DETERMINANTS OF PAIN AND DISABILITY IN OSTEOARTHRITIS

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1. INTRODUCTION

Osteoarthritis

Osteoarthritis (OA) is a chronic musculoskeletal disorder, which can affect multiple joints in the human body. The most common joints to be affected are the hip (coxarthrosis) and knee (gonarthrosis) and joints in the hand, especially the first carpometacarpal joint. Osteoarthritis is characterized by a destabilisation of the normal process of simultaneous degradation and synthesis of articular cartilage, chondrocytes, extracellular matrix and subchondral bone (1): progressive loss of articular cartilage is combined with stiffening of the underlying subchondral bone (sclerosis) and development of lateral outgrowths of bone in the joint margins (osteophytes) (2). In later stages of the disease, pathological changes may also take place in other tissues of the joint and its surroundings, such as synovial membrane, capsula, ligaments and muscles, potentially leading to instability of the joint, capsular restriction and muscle atrophy (3,4). These changes result in a reduced capability to carry a load of joints affected by OA, which will often result in functional disability.

Traditionally, osteoarthritis was regarded a consequence of the natural wear and tear of joints due to life-long use. However, in recent years a number of other risk factors for the development and progression of OA beside age have been identified. In a review of other studies, Felson and Zhang (5) identified a number of systemic and local biomechanical factors associated with the pathogenesis of OA. Beside age, the systemic factors included female gender, racial characteristics, genetic suspectibility, high bone density and nutritional factors (especially a lack of vitamin C intake). The biomechanical factors included major joint trauma, prolonged repetitive joint use, obesity, joint deformity and muscle weakness.

As a chronic progressive condition with a multi-factorial etiology, it has proven difficult to clinically define and diagnose the disease. Currently there are two main systems of classifying OA. The system of Kendall and Lawrence is based on radiographically visible changes in the cartilage and subchondral bone (6). Many studies on OA have used their grading scheme, with the definite presence of osteophytes indicating that the joint was indeed affected by OA (2). This has become known as radiological OA (ROA). In more recent years, the American College of Rheumatology has issued clinical criteria for the classification of OA of the knee and hip (7,8). These criteria are given in Table 1. According to the authors, the criteria for knee-OA yield a sensitivity (proportion of true positives) of 0.95, and a specificity (proportion of true negatives) of 0.69 (7), whereas the criteria for hip-OA have a sensitivity of 0.86 and a specificity of 0.75 (8). Although these criteria were first devised in secondary (intramural) health care, they are now commonly used in primary health care as well.

Table 1: Clinical criteria of the ACR for the classification of OA of the knee and hip

Knee	Hip
Knee Pain and at least 3 of the following 6: Age > 50 years Morning stiffness < 30 minutes Crepitus Bony tenderness Bony enlargement No palpable warmth	Hip pain and Hip internal rotation < 15° and ESR ≤ 45 mm/hour (if no ESR available: substitute hip flexion ≤ 115°) OR Hip pain and Hip internal rotation > 15° and Pain on hip internal rotation and Morning stiffness of the hip ≤ 60 minutes and Age > 50 years

ESR = Erythrocyte Sedimentation Rate.

Osteoarthritis is the most common joint disorder in The Netherlands and throughout the western world (2). It has been stated that ROA is present in over 80% of people aged 75+ (2). In a Dutch population study it was reported that in 30% of men and 40% of women over age 65, radiological evidence of knee-OA could be obtained (9). These figures are consistent with other studies, which all report a prevalence of radiographic changes indicating knee-OA of up to 40% in the oldest age groups (2). Hip-OA appears to be less common than knee-OA, with reported prevalence ranging from 7% to 25% for adults aged 55 and over (10). However, for both types of OA the prevalence of symptomatic OA is much lower. It has been stated that 50% of those identified with ROA remain asymptomatic, i.e., do not develop clinical symptoms (predominantly pain and loss of function) attributable to OA (11). Nevertheless, this still means that the estimated prevalence of symptomatic OA in elderly people is about 20% for knee-OA and 10% for hip-OA.

Determinants of pain and disability in knee- and hip-OA

Pain is the predominant symptom in both knee- and hip-OA. This is also reflected in the clinical criteria for diagnosing OA as issued by the American College of Rheumatology (7,8): pain must be present in order to diagnose OA (see Table 1).

A second feature closely associated with OA is functional disability. In the International Classification of Functioning and Disability (ICIDH-2), disability is defined as an umbrella term for problems in body structure or functioning, activities performed by an individual or participation of an individual in his environment (12). In this thesis, functional disability (or physical disability) represents disability associated with basic physical everyday-tasks which involve the lower extremities, such as walking, stair-climbing, reclining and sitting down. OA is widely regarded to be the most common cause of functional disability in Europe and North America (11,13).

It is a common observation that in patients with symptomatic OA the severity of the radiographic changes only plays a minor role in explaining pain and disability (13-15). Other factors have been proposed as determinants of pain and disability in patients with OA. This includes both physical and psychosocial factors.

Muscle weakness has been hypothesized to be an important physical determinant of pain and disability. Several studies have identified a strong link between muscle weakness and pain and disability (13-16). Theoretically, muscle weakness leads to instability of joints, as the weakened muscles become less able to control the joint. This instability may cause strain on tissues in and near the joint, causing pain (14). Also, weakness of muscles in the lower extremities leads to functional disability: the ability of patients to walk, climb stairs etc. will be affected by insufficient strength of leg muscles (17).

A second physical factor that has been proposed as a determinant of disability is joint mobility, although considerably less studies on this topic are available. Odding et al. (18) reported that restricted flexion of the hips and restricted flexion of the knees were strong risk factors for functional disability in a general population of elderly. Two other population studies have reported associations between range of joint motion and functional disability (19,20).

Next to physical factors, psychosocial factors may play a role in determining pain and disability in patients with hip- or knee-OA. More specifically, the way in which patients cope with their condition may have considerable consequences. Lazarus and Folkman defined coping as "efforts to manage specific external and/or internal demands that are appraised as taxing or exceeding the resources of the person" (21). Patients can use different strategies to cope with the pain and discomfort associated with a chronic disease, such as OA. A general distinction can be made between passive and active coping styles. Passive coping styles refer to, often emotional, strategies aimed at avoiding the problem at hand (e.g., avoiding physical activity, catastrophizing, wishful thinking), whereas active coping styles are often more rational and refer to strategies aimed at dealing with the problem at hand (e.g., information seeking, pacing but not ceasing physical activity, transforming pain) (22, 23).

Especially the use of passive coping strategies has been shown to be associated with pain and disability in patients with knee-OA (24, 25). As these were all cross-sectional studies, no causal relationship between the use of passive coping strategies and pain and disability could be established. However, in other groups of chronic pain patients, most notably rheumatoid arthritis and chronic low back pain, longitudinal studies have provided evidence for the role of passive coping strategies as determinants of pain and disability (22, 23, 26). In two of these studies, avoidance of activity was identified as the passive coping style most closely related to an increase in disability (22,23).

The avoidance model

It has been proposed that the effect of avoiding physical activity on disability can be explained using the conceptual framework of the avoidance model (27). As shown in Figure I, according to this theory a patient will initially experience pain during activity. As a consequence, the patient will fear that renewed physical activity will increase the pain, and will therefore start avoiding physical activity. In the short term, pain can be reduced by avoiding physical activity. In the long term, however, low activity levels will result in a deterioration of the patient's physical condition, most notably in muscle weakness. Due to this muscle weakness, the stability of joints will be affected, and the joints' ability to carry a load will be reduced. This results in increased disability. Consequently, the patient will avoid activity even more, thus entering a downward spiral towards increasing physical disability.

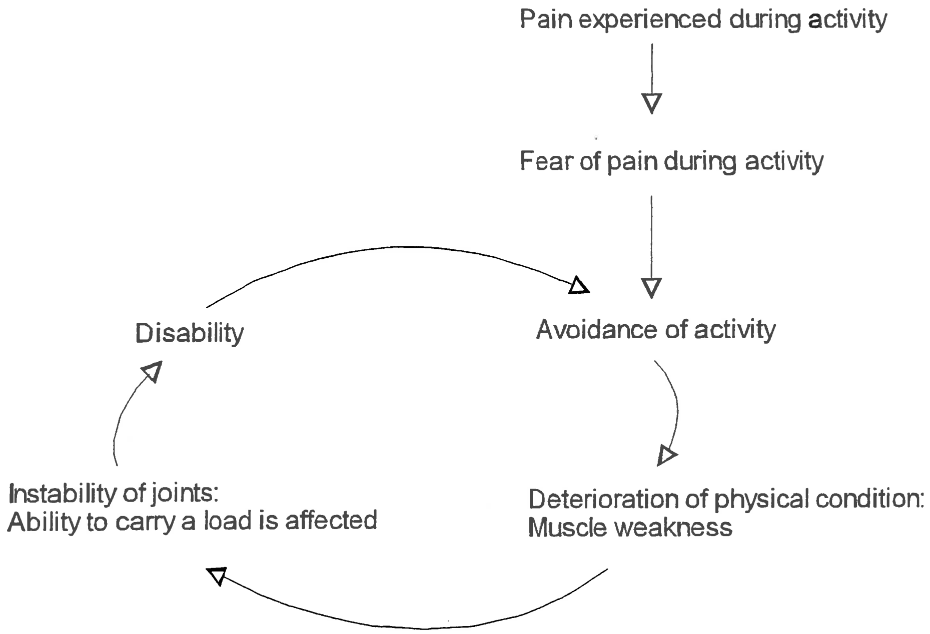

Figure 1: The Avoidance Model

A vital pathway in this model is the way in which avoidance leads to disability: first avoidance of activity leads to muscle weakness. Because of the weakness of the surrounding muscles, the capability of joints to carry a load is reduced. This then leads to functional disability. According to the model, muscle weakness therefore acts as a mediator of the relationship between avoidance of activity and functional disability. Formally, a mediator is defined as a factor which "represents the generative mechanism through which the focal independent variable is able to influence the dependent variable of interest" (28). Thus, in the avoidance model, muscle strength is the mediator between avoidance of activity and functional disability.

The aim of this thesis is to establish the validity of this pathway in the avoidance model. However, some other issues need to be addressed before the avoidance model can be studied in depth. A key issue is the way in which factors present in the model are assessed. This applies in particular to the assessment of disability and muscle strength.

The assessment of disability and muscle strength

Physical disability is normally assessed in one of two ways: using an observational method or by means of a self-report method (questionnaire). When using an observational method, a patient is asked to perform a number of tasks related to physical disability (e.g., walking, stair climbing, reclining). The performance is rated by an observer, normally an experienced medical professional (e.g., a physical therapist). The observer's rating is used as a score for disability. The second way to assess disability is by presenting a questionnaire to the patient himself. By filling out this questionnaire, the patient gives his own assessment of his level of disability.

Theoretically, observational methods give insight into a patient's actual level of functioning, since they are not contaminated by the views of the patient concerning his functional disability. On the other hand, the patient's own view is normally of considerable interest. Self-report methods do reflect these views. Additionally, self-report methods may be better at establishing a patient's functional capabilities in his own daily living environment than observational methods, which normally take place in laboratories. However, it has been stated that psychological factors heavily affect a patient's perception of symptoms of his disease (29). This would impede attempts to establish the level of disability by self-report.

Given these differences between observational and self-report methods, it has been argued that these two types of instruments measure two different dimensions of disability (30, 31). So far this claim has not been substantiated, however.

Next to disability, questions can also be asked concerning the assessment of muscle strength. A number of muscle actions around the knee and hip (e.g., flexion of the knee, internal rotation of the hip) are involved in daily activities such as walking. Assessing all of these muscle actions leads to a considerable amount of available data. The question arises how these data on muscle strength should be treated in statistical analyses. On the one hand, a parsimonious approach can be chosen. Using one or a few aggregated indicators for muscle strength means analyses are more transparent, and results are more easily interpreted. On the other hand, a parsimonious approach is only justified if the strength of different muscle actions is similarly related to other variables of interest, such as disability. If different muscle actions are differently related to disability, the use of a single indicator would lead to loss of vital information. It is therefore important to find the optimal trade-off between parsimony (using as few separate indicators for muscle strength as possible) and content (no loss of vital information on muscle strength).

Aim and scope of this thesis

The aim of this thesis is to establish the validity of the pathway in the avoidance model in which avoidance of activity leads to functional disability through muscle weakness, in patients with OA of the hip or knee. To this aim, a number of separate research questions will be addressed. The first research question addresses the issue of how to assess disability. The second question deals with the reduction of muscle strength data into indices for muscle strength.

A similar topic is raised in the third research question of this thesis. This question addresses the role of joint mobility as a determinant of disability in OA. Before the relationship between joint mobility and disability can be studied, it first needs to be established to which degree data on range of motion of different joint actions can be reduced into a limited number of indicators for joint mobility. Although joint mobility is not a factor in the avoidance model, restricted range of joint motion is regarded an important determinant of disability in patients with OA. This relationship has rarely been studied in depth, however. Therefore, a study on range of joint motion and its role as a determinant of disability has been included in this thesis.

The fourth research question concerns the role of different coping styles, most notably avoidance of activity, as a determinant of pain and disability in patients with OA. The fifth research question focuses on the main theme of this thesis: the mechanism through which avoidance of activity acts as a determinant of disability.

These five research questions are listed below.

1. *When assessing functional disability in patients with knee-OA or hip-OA, is there a difference in information obtained by observational methods on the one hand and self-report methods (questionnaires) on the other hand?*

It has been stated that observational methods and questionnaires assess different dimensions of disability. This assumption will be tested in both a cross-sectional and a longitudinal design. To address this question, the internal consistency and construct validity (in a cross-sectional design) and responsiveness (in a longitudinal design) of an observational method for the assessment of physical disability will be established. To establish the validity and responsiveness of the observational method, the information provided by the observational method will be compared with the results of a number of questionnaires.

2. *Is it possible to adequately describe the level of muscle strength of a patient with OA of the knee or hip using a single indicator or should a distinction between different (subsets of) muscle actions be maintained?*

This second question deals with the use of muscle strength data in statistical analyses. It is unclear whether muscle strength can be represented by a single index at the level of the patient (i.e., all muscle actions can be aggregated into a single indicator for a patient's level of muscle strength), or whether the strength of different muscle actions should be seen as separate entities, with their own specific impact on a patient's level of functioning. The optimal trade-off between parsimony (as few separate indicators as possible) and detail (no loss of vital information) needs to be established.

3. *Is impaired range of joint motion a determinant of physical disability in patients with OA of the hip or knee?*

Next to muscle weakness, impaired range of joint motion is regarded the second important physical determinant of disability in patients with OA. A limited number of studies on the relationship between range of joint motion and disability have been executed. None of these studies specifically involved a population of OA-patients (18-20). Because of this lack of specific research, a study on the relationship between impaired range of motion and disability is included in this thesis. In this study, first the inter-relationships between the various joint actions of the knee and hip will be established, analogous to the research question #2 concerning muscle strength. For joint mobility, like muscle strength, the question can be asked whether range of motion of a specific joint action can be regarded a representation of a general physical characteristic of the patient (his overall sinuosity), or whether the range of motion of a single joint action, or subsets of joint actions, should be treated as separate entities.

After establishing the inter-relationships of the range of motion of different joint actions, the role of range of joint motion as a determinant of disability in patients with OA can then be determined.

4. *Is the passive coping style of avoidance of physical activity a determinant of pain and disability in patients with OA of the knee or hip?*

Avoidance of activity has been identified as a determinant of pain and physical disability in different groups of chronic pain patients, such as rheumatoid arthritis (22, 23) and low back pain (26). It is expected that these relationships can also be established in patients with OA of the knee or hip. Two studies identified avoidance of activity as the most important one among the different coping styles in explaining disability (22, 23). To assess the relative importance of avoidance of activity as a determinant of pain and disability, other coping styles will also be included in this study. As its aim is to establish a causal relationship between the use of coping styles and pain and disability, these analyses will be performed using a longitudinal design.

5. *If avoidance of activity is a determinant of disability in patients with OA of the hip or knee, is this relationship mediated by muscle weakness?*

This is the main research question of this thesis, as it addresses the core of the avoidance model. According to the avoidance model, avoidance of activity leads to muscle weakness, which in turn leads to an increase in physical disability. This means that, theoretically, muscle weakness acts as a mediator in the relationship between avoidance of activity and disability. The mediating role of muscle weakness in this relationship will be assessed using both cross-sectional and longitudinal analyses.

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2. INTERNAL CONSISTENCY AND VALIDITY OF AN OBSERVATIONAL METHOD FOR ASSESSING DISABILITY IN MOBILITY IN PATIENTS WITH OSTEOARTHRITIS

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ABSTRACT

Objective: To establish the internal consistency and validity of an observational method for assessing disability in mobility in patients with osteoarthritis (OA).

Methods: Data were obtained from 198 patients with OA of the hip or knee. Results of the observational method were compared with results of self-report methods (questionnaires) on disability in mobility.

Results: Both Cronbach's α and Mokken Scale Analysis indicate that the method is internally consistent. Using factor-analysis, observed and self-reported disability in mobility were found to be closely associated, and could not be differentiated.

Conclusions: The observational method is internally consistent and it indeed measures disability in mobility (high convergent validity), but observations and self-report seem to yield largely equivalent information (low divergent validity). This raises questions regarding the simultaneous use of both observational and self-report methods in the assessment of disability in mobility in OA patients.

Key words: osteoarthritis, disability, mobility, assessment

Osteoarthritis (OA) of the hip or knee can be a severely disabling condition. Especially problems concerning patients' mobility are frequently reported (1-4). These problems concern walking, rising and sitting down and getting into and out of bed. Disability in mobility can be assessed by both observational and self-report methods (questionnaires). Both kinds of methods have their own specific advantages and disadvantages. Observational methods provide information about a patient's actual state of disability. They have the advantage of not being influenced by subjective factors, such as a patient's expectations and beliefs. On the other hand, they yield no information on the patient's own point of view concerning his situation. Self-report methods reflect the patient's opinion, which frequently is of essential interest. Also, self-report methods may be better suited to evaluate a person's functional capability in his own environment. However, information obtained by self-report methods could be highly influenced by subjective factors. Watson and Pennebaker (5) argued that psychological factors heavily affect the way in which patients perceive symptoms of their disease. This impedes attempts to assess the actual level of functioning of patients. Therefore, it has been argued that observational and self-report methods measure two different dimensions of disability (6-7).

The aim of this study was to examine the clinimetrical quality of an observational method for the assessment of disability in mobility in OA patients. This instrument is an adaptation of the method first described by Keefe and Block (8-9). In this method, a patient is instructed to perform a series of standardized tasks. These tasks include walking, sitting down, bending and reclining. The performance of these tasks is recorded on videotape. The videotape is watched by an observer, who scores a number of items related to mobility. In its original form, this method was developed to assess pain behaviour (8). Next to items related to mobility, it also comprised a number of items assessing non-verbal expression of pain. This last group of items has been left out of the present method. In the present form, only items assessing disability in mobility are included. Earlier reports on this method have shown that the inter-observer reliability is good (8, 10)

In the present study the internal consistency of the method is established, i.e. the assumption that the method is unidimensional is tested. A second objective of the present study was to determine the validity of the test. It was expected that the results of the observational method would correlate - to a certain extent - with questionnaires measuring disability in mobility. This would establish the concurrent or convergent validity of the observational method. But it was also expected that the questionnaires would be more closely correlated to each other than to the observational method. This would indicate that observational and self-report methods indeed measure different aspects of disability in mobility. These findings would establish the divergent validity of the observational method. Divergent validity is the degree to which a test yields information that is not picked up by other methods. Finally, it was expected that observational assessment of disability in mobility would not be correlated with disability in activities involving the upper extremities, such as washing or dressing ('self care'). This would establish another aspect of the divergent validity of the observational method.

METHODS

Subjects

Data were obtained from a randomized clinical trial into the effectiveness of exercise therapy in patients with osteoarthritis of the hip or knee (11). Patients were included if they were diagnosed to have OA of the knee or hip according to the classification criteria of the American College of Rheumatology (12-13). 200 Patients participated in the trial. Data for the present study were obtained at the onset of the trial (baseline). Of two patients data on one or more of the tests presented below were incomplete. These patients' data were excluded from the analyses, resulting in 198 patients in the analyses.

Observed disability in mobility

Observational assessments of disability in mobility were made by watching videotape recordings of patients' performance on a series of standardized tasks, using an adaptation of the method described by Keefe and Block (8). These tasks were: walking: walking up and down in a straight line directly in front of the camera; sitting down: seating oneself in a chair, height 45 cm, with arm-rests, height 67 cm; reclining: lying down on a bench, height 53 cm; bending: lifting a 2 kg weight off the ground, coming to a full erect position, putting the weight back in its original place on the ground.

Apart from bending, these tasks were performed for one minute. This meant that subjects were asked to walk for one minute, or to remain in a seated or reclined position after conclusion of the task until the end of the minute. The bending task was finished when the patient stood fully erect after putting the weight back in its original position. The videotaped recordings of the performance of the tasks were watched and scored for the following four items:

- Guarding: abnormally slow, stiff, interrupted or rigid movement while walking (9). Possible score range for this item was 0-2.
- Rigidity: excessive stiffness of the affected joint or extremity while bending, reclining or sitting down (9). Possible score range for this item was 0-5.

- Stand-to-sit time. The time (in seconds) it takes a subject to change from a standing to a seated position. Timing began when a patient crossed a line 50 cm in front of the chair and ended when his back touched the back of the chair, or when the patient sat at ease without showing any intention to move further backwards.
- Stand-to-recline time: the time (in seconds) it takes a subject to change from a standing to a reclined position. Timing began when a patient crossed a line 50 cm in front of the bench, and was ended when his head touched the pillow.

All items were scored by trained observers. They watched the first twenty seconds of the task, and then had ten seconds to score either 'yes' or 'no' for guarding and rigidity (presence or absence of guarding or rigidity), after which they watched another twenty seconds of the task and had again ten seconds to score. 'Guarding' was not scored when the subject had to turn during the walking-task. Bending was scored once, after conclusion of the task. In addition, they scored the stand-to-sit time and the stand-to-recline time.

A fifth item was scored independently from the videotaped performances described above. This was the 5 m-walking time: the time (in seconds) it takes a subject to walk five meters. Because of the distance involved, this task was not videotaped, but scored directly while the patient was performing the task. In order to minimize the influence of acceleration and deceleration at the onset and conclusion of the task, subjects walked a total of 8 meters, the 5-m walking time was scored during the middle five meters.

An overall score for observed disability in mobility was computed, taking into account guarding, rigidity, stand-to-sit time, stand-to-recline time and 5-m walking time. In order to correct for skewed distributions, scores on the three movement times were ranked into ten categories per item. After that, scores on all items were standardized (z-scores) to exclude weighting problems due to different score ranges. These standardized item-scores were then added up to obtain the overall score.

Self-reported disability in mobility

To assess self-reported disability in mobility, five subscales of four questionnaires were used. These included:

- IRGL mobility subscale. The IRGL (Influence of Rheumatic Disease on General Health and Lifestyle) (14) is a Dutch adaptation of the Arthritis Impact Measurement Scales (AIMS) (15). This subscale has seven items. Two items are general statements ("Because of my health I spent most of the day indoors" and "Because of my health, during the daytime I spent most of the time in a chair"), while the other five address disability in going upstairs, riding a bicycle and walking (such as 'I was able to walk for 30 to 60 minutes'). All items have a score range from 1 ('hardly ever') to 4 ('nearly all the time'). The IRGL is a 'positive' questionnaire, i.e. it measures 'ability' rather than 'disability'. To facilitate interpretation, scores on this test have been reversed to obtain a 'disability-score'.

- Nottingham Health Profile mobility subscale. This subscale consists of eight statements concerning problems with mobility (e.g., 'I find it hard to bend', 'I'm unable to walk at all'), to which a patient can answer 'yes' or 'no'. Because of differences in the severity of the statements, they are weighted. This leads to a score range of 0-100, in which '100' represents an affirmative response to each statement (16).
- EuroQoL mobility subscale. This scale considers health status in the previous week. The mobility subscale consists of one item. This item has three statements of which patients must pick one that describes their situation during the previous week. These statements are: no problems walking about; unable to walk about without a stick, crutch or walking frame; confined to bed. A patient's test score equals 1 if he picks the first statement, 2 if he picks the second and 3 if the third is chosen. (17-18).
- The QR&S (Questionnaire Rising and Sitting down) (19). This questionnaire consists of two subscales. One subscale ("QR&S high chair") focuses on rising from and sitting down on high chairs (including the lavatory and sitting on a bed), while the other ("QR&S low chair") focusses on rising from and sitting down on low chairs, such as easy chairs, carseats and sofas. The questionnaire consists of statements, which can be answered affirmatively. 14 statements form the "high chair" subscale (7 statements on rising, 6 on sitting down, 1 on being seated), and 9 statements form the "low chair" subscale (4 statements on rising and 5 on sitting down).

Self-reported disability in self care

Two questionnaires were used to assess subjects' level of self care. These questionnaires focus on activities related to the upper extremities, such as dressing or washing. These were:

- IRGL dexterity subscale (14). This subscale consists of eight items, such as 'I was able to button up my blouse/shirt', 'I was able to lace up my shoes'. Equivalent to the IRGL mobility subscale, itemscores ranged from 1 ('hardly ever') to 4 ('nearly all the time'). To facilitate interpretation, its scores have been reversed.
- EuroQoL self care subscale. Equivalent to the mobility subscale, the self care subscale has one item with three statements. These are: no problems with self care; unable to dress self; unable to feed self. A patient's test score equals 1 if

he picks the first statement, 2 if he picks the second and 3 if the third is chosen. (18).

Statistical analyses

The internal consistency of the observational method was assessed by computing Cronbach's alpha, and by performing Mokken Scale Analysis. Mokken Scale Analysis is a non-parametric method for assessing the scalability of a set of polytomous items (20-21). It computes a scalability coefficient (Loevinger's H, a value of 0.5 or higher represents 'good' scalability), a reliability coefficient (Rho, a value of at least 0.75 is required) and a hierarchical order for the level of difficulty of the items. This means that a patient with a given score on a specific item is expected to score higher on any item lower in the order. All items entering the Mokken procedure must have the same score range. Therefore, in the Mokken analysis only, the raw item scores were transformed. 'Rigidity' was chosen as the 'base-item'. This item had a possible score range of 0-5. The scores on the three movement times were transformed into six categories to obtain the same range. 'Guarding'-scores originally ranged from 0 to 2. These scores were multiplied by 2 to obtain the possible scores of 0, 2 and 4. These scores were considered to fit into a 0-5 range.

Spearman's rank correlation coefficients were computed to test relationships between methods. To minimize the chance of making Type I-errors, the significance level for individual coefficients was set at $\alpha=0.001$. Correlations of approximately 0.40 to 0.60 were expected between observed disability in mobility and the various self reported disability in mobility questionnaires.

Using the Spearman rank correlation coefficients, oblique factor-analysis was performed to test the assumption that observed mobility, self-reported mobility and self-reported self care address three separate aspects of disability. Oblique factor-analysis allows for correlations between factors. Since a relationship between observed and self-reported disability in mobility was expected, this was the best suitable form of factor-analysis.

All statistical analyses were carried out using SPSS/PC+ version 5.0, except for the Mokken analysis, which was performed in MSP, version 3.0 . MSP is a computer program specifically designed to perform Mokken analysis.

RESULTS

Patient characteristics

Mean age, proportion of males and females and location of OA are given in Table 1. This table also features mean raw data scores on the items of the observational method, mean sum score on the observational method and mean scores on all questionnaires. All scores are 'disability scores', i.e. a higher score means a higher level of disability.

Internal consistency

The analyses show that the observational method is internally consistent. Cronbach's alpha equals 0.84. The results of the Mokken Scale Analysis are given in Table 2. The scalabilty coefficient (Loevinger's H) has a value of more than 0.5, which represents 'good scalability'. In Table 2 the items are ordered hierarchically according to their means. According to the hierarchical order, 'rigidity' is the 'easiest' item. This means that patients show relatively few signs of stiffness . Patients score lower on 'rigidity' (showing no stiffness) than on the three movement tasks.

Correlations

Spearman rank correlation coefficients between methods are given in Table 3. Significant relationships were found between observed disability in mobility and all five questionnaires on self-reported disability in mobility. However, the correlation between observed disability in mobility and the EuroQoL mobility subscale is much lower than that between observed disability in mobility and the other four questionnaires on disability in mobility. This pattern is also visible among the mobility questionnaires themselves. The NHP, IRGL and QR&S subscales all correlate with each other, but show a much less distinct correlation with the EuroQoL mobility subscale. Observed mobility also correlates with the IRGL dexterity subscale, but not with the EuroQoL self care subscale.

Factor-analysis

The oblique factor-analysis of the observed disability in mobility sum-score, the five mobility questionnaires and the two self care questionnaires produced two factors instead of the assumed three. Total proportion of variance accounted for was 57.8%. Factor 1 accounted for 44.6% of variance, factor 2 accounted for the remaining 13.2%. The factor loadings in Table 4 show that the first factor comprises both the observational and self-report methods concerning disability in mobility. The second factor is loaded by the two self care questionnaires. The two factors can therefore be regarded as a 'mobility-factor' and a 'self care-factor'. It is also notable that the EuroQoL mobility subscale hardly correlates with any of the two factors.

A number of complementary factor-analyses have been performed (results not presented here). These included subgroup-analyses by gender and involving only patients suffering from hip-OA, or patients suffering from knee-OA. Also, analyses were performed excluding the two questionnaires addressing disability in self care. Finally, an analysis was performed, including all questionnaires and the observational method, with a mandatory three factor-solution. The results of all of these analyses were consistent with the factor structure presented here. Analysing on subgroups based on gender or type of arthritis did not lead to different results than presented here.

Table 1: Patient Characteristics (N=198)

	mean	±	sd	range	n	%
Gender						
Male					43	21.7
Female					155	78.3
Osteoarthritis of						
Hip					69	34.8
Knee					119	60.1
Both					10	5.1
Age	68.0	±	8.9	39 - 84		
Disability in Mobility: Observations						
Sum Score	0.0	±	3.9	-6.2 - 13.4		
1. 5 m-walking time (s)	5.3	±	1.7	2.9 - 18.6		
2. sit time (s)	3.6	±	1.1	1.9 - 8.0		
3. recline time (s)	7.2	±	2.9	2.5 - 19.4		
4. guarding	1.1	±	1.0	0 - 2		
5. rigidity	1.0	±	1.1	0 - 4		
Disability in Mobility: Self-report						
IRGL Mobility	-20.3	±	5.6	-28 - -7		
NHP Mobility	27.4	±	16.8	0 - 87.4		
EuroQoL Mobility	1.8	±	0.4	1 - 2		
QR&S "high chair"	2.8	±	2.6	0 - 10		
QR&S "low chair"	5.2	±	3.2	0 - 10		
Disability in Self Care: Self-report						
IRGL Dexterity	-29.7	±	3.6	-32 - -10		
EuroQoL Self Care	1.1	±	0.3	1 - 2		

Table 2: Mokken Scale Analysis of the items of the observational method

Item	Mean Score	Scalability coefficient (H)
1. Rigidity	1.00	0.56
2. Guarding	2.22	0.51
3. 5 m-walking time	2.48	0.53
4. Stand-to-Recline time	2.50	0.55
5. Stand-to-sit time	2.54	0.49
Scale		0.53
Reliability coefficient: Rho = 0.82		

Table 3: Correlations

	Disability in mobility: observations	Disability in mobility: self-report			disability in self care: self-report			
		NHP mobility	EuroQoL mobility	IRGL mobility	QR&S high chair	QR&S low chair	EuroQoL self-care	IRGL dexterity
Disability in mobility: observations								
Disability in mobility: self-report								
NHP mobility	.55*							
EuroQoL mobility	.29*	.38*						
IRGL mobility	.54*	.58*	.21					
QR&S high chair	.51*	.57*	.18	.51*				
QR&S low chair	.54*	.54*	.21	.51*	.78*			
Disability in self-care: self-report								
EuroQoL self-care	.13	.28*	.08	.10	.20	.12		
IRGL dexterity	.42*	.41*	.25*	.47*	.34*	.36*	.17	

(* : $p < .001$)

Table 4: Factor-loadings of the factor-analysis

	Factor 1	Factor 2
Disability in Mobility: Observations	.79	-.01
Disability in Mobility: Self-report		
IRGL Mobility	.75	.06
EuroQoL Mobility	.31	.21
NHP Mobility	.72	.21
QR&S "high chair"	.84	.06
QR&S "low chair"	.89	.18
Disability in Self Care: Self-report		
IRGL Dexterity	.11	.67
EuroQoL Self Care	-.09	.84

DISCUSSION

The aim of the present study was to establish the consistency and validity of an observational method for assessment of disability in mobility in patients with osteoarthritis of the hip or knee. Four assumptions concerning the clinimetrical quality of the method were tested.

The method is internally consistent, i.e. it has a unidimensional construct (first assumption). Both Cronbach's alpha and the reliability coefficient Rho had values well above the minimum required for consistent methods. Also, Loevinger's H was above 0.5, which represents good scalability. To our knowledge, the internal consistency of this method had not been established before.

The observational method correlates significantly with all questionnaires addressing disability in mobility. It can therefore be concluded that the observational method is convergently valid (second assumption). The existence of a close relationship between self-report and observational methods is consistent with the findings of earlier research (7,22-23). It is, however, notable that the correlations between observed and self-reported disability in mobility do not exceed 0.60. A possible explanation for this is the imperfect overlap between the items of the self-report methods on the one hand and the observational method on the other hand.

A total of three factors was expected to be present among the methods: a factor addressing observed disability in mobility, a factor concerning self-reported disability in mobility and a factor addressing (self-reported) disability in self-care. It was expected that these three factors would be related to each other. This was accounted for by using oblique factor-analysis. The factor-analysis revealed the existence of only two factors. One factor comprised observed and self-reported disability in mobility. The other factor comprised self-reported disability in self care. These results show that there is a clear differentiation between methods that address disability in activities related to the upper extremities ('self care') and activities related to the lower extremities ('mobility'). This partly establishes the divergent validity of the observational method: it yields information on mobility, which is different from information on self-care gathered by questionnaires (fourth assumption). However, the observational method and questionnaires on disability in mobility do not exhibit enough contrast to separate them into two different factors. Apparently, the assumption that observed and self-report methods address two different aspects of disability in mobility is not supported (third assumption). This finding is consistent with those of Hidding et al (24), who found that 'mode of assessment' (self-report or observations) is hardly a source of variability in functional disability scores in patients with Ankylosing Spondylitis or Rheumatoid Arthritis. On the other hand, it has also been argued that observations and self-reports are complementary to each other. According to Guralnik et al. (23) observations yield information that cannot be obtained by self-reports. The results of the present study do not support this. The lack of correlations between observations and self-reports in excess of 0.60 can be attributed to imperfect overlap between the items of both kinds of methods, and errors of measurement in general.

This gives rise to questions on the good of using both observational and self-report measures when assessing disability in mobility. In the present study, these methods were found to yield largely the same information. Of course, replication of this finding is called for, in both OA-patients and other groups of patients. Also, other methods (both observational and self-report) than the ones chosen in the present study, with their own psychometrical attributes, could show a different relationship. In addition, there remains a possibility that observed and self-reported disability in mobility are differentially related to other variables. For example, subjective factors, such as patients' expectations, could be expected to be more closely related to self-report than to observation (5). Also, in certain subgroups of patients there may be substantial differences in the outcome of observational assessment and assessment by self-report (23). Similarly, interventions may differentially affect observations and self-report (25). In the present study, the responsiveness (sensitivity to change, specificity to change) and predictive validity of the instruments have not been established. Whether self-report and observational methods differ in those respects needs to be established in future research. It has been stated that observations are a better predictor of mortality and nursing home admission than self-reports (23). Nevertheless, the present cross-sectional results do not support the good of using both observational and self-report measures for the assessment of disability in OA-patients.

Questions can be raised regarding the validity of the EuroQoL scale. The mobility subscale shows few significant relationships with other questionnaires on disability in mobility. Its correlation with the observational method is also considerably lower than that of the other disability in mobility questionnaires. This can probably be attributed to the single question format of the subscales of the EuroQoL questionnaire.

In summary, in this cross-sectional study the observational method has been shown to be internally consistent. Observed and self-reported disability were found to be associated. Therefore, the observational method does indeed seem to measure disability in mobility (high convergent validity), but observation and self-report seem to yield largely equivalent information (low divergent validity). This result raises questions regarding the simultaneous use of both observation and self-report in the assessment of disability in mobility in patients with OA of the hip or knee.

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3. RESPONSIVENESS OF OBSERVATIONAL AND SELF-REPORT METHODS FOR ASSESSING DISABILITY IN MOBILITY IN PATIENTS WITH OSTEOARTHRITIS

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ABSTRACT

Objectives: To establish the responsiveness of observational and self-report methods for the assessment of disability in mobility.

Methods: Data of 186 patients with hip-OA or knee-OA were used. Data from one observational method and four self-report methods for the assessment of disability in mobility were collected in week 0 and 12 weeks later. Using correlations and factor analysis, the relationship among changes in these five methods was established.

Results: Inter-correlations between change scores of the self-report methods ranged from 0.12 to 0.34. Correlations between the observational method on the one hand and self-report methods on the other hand ranged from 0.14 to 0.26. In the factor analysis, both self-report methods and the observational method loaded on the same factor.

Conclusion: In a longitudinal design, no evidence for the differential responsiveness of observational and self-report methods was obtained. Due to the advantages of questionnaires (easier to use, less time-consuming, less of a burden to subjects), this implies that the use of self-report methods is to be prefered over observational methods.

Key Words: Osteoarthritis, Disability, Responsiveness

Osteoarthritis (OA) of the knee or hip is associated with a substantial decrease in the ability to carry a load of the affected joints (1,2), which results in disability in mobility (3-5). The level of disability in mobility can be assessed by either observational methods (e.g., the 50-feet walking time) or self-report methods (questionnaires). These two kinds of methods have been hypothesized to measure different dimensions of disability (6-9). Theoretically, observations made by an expert therapist or researcher give insight into a patient's actual state of disability, without being influenced by subjective factors, such as a patient's beliefs. On the other hand, observations do not take a patient's own point of view into account concerning the level of his functional ability. Self-report methods do reflect the patient's opinion , which frequently is of essential interest. However, it has been argued that psychological factors heavily affect the way in which patients perceive symptoms of their disease, which impedes attempts to assess the actual level of functioning of patients (10).

So far conclusive evidence that questionnaires and observational methods indeed provide different information has been scarce (11-13). Even when it was specifically stated that questionnaires and observational methods differ in this respect, this claim was not conclusively supported by the results (6-9). Consequently, the recommendations of OMERACT on which instruments to use in osteoarthritis clinical trials aim at using self-report methods, and question the use of observations (14). On the other hand, in a randomized clinical trial into the effectiveness of exercise therapy on osteoarthritis, an effect of the intervention was found on observed but not self-reported disability (15). This result suggests that observational and self-report methods do measure different aspects of disability in mobility.

Most studies which failed to find a difference in information provided by self-report methods and observational methods had a cross-sectional design (6-9, 11-13), whereas the trial which yielded different results for self-report and observational methods had a longitudinal design (15). Therefore, it is possible that the main difference between questionnaires and observational methods lies in their ability to detect relevant changes over a period of time, or responsiveness (16, 17).

In the present study the responsiveness of observational and self-report methods for assessing disability in mobility is determined. Various approaches towards the assessment of responsiveness are available. Ideally, responsiveness is assessed by comparing the results of the test of interest to the results of an instrument known to be responsive, i.e. the Gold Standard (16). In the present study, however, such an approach is not viable, because there is no Gold Standard for disability in mobility available. In the absence of a “true” Gold Standard for disability in mobility, the change in global health status as perceived by the patient himself (“global perceived effect”) is frequently used as gold standard. Recently, the latter approach has been severely criticized, mainly because global perceived effect and similar measures may be affected by other changes in health status and subjective bias, and may not be reliable (18). An alternative way of assessing responsiveness is to study longitudinal construct validity (19). In this approach, the longitudinal convergent validity (i.e., changes in scores of similar instruments are expected to correlate with each other) and the longitudinal divergent validity (i.e., changes in scores of dissimilar instruments are expected not to correlate with each other) are established. This means that it is expected that scores of similar instruments will demonstrate similar change over time, whereas scores of dissimilar instruments will display differential change over time.

The aim of the present study was to establish the responsiveness of observational and self-report methods for assessing disability in mobility, by studying their longitudinal convergent and divergent validity. One observational and four self-report methods were used. It was expected that, in a longitudinal design, (i) changes in scores on the self-report measures would be mutually correlated and that (ii) changes in the score on the observational method would not be associated with changes on self-report methods.

METHODS

Subjects

Data were obtained from a randomized clinical trial into the effectiveness of exercise therapy in patients with osteoarthritis of the hip or knee (15) Patients were included in the trial if they were diagnosed to have OA of the knee or hip according to the clinical classification criteria of the American College of Rheumatology (20,21). 200 Patients participated in the trial. Of 14 patients data on one or more of the instruments presented in this study were missing. These patients' data were excluded from the analyses. This resulted in 186 patients in the analyses, including patients from both the experimental and control group of the trial.

During the trial, the level of disability in these patients was assessed at regular intervals. For the present study, data were used from the tests at the onset of the trial (baseline, week 0), and immediately after the conclusion of the intervention period, at week 12. Due to the intervention period between these two measurements, it was expected that the change in disability in these patients would be most pronounced between week 0 and week 12.

Observed disability

Observational assessments of disability in mobility were made by watching video-taped recordings of patients' performance on a series of standardized tasks (12). These tasks included walking, sitting down, reclining, and bending. Five items were scored to assess the level of disability: three movement times (walking time, stand-to-sit time and stand-to-recline time) and two items rated by the observer (the levels of rigidity and caution, or guarding, during the execution of a task; both were rated on a 0-5 scale). All items were scored by *two* trained observers. For a more detailed description of this observational method we refer to Steultjens et al. (12).

An overall score for observed disability in mobility was computed, taking into account guarding, rigidity, stand-to-sit time, stand-to-recline time and 5-m walking time. In order to correct for skewed distributions, scores on the three movement times were ranked into ten categories per item. After that, scores on all items were standardized (z-scores) to exclude weighting problems due to different score ranges. These standardized item-scores were then added up to obtain the overall score. This overall score has been shown to be internally consistent and to have good cross-sectional convergent validity (12). Both the intra-rater reliability and inter-rater reliability were established in the same group of patients featured in the present study. The Pearson correlation coefficients equalled 0.90 and 0.78 for intra- and inter-rater reliability, respectively.

To be able to identify changes in week 12 (compared to week 0), the z-scores for the data of week 12 were computed as if they were taken at baseline (i.e., mean and standard deviation of the scores at baseline were used in the computation of z-scores for data from week12).

Self-reported disability

To assess self-reported disability in mobility, four subscales of three questionnaires were used. These included:

- IRGL mobility subscale. The IRGL (Influence of Rheumatic Disease on General Health and Lifestyle; 22,23) is a Dutch adaptation of the Arthritis Impact Measurement Scales (AIMS) (24). This subscale has seven items. Two items are general statements ("Because of my health I spent most of the day indoors" and "Because of my health, during the daytime I spent most of the time in a chair"), while the other five address disability in going upstairs, riding a bicycle and walking (such as 'I was able to walk for 30 to 60 minutes'). All items have a score range from 1 ('hardly ever') to 4 ('nearly all the time'). The IRGL is a 'positive' questionnaire, i.e. it measures 'ability' rather than disability. To facilitate interpretation, scores on this test have been reversed to obtain a 'disability-score'.

- Nottingham Health Profile mobility subscale. This subscale consists of eight statements concerning problems with mobility (e.g., 'I find it hard to bend', 'I'm unable to walk at all'), to which a patient can answer 'yes' or 'no'. Because of differences in the severity of the statements, they are weighted. This leads to a score range of 0-100, in which '100' represents an affirmative response to each statement (25).
- The QR&S (Questionnaire Rising and Sitting down) (26). This questionnaire consists of two subscales. One subscale ("QR&S high chair") focuses on rising from and sitting down on high chairs (including the lavatory and sitting on a bed), while the other ("QR&S low chair") focuses on rising from and sitting down on low chairs, such as easy chairs, carseats and sofas. The questionnaire consists of statements, which can be answered affirmatively. For both subscales of the QR&S, the scores are converted to fit into a 0-10 range.

In order to make the scores of the four questionnaires directly comparable to each other and the observational method, the sum scores of the questionnaires were standardized (z-scores), in the same manner as the observational method.

Statistical analyses

For all five tests of interest, a change score was computed. This was done by subtracting the baseline score from the score at week 12 (change score = score(week 12) - score(week 0)). This means that a negative change score signifies a lower level of disability (i.e., improvement) at week12 compared to the baseline-level of disability.

Using the standardised change scores of the observational method and the four questionnaires, Spearman rank correlation coefficients were computed to test the longitudinal relationships between the methods. Using the Spearman correlation coefficients, oblique factor analysis was performed on the change scores of the five tests of interest. Given the assumption that observations and self-reports address different aspects of disability, two factors were expected to be present: one comprising the observational method, and one primarily loaded by the four questionnaires. Oblique factor-analysis allows for correlations between factors. Since a relationship between observed and self-reported disability in mobility was expected, this was the best suitable form of factor-analysis.

RESULTS

Table 1 features proportion of males and females and proportion of knee- and hip-OA in the study population, and mean age of the study population. In Table 2, the baseline scores, scores at week 12 and change scores for the observational method and the self-report methods are given in this table. For the four questionnaires, first the usual change score is given, and then the standardized change score. The standardized change scores of the four questionnaires and the observational method are directly comparable to each other. The average standardized change of the observational method and two questionnaires, the NHP and IRGL, are similar to each other. The average standardized change of the two subscales of the QR&S is lower than the change of the other instruments. However, the variability in the standardized change scores is similar for all instruments. There is little difference in the standard deviation of the change scores of the five methods. For all methods, the average change is not very large. However, there is substantial variation in the level of change between patients, which is a vital requisite when studying responsiveness.

Table 1: Patient characteristics

	mean	±	sd	range	n	%
Gender						
Male					37	19.9
Female					149	80.1
Osteoarthritis of						
Hip					66	35.5
Knee					112	60.2
Both					8	4.3
Age (in years)	67.9	±	8.7	39 , 84		

The Spearman rank correlations between the change scores of the methods are given in Table 3. Significant relationships were found between the observational method and three of the four questionnaires, i.e., the IRGL and NHP Mobility subscales, and the Low Chair subscale of the QR&S. The IRGL and NHP Mobility subscales also correlated with each other, and the two subscales of the QR&S correlated with each other. However, neither of the two subscales of the QR&S showed significant relationships with the other two self-report methods.

Table 2: Scores and change scores of observational and self-report methods for disability in mobility

	Week 0	Week 12	Change	Standardized Change
	mean ± sd	mean ± sd	mean ± sd	mean ± sd
Observed disability in mobility[1,2]	0.0 ± 1.0		-0.1 ± 1.0	-0.13 ± 0.73
Self-reported disability in mobility[1]				
NHP Mobility (0-100)	26.7 ± 16.5	24.5 ± 16.9	-2.2 ± 13.9	-0.13 ± 0.84
IRGL Mobility (-28 - -7)	-20.4 ± 5.5	-21.1 ± 5.7	-0.6 ± 4.3	-0.12 ± 0.77
QR&S High chair (0-10)	2.8 ± 2.6	2.7 ± 2.7	-0.1 ± 1.8	-0.04 ± 0.70
QR&S Low chair (0-10)	5.2 ± 3.3	5.1 ± 3.6	-0.1 ± 2.5	-0.02 ± 0.75

1 All scores are disability scores, i.e. a higher score represents a higher level of disability

2 For the observational method, the normal change score is identical to the standardized change score, and is therefore not given

Table 3: Spearman rank correlations between change scores of the methods

	Observational method	NHP mobility	IRGL mobility	QR&S high chair
Self-report methods				
NHP Mobility	.20*			
IRGL Mobility	.26*	.25*		
QR&S High chair	.14	.16	.18	
QR&S Low chair	.21*	.12	.14	.34*

* p<.01

Table 4: Factor loadings

	Factor 1	Factor 2
Observational method	0.65	-0.27
Self-report methods		
NHP Mobility	0.70	-0.13
IRGL Mobility	0.74	-0.19
QR&S High chair	0.23	-0.80
QR&S Low chair	0.21	-0.83

The results of the oblique factor analysis of the change scores of the observational method and the four questionnaires feature in Table 4. Two factors were present. The total proportion of variance accounted for by these two factors was 56.6%. Factor 1 accounted for 36.1% of variance; factor 2 accounted for the remaining 20.5%. The correlation between the two factors was -0.27 ($p<0.01$). However, contrary to the expectation, these two factors did not represent an observational factor and a self-report factor. Instead, factor 1 mainly comprised the observational method and the Mobility subscales of both NHP and IRGL. The second factor was predominantly loaded by the two subscales of the QR&S: High Chair and Low Chair.

The analyses presented here were repeated in two subgroups, one comprising all patients with OA of the knee and one consisting of all patients with OA of the hip. The results of these subgroup-analyses were similar to the results in the combined group of hip-OA and knee-OA patients, presented here.

DISCUSSION

The aim of the present study was to compare the responsiveness of observational and self-report methods for assessing disability in mobility in patients with OA of the hip or knee. To this end, the longitudinal convergent and divergent validity of these methods was assessed.

It was expected that the change scores of four self-report methods (questionnaires) would be mutually correlated (convergent validity), while the change scores of the observational method would not be correlated with self-report methods (divergent validity). Indeed, the correlations between the observational method on the one hand and the self-report methods on the other hand were rather low. However, these correlations were not lower than the inter-correlations among the questionnaires. Also, the results of the factor analysis indicated that changes on the observational method could not be consistently differentiated from changes on two questionnaires (NHP and IRGL). It can therefore be concluded that there was no systematic divergence between the observational method on the one hand, and the questionnaires on the other hand. Thus, no evidence was obtained for the differential responsiveness of observational and self-report methods. Instead, evidence for the systematic convergence of questionnaires and the observational method was found. Because questionnaires are easier to use, less time-consuming and less of a burden to patients, it seems that questionnaires should be prefered over observational methods. This confirms the recent recommendation of OMERACT to use questionnaires, and to not use observational methods (14).

The correlations between the observational and self-report methods were low, and did not exceed 0.26. In an earlier cross-sectional study the relationships between the observational method and the questionnaires, and the relationships among the questionnaires, were found to be distinctively stronger. The cross-sectional correlations between the methods all exceeded 0.50 (12) . There are two likely explanations for the lower correlations among the methods, both of which address sources of random variation between the methods: the amount of error present in the various scores and differences in item content between the methods.

Change scores were obtained by subtracting the baseline score from the score at week 12. Both these scores (baseline and week 12) have their own random error of measurement. Subtracting two scores containing a random error means that the resulting score is relatively largely influenced by random error. This presence of relatively large random variability in all the change scores can explain the low correlations between the various methods in the present longitudinal study.

A second possible explanation for the low correlations between the instruments is a difference in item content between the various methods. Both the NHP and IRGL mobility subscales and the observational method address walking as an important aspect of mobility, whereas the QR&S focuses entirely on rising, sitting down and reclining. Changes in the two subscales of the Questionnaire Rising and Sitting down (QR&S) were associated with each other only, and were not related to the two other questionnaires and the observational method. Thus, despite these questionnaires all measuring disability in mobility, subtle differences in the content of the items may cause differential responsiveness in these questionnaires. This notion is also supported by recent analyses on the Western Ontario and McMaster Universities Osteoarthritis Index (WOMAC), which revealed that specific disabilities which are part of general disability in mobility (such as walking down the stairs and getting into and out of a bath) had low correlations with other aspects of disability in mobility in patients with OA (27). This implies that item selection is an important source of divergence between different instruments.

An explanation for the isolated position of the QR&S in the present study might be that the two subscales of the QR&S have exactly the same format (wording of the items, response-categories), which may have contributed to the correlation between the change scores of these two subscales. Finally, the average standardized change of the two subscales of the QR&S was lower than the change of the other instruments (see Table 2). The relative stability of the subscales of the QR&S could be a sign of a relatively low responsiveness of the QR&S in this group of patients with osteoarthritis. In the absence of a true gold standard, this interpretation can not be substantiated, however.

The observational method used in the present study is an extensive method. Other, less resource- intensive, methods are available for observational assessment of disability. The disadvantages of observational methods mentioned in this paper apply less to these simpler methods. A more parsimonious approach to the observational assessment of disability, for instance by limiting the measurements to just the 5m walking time, could still be useful.

Disability in mobility was clearly present in the study population. However, on average, patients showed a moderate level of disability. As can be seen in Table 2, for three out of four questionnaires the average score was in the bottom half of the scale, for both measurements. It is possible that the actual level of disability affects the observed associations between the various disability measures. This could mean that the associations between the disability measures found in the present study, would be different in patients with more pronounced disability. However, in our opinion, it is unlikely that this would lead to systematic divergence between questionnaires on the one hand and observational methods on the other hand. Nevertheless, caution should be observed when generalising the results of the present study to other patient groups.

Earlier research on the responsiveness of disability measures has been scarce in patients with OA. In a study in patients with musculoskeletal disorders, the NHP mobility subscale was shown to be adequately responsive (28). In general, the available studies show various well-known disability measures (such as the SF36, Roland Disability Questionnaire, Sickness Impact Profile) to be responsive (28-30). It has also been reported that disease-specific measures are more responsive than generic measures (31). However, in most cases the ability of the instruments to detect clinically important differences (responsiveness) was based on a comparison with a gold standard, such as global perceived effect (28-31). The use of such external criteria as absolute measures of change is questionable (18). Also, recently the stability of the resulting estimates of instrument responsiveness has been questioned (32). Approaches to establishing the clinimetrical quality of instruments in longitudinal designs which do not utilize such external criteria as absolute measures of change, may therefore prove to be more accurate. The use of factor analysis, as presented in this paper, appears to be a useful alternative.

In conclusion, no evidence was obtained in support of a differential responsiveness of observational and self-report methods for assessing disability in mobility in OA-patients. We have previously shown that that these measures yield largely equivalent information in a cross-sectional design (12). Because of the advantages of questionnaires, this implies that in general the use of self-report methods for the assessment of disability is to be prefered over extensive observational methods in OA.

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4. MUSCLE STRENGTH, PAIN AND DISABILITY IN PATIENTS WITH OSTEOARTHRITIS

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ABSTRACT

Objective: Reduced muscle strength is regarded a risk factor for pain and disability in osteoarthritis. Currently, various indices for muscle strength are used when assessing determinants of pain and disability. The goal of the present study was to evaluate these indices of muscle strength.

Design: Isometric muscle strength was measured for 16 muscle actions around the knees and hips in 52 patients with OA of the hip and 70 patients with OA of the knee. Various indices of muscle strength were derived from these measurements, applying five alternative approaches. These approaches ranged from a single overall index to a set of 16 separate indices. The internal consistency of these indices was determined (Cronbach's α); and it was determined to what extent these indices could reveal the association between reduced muscle strength on the one hand and pain and disability on the other hand.

Results: Internal consistency was satisfactory for all indices (Cronbach's $\alpha > 0.74$). As expected, reduced muscle strength was associated with increased disability, but no clear relationship could be established between muscle weakness and pain. The strength of these associations did not depend on the approach used to derive the indices for muscle strength.

Conclusions: The indices did not show major differences with regard to internal consistency or the extent to which the association with pain and disability could be revealed. For reasons of parsimony, the most suitable approaches seem to be the ones resulting in few indices, i.e., the patient-based and averaged joint approach.

Keywords: Osteoarthritis, Muscle Strength, Assessment

Reduced muscle strength is frequently observed in patients with osteoarthritis (OA) of the hip or knee (1-6). Loss of muscle strength is an important determinant of pain and disability in patients with OA (7-11). In the treatment of OA, especially in exercise therapy, improving muscle strength is regarded one of the most important mechanisms towards reducing pain and disability (12, 13). Therefore, in research into the effectiveness of various treatments for OA, muscle strength is often chosen as one of the outcome measures.

The assessment of muscle strength involves measurement of a multitude of muscle actions, at a number of anatomical sites (i.e., different joints). The large amount of data, resulting from these measurements, usually has to be reduced into one or more indices (sum scores). A number of different approaches to the calculation of indices can be chosen. These approaches differ in their level of reduction, from an approach where the measurements of all different muscle actions are reduced into one overall index for muscle strength, to an approach where no data reduction is performed and every muscle action is used as a separate index for muscle strength. Approaches that result in a large amount of indices have the advantage that due to their high level of detail they may reveal relationships that would otherwise be obscured. On the other hand, such a level of detail can also result in an unnecessary amount of indices. This could lead to results that are difficult to interpret. So, a level of detail needs to be maintained that prevents the loss of vital information, while at the same time a low number of indices is advantageous for ease of use and interpretation of results.

Thus, the question arises whether the way in which data are reduced into indices affects the observed relationship between muscle strength and pain and disability. The goal of the present study was to determine which approach to the reduction of data into indices for muscle strength would provide the optimal trade-off between parsimony (as few indices as possible) and detail (no loss of vital information). To this aim, first the internal consistency of the indices resulting from various approaches was determined. Second, the relationship was assessed between the indices of muscle strength on the one hand and pain and disability on the other hand. The relationship between muscle weakness and both pain and disability in OA has been well established (9,14,15). An index, or indices, for muscle strength should be able to accurately reveal the existence of these relationships. Thus, it was determined to what extent these indices could reveal the association between muscle strength on the one hand and pain and disability on the other.

METHODS

Subjects

Data were obtained from a randomized clinical trial into the effectiveness of exercise therapy in patients with OA of the hip or knee (16). Patients were included if they were diagnosed to have OA of the hip or knee according to the clinical classification criteria of the American College of Rheumatology (17-18). 200 Patients participated in the trial. These were mainly patients with relatively mild symptoms of OA. For the present study, data were only used from patients diagnosed with either OA of one knee or OA of one hip. Patients diagnosed with OA at more than one site were excluded from the analyses presented here. This resulted in 122 patients in the analyses, 70 patients with unilateral knee-OA and 52 patients with unilateral hip-OA. Data for the present study were obtained at the onset of the trial (baseline).

Muscle strength

Isometric muscle strength was measured with a hand-held dynamometer (19), the MicroFet (Hoggan Health Industries, Draper, Utah). Hand-held dynamometry has been shown to be a reliable method of assessing muscle strength (20-23), and the dynamometer has the advantage of being small and easy to use. Make tests, or "doctor initiated" methods (21), were used. This means that the research assistant holds the dynamometer steady, while the patient exerts maximum force against it (24). After one initial attempt, during which the patient could get used to the required movement, patients were asked to build their effort to a maximum in the first two seconds of the test, and then to maintain this maximum force for three seconds. The peak force registered during those three seconds was recorded as the patient's maximum force. Muscle strength in Newton (N) was measured bilaterally for 8 muscle actions: flexion, extension, external rotation, internal rotation, abduction and adduction of the hip, and flexion and extension of the knee. Each muscle action was measured once. All muscle strength tests were performed within one session. During this session, other physical examinations were also carried out. This provided sufficient time between the muscle strength tests to avoid fatigue in the patients. During the tests, no fatigue or excessive pain (i.e., more pain than normal) was present as assessed by the patients themselves.

Starting positions were analogous to Kendall et al. (25). The protocol of Kendall et al provides starting positions for both patient and therapist, and also prescribes the positioning of the dynamometer. In case a patient is unable to adopt a prescribed position, the protocol also provides an alternative position. The tests were carried out by two experienced physical therapists. Prior to the trial, the interrater reliability was established using 10 healthy subjects and 10 patients with OA. The inter rater reliability was satisfactory for all muscle actions (Pearson's R exceeding 0.75 for all actions).

These measurements resulted in 16 items for muscle strength: 8 actions, both left and right. First, these measurements were corrected for body mass by dividing them by the patient's weight. After that, for all 16 items z-scores were computed to exclude problems due to different score ranges between items. A distinction was made between affected and unaffected joints (e.g., z-scores were computed for flexion of the affected knee and the unaffected knee). For patients with hip-OA, a distinction for the knees was made between knees ipsilateral and contralateral to the affected hip. Likewise, for knee-OA patients, hips were divided into ipsilateral and contralateral to the affected knee.

A number of different approaches was chosen to compute indices for muscle strength. These were:

- Patient-based approach: 1 index; all 16 items added up to obtain a single index for muscle strength.
- Averaged joint approach: 2 indices; a separate index was computed for muscle strength around the knee (comprising 4 items: flexion and extension for both the affected and unaffected knee), and for muscle strength around the hip (comprising 12 items: all six muscle actions for both the affected and unaffected hip).
- Single joint approach: 4 indices; separate indices were made for the affected knee, unaffected knee, affected hip and unaffected hip. The knee indices consisted of two items each, while the hip indices had 6 items each.
- Averaged muscle action approach: 8 indices; they were made for each action by adding up the same action for the affected and unaffected joint (e.g., flexion of the affected hip and unaffected hip were added up to obtain a score for flexion of the hip). This resulted in separate indices for flexion, extension, external rotation, internal rotation, abduction and adduction of the hip, and flexion and extension of the knee.
- Single muscle action approach: no indices were computed, instead all 16 items were regarded as separate variables.

Pain and disability

Pain as experienced by patients was assessed using a visual analogue scale (VAS; 0-100 mm), in which 0 mm represents no pain at all and 100 mm represents 'the worst pain I can imagine'. Patients were asked to rate their overall pain in the past week.

Observed disability was determined by watching videotaped performances of patients on a series of standardised tasks, using an adaptation of the method described by Keefe (26-28). An overall-score for observed disability was calculated based on five items: three movement times (5m-walking time, stand-to-sit time and stand-to-recline time), and two measures for the quality of the performance (level of guarding, level of rigidity). The overall-score for observed disability was constructed as follows. First of all, for the three movement times scores were transformed into ten categories (each containing 10% of scores), to correct for skewed distributions. Then z-scores were computed for all five items (movement times and qualitative assessments), to avoid weighting problems due to differences in score range between the items. These z-scores were added up to obtain an overall-score for observed disability per patient. By definition, z-scores have a mean of 0, which means that an overall score constructed from z-scores will also have a mean of 0. A higher overall-score means a higher level of disability. All items were scored by trained observers. This overall-score has been shown to be internally consistent and valid (29).

Next to observed disability, self-reported disability was also assessed. To this end, the mobility subscale of the IRGL was used. The IRGL (Influence of Rheumatic Disease on General Health and Lifestyle) is a Dutch adaptation of the Arthritis Impact Measurement Scales (AIMS) (30). This subscale has seven items. Two items are general statements concerning disability in mobility, while the other five address disability in climbing stairs, riding a bicycle and walking. The IRGL is a 'positive' questionnaire, i.e. it measures 'ability' rather than 'disability'. To facilitate interpretation, scores on this test have been reversed to obtain a 'disability-score'. After the reversal, the score range for this test is -28 (minimum disability) to -7 (maximum disability).

Statistical analyses

All analyses were performed using two subgroups of patients: one group comprising all patients with knee-OA, and another group comprising all patients with hip-OA.

An initial comparison between muscle strength around the affected or ipsilateral joint and muscle strength around the unaffected or contralateral joint was made using t-tests. To assess the inter-relationships between the various muscle actions, first Pearson correlation coefficients were computed between all muscle actions. Next to that, factor analysis was performed. With the factor analysis, the unidimensionality of the pool of muscle actions could be assessed (i.e., can strength for a number of muscle actions be regarded as representations of the same entity, or should a clear distinction be made between, for instance, muscle strength around the hip and muscle strength around the knee?). The inclusion criterion for factors in this analysis was an eigenvalue > 1.

The internal consistency of the several indices for muscle strength was assessed by computing Cronbach's α. A value of 0.80 or more represents high internal consistency (31).

Multiple regression analyses were performed with pain, observed disability and self-reported disability as dependent variables. The indices for muscle strength, which had been computed using various approaches (see above), were the independent variables. A separate analysis was performed for each combination of a dependent variable (i.e., pain or self reported or observed disability) and an approach for computing indices for muscle strength (e.g., patient-based, joint-based). These regression analyses could establish the relationships between muscle strength on the one hand and pain and disability on the other hand.

All statistical analyses were carried out using SPSS version 8.0.

RESULTS

Patient characteristics

Table 1 features the mean age and proportion of males and females in the study population. Also, raw item scores of the 16 muscle actions (corrected for the patient's body weight), mean sum score on observed disability and self-reported disability, and mean pain-score are given for both the knee-OA and hip-OA group.

A significant difference between muscle strength around affected joints and around unaffected joints was found for five muscle actions. In four cases, muscle strength around affected joints was significantly lower than around unaffected joints, in one case (adduction of the hip) strength around the affected joint was better than around the unaffected joint. For two muscle actions, a significant difference in strength was found between ipsilateral and contralateral joints (e.g., in patients with knee-OA, muscle strength for flexion of the ipsilateral hip was significantly lower than for flexion of the contralateral hip).

On average, patients in the knee-OA group had decreased muscle strength compared to patients in the hip-OA group, including decreased muscle strength around the hip.

Inter-item relationships

Pearson correlation coefficients between the 16 muscle actions for both subgroups are presented in Table 2 and 3. All coefficients are significantly greater than zero ($p=.000$ for all coefficients). The highest correlations were found between the lateral and contralateral side of the same muscle action (e.g., flexion of the affected and unaffected hip). These correlations range from 0.74 to 0.88 in the hip-OA group and

.67 to .84 in knee-OA group. Correlations between two ipslateral muscle actions (e.g., flexion and extension of the affected hip) tend to be slightly higher than correlations between a lateral and different contralateral muscle action (e.g., flexion of the affected hip and extension of the unaffected hip).

Internal consistency

The internal consistency of the various indices was assessed computing Cronbach's Alpha. The results for both subgroups of patients are presented in Tables 4 and 5. Since the single muscle action approach does not use an index that consists of more than one item, this approach was not included in these analyses. The internal consistency is high for nearly all indices. Only the indices for muscle strength around the knee in the single joint approach scored below 0.80, indicating satisfactory but not high internal consistency.

For both subgroups, factor analysis was also performed, to assess the unidimensionality of the complete set of muscle actions. For both the knee-OA and hip-OA group, one factor was identified by the analysis. In the hip-OA group, this factor accounted for 64.9% of variance between the items, with factor loadings of the separate muscle actions ranging form .75 to .89. The results in the knee-OA group were similar: the one factor accounted for 66.2% of variance, with factor loadings ranging from .76 to .89.

The relationship with pain and disability

The results of the various multiple regression analyses are shown in Table 6 and 7. These tables feature - for each approach separately - the fraction of variance accounted for (r^2). In both groups, similar results are found for the various approaches. The strength of the relationship between observed disability and muscle strength ranges from 0.173 to 0.227 in the hip-OA group and from 0.146 to 0.205 in the knee-OA group. The same applies to self-reported disability (r^2 ranging from 0.136 to 0.174 in the hip-OA group, and from 0.130 to 0.178 in the knee-OA group). Only a minimal relationship could be established between pain and muscle strength. Again, no major differences were found between the various approaches in either of the two subgroups. In all cases, the relationship between muscle strength and pain and disability was a negative one: muscle weakness (i.e., lower muscle strength) was associated with more disability and, to a lesser extent, pain.

The analyses presented in this paper were also carried out without taking the actual location of OA into account. In these additional analyses, a distinction between left and right was made, instead of between affected and unaffected joints (e.g., for the single joint approach, separate indices were constructed for the left and right knee, rather than the affected and unaffected knee). The results of these additional analyses were remarkably similar to the results presented here. The results of these additional analyses can be found on the Internet, at http://www.nivel.nl/muscle/index.html.

Table 1: Patient Characteristics

	Hip-OA	Knee-OA
Number of patients	52	71
Demographics:		
Sex		
Female	34 (65.4%)	57 (80.3%)
Male	18 (34.6%)	14 (19.7%)
Age (in years)	67.8 ± 9.2	68.2 ± 8.9
Weight (in kg)	76.2 ± 11.3	78.1 ± 11.8
Pain and disability:		
Overall pain in the past week (0-100)	39.4 ± 25.8	44.4 ± 29.4
Observed disability (composite z-score)	-0.18 ± 0.99	0.08 ± 0.94
Self-reported disability (-7 to –28)	-21.9 ± 4.9	-19.9 ± 6.0
Muscle strength (in N/kg):		
Hip flexion*°		
Affected / ipsilateral	2.22 ± 0.81	2.03 ± 0.75
Not affected / contralateral	2.50 ± 0.82	2.31 ± 0.77
Hip extension		
Affected / ipsilateral	1.64 ± 0.81	1.43 ± 0.69
Not affected / contralateral	1.72 ± 0.72	1.45 ± 0.70
Hip internal rotation		
Affected / ipsilateral	1.75 ± 0.66	1.57 ± 0.57
Not affected / contralateral	1.81 ± 0.62	1.68 ± 0.58
Hip external rotation°		
Affected / ipsilateral	1.40 ± 0.43	1.27 ± 0.45
Not affected / contralateral	1.46 ± 0.39	1.39 ± 0.42
Hip adduction*		
Affected / ipsilateral	2.15 ± 0.77	1.75 ± 0.73
Not affected / contralateral	2.01 ± 0.74	1.82 ± 0.72
Hip abduction*		
Affected / ipsilateral	2.15 ± 0.72	2.28 ± 0.82
Not affected / contralateral	2.39 ± 0.70	2.34 ± 0.80
Knee flexion*		
Ipsilateral / affected	1.28 ± 0.46	1.15 ± 0.42
Contralateral / not affected	1.28 ± 0.41	1.22 ± 0.42
Knee extension*		
Ipsilateral / affected	2.23 ± 0.67	2.03 ± 0.69
Contralateral / not affected	2.30 ± 0.67	2.27 ± 0.72

* significant difference in muscle strength between affected and unaffected joint ($p<.05$)
° significant difference in muscle strength between ipsilateral and contralateral joint ($p<.05$)

Table 2: Inter-correlations of strength of muscle actions in patients with hip-OA

	Hip Flexion		Hip Extension		Hip Internal Rotation		Hip External Rotation		Hip Adduction		Hip Abduction		Knee Flexion		Knee Extension	
	aff.	unaff.	aff.	unaff.	aff.	unaff.	aff.	unaff.	aff.	unaff.	aff.	unaff.	ipsi	contra	ipsi	contra
Hip Flexion																
affected side																
unaffected side	0.79															
Hip Extension																
affected side	0.54	0.48														
unaffected side	0.61	0.65	0.80													
Hip Internal Rotation																
affected side	0.55	0.49	0.55	0.54												
unaffected side	0.53	0.53	0.45	0.56	0.74											
Hip External Rotation																
affected side	0.70	0.63	0.51	0.59	0.70	0.79										
unaffected side	0.52	0.64	0.50	0.59	0.62	0.77	0.79									
Hip Adduction																
affected side	0.77	0.72	0.56	0.67	0.61	0.66	0.77	0.72								
unaffected side	0.66	0.64	0.65	0.66	0.69	0.69	0.73	0.72	0.86							
Hip Abduction																
affected side	0.58	0.57	0.39	0.49	0.64	0.66	0.66	0.63	0.70	0.67						
unaffected side	0.66	0.57	0.55	0.56	0.52	0.57	0.64	0.63	0.73	0.74	0.75					
Knee Flexion																
ipsilateral side	0.70	0.71	0.50	0.57	0.49	0.44	0.55	0.62	0.64	0.56	0.52	0.59				
contralateral side	0.59	0.68	0.47	0.54	0.44	0.47	0.60	0.71	0.65	0.56	0.54	0.52	0.88			
Knee Extension																
ipsilateral side	0.69	0.56	0.45	0.58	0.76	0.75	0.75	0.63	0.68	0.65	0.61	0.62	0.54	0.47		
contralateral side	0.57	0.57	0.36	0.61	0.67	0.74	0.69	0.70	0.67	0.58	0.62	0.55	0.54	0.53	0.85	

All correlations: $p<.01$

Table 3: Inter-correlations of strength of muscle actions in patients with knee-OA

	Hip Flexion		Hip Extension		Hip Internal Rotation		Hip External Rotation		Hip Adduction		Hip Abduction		Knee Flexion		Knee Extension	
	Ipsi	contra	ipsi	contra	ipsi	contra	ipsi	contra	ipsi	contra	ipsi	contra	aff.	unaff.	aff.	unaff.
Hip Flexion																
ipsilateral side																
contralateral side	0.84															
Hip Extension																
ipsilateral side	0.58	0.53														
contralateral side	0.54	0.59	0.80													
Hip Internal Rotation																
ipsilateral side	0.60	0.46	0.40	0.37												
contralateral side	0.66	0.76	0.53	0.61	0.67											
Hip External Rotation																
ipsilateral side	0.77	0.71	0.53	0.49	0.73	0.77										
contralateral side	0.70	0.72	0.56	0.55	0.62	0.82	0.80									
Hip Adduction																
ipsilateral side	0.73	0.64	0.50	0.52	0.60	0.61	0.72	0.60								
contralateral side	0.72	0.69	0.46	0.54	0.58	0.75	0.72	0.70	0.80							
Hip Abduction																
ipsilateral side	0.67	0.66	0.48	0.51	0.54	0.64	0.67	0.66	0.71	0.76						
contralateral side	0.69	0.68	0.53	0.57	0.54	0.71	0.72	0.74	0.71	0.76	0.83					
Knee Flexion																
affected side	0.81	0.80	0.60	0.64	0.51	0.71	0.75	0.70	0.71	0.70	0.65	0.69				
unaffected side	0.65	0.77	0.55	0.61	0.30	0.66	0.59	0.63	0.56	0.55	0.50	0.60	0.80			
Knee Extension																
affected side	0.64	0.60	0.50	0.45	0.55	0.63	0.66	0.66	0.65	0.69	0.70	0.73	0.73	0.51		
unaffected side	0.59	0.68	0.37	0.42	0.42	0.76	0.58	0.66	0.57	0.69	0.60	0.63	0.68	0.63	0.77	

All correlations: $p<.01$

Table 4: Internal consistency of indices of muscle strength in patients with hip-OA

		n_I	Cronbach Reliability (α)
Patient-based		16	0.97
Averaged joint	Hip	12	0.96
	Knee	4	0.88
Single joint	Hip affected	6	0.91
	Hip not-affected	6	0.92
	Knee ipsilateral	2	0.75
	Knee contralateral	2	0.74
Averaged muscle action	Hip flexion	2	0.87
	Hip extension	2	0.91
	Hip external rotation	2	0.87
	Hip internal rotation	2	0.86
	Hip abduction	2	0.90
	Hip adduction	2	0.91
	Knee flexion	2	0.92
	Knee extension	2	0.87

n_I = number of items per index

Table 5: Internal consistency of indices of muscle strength in patients with knee-OA

		n_I	Cronbach Reliability (α)
Patient-based		16	0.97
Averaged joint	Knee	4	0.90
	Hip	12	0.96
Single joint	Knee affected	2	0.85
	Knee not-affected	2	0.78
	Hip ipsilateral	6	0.91
	Hip contralateral	6	0.93
Averaged muscle action	Knee flexion	2	0.90
	Knee extension	2	0.85
	Hip flexion	2	0.91
	Hip extension	2	0.89
	Hip external rotation	2	0.89
	Hip internal rotation	2	0.87
	Hip abduction	2	0.91
	Hip adduction	2	0.89

n_I = number of items per index

Table 6: Results of multiple regression analyses: fraction of variance accounted for per approach (Hip-OA)

	Observed Disability	Self-reported Disability	Pain
Patient-based	0.227	0.143	0.036
Averaged joint	0.218	0.148	0.039
Single joint	0.184	0.136	0.043
Averaged muscle action	0.195	0.142	0.029
Single muscle action	0.173	0.174	0.067

Table 7: Results of multiple regression analyses: fraction of variance accounted for per approach (Knee-OA)

	Observed Disability	Self-reported Disability	Pain
Patient-based	0.205	0.178	0.025
Averaged joint	0.205	0.177	0.042
Single joint	0.185	0.165	0.056
Averaged muscle action	0.146	0.130	0.053
Single muscle action	0.202	0.177	0.057

DISCUSSION

Various approaches are available to the calculation of indices of muscle strength in patients with OA of the hip or knee. The goal of the present study was to determine which approach to the reduction of data into indices for muscle strength would provide the most useful and accurate set of indices. Such an approach would have the optimal trade-off between parsimony (as few indices as possible) and detail (no loss of vital information). To this aim, first the inter-relationships between the various muscle actions and the internal consistency of the indices for muscle strength were established. Next to that, the relationship of the various indices with pain and disability was established. With regard to the inter-relationships between muscle actions, the correlations showed that muscle strength for various actions is closely inter-related. As the factor analyses have shown, strength of singular muscle actions can be regarded as representations of one unidimensional construct. With respect to both the internal consistency and the relationship with pain and disability there were only minor differences between the indices resulting from various approaches. The internal consistency of the indices all reached an acceptable level. Also, in the regression analyses, the strength of the relationship between the indices of muscle strength and pain and disability was hardly influenced by the choice of approach. Isometric muscle strength is negatively associated with disability, in all of the approaches chosen. This is consistent with earlier findings (9,14,15). Variations in muscle strength were found to account for some 15-20% of the variance in disability.

It was found that muscle strength around affected joints was decreased compared to muscle strength around unaffected joints. However, the relationship between muscle strength and pain appears not to be primarily determined by muscle strength around affected joints. As demonstrated, muscle strength in these patients can be regarded a trait at the level of the patient. Hence, the relationship between disability and muscle strength appears to be determined by muscle strength in general. Nevertheless, the difference in muscle strength around affected and unaffected joints is an important clinical feature of OA, which should not be neglected. Therefore, using more detailed indices for muscle strength, which distinguish between muscle strength around affected and unaffected joints, is appropriate.

In our opinion, the strength of the relationship between muscle weakness and disability (15-20% variance-accounted-for) marks muscle weakness as an important entity within the multi-factorial framework of determinants of disability. No clear relationship was identified between pain and muscle strength. Muscle weakness was weakly associated with more pain, with muscle strength accounting for, on average, 5% of the variance in pain levels. A stronger relationship was expected to be found, and indeed has been found in an earlier study involving the same group of patients (32). In the present study, a substantial number of patients were excluded because they were diagnosed with bilateral OA. The resulting smaller group sizes, and therefore decreased statistical power, may be responsible for the inability of the present study to identify a clear relationship between muscle strength and pain.

The results seem to indicate that all of the aforementioned approaches are generally acceptable. For reasons of parsimony, approaches resulting in relatively few indices, such as the patient-based or averaged joint approach, may therefore be the best option. These approaches result in just one or two indices, which makes analysing and interpreting the results less complicated. The use of a patient-based approach is also consistent with Zhang et al. (33), who stated that when an overall assessment of a patient's level of functioning is made, only a patient-based analysis is appropriate. Of course, the choice of which approach is most suitable depends on the specific design and aims of a given study. The present study has shown that each approach yields consistent indices, and that the manner in which data reduction is carried out does not interfere heavily with the outcomes of statistical analyses. In the context of research on the relationship between muscle strength and pain and disability in OA-patients, it is concluded that the most suitable approach seems to be either a single overall index (patient-based approach) or two separate indices, one for muscle strength around the hip and one for muscle strength around the knees (averaged joint approach).

For clinicians, whose interests may differ from researchers', these results may have other implications. Given the clear inter-relationships between various muscle actions, measuring a limited number of muscle actions will provide accurate information on a patient's overall level of muscle strength. The present study has shown that the same muscle actions on the ipsilateral and contralateral side are very closely related to each other, and that the entire pool of muscle actions around the knees and hips can be regarded a unidimensional trait of the patient. Although closely associated, muscle strength around affected joints is generally decreased compared to muscle strength around unaffected joints (34). Overall, these findings suggest it is sufficient to assess a limited number of muscle actions.

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5. RANGE OF JOINT MOTION AND DISABILITY IN PATIENTS WITH OSTEOARTHRITIS OF THE KNEE OR HIP

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ABSTRACT

Objective: To establish the relationships between the range of joint motion (ROM) and disability in patients with osteoarthritis (OA) of the knee or hip. Two related issues were addressed: (1) the inter-relationships between ROMs of joint actions, and (2) the relationship between ROM and disability.

Methods: Data on 198 patients with OA of the knee or hip were used. The ROM was assessed bilaterally, using a goniometer. Disability was assessed using a self-reporting method (questionnaire) and an observational method. Correlation and factor analysis were used to establish the inter-relationships between the ROMs of joint actions. Correlation and multiple regression analysis were carried out to establish the relationships between ROM and disability.

Results: Close inter-relationships were found between the ROMs of the same joint action of the lateral and contralateral joints; inter-relationships between ROMs of different joint actions were substantially weaker. Low ROMs were associated with high levels of disability, both self-reported and observed. Some 25% of the variation in disability levels could be accounted for by differences in ROM. In both knee and hip OA patients, flexion of the knee and extension and external rotation of the hip were found to be most closely associated with disability.

Conclusion: Restricted joint mobility, especially in flexion of the knee and extension and external rotation of the hip, appears to be an important determinant of disability in patients with OA

Keywords: osteoarthritis, range of motion, disability

Physical disability is frequently reported in patients with osteoarthritis (OA) (1). However, the disabled condition of these patients can only partially be explained by the degeneration of joints affected by OA (2-5). A number of other factors have been proposed as possible explanations for the level of disability in these patients (2,6). This includes physical factors, such as reduced range of motion (ROM) of joints (2).

Relationships between range of joint motion in general and disability have been reported. Dunlop et al (7) identified joint impairment as a predictor for disability. In their study, the presence of impaired range of motion was one of the factors defining joint impairment, but other factors, such as tenderness, swelling or pain during motion were also used in the assessment of joint impairment. Thus, this study did not identify a separate association between joint mobility and disability. In a study among Swedish elderly, strong correlations were found between ROM of knee- and hip-joints and disability (8).In an other population study, Odding et al. (9) found that restricted flexion of the hips and restricted flexion of the knees were strong risk factors for locomotor disability (disability in activities primarily involving the lower extremities, such as walking, stair climbing, rising from and sitting down in a chair). Recently, Escalante et al. (10) have reported that impaired hip flexion was associated with a decrease in walking velocity in a population of elderly.

In patients with OA, the existence of this relationship has also been reported. In a review of previous studies, Dekker et al. (2) identified impaired joint mobility in general as a factor in the development of disability in patients with OA. However, to our knowledge, in recent years no further research on this topic has been published. The role of physical functioning as a determinant of disability in OA has been widely investigated, but research has primarily been focussed on the impact of decreased muscle strength on disability in OA (4,5,11-13). The relationship between joint mobility and disability in OA has not been established in more detail than the global statement of Dekker et al. (2). The impact of restricted mobility of specific joint actions on disability has not been established in patients with OA. Establishing these relationships in detail could provide essential information for both the assessment and management of disability in patients with OA.

The overall goal of the present study was to further establish the relationship between ROM and disability in patients with OA of the knee or hip. To this aim, two related issues were addressed. First, the inter-relationships between ROM of all joint actions of the knees and hips were studied. Before studying the role of ROM as a determinant of disability, it is of vital importance to have insight in the nature of ROM in OA. Can ROM be regarded a unidimensional trait of an OA patient (i.e., ROM of all joint actions are closely inter-related and can be regarded representations of one trait: a patient's overall sinuosity)? Or must ROM of separate joint actions be treated as separate entities, with their own specific impact on a patient's level of physical ability? Second, and only after establishing the best level of aggregation for ROM-data, the relationship between ROM and disability in OA was then studied.

METHODS

Subjects

Data were obtained from a randomized clinical trial into the effectiveness of exercise therapy in patients with osteoarthritis of the hip or knee (14). Patients were included if they were diagnosed to have OA of the knee or hip according to the classification criteria of the American College of Rheumatology (15,16). 200 Patients participated in the trial. Data for the present study were obtained at the onset of the trial (baseline). Of two patients data on one or more of the tests presented below were incomplete. These patients' data were excluded from the analyses, resulting in 198 patients in the analyses.

Range of motion

Assisted active ROM was measured bilaterally for the hip and knee, using a goniometer. ROM measurements were scored in degrees. Measurements were taken according to Norkin and White (17). These measurements were taken for all possible actions of the joint, i.e. flexion, extension, internal and external rotation, abduction and adduction of the hip, and flexion and extension of the knee. For each action, the protocol provided starting positions for both patient and therapist, reference points for the pivot and distal points of the goniometer, and a horizontal (defining positive and negative values for ROM). In case a patient was not able to adopt a prescribed starting position, the protocol provided an alternative starting position. By supplying a protocol for starting positions for the patient and therapist and positioning of the goniometer, the reliability of the ROM-measurements was increased (18). The full protocol for taking ROM-measurements can be found on the Internet, at http://www.nivel.nl/english/range_of_motion/protocol_ROM.html. The tests were carried out by two experienced physical therapists. Prior to the measurements used for the present study, the inter-rater reliability was established using 10 healthy subjects and 10 patients with OA. The inter-rater reliability was satisfactory for all joint actions (Pearson's R exceeding 0.75 for all actions).

Disability

Two instruments for the assessment of disability were used: one observational method and one questionnaire. Observed disability was determined by watching videotaped performances of patients on a series of standardized tasks (19). These standardized tasks included: walking, sitting down into a chair, reclining onto a bed and bending over to pick a weight up from the floor. Trained observers scored the performance of the patients. The observers scored five items: three movement times (5m-walking time, stand-to-sit time and stand-to-recline time), and two measures for the quality of the performance (level of guarding, level of rigidity). Based on these five items, an overall score for observed disability was calculated (for details, see Steultjens et al., 19) A higher overall-score means a higher level of disability. This overall-score has been shown to be internally consistent and valid (19).

Next to observed disability, self-reported disability was also assessed. To this end, the mobility subscale of the IRGL-questionnaire was used. The IRGL (Influence of Rheumatic Disease on General Health and Lifestyle) is a Dutch adaptation of the Arthritis Impact Measurement Scales (AIMS) (20,21). This subscale has seven items. Two items are general statements concerning disability in mobility, while the other five address disability in climbing stairs, riding a bicycle and walking. The IRGL is a 'positive' questionnaire, i.e. it measures 'ability' rather than 'disability'. To facilitate interpretation, scores on this test have been reversed to obtain a 'disability-score'. After the reversal, the score range for this test is -28 (minimal disability) to -7 (maximal disability).

Statistical analyses

To establish the inter-relationships of the ROM scores of the joint actions, Pearson correlation coefficients were computed and factor analysis was performed. Both types of analysis assess the associations between items (in this case, ROM of joint actions), but correlation coefficients only provide information for pairs of items, whereas with factor analysis it is possible to assess the inter-relationships within the complete pool of items. The number of factors resulting from the factor analysis provides insight into the dimensionality of ROM. If the analysis produces one factor, containing all ROM items, joint ROM can be regarded a unidimensional trait of the patient (i.e., ROM of different joint actions are all representations of the same trait: a patient's sinuosity). If more factors are identified, effectively isolating subgroups of closely associated items, joint ROM cannot be regarded a trait of the patient, but a trait at a lower level (e.g., of the joint or the joint action). In the analysis, factors are identified if they can account for a significant amount of variance within the item pool (represented by an eigenvalue > 1).

The relationship between range of joint motion and locomotor disability was assessed using both bivariate and multivariate analyses. Pearson correlation coefficients were computed to establish the bivariate relationships between ROM for the joint actions on the one hand and the two disability measures on the other hand. Next to that, stepwise multiple regression analyses were performed, using observed disability and self-reported disability as dependent variables. The ROM-scores were the independent variables. Stepwise regression shows which of the ROM-scores are predominantly responsible for the relationship between joint ROM on the one hand and disability on the other hand. The inclusion criterion for the regression analyses was $p<0.05$ (p-in = 0.05), the exclusion criterion was $p>0.10$ (p-out = 0.10). The bivariate and multivariate analyses were carried out using the total group of patients (including both hip-OA and knee-OA patients), and also using subgroups of hip-OA or knee-OA patients only. All analyses were carried out using SPSS for Windows 8.0

RESULTS

Patient characteristics

Table 1 features the patient characteristics, average scores on observed and self-reported disability, and average ROM-scores for all joint actions. A negative ROM-score means a patient was not able to reach the position defined as horizontal by the protocol for ROM-measurements (17). A substantial group of patients was not able to reach the defined horizontal for extension of the knee or hip. This is known as flexion contracture. Flexion contracture of both the knee and hip was present in 25.5% of patients. Flexion contracture of the knee was present in an additional 31.5% of patients, whereas flexion contracture of the hip was found in another 15.5% of patients. In total, in 72.5% of patients flexion contracture was observed of either the knee or hip or of both the knee and hip.

Table 1: Patient characteristics (N=198)

	mean	±	sd	range	n	%
Gender						
Male					43	21.7
Female					155	78.3
Osteoarthritis of						
Hip					69	34.8
Knee					119	60.1
Both					10	5.1
Age (in years)	68.0	±	8.9	39 - 84		
Disability						
Observed disability	0.0	±	3.9	-6.2 - 13.4		
Self-reported disability (IRGL)	-20.3	±	5.6	-28 - -7		

Range of Motion
(in degrees; mean ± sd and range)

	Left			Right		
Knee flexion	136.3	± 11.4	(65-155)	136.3	± 10.6	(90- 53)
Knee extension	0.3	± 5.3	(-20- 13)	0.2	± 5.2	(-19- 12)
Hip flexion	115.4	± 12.3	(25-146)	115.3	± 11.7	(60-148)
Hip extension	2.3	± 7.3	(-18- 30)	2.2	± 7.6	(-15- 30)
Hip adduction	11.8	± 4.7	(0- 25)	12.2	± 4.6	(-2- 22)
Hip abduction	18.3	± 6.9	(-3- 40)	17.2	± 7.5	(-7- 45)
Hip internal rotation	29.4	± 9.5	(-5- 50)	28.6	± 10.2	(-15- 52)
Hip external rotation	34.8	± 9.6	(-5- 60)	34.5	± 9.2	(10- 56)

Inter-relationships of joint actions

The Pearson correlation coefficients between the ROM-values of all joint actions of the knees and hips are presented in Table 2. The highest correlations were found between the identical actions of the lateral and contralateral joint (e.g., flexion of the left hip and flexion of the right hip), these correlations ranged from .35 to .82. Especially for the joint actions of the hip in the transversal and sagittal plane (flexion, extension and internal and external rotation), these correlations were rather high, ranging from .62 to .82. The correlations among other joint actions (i.e., other than identical actions of the lateral and contralateral joint) were substantially lower, ranging from -.16 to .44.

In the factor-analysis, five factors were identified. In total, these factors accounted for 66.5% of variance. The factor structure is presented in Table 3. This table features the factor structure of the variables for joint mobility. Factor loadings < 0.40 are excluded from this table. All five factors are loaded by identical muscle actions of the lateral and contralateral joint (e.g., factor 1 is loaded by flexion of both the left and right hip). Both joint actions of the knee load on the same factor. The other four factors are loaded by the six joint actions of the hip.

Although there are also some significant relationships between different joint actions (e.g., flexion and internal rotation of the hip), the correlation coefficients and factor structure primarily reveal a very close relationship between ROM of the same joint action of the lateral and contralateral joint. Because of this finding, the average ROM of the same lateral and contralateral joint action was calculated and used as ROM value for that joint action (e.g., the average ROM value of flexion of the left knee and flexion of the right knee was calculated and used as ROM value for flexion of the knee) in the subsequent analyses on the association between range of motion on disability.

Range of joint motion and disability

Pearson correlation coefficients between the ROM of joint actions on the one hand and disability on the other hand are presented in Table 4. Generally, there was a negative relationship between ROM and disability. This means that a decreased ROM is associated with an increase in disability. However, not all joint actions seemed to be related to the level of disability in these patients. Extension, abduction and external rotation of the hip, and flexion of the knee were primarily associated with disability (both self-reported and observed), in both patients with hip-OA and patients with knee-OA. To a lesser degree, significant relationships were also found between ROM of flexion of the hip and disability.

The results of the multiple regression analyses are presented in Tables 5 and 6. Each table features the results of both the total group and subgroups comprising only hip-OA or knee-OA patients. Again, the level of disability was found to be dependent on the level of joint mobility. For both observed and self-reported disability, between 20%-25% of the variance in disability was accounted for by the level of joint mobility of patients. The pattern found in subgroups of hip-OA and knee-OA patients was largely equivalent to that found in the total group. Extension and external rotation of the hip were primarily responsible for the association between joint ROM and disability. Flexion of the knee was a third important joint action in this relationship.

Table 2: Inter-relationships of ROM of joint actions: Pearson correlation coefficients

		K extension		K flexion		H flexion		H extension		H abduction		H adduction		H ext. rot.		H int. rot	
		L	R	L	R	L	R	L	R	L	R	L	R	L	R	L	R
K ext.	L																
	R	.50															
K flex.	L	.36	.19														
	R	.07	.41	.50													
H flex.	L	-.02	.07	.17	.27												
	R	-.16	-.03	.05	.14	.71											
H ext.	L	.21	.13	.13	.15	.14	.16										
	R	.12	.14	.08	.11	.08	.21	.82									
H abd.	L	.11	.07	.07	.19	.33	.27	.33	.30								
	R	-.11	-.05	-.01	.01	.20	.42	.28	.39	.50							
H add.	L	.07	.12	.11	.08	.17	.09	.07	-.00	-.10	.05						
	R	-.05	.08	.01	.10	-.05	.14	.12	.12	-.05	-.02	.35					
H ext. rot	L	.03	.07	.19	.21	.41	.26	.03	.01	.40	.27	.07	-.02				
	R	-.13	-.08	.10	.16	.29	.37	.01	.05	.28	.41	-.04	.10	.68			
H int. rot	L	.05	.07	.13	.14	.36	.31	.16	.09	.26	.18	.40	.23	.01	.05		
	R	-.07	.05	.09	.16	.26	.44	.12	.17	.20	.40	.25	.32	.19	.18	.63	

Table 3: Inter-relationships of ROM of joint-actions: results of the factor analysis

	Factor 1	Factor 2	Factor 3	Factor 4	Factor 5
Left knee extension	-	.70	-	-	-
Right knee extension	-	.72	-	-	-
Left knee flexion	-	.70	-	-	-
Right knee flexion	-	.64	-	-	-
Left hip flexion	.82	-	-	-	-
Right hip flexion	.74	-	-	-	-
Left hip extension	-	-	-	.88	-
Right hip extension	-	-	-	.93	-
Left hip adduction	-	-	.62	-	-
Right hip adduction	-	-	.87	-	-
Left hip abduction	-	-	-	.44	-
Right hip abduction	-	-	-	.52	-
Left hip internal rotation	.75	-	-	-	-
Right hip internal rotation	.57	-	-	-	-
Left hip external rotation	-	-	-	-	.85
Right hip external rotation	-	-	-	-	.92

Table 4: Pearson correlation coefficients between ROM and disability

	Observed disability			Self-reported disability		
	Total group (n=198)	Hip-OA (n=79)	Knee-OA (n=129)	Total group (n=198)	Hip-OA (n=79)	Knee-OA (n=129)
Knee flexion	-.28**	-.34**	-.23*	-.32**	-.34**	-.29*
Knee extension	-.07	-.16	-.01	-.19	-.20	-.13
Hip flexion	-.25*	-.15	-.26*	-.16	-.09	-.23*
Hip extension	-.35**	-.52**	-.29**	-.34**	-.29*	-.30**
Hip adduction	-.03	-.01	-.05	-.02	-.04	-.08
Hip abduction	-.33**	-.33*	-.36**	-.23*	-.16	-.30**
Hip int. rotation	-.16	-.07	-.22	-.09	-.30*	-.20
Hip ext. rotation	-.31**	-.26	-.23*	-.31**	-.20	-.35**

* $p<0.01$.
** $p<0.001$.

Table 5: Multiple regression analyses. Dependent variable: Observed disability

	Total group (n=198)	Hip-OA only (n=79)	Knee-OA only (n=129)
Fraction variance-accounted-for (r^2)	0.239	0.272	0.232
β			
Knee flexion	-.183**	NS	-.154
Knee extension	NS	NS	NS
Hip flexion	NS	NS	NS
Hip extension	-.313**	-.516**	-.201*
Hip adduction	NS	NS	NS
Hip abduction	NS	NS	-.186
Hip internal rotation	NS	NS	NS
Hip external rotation	-.263**	-.244**	-.224*

NS = Not Significant; variable was not entered into the equation.
* $p<0.05$.
** $p<0.01$.

Table 6: Multiple regression analyses. Dependent variable: Self-reported disability (IRGL)

	Total group (n=198)	Hip-OA only (n=79)	Knee-OA only (n=129)
Fraction variance-accounted-for (r^2)	0.248	0.206	0.255
β			
Knee flexion	-.221**	-.197	-.195*
Knee extension	NS	NS	NS
Hip flexion	NS	NS	NS
Hip extension	-.298**	-.232*	-.279**
Hip adduction	NS	NS	NS
Hip abduction	NS	NS	NS
Hip internal rotation	NS	NS	NS
Hip external rotation	-.256**	-.229*	-.331**

NS = Not Significant; variable was not entered into the equation.
* $p<0.05$.
** $p<0.01$.

The subgroup-analyses presented here were repeated, with exclusion of all ten patients diagnosed with both hip-OA and knee-OA. This yielded results equivalent to those featured in Tables 5 and 6. The results of these additional subgroup-analyses are therefore not presented here.

To test whether the presence of flexion contractures could be responsible for the relationship between joint ROM and disability, the regression analyses were also repeated with two dichotomous (present / absent) variables for flexion contracture of the knee or hip included as potential independent variables. However, the inclusion of flexion contracture as potential determinant of disability did not change the outcome of these analyses; the results were equivalent to those presented in Table 5 and 6.

DISCUSSION

The overall goal of the present study was to assess the relationship between range of joint motion and disability in patients with OA of the hip or knee. To this end, first the inter-relationships of ROM for all joint actions of the knee and hip were established in patients with OA. Clearly, joint ROM cannot be regarded as a unidimensional physical characteristic of patients with OA: close relationships were found for the same joint action on the lateral and contralateral side, but relationships between ROM of different joint actions were much weaker. Because of this result, ROM's of the same joint action on the lateral and contralateral side were added up and used in subsequent analyses. Next, range of joint motion was found to be closely associated with the level of disability in these patients. On average 20%-25% of the variance in disability could be attributed to differences in range of motion. The ROM of three specific joint actions showed the strongest relationship with disability. These were extension and external rotation of the hip and flexion of the knee.

The inter-relationships between the ROM of different joint actions were assessed using both correlational and factor-analyses. The most important finding was the clear inter-relationship found for the same joint action of the lateral and contra-lateral joint. High correlations between identical actions of the lateral and contralateral joint have also been reported by Escalante et al. (10). Next to that, in the factor analysis, the six different joint actions of the hip were clustered into four factors. Extension and abduction loaded on the same factor, while flexion and internal rotation loaded on another factor. The other two factors found for hip-ROM were both loaded by a single joint action of the hip. The clustering of two different joint actions into one factor may be explained by the dependence of both joint actions on the same muscles. A number of muscles are responsible for more than one joint action of the hip, mainly combining flexion or extension with medial or lateral rotation (22).

No major differences were found between patients with hip-OA and knee-OA with regard to relationships of joint ROM with disability. For both subgroups, the results were similar. Extension and external rotation of the hip showed significant relationships with disability in patients with knee-OA. Likewise, flexion of the knee was associated with disability in hip-OA patients. Although there were some (10 in total) patients present in the study population with both types of OA (who were therefore included in both subgroups), these patients were not responsible for the existence of these relationships in this population. The results of the subgroup-analyses did not change significantly when patients with both types of OA were excluded.

Some issues need to be addressed concerning the methodology of this study and its impact on the results presented here. Joint ROM can depend on factors other than articular deformation.. Certain motions may be too painful for patients to complete, or they may lack the muscle strength to maintain the joint action. This results in impaired ROM, without there being a clear articular cause for this impairment. In the present study, the impact of muscle strength on ROM was minimized by determining assisted active ROM. This means that the therapist provided support against the pull of gravity, but no support for the completion of the joint action. The patient had to carry out the motion by himself, using muscle strength to increase the angle, but did not have to use his muscle strength to keep his limb in position. Of course this minimizes, but does not eliminate, the impact of muscle strength on ROM. Neither does this eliminate the influence of pain during motion on ROM, or does it take clinical status of the joint (whether or not it is diagnosed with OA) into account. However, the aim of the present study was to reveal the role of ROM as a determinant of functional disability. Joint ROM has its own determinants, but whether impaired ROM is caused by pain during motion, insufficient muscle strength, or intra- and peri-articular deformation due to OA, is a question beyond the scope of this study. More in-depth research needs to be carried out to provide insight into this complex matter.

To our knowledge, other research on the topic of joint mobility, especially the range of joint motion of specific joint actions in patients with OA, and disability has been scarce. In a population survey, Odding et al. (9) found that restricted ROM of a number of joint actions was associated with the presence of locomotor disability. The strongest relationships were found for flexion of the knee and flexion of the hip. Significant, yet weaker, relationships with disability were also found for internal and external rotation of the hip. This is partially consistent with the present study's findings. Both studies find a relation between the presence of disability and restricted range of joint motion in the sagittal (flexion of the knee and flexion or extension of the hip) and transversal planes (internal and external rotation of the hip). One major difference, however, is that in the present study extension of the hip was found to be closely associated with disability, whereas Odding et al. (9), and also Escalante et al. (10), identified impaired flexion of the hip as a strong risk factor for disability. This may be attributable to differences between the studies in the population under survey: for the present study data were used from patients with OA of the hip or knee, whereas the other two studies were surveys in a general population of elderly. It is possible that the relationship between joint ROM and disability is different in OA, compared to the general population, for instance because of the presence of flexion contractures in patients with OA. Another possibility is that differences between the studies in the protocol for taking ROM measurements (in defining the horizontal, starting positions etc.) have led to different results.

It can be concluded that there is a clear relationship between joint ROM and disability in patients with OA of the knee or hip. However, these relationships do not apply to all joint actions of the knee or hip. Flexion of the knee and extension and external rotation of the hip were found to be most strongly associated with disability. Restricted joint ROM appears to be an important risk factor for the occurrence of locomotor disability in patients with OA.

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6. COPING, PAIN AND DISABILITY IN OSTEOARTHRITIS: A LONGITUDINAL STUDY

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ABSTRACT

Objective: To establish the role of coping styles as prospective determinants of pain and disability in patients with osteoarthritis of the knee or hip.

Methods: Data were used from 71 patients with OA of the hip and 119 patients with OA of the knee. Using regression analysis, relationships were established between the use of active and passive coping styles and the level of pain and disability 36 weeks later

Results: In patients with knee-OA, the passive coping style of resting was found to predict a higher level of disability 36 weeks later after controlling for the baseline level of disability. In the same manner, the active coping style of transforming pain was found to predict higher levels of pain 36 weeks later, also in patients with knee-OA. In patients with hip-OA, no significant relationships between coping styles and pain and disability were found.

Conclusion: The role of resting as a prospective determinant of disability, previously reported in patients with other chronic disorders, could also be established for knee-OA, but not hip-OA. Transforming pain was found to be a risk factor for pain in knee-OA.

Keywords: Coping, Pain, Disability, Osteoarthritis

Pain and physical disability are major symptoms in osteoarthritis (OA), profoundly affecting everyday life of patients. However, the level of articular degeneration, central to the syndrome of osteoarthritis, is only weakly related to the level of pain and disability experienced by patients (1-3).

One of the factors which may influence the level of pain and disability is the way in which patients cope with their chronic condition. Several studies, in patients with various chronic disorders, have shown that patients who use passive coping styles, such as catastrophizing, worrying and avoidance of physical activity report higher levels of pain and disability (3-10). In a longitudinal study in patients with rheumatoid arthritis (RA), Evers et al (11) found that frequent use of passive coping styles predicted a higher level of disability 1 year later. Similar findings were reported by van Lankveld et al. (12), also in RA-patients, and Vlaeyen et al., in patients with chronic low back pain (LBP) (13). The relationship between passive coping styles and pain and disability may be explained through the impact of avoidance of activity on the physical condition of the patient. Patients tend to avoid activity, fearing that activity will result in more pain. However, because of their inactivity, their physical condition deteriorates, resulting in muscle weakness and instability of joints. This, in turn, will lead to more pain and disability (14). With pain and disability increasing, the patient will avoid activity even more, thus entering a downward spiral towards increasing pain and disability.

Less attention has been given to the impact of active coping styles on the level of pain and disability. Positive associations have been reported between the active coping style of transformation of pain (diverting attention from pain) and low levels of pain and disability in RA patients (15). However, in general, studies have focussed on the importance of the use of passive coping styles rather than the use of active coping styles.

The aim of the present study was to establish the role of coping styles as prospective determinants of pain and disability in patients with OA of the knee or hip. It was expected that resting (avoidance of activity) would determine pain and disability: frequent use of resting as a means to cope with OA was expected to result in higher levels of pain and disability.

METHODS

Subjects

Data were used from a randomized clinical trial into the effectiveness of exercise therapy in patients with OA (16). Patients were admissible for the trial if they were diagnosed with OA of the knee or hip according to the classification criteria of the

American College of Rheumatology (17,18). There were 200 patients participating in the trial. Of those patients, ten were diagnosed with both knee-OA and hip-OA. These ten patients were excluded form the analyses presented here. Data of all 190 remaining patients were used in this paper. This included patients from both the intervention and control group of the trial. Data used in the present study were obtained at the onset of the trial (baseline) and at the end of the follow-up period, 36 weeks later.

Coping styles

To assess the deployment of different coping strategies by these patients, three subscales of the Pain Coping Inventory (PCI) were used (19). These three subscales included:

- PCI Resting. This subscale consists of five items, which assess the level to which patients avoid physical activity when in pain ("I cease my activities", "I avoid physical exercise")
- PCI Pain Transformation. This subscale comprises four items which focus on distracting their attention from pain, such as "I pretend the pain is not there" and "I pretend the pain is less severe".
- PCI Lowering Demands. The three items of this subscale assess the extent to which patients lower the demands of their activities ("I continue with less effort", "I continue at a slower pace", and "I continue with less precision").

The PCI Resting subscale determines the level to which this specific passive coping strategy is used, whereas both the PCI Pain Transformation and PCI Lowering Demands subscale assess the utilization of active coping strategies (19). On all three subscales, a higher score means the coping strategy associated with the subscale is utilized more when in pain. The PCI has been shown to be a reliable and valid instrument in different groups of chronic pain patients (19).

Next to the use of different coping strategies, the presence of fear avoidance beliefs about physical activity was determined, using the Fear Avoidance Beliefs Questionnaire (FABQ) (20). The FABQ consists of four statements which can be rated on a 7-point Likert-scale. A higher score means a more pronounced presence of fear avoidance beliefs towards physical activity. The items of the FABQ, which was originally developed for back pain patients, were adapted for use in patients with OA of the hip or knee (21).

All the data of the instruments presented above were obtained at baseline.

Pain and disability

The level of pain and physical disability were assessed at both baseline and the end of the follow-up period, 36 weeks later. Pain was measured with a VAS, range 0-100mm. Patients were asked to rate their overall pain in the past week. Physical disability was measured using an observational method (22). The level of disability was determined by watching videotaped performances on a number of standardized tasks. These tasks included: walking, sitting down into a chair, reclining onto a bed and bending to pick up a weight from the floor. Trained observers assessed the performance of the patients. They scored five items: three movement times (5m-walking time, stand-to-sit time, stand-to-recline time) and two qualitative measures (the level of guarding and the level of rigidity during the performance). Based on these five items, an overall score for observed disability was computed. This overall score has been shown to be internally consistent and valid. For a more detailed description of this method, see Steultjens et al. (22).

Statistical analyses

The role of coping as a determinant of pain and disability in OA was assessed using both bivariate and multivariate techniques. First, Pearson correlation coefficients were computed between the coping measures at baseline on the one hand and pain and disability at follow-up on the other hand.

Next to that, multiple regression analyses were performed, in which pain and disability at follow-up featured as dependent variables. These regression analyses were carried out in three steps. First, two control variables were entered into the regression equation: the baseline value of the dependent variable (either pain or disability) and intervention group. Intervention group was included in the equation to control for the influence of the treatment received within the framework of the trial in which the patients had participated. The baseline value of the dependent variables was taken into account to control for possible systematic differences in the baseline levels of pain and disability between patients utilizing different coping strategies.

In the second step of the regression analyses, clinical and demographic variables were presented to the equation in a stepwise procedure. This included the radiological status of the patients (ROA) (16-18), the duration of complaints as reported by the patient's general practitioner, age, sex and Body Mass Index (BMI; body weight divided by the square of the height). For this stepwise procedure, the inclusion criterion was $p < 0.05$ (p-in = 0.05) and the exclusion criterion was $p > 0.10$ (p-out = 0.10).

In the third and final step, the coping measures (the three subscales of the PCI and the FABQ) were presented to the equation, also in a stepwise procedure. The same in- and exclusion criteria were used as in the previous step of the analysis (p-in = 0.05, p-out = 0.10).

All analyses were performed with two subgroups comprising patients with only hip-OA or patients with only knee-OA. On some patients data were missing on one or more of the variables described above. In the analyses, these missing values were excluded by means of pairwise deletion.

RESULTS

Patient characteristics

Table 1 features the patient characteristics, mean scores for pain and disability and for the coping styles, for both the hip-OA and knee-OA patients. All measurements were taken at baseline, with pain and disability also being assessed at follow-up, 36 weeks later. In general, hip- and knee-patients were rather similar. There were only three significant differences between the patients with hip-OA and knee-OA: on average, patients with hip-OA reported less pain at baseline; patients with hip-OA had a shorter duration of complaints; and patients with hip-OA were less obese (significantly lower BMI).

Correlations between coping styles and pain and disability

Pearson correlation coefficients between the three coping styles and fear avoidance beliefs at baseline on the one hand, and pain and disability at follow-up on the other hand, are given in Table 2. Two significant relationships were identified, both in the group of patients with OA of the knee: resting was associated with higher levels of disability 36 weeks later, and pain transformation was associated with higher levels of pain 36 weeks later.

Table 1: Patient characteristics

	Hip-OA (n=71)		Knee-OA (n=119)	
Sex				
Male	21	29.6%	19	16.0%
Female	50	70.4%	100	84.0%
Age (in years)	68.1 ± 8.5		68.5 ± 8.9	
Body Mass Index (in kg/m²)	27.5 ± 4.0*		28.6 ± 3.9*	
Duration of complaints (in weeks)	74.4 ± 124.7*		118.6 ± 200.6*	
Radiological OA				
Joint space narrowing	44	62.0%	75	63.0%
Osteophytes	56	78.9%	77	64.7%
Trial intervention group				
Experimental	40	49.4%	62	48.1%
Control	41	50.6%	67	51.9%
Observed disability				
Baseline (t0)	0.0 ± 1.0		0.0 ± 1.0	
Week 36 (t1)	-0.2 ± 0.8		-0.1 ± 1.1	
Pain last week (VAS in mm)				
Baseline	40.0 ± 24.6*		48.5 ± 28.1*	
Week 36	33.7 ± 25.5		31.9 ± 30.5	
Coping styles				
PCI: Pain transformation (range: 4-16)	9.1 ± 2.4		9.0 ± 2.8	
PCI: Lowering demands (range: 3-12)	6.3 ± 2.0		6.6 ± 2.0	
PCI: Resting (range: 5-20)	11.4 ± 3.3		11.3 ± 3.1	
Fear Avoidance Beliefs Questionnaire (range: 0-24)	13.2 ± 6.2		14.3 ± 6.5	

* significant difference between hip-OA and knee-OA ($p < .05$).

Table 2: Pearson correlation coefficients between coping measures (at baseline) and pain and disability (at follow-up)

	Hip-OA		Knee-OA	
	Pain	Disability	Pain	Disability
PCI: Pain transformation	.21	-.02	.34*	.20
PCI: Lowering demands	.05	.18	.11	.04
PCI: Resting	.02	.24	.22	.42*
Fear Avoidance Beliefs	.04	.24	-.08	.16

* p <.05.

Table 3: Coping styles (at baseline) as determinants of pain (at follow-up) in OA

	Hip-OA		Knee-OA	
Variance-accounted-for	r^2 = .039		r^2 = .249	
Block 1: method Enter:	β	p	β	p
Control variables				
Pain at baseline	.132	.281	.352	.000
Trial intervention group	-.172	.162	-.139	.096
Block 2: method Stepwise:	β	p	β	p
Demographic variables				
Radiological OA	-	-	-	-
Duration of complaints	-	-	-	-
Age	-	-	-	-
Sex	-	-	-	-
Body Mass Index	-	-	-	-
Block 3: method Stepwise:	β	p	β	p
Coping styles				
PCI: Pain transformation	-	-	.206	.003
PCI: Lowering demands	-	-	-	-
PCI: Resting	-	-	-	-
Fear Avoidance Beliefs	-	-	-	-

- = variable did not meet the inclusion criterion in the stepwise procedure.

Table 4: Coping styles (at baseline) as determinants of disability (at follow-up) in OA

	Hip-OA		Knee-OA	
Variance-accounted-for	r^2 = .329		r^2 = .292	
Block 1: method Enter:	β	p	β	p
Control variables				
Disability at baseline	.570	.000	.381	.000
Trial intervention group	.230	.820	-.116	.163
Block 2: method Stepwise:	β	p	β	p
Demographic variables				
Radiological OA	-	-	-	-
Duration of complaints	-	-	-	-
Age	-	-	-	-
Sex	-	-	-	-
Body Mass Index	-	-	-	-
Block 3: method Stepwise:	β	p	β	p
Coping styles				
PCI: Pain transformation	-	-	-	-
PCI: Lowering demands	-	-	-	-
PCI: Resting	-	-	.219	.024
Fear Avoidance Beliefs	-	-	-	-

- = variable did not meet the inclusion criterion in the stepwise procedure.

Coping as a determinant of pain and disability

The same two relationships that were found in the univariate analyses, were also identified in the multiple regression analyses: in patients with knee-OA, resting predicted a higher level of disability 36 weeks later, and pain transformation predicted a higher level of pain 36 weeks later (see Table 3 and Table 4). No other variables reached significance in the regression analyses, apart form pain and disability at baseline. These variables were entered first into the regression model to control for the baseline level of pain and disability.

For the group of patients with hip-OA, no relationships between coping and pain and disability could be established. In the regression analysis where pain was the dependent variable, however, the active coping style of pain transformation had a p-value of .066, which was just outside the inclusion criterion of p=.05.

DISCUSSION

The aim of the present study was to establish the role of coping as a determinant of pain and disability in patients with OA of the knee or hip. To this aim, the relationships were determined between the use of various coping styles at baseline and the level of pain and disability at follow-up, 36 weeks later. The present study is the first to test these relationships in OA-patients in a *longitudinal* design. It was expected that the passive coping style of resting would predict pain and disability in these patients. This expectation was met by the results in knee-OA. In patients with knee-OA, after controlling for the radiological severity of OA and clinical and demographic variables, resting was a predictor for the level of disability (but not pain) 36 weeks later. The level of pain on follow-up was found to be also dependent on another coping style, pain transformation (diverting attention from pain). In patients with hip-OA, no coping style could be identified which predicted future levels of pain or disability.

The finding that frequent use of resting (passivity) as a coping strategy is a risk factor for high levels of disability in patients with knee-OA, is consistent with several studies among patients with other chronic disorders (11-13). This relationship, previously found in patients with RA and chronic low back pain, has now also been established for patients with knee-OA. The impact of passivity on physical disability can be explained by the effect passivity has on the physical condition of patients. Avoidance of activity is supposed to lead to muscle weakness, which means that there is less potential for muscular control of joints. This results in instability of joints, affecting their ability to carry a load. Instability thus induces physical disability.

It is a well-known fact that the level of articular degeneration of joints is not a prominent factor in explaining the level of physical disability present in patients with knee-OA. The present study confirms this observation. Therefore, other factors need to be taken into account in order to explain the level of disability. The present study demonstrates that the level of disability depends - to a certain extent - on resting: patients who cope with OA by passivity frequently tend to develop a higher level of disability.

In the group of patients with knee-OA, transforming pain was identified as a risk factor for higher future levels of pain. Other studies on the association between active coping and pain, on other groups of chronic pain patients (such as RA or LBP) have mostly reported a beneficial effect of the use of active coping strategies on pain levels (15, 23). An explanation for these contradictory results may be that seeking distraction from pain in itself is beneficial, but that excessive use of this strategy is more detrimental. Excessive use of pain transformation as a coping style may mean ignoring warning signs that the patient is doing too much for the affected joint to bear, thus leading to more serious injury and pain.

Coping could not be established as a determinant of pain and disability in the group of hip-OA patients. Partially, this appears to be caused by insufficient statistical power due to the smaller size of this group (71 patients, as compared to 119 patients in the knee-OA group). In the regression analysis with pain as the dependent variable, using the data from the hip-OA patients, pain transformation did not meet the inclusion criterion by a rather small margin ($p=.066$, with the inclusion criterion at $p=.05$). It is likely that with a slightly larger group (i.e., higher statistical power), the same relationship between the use of pain transformation and pain would have been found as in knee-OA patients. However, the absence in the hip-OA group of the relationship between resting at baseline and disability at follow-up, as found in the knee-OA group, could not be attributed to insufficient statistical power. It has been stressed that determinants of pain and disability in OA, including psychosocial determinants, need not be the same for different types of OA, such as knee-OA and hip-OA (24,25). It is notable that at baseline the patients with hip-OA reported less pain than the patients with knee-OA. Also, the hip-OA group was on average less obese and had suffered from OA-linked complaints for a shorter period than the knee-OA patients. This may indicate that the patients with hip-OA in the present study were on average in a better physical conditions than the knee-OA patients. A better physical condition is a protection against the theoretical pathway through which passivity leads to more disability (14).

An important question is whether the deployment of coping styles by patients can be therapeutically improved (i.e., is it possible to teach patients not to rely on passive coping strategies?). In a longitudinal study, Keefe et al. showed that knee-OA patients who had taken part in coping skills training had less disability and pain than other patients, up to six months follow-up (26,27). Thus, it appears that an intervention aimed at improving coping skills can indeed be effective; such an intervention is able to improve a patient's functional status.

In summary, it can be concluded that the use of specific coping styles predicts future pain and disability in patients with OA. Frequent use of resting (passivity) as a coping style was found to predict higher levels of disability 36 weeks later in patients with knee-OA. Pain transformation was found to predict more pain in knee-OA patients. The latter relationship is likely to exist in patients with hip-OA as well.

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7. AVOIDANCE OF ACTIVITY AND DISABILITY IN PATIENTS WITH OSTEOARTHRITIS OF THE KNEE: THE MEDIATING ROLE OF MUSCLE STRENGTH

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Submitted

ABSTRACT

Objective: Avoidance of activity is hypothesized to lead to muscle weakness and consequently physical disability. This study aimed at validating the avoidance model by establishing the mediating role of muscle weakness in the relationship between avoidance of activity and physical disability in patients with osteoarthritis (OA) of the knee.

Methods: Data were used of 107 patients with OA of the knee. In both a cross-sectional and longitudinal design, a series of regression analyses was performed to establish the mediating role of muscle weakness. Mediation could be established if the effect of avoidance on disability diminished after taking muscle strength into account.

Results: In the cross-sectional analyses, avoidance of activity initially accounted for 15.2% of variance in disability. After taking muscle strength into account, this was lowered to 6.9%. In the longitudinal analyses, 13.9% of variance in disability was accounted for by avoidance of activity. This was reduced to 8.5% when muscle strength was taken into account.

Conclusion: Evidence was obtained for the mediating role of muscle weakness in the relationship between avoidance of activity and disability in patients with knee-OA.

Physical disability is one of the major consequences of osteoarthritis (OA) of the knee (1,2). The way in which patients cope with their condition, especially the passive pain coping style of avoiding physical activity ("resting"), has been found to be associated with a higher level of disability (3,4). When using this coping style, a patient tends to avoid physical activity in order to prevent an increase in pain or onset of a new painful period. In different groups of chronic pain patients, most notably patients with rheumatoid arthritis (5,6) and low back pain (7), the role of avoidance of activity as a determinant of disability has been established. Recently, this relationship has been established also among patients with OA of the knee (8).

It has been proposed that the effect of avoidance of activity on disability can be explained using the conceptual framework of the avoidance model (9). As shown in Figure I, the theory states that a patient tends to avoid physical activity because of a (feared or actual) increase in pain. In the short term, pain can be reduced by avoiding physical activity. In the long term, however, low activity levels will result in a deterioration of physical condition, especially in muscle weakness. Due to this muscle weakness, joints become less stable and the joints' ability to carry a load is reduced. This results in increased disability. Consequently, the patient avoids activity even more, thus entering a downward spiral towards increasing physical disability.

A vital pathway in the model is the relationship between avoidance of activity and disability. According to the model, this relationship is mediated by muscle strength. Baron and Kenny (10) define a mediator as a factor which "represents the generative mechanism through which the focal independent variable is able to influence the dependent variable of interest". In this case, the muscle weakness resulting from inactivity is the mechanism through which the avoidance of activity exerts its influence on the level of physical disability. Establishing the role of muscle strength as a mediator for the relationship between avoidance of activity and disability would therefore be an important step in the validation of the avoidance model.

The general aim of the present study was to assess, in both a cross-sectional and longitudinal design, the validity of the avoidance model in patients with OA of the knee. More specifically, the aim was to establish the mediating role of muscle strength in the relationship between avoidance of activity and physical disability. It was hypothesized that avoidance of activity affects disability through the pathway identified by the avoidance model: avoidance of activity leads to reduced muscle strength, which in turn leads to more physical disability.

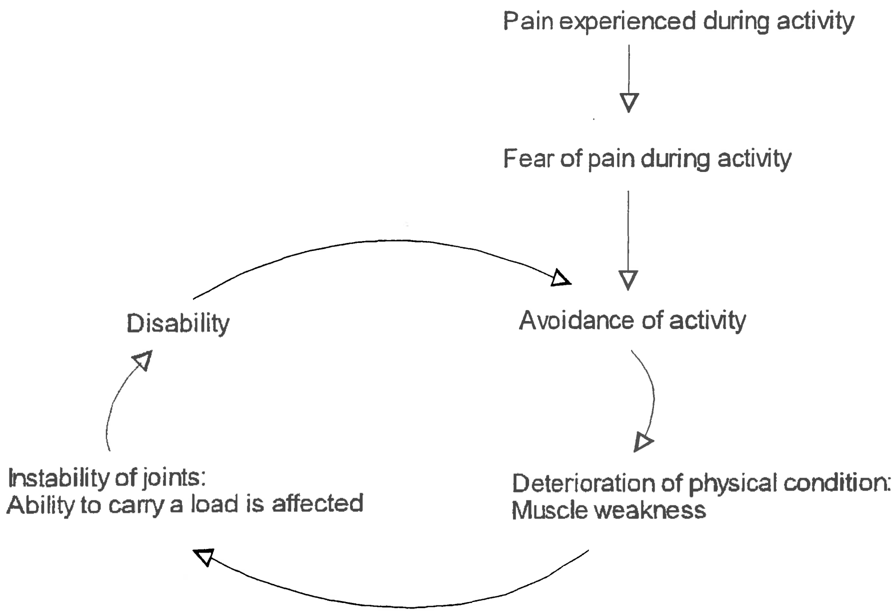

Figure 1: The Avoidance Model

PATIENTS AND METHODS

Patients

Data were obtained from patients with OA of the knee, who participated in a randomized clinical trial into the effectiveness of exercise therapy (11). All patients were diagnosed with knee-OA according to the criteria of the American College of Rheumatology (12). In total, 127 patients with knee-OA participated in the trial. Of those patients, 10 were also diagnosed with OA of the hip. These patients' data were excluded from the analyses presented here. For an additional10 patients data on the measures listed below were missing. These patients were also excluded form the present study, resulting in 107 patients in the analyses. In the present study, data were used from measurements taken directly after conclusion of the intervention period of the trial (which will be referred to as 'baseline' in the present study), and from measurements taken at the final follow-up of the trial, 24 weeks later (which will be referred to as 'follow-up' in the present study).

Measures

Avoidance of activity

The coping style of avoidance of activity was assessed using the Resting subscale of the Pain Coping Inventory (13). This subscale consists of five items, which assess the level to which patients avoid activity when experiencing pain. Items are answered on a 4-point scale, with a higher score indicating a more frequent use of resting as a strategy for coping with pain. The sum of scores on all five items is used as the score for avoidance of activity. A higher score means a more frequent use of avoidance of activity as a coping style. The PCI has been shown to be a reliable and valid measure for pain coping in different groups of chronic pain patients (13). Data on avoidance of activity were collected at baseline.

Disability

Physical disability was measured using an observational method (11, 14). The level of disability was determined by scoring videotaped performances on a number of standardized tasks. These tasks included: walking, sitting down into a chair, reclining onto a bed and bending to pick up a weight from the floor. Trained observers assessed the performance of the patients. They scored five items: three movement times (5m-walking time, stand-to-sit time, stand-to-recline time) and two qualitative measures (the level of guarding and the level of rigidity during the performance). Based on these five items, an overall score for observed disability was computed. This overall score has been shown to be internally consistent and valid. A higher score means a higher level of disability. For a more detailed description of this method, see Steultjens et al. (14). Data on disability were used from both baseline and follow-up.

Muscle strength

Isometric muscle strength was assessed using a hand-held dynamometer (15). Make tests, or "doctor initiated" methods (16), were used. This means that the research assistant holds the dynamometer steady, while the patient exerts maximum force against it (17).Muscle strength in Newton (N) was determined for flexion and extension of the most painful knee. Starting positions were analogous to Kendall et al. (18). The protocol of Kendall et al. provides starting positions for both patient and therapist, and also prescribes the positioning of the dynamometer. In case a patient is unable to adopt a prescribed position, the protocol also provides an alternative position. The tests were carried out by two experienced physical therapists. Prior to the study, the inter-rater reliability was established using 10 healthy subjects and 10 patients with OA. The inter-rater reliability was satisfactory (Pearson's R exceeding 0.75 for both flexion and extension).

These measurements were corrected for body mass by dividing them by the patient's weight. The flexion and extension scores were transformed into z-scores, in order to exclude problems due to different ranges of scores for flexion and extension. The resulting two z-scores per patient were then added up to obtain one sum-score for muscle strength around the affected knee. This has been shown to be a reliable and valid indicator for muscle strength (19). Data on muscle strength were collected at baseline and follow-up. For use in the longitudinal analyses, the average value of muscle strength at baseline and follow-up was calculated, as a measure of the level of muscle strength in the intervening period.

Statistical analyses

To establish whether muscle strength mediates between avoidance of activity and physical disability, regression analyses were performed analogous to Baron and Kenny (10). Baron and Kenny state that to establish mediation, three consecutive regression analyses should be performed (see Figure 2):

1. regress the mediator (muscle strength) on the independent variable (avoidance of activity)
2. regress the dependent variable (disability) on the independent variable (avoidance of activity)
3. regress the dependent variable (disability) on both the mediator (muscle strength) and the independent variable (avoidance of activity)

The mediating role of muscle strength is established if the following criteria are met: avoidance of activity affects muscle strength in analysis #1; avoidance of activity affects disability in analysis #2; muscle strength affects disability in analysis #3; and, in analysis #3, the influence of avoidance of activity on disability must be weaker than in analysis #2.

These analyses were performed in both a cross-sectional and a longitudinal design. In the cross-sectional design, all data used (on both avoidance of activity, disability and muscle strength) were collected at baseline. In the longitudinal design, baseline data on avoidance of activity were used. For muscle strength, the average of the measurements at baseline and follow-up was used. For disability, data collected at follow-up were used. This design does justice to the longitudinal character of the avoidance model (avoidance of activity results in disability over time through the diminishing of muscle strength).

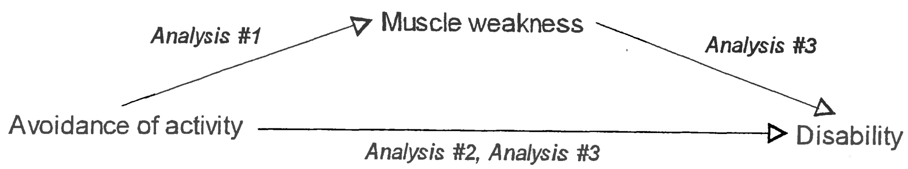

Figure 2: Analytical model for establishing mediation

RESULTS

Patient characteristics and mean scores on avoidance of activity, muscle strength and disability are featured in Table 1.

Table 1: Patient characteristics (n=107)

Sex			
Male		17	16.0%
Female		90	84.0%
Age (in years)		68.7 ± 8.6	
Avoidance of activity			
PCI: Resting (range: 5-20)		10.7 ± 2.9	
Muscle strength (in N/kg)			
Baseline	Flexion	1.2 ± 0.4	
	Extension	2.0 ± 0.7	
Follow-up	Flexion	1.2 ± 0.4	
	Extension	2.0 ± 0.6	
Observed disability (z-score)			
Baseline		0.0 ± 1.0	
Follow-up		-0.1 ± 1.1	

Cross-sectional analyses

The results of the cross-sectional analyses are presented in Table 2. All data used in these analyses were collected at baseline. All criteria for establishing mediation were met. In analysis #1, avoidance of activity was associated with reduced muscle strength. In analysis #2, avoidance of activity was associated with higher disability levels. In analysis #3, reduced muscle strength was associated with more disability, and the impact of avoidance of activity on disability was lowered, compared to analysis #2. The regression coefficient β for avoidance of activity was 0.270, compared to 0.390 in analysis #2. Similarly, in analysis #2, avoidance of activity accounted for 15.2% of variance in disability levels. In analysis #3 the fraction of variance in disability accounted for by avoidance of activity decreased to 6.9%.

Table 2: Establishing mediation in a cross-sectional design

Analysis	Dependent variable	Independent variable(-s)	Regression coefficient (β)	r^2
#1	Muscle strength			0.077
		Avoidance of activity	-0.278 (p=.004)	
#2	Disability			0.152
		Avoidance of activity	0.390 (p=.000)	
#3	Disability			0.265
		Muscle strength	-0.366 (p=.000)	0.196
		Avoidance of activity	0.274 (p=.002)	0.069

Longitudinal analyses

The results of the longitudinal analyses are presented in Table 3. Analogous to the results of the cross-sectional analyses, all criteria for establishing mediation were met. In analysis #1, avoidance of activity at baseline predicted lower average muscle strength from baseline to follow-up. In analysis #2, avoidance of activity at baseline predicted higher levels of disability at follow-up, 24 weeks later. Analysis #3 revealed an effect of reduced muscle strength on disability at follow-up. In this analysis, the effect of avoidance of activity on disability was reduced compared to analysis #2: the β-value for avoidance of activity in analysis #3 was 0.303, compared to 0.372 in analysis #2. Similarly, the fraction of variance in disability accounted for by avoidance of activity went down to 8.5%, from 13.9% in analysis #2.

Table 3: Establishing mediation in a longitudinal design

Analysis	Dependent variable	Independent variable(-s)	Regression coefficient (β)	r^2
#1	Muscle strength			0.079
		Avoidance of activity	-0.281 (p=.003)	
#2	Disability			0.139
		Avoidance of activity	0.372 (p=.000)	
#3	Disability			0.196
		Muscle strength	-0.334 (p=.000)	0.112
		Avoidance of activity	0.303 (p=.001)	0.085

DISCUSSION

The aim of the present study was to establish the role of muscle strength as a mediator in the relationship between avoidance of activity and physical disability in patients with OA of the knee. The mediator function of muscle strength was predicted from the avoidance model, which offers a biobehavioral explanation for disability experienced by chronic pain patients, such as knee-OA patients (9). The model states that patients tend to avoid activity, fearing that activity will lead to an increase in pain. Due to this inactivity, their physical condition worsens, most notably it will lead to muscle weakness. This in turn results in instability of joints, and a decreased ability of joints to carry a load. The end result is increased disability. According to the model, the influence of avoidance on disability is exerted through a reduction of muscle strength, i.e., reduced muscle strength is the mediator for the relationship between avoidance of activity and disability. This mediator hypothesis was tested in both a cross-sectional and a longitudinal design, in the manner proposed by Baron and Kenny (10).

In both designs, evidence was obtained in favour of the mediation hypothesis. Avoidance of activity was found to be a predictor of both muscle weakness and functional disability. When muscle strength was taken into account, the effect of avoidance of activity on disability decreased. This is a clear indication of the mediating role of muscle strength in the relationship between avoidance of activity and disability (10). These results confirm the avoidance model (9)

Although evidence was obtained in favour of the mediating role of muscle strength, it could not completely account for the effect of avoidance of activity on functional disability. In the longitudinal analyses, after controlling for muscle strength, avoidance of activity still accounted for 8.5% of variance in disability levels. Similar findings resulted from the cross-sectional analyses, where avoidance of activity independently from muscle strength, accounted for 6.9% of variance in disability levels. This may indicate that there is a second pathway via which avoidance of activity has an influence on disability. It is possible that inactive patients develop low self-beliefs about their capabilities (low self-efficacy), which is expressed in the inability to perform certain every day tasks (functional disability) without there being clear physical reasons for this inability. Several studies, in different groups of patients, have provided evidence that inactivity is associated with low self-efficacy (20), and that low self-efficacy leads to disability (21-25).

An important issue is the role of pain in the avoidance model. The mechanism through which avoidance of activity leads to disability is initially triggered by pain: patients start avoiding activity because they experience pain. In later stages, fear of pain rather than pain itself keeps fuelling the mechanism. In a study of chronic back pain patients, Crombez et al. found that fear of pain was more disabling than actually experienced pain itself (26). Thus, although pain is not in itself represented in the circular part of the avoidance model (as shown in Figure I), pain does initially play a key role in the avoidance model. Further research should elaborate on the role of pain and fear of pain.

In conclusion, the present study demonstrates that the effect of avoidance of activity on disability can be (partially) explained by the avoidance model: avoidance leads to muscle weakness; which in turn leads to disability.

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8. GENERAL DISCUSSION

In this thesis, a number of studies have been presented in which the role of physical and psychosocial factors as determinants of pain and disability in patients with OA of the knee or hip was established. The aim of this thesis was to establish the way in which avoidance of activity leads to physical disability, as predicted from the theoretical framework of the avoidance model (1). To this aim, a number of research questions were addressed.

The assessment of disability: observations versus self-report

The first research question of this thesis concerned the assessment of physical disability. Disability is normally assessed in one of two ways: by a patient's self-report, or by observations made by a medical professional. Theoretically, observational assessment of disability would provide information on a patient's actual level of functioning, whereas assessment by self-report would reflect a patient's own opinion. Given this theoretical difference in information provided by these two kinds of methods, it has been argued that observations and self-reports measure two different dimensions of disability (2-3). This led to the first research question of this thesis:

When assessing functional disability in patients with OA of the knee or hip, is there a difference in information obtained by observational methods on the one hand and self-report methods (questionnaires) on the other hand?

This question was addressed in two separate studies. In Chapter 2, the construct validity of an observational method for the assessment of functional disability was established. This was done by comparing, in a cross-sectional design, the data obtained using the observational method with data obtained by a number of questionnaires. In Chapter 3 these analyses were repeated using a longitudinal design. The responsiveness of the observational method was assessed by comparing the observed change in disability in a 12-week period to the change in self-reported disability in the same period.

In Chapter 2, observed disability in mobility was found to be clearly associated with self-reported disability in mobility, but not with self-reported dexterity (disability associated with the upper extremities, such as washing and dressing). This meant the convergent validity of the observational method could be established: the information on disability provided by the observational method was similar to information picked up by questionnaires. The divergent validity of the observational method could only be partially established. The data obtained using the observational method were dissimilar from the data on self-reported dexterity, but could not be differentiated from self-reported disability in mobility.

These analyses were repeated in a longitudinal design, to investigate whether observational and self-report methods differ in their responsiveness. This would mean that the two kinds of methods differ in the signs of change in a patient's level of disability that they are able to identify. The analyses with longitudinal data, featured in Chapter 3, were also unable to differentiate between observational and self-report methods. The change in disability identified by the observational method was similar to the change in disability as picked up by the self-report methods. This means that in both a cross-sectional and a longitudinal design, the claim that observational and self-report methods measure different dimensions of disability could not be substantiated. Thus, in general, it is unnecessary to use an extensive observational method when assessing functional disability. This is also consistent with the recommendations of OMERACT, which support the assessment of disability by self-report over observations (4).

Physical factors as determinants of pain and disability in OA of the knee or hip

The second and third research question concerned the role of two physical factors as determinants of pain and functional disability in patients with OA of the knee or hip. Question #2 addressed the use of muscle strength data:

Is it possible to adequately describe the level of muscle strength of a patient with OA of the knee or hip using a single indicator or should a distinction between different (subsets of) muscle actions be maintained?

The role of muscle strength as a determinant of pain and disability in knee-OA and hip-OA has been previously established and extensively described elsewhere (5-8). However, most studies on this topic limited muscle strength to the large muscle groups of the upper leg: m.quadriceps femoris and the hamstrings. These muscle groups are major contributors to most basic activities related to physical disability as it is used in this thesis (i.e., disability related to activities primarily involving the lower extremities, such as walking, stair climbing and reclining). Nevertheless, the question remained whether the strength of these large muscle groups alone could adequately indicate the general level of muscle strength of a patient with knee-OA or hip-OA. The second research question of this thesis aimed at establishing the optimal way of using muscle strength data of OA-patients in statistical analyses. This optimum would represent a trade-off between a parsimonious approach and a detailed approach. In the parsimonious approach, the transparency of statistical analyses, and ease of interpretation of the results is guaranteed by using only one indicator for muscle strength. The aim of the detailed approach is to prevent loss of information from data aggregation by maintaining a high level of segregation, resulting in a large number of separate indicators for muscle strength.

To this aim, first the inter-relationships between the strength of 16 muscle actions around the knees and hips were established. For both patients with knee-OA and hip-OA, all muscle actions were found to be closely associated. Secondly, the relationship between muscle strength and pain and disability was established, using different levels of aggregation for muscle strength data (varying from one patient-level index containing the sum-score of all 16 muscle actions, to an approach with 16 separate indicators, one for each muscle action). In both patients with knee-OA and patients with hip-OA, the level of aggregation of muscle strength data did not affect the strength of the relationship between muscle strength on the one hand and pain and disability on the other hand.

These results, featured in Chapter 4 of this thesis, indicated that any level of aggregation of lower extremity muscle strength data is generally acceptable in patients with knee-OA or hip-OA. This would mean that for reasons of parsimony, using one index for muscle strength, combining data on all muscle actions, would be the preferred option. However, it is a common finding that in patients with OA muscle strength around the affected joint is decreased compared to muscle strength around the contralateral joint. This finding was reproduced in Chapter 4: in patients with knee-OA, muscle strength for flexion and extension of the affected knee was lower than for the contralateral knee. In hip-OA, muscle strength was lower around the affected hip than around the contralateral hip for four of the six muscle actions. Aggregating these muscle strength data to a level where a distinction between affected and unaffected joints is no longer made, did not lead to a weakening of the relationship between muscle strength on the one hand and pain and disability on the other hand. However, the difference in strength between muscles surrounding affected and unaffected joints is an important clinical feature of OA, and is acknowledged as such in medical practice. It was therefore judged that combining muscle strength around affected joints with muscle strength around unaffected joints would lead to an unacceptable loss of vital information. This means that the highest acceptable level of aggregation is to combine the strength of all muscle actions of one joint into an index for muscle strength around that joint.

Given the clear inter-relationships between the strength of different muscle actions, it appears unnecessary to collect extensive strength readings of multiple muscle actions when a global assessment of a patient's muscle strength is made. Measuring the strength of a limited number of muscle actions will adequately reveal the level of muscle strength of a patient. It can therefore be concluded that limiting the assessment of muscle strength in patients with OA of the hip or knee to the major muscle groups of the upper leg does not affect the validity of studies on the role of muscle strength in OA.

A second physical factor which has been proposed as a determinant of disability in knee-OA and hip-OA is range of joint motion (ROM) (6). Contrary to muscle strength, however, only few studies have been published concerning ROM, none of which specifically featured a group of OA-patients (9-11). For this reason, this thesis included a more fundamental research question on the role of ROM as a determinant of disability in knee-OA and hip-OA:

Is impaired range of joint motion a determinant of physical disability in patients with OA of the hip or knee?

To address this research question, two separate, but related, issues featured in Chapter 5 of this thesis. First, the inter-relationships between ROM of different joint actions were established, analogous to research question #2 of this thesis, concerning muscle strength. It was found that there were no clear inter-relationships between ROM of different joint actions, other than the relationship between the same action of the lateral and contralateral joint. This meant that, contrary to the findings concerning muscle strength, it was not possible to adequately represent a patient's level of joint ROM by aggregating data on ROM of several joint actions. Given the close inter-relationships between ROM of the same joint action of the lateral and contralateral joint, combining the ROM scores of the two joints into one score for ROM per joint action was the only acceptable option for data-reduction.

In the analyses concerning muscle strength, it was found that the strength of all muscle actions, of different joints, were closely inter-related. Using ROM-data, only the same joint action of the lateral and contralateral joint were found to be closely inter-related. So, apparently muscle strength can be regarded a general characteristic of a patient, whereas ROM is a local characteristic of the joint. This difference may be explained through the factors which influence muscle strength and ROM. Possibly, the level of muscle strength depends primarily on factors linked to a patient's overall physical condition, which means different muscles will be similarly affected by these factors. On the other hand, joint ROM may be mainly determined by the anatomy and other characteristics of the joint, and other structures responsible for controlling joint movement (tendons, ligaments etc.). The clear inter-relationships between ROM of the same action of the lateral and contralateral joint indicate that such factors will in general exert their influence symmetrically (i.e., the effect on ROM will be similar in both joints).

The second issue addressed in Chapter 5 aimed at determining the role of joint ROM as a determinant of disability in patients with OA of the knee or hip. In both patients with hip-OA and patients with knee-OA, ROM of extension and external rotation of the hip were found to be the two predominant determinants of disability. In patients with knee-OA, flexion of the knee was also found to be a determinant of disability, together with abduction of the hip. This implies that, regardless of the precise location of OA (hip or knee), impaired ROM of certain joint actions of the hip is an important determinant of disability. This may be explained by the fact that in many basic physical activities, movement of the hip plays a central role, more so than movement of the knee joint. This could explain why, even in patients with knee-OA, ROM of the hip is more closely related to disability than knee-ROM.

No attempt was made to establish the role of joint ROM as a determinant of pain. There is no theoretical basis for the existence of such a relationship between ROM and pain. On the contrary, pain may be an important determinant of joint ROM. Joint motion can induce pain, which forces the patient to stop moving before the full ROM (as determined by the anatomical lay-out of the joint) is reached. In this way, the actual joint ROM is determined by the maximum joint angle the patient can adopt in a painless (or not too painful) manner.

In Chapter 4 it was found that muscle strength, on average, accounted for close to 20% of variance in disability in OA-patients. In Chapter 5, joint ROM accounted for some 25% of variance in disability. However, this does not mean it can be concluded that, together, these two physical factors account for nearly 45% of variance in disability in patients with OA. It is possible that muscle strength partially determines joint ROM. If a patient lacks the muscle strength to complete his movement, this will be reflected in a low ROM. The method used to measure ROM in this thesis aims at minimizing the impact of muscle strength on ROM (12), but does not completely eliminate it. It is therefore possible that, after controlling for muscle strength, the impact of joint ROM on disability (as expressed in the variance in disability accounted for by joint ROM) will be somewhat decreased, compared to the results in Chapter 5.

The analyses presented in Chapter 4 and Chapter 5, concerning the role of muscle strength and joint ROM as determinants of pain and disability in OA, were all cross-sectional analyses. Both the muscle strength and ROM readings, and the data on pain and disability, were collected simultaneously. Strictly speaking, it was therefore not possible to identify a causal relationship between muscle weakness and increased pain and disability, and between impaired ROM and disability. These physical factors were only found to be closely associated with pain (muscle strength) and disability (muscle strength, ROM). However, the most likely explanation for these associations is that muscle weakness induces pain and disability, and that impaired ROM leads to disability.

It was therefore concluded that two physical factors are major determinants of physical disability in patients with knee-OA or hip-OA: muscle strength in general, and ROM of specific joint actions of the hip. In patients with knee-OA, ROM of flexion of the knee is also a determinant of physical disability. Muscle strength can also be regarded as a determinant of pain in patients with OA..

Avoidance of activity as a determinant of pain and disability in OA

The fourth and fifth research question of this thesis addressed the role of avoidance of activity as a determinant of pain and disability in patients with OA of the knee or hip. According to the theoretical framework of the avoidance model, avoidance of activity will lead to increased disability. First, avoiding activity results in a deterioration of physical condition, expressed in muscle weakness. Due to muscle weakness, the stability of joints is affected. This instability of joints means that patients will be less able to carry out basic activities, since the ability of the joint to carry a load is reduced. The aim of this thesis was to establish the validity of this pathway in the avoidance model (see Figure 1 in Chapter 1).

Before the avoidance model could be studied in depth, first the role of avoidance of activity as a determinant of disability in OA had to be established:

Is the passive coping style of avoidance of activity a determinant of pain and disability in patients with OA of the knee or hip?

The role of avoidance of activity as a determinant of disability was explored in Chapter 6 of this thesis. To assess the relative importance of avoidance of activity, the analyses also featured two other coping styles: pain transformation (distracting attention from pain) and lowering demands (continuing with activity with reduced intensity when in pain). These are both active coping styles, meaning more rational and oriented towards problem solving, whereas avoidance of activity is a passive, problem-avoiding, coping style.

In patients with OA of the knee, avoidance of activity was identified as a determinant of functional disability. Avoiding physical activity at baseline predicted more disability 36 weeks later. This finding was consistent with a number of other studies, in other groups of chronic pain patients (13-15). However, this relationship between avoidance and disability could not be identified in patients with OA of the hip. It has been stated that OA of the hip and OA of the knee are two distinctly different conditions, both with their own specific set of associated problems (16). It is therefore not unlikely that there is a difference between the two conditions in the factors that determine disability. Alternatively, the strength of the relationship between disability and a specific determinant of disability may be different in hip-OA and knee-OA. This would explain the difference between the two patient groups in the results found.

A second option is that the two subgroups of patients with OA of the hip and patients with OA of the knee in the study population differed on other characteristics, which could have influenced the observed relationship between avoidance of activity and disability differentially. There were some significant differences between the hip-OA and knee-OA group in this study: the patients with OA of the knee were on average more obese, had a longer duration of complaints linked to OA, and initially reported more pain than patients with hip-OA. There was no significant difference between the groups in the frequency of use of avoidance of activity as a means of coping with OA, nor in the level of disability. The observed differences could indicate that on average the patients with OA of the hip were in a better physical condition than the knee-OA patients. As a better physical condition is a protection against disability, this could mean the relationship between avoidance of activity and disability was weaker in this group of hip-OA patients than in the patients with knee-OA present in the study population.

Based on the results presented in Chapter 6, it would be premature to conclude that there is a significant difference between patients with OA of the knee and patients with OA of the hip in the way coping styles affect the level of disability. Additional research will have to provide insight into differences and similarities between the consequences of different types of OA.

The fourth research question also covered the role of avoidance of activity as a determinant of pain. A few studies have reported a relationship between avoidance of activity and pain (17-21). Theoretically, avoidance of activity could cause pain by a similar pathway as described in the avoidance model for the relationship between avoidance of activity and disability. Avoidance results in muscle weakness, which leads to instability of joints. The instability of the joint may cause strain in innervated tissues near the joint, leading to pain (1).

However, the role of avoidance of activity as a determinant of pain in patients with OA of the knee or hip could not be established. Apparently, the deterioration of physical condition, as hypothesized to result from avoiding activity, is not clearly expressed in an increase in pain.

In patients with OA of the knee, it was found that transforming pain predicted higher levels of pain 36 weeks later. In patients with OA of the hip, this relationship could not be established. However, this appeared to be caused by an insufficient number of hip-OA patients in the analyses to detect this relationship (insufficient statistical power). The margin by which pain transformation failed the inclusion criterion in the regression analysis into determinants of pain in hip-OA was very small (p-value 0.066, inclusion criterion: $p \leq 0.05$). It is therefore likely that the relationship between pain transformation and pain, as found in patients with OA of the knee, did also exist in the smaller group of patients with hip-OA.

It was not expected that frequent use of pain transformation would be associated with increased pain in patients with OA. In general, the use of active coping strategies is regarded to be beneficial (22). It is possible that the use of active coping styles in itself is beneficial. In utilizing active coping styles, the patient attempts to minimize the consequences of his disease. However, at a certain point this may result in ignoring warning signs that the patient exceeds the maximum load the affected joint is able to bear. This could ultimately lead to a considerable increase in pain. This propagates the existence of a curvilinear relationship between pain transformation and pain. Initially, transforming pain is beneficial and reduces pain, but excessive use of pain transformation as a means of coping with pain, results in more pain.

The role of avoidance of activity as a determinant of disability could be established in patients with OA of the knee. This meant the fifth research question of this thesis could be addressed in this group:

If avoidance of activity is a determinant of disability in patients with OA of the knee, is this relationship mediated by muscle weakness?

Addressing this research question would provide insight in the mechanism through which avoidance of activity leads to disability, as predicted by the avoidance model (1): avoidance results in a deterioration of physical condition, especially muscle weakness. Muscle weakness leads to instability of joints, which restricts the ability of the joint to carry a load, resulting in physical disability. In this model, muscle strength is the mediator of the relationship between avoidance of activity and disability. A mediator is defined as a factor which "represents the generative mechanism through which the focal independent variable is able to influence the dependent variable of interest" (23). In Chapter 7, both cross-sectional and longitudinal analyses were performed to establish this mechanism. These analyses comprised a series of three consecutive regression analyses. To establish the mediating role of muscle strength, a number of criteria had to be met. First of all, avoidance of activity had to affect muscle strength. Next, avoidance also had to have an effect on disability. The most important criterion was that the effect of avoidance of activity on disability would decrease when muscle strength was taken into account. In both the cross-sectional and longitudinal analyses, these criteria were met. Avoidance of activity initially accounted for 15.2% of variance in disability in the cross-sectional analyses. After controlling for muscle strength, this decreased to 6.9%. In the longitudinal analyses, Avoidance of activity at baseline accounted for 13.9% of variance in disability 24 weeks later. This fell to 8.5% when controlling for the average level of muscle strength in this period. It could therefore be concluded that muscle strength is a mediator of the relationship between avoidance of activity and disability in patients with OA of the knee.

The analyses presented in Chapter 7 did not include data on the instability of joints. In the avoidance model, instability of joints is caused by muscle weakness and directly precedes disability. However, data on the level of instability of joints were not available. Therefore, the role of joint instability could not be addressed in this thesis. It is questionable whether instability of joints is entirely responsible for the impact of muscle weakness on disability. It can be argued that insufficient muscle strength in itself causes disability. Muscle weakness may prevent the execution of certain tasks (e.g., lifting a weight) and limits endurance, thereby inducing disability.

The mechanism as described in the avoidance model could not completely account for the relationship between avoidance of activity and disability. When controlling for muscle strength, avoidance of activity still accounted for 6.9% of disability in the cross-sectional analyses, and 8.5% of disability in the longitudinal analyses. This implies that there may be another pathway in which avoidance affects disability. There is some evidence that physical inactivity is associated with low self-beliefs about physical capability (low self-efficacy) (24). It has been frequently reported that low self-efficacy is associated with a higher level of disability (25-29). The patient's low self-efficacy would then be expressed in functional disability, while the patient's physical condition does not justify a high level of disability.

Pain is an important factor in the avoidance model. The onset of avoidance of activity can be attributed to pain experiences during activity. Since activity is consequently evaded, it is not pain actually experienced during activity, but fear of pain which will cause low activity levels in these patients. It has been reported that fear of pain is more disabling than pain itself (30). In this manner, pain and fear of pain do play a key role in the avoidance model. However, more research needs to be carried out to establish the precise impact of pain and pain-related fear.

In this thesis, a parsimonious version of the avoidance model was presented, in which the featured relationships were limited to those directly related to the manner in which avoidance of activity affects disability (see Figure 1 in Chapter 1). It is likely that there will be more relationships between the factors than featured in this version of the model. For instance, disability may lead to more pain, and in turn pain may have a direct effect on disability. Including these relationships in the model would do more justice to the complexity of reality. On the other hand, this would diminish the transparency of the model with regard to the relationships under study in this thesis. Therefore, it was decided to limit the description of the avoidance model to those relationships directly linked to the way in which avoidance of activity develops, and leads to increased physical disability.

Implications for the therapeutical management of OA

The studies presented in this thesis have some implications for the therapeutical management of OA. The previously reported finding that the level of articular degradation of joints is not strongly related to the level of pain and disability (6-8) has been confirmed in Chapter 6 of this thesis. This implies that in the therapeutical management of OA, next to attempting to control the level of articular degradation, other goals should be pursued in order to improve a patient's health status. In addition, efforts should focus on an integration of pain control, improving physical fitness and discouraging maladaptive coping.

Pain is the predominant feature of symptomatic OA, and is hypothesized to play a key role in the deterioration of a patient's physical condition. Patients avoid activity out of fear for pain, based on earlier experience. Nevertheless, the importance of physical fitness in the prevention of pain and disability has been clearly established. Even though physical activity could initially induce some pain, physical activity reduces pain and disability in the long term.

Given the negative short-term effect of activity on pain, isolated physical activity may only strengthen the patient's belief that activity should be avoided. Frequent physical activity, preferably in a controlled manner (e.g., through exercise therapy) should therefore be combined with active pain control, for instance through the use of medication. In this way, the negative immediate effect of physical activity on pain can be minimized, while maintaining the beneficial long term effect.

An important aspect of therapeutical physical activity is that it should not induce additional damage to the joint. This means that during physical exercise, strain on the joint should be minimized. This can be accomplished by focussing on the use of isometric muscle strength exercises. An isometrically contracting muscle does not generate an external pull on the joint, thereby limiting the negative consequences of physical activity to the joint.

Integrating physical activity with active pain control may also modify the patient's belief that activity results in pain and that activity should therefore be avoided. Next to that, the use of passive, maladaptive coping strategies may also be actively challenged. Keefe et al obtained promising results in patients with OA of the knee, using a coping skills training program (31). These patients reported lower levels of disability than other patients, up to six months after completion of the program. By simultaneously challenging both pain, avoidance of activity and muscle weakness, the vicious circle resulting in increased disability as depicted by the avoidance model, could be broken.

Methodological issues

An important methodological issue in the thesis was the assessment of instrument responsiveness. In Chapter 3, the responsiveness of an observational method for the assessment of disability in mobility was established. To this aim, longitudinal data obtained with the observational method were directly compared to longitudinal data provided by self-report methods, using factor analysis.

This analytical approach mirrors the assessment of construct validity in cross-sectional studies, such as performed in Chapter 2 of this thesis. The notion of viewing instrument responsiveness as longitudinal construct validity was originally put forth by Deyo et al. (32). However, in recent years the design of studies into responsiveness has focussed on the use of a Gold Standard (GS), to which the tests of interest could be compared (33). If a true GS (i.e., a test which classifies all patients correctly as improved or non-improved) is available, this is the preferred option. However, there is no true GS available for physical disability. Using as a GS an indicator for disability which does not adequately reflect the change in the level of physical disability over a period of time, diminishes the validity of these studies into instrument responsiveness. For the test of interest, to equal the imperfect assessment of the GS will be seen as the best possible outcome. Any disagreement between the GS and the test of interest will be regarded as a lack of responsiveness in the test of interest, while it is possible that this instrument is actually more responsive than the GS.

A commonly used GS for change in physical disability is global perceived effect: a patient is asked to globally rate the change in his health status during the period under study, often in a single item format. This creates additional problems for the assessment of the responsiveness of the test of interest. Firstly, there is no one-on-one relationship between global perceive effect and a change in disability. A patient's health status will be profoundly affected by other factors than the level of disability, such as pain, the medical management of the disease and the patient's behavioural patterns. This invalidates global health status as an indicator for physical disability. Secondly, it has been argued that directly asking a patient to rate change will result in an unreliable estimate. Patients will have to compare their present status with the past, and quantify the difference between the two moments. It has been questioned whether patients are able to do this with sufficient precision (34,35).

Another problem is specifically related to the single-item format which is often used to assess global perceived effect and similar measures. It can be argued that such an indicator lacks the discriminative power needed to correctly differentiate between improved and non-improved patients. An example of the insufficient discriminative power of single-item indicators may also be present in Chapter 2 of this thesis. One of the self-report methods featured in Chapter 2 was the mobility subscale of the EuroQoL questionnaire, which has a single item with three response categories. The EuroQoL mobility subscale correlated poorly with the other questionnaires on disability. This was probably due to the lack of variance in the study population with regard to the responses on the EuroQoL mobility subscale, compared to the other questionnaires.

Given the reservations mentioned above, comparison with a GS was judged to be an inappropriate approach to establishing the responsiveness of disability measures. Instead, it was decided to directly compare the observational method and questionnaires to each other. This would establish the longitudinal construct validity of the observational method. A possible criticism of this approach is, that information that a change in disability is present in the study population is not available. If an appropriate GS is available, it provides information on the magnitude of change present in the study population. When directly comparing tests of interest, as in Chapter 3 of this thesis, there is no external information on the magnitude of change in the study population. Theoretically, this could mean that instrument responsiveness is studied in the absence of change, which would undermine the usefulness of this approach. Any "change" in a patient's level of disability as recorded by any of the instruments would then merely be random variation (noise), and a comparison between the different methods would only be based on this noise. However, it is unlikely that a consistent set of relationships can emerge from analyses performed using change scores purely based on random variation. Even when beforehand the magnitude of change in disability is not known, it is therefore possible to directly compare change scores of different methods for establishing disability, in order to assess their responsiveness. This line of reasoning reflects the way in which the construct validity of an instrument is established in cross-sectional studies. Construct validity can be established by identifying relationships between instruments, which were theoretically predicted to exist, without having a priori information on the signal-to-noise ratio (reliability) and validity of all separate instruments involved in the analyses.

The relatively dominant presence of random variation in change scores does present a problem in research into instrument responsiveness, however. A change score is the result of a subtraction. The subtraction of two measurements of equal magnitude (such as a baseline and follow-up measurement of disability) results in a relatively small true value with a double amount of measurement error. This results in a low signal-to-noise ratio. Due to a low signal-to-noise ratio, existing associations between instruments will be obscured. This is notable in the correlation matrix featured in Table 2 of Chapter 3. The correlations between the change scores of the various instruments are very low, especially when compared to the cross-sectional correlations between the same instruments as featured in Table 3 of Chapter 2.

All studies presented in this thesis were based on secondary analyses on data collected in a randomized clinical trial (RCT) into the effectiveness of exercise therapy in patients with OA of the knee or hip (36). The context in which these data were collected, complicated the design of a number of analyses presented in this thesis, in particular the longitudinal analyses featured in Chapter 6 and 7.

The main complicating factor was the intervention applied in the trial. During the first 12 weeks of the registration period of the trial, the experimental group received exercise therapy, which was specifically aimed at improving physical fitness (muscle strength, range of motion, stability of joints and aerobic capacity) and reducing pain and disability (37). This presented a major interference with the natural course of the consequences of OA, potentially obscuring the normal effect of avoidance of activity and muscle weakness on pain and disability.

This problem was dealt with in two separate ways. In Chapter 6, regression analyses were presented aimed at establishing the role of coping styles as determinants of pain and disability. To this aim, the effect of the use of coping styles at baseline on disability 36 weeks later was assessed. The intervention period of the trial coincided with the first 12 weeks of this 36-week period between baseline and follow-up. To control for the effect of the intervention on disability, a dichotomous variable for intervention group (experimental or control) was included in the regression equation. Intervention group did not have a significant effect on pain or disability in any of the regression analyses featured in Chapter 6. It can therefore be concluded that this intervention did not interfere with the relationships between coping styles and disability, as identified in these analyses.

In Chapter 7, the role of muscle strength as a mediator in the relationship between avoidance of activity and disability was assessed using a series of regression analyses. Including a control variable for intervention groups, analogous to Chapter 6, in each of these regression analyses would have heavily affected the transparency of these analyses. It was therefore decided to limit these analyses to a 24-week period, excluding the 12 weeks of the intervention period. The assessments of avoidance of activity, muscle strength and disability made directly after conclusion of the intervention period served as the baseline values in these analyses. In this way, the trial interventions did not affect these analyses.

However, it is possible that participating in the trial did make patients more aware of the importance of physical fitness in the prevention of pain and disability, compared to the general population of patients with OA. In this way, the trial may have had an indirect effect on the results presented in this thesis. Such an effect would have resulted in weakening of the relationship between avoidance of activity, muscle weakness and disability, however, since muscle weakness would be less common in these patients. This would have obscured rather than highlighted the relationship between avoidance of activity and disability. It can therefore be concluded that this does not invalidate the finding that avoidance of activity affects disability through muscle weakness.

Implications for future research

A number of recommendations for future research into the consequences of OA can be given, based on the experiences gained from the studies featured in this thesis. These recommendations address various issues related to the design of consecutive research projects.

The selection of patients will be a key issue in the design of consecutive studies. First of all, attention will have to be paid to the inclusion of a sufficient number of patients. This does not only apply to the size of the entire study population, but in particular to subgroups of patients with specific types of OA. In Chapter 6 of this thesis, the role of the active coping style of pain transformation as a determinant of pain could not be established in patients with OA of the hip. It is highly likely that this relationship did exist in this group of patients, but could not be revealed due to the insufficient number of patients with hip-OA in the study.

A second aspect of patient selection is the duration of OA-linked complaints in the selected patients. Ideally, the study population would consist of recently diagnosed patients. In the study population of this thesis, the average duration of complaints was more than one year. In this year, most patients will have made the transition from a relatively healthy, pain-free and non-disabled state to the average state of substantial pain and moderate disability presented throughout this thesis. This first year of complaints may especially be a crucial year for the initiation of the process described in the avoidance model. In the first year, many patients will start to experience pain during activity, and will begin avoiding activity as a result. It will be more difficult to reveal this process in patients who have been diagnosed with OA for a considerable period, because some significant changes will already have taken place in the first stages of OA.

A topic closely linked to the selection of early patients is the monitoring of change. More than determining a patient's state at a given moment, it will be of paramount importance to gain insight in the course of the patient's situation.

The assessment of change is complicated by a number of factors. Firstly, when monitored over a prolonged period of time, in all probability the change in a patient's health status will not be linear. Neglecting the non-linearity of change may obscure the processes responsible for temporal variations in health status, thus compromising the identification of determinants of health status and the mechanisms responsible for health status changes.

If change is assumed to be non-linear, the assessment of change requires the availability of longitudinal data from a number of measurements, collected over a prolonged period. Having these data available means the pattern of fluctuations in health status can be assessed more precisely. Most probably, financial and practical constraints will limit the number of repeated measurements, and extend the period in-between two measurements.

In order to be able to reliably assess change in health status, it is of vital importance that the instruments used are responsive. It has been stated that disease-specific instruments are more responsive than generic measures (38). On the other hand, using generic measures will enhance the external validity (i.e., the possibility of generalising the results to other patient groups than the study population) of the study. Responsive generic instruments should therefore be considered for inclusion in the study. However, the assessment of instrument responsiveness remains one of the most complicated topics in clinimetrics, as shown in Chapter 3 of this thesis. This can mainly be attributed to the absence of gold standards for most vital characteristics of health status, such as physical disability. Therefore, the identification of responsive instruments, most suited to picking up change, will require additional research.

Finally, modelling non-linear change, and establishing determinants of non-linear change, will require the use of sophisticated statistical techniques, such as multi-level analysis for repeated measures (39).

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SUMMARY

The aim of this thesis was to identify determinants of pain and functional disability in patients with osteoarthritis (OA) of the knee or hip. In particular, the objective was to establish the role of avoidance of activity as a determinant of pain and disability in OA, and to identify the mechanism through which avoidance of activity results in disability. The supposed mechanism is provided by the avoidance model. Initially, patients with OA experience pain during activity. As a result, patients will fear that more physical activity will lead to more pain, and will therefore avoid physical activity. In the short term, avoiding activity can lead to a reduction in pain. In the long term, however, a lack of physical activity results in a deterioration of the patient's physical condition, especially in muscle weakness. Muscle weakness affects the stability of joints, reducing the ability of the joints to carry a load. This leads to functional disability. Consequently, the patient avoids activity even more, thus entering a downward spiral towards increasing disability.

Before the role of avoidance of activity as a determinant of pain and disability, and the validity of the avoidance model, could be studied, some issues had to be resolved concerning the assessment of two other factors present in the avoidance model: disability and muscle strength. Next to that, the role of range of joint motion (ROM) as a determinant of disability was also studied. Joint ROM is not a factor in the avoidance model. However, impaired ROM is regarded an important physical determinant of disability in OA, although this relationship has rarely been studied in depth. Therefore, a study on the role of ROM as a determinant of disability in OA was included in this thesis.

The following research questions were addressed in this thesis:

1. When assessing functional disability in patients with knee-OA or hip-OA, is there a difference in information obtained by observational methods on the one hand and self-report methods on the other hand?
2. Is it possible to adequately describe the level of muscle strength of a patient with OA of the knee or hip using a single indicator, or should a distinction between different (subsets of) muscle actions be maintained?
3. Is impaired range of joint motion a determinant of physical disability in patients with OA of the hip or knee?
4. Is the passive coping style of avoidance of physical activity a determinant of pain and disability in patients with OA of the knee or hip?

5. If avoidance of activity is a determinant of disability in patients with OA of the hip or knee, is this relationship mediated by muscle weakness?

The first research question was addressed in Chapter 2 and Chapter 3 of this thesis.

In Chapter 2, the internal consistency and validity of an observational method for assessing disability in mobility was established. To this aim, for 198 patients with hip-OA or knee-OA, results of the observational method were compared with results of self-report methods (questionnaires). The analyses revealed that the observational method was internally consistent. Using factor analysis, observed and self-reported disability in mobility were found to be closely associated and could not be differentiated. This established the convergent validity of the observational method, but did not establish the divergent validity of the observational method (observations and self-report appeared to yield largely equivalent information).

The aim of Chapter 3 was to establish the responsiveness of observational and self-report methods for the assessment of disability in mobility. Of 186 patients with OA of the knee or hip, data on disability were collected on two occasions with a 12-week interval, using one observational method and four self-report methods. Correlations and factor analysis were used to establish the relationship among changes in these five methods. It was found that the inter-correlations between changes in questionnaires were not substantially higher than the correlations between the observational method on the one hand and the self-report methods on the other hand. In the factor analysis, both the observational method and self-report methods loaded on the same factor. In Chapter 3, no evidence was therefore obtained for the differential responsiveness of observational and self-report methods.

In a cross-sectional design, information obtained using the observational method could not be differentiated from information on disability provided by questionnaires (Chapter 2). In a longitudinal design, a difference in responsiveness between the observational method on the one hand and self-report methods on the other hand could not be established. These results raise questions regarding the simultaneous use of both observational and self-report methods for the assessment of disability in patients with OA. Due to the advantages of questionnaires (easier to use, less time-consuming, less of a burden to patients), this implies that the use of self-report methods is to be preferred over observational methods.

Chapter 4 addressed the use of muscle strength data in statistical analyses (research question #2). Reduced muscle strength is regarded a risk factor for pain and disability in osteoarthritis. It was unclear whether muscle strength could be represented by a single index at the level of the patient (i.e., all muscle actions can be aggregated into a single indicator for a patient's level of muscle strength), or whether the strength of different muscle actions should be seen as separate entities, with their own specific impact on a patient's level of functioning. This meant the optimal trade-off between parsimony (as few separate indicators as possible) and detail (no loss of vital information) had to be established.

Isometric muscle strength was measured for 16 muscle actions around the knees and hips in 52 patients with OA of the hip and 70 patients with OA of the knee. Various indices of muscle strength were derived from these measurements, applying five alternative approaches. These approaches ranged from a single overall index to a set of 16 separate indices. First, the internal consistency of these indices was determined. It was found that the internal consistency was satisfactory for all indices (Cronbach's $\alpha > 0.74$). Next to that, it was determined to what extent these indices could reveal the association between reduced muscle strength on the one hand and pain and disability on the other hand. As expected, reduced muscle strength was associated with increased disability, but no clear relationship could be established between muscle weakness and pain. The strength of these associations did not depend on the approach used to derive the indices for muscle strength.

The indices did not show major differences with regard to internal consistency or the extent to which the association with pain and disability could be revealed. For reasons of parsimony, approaches resulting in few indices appeared to be most useful. However, muscle strength was found to be significantly reduced around affected joints, compared to muscle strength around unaffected joints. Therefore, the most suitable approach for reducing muscle strength data into indices was judged to be an approach that results in as few indices as possible, but with separate indices for muscle strength around affected and unaffected joints.

The role of joint ROM as a determinant of disability in OA (research question #3) was addressed in Chapter 5. Data on ROM of 16 joint actions of the knee and hip were collected in 198 patients with OA of the knee or hip. First, the inter-relationships between the ROM of different joint actions were assessed by calculating correlation coefficients and performing factor analysis. Second, the relationship between ROM and physical disability was established using regression analyses.

Range of motion of the same joint action of the lateral and contralateral joint were found to be closely associated. No other clear inter-relationship between ROM of joint actions could be established. In both patients with OA of the hip and patients with OA of the knee, ROM of extension and external rotation of the hip were found to be most closely associated with disability.

Chapter 6 looked into the fourth research question of this thesis. The aim was to establish the role of coping styles, in particular the passive coping style of avoidance of activity as prospective determinants of pain and disability in patients with osteoarthritis of the knee or hip. Data were used from 71 patients with OA of the hip and 119 patients with OA of the knee. Using regression analysis, relationships were established between the use of coping styles and the level of pain and disability 36 weeks later.

In patients with knee-OA, avoidance of activity was found to predict a higher level of disability 36 weeks later after controlling for the baseline level of disability. In the same manner, the active coping style of transforming pain was found to predict higher levels of pain 36 weeks later, also in patients with knee-OA. In patients with hip-OA, no significant relationships between coping styles and pain and disability were found.

In this way, the role of avoidance of activity as a prospective determinant of disability, previously reported in patients with other chronic disorders, could also be established for knee-OA, but not hip-OA.

Given the results of Chapter 6, the fifth research question, regarding the mechanism through which avoidance of activity leads to disability, could be addressed in patients with OA of the knee. This was done in Chapter 7. The aim was to establish the mediating role of muscle weakness in the relationship between avoidance of activity and physical disability, as predicted by the avoidance model. Data were used of 107 patients with OA of the knee. In both a cross-sectional and longitudinal design, a series of regression analyses was performed to establish the mediating role of muscle weakness. Mediation could be established if the effect of avoidance on disability diminished after taking muscle strength into account.

In the cross-sectional analyses, avoidance of activity initially accounted for 15.2% of variance in disability. After taking muscle strength into account, this decreased to 6.9%. In the longitudinal analyses, 13.9% of variance in disability was accounted for by avoidance of activity. This was reduced to 8.5% when muscle strength was taken into account. In this manner, evidence was obtained for the mediating role of muscle weakness in the relationship between avoidance of activity and disability in patients with knee-OA.

A general discussion of the results presented in this thesis was provided in Chapter 8. This chapter featured an extensive discussion of the methodology used to assess instrument responsiveness in Chapter 3. Also, the differences between muscle strength and range of joint motion as determinants of disability in OA were discussed. A third topic was the implications of the results presented in Chapter 6 and 7 for the validity of the avoidance model. Some implications for the therapeutical management of OA were presented. It was concluded that therapy would benefit from an integration of active pain control, improvement of physical fitness and the discouragement of maladaptive coping.

All analyses presented in this thesis were secondary analyses on data collected previously. The consequences of analysing existing data for the design of the studies presented in this thesis was also discussed. Finally, some implications for future research were given. These recommendations focussed on the selection of early patients. Also, the importance of monitoring change was stressed. The difficulties associated with the monitoring of change were discussed.

SAMENVATTING

In dit proefschrift is onderzoek beschreven naar determinanten van pijn en functionele beperkingen in patiënten met artrose van de knie (gonartrose) of heup (coxartrose). In het bijzonder was het doel van dit onderzoek het bepalen van de rol van het vermijden van fysieke inspanning als determinant van pijn en beperkingen, en om het mechanisme bloot te leggen dat verantwoordelijk is voor de invloed van het vermijden van fysieke activiteit op de ernst van functionele beperkingen. Het veronderstelde mechanisme wordt beschreven in het vermijdingsmodel. In het begin zullen patiënten met artrose pijn ondervinden tijdens fysieke activiteit. Hierdoor ontstaat de angst dat meer fysieke activiteit zal leiden tot meer pijn; patiënten zullen hierdoor fysieke inspanning gaan vermijden. Op de korte termijn kan dit vermijdingsgedrag een reductie in pijn opleveren. Op de lange termijn daarentegen, zal een gebrek aan fysieke inspanning een afname in de lichamelijke conditie tot gevolg hebben, die zich met name uit in spierzwakte. Spierzwakte tast de stabiliteit van gewrichten aan, waardoor de belastbaarheid van de gewrichten vermindert. Dit leidt tot functionele beperkingen. Als gevolg hiervan zal de patiënt fysieke activiteit nog sterker gaan vermijden. Op deze manier ontstaat een neerwaartse spiraal met toenemende functionele beperkingen.
Voordat de rol van vermijding van inspanning als determinant van pijn en beperkingen, en daaraan gekoppeld de validiteit van het vermijdingsmodel, kon worden onderzocht, diende eerst bepaald te worden op welke manier omgegaan moet worden met twee andere factoren in het vermijdingsmodel: spierkracht en functionele beperkingen. Daarnaast werd ook onderzocht wat de rol van gewrichtsmobiliteit (de maximale bewegingsuitslag van gewrichten) is bij functionele beperkingen in patiënten met artrose. Gewrichtsmobiliteit is geen factor in het vermijdingsmodel. Wel wordt deze factor algemeen gezien als een belangrijke determinant van functionele beperkingen. Desondanks is hier zelden onderzoek naar verricht. Om deze reden is een studie naar het verband tussen gewrichtsmobiliteit en functionele beperkingen in dit proefschrift opgenomen.

De volgende vijf onderzoeksvragen kwamen aan bod in dit proefschrift:

1. Is er verschil tussen observatiemethodes en vragenlijsten in verkregen informatie bij het bepalen van functionele beperkingen in patiënten met artrose van de heup of knie?
2. Is het mogelijk om de spierkracht van een patiënt met artrose van heup of knie adequaat te beschrijven door middel van een enkelvoudige index, of is het nodig om onderscheid te maken tussen verschillende (subsets van) bewegingsrichtingen?
3. Is een verminderde gewrichtsmobiliteit een determinant van functionele beperkingen in patiënten met artrose van heup of knie?
4. Is het frequent toepassen van de passieve coping-strategie “vermijden van lichamelijke inspanning” een determinant van pijn en functionele beperkingen in patiënten met artrose van heup of knie?
5. Als het zo is dat het vermijden van lichamelijke inspanning een determinant van functionele beperkingen is, is spierzwakte dan de mediator in deze relatie?

De eerste onderzoeksvraag was onderwerp van hoofdstuk 2 en 3 van dit proefschrift.

Het doel van hoofdstuk 2 was om de interne consistentie en validiteit te bepalen van een observatiemethode voor functionele beperkingen. hiertoe werden bij 198 patiënten met artrose van heup of knie de resultaten van de observatiemethode vergeleken met de resultaten van een aantal vragenlijsten. De observatiemethode bleek intern consistent te zijn. Uit een factor analyse bleek dat de resultaten van de observatiemethode nauw samenhingen met de resultaten van vragenlijsten; op grond van de resultaten kon geen systematische differentiatie tussen de beide soorten instrumenten aangebracht worden. Hiermee kon wel de convergente validiteit van de observatiemethode worden vastgesteld, maar niet de divergente validiteit. Geconcludeerd werd dat observatiemethodes en vragenlijsten grotendeels gelijke informatie opleveren.

Het doel van hoofdstuk 3 was om de responsiviteit van de observatiemethode en vier vragenlijsten te bepalen. Bij 186 patiënten werden twee maal, met een tussenperiode van 12 weken, data met betrekking tot functionele beperkingen verzameld. Met behulp van correlaties en factor analyse werden de relaties tussen de vijf instrumenten met betrekking tot de geregistreerde veranderingen (verschilscores) in functionele beperkingen bepaald. De samenhang tussen de verschilscores van de vragenlijsten onderling was niet groter dan de samenhang tussen de verschilscore van de observatiemethode enerzijds en de vragenlijsten anderzijds. Ook in de factor analyse werd geen systematische divergentie tussen de twee soorten instrumenten gevonden. In hoofdstuk 3 werd dientengevolge geen bewijs gevonden voor differentiële responsiviteit van observatiemethodes en vragenlijsten.

In een cross-sectioneel design bleek het niet mogelijk een differentiatie aan te brengen tussen informatie verkregen door observaties en informatie verkregen door vragenlijsten (hoofdstuk 2). In een longitudinaal design kon geen systematisch verschil in responsiviteit gevonden worden tussen observaties enerzijds en vragenlijsten anderzijds. Deze bevindingen plaatsen vraagtekens by het simultaan gebruiken van zowel observatiemethodes als vragenlijsten voor het bepalen van functionele beperkingen in patiënten met artrose. Gezien de voordelen van vragenlijsten (gebruiksgemak, minder tijdsintensief, minder belastend voor patiënten) dienen deze de voorkeur te krijgen boven observatiemethodes.

Hoofdstuk 4 behandelde het gebruik van spierkracht-data in statistische analyses (onderzoeksvraag 2). Spierzwakte wordt beschouwd als een risicofactor voor pijn en beperkingen in artrose. Het was onduidelijk of spierkracht adequaat weergegeven kan worden door middel van een enkelvoudige index (d.w.z., spierkracht voor alle bewegingsrichtingen wordt samengevoegd tot één indicator voor spierkracht op het niveau van de patiënt), of dat onderscheid gemaakt dient te worden tussen verschillende bewegingsrichtingen, die elk een afzonderlijke invloed hebben op het functioneren van de patiënt.

Isometrische spierkracht werd gemeten voor 16 bewegingsrichtingen rond de knie en heup in 52 patiënten met artrose van de heup en 70 patiënten met artrose van de knie. Verschillende indicatoren voor spierkracht werden berekend, volgens vijf verschillende benaderingen. Deze benaderingen varieerden van een enkelvoudige indicator gebaseerd op spierkracht in alle 16 bewegingsrichtingen tot een set van 16 afzonderlijke indices. Allereerst werd de interne consistentie van deze indices bepaald. Deze was voldoende voor alle indices (Cronbach's $\alpha > 0.74$). Daarnaast werd onderzocht in hoeverre deze indices in staat waren de relatie tussen spierkracht enerzijds en pijn en beperkingen anderzijds bloot te leggen. Volgens verwachting bleek spierzwakte geassocieerd te zijn met functionele beperkingen. Daarentegen kon geen duidelijke relatie worden vastgesteld tussen spierzwakte en pijn. De sterkte van deze associaties was niet afhankelijk van de gekozen benadering voor het construeren van de indicatoren voor spierkracht.

Er waren geen duidelijke verschillen tussen de indices met betrekking tot de interne consistentie of de associatie met pijn en functionele beperkingen. Uit oogpunt van spaarzaamheid bleken benaderingen die resulteren in zo weinig mogelijk indices het meest bruikbaar. Er werd echter een significant verschil in spierkracht gevonden tussen bewegingsrichtingen rond door artrose aangedane gewrichten enerzijds en bewegingsrichtingen rond niet-aangedane gewrichten anderzijds. Daarom werd geconcludeerd dat een benadering die resulteert in zo weinig mogelijk indicatoren, maar die wel onderscheid maakt tussen aangedane en niet-aangedane gewrichten, de voorkeur verdient.

De rol van gewrichtsmobiliteit als determinant van functionele beperkingen in artrose kwam aan bod in hoofdstuk 5. Data betreffende gewrichtsmobiliteit werden verzameld voor 16 bewegingsrichtingen van de heup en knie bij 198 patiënten met artrose. De samenhang tussen de gewrichtsmobiliteit in de 16 bewegingsrichtingen werd bepaald met behulp van correlaties en factor analyse. Daarnaast werd via regressie analyse de relatie tussen gewrichtsmobiliteit en functionele beperkingen bepaald.

Identieke bewegingsrichtingen van het laterale en contralaterale gewricht bleken nauw met elkaar samen te hangen met betrekking tot mobiliteit. Behoudens deze bevindingen werden geen andere duidelijke associaties tussen verschillende bewegingsrichtingen gevonden. In zowel patiënten met artrose van de knie als patiënten met artrose van de heup bleken extensie en exorotatie van de heup het sterkst samen te hangen met functionele beperkingen.

Hoofdstuk 6 betrof de vierde onderzoeksvraag van dit proefschrift. Het doel van dit hoofdstuk was om de rol van coping strategieën, en met name de passieve coping strategie "vermijden van fysieke activiteit", als determinanten van pijn en functionele beperkingen in artrose. Hoervoor werden gegevens gebruikt van 71 patiënten met artrose van de heup en 119 patiënten met artrose van de knie. Door middel van regressie analyse werden de relaties tussen het gebruik van coping strategieën enerzijds en het niveau van pijn en beperkingen 36 weken later anderzijds vastgesteld.

In patiënten met artrose van de knie werd gevonden dat het vermijden van fysieke activiteit een voorspeller was voor het verslechteren van de functionele beperkingen 36 weken later. Eveneens in patiënten met artrose van de knie werd gevonden dat het transformeren van pijn aanleiding gaf tot meer pijn na 36 weken. In patiënten met artrose van de heup werden geen significante relaties gevonden tussen coping strategieën en pijn en beperkingen.

De rol van vermijden van fysieke inspanning als determinant van functionele beperkingen, zoals eerder gevonden in andere groepen chronische pijn patiënten, werd hiermee ook vastgesteld in patiënten met artrose van de knie, maar niet in patiënten met artrose van de heup.

Gezien de resultaten van hoofdstuk 6 kon de laatste onderzoeksvraag, betreffende het mechanisme waardoor vermijden van activiteit leidt tot functionele beperkingen, bestudeerd worden in patiënten met artrose van de knie. Deze studie werd uitgevoerd in hoofdstuk 7. Het doel was hier om de rol van spierzwakte als mediator in de relatie tussen het vermijden van activiteit en functionele beperkingen, zoals geformuleerd in het vermijdingsmodel, vast te stellen. Hiervoor werden data gebruikt van 107 patiënten. Een serie regressie analyses werd uitgevoerd in zowel een cross-sectioneel als longitudinaal design. De mediërende rol van spierzwakte kon worden vastgesteld als het effect van het vermijden van activiteit op functionele beperkingen afnam wanneer spierzwakte in het analytisch model werd opgenomen.

In de cross-sectionele analyses verklaarde het vermijden van activiteit in eerste instantie 15,2% van de variantie in beperkingen. Nadat de invloed van spierzwakte was verrekend, daalde dit tot 6,9%. In de longitudinale analyses reduceerde het includeren van spierzwakte in het verklaringsmodel het percentage door vermijding verklaarde variantie in beperkingen van 13,9% tot 8,5%. Hiermee werd bewijs verkregen dat spierzwakte inderdaad de mediator is in de relatie tussen het vermijden van fysieke inspanning en functionele beperkingen in artrose van de knie.

Een algemene discussie van de resultaten van dit proefschrift werd gegeven in hoofdstuk 8. Dit hoofdstuk bevatte een uitgebreide discussie van de in hoofdstuk 3 gehanteerde methodologie voor het bepalen van de responsiviteit van meetinstrumenten. Daarnaast werden de verschillen tussen spierkracht en gewrichtsmobiliteit besproken met betrekking tot de rol van beide factoren als determinanten van functionele beperkingen. Een derde onderwerp was de implicaties van de in hoofdstuk 6 en 7 gepresenteerde resultaten voor de validiteit van het vermijdingsmodel. Ook werden enkele implicaties voor de therapeutische zorg aan artrosepatiënten gegeven. Geconcludeerd werd dat een integratie van actieve pijnbestrijding, verbetering van de fysieke conditie en het ontmoedigen van passieve coping strategieën de basis voor therapie zou kunnen verzorgen.

Alle in dit proefschrift gepresenteerde analyses betroffen secundaire analyses op eerder verzamelde data. De consequenties van het gebruiken van bestaande data voor de opzet van de in dit proefschrift gepresenteerde studies werden besproken. Als laatste werden enkele aanbevelingen voor vervolgonderzoek gegeven. Deze aanbevelingen richtten zich vooral op de selectie van recent gediagnosticeerde patiënten. Daarnaast werd het belang van het monitoren van verandering benadrukt.

DANKWOORD

De jongeman die in september 1990, gedreven door zijn fascinatie voor de mens als machine, naar Limburg trok om bewegingswetenschapper te worden, kon niet bevroeden dat dit tien jaar later zou leiden tot een proefschrift met prominent op de omslag de ietwat versleten en verroeste schokdemper van zijn oude Suzuki motorfiets als symbool voor artrose. Dat het zover is gekomen, is natuurlijk deels te wijten aan die ene verklarende factor waar wetenschappers allergisch voor zijn: het toeval. Daarnaast is dit boek het resultaat van grote inspanningen en kleine duwtjes in de rug van velen.

Bovenaan de lijst staat Joost. In 1996 kwam ik op het Nivel binnen als jouw wetenschappelijk assistent, volledig onwetend van je verborgen agenda. Een klein jaar "wat secundaire analyses" doen, leek me een prima start van een carrière als onderzoeker, dan kon ik ondertussen eens rondkijken voor ergens een promotieplaats wellicht. Dat laatste bleek dus al snel onnodig. Afgezien van een eenmalige reis naar Amsterdam om jouw geleende toga weer bij de pedel in te leveren, kenmerkte de baan zich door veel wetenschap en weinig assistent. Op het gebied van onderzoek doen en, vooral, het opschrijven van de bevindingen heb ik zeer veel van je opgestoken. Dat onze samenwerking altijd in een prettige sfeer is verlopen is de slagroom op de taart. Ik ben dan ook blij dat ons eerste "buiten-proefschriftelijke" artikel alweer in de steigers staat.

Hans, als tweede promotor heb je een cruciale bijdrage geleverd aan de totstandkoming van dit proefschrift. Onze gesprekken waren niet zo talrijk, maar inhoudelijk waren ze van groot belang voor de inhoud van de diverse hoofdstukken. Een van je andere promovendi bedankt je in haar eigen proefschrift voor je "scherpe opmerkingen en ideeën", en daar wil ik me van harte bij aansluiten.

De resultaten in dit proefschrift zijn geheel afkomstig uit secundaire analyses op data verzameld tijdens een trial naar de effectiviteit van oefentherapie bij artrose. Bij het opzetten en uitvoeren van de hier beschreven studies verkeerde ik dus in de grote luxe dat de benodigde data reeds beschikbaar waren. Margriet, niet alleen de gegevens, maar ook je nauwgezette verslaglegging, hebben me veel geholpen. Om nog niet te spreken van het feit dat je geruime tijd als kamergenoot de ideale vraagbaak was op een meter afstand. En kijk! Mijn boekje is ook af!

Leo Roorda en Rob Oostendorp zijn betrokken geweest bij de totstandkoming van delen van dit proefschrift. Mijn dank voor jullie bijdragen en nuttig commentaar op conceptversies van artikelen.

Het belang van duwtjes in de rug moet niet onderschat worden. En in zekere zin werd het eerste duwtje gegeven ruim een jaar voordat ik aan dit proefschrift begon te werken. Zonder mijn werkervaring op het AZU was ik wellicht nooit op het Nivel terechtgekomen; en was dientengevolge dit proefschrift er nooit geweest. Esther, bedankt dat je me toen, in 1995, die kans bood. Het heeft zelfs nog een gezamenlijke Franstalige publicatie opgeleverd. Inmiddels lijken onze loopbanen op curieuze wijze met elkaar verweven. Dezelfde werkgever, dezelfde promotor. Over een paar jaar sta je weer naast je kleine broertje op de academische ladder. Tot die tijd is het hard werken. Mocht je in nood komen, dan weet je in ieder geval waar je je lastige kroost even kunt dumpen.
Morele steun kan elke promovendus goed gebruiken. Mijn dank gaat uit naar kamergenoten Marion en Esmée, "de kroeg in met" Ger & Harald, andere collega's van het Nivel, met name in het themagebied Paramedische Zorg en het Projectteam NS2, collega's van RIVM-CZO (met een speciaal bedankje aan Gert Westert voor het adequaat runnen van een dagopvang voor Flexwet-slachtoffers), en alle anderen, van naaste familie tot hele vage kennissen, die belangstellend hebben geïnformeerd naar de voortgang van dit boek. Een enkele keer uit een misplaatste neiging te willen aanschurken tegen de bijbehorende titel wellicht, maar meestal met oprechte interesse; soms is het nodig om je te realiseren dat het schrijven van een proefschrift minder vanzelfsprekend is dan het binnen de muren van een wetenschappelijk instituut lijkt.
Als cynische grap ten koste van de moegeschreven promovendus wint op het allerlaatst vorm het toch nog van inhoud. Richard, bedankt voor je hulp bij het vormgeven van de buitenkant van dit boek. Marina, bedankt voor het uit handen nemen van de vervelendste klus: het lay-outen van de binnenkant.
Lang heb ik getwijfeld over het al dan niet opnemen van dit laatste bedankje. Het is min of meer traditie om het op deze plaats te zetten (en "before every thinking man stands a questioning woman", qua parafraseren). Maar verdient iemand die afwassen en het huis schilderen oneindig veel belangrijker vindt dan het schrijven van een boek vol abstract theoretisch geklets het eigenlijk wel om hier bedankt te worden? Vooruit maar. Bedankt, hè, gij daor.

CURRICULUM VITAE

Martijn Steultjens werd geboren op 4 april 1972 te Eersel, alwaar hij achttien jaar later op het plaatselijke Rythoviuscollege het VWO afrondde. Daarop startte hij in 1990 met de studie Gezondheidswetenschappen aan de toenmalige Rijksuniversiteit Limburg, de huidige Universiteit van Maastricht. Na het behalen van het propedeutisch examen in 1991 vervolgde hij deze studie met de afstudeerrichting Bewegingswetenschappen, waarin hij in 1996 het doctoraalexamen behaalde.
Sinds september 1996 is hij als onderzoeker werkzaam op het Nederlands Instituut voor Onderzoek van de Gezondheidszorg (Nivel) te Utrecht. Naast het in dit proefschrift opgenomen onderzoek heeft hij daar diverse studies verricht op het gebied van paramedische zorg en, recentelijk, huisartsenzorg.
Deze aanstelling bij het Nivel werd in 1999 drie maanden onderbroken door een aanstelling als onderzoeker bij het Centrum voor Zorgonderzoek (CZO) van het Rijksinstituut voor Volksgezondheid en Milieu (RIVM) te Bilthoven. Daar heeft hij onderzoek gedaan naar regionale verschillen in de incidentie van ziekenhuisopnames.