

# Physical activity, participation and health-related quality of life in chronic fatigue syndrome and multiple osteochondromas

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## Summary

Chronic fatigue and pain are prevalent symptoms in the adult general population (approximately >20%), debilitating in nature and often co-occurring in various chronic conditions. Both symptoms have been found to negatively affect health-related quality of life (HRQOL), one of the primary outcomes of healthcare and rehabilitation. HRQOL is seen as a comprehensive concept that can be influenced by biological factors, such as fatigue and pain, psychological as well as social factors. Despite the established high prevalence and negative effects of fatigue, it is not always taken into account in clinical research on chronic pain, and its importance is frequently overlooked in clinical practice. This thesis is therefore interested in the impact of chronic fatigue in patients who also experience chronic pain.

Fatigue is proposed as a transdiagnostic rather than a disease-specific symptom. To explore this hypothesis, two distinct patient populations experiencing chronic fatigue and pain were included in this doctoral thesis, patients with chronic fatigue syndrome (CFS) and patients with multiple osteochondromas (MO). Because HRQOL is an important health care outcome, this doctoral thesis is centered around the exploration of disease-specific and transdiagnostic bio-, psycho-, social determinants of HRQOL in patients with chronic fatigue and pain, and specifically taking the physical activity level into account due to its established positive relationship with health in the general population.

This thesis consists of three general aims:

1. To explore HRQOL in patients with CFS and MO;
2. To explore the physical activity level in patients with CFS and MO;
3. To identify transdiagnostic and disease-specific determinants of HRQOL.

The first part focuses on measurement instruments to evaluate activity limitations and participation restrictions, which are considered more objectively measured subitems of HRQOL, and the physical activity level and pattern in patients with CFS.

**Chapter 2** describes the results of a systematic review on measurement instruments to evaluate activity limitations and participation restrictions in patients with CFS. The main objectives of this study were to (1) explore which measurement instruments are currently used to evaluate activity limitations and participation restrictions in patients with CFS, (2) gather information on the psychometric properties of these measurement instruments in patients with CFS, and (3) determine which of these measurement instruments are suited to use in patients with CFS.

A total of 71 studies and 38 unique measurement instruments were identified, but only eight studies evaluated the psychometric properties of five measurement instrument. Psychometric studies of the Chronic Fatigue Syndrome-Activities and Participation Questionnaire (CFS-APQ), Work and Social Adjustment Scale (WSAS), Euroqol Questionnaire (EQ-5D), World Health Organization quality of life assessment instrument (WHOQOL-100) and Short-Form 36 (SF-36) were critically appraised. Based on available evidence, it could only be concluded that the psychometric properties of measurement instruments used in scientific research with patients with CFS are insufficiently evaluated

within this population. Development of new measurement instruments is strongly discouraged, given the high number of measurement instruments found ( $n = 38$ ). Instead, it is recommended that future research evaluates the unknown psychometric properties and repeats the studies of poor methodological quality with sound methodology to provide strong evidence of the quality of a psychometric property. An interesting observation is that all identified measurement instruments are self-reported, which have been found to have limited value in patients with CFS because they tend to rate their functioning worse than it actually is. Even though subjective reporting of one's functioning reflects important information for healthcare professionals, it is suggested that it be supplemented by more objective measures such as detailed assessment and observations performed by healthcare professionals.

**Chapter 3** describes the results of a systematic review on measurement instruments to evaluate the physical activity level and pattern in patients with CFS/ME. The main objectives of this study were to (1) systematically review the literature for measures or scales capable of evaluating the activity level and/or pattern that were used in patients with CFS/ME, and (2) critically appraise the psychometric properties of identified measures or scales in patients with CFS/ME. A total of 50 articles and 15 unique measurement instruments were identified, but only two studies examined the psychometric properties of three measurement instruments: the Chronic Fatigue Syndrome–Activity Questionnaire (CFS-AQ), Activity Pattern Interview (API) and International Physical Activity Questionnaire-Short-Form (IPAQ-SF). Based on these results, all three measurement instruments are considered equally (in)valid and further research is recommended to evaluate the psychometric properties of existing measures or scales.

Even though activity monitors were most frequently used in research ( $n = 29$ ), there is lack of studies investigating their psychometric properties in patients with CFS/ME. In addition, the CFS-AQ, API and IPAQ-SF are all self-reported measurement instruments reflecting patients' perception of their performed physical activity, rather than providing an accurate representation of their actual physical activity level. Activity monitors have been found capable of objectively capturing the physical activity level in the general population, but to date it is unclear whether they are reliable and valid to use in patients with CFS/ME. Given the added value of activity monitors to accurately measure patients' physical activity levels, evaluation of their psychometric properties in patients with CFS/ME is recommended.

Self-reported measures, more specifically activity diaries, are capable of capturing more information on patients' physical activity level, such as type, context and meaningfulness of an activity. Therefore, in addition to objective measures, they provide useful information for healthcare professionals to select appropriate treatment interventions. However, the systematic review of **chapter 3** did not identify a self-reported measurement instrument suitable for use in patients with CFS/ME. Therefore, the aim of **chapter 4** was to evaluate the ability of a detailed self-reported activity diary based on an instantaneous registration of activities to measure the PAL in female patients with CFS by comparing it to an activity monitor (Actical). Additional objectives were to

compare potential discrepancies between subjective (activity diary) and objective (Actical) measurement instruments in patients with CFS versus healthy controls and, in case of discrepancies between both measurement instruments, to investigate whether and which illness-related complaints, HRQOL domains or demographic factors are associated with these discrepancies. The results showed that female patients with CFS are less able to record their physical activity level with an activity diary in comparison to healthy controls. In both patients with CFS and healthy controls, younger persons tended to underestimate their physical activity level, while older persons tended to overestimate their physical activity level with an activity diary. No other factors were significantly associated with the discrepancies found between the two measurement instruments. In conclusion, the proposed detailed self-reported activity diary cannot replace activity monitoring to assess the physical activity level in patients with CFS, but may provide additional information about the perceived activity. Further exploration of factors associated with the discrepancy between self-reported and objective measurement instruments could facilitate the development or adaptation of a self-reported measurement that can be used complementary to an objective measurement instrument by encompassing more detailed information about patients' perceived activities.

The second part of this thesis focuses on the physical activity level and HRQOL in patients with MO, and transdiagnostic and disease-specific determinants of HRQOL.

**Chapter 5** describes the results of an explorative study on HRQOL and the PAL of patients with MO. The aims of the study were to (1) identify the physical activity level and HRQOL of patients with MO and compare them to reference scores of the healthy population, and (2) determine which illness-related symptoms, sociodemographic or psychological factors are associated with patients' physical activity level, and physical and mental HRQOL. A significantly lower physical activity level and physical HRQOL in patients with MO compared to reference scores of healthy controls was confirmed, but mental HRQOL did not differ. Surprisingly, the physical activity level was not associated with physical HRQOL, opening the debate on whether increasing the physical activity level is important when it comes to physical HRQOL or whether more emphasis should be placed on enabling personally relevant activities. Additionally, a higher educational level was positively related to physical HRQOL which fits the hypothesis that a higher educational level may be related to higher self-efficacy, which in turn is positively related to HRQOL. The negative association of fatigue with both physical and mental HRQOL confirms the hypothesis that fatigue is a prevalent symptom in patients with chronic pain and sometimes even more debilitating than pain. An interesting result was that having a paid job was associated with a higher physical activity level and mental HRQOL and seems to call for increased attention towards employment and vocational rehabilitation.

**Chapter 6** builds further on findings of **chapter 5** to identify transdiagnostic and disease-specific determinants of HRQOL. **Chapter 6** included age, gender, disease duration, pain, fatigue, depression, pain catastrophizing and the physical activity level as possible determinants of HRQOL. Based on the results, fatigue, pain, pain catastrophizing, depressive feelings and the physical activity level can be considered

transdiagnostic determinants of HRQOL, but the magnitude of their association with HRQOL appears to differ depending on the underlying disease. Per unit increase in the physical activity level, the general health subscale increased by 2.4 more in patients with MO compared with patients with CFS. This contributes to the question whether merely increasing the physical activity level leads to a clinically relevant change in HRQOL or whether enabling personally relevant activities might have a more direct positive relationship with HRQOL, especially in patients with CFS. The identified transdiagnostic determinants, i.e. fatigue, pain, pain catastrophizing and depressive feelings, were less severe in patients with MO than in patients with CFS. However, results showed that when patients with MO do experience severe fatigue, pain, pain catastrophizing or depressive feelings, these symptoms are significantly and negatively related to various domains of HRQOL. This advocates for a timely and systematic evaluation of these determinants in clinical practice in patients with chronic fatigue or pain. In addition, exploring whether increasing personally relevant activities improves patients' HRQOL is recommended.

**Chapter 7** contains the general discussion in which the findings are summarized and discussed. Based on the results of this thesis, it has become clear that fatigue and pain are co-occurring symptoms that may not be overlooked in patients presenting with either chronic fatigue or chronic pain. Patients may report significant reductions in their physical activity level, mental and physical HRQOL and several determinants negatively related herewith were identified. With respect to clinical practice, the importance of investigating fatigue, pain, depression, pain catastrophizing and the physical activity level in patients presenting with chronic fatigue or pain is discussed. It is proposed that timely and systematic evaluation of psychological factors may lead to early recognition of developing symptoms, which allows for timely treatment, referral to specialized help and may prevent exacerbation. Also, improvement of mental health literacy may contribute to prevention and early recognition of developing symptoms, which may also lead to better self-management. To improve HRQOL, it is recommended to focus on increasing personally relevant activities and self-efficacy rather than solely on increasing the physical activity level. Implications for research include that psychometric properties of measurement instruments to evaluate activity limitations, participation restrictions or the physical activity level or pattern in patients with CFS are currently considered insufficient and should be examined further. Especially examination of activity monitors in patients with CFS requires urgent attention, because complementary use of a self-reported and objective measurement instrument is recommended but requires reliable and valid measurement instruments. Until then, results should be interpreted and used with caution. The concluding paragraph provides directions for future impact: measuring the physical activity level and HRQOL in clinical practice and related future research needs; opportunities for a preventive approach to prevent psychological symptoms, activity limitations and participation restrictions; and needs regarding the expansion of occupational therapy in primary care to provide appropriate treatment to patients with chronic fatigue and pain.