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The Essence of Health and Disability in Persons with Chronic Widespread Pain

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Summary

Functioning and disability represent the core experience in patients suffering from Chronic Widespread Pain (CWP). This experience can now be systematically described with the International Classification of Functioning, Disability and Health (ICF). The ICF is a classification system including more than 1400 so-called categories which describe aspects of functioning and health based upon a comprehensive bio-psycho-social understanding of illness. However, due to its comprehensiveness, this classification is difficult to use in clinical practice. Therefore, ICF Core Sets have been developed for several different health conditions, including CWP. There are two Core Sets for CWP: the Comprehensive ICF Core Set consisting of a short list of categories relevant to most patients with CWP and the Brief ICF Core Set - a shorter more compact version of the Comprehensive ICF Core Set.

This doctoral project consists of two studies exploring the relevance of the aspects of functioning, disability and health as they are described in the ICF Core Sets for CWP. The first study uses qualitative methodology to investigate to what extent the Comprehensive ICF Core Set for CWP represents aspects of functioning important to patients with Fibromyalgia (FMS). 54 out of the 67 categories of the current version of the Comprehensive ICF Core Set for CWP could be confirmed by focus groups and 48 additional categories, not covered in the current version, were identified. The second study explores to what extent the Brief ICF Core Set for CWP reflects the most important core elements of functioning when looking at its content from a statistical perspective. Regression analyses using data from a cross-sectional multicenter international study were performed. 11 categories relevant for describing self-perceived general health in patients with CWP were identified in the regression models. These categories were identical or similar to the set of categories included in the Brief ICF Core Set for CWP. This doctoral thesis adds evidence to the question of what the essence of functioning and disability in CWP is, based on the patient perspective and the application of a statistical approach. The results are a valuable contribution to pinpointing the most important aspects of functioning in CWP. However, the ICF Core Sets still need further validation in order to come to a consensus regarding the core elements.

Zusammenfassung

Das Erleben von Funktionsfähigkeit und Behinderung prägt die Erfahrungen von Patienten mit chronischem Ganzkörperschmerz (CWP). Diese Erfahrungen können jetzt systematisch durch die Internationale Klassifikation für Funktionsfähigkeit, Behinderung und Gesundheit (ICF) beschrieben werden. Die ICF ist ein Klassifikationssystem, das mehr als 1400 Kategorien beinhaltet, die Aspekte von Funktionsfähigkeit und Behinderung basierend auf einem bio-psycho-sozialen Verständnis von Erkrankungen beschreiben. Allerdings ist diese Klassifikation sehr umfassend und daher im klinischen Alltag schwierig anzuwenden. Aus diesem Grund sind Kerndatensätze (*Core Sets*) für einige Erkrankungen, einschließlich CWP, entwickelt worden. Für CWP existieren 2 *ICF Core Sets*. Das *Comprehensive ICF Core Set for CWP* besteht aus einer kurzen Liste von Kategorien, die für die meisten Patienten mit CWP relevant sind. Das *Brief ICF Core Set* ist eine kompakte, verkürzte Version des *Comprehensive ICF Core Set for CWP*.

Diese kumulative Dissertation besteht aus zwei Studien, die relevante Aspekte von Funktionsfähigkeit, Behinderung und Gesundheit bei CWP auf der Basis der ICF erforschen. Bei der ersten Studie handelt es sich um eine qualitative Studie, die untersucht, inwieweit die aktuelle Version des *Comprehensive ICF Core Set for CWP* diejenigen Aspekte von Funktionsfähigkeit enthält, die für Patienten mit Fibromyalgie (FMS) wichtig sind. Von 67 ICF-Kategorien in der aktuellen Version des *Comprehensive ICF Core Set for CWP* konnten 54 davon in Fokusgruppen bestätigt werden. Zusätzlich wurden 48 ICF-Kategorien identifiziert, die nicht in der aktuellen Version enthalten sind. Die zweite Studie erforscht anhand einer statistischen Analyse, inwieweit das *Brief ICF Core Set for CWP* die Kernelemente von Funktionsfähigkeit bei FMS reflektiert. Hierfür wurden die Daten einer multizentrischen, internationalen Querschnittstudie regressionsanalytisch ausgewertet. In den berechneten Regressionsmodellen wurden 11 ICF-Kategorien identifiziert, die für die Beschreibung der subjektiven allgemeinen Gesundheit bei Patienten mit CWP relevant sind. Diese ICF-Kategorien sind identisch oder ähnlich mit den Kategorien des *Brief ICF Core Set for CWP*. Diese Dissertation integriert die Patientenperspektive und eine statistische Analyse um wichtige Aspekte von Funktionsfähigkeit, Behinderung und Gesundheit in CWP zu erforschen. Die Ergebnisse sind ein wertvoller Beitrag, die wichtigsten Kernelemente bezüglich der

Funktionsfähigkeit bei CWP zu identifizieren. Allerdings sollten die ICF Core Sets in weiteren Studien validiert werden, um einen Konsens bezüglich der Kernelemente der Funktionsfähigkeit bei CWP zu erzielen.

Content

I Introduction

II Research articles

1. Background

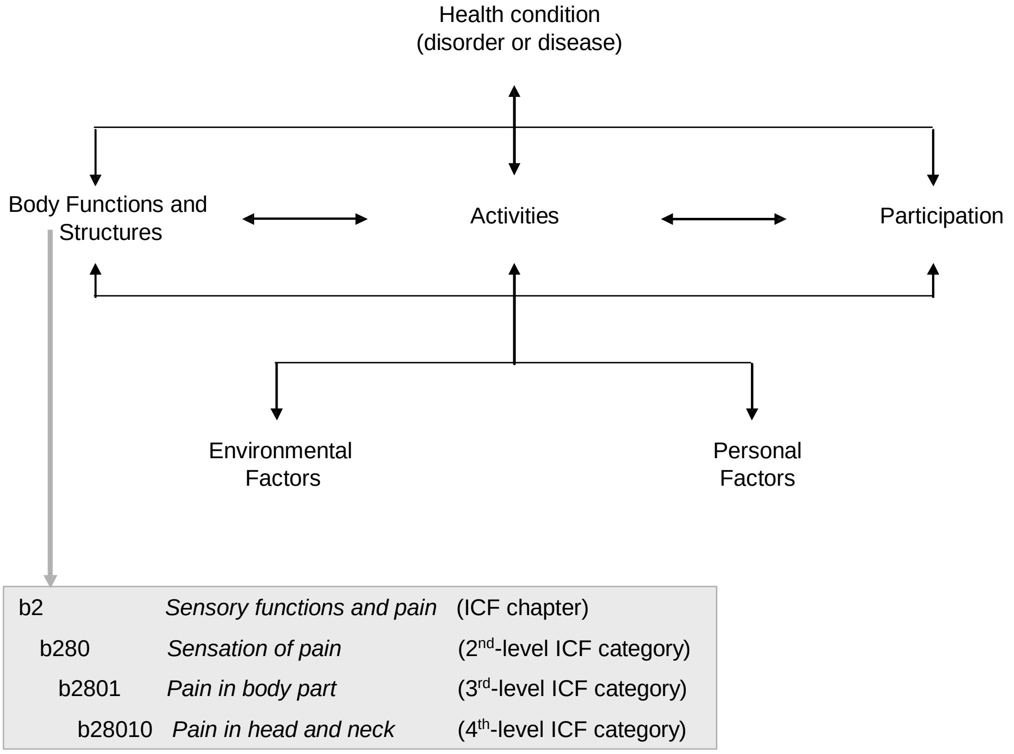
Chronic Widespread Pain (CWP) is a common musculoskeletal disorder affecting an estimated 10 to 12 percent of the population [1, 2]. CWP is associated with significant disability and functional impairment disrupting many aspects of daily life such as managing everyday routines and activities [3, 4]. Functioning and disability represent the core experience in patients suffering from CWP. One major challenge is to capture the essence of functioning and disability and to implement this knowledge in clinical practice and research.

A number of patient reported outcomes addressing areas of functioning and disability relevant in CWP have been developed. These instruments were developed according to the medical perspective based upon the assumption that salient patient problems in functioning are caused by the illness. The influence of the environment and personal factors is still rarely taken into account in these instruments [5, 6], even though several studies show that patients' experience of functioning is determined not only by the health condition, but also by personal factors and the interaction with the environment [7, 8, 9, 10].

The essential role of functioning has been acknowledged by initiatives such as the OMERACT (Outcome Measures in Rheumatoid Arthritis Clinical Trials), with the goal of defining what should be measured and how, across the spectrum of rheumatology intervention and observational studies [11]. Fibromyalgia Syndrome (FMS), an extreme form of CWP, is one of the several conditions addressed in OMERACT conferences. The core domains agreed upon in OMERACT are pain, fatigue, sleep disturbance, multidimensional function, health-related quality of life, mood disorders and, cognitive dyscognition. [12]. Although functioning is one of the core domains, OMERACT has not yet determined how to define functioning in CWP and FMS. The question as to what is really relevant in functioning is still open.

A complementary initiative addressing functioning and disability is the development of International Classification of Functioning, Disability and Health (ICF) Core Sets for chronic conditions. The ICF was endorsed by the WHA as a member of the World Health Organisation (WHO) Family of International Classifications. This initiative is important because the ICF is the current WHO framework for describing and assessing functioning and disability. It integrates the bio-psycho-social perspective of functioning and disability in illness and establishes the basis for a more comprehensive description of the experience of patients suffering from a determined disease. Based on this perspective, functioning with its components *Body Functions, Body Structures, Activities and Participation* and *Environmental and Personal Factors* is seen in relation to the health condition.

Figure 1 The bio-psycho-social perspective and the International Classification of Functioning, Disability and Health (ICF).



The ICF contains 1424 ICF categories, each allotted to the named components of the classification with the exception of the component *Personal Factors*, which has not yet been classified. Each ICF category is denoted by a code composed of a letter that refers to the components of the classification (b: *Body Functions*; s: *Body Structures*; d: *Activities and Participation* and e: *Environmental Factors*) and is followed by a numeric code starting with the chapter number (one digit), followed by the 2nd level (two digits) and the 3rd and 4th levels (one digit each) (see fig. 1).

Based on the ICF classification, ICF Core Sets have been developed for several chronic diseases in a worldwide, comprehensive consensus process over the last few years. ICF Core Sets are a selection of categories out of the 1424 ICF categories, which set out to reflect important aspects of functioning, characteristic of a certain illness under consideration of influential *Environmental Factors*. This development allows the description of functioning and disability of CWP using the ICF as a standard.

For CWP, two ICF Core Sets have already been developed: the Comprehensive ICF Core Set and the Brief ICF Core Set. The Comprehensive ICF Core Set for CWP is a set of ICF categories which makes possible a comprehensive description of functioning of persons with CWP in multidisciplinary assessments. It includes as few categories as possible to be practical, but as many as necessary to describe the aspects of functioning relevant to persons with CWP. In total, the Comprehensive ICF Core Set for CWP includes 65 categories at the 2nd level and 2 categories at the 3rd level of the classification (23 *Body Functions,* 1 *Body Structure*, 27 *Activities and Participation*, 16 *Environmental Factors)* [15].

The **Comprehensive ICF Core Set for CWP** is now undergoing worldwide testing and validation using a number of approaches including an international multicentre validation study and validation from the perspective of health professionals. Another key aspect is the validation from the patient perspective – those who actually experience the disease. While the patient perspective has been implicitly included in the development of ICF Core Sets [13], the patients are now explicitly involved in the process of the validation of this ICF Core Set. One objective of this doctoral thesis is to explore the patient perspective of persons with FMS and to see whether these aspects are covered in the Comprehensive ICF Core Set for CWP. I chose qualitative methodology involving focus groups because it provides the possibility of exploring the perspective of those who experience a health problem and is especially useful for studies involving complex issues that entail many levels of feeling and experience. The idea behind focus group methodology is that group processes can help people explore and clarify their views while the non-directive nature of focus groups allow for participants to comment, explain, disagree and share experiences and attitudes [14, 15]. FMS is in itself an extremely complex illness, therefore focus groups seemed the most appropriate for capturing all aspects of everyday life important to patients with FMS.

The **Brief ICF Core Set for CWP** is a shorter, more compact version of the respective Comprehensive ICF Core Set for CWP and address only those aspects of functioning that are essential. It includes the fewest number of ICF categories that can serve as a minimal standard for the reporting of functioning and health of persons with CWP for clinical studies and clinical encounters. The Brief ICF Core Set can be used internationally for the reporting of functioning in persons with CWP in any care setting and across sectors such as health, social welfare, education and labour and research [16]. The current version of the Brief ICF Core Set for CWP includes a total of 26 categories (10 *Body Functions,* 10 *Activities and Participation*, six *Environmental Factors)*. This ICF Core Set also needs validation using statistical methods in order to complement expert opinion based upon which it was developed. Therefore, the second objective of this doctoral thesis is to identify whether the Brief ICF Core Set for CWP reflects the most important aspects. I used statistical methodology, specifically regression modelling, to identify which ICF categories explain the most variance of the experience of health in CWP and compared the identified categories to the categories of the Brief ICF Core Set for CWP which was decided based upon expert opinion. Instead of regression modelling, other methods, such as simply ranking the strength of the association after using parametric or non-parametric correlation statistics (Spearman or Pearson Coefficients) could be considered when selecting ICF categories in relation to a broad concept, like health. However, regression modelling enables a better understanding of the relative contribution of individual ICF categories in relation to others [17].

2. Objective of the doctoral thesis

The overall objective of this doctoral thesis is to explore the essence of functioning and disability in persons with CWP from different perspectives. In detail, this thesis explores the following questions: 1) to what extent does the Comprehensive ICF Core Set for CWP represent aspects of functioning important to patients with FMS and 2) to what extent does the Brief ICF Core Set for CWP reflect the most important core aspects of functioning when looking at its content from a purely statistical perspective? Two studies using different methodologies were conducted to explore these questions. Study 1 consists of a qualitative study in which the patient perspective was investigated. Study 2 follows a statistical approach using data from a cross-sectional multicenter international study.

3. Summary of study 1

In the first study I considered the question of to what extent the Comprehensive ICF Core Set for CWP represents aspects of functioning important to patients with FMS. The specific aims were (1) to explore the aspects of functioning and health important to patients with FM using focus group methodology and (2) to examine to what extent these aspects are represented by the current version of the *Comprehensive ICF Core Set for CWP*. Qualitative methodology, which provides the possibility of exploring the perspective of those who experience a health problem, i.e. the patient perspective [18, 19] was used. One of the most broadly used techniques in qualitative research is the focus group methodology [20, 21, 22]. Focus groups are “carefully planned series of discussions designed to obtain perceptions in a defined area of interest, in a permissive, non-threatening environment” (p. 5) [23]. They are especially useful for studies involving complex issues that entail many levels of feeling and experience [24].

A total of 33 participants were included in six focus groups. The participants all had FMS diagnosed according to the American College of Rheumatology [25]. In addition, the patients had to be at least 18 years of age and have sufficient linguistic skills of the German language. Patients who had psychiatric disorders or have had surgery briefly before participation in the study were excluded. The focus groups were performed according to focus group guidelines in which the titles of the chapters of the Comprehensive ICF Core Set for CWP and open-ended questions were presented visually to the participants with a Power-Point presentation. At the end of each focus group, a summary of the main results was given back to the group to enable the participants to verify and amend emergent issues. The focus groups were digitally recorded and transcribed verbatim.

For the qualitative analysis of the data, the meaning condensation procedure [26] was used. In the first step, the transcripts of the focus groups were read through to get an overview of the collected data. In the second step, the data were divided into units of meaning, and the theme that dominated a meaning unit was determined. In the third step, the concepts contained in the meaning units were identified. The identified concepts were then linked to ICF categories based on established linking rules [27, 28], which enable the linking of concepts to ICF categories in a systematic and standardized way. An ICF category of the Comprehensive ICF Core Set for CWP was regarded as confirmed if the respective category emerged from the focus groups.

A total of 1686 relevant concepts were identified in the focus groups. These concepts were linked to 247 different ICF categories of the 1st to the 4th level. There were 277 concepts which could not be linked to ICF categories (e.g. quality of life in general, aspects of coping, disease management, time-related aspects, and variability of functioning). Of these, 143 of them could be allotted to the component *Personal Factors*, which has not yet been classified. 90 topics were not covered in the ICF classification and were defined as “not covered” (nc) and 44 concepts were labelled “not definable” (nd).

Relevant outcomes of treatment in CWP from the patient perspective such as pain, fatigue, sleep disorders, psychological distress, lack of muscle power, difficulties changing and maintaining a body position and difficulties carrying out a daily routine were confirmed [29, 30]. Pain proved to be the central topic reported by patients, with a total of 159 meaning units linked to the ICF Core Set category *b280 sensation of pain*. Interestingly, apart from pain, the most outstanding theme reported by participants in all six focus groups was the attitude of others regarding FMS. The patients described often feeling left alone with their illness, due to a lack of understanding and acceptance from others. Several patients reported feeling as if FM is not accepted as a legitimate illness by some doctors and health care professionals and is often trivialised by friends, relatives and colleagues, thus adding to the burden of pain and exhaustion. Fifty concepts concerning negative attitudes of others regarding the illness were linked to the corresponding ICF Core Set categories and forty-six additional concepts were linked to the 1st level category *attitudes.* Several studies report similar findings such as patients’ experiences of stigma [31, 32, 33, 34] and studies documenting controversy as to the existence, classification and acceptance of FMS by health care professionals [35, 36, 37, 38].

In total, 54 out of the 67 categories included in the Comprehensive ICF Core Set for CWP were confirmed by the participants of the focus groups. Forty-eight additional 2nd level categories which are not included in the current version of the Comprehensive ICF Core Set for CWP were identified in the focus groups. Most of the additional categories stem from the component *Body Functions* (n=23) followed by *Activities and Participation* (n=15). Ten additional categories were reported by the participants related to the component *Environmental Factors*. No additional categories from the component *Body Structures* were identified.

Thirteen categories in the ICF Core Set for CWP were not at all mentioned by the focus groups. Most of the categories belonged to the component *Body Functions* and included *b122 global psychosocial functions, b147 psychomotor function*, *b1602 content of thought*, *b260 proprioceptive function*, *b430 haematological system functions* and *b640 sexual functions*. Some concepts were linked to similar categories, e.g. 18 meaning units were linked to the category *d230 carrying out daily routine* instead of *d220 undertaking multiple tasks*.

Forty-eight additional 2nd level categories that are not covered in the current version of the *Comprehensive ICF Core Set for CWP* were raised. Several of these categories have to do with difficulties in cognitive functioning such as problems with short and long term memory, learning and applying knowledge and difficulties thinking, hearing, listening and reading. Further topics not yet covered are the use of the hands and arms, functions of the digestive system and several issues belonging to the not yet classified component *Personal Factors*, such as feeling ashamed, feeling stressed and exhausted, perfectionism and fear of losing one’s job due to the illness.

To summarize, the current version of the Comprehensive ICF Core Set for CWP does represent areas of functioning important to patients with FMS since most categories of the existing version could be confirmed by the focus groups (54 out of 67). However, additional categories not represented in the *Comprehensive ICF Core Set for CWP* emerged from the focus groups and should be considered for inclusion in the final version of the Comprehensive ICF Core Set for CWP. Several concepts important to patients with FMS could be allotted to the component *Personal Factors*, which has not yet been classified. This reflects the importance of including this component in further development. This ICF Core Set aims to be used as the basis of multidisciplinary assessments, for example in rehabilitation. Patient centeredness is an important and valuable concept in rehabilitation and in the medical field in general, taking into account patients’ wants, needs and preferences [39, 40]. The results of this study contribute to a patient centered approach in further development of the ICF.

The qualitative study described above was published as follows:

Hieblinger R, Coenen M, Stucki G, Winkelmann A, Cieza A. *Validation of the International Classification of Functioning, Disability and Health Core Set for chronic widespread pain from the perspective of fibromyalgia patients.* Arthritis Res Ther 2009, 11(3):pR67

4. Summary of study 2

In this study the question was to what extent the Brief ICF Core Set reflects the most important core aspects of functioning when performing analyses from a purely statistical perspective. The study was performed in cooperation with the WHO and included a convenience sample of 452 patients with CWP from 22 study centers in 10 different countries. The specific aims were (1) to identify which ICF categories explain the most variance of the experience of health in CWP and (2) to compare the identified ICF categories to the ICF categories of the Brief ICF Core Set for CWP which was decided based upon expert opinion.

In addition to collecting sociodemographic data and disease-specific characteristics, the data was collected using (1) the Comprehensive ICF Core Set for CWP with its total of 67 ICF categories and (2) the Medical Outcome Study Short Form 36 (SF-36) with its eight multi-item scales and two summary scales (Physical Component Summary Score (PCS), Mental Component Summary Score (MCS)). Multiple regression models to explain general health were calculated. The first question of the SF-36, which addresses health in general, was selected as the dependent variable for data analysis. Independent variables were the ICF categories of the Comprehensive ICF Core Set for CWP. Since there were a large number of potential independent variables, bivariate analysis were performed to reduce the number of those variables which could enter the multivariate model. Correlation of the ICF categories of the Comprehensive ICF Core Set for CWP and general health were therefore performed using the Pearson product-moment correlation coefficient (r).

The results of the bivariate analysis showed that a total of 27 categories from the 67 categories of the Comprehensive ICF Core Set for CWP correlated with general health as represented by item one of the SF-36*.* The highest correlation was found in *d910 community life (*r=.45; p=.000) in the component *Activities and Participation.* In the component *Body Functions, b280 sensation of pain* correlated highest (r=.43; p=.000).

The ICF categories to be entered in an initial regression model using backwards regression analysis were selected according to their correlation with item one of the SF-36 and in accordance with the ICF structure. Age and the ICF categories *b152 emotional functions, b280 sensation of pain, b710 mobility of joint functions, d845 acquiring, keeping and terminating a job, d910 community life* and *e460 societal attitudes* remained in the final model and explained 33 percent of the variance of general health.

A further set of regression models was performed because in some ICF chapters, categories within the same chapter highly inter-correlated with one another which can lead to unclarity regarding the true association between the respective variables and general health. The categories which correlated with other categories from the same chapter were therefore systematically substituted in additional models. As a result, *b1602 content of thought*, *b126 temperament and personality functions, b147 psychomotor functions, d850 remunerative employment and d920 recreation and leisure* remained in their respective models. These categories explained between 33 and 35 percent of the total variance of general health. Therefore, not only the categories identified in the initial regression model, but also these five categories are considered relevant to the experience of health in CWP.

Altogether, a total of 11 categories relevant for describing self-perceived general health in patients with CWP were identified in the regression models. I then compared these categories with the set of categories included in the Brief ICF Core Set for CWP. Six ICF categories remaining in the regression models were included in the Brief ICF Core Set for CWP: *b152 emotional functions, b280 sensation of pain, b147 psychomotor functions, b1602 content of thought, d850 remunerative employment* and *d920 recreation and leisure.* ICF categories selected in the multivariate analysis but not included in the Brief ICF Core Set were: *b126 temperament and personality functions, b710 mobility of joint functions, d845* *acquiring, keeping and terminating a job, d910 community life* and *e460 societal attitudes*. However, all of these categories resemble categories within the same chapter which are included in the Brief ICF Core Set. For example, *temperament and personality functions,* confirmed in the regression models, is from the same chapter (*mental functions*) as six other categories included in the Brief ICF Core Set. These 11 categories best differentiate among different levels of self-perceived health and should therefore be considered for use in studies in addition to the categories of the Brief ICF Core Set for CWP.

The study described above was submitted and accepted for publication as follows:

Hieblinger R, Coenen M, Stucki G, Winkelmann A, Cieza A. *Identification of essential elements of functioning in chronic widespread pain based on a statistical approach.* Am J Phys Med Rehabil 2011, 90(12):979-91.

5. Discussion and Outlook

This doctoral thesis adds evidence to the question of what the essence of functioning and disability in CWP is. CWP is an extremely complex condition with a large range of physical and mental symptoms affecting numerous areas of life. Patient centeredness is an important concept in health care and the patient perspective is a key element in gaining a fuller understanding of functioning, disability and health in this complex illness. Since the Comprehensive ICF Core Set for CWP is the basis for multidisciplinary care in CWP it is essential that all aspects relevant from the patient perspective are taken into account. Regarding the qualitative study, light was shed upon whether the ICF Core Set for CWP is an appropriate tool to be used to address all relevant problems of the patients in a multidisciplinary comprehensive assessment such as rehabilitation. Since most of the categories were confirmed I can say with confidence that the ICF Core Set for CWP is an adequate reference for a patient centered approach. However, *Personal Factors* are also a key area reflected in the patient perspective. Therefore, development and inclusion of this important component in the ICF is essential. I also suggest conducting further studies involving the patient perspective of all CWP patients – not only those with FMS. It may even be considered developing an additional ICF Core Set specifically for FMS because this illness poses special challenges such as stigmatization and negative attitudes of others towards the illness.

Regarding the Brief ICF Core Set for CWP, the results of this doctoral thesis are of special interest because they may contribute to helping clinicians and health professionals obtain a quick efficient overview regarding the level of functioning of their patients in those essential areas that best differentiate among various levels of functioning. The Brief ICF Core Sets can be used as minimal standards for the reporting of functioning and environmental factors for research and clinical encounters. With this study a set of 11 ICF categories essential for describing functioning and disability in patients with CWP were identified. These ICF categories could represent a first proposal for a reduction of the Brief ICF Core Sets for CWP or it may be considered substituting certain ICF categories currently included in the Brief ICF Core Set for similar categories, which may reflect functioning and disability in CWP more accurately. According to the FM OMERACT 8 workshop, the key domains in CWP to be investigated in studies and endorsed by both clinician-investigators and patients are *pain, fatigue, sleep disturbance, multidimensional function, health-related quality of life, mood disorders* and*, cognitive dyscognition* [41]*.* Comparing my results with these domains, all 11 categories found as a result of the regression models are represented in these key OMERACT domains. The Brief ICF Core Set for CWP is not only being used in studies but also as part of the revision process of the ICD-11. A newly developed axis called functioning properties serves as a link to allow for joint usage of the ICD and the ICF. These functioning properties are proposed to be included in the ICD revision process [42]. Therefore these results may be a valuable contribution to pinpointing the most important aspects of functioning in CWP, which can be compared to the functioning properties used in the revision process of the ICD.

Although the two studies focused on different aspects, the results are complimentary. Both point out the most essential aspects of functioning in patients with CWP. The results also provide evidence for the further development of the ICF based on the results of the qualitative study in which areas of functioning have been identified that are not yet included in the ICF. These areas can now be included in the update process of the ICF which has already started and for which German physicians and ICF users are called upon to participate in the process [43]. The validation of the ICF Core Sets will be part of the process of implementing the ICF in clinical practice. There are examples illustrating the significance of the ICF in general. The International Society for Physical and Rehabilitation Medicine (ISPRM) has adapted the ICF Core Sets as a tool for addressing functioning and disability and recommends using the ICF Core Sets in clinical practice. This worldwide implementation will take place over the years to come. In light of the new requirement of the German pension insurance to introduce the ICF in clinical rehabilitation practice, the ICF Core Set could be used as an approach in assessment, assignment, intervention and evaluation of a given illness. In the literature there are some examples of how this implementation can take place [44].

This thesis is a contribution to a holistic approach in assessing essential aspects of functioning in CWP because it includes the influence of the environment and personal factors. Nevertheless, the implementation of qualitative methodology as used in the first study has some limitations. The nature of qualitative methodology could make it difficult to determine the validity and reliability of the data. Confounding factors such as medication may have an effect on the state of being of the participants. However, I chose qualitative methodology because it allows for an in-depth examination of complex issues which in essence is characteristic for CWP and covers areas not possible using quantitative methods. The ICF Core Sets still need further validation to come to a consensus regarding the core elements. More research including both the patient perspective as well as statistical approaches is needed to gain more evidence.

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II Research articles

Research article 1: The Validation of the International Classification of Functioning, Disability and Health Core Set for Chronic Widespread Pain from the perspective of fibromyalgia patients

Validation of the International Classification of Functioning, Disability and Health (ICF) Core Set for Chronic Widespread Pain from the perspective of Fibromyalgia patients

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International Classification of Functioning, Disability and Health (ICF)
Patient perspective
Comprehensive ICF Core Set
Chronic Widespread Pain
Fibromyalgia
Focus group

Abstract

Introduction

Functioning is recognized as an important study outcome in Chronic Widespread Pain (CWP). The *Comprehensive ICF Core Set for CWP* is an application of the International Classification of Functioning, Disability and Health (ICF) with the purpose of representing the typical spectrum of functioning of patients with CWP. The objective of the study was to add evidence to the validation of the *Comprehensive ICF Core Set for CWP* from the patient perspective. The specific aims were to explore the aspects of functioning and health important to patients with Fibromyalgia, and to examine to what extent these aspects are represented by the current version of the *Comprehensive ICF Core Set for CWP.*

Methods

The sampling of patients followed the maximum variation strategy. Sample size was determined by saturation. The focus groups were digitally recorded and transcribed verbatim. The meaning condensation procedure was used for qualitative data analysis. After qualitative data analysis, the identified concepts were linked to ICF categories.

Results

Thirty-three patients participated in six focus groups. Fifty-four ICF categories out of 67 categories of the *Comprehensive ICF Core Set for CWP* were reported by the patients. Forty-eight additional categories which are not covered in the *Comprehensive ICF Core Set for CWP* were raised.

Conclusion

Most ICF categories of the existing version of the Comprehensive ICF Core Set for CWP could be confirmed from the patient perspective. However, several categories not included in the Core Set emerged and should be considered for inclusion.

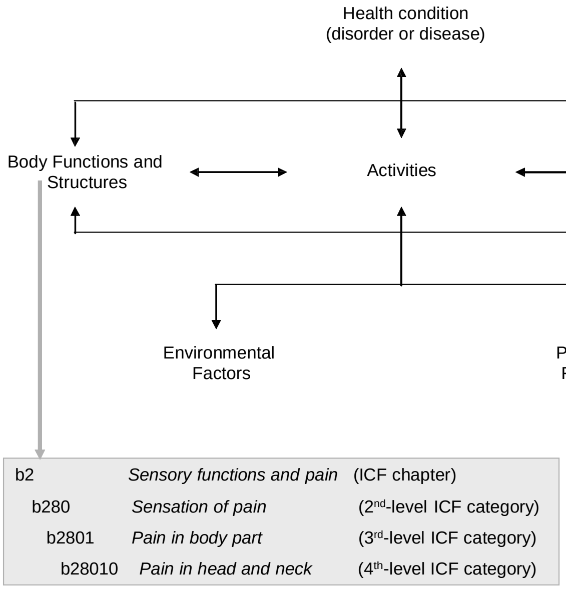
Introduction

The perspective of functioning, disability and health of the World Health Organization (WHO) [1] establishes the basis for a comprehensive description of the experience of patients suffering from a determined disease. This perspective recognizes different aspects of health from a biological, individual and social perspective, providing for a coherent view of illness [2]. This holistic approach guided the development of the International Classification of Functioning, Disability and Health (ICF), which was approved by the World Health Assembly (WHA) in May 2001. Since the ICF has been developed in a worldwide, comprehensive process and was endorsed by the WHA as a member of the WHO Family of International Classifications, it is likely to become the generally accepted framework to describe functioning, disability and health from a bio-psycho-social perspective.

Based on the bio-psycho-social perspective, the ICF classification contains the so-called ICF components *Body Functions, Body Structures, Activities and Participation* as well as the contextual factors *Environmental* and *Personal Factors* (see fig. 1). Both functioning and disability represent the result of the interaction between *Body Functions, Body Structures,* and *Activities and Participation* of an individual with a health condition and the contextual factors of that individual. The ICF classification contains more than 1400 so-called ICF categories, each allotted to the named components of the classification with the exception of the component *Personal Factors*, which has not yet been classified. Each ICF category is denoted by a code composed of a letter that refers to the components of the classification (b: *Body Functions*; s: *Body Structures*; d: *Activities and Participation* and e: *Environmental Factors*) and is followed by a numeric code starting with the chapter number (one digit), followed by the 2nd level (two digits) and the 3rd and 4th levels (one digit each) (see fig. 1).

All member states of the WHO are now called upon to implement the ICF in multiple sectors that, besides health, include: education, insurance, labour, health-and-disability policy, statistics, etc. However, the ICF has to be tailored to suit these specific applications [3]. To address the issue of feasibility regarding the over 1400 ICF categories, ICF Core Sets have been developed in a formal-decision-making and consensus-based process integrating evidence gathered from preparatory studies for a number of most burdensome, chronic health conditions. ICF Core Sets represent a selection of ICF categories out of the whole classification which can serve as minimal standards for the reporting of functioning and environmental factors for clinical studies and clinical encounters (Brief ICF Core Set) or as standards for multiprofessional, comprehensive assessment (Comprehensive ICF Core Set). Since the ICF Core Sets address aspects within all the components of the ICF (*Body Functions, Body Structures*, *Activities and Participation*, *Environmental Factors*) they present a broad perspective that may reflect the whole health experience of patients.

Figure 1 The bio-psycho-social perspective and the ICF



All member states of the WHO are now called upon to implement the ICF in multiple sectors that, besides health, include: education, insurance, labour, health-and-disability policy, statistics, etc. However, the ICF has to be tailored to suit these specific applications [4]. To address the issue of feasibility regarding the over 1400 ICF categories, ICF Core Sets have been developed in a formal-decision-making and consensus-based process integrating evidence gathered from preparatory studies for a number of most burdensome, chronic health conditions. ICF Core Sets represent a selection of ICF categories out of the whole classification which can serve as minimal standards for the reporting of functioning and environmental factors for clinical studies and clinical encounters (Brief ICF Core Set) or as standards for multiprofessional, comprehensive assessment (Comprehensive ICF Core Set). Since the ICF Core Sets address aspects within all the components of the ICF (*Body Functions, Body Structures*, *Activities and Participation*, *Environmental Factors*) they present a broad perspective that may reflect the whole health experience of patients.

One of the several health conditions for which ICF Core Sets were developed is Chronic widespread pain (CWP). The *Comprehensive ICF Core Set for CWP* describes the typical spectrum of problems in functioning among patients with CWP. Additionally, it provides an ideal basis from which to define theoretically sound models of functioning and disability in patients with CWP. The current version of the *Comprehensive ICF Core Set for CWP* includes 65 ICF categories at the 2nd level and 2 ICF categories at the 3rd level of the classification.

Fibromyalgia (FM), a health condition with unclear aetiology, is one of several related disorders, classified under CWP and is characterized by pain, fatigue, sleeping problems, mood disorder and several other symptoms as well [5]. An estimated 0.5 to 4 percent of the population suffers from FM [6]. Working, managing daily tasks and functioning in general can become a tremendous burden for the sufferers. Functioning represents the core of the patients' experience and is determined and influenced by their interaction with the environment and their own personal characteristics - not only by the health condition [7, 8, 9, 10]. This is reflected by patients themselves and also by health professionals treating patients with FM. However, due to the multidimensional nature of FM, experts in general have recognized the difficulty of finding standardized measures, thus limiting the progress in therapeutic approaches.

The lack of standardized or validated outcome measures for FM has caused uncertainty regarding which key domains of the condition should be measured. This has been acknowledged by initiatives such as the Outcome Measures in Rheumatoid Arthritis Clinical Trials (OMERACT), the goal being to define what should be measured and how, across the spectrum of rheumatology intervention and observational studies [11]. OMERACT FM workshops have been held with the objective of standardizing and improving the quality of outcome research in FM by identifying and prioritizing domains [12, 13]. The ICF can help specify OMERACT domains by serving as a conceptual model to define functioning.

The *Comprehensive ICF Core Set for CWP* is now undergoing worldwide testing and validation using a number of approaches including an international multicentre validation study and validation from the perspective of health professionals. Since patients were not directly included in the development of the ICF Core Sets, they are now explicitly involved in the validation of ICF Core Sets to establish the patient perspective in this process. As standards of functioning and health in research and clinical practice, the ICF Core Sets have to show that they address the perspective of those who experience the disease.

Qualitative methodology provides the possibility of exploring the perspective of those who experience a health problem, i.e. the patient perspective [14, 15]. Qualitative methods are now widely used and increasingly accepted in health research and health-related sciences [16, 17, 18]. One of the most broadly used techniques in qualitative research is the focus group methodology [19, 20, 21]. Focus groups are “carefully planned series of discussions designed to obtain perceptions in a defined area of interest, in a permissive, non-threatening environment” (p. 5) [22]. They are especially useful for studies involving complex issues that entail many levels of feeling and experience [23]. “The basic goal in conducting focus groups is to hear from the participants about the topics of interest to the researcher" (p. 11) [24]. The idea behind this methodology is that group processes can help people explore and clarify their views [25]. The non-directive nature of focus groups allows participants to comment, explain, disagree and share experiences and attitudes [26].

The objective of the study was to add evidence to the validation of the *Comprehensive ICF Core Set for CWP* from the perspective of patients with FM. The specific aims were to explore the aspects of functioning and health important to patients with FM using focus group methodology and to examine to what extent these aspects are represented by the current version of the *Comprehensive ICF Core Set for CWP*.

Materials and Methods

Design

We conducted a qualitative study with patients suffering from FM using focus groups. The study was approved by the Ethics Commission of the medical faculty of the Ludwig-Maximilian University, Munich.

Participants

Persons with Fibromyalgia from three different sources (the FM day clinic of the Department of Physical Medicine and Rehabilitation of the Ludwig-Maximilian University Munich, the waiting list of the same clinic, and patients from a German self-help group of FM sufferers ("Deutsche Rheuma-Liga e.V.") were contacted and asked whether they would like to participate in the study. A sample was selected based on the maximum variation strategy [27] from the pool of patients who answered positively, the two criteria being disease duration and age. Only participants with FM diagnosed according to the American College of Rheumatology [28] and who gave written informed consent according to the Declaration of Helsinki 1996 were definitely selected.

Sample Size

The sample size was determined by saturation [29]. Saturation refers to the point at which an investigator obtains sufficient information from the field [24] (see *data analysis: saturation of data*).

Material

Participants filled out a patient questionnaire including sociodemographic and disease related variables. An established topic guide with guidelines describing how to prepare and perform the focus group sessions as well as open-ended questions was applied [30]. During the focus group sessions, a visual presentation of the open-ended questions was used for better comprehension.

Data collection

All focus groups were conducted in a non-directive manner by the same moderator (RH) and one group assistant (MC). The moderator and group assistant were psychologists with expertise in the ICF and in conducting group processes.

According to the topic guide patients were asked (a) which FM-related problems of their body functions they were experiencing, (b) which body structures were involved, (c) which limitations of activities and restrictions in participation were significant to them and (d) which environmental factors and (e) which personal factors were barriers or facilitators for them. In addition to these open-ended questions – representing the ICF components - all titles and definitions of the ICF chapters of which categories are included in the *Comprehensive ICF Core Set for CWP* were presented. Patients were then encouraged to describe in their own words any problems they personally experienced related to each specific ICF chapter. To gain more information relevant to the participants, they were asked after the presentation of all chapter titles of each of the ICF components, if they thought anything was missing (Table 1 also presents examples for the ICF chapters).

At the end of each focus group session, a summary of the main results was given back to the group to enable the participants to verify and amend emergent issues.

The focus group sessions were digitally recorded and transcribed verbatim. The assistant observed the process within the group session and took field notes according to a standardized coding schema. Field notes refer to descriptive observations of the group interaction and of the topics of discussion. To review the course of the focus group, a debriefing with the moderator and assistant took place after each focus group.

Table 1 **Open-ended questions of the focus group; including a brief example from the component *Activities & Participation***

Open-ended questions
If you think about your body and mind, what does not work the way it is supposed to?
If you think about your body, in which parts are your problems?
If you think about your daily life, what are your problems in this area?
*- The next area is called Mobility. This area involves everything having to do with movement. If you think about your daily life, what are your problems in this area? **
*- The next area is called self-care. If you think about your daily life, what are your problems in this area? **
- *….*
- Can you think of anything else missing in this area regarding your daily life?
If you think about your environment and your living conditions, what do you find helpful or supportive?
If you think about your environment and your living conditions, what barriers do you experience?

* These detailed questions were used in all components for all chapters containing ICF categories in the *Comprehensive ICF Core Set for CWP*.

Data analysis

Qualitative analysis

The meaning condensation procedure [30] was used for the qualitative data analysis. In the first step, the transcripts of the focus groups were read through to get an overview of the collected data. In the second step, the data were divided into meaning units, and the theme that dominated a meaning unit was determined. A meaning unit was defined as a specific unit of text, either a few words or a few sentences with a common theme [31]. Therefore, a meaning unit division did not follow linguistic grammatical rules. Rather, the text was divided where the researcher discerned a shift in meaning [31]. In the third step, the concepts contained in the meaning units were identified. A meaning unit could contain more than one concept.

Linking to the ICF

The identified concepts were linked to ICF categories based on established linking rules [32, 33] in a systematic and standardized way. According to these linking rules, health professionals trained in the ICF are advised to link each concept to the ICF category representing this concept most precisely.

Saturation of data

Saturation was defined as the point during data collection and analysis in which the linking of the concepts of two consecutive focus groups each reveal less than 5 percent additional ICF categories in relation to the number of ICF categories contained in the *Comprehensive ICF Core Set for CWP* which were identified in the respective previous focus group.

Confirmation of ICF categories

An ICF category of the *Comprehensive ICF Core Set for CWP* was regarded as confirmed if the respective ICF category had been identified after linking the information recorded from the focus groups to the ICF.

Additional ICF categories

All ICF categories identified in the focus groups which are included in the ICF but not in the current version of the *Comprehensive ICF Core Set for CWP* are reported as additional categories. To allow for a quick overview only 2nd level ICF categories are presented in the tables.

Accuracy of the analysis

To ensure the accuracy of data analysis two strategies were conducted: First, *multiple coding*, which refers to performing the qualitative analysis and the linking to the ICF of the first focus group by two health professionals. The two health professionals compared their data analysis and documented the discussion. Second, *peer review*, which refers to analyzing and linking random samples of 15 percent of the transcribed text and 15 percent of the identified concepts (of the first health professional) by a second health professional. The degree of agreement between the two health professionals regarding the linked ICF categories was calculated by kappa statistic with 95%-bootstrapped confidence intervals [34, 35]. The values of the kappa coefficient generally range from 0 to 1, whereas 1 indicates perfect agreement and 0 indicates no additional agreement beyond what is expected by chance alone. The Kappa analysis was performed with SAS for windows V9.1.

Results

Description of the focus groups

A total of 33 participants were included in six focus groups. Participants' characteristics are summarized in table 2. The focus group sessions lasted from 70 to 115 minutes (mean 1:40 h) including a short break.

Table 2 Characteristics of participants

Characteristics of participants	
Age, years (M, range)	54,4 (36 - 69)
Gender (n female/male)	30/3
Disease duration (based on date of diagnosis), years (M, range)	3,06 (0-17)
Living alone (n)	9
Employment status (n)	
Paid employment/ self-employed	16
Homemaker	3
Unemployed (for health reasons)	5
Unemployed (for other reasons)	3
Pensioned	5
Pensioned do to CWP	1

Qualitative analysis and linking

A total of 1686 concepts were identified in the focus groups. These concepts were linked to 247 different ICF categories of the 1st to the 4th level. There were 277 concepts which could not be linked to ICF categories. Of these, 143 could be allotted to the component *Personal Factors* (e.g. aspects of coping, disease management,) and 90 concepts were not comprised in the ICF classification, therefore defined as “not covered” (nc) (e.g. time-related aspects, benefits of heat or exercise). 44 concepts were labelled “not definable” (nd), which means that the concept is too unspecific to be assigned to a concrete ICF category (e.g. quality of life in general).

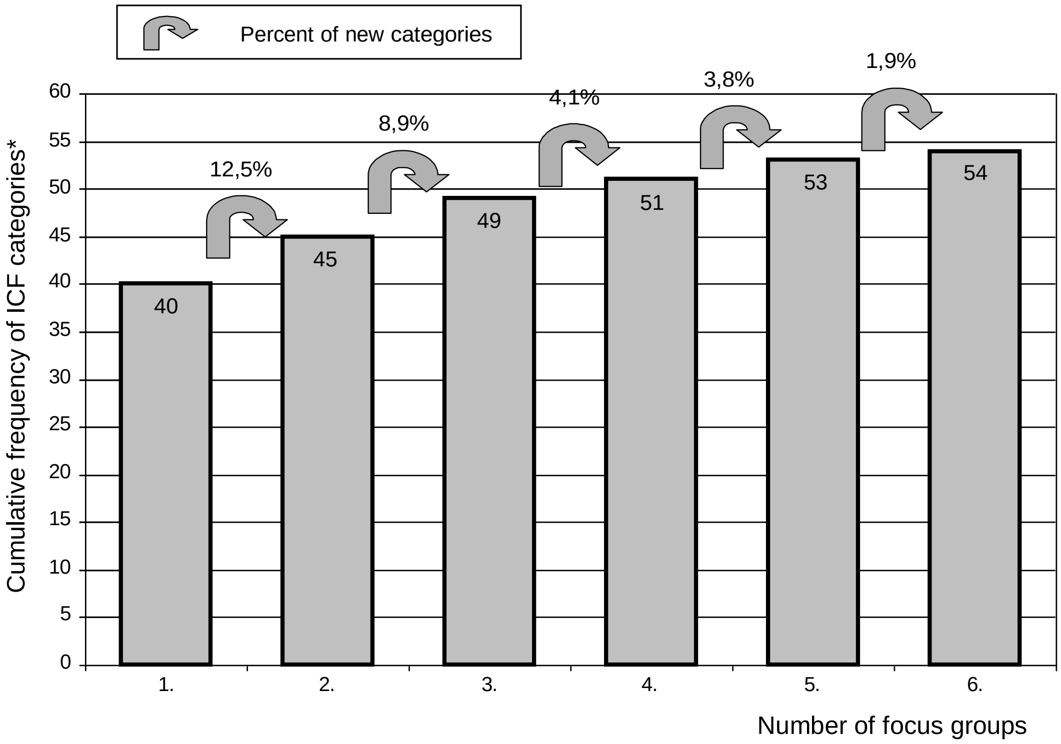
Some concepts named by the participants were more specific than the corresponding most specific ICF category. For example, the participants reported several issues pertaining to the pain quality (pressure pain, rest pain, stabbing pain), which are not specifically covered by the existing ICF categories at that level of detail. Therefore, all of these concepts referring to different qualities of pain were linked to the ICF category *b280 sensation of pain*.

Saturation of data

Regarding the ICF categories of the *Comprehensive ICF Core Set for CWP,* saturation of data was reached after conducting six focus groups (see figure 2).

Figure 2 **Saturation of the qualitative data in the focus groups**

**cumulative frequency of identified ICF categories of the Comprehensive ICF Core Set for CWP*



Confirmation of the Comprehensive ICF Core Set for CWP

In total, 54 out of the 67 ICF categories included in the *Comprehensive ICF Core Set for CWP* were confirmed by the participants: Fifteen out of the 23 categories of *Body Functions*, the one category of *Body Structures*, 25 out of the 27 categories of *Activities and Participation* and 13 out of the 16 categories of *Environmental Factors* (tables 3 – 5).

Table 3 ***Body Functions*** **(b) and** ***Body Structures*** **(s): Participants' reporting of ICF categories**

ICF code	ICF category title	Number of linked concepts
ICF categories of the Comprehensive ICF Core Set for CWP		
b122	Global psychosocial functions	not reported
b126	Temperament and personality functions	38
b130	Energy and drive functions	51
b134	Sleep function	13
b140	Attention functions	14
b147	Psychomotor function	not reported
b152	Emotional functions	42
b1602	Content of thought	not reported
b164	Higher-level cognitive functions	not reported
b180	Experience of self and time functions	2
b260	Proprioceptive function	not reported
b265	Touch function	17
b270	Sensory functions related to temperature and other stimuli	8
b280	Sensation of pain	159
b430	Haematological system functions	not reported
b455	Exercise tolerance functions	18
b640	Sexual functions	not reported
b710	Mobility of joint functions	11
b730	Muscle power functions	19
b735	Muscle tone functions	2
b740	Muscle endurance functions	not reported
b760	Control of voluntary movement functions	1
b780	Sensations related to muscles and movement functions	40
s770	Additional musculoskeletal structures related to movement	1
Additional ICF categories		
b114	Orientation functions	2
b144	Memory functions	32
b210	Seeing functions	5
b220	Sensations associated with the eye and adjoining structures	4
b230	Hearing functions	12
b240	Sensations associated with hearing and vestibular function	8
b250	Taste function	4
b255	Smell function	2
b420	Blood pressure functions	6
b440	Respiration functions	2
b450	Additional respiratory functions	1
b460	Sensations associated with cardiovascular and respiratory functions	11
b510	Ingestion functions	6
b515	Digestive functions	3
b525	Defecation functions	10
b535	Sensations associated with the digestive system	3
b620	Urination functions	19
b650	Menstruation functions	2
b770	Gait pattern functions	2
b810	Protective functions of the skin	1
b820	Repair functions of the skin	1
b830	Other functions of the skin	2
b840	Sensations related to the skin	1

Table 4 ***Activities and Participation*** **(d): Participants' reporting of ICF categories**

ICF code	ICF category title	Number of linked concepts
ICF categories of the Comprehensive ICF Core Set for CWP		
d160	Focusing attention Solving	5
d175	problems Undertaking	not reported
d220	multiple tasks Carrying out	not reported
d230	daily routine	18
d240	Handling stress and other psychological demands	5
d410	Changing basic body position	44
d415	Maintaining a body position	20
d430	Lifting and carrying objects	18
d450	Walking	20
d455	Moving around	12
d470	Using transportation	1
d475	Driving	19
d510	Washing oneself	6
d540	Dressing	19
d570	Looking after one's health	2
d620	Acquisition of goods and services	4
d640	Doing housework	39
d650	Caring for household objects	5
d660	Assisting others	3
d720	Complex interpersonal interactions	2
d760	Family relationships	3
d770	Intimate relationships	9
d845	Acquiring, keeping and terminating a job	5
d850	Remunerative employment	3
d855	Non-remunerative employment	2
d910	Community life	1
d920	Recreation and leisure	30
Additional ICF categories		
d110	Watching	3
d115	Listening	5
d155	Acquiring skills	9
d163	Thinking	1
d166	Reading	3
d210	Undertaking a single task	1
d310	Communicating with – receiving – spoken messages	2
d330	Speaking	3
d360	Using communication devices and techniques	2
d440	Fine hand use	27
d445	Hand and arm use	15
d465	Moving around using equipment	5
d520	Caring for body parts	14
d630	Preparing meals	7
d740	Formal relationships	2

Table 5 ***Environmental Factors* (e): Participants' reporting of ICF categories**

ICF code	ICF category title	Number of linked concepts
ICF categories of the Comprehensive ICF Core Set for CWP		
e1101	Drugs	8
e310	Immediate family members	23
e325	Acquaintances, peers, colleagues, neighbours and community members	6
e355	Health professionals	9
e410	Individual attitudes of immediate family members	12
e420	Individual attitudes of friends	6
e425	Individual attitudes of acquaintances, peers, colleagues, neighbours and community members	4
e430	Individual attitudes of people in positions of authority	2
e450	Individual attitudes of health professionals	29
e455	Individual attitudes of other professionals	2
e460	Societal attitudes	not reported
e465	Social norms, practices and ideologies	not reported
e570	Social services, systems and policies	2
e575	General social services, systems and policies	not reported
e580	Health services, systems and policies	36
e590	Labour and employment services, systems and policies	3
Additional ICF categories		
e110	Products or substances for personal consumption	10
e115	Products and technology for personal use in daily living	39
e225	Climate	11
e320	Friends	3
e330	People in positions of authority	1
e340	Personal care providers and personal assistants	1
e415	Individual attitudes of extended family members	2
e555	Associations and organisational services, systems and policies	13
e560	Media services, systems and policies	4
e595	Political services, systems and policies	2

Additional categories

Forty-eight additional 2nd level ICF categories which are not included in the current version of the *Comprehensive ICF Core Set for CWP* were identified in the focus groups (tables 3-5). Most of the additional ICF categories stem from *Body Functions* (n=23) followed by *Activities and Participation* (n=15). Ten additional ICF categories reported by the participants related to *Environmental Factors*. No additional ICF categories from *Body Structures* were identified. Twenty-two further 3rd and 4th level ICF categories emerged (not shown in tables) mainly from the *Activities and Participation* chapter '*Mobility*' (e.g. *d4401 grasping*, *d4153 maintaining a sitting position*, *d4552 running*), and the *Body Functions* chapters '*Mental functions*' (e.g. *b1300 energy level*, *b1301 motivation*, *b1343 quality of sleep*) and '*Neuromusculoskeletal and movement-related functions*' (e.g. *b7801 sensation of muscle spasm*, *b7300 power of isolated muscles and muscle groups*).

Accuracy of the analysis

The kappa coefficient for the agreement between the two investigators (peer review) was 0.76. The 95%-bootstrapped confidence interval was 0.70 – 0.82.

Discussion

Most ICF categories of the current version of the *Comprehensive ICF Core Set for CWP* could be confirmed from the patient perspective by FM patients. This study also confirmed relevant outcomes of treatment in CWP and FM, respectively from the patient perspective such as pain, fatigue, sleep disorders, psychological distress, lack of muscle power, difficulties changing and maintaining a body position and difficulties carrying out a daily routine [36, 37]. Pain proved to be the central topic reported by patients, with a total of 159 concepts linked to the ICF Core Set category *sensation of pain* (b280).

Apart from pain, the most outstanding theme reported by participants was the attitude of others regarding FM. The patients describe often feeling left alone with their illness, due to a lack of understanding and acceptance from others. Several patients reported feeling as if FM is not accepted as a legitimate illness by some doctors and health care professionals and is often trivialised by friends, relatives and colleagues, thus adding to the burden of pain and exhaustion. Fifty-five concepts concerning negative attitudes of others regarding the illness were linked to the corresponding ICF Core Set categories (e410, e420, e425, e430, e450, e455). Forty-six additional concepts were linked to the 1st level ICF category *attitudes* (e4). Several studies report similar findings such as patients' experiences of stigma [38, 39, 40, 41] and studies documenting controversy as to the existence, classification and acceptance of FM by health care professionals [42, 43, 44, 45].

Thirteen ICF categories in the *Comprehensive ICF Core Set for CWP* were not at all mentioned by the focus groups. Most of the ICF categories belonged to *Body Functions* and included *global psychosocial functions* (b122), *psychomotor function* (b147), *content of thought* (b160 2), *proprioceptive function* (b260) and *haematological system functions* (b430). Some categories were not confirmed but were linked to similar categories, e.g. 18 concepts were linked to the category *carrying out daily routine* (d230) instead of *undertaking multiple tasks* (d220), and 9 concepts were linked to *intimate relationships* (d770) instead of *sexual functions* (b640). Sometimes the participants made more specific statements which were linked to similar ICF categories, e.g. although the ICF Core Set category *societal attitudes* (e460) was not linked, several statements were linked to categories e410 through e455 specifying individual attitudes (e.g. individual attitudes of friends, colleagues, people in positions of authority, health professionals).

Forty-eight additional 2nd level ICF categories that are not covered in the current version of the *Comprehensive ICF Core Set for CWP* were raised. Most of the additional ICF categories belong to *Body Functions* followed by *Activities and Participation* and *Environmental Factors.* Some of these additional ICF categories need special discussion. Several concepts deal with difficulties in cognitive functioning. Thirty-two concepts were linked to the *Body Functions* category *memory functions* (b144). The patients reported problems with short and long term memory such as absorbing, storing, and recalling information. Learning and applying knowledge was also perceived as challenging for the participants. Difficulties acquiring skills, thinking, hearing, listening and reading were frequently reported by the focus group participants. Poor memory performance and problems in cognitive functioning in FM sufferers has been well documented and is in accordance with other studies [46, 47, 48, 49]. Sensations associated with hearing, such as tinnitus and dizziness were also reported by the participants, as in other studies [50, 51].

The use of the hands and arms is a further topic not included in the *Comprehensive ICF Core Set for CWP* and which FM-sufferers experience as very burdening. The participants reported difficulties in grasping, picking up and manipulating objects with their hands and pulling, reaching and turning or twisting the arms, making everyday activities and tasks very difficult to fulfil. Twenty-seven and fifteen concepts were linked to *fine hand use* (d440) and *hand and arm use (d445)*, respectively.

An additional topic found among the participants but not included in the *Comprehensive ICF Core Set for CWP* was functions of the digestive system. Such problems included difficulties with salivation, swallowing and digesting food. Urinal and intestinal irregularities were frequently reported and experienced as extremely hindering, affecting numerous activities and the participation in sports and social engagements. Irritable bowel syndrome and urinary problems in FM sufferers are reported in other studies as well [52, 53, 54]. Twenty-two concepts were linked to functions related to the digestive system (b510-b535). Some participants mentioned feeling as if fingers, hands, feet or legs were swollen although swelling was not always visible. These concepts were labelled “not definable” (nd). Other topics had to do with *Environmental Factors* not covered in the ICF classification (labelled nc). Numerous patients mentioned the benefits of heat, such as using hot or warm water to sooth aching body parts. Several others recognized the importance of exercise in coping with pain and fatigue.

The characteristics of the sample in this study (gender, age, disease duration) are comparable to samples in other national [55] and international studies [56].

It is important to mention, that several strategies were used to improve and verify the trustworthiness of the data analysis. *(1) Triangulation* ensured the comprehensiveness of data. We included data triangulation by using two data analysts (investigator triangulation: multiple coding) [57, 58]. *(2) Reflexivity* was assured by conducting a research diary for the documentation of memos concerning the design, data collection and data analysis. *(3) Clear exposition* was used establishing guidelines for conducting the focus groups (including open-ended questions), verbatim transcription, and linking rules [33, 34]. *(4)* Finally, *peer review* as described earlier. The kappa coefficient of 0.76 for the accuracy of the peer review is remarkably high compared to other studies reporting kappa statistics and can be regarded as ‘substantial agreement’ [28, 59, 60].

There are some limitations of this study that need special mentioning. The sample consists primarily of German residents. To establish a cross-cultural perspective we suggest that our methods be used in similar studies in other countries. Second, FM is a subtype of CWP, and may not be representative of all CWP conditions. Other ICF categories may have emerged if focus groups had been conducted with other CWP illnesses such as Chronic Fatigue or Gulf War Syndrome. The controversy concerning the existence, classification and acceptance of FM interferes with the patients need to be recognized and taken seriously with their illness. This may exacerbate symptoms and add to the burden of pain and exhaustion. Third, the linking process was performed by two psychologists according to established linking rules [33, 34]. However, it remains unclear whether other health professionals would have decided differently. Finally, we conducted six focus groups following the strategy of saturation during data analyses, with the criteria of two consecutive focus groups each revealing less than five percent additional ICF categories in relation to the number of ICF categories of the *Comprehensive ICF Core Set for CWP* identified in the respective previous focus group. Participants in a seventh focus group might still report new themes and concepts not yet addressed.

Initiatives such as the OMERACT address the challenge of standardizing and improving the quality of outcomes research by finding a common terminology and a common model of functioning and disability. The OMERACT FM workshop agreed upon the most important key domains to measure in FM. Some of the key domains mentioned are pain, patient global sense of well-being, fatigue, multidimensional aspects of functioning, sleep, depression, and treatment side effects. These domains are included in the *Comprehensive ICF Core Set for CWP* which can in turn be used as a basis for the further specification of OMERACT domains and the development of new instruments to assess functioning for research. A further key research objective of the OMERACT initiative will be to include the patient perspective on what represents a clinically meaningful change in a domain or the syndrome as a whole. This study can help enhance the knowledge of FM by including the patient perspective. However, further research in the context of the development and confirmation of ICF Core Sets is needed. The results of this study will be presented at an international WHO conference and will be taken into account for the decision on the final version of the *Comprehensive ICF Core Set for CWP*.

Conclusions

It is important to consider the patient perspective for the validation of the *Comprehensive ICF Core Set for CWP*. Most ICF categories of the existing version of the *Comprehensive ICF Core Set for CWP* could be confirmed by focus groups with FM patients. Several additional categories not represented in the *Comprehensive ICF Core Set for CWP* emerged from the focus groups and should be considered for inclusion in the final version.

List of abbreviations used

CWP	Chronic Widespread Pain
FM	Fibromyalgia
ICF	International Classification of Functioning, Disability and Health
OMERACT	Outcome Measures in Rheumatoid Arthritis Clinical Trials
WHO	World Health Organization
WHA	World Health Assembly

Authors´ contributions

RH conceived and organized the study and drafted the manuscript. MC participated in the performance of the focus groups, the data analysis and was involved in the peer review. GS was responsible for the overall design of the development and the validation of ICF Core Sets. AW guided the study with his input on fibromyalgia. AC participated in the development of the study design and accompanied the study implementation.

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Research article 2: Identification of essential elements of functioning in Chronic Widespread Pain based on a statistical approach

Identification of essential elements of functioning in Chronic Widespread Pain based on a statistical approach

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Abbreviated title: Identification of essential elements of functioning in CWP

Keywords:

- International Classification of Functioning, Disability and Health (ICF)
- Brief ICF Core Set
- Chronic Widespread Pain
- Fibromyalgia

Abstract

Objective

To study the most relevant ICF categories for describing functioning and disability in patients with CWP. Specific aims: (1) to identify which ICF categories explain the most variance of the experience of health in CWP and (2) to compare the identified ICF categories to the ICF categories of the Brief ICF Core Set for CWP.

Design

The ICF categories entered in an initial regression model were selected according to their correlation with item one of the SF-36. Based on an initial regression model, additional regression models were performed by systematically substituting the ICF categories included in the initial model with ICF categories from the same chapter with which they highly correlated.

Results

11 categories were identified. Six of them are included in the Brief ICF Core Set for CWP.

Conclusions

The majority of categories identified in the regression models are similar to the domains identified in OMERACT workshops and are represented in the Brief ICF Core Set for CWP, either directly or in ICF categories from the same chapters. Based on the 11 identified categories, clinicians and health professionals can obtain an efficient overview regarding the level of functioning of their patients in those essential areas that best differentiate among various levels of functioning.

Introduction

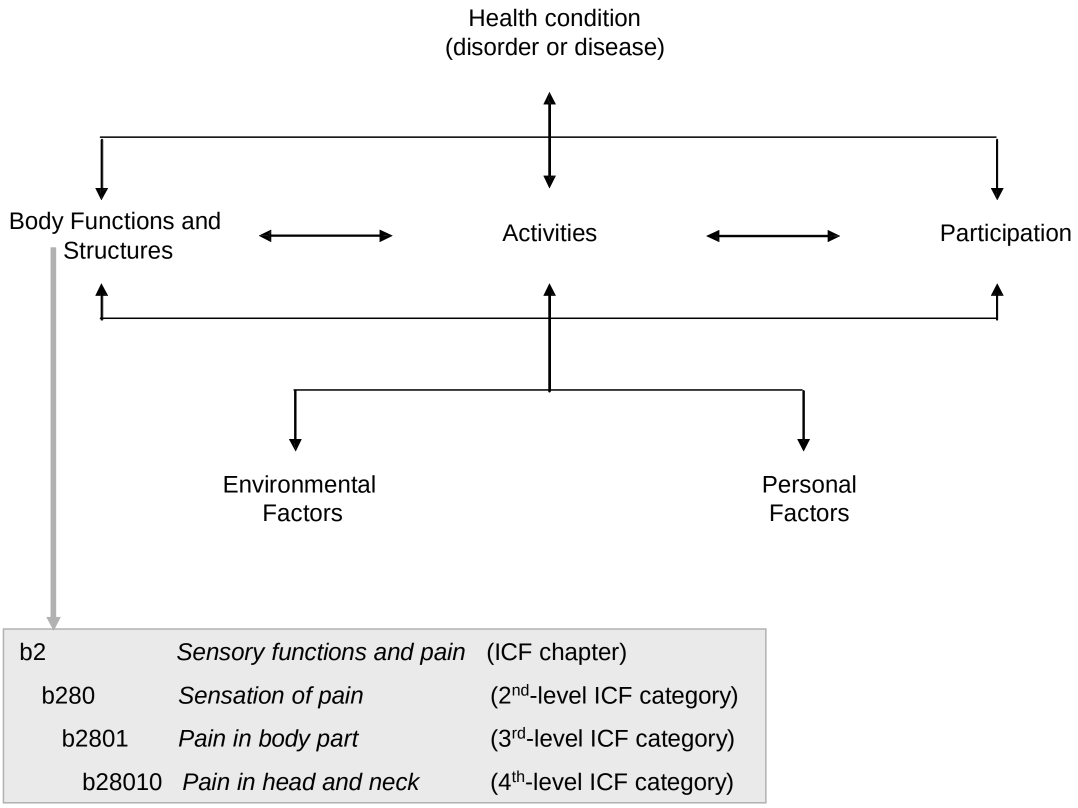
Chronic Widespread Pain (CWP) is a debilitating musculoskeletal disorder commonly found in primary care. CWP has been described as one of the main health problems in the western world [1] and is estimated to affect 11 to 13 percent of the population [2, 3, 4]. According to the criteria of the American College of Rheumatology (ACR), patients who have persistent pain in at least two contra lateral quadrants of the body and in the axial skeleton for at least three months are classified as having CWP [5]. Functioning and disability represent the core experience in patients suffering from CWP. Apart from pain, sufferers experience psychological distress, fatigue, poor sleep and a range of other debilitating symptoms [5, 6]. CWP is very much associated with the inability to continue working, ultimately leading to the experience of restriction in many other areas of life [7, 8, 9, 10].

The essential role of functioning has been acknowledged by initiatives such as the OMERACT (Outcome Measures in Rheumatoid Arthritis Clinical Trials), with the goal of defining what should be measured and how, across the spectrum of rheumatology intervention and observational studies [11]. Fibromyalgia (FM), an extreme form of CWP, is one of the several conditions addressed in OMERACT workshops [12, 13]. Also, the large number of patient-reported outcomes (e.g. Pain Disability Index (PDI) [14], Fibromyalgia Impact Questionnaire (FIQ) [15]) addressing the areas of functioning relevant to patients with CWP and the large number of studies including these instruments recognize the importance of functioning in the outcome assessment of CWP [16].

Both, the OMERACT initiative as well as the development of patient-reported outcomes were initiated according to the medical perspective [17], and are therefore based on the assumption that salient patient problems in functioning are caused by the illness. However, a consistent holistic concept necessary to describe and assess aspects of functioning in CWP is developing only slowly among scientist and clinicians in the field. The influence of the environment and personal factors is still rarely taken into account [18, 19]. However, several studies show that patients’ experience of functioning is determined by their interaction with the environment and personal factors - not only by the health condition [20, 21, 22, 23].

The bio-psycho-social perspective of functioning and disability of the World Health Organization (WHO) establishes the basis for a comprehensive description of the experience of patients. This perspective has been concretized with the International Classification of Functioning, Disability and Health (ICF). The ICF contains all concrete domains (i.e. ICF categories) necessary in describing functioning. Based on this perspective, functioning with its components *Body Functions, Body Structures, Activities and Participation* and *Environmental Factors* is seen in relation to the health condition (figure 1). Functioning denotes the result of the interaction between an individual with a health condition and the contextual factors (*Environmental Factors* and *Personal Factors*) of that individual.

Figure 1 **The bio-psycho-social perspective and the ICF**



b2	*Sensory functions and pain*	(ICF chapter)
b280	*Sensation of pain*	(2nd-level ICF category)
b2801	*Pain in body part*	(3rd-level ICF category)
b28010	*Pain in head and neck*	(4th-level ICF category)

There are some initiatives to integrate the ICF in the OMERACT process based on so-called ICF Core Sets [24]. ICF Core Sets represent a selection of ICF categories out of the whole classification which can serve as standards for multiprofessional, comprehensive assessment (Comprehensive ICF Core Set) or as minimal standards for the reporting of functioning and environmental factors for clinical studies and clinical encounters (Brief ICF Core Set). ICF Core Sets for CWP based on expert opinion have already been defined in a formal-decision-making and consensus-based process integrating evidence gathered from preparatory studies [25].The current version of the Brief ICF Core Set for CWP includes a total of 26 categories (10 *Body Functions,* 10 *Activities and Participation*, six *Environmental Factors)*. The ICF categories of the Brief ICF Core Set for CWP are shown in table 5. An open question is still to what extent these 26 ICF categories represent the essential aspects of functioning in CWP and whether all of them should remain in the final Brief ICF Core Sets for CWP. If a reduction of the categories is achieved, a description of the functioning level of patients with CWP could be performed in an efficient way in clinical practice.

Thus, the objective of this investigation was to study the most relevant ICF categories for describing functioning and disability in patients with CWP using a statistical approach. The specific aims were (1) to identify which ICF categories explain the most variance of the experience of health in CWP and (2) to compare the identified ICF categories to the ICF categories of the Brief ICF Core Set for CWP which was decided based upon expert opinion.

Materials and Methods

Study design

The study was an international multi-center, cross-sectional study. Participants gave written informed consent according to the Declaration of Helsinki 1996. The study protocol and the informed consent forms were approved by the responsible Ethics Committees in each involved center and country.

Subjects

We used data from a convenience sample of patients with CWP included in a multicenter international cross-sectional study performed in cooperation with the World Health Organization (WHO). The patients for this study was derived from 22 study centers in 10 different countries including Austria, Argentina, Brazil, Germany, Hungary, Italy, Kuwait, Philippines, Spain and Switzerland.

Inclusion criteria for patients were: diagnosis of CWP according to the ACR criteria, at least 18-years-of-age, sufficient knowledge of the official language of the corresponding country, comprehension of the purpose of the study, and signed informed consent.

Measures

The following measures were used:

(1) The *Comprehensive ICF Core Set for CWP*: The *Comprehensive ICF Core Set for CWP* has a total of 67 ICF categories. To evaluate the extent of a patient’s problem in each of the ICF categories, a generic qualifier scale was used. The qualifier scale of the components *Body Functions*, *Body Structures* and *Activities and Participation* has five response options, each ranging from 0 to 4 representing no, mild, moderate, severe or complete impairment. The qualifier scale of the component *Environmental Factors* has nine response options. A specific environmental factor can be a barrier (-1 to -4), or a facilitator (1 to 4), or can have no influence (0) on a patient’s life. If a factor does have an influence, the extent of the

influence (positive or negative) can be coded as mild, moderate, severe or complete. The response option “8 (not specified)” is used when the available information is not sufficient to determine the severity of the problem, and “9 (not applicable)”, when a category is not applicable.

(2) The Medical Outcome Study Short Form 36 (SF-36): The SF-36 [26] includes eight multi-item scales containing two to ten items each and a single item to assess health transition. The scales cover the dimensions of physical health, mental health, social functioning, role functioning, general health, pain, and vitality. Two summary scales can be obtained (Physical Component Summary Score (PCS), Mental Component Summary Score (MCS)).

In addition, sociodemographic data (date of birth, gender, living situation, current work status) and disease-specific characteristics (disease duration based on date of diagnosis, CWP-related comorbidities) were collected.

Data collection

Patient recruitment and data collection were performed by health professionals at each study center. The health professionals were trained in either a structured one-day workshop or using a training video provided by the WHO ICF Collaborating Center at the Ludwig-Maximilian University, Munich to familiarize them with the ICF classification and the data collection. Data were collected during clinical routine. Health professionals were instructed to take all the clinical data they had about the patients into consideration when coding the extent of a patient’s problem in each of the ICF categories using the qualifier scale. If this information was not sufficient to perform the rating, they were requested to consult the patient. Data collection at the respective study centers were performed from September 2004 to June 2007.

Data analysis

Descriptive analysis

Descriptive statistics were used to define the study population. Description of the health status of the patients was based on PCS and MCS Scores of the SF-36.

Bivariate analysis and data preparation

The first question of the SF-36 (SF-36_1), which addresses health in general (“In general, would you say your health is (excellent, very good, good, fair, or poor)?”), was selected as dependent variable.

To select the variables that should enter in the multivariate analysis the correlation of the ICF categories of the Comprehensive ICF Core Set for CWP and SF-36_1 were performed using the Pearson product-moment correlation coefficient (r).

Empirical work has consistently shown that the SF-36_1 question requires recalibration, since the intervals between adjacent response categories are unequal. Therefore, the item scale values were transformed as follows: excellent=5.0, very good=4.4, good=3.4, fair=2.0 and poor=1.0 [26].

In addition, the ICF categories were prepared for further analysis. As the ICF qualifiers “8 (not specified)” and “9 (not applicable)” cannot be integrated into the ordinal scale of the ICF qualifiers, they were considered missing values.

Multivariate analysis

Multiple regression models to explain general health (SF-36_1) were calculated based upon the following procedure:

Development of initial regression models

The selection of the ICF categories to be entered in a regression model occurred in four steps:

(1) Preselection: Based on the results of the bivariate analysis, all ICF categories which significantly correlated above r=0.3 with SF-36_1 were selected.

(2) Selection of candidate categories on chapter level: (2a) If only one ICF category was left within the chapter after step (1) this category was selected to enter the next steps. (2b) If two categories were left within the chapter after step (1), both categories were selected. (2c) If more than two categories were left within the chapter after step (1) backwards regression analysis were performed to obtain a further preselection on chapter level.

(3) Selection of candidate categories on component level: Based on the categories selected in step (2) a regression model (backwards selection) was computed per component.

(4) Final model: Based on the ICF categories selected in step (3) a final model was calculated by backwards selection.

All performed regression models included age as a control variable. As the variance inflation factor (VIF) was below 4 in all models we consider collinearity to be no problem in our analysis.

Development of additional regression models

In addition to the final initial models, a further set of regression models explaining general health (SF-36_1) was performed. In a first additional regression model the variables which resulted from the initial final model (step (4)) plus the variables gender and living situation were entered. Then, in further models we systematically substituted the ICF categories with ICF categories from the same chapter with which they were highly correlated (above r=0.5), starting with the ICF category which correlated highest with the SF-36_1.

Brief ICF Core Set for CWP

We compared the categories selected as a result of the regression analysis with the categories in the Brief ICF Core Set for CWP.

Results

Data from 452 patients were collected. The patients' mean age was 49 years (SD=12.0) and 76.4 percent of the patients were female (n=346). Characteristics of participants are shown in Table 1.

Table 1 Characteristics of participants

Characteristics of participants		
Age, year	M (SD; range)	49.1 (12.0; 17 - 86)
Gender	n female (%)	346 (76.5)
Disease duration, years (based on date of diagnosis)	M (SD)	4.8 (5.9)
Living alone	n (%)	94 (20.8)
Paid employment/ self-employed	n (%)	250 (48.1)
Homemaker	n (%)	65 (14.3)
Retired	n (%)	70 (15.5)
Unemployed (for health reasons)	n (%)	37 (8.2)
Unemployed (for other reasons)	n (%)	20 (4.4)
Pensioned due to CWP	n (%)	38 (8.4)
SF-36 PCS Score (n=420)	M (SD)	32.4 (8.2)
SF-36 MCS Score (n=420)	M (SD)	40.8 (11.3)

Bivariate analysis

In the bivariate analysis a total of 27 ICF categories from the 67 categories of the Comprehensive ICF Core Set for CWP correlated above r=0.3 with general health (SF-36_1). The highest correlation was found in *d910 community life (*r=.45; p=.000) in the component *Activities and Participation.* In the component *Body Functions, b280 sensation of pain* correlated highest (r=.43; p=.000). The one single category from the component *Body Structures* as well as all 16 categories from the component *Environmental Factors* did not correlate above r=0.3 with general health (correlations shown in table 2).

Table 2 **Bivariate analysis of the ICF Core Set for CWP: ICF categories in bold print preselected for regression models**

ICF code	ICF category title	r (p)	ICF code	ICF category title	r (p)
Body Functions					
b122	Global psychosocial functions	.27 (.000)	**b126**	**Temperament and personality functions**	**.35** (.000)
b130	**Energy and drive functions**	**.36** (.000)	**b134**	**Sleep function**	**.37** (.000)
b140	**Attention functions**	**.38** (.000)	**b147**	**Psychomotor function**	**.33** (.000)
b152	**Emotional functions**	**.39** (.000)	**b1602**	**Content of thought**	**.35** (.000)
b164	**Higher-level cognitive functions**	**.31** (.000)	b180	Experience of self and time functions	.24 (.000)
b260	Proprioceptive function	.27 (.000)	b265	Touch function	.24 (.000)
b270	Sensory functions related to temperature and other stimuli	.17 (.001)	**b280**	**Sensation of pain**	**.43** (.000)
b430	Haematological system functions	.14 (.004)	b455	Exercise tolerance functions	.20 (.000)
b640	Sexual functions	.29 (.000)	**b710**	**Mobility of joint functions**	**.35** (.000)
b730	**Muscle power functions**	**.34** (.000)	**b735**	**Muscle tone functions**	**.34** (.000)
b740	**Muscle endurance functions**	**.30** (.000)	b760	Control of voluntary movement functions	.21 (.000)
b780	**Sensations related to muscles and movement functions**	**.30** (.000)			
Body Structures					
s770	Additional musculoskeletal structures related to movement	.29 (.000)			
Activities & Participation					
d160	**Focusing attention**	**.36** (.000)	d570	Looking after one's health	.20 (.000)
d175	Solving problems	.28 (.000)	d620	Acquisition of goods and Services	.29 (.000)
d220	**Undertaking multiple tasks**	**.33** (.000)	d640	Doing housework	.27 (.000)
d230	**Carrying out daily routine**	**.31** (.000)	d650	Caring for household objects	.22 (.000)
d240	**Handling stress and other psychological demands**	**.41** (.000)	d660	Assisting others	.28 (.000)
d410	**Changing basic body position**	**.33** (.000)	d720	Complex interpersonal interactions	.30 (.000)
d415	Maintaining a body position	.29 (.000)	**d760**	**Family relationships**	**.33** (.000)
d430	Lifting and carrying objects	.24 (.000)	d770	Intimate relationships	.22 (.000)
d450	**Walking**	**.31** (.000)	**d845**	**Acquiring, keeping and terminating a job**	**.43** (.000)
d455	Moving around	.28 (.000)	**d850**	**Remunerative employment**	**.42** (.000)
d470	Using transportation	.21 (.000)	**d855**	**Non-remunerative employment**	**.42** (.000)
d475	Driving	.29 (.000)	**d910**	**Community life**	**.45** (.000)
d510	Washing oneself	.23 (.000)	**d920**	**Recreation and leisure**	**.34** (.000)
d540	**Dressing**	**.32** (.000)			
Environmental Factors					
e1101	Drugs	.03 (.604)	e450	Individual attitudes of health professionals	-.18 (.000)
e310	Immediate family members	.07 (.140)	e455	Individual attitudes of other professionals	-.10 (.050)
e325	Acquaintances, peers, colleagues, neighbours and community members	-.11 (.019)	e460	Societal attitudes	-.26 (.000)
e355	Health professionals	-.15 (.001)	e465	Social norms, practices and ideologies	-.17 (.001)
e410	Individual attitudes of immediate family members	.06 (.214)	e570	Social services, systems and policies	-.18 (.000)
e420	Individual attitudes of friends	-.07(.165)	e575	General social services, systems and policies	-.17 (.001)
e425	Individual attitudes of acquaintances, peers, colleagues, neighbours and community members	-.18 (.000)	e580	Health services, systems and policies	-.08 (.108)
e430	Individual attitudes of people in positions of authority	-.20 (.000)	e590	Labour and employment services, systems and policies	-.19 (.001)

Multivariate analysis

Initial regression model

In step (1) 27 ICF categories from the components *Body Functions* and A*ctivities and Participation* (see bivariate analysis), as well as the variable age, were selected. Fourteen of these categories belonged to the component *Body Functions* and 13 to the component *Activities and Participation.* As described in step (2a) and (2b), the following ICF categories were identified: *b280 sensation of pain, d160 focusing attention, d410 changing basic body position, d450 walking, d540 dressing, d760 family relationships, d910 community life, d920 recreation and leisure* (see table 2). According to step (2c), the backwards multiple regression model was computed, including the ICF categories of the chapters in which more than two ICF categories were selected in step (1). Two *Body Functions* chapters (b1 *Mental functions, b7 Neuromusculoskeletal and movement-related functions*) and two *Activities and Participation* chapters (d2 *General tasks and demands, d8 Major life areas*) included more than two ICF categories. In step (3), backwards regression models were computed including the categories resulting from steps (2a) and (2b) from the components *Body Functions* and *Activities and Participation* respectively (see table 3).

In step (4) the final initial model was calculated including age and all ICF categories which remained in their respective previous models according to step (3). To ensure inclusion of all four ICF components, the one single category from the component *Body structures* (*s770 additional musculoskeletal structures related to movement*) was included, although the correlation with item one of the SF36 was below r=0.3. Also keeping in mind the holistic perspective of the ICF, the category *e460 societal attitudes* which had the highest correlation (r=-0.26) in the component *Environmental Factors* with SF36_1 was also included in the final model. However, no categories from this component correlated >0.3 with SF36_1.

In the final model of the initial regression model, age and the ICF categories *b152 emotional functions, b280 sensation of pain, b710 mobility of joint functions, d845 acquiring, keeping and terminating a job, d910 community life* and *e460 societal attitudes* remained in the model and explained 33 percent of the variance of general health (SF-36_1) (see table 3). The categories *b1602 content of thought, d240 handling stress and other psychological demands* and *d410 changing basic body position* did not remain in the final initial model.

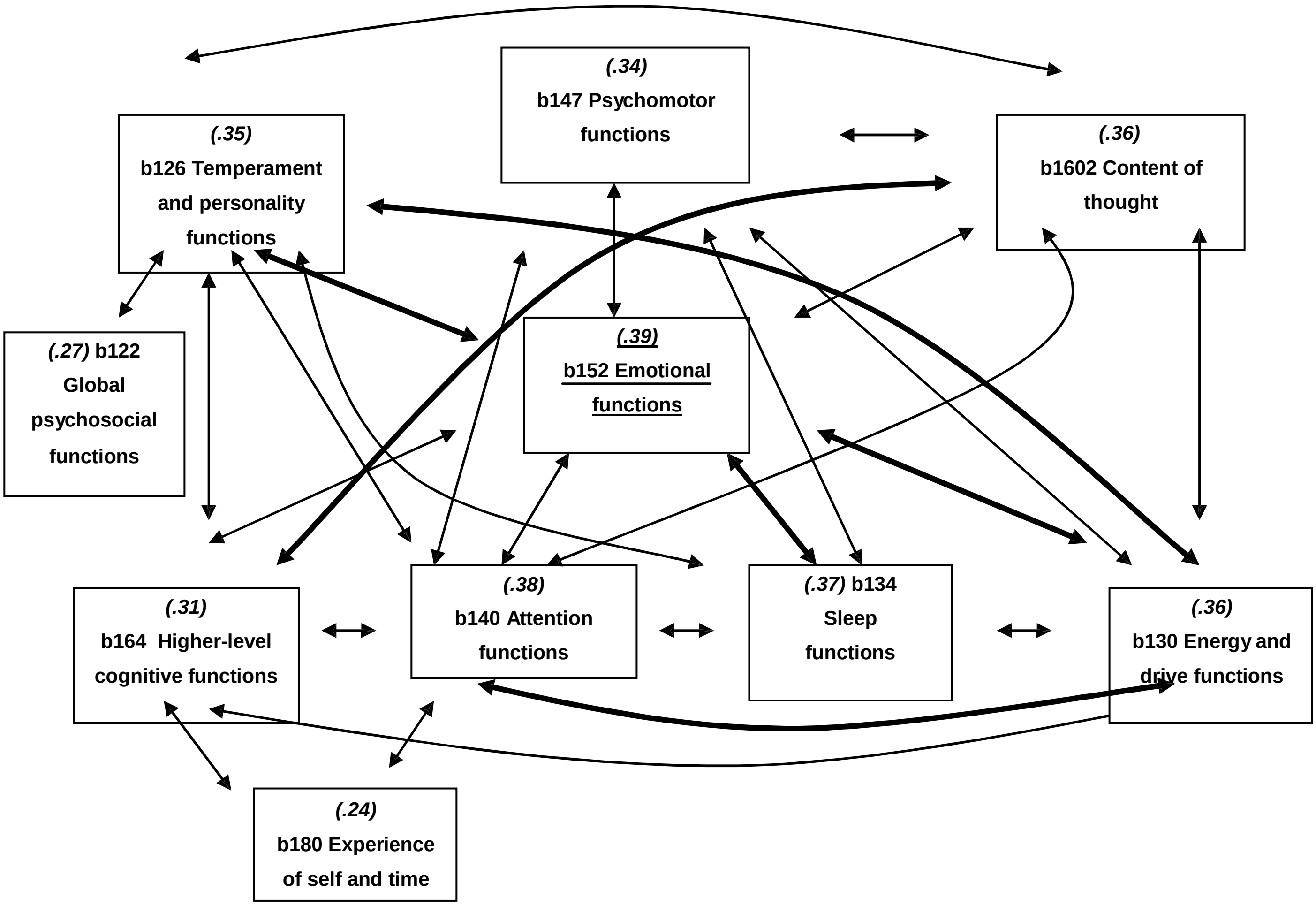
Table 3 **Results of initial regression models**

	Selection on chapter level								Selection on component level				Final Model	
	Model I		Model II		Model III		Model IV		Model V		Model VI		Model VII	
Variables	β	SE	β	SE	β	SE	β	SE	β	SE	β	SE	β	SE
Age	**-.013**	**.003**	**-.011**	**.004**	**-.015**	**.003**	**-.020**	**.003**	**-.008**	**.003**	**-.015**	**.003**	**-.011**	**.004**
b126 Temperament and personality functions														
b130 Energy and drive functions														
b134 Sleep functions	**-.086**	**.046**												
b140 Attention functions	**-.127**	**.051**												
b147 Psychomotor functions														
b152 Emotional functions	**-.124**	**.054**							**-.114**	**.049**			**-.107**	**.052**
b1602 Content of thought	**-.128**	**.056**							**-.137**	**.051**				
b164 Higher-level cognitive functions														
b710 Mobility of joint functions			**-.149**	**.050**					**-.119**	**.046**			**-.094**	**.049**
b730 Muscle power functions			**-.109**	**.056**										
b735 Muscle tone functions			**-.104**	**.052**										
b740 Muscle endurance functions														
b780 Sensations related to muscles & movement funct.			**-.095**	**.047**										
b280 Sensation of pain									**-.176**	**.040**			**-.117**	**.042**
d220 Undertaking multiple tasks														
d230 Carrying out daily routine					**-.101**	**.046**								
d240 Handling stress and other psychological demands					**-.245**	**.043**					**-.159**	**.042**		
d845 Acquiring, keeping and terminating a job							**-.241**	**.040**			**-.160**	**.039**	**-.115**	**.041**
d850 Remunerative employment														
d855 Non-remunerative employment							**-.086**	**.046**						
d160 Focusing attention														
d410 Changing basic body position											**-.116**	**.043**		
d450 Walking														
d540 Dressing														
d760 Family relationships														
d910 Community life											**-.148**	**.045**	**-.158**	**.049**
d920 Recreation and leisure														
e460 Societal attitudes													**.062**	**.031**
s770 Additional musculoskeletal struct. rel. to movement														
R²	**.24**		**.21**		**.20**		**.21**		**.28**		**.32**		**.33**	

Additional regression models

In the first additional regression model, the variables which resulted from the initial final model (step (4)) plus the variables gender and living situation were entered. In the additional regression models 2 through 8, we substituted *b152 emotional functions* with all ICF categories from the same chapter with which it correlated above r=0.5 (see figure 2).

Figure 2 **Inter-correlated ICF categories in chapter 1 mental functions. Values between brackets and cursive represent the correlation of the corresponding ICF category with item 1 of the SF-36. Correlations over 0.5 displayed in table below. Thick lines represent correlations over 0.6.**



Inter-correlations of the ICF categories in chapter 1 mental functions.

	b122	b126	b130	b134	b140	b147	b152	b1602	b164	b180
b122 Global psychosocial functions		.55								
b126 Temperament and personality functions			.66	.51	.58		.66	.56	.58	
b130 Energy and drive functions				.58	.64	.52	.64	.51	.52	
b134 Sleep functions					.53	.53	.64			
b140 Attention functions						.52	.59	.56	.59	.52
b147 Psychomotor functions							.52	.53		
b152 Emotional functions								.51	.52	
b1602 Content of thought functions									.65	
b164 Higher level cognitive functions										.52
b180 Experience of self and time functions										

Then, *b710 mobility of joint functions* was substituted with *b730 muscle power functions, b735 muscle tone functions* and *b740 muscle endurance functions* (models nine through 12). Finally, *d845 acquiring, keeping and terminating a job* was substituted with *d850 remunerative employment* and *d855 non-remunerative employment* and *d910 community life* was substituted with *d920 recreation and leisure* (models 13 through 14 and model 15 respectively). The ICF categories *b1602 content of thought*, *b126 temperament and personality functions, b147 psychomotor functions, d850 remunerative employment and d920 recreation and leisure* which remained in the respective models, explained between 33 and 35 percent of the total variance of item one of the SF-36 (see table 4).

Table 4 Results of additional regression models

Model	*substituted by (s.b.)			**r	ICF categories in the respective models						R²
Model 1					**b152**	**b280**	**b710**	**d845**	**d910**	**e460**	.33
Model 2	b152	s.b.	b140	.38	b140	**b280**	**b710**	**d845**	**d910**	**e460**	.34
Model 3	b152	s.b.	b134	.37	b134	**b280**	**b710**	**d845**	**d910**	**e460**	.35
Model 4	b152	s.b.	b130	.36	b130	**b280**	**b710**	**d845**	**d910**	**e460**	.34
Model 5	b152	s.b.	b1602	.36	**b1602**	**b280**	**b710**	**d845**	**d910**	**e460**	.35
Model 6	b152	s.b.	b126	.35	**b126**	**b280**	**b710**	**d845**	**d910**	**e460**	.34
Model 7	b152	s.b.	b147	.34	**b147**	**b280**	**b710**	**d845**	**d910**	**e460**	.34
Model 8	b152	s.b.	b164	.31	b164	**b280**	**b710**	**d845**	**d910**	**e460**	.34
Model 9	b710	s.b.	b730	.34	**b152**	**b280**	b730	**d845**	**d910**	**e460**	.34
Model 10	b710	s.b.	b735	.34	**b152**	**b280**	b735	**d845**	**d910**	**e460**	.34
Model 11	b710	s.b.	b740	.30	**b152**	**b280**	b740	**d845**	**d910**	**e460**	.34
Model 12	b710	s.b.	b780	.30	**b152**	**b280**	b780	**d845**	**d910**	**e460**	.34
Model 13	d845	s.b.	d850	.42	**b152**	**b280**	**b710**	**d850**	**d910**	**e460**	.33
Model 14	d845	s.b.	d855	.42	**b152**	**b280**	**b710**	d855	**d910**	**e460**	.33
Model 15	d910	s.b.	d920	.34	**b152**	**b280**	**b710**	**d845**	**d920**	**e460**	.34

ICF categories in bold print remained in the respective models

* Alternative ICF category which substituted the category from the same chapter in Model 1 (with which it highly correlated)

** Correlation of the alternative category with SF-36 1

Comparison with the Brief ICF Core Set for CWP

Six ICF categories remaining in the final initial and additional regression models respectively were included in the Brief ICF Core Set for CWP: *b152 emotional functions* and *b280 sensation of pain* of the initial final model and *b147 psychomotor functions, b1602 content of thought, d850 remunerative employment* and *d920 recreation and leisure* from the additional regression models. In addition, the following ICF categories were selected in the regression analysis but not included in the Brief ICF Core Set: *b126 temperament and personality* *functions, b710 mobility of joint functions, d845 acquiring, keeping and terminating a job, d910 community life* and *e460 societal attitudes* (see table 5).

Table 5 ICF categories included in the Brief ICF Core Set for CWP and selected in the regression analysis

ICF categories ICF code	ICF category title	included in Brief ICF Core Set	Selected in regression models
b126	Temperament and personality functions		+
b130	Energy and drive functions	x	
b134	Sleep functions	x	
b140	Attention functions	x	
b147	Psychomotor functions	x	+
b152	Emotional functions	x	+
b1602	Content of thought	x	+
b280	Sensation of pain	x	+
b455	Exercise tolerance functions	x	
b710	Mobility of joint functions		+
b730	Muscle power functions	x	
b760	Control of voluntary movement functions	x	
d175	Solving problems	x	
d230	Carrying out daily routine	x	
d240	Handling stress and other psychological demands	x	
d430	Lifting and carrying objects	x	
d450	Walking	x	
d640	Doing housework	x	
d760	Family relationships	x	
d770	Intimate relationships	x	
d845	Acquiring, keeping and terminating a job		+
d850	Remunerative employment	x	+
d910	Community life		+
d920	Recreation and leisure	x	+
e1101	Drugs	x	
e310	Immediate family	x	
e355	Health professionals	x	
e410	Individual attitudes of immediate family members	x	
e420	Individual attitudes of friends	x	
e460	Societal attitudes		+
e570	Social security services, systems and policies	x	

\+ confirmed in regression model

Discussion

With this study a set of 11 ICF categories essential for describing functioning and disability in patients with CWP were identified. These ICF categories represent a first proposal for a reduction of the Brief ICF Core Sets for CWP.

Six out of the 11 ICF categories were already included in the Brief ICF Core Set for CWP. These categories are *psychomotor functions, emotional functions, content of thought, sensation of pain, remunerative employment* and *recreation and leisure.* Categories not included in the Brief ICF Core Set for CWP were *temperament and personality functions, mobility of joint functions, acquiring, keeping and terminating a job, community life* (which also correlated highest with general health) and *societal attitudes.* However, all of these categories resemble categories within the same chapter which are included in the Brief ICF Core Set. For example, *temperament and personality functions* confirmed in the regression models is from the same chapter (*mental functions*) as six other categories included in the Brief ICF Core Set. Category *mobility of joint functions* confirmed in the regression models is comparable with *muscle power functions* included in the Brief ICF Core Set. During the consensus conference on the ICF Core Sets, the experts were forced to select among ICF categories that are highly related and belong to the same chapter. Experts frequently expressed their difficulty deciding among categories that are similarly important [24]. The results of this investigation may stress the complementarities and necessity of both approaches, i.e. to follow a statistical approach to inform a content-oriented expert approach.

Regarding the content of the categories selected, some have to be discussed in detail. As pain is the cardinal symptom in CWP, it is not surprising that the category *sensation of pain,* was identified and also correlated highest with general health in the *Body Functions* component. In addition, the category *emotional functions* was identified in the regression model. Emotional functions and psychological distress, especially depression and anxiety are frequent in chronic pain patients [27, 28, 29] and studies show that pain and depression are strongly associated [30]. It is widely agreed that depression contributes to suffering and disability [31]. The category *psychomotor functions* also remained in the regression model. Psychomotor functions include moving and speaking slowly, agitation and restlessness. Restless Legs Syndrome is very common in CWP, with prevalence rates as high as 64 percent in FM patients [32, 33]. The category *content of thought* reflects having distressing thoughts and/or images and somatisation. Several studies have focused on the construct of somatisation and pain and show that somatisation is a risk factor in the development of CWP [34, 35]. Furthermore, the categories *remunerative employment* and *recreation and leisure* were identified. Studies document CWP as a frequent cause for sick leave, disability pension, work dissatisfaction [36], and prominent effects on work status in patients with CWP

[37]. Involvement in social activities and recreation and engagement in all aspects of community life is strongly affected in chronic pain patients [38].

The results of the bivariate analysis showed that a total of 27 categories from the 67 categories of the Comprehensive ICF Core Set for CWP correlated with general health as represented by item one of the SF-36. To ensure inclusion of all four ICF components in the initial regression model, the one single category from the component *Body Structures* (*additional musculoskeletal structures related to movement*) and the category from the component *Environmental Factors* with the highest correlation with item one of the SF-36 (*societal attitudes)* were included, although the correlation was below r=0.3. In the final model of the initial regression, age and the categories *emotional functions, sensation of pain, mobility of joint functions, acquiring, keeping and terminating a job, community life* and *societal attitudes* remained in the model. Interestingly, the category *societal attitudes* remained in the model even though the correlation with item one of the SF-36 was below r=0.3. This finding poses the question whether or not the inclusion of at least one category from each chapter of the Comprehensive ICF Core Set – regardless of the correlation with item one of the SF-36 – may have resulted in further relevant categories remaining in the regression model.

In some ICF chapters, categories within the same chapter highly inter-correlated with one another. This can lead to unclarity regarding the true association between the variables and general health because the categories are intertwined. That is why a further set of regression models was performed by substituting the categories which correlated with other categories from the same chapter. For example, *emotional functions* (as the category within its chapter which correlated highest with the SF-36) was substituted by seven categories from the same chapter with which it correlated above p=0.5. Furthermore, all other categories which were highly inter-correlated within the chapters were included in this set of regression models. As a result of the regression, the categories which remained in their respective models were *temperament and personality functions, psychomotor functions, content of thought, remunerative employment and recreation and leisure.* Therefore, not only the categories identified in the initial regression model, but also these five categories, which resulted as a result of the additional regression models, are considered relevant to the experience of health in CWP.

According to the FM OMERACT 8 workshop, the key domains to be investigated in studies and endorsed by both clinician-investigators and patients are *pain, fatigue, sleep disturbance, multidimensional function, health-related quality of life, mood disorders* and*, cognitive dyscognition* [13]. Comparing our results with these domains, all 11 categories found as a result of the regression models are represented in the key OMERACT domains, either specifically or within a domain subsuming more than one element of functioning (such as *multidimensional function*). Category *emotional functions* corresponds with OMERACT domain *mood disorders*, although *emotional functions* reflects emotion on a broader level. Not surprisingly, the regression models and the OMERACT workshop identified *pain* as a core domain. Category *mobility of joint functions* may be represented within the rather unspecific OMERACT domain *multidimensional function,* which reflects multiple aspects of functioning, including physical, vocational, role and emotional function [12]. Our findings may add knowledge for future OMERACT workshops in the specification and operationalization of the key domain *multidimensional function*. The categories *acquiring, keeping and terminating a job, remunerative employment, community life, recreation and leisure* and *societal attitudes* identified as a result of our analysis are not specifically mentioned in OMERACT as essential domains in CWP. However, they may be embodied in OMERACT domains *multidimensional function* and/or *health-related quality of life*, which subsume a variety of domains reflecting the overall sense of well being [13]. We also identified the categories *psychomotor functions* and *content of thought,* both of which may be represented in OMERACT domain *dyscognition,* subsuming “problems with attention or concentration”, “disorganized thinking”, and “memory problems” [13]. Key OMERACT domains, which were not reflected as a result of the regression models, were *fatigue* and *sleep disturbance.* This finding again stresses the complementarities of both a purely statistical method and an expert-based method. The ICF categories b130 *energy and drive* and b134 *sleep functions* may be considered for inclusion in the final Brief ICF Core Set for CWP as was originally proposed by the experts of the conference.

There are some further limitations in this study which need to be addressed. First, although the data collected included patients from 10 different countries, almost half of them were German residents. If all countries had been represented more equally, the results may have differed. Second, the Comprehensive ICF Core Set for CWP, used to assess our data, includes 67 defined categories. However, ICF categories not included in the Comprehensive ICF Core Set for CWP may also represent relevant areas of functioning in CWP. Third, item one of the SF-36 is one of the most widely investigated single items referring to health in general. Moreover, it has high face-validity. However, as a simple and straightforward measure, it may be subjective and too broad. Similar analyses have to be performed with alternate items to get a more precise picture of what is being assessed. Fourth, it is important to emphasize that this study represents only one of many perspectives based on which the content validity of the ICF Core Set for CWP is being studied. Another essential element is the patient perspective, which was recently studied based on focus groups of FM patients [39]. It is envisioned that all different perspectives will be integrated to come up with a final version of the ICF Core Set for CWP. Finally, no assessment instruments were used to assess the extent of the problems of the CWP patients in each of the areas addressed in the ICF categories of the ICF Core Set. The coding of the health professionals was based on the clinical information they had regarding the patients and the information provided to them directly by the patients. The results of this investigation should be validated with data collected using clinical measures, the results of which are then transferred to the qualifier scale.

Conclusions

These ICF categories represent a first proposal for a reduction of the Brief ICF Core Set for CWP. All categories identified based on regression models are similar to the domains identified in OMERACT workshops and are represented in the Brief ICF Core Set for CWP, either directly or in ICF categories from the same chapters. It may be considered substituting certain ICF categories currently included in the Brief ICF Core Set for similar categories, which may reflect functioning and disability in CWP more accurately. Based on the 11 categories identified in this study, clinicians and health professionals can obtain an efficient overview regarding the level of functioning of their patients in those essential areas that best differentiate among various levels of functioning.

List of abbreviations used

CWP	Chronic Widespread Pain
ICF	International Classification of Functioning, Disability and Health
WHO	World Health Organization
OMERACT	Outcome Measures in Rheumatoid Arthritis Clinical Trials
FM	Fibromyalgia

Authors´ contributions

RH performed the statistical analysis and drafted the manuscript. MC participated in the data analysis. GS was responsible for the overall design of the development and the validation of ICF Core Sets. AW guided the data analysis with his input on fibromyalgia. AC participated in the development of the study design and accompanied the study implementation.

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FIGURE LEGENDS

Figure 1: The bio-psycho-social perspective and the ICF

Figure 2: Inter-correlated ICF categories in chapter b1 *mental functions* Values between brackets and cursive represent the correlation of the corresponding ICF category with item 1 of the SF-36. Correlations over r=0.5 displayed in table below. Thick lines represent correlations over r=0.6.