren we de kwaliteit van de vragenlijst binnen een onderzoek onder 246 Nederlandse patiënten met AS. De SRPQ omvat vragen omtrent participatie in elf rollen en één algemeen participatie item en gaat daarbij telkens vier dimensies na: het belang van de rol, tevredenheid met het vermogen om te participeren in de rol, tevredenheid met de besteedde tijd aan de rol en tenslotte de ervaren moeilijkheid bij het uitvoeren. Alle scores variëren van 1 (helemaal niet belangrijk, tevreden, niet mogelijk) tot 5 (heel erg belangrijk, tevreden, helemaal geen moeilijkheden). De vertaling werd uitgevoerd met behulp van de ‘dual panel’ methode en vereiste slechts kleine aanpassingen. Er was een goede samenhang te zien tussen de sociale rollen binnen de vier afzonderlijke dimensies. Tussen de verschillende dimensies, was er een sterke overeenkomst tussen de dimensies tevredenheid met ‘tijd’ en ‘vermogen’ ($r=0.85$). De betrouwbaarheid van herhaalde metingen werd getest bij 31 patiënten met stabiele ziekte en werd bevredigend bevonden. Overeenkomsten met participatiedomeinen uit andere participatie vragenlijsten of vragenlijsten omtrent ziekte activiteit, lichamelijk en mentaal functioneren waren zoals verwacht op zijn minst matig ($r=0.41$ tot 0.75) voor de dimensies ‘roltevredenheid’ en ‘moeilijkheden bij uitvoeren van rollen’, maar zwak voor ‘rolbelang’ ($r \leq 0.40$). Ook het onderscheidend vermogen van de SRPQ tussen patientengroepen is onderzocht door te kijken naar de verschillen in de mate

waarop gescoord werd binnen vijf categorieën van zelfgerapporteerde gezondheidstoestand (op basis van de Short-Form 36), deze was goed voor alle dimensies. Een belangrijke bevinding was dat de vraag naar ‘algemene participatie’ voor elk van de 4 dimensies, van een vergelijkbare kwaliteit was als het gemiddelde van alle 11 rollen. We concludeerden dat de Nederlandse versie van de SRPQ beschikbaar is om sociale participatie van patiënten met AS beter te leren begrijpen. De dimensie ‘rolbelang’ meet een ander aspect van participatie en moet gemeten blijven worden. De dimensie tevredenheid met de tijd lijkt overbodig en kan, zoals in een eerdere publicatie werd gesuggereerd, worden verwijderd uit de vragenlijst. Tenslotte, de algemene participatie vraag is een goede algemene maatstaf voor het meten van sociale participatie binnen elke dimensie. Dankzij dit onderzoek was er nu een goede vragenlijst beschikbaar die in **hoofdstuk 3** kon worden afgenomen bij 246 patiënten met AS en 510 mensen uit de algemene bevolking (controlegroep), met een vergelijkbare leeftijds- en geslachtsverdeling. Naast het invullen van de SRPQ rangschikten alle deelnemers hun drie belangrijkste rollen. De rangorde van rolbelang, de gemiddelde SRPQ-dimensiescores en de kloof tussen ‘belang’ en ‘tevredenheid met vermogen’ van rollen werden vergeleken tussen patiënten en de controlegroep. Zowel patiënten als de controles uit de algemene bevolking, rangschikten ‘intieme relaties’, ‘relaties met (groot / stief-) kinderen’ en ‘werk’ als de drie belangrijkste rollen. Vergeleken met de controlegroep, scoorden patiënten over de 11 rollen in de SRPQ gemiddeld een hoger ‘belang’ (3.75 versus 3.43 op een ‘beste’ score van 5), een lagere ‘tevredenheid met vermogen’ (3.19 versus 3.58) en meer ‘fysieke moeilijkheden’ (3.87 versus 4.67). De grootste kloof tussen ‘belang’ en ‘tevredenheid met het vermogen’ voor patiënten in vergelijking met controles werden gezien voor vrijetijdsactiviteiten die lichamelijke inspanning vergen, hobby’s, reizen en vakantie. Hierin gaven patiënten een hogere belangscore maar vooral ook een lagere tevredenheid aan. Naarmate in de samenleving steeds meer de nadruk wordt gelegd op een ieders individuele verantwoordelijkheid om volledig deel te nemen aan sociale rollen, suggereren de huidige gegevens dat zorgverleners meer aandacht moeten besteden aan vermogenskloof die patiënten met AS ervaren mede door het grote belang dat zij hechten aan hun sociale rollen.

Om inzicht te verkrijgen in de relatie tussen sociale participatie en de ervaren levens- tevredenheid van patienten, hebben we in **hoofdstuk 4** het verband onderzocht tussen verschillende aspecten van sociale participatie en levenstevredenheid bij 246 patiënten met AS in vergelijking met 510 deelnemers uit een controlegroep. Voor mensen met AS en de controlegroep werd de overeenkomst tussen SRPQ-rol domeinen en de tevredenheid met het leven in het Engels Satisfaction With Life (SWL)-schaal onderzocht voor elke participatiedimensie afzonderlijk, in de totale en de werkende populatie, waarbij rekening werd gehouden met relevante versturende factoren zoals leeftijd, geslacht, opleiding en inkomen. Patiënten waren vaker (uiterst) ontevreden over het leven in vergelijking met mensen uit de algemene bevolking die geen reumatische aandoening hadden (17.9% versus 8.6%). Minder fysieke moeilijkheden en een hogere tevredenheid betreffende interpersoonlijke

relaties en vrije tijdsbesteding waren geassocieerd met een hogere levenstevredenheid en dit was iets sterker bij patiënten dan bij controles. Bij werkende controles maar niet bij werkende patiënten, was ook tevredenheid met het vermogen om hun werk uit te voeren onafhankelijk geassocieerd met levenstevredenheid. De bevindingen van deze studie benadrukken het belang van sociale rollen en suggereren dat het de moeite waard zou kunnen zijn om patiënten te stimuleren actief te investeren in bevredigende relaties binnen de familie- en vriendenkring.

Deel II Lichaamsbeweging en lichaamssamenstelling

In het tweede deel van dit proefschrift hebben we lichaamsbeweging en lichaamssamenstelling onderzocht bij patiënten met AS, met de nadruk op mogelijke verschillen met mensen uit de algemene bevolking (controlegroep), met een vergelijkbare leeftijds- en geslachtsverdeling. In **hoofdstuk 5** werden 24 patiënten met AS en 24 controles vergeleken met betrekking tot drie benaderingen van functioneren: zelf-gerapporteerde ervaren *moeilijkheden* met het uitvoeren van lichamelijke activiteiten, gemeten met de Bath AS Functional Index (BASFI), zelf gerapporteerde *hoeveelheid* lichaamsbeweging (Baecke-vragenlijst) en de objectief gemeten hoeveelheid lichaamsbeweging (bewegingsmeter). Beide groepen omvatten 14 (58%) mannen en de gemiddelde leeftijd was 48 jaar. Patiënten scoorden significant slechter op de moeilijkheden met zelf-gerapporteerde lichamelijke activiteiten gemeten met de BASFI dan hun gezonde leeftijdsgenoten, terwijl de hoeveelheid lichaamsbeweging gemeten met de Baecke vragenlijst en de bewegingsmeter niet verschilden tussen groepen. De mate van overeenkomst tussen benaderingen van fysiek functioneren waren laag ($r = <0.4$) tussen de BASFI en Baecke vragenlijsten tot matig tussen de bewegingsmeter met de BASFI en de Baecke vragenlijst ($r = 0.41$ tot 0.75). De BASFI kwam overeen met ziekteactiviteit ($r = 0.49$) en ervaren fysieke vermoeidheid (0.73). De Baecke kwam niet overeen met ziekte activiteit, maar wel met fysieke en activiteit gerelateerde vermoeidheid ($r = 0.54$ en $r = 0.54$). De totale hoeveelheid lichaamsbeweging gemeten met de bewegingsmeter kwam niet overeen met een van deze op ervaring gebaseerde gezondheidsmaten. We concludeerden dat verschillende benaderingen van het concept fysiek functioneren bij patiënten met AS verschillende informatie verschaffen. Vergeleken met de controles rapporteerden patiënten met AS meer moeilijkheden bij het uitvoeren van lichamelijke activiteiten en ook meer vermoeidheid, maar bewogen wel evenveel.

In **hoofdstuk 6** werden de totale hoeveelheid lichaamsbeweging en tijd besteed binnen verschillende intensiteiten van beweging, tussen 135 patiënten met AS en een controle groep met 99 deelnemers vergeleken en factoren gerelateerd met lichaamsbeweging onderzocht. Aan de deelnemers werd gevraagd om gedurende zeven dagen een bewegingsmeter te dragen en een reeks vragenlijsten in te vullen. Patiënten en de controles verschilden niet wat betreft de totale hoeveelheid lichaamsbeweging of de tijd die doorgebracht werd in sedentaire of lichte intensiteiten van beweging. Echter brachten patiënten minder tijd door in matige tot intensieve intensiteiten van beweging. Bij zowel

patienten als controles kwamen een slechtere zelf-gerapporteerde fysieke gezondheid (gemeten met een sub schaal van de Short Form-36) en een hogere BMI score het meeste overeen met een lagere totale hoeveelheid lichaamsbeweging. Verder onderzoek naar de kenmerken van patienten met AS in relatie tot tijd die doorgebracht werd in matige tot intensieve intensiteiten, toonde dat een hogere BMI, meer moeilijkheden met fysiek functioneren (BASFI) maar ook een langere ziekteduur invloed hadden op deze zwaardere beweegintensiteiten. Boeiend was dat patiënten van 52 jaar en ouder met een hogere ziekteactiviteit meer tijd doorbrachten in lichte activiteiten en minder in zittende activiteiten in vergelijking met patiënten van dezelfde leeftijd met een lagere ziekteactiviteit. Samenvattend zagen we dat patiënten met AS niet relevant verschillen met mensen uit de algemene bevolking op basis van de totale hoeveelheid (objectief gemeten) lichaamsbeweging. Echter bewogen zij wel minder vaak matig tot intensief waardoor de beweegnorm minder vaak werd behaald vergeleken met mensen uit de algemene bevolking. Het kan daarom relevant zijn om de causale relatie tussen bewegen, gezondheid en ziekteactiviteit verder te onderzoeken.

In **hoofdstuk 7** wordt een systematisch overzicht gegeven van de literatuur betreffende (a) lichaamssamenstelling van patiënten met reumatoïde artritis (RA) of spondyloarthritis (SpA) in vergelijking met controle populaties, (b) factoren die een verband hebben met (een verandering in) vetvrije massa, en (c) effecten van medicamenteuze en niet- medicamenteuze interventies op lichaamssamenstelling bij beide aandoeningen. MEDLINE/ PubMed werd doorzocht en twee onafhankelijke beoordelaars hebben sterke en zwakke punten van geselecteerde geëvalueerd. Resultaten werden gepresenteerd als narratieve samenvattingen per ziekte (RA of SpA) en doel van de studie (beschrijvend, verklarend of experimenteel). In RA rapporteerden 17 van de 23 onderzoeken een lagere vetvrije massa in vergelijking met controles, maar in SpA werd dit gevonden in slechts één van de vijf onderzoeken. De observationele onderzoeken bij RA onthulden dat leeftijd, het vrouwelijk geslacht, ziekteactiviteit (op diverse manieren gemeten), ziekteduur en moeilijkheden bij het uitvoeren van dagelijkse activiteiten allemaal onafhankelijk geassocieerd waren met een lagere vetvrije massa. De uitgevoerde experimentele onderzoeken suggereerden dat krachttraining resulteerde in een toename van vetvrije massa. In geen van de beperkte hoeveelheid onderzoeken werd gemeld dat een medicamenteuze behandeling of dieetinterventies een effect had op vetvrije massa. Zeer beperkt onderzoek bij patiënten met SpA lieten een effect zien van anti-TNF alfa therapie op vetvrije massa. Samenvattend toonde deze systematische literatuurstudie dat de vetvrije massa van patiënten met RA lager is dan die van controles, maar nog steeds omkeerbaar is met krachttraining. Echter, bewijs bij patiënten met SpA was schaars en wees niet op verschillen in vetvrije massa. Meer onderzoek naar de lichaamssamenstelling en gerelateerde factoren bij patiënten met SpA wordt aanbevolen.

Algemene conclusie

De studies die uitgevoerd zijn in het kader van dit proefschrift geven meer inzicht in de sociale participatie van patiënten met AS. Er werd aangetoond dat patiënten met AS minder levenstevredenheid ervaren, maar ook dat sociale participatie is gerelateerd aan een hogere levenstevredenheid. We willen een discussie openen of en hoe we in de zorg meer aandacht moeten besteden aan sociale rol participatie. Nieuwe inzichten werden ook verkregen over objectief gemeten patronen van lichaamsbeweging en laten zien dat patiënten in totaal niet minder bewegen dan mensen uit de algemeen bevolking maar wel minder vaak matig tot intensief waardoor de beweegnorm minder vaak werd behaald. Vervolgonderzoek moet uitwijzen welke intensiteiten van beweging in het dagelijks leven gerelateerd zijn met positieve of eventueel negatieve uitkomsten van ziekte gerelateerde maten zoals progressie in radiologisch aantoonbare bot- en gewrichtsafwijkingen.

Valorisation addendum

Background

Participation and physical activity (PA) receive much attention in our society. Over the past years, the Dutch society has shifted or at least attempted to shift from a care taking-policy (zorgstaat) into a society in which participation of citizens has a more central place. Arguments in favor of this change were that the government policy insufficiently enabled individuals to take responsibilities in overcoming their health-related limitations in important aspects of life, such as work. Also, the government was ambitious in striving for a more cohesive community in which initiatives of people helping, aiding or assisting predominantly people with a disease or limitation were stimulated. However, little research had been done on attitudes and challenges of persons with chronic diseases in relation to participation in social roles (1, 2).

A prerequisite to participate in social roles is the ability to perform PA. That is, almost all participation areas such as work, social life, sports, traveling, care taking activities, and hobbies require movement of the body. Moreover, low PA has a negative impact on health. Worldwide, the consequences of insufficient PA may have caused major health-, economic- and societal problems, and this may be particularly true for patients with chronic diseases (3).

This thesis contributes to the evidence base of participation and PA in patients with ankylosing spondylitis (AS). The chronic auto-inflammatory rheumatic disease AS, has an onset in the third decade of life, characterized by pain, stiffness and mobility impairments in the spine and often also in the large joints. The early age at onset implies that patients with AS usually perform a relatively active role in society. Results of our research into social role participation, physical activity (PA) and body composition are described in chapters 2–7(4-7). The findings, research approaches and areas for further research are discussed in chapter 9. Here we describe the valorisation of the research presented in this thesis, which is defined as “the process of value creation from knowledge, by making it applicable and available for economic or societal utilisation, and by translating it in the form of new business, products, services, or processes”.

Value of the findings related to social role participation and physical activity

In the Social Participation in Ankylosing Spondylitis Study (SPASS) study, patient research partners of outcome studies of the department of Rheumatology of the MUMC, were involved. Especially in the organizational stage of the study, the partners had an important role in carefully selecting which participation questionnaire, among available (English) instruments, would best reflect participation needs of Dutch patients with AS. The Social Role Participation Questionnaire (SRPQ) was preferred, because it captures most comprehen-

sively essential aspects of participation in patients with a chronic disease like ankylosing spondylitis (AS). Specifically, it addresses the full diversity of roles relevant to patients and focuses on different aspects/dimensions such as importance, difficulties and satisfaction. The collaboration with patients is worthwhile and contributed to the face validity of the final findings. The translated and validated Dutch version of the full-length SRPQ as well as a short version, which has been developed following the work in this thesis, are now freely accessible for researchers and provide a well-performing instrument to measure participation in AS, and likely also for patients with other chronic conditions. Momentarily, the short version of the SRPQ is being applied in a larger Dutch longitudinal study.

For daily practice there is an ongoing discussion among health professionals whether and which participation roles should be measured. Until now, there is only consensus to ask patients about their work participation, which, in contrast to other roles, has a more apparent relation with health impairments and has clear economic relevance for the individual and society when accounting for the increased risk for sick-leave and work-disability of persons with AS. On that line, several interventions have been developed to support 'persons at risk for adverse work outcome'. The amount of evidence on the importance and prevention of adverse work outcome in persons with rheumatic diseases resulted in 2015 in the guideline 'Worker Participation and Rheumatoid Arthritis', by the Dutch Society of Rheumatology (Nederlandse Vereniging voor Reumatologie; NVR (8). During the period of this thesis, in 2013, data from the SPASS study, provided a broader view on social participation and were presented during the annual scientific meeting of the NVR. Also a special discussion session was organized around social role participation, opening the conversation whether other roles in addition to work participation should receive attention of the clinical team, for example in an annual review of the patient. It was felt that (indirect) benefits of social role participation on life satisfaction could be brought under the attention of patients as part of self-management programs.

Regarding PA, results of the studies reported in this thesis showed that patients with AS perform overall similar total amounts of PA, but only 47% of the patients as compared to 60% of the directly compared controls met the recommended minimum amount of 150 min/week moderate to vigorous PA (MVPA). Objectively assessed evidence of habitual PA and patterns of patients with AS have been indicated in this thesis. The relevance of patients performing less MVPA might also be related to possible disease related symptoms like involuntarily accelerated muscle loss, which is more common in several chronic diseases like rheumatoid arthritis (RA). That is, strength training, which is usually performed in intensities which are at least moderate PA, seem to have a protective effect against possible disease related muscle loss. This is in line with the recently proposed renewed Dutch guidelines for PA by the Dutch health council, who stressed the importance of performing musculoskeletal strength exercises on a weekly basis (9). As there is substantial evidence that (moderate) exercise helps to maintain known muscle loss in RA, increasing time spent

in moderate PA might improve muscle mass and protect the aging patients against sarcopenia and frailty.

Further, it was striking to find that patients experience considerably more difficulty and fatigue in performing daily tasks and activities when compared to controls, despite a similar amount of total PA at lower intensity. These findings have led to more research, studying the basic relationship of fatigue, PA and basal metabolic energy expenditure and cardiovascular fitness. This research aims to contribute to evidence based insight and interventions related to fatigue of patients with AS.

With this thesis, we addressed important aspects of the lives of patients with the chronic disease AS. Hopefully this work contributes to appreciate the importance of PA as well as other social roles alongside work participation in patient-related care and research.

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Curriculum Vitae

Simon van Genderen werd op 22 mei 1985 geboren in Nijmegen. Na het behalen van zijn diploma (HAVO) economie en maatschappij aan de Stedelijke Scholen Gemeenschap Nijmegen, volgde hij de gecombineerde MBO/HBO opleiding CIOS-Sport Gezondheid en Management aan het Rijn- IJssel College en de Hogeschool van Arnhem en Nijmegen. Nadat hij deze beide in 2008 succesvol had afgerond begon hij in 2009 aan de Master Sports and Physical Activity Interventions aan de Universiteit van Maastricht, waarvan hij in 2010 het diploma behaalde.

In 2011 begon hij aan zijn promotietraject op de afdeling reumatologie, CAPHRI, the School for Public Health and Primary Care en de afdeling humane biologie, van het Maastricht Universitair Medisch Centrum onder leiding van Prof. dr. Annelies Boonen, Prof. dr. Robert Landewé en dr. Guy Plasqui. Tijdens zijn promotieonderzoek heeft hij diverse cursussen gevolgd om zijn wetenschappelijke competenties verder te ontwikkelen. Zijn onderzoeksresultaten werden op nationale en internationale congressen gepresenteerd en zijn wetenschappelijke inspanningen resulteerden in dit proefschrift. Na in 2015 een jaar te hebben gewerkt als profielloos Sport Beweging en Gezondheid en als sportmanager aan het Graafschap College en bij Sportservice Doetinchem, werkt hij sinds 2016 als docent en onderzoeker aan de Hogeschool van Arnhem en Nijmegen, Instituut Sport- en Bewegingsstudies binnen het expertiseteam Sports Economics & Strategic Sports Management (SESS).

List of publications

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