Exploring functional limitations in people with traumatic and nontraumatic neck pain

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Martijn Stenneberg

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Colophon

Exploring functional limitations in people with traumatic and nontraumatic neck pain
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**Exploring functional limitations in people
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PROEFSCHRIFT

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Voor mijn moeder

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Chapter 1

GENERAL INTRODUCTION

Neck Pain

Epidemiology

Neck pain is a common musculoskeletal disorder affecting all age groups of the general population, and is an important reason for consulting general practitioners and physiotherapists [1–6]. The 1-year prevalence of neck pain ranges from 30 to 50% throughout the world, with a point prevalence of about 14% [3,4,7–9]. The prevalence of neck pain is higher in high-income countries [7,9]. Neck pain, along with low back pain and shoulder pain [10], is one of the most prevalent musculoskeletal disorders in The Netherlands. In the general population, about 20% to 70% of the adults will experience neck pain at some time during their lives, and prevalence is highest during middle age [4,7–9,11]. Women have a higher risk of neck pain [3,8]. Over the last 10 years, the global prevalence of neck pain of more than 3 months duration has increased by 21%, and it is expected that, with people living longer [3,12], incidence will increase even further over the coming decades.

A substantial part of people with neck pain develops chronic pain and disability [13–15]. Compared to other global diseases, neck pain is one of the leading causes of disability around the world [15–17], and low back and neck pain combined is the first primary cause of disability [15,18]. Years lived with disability caused by neck pain increased by 21% over the last 10 years [12,15]. Neck pain is also a disabling condition in workers, and the economic burden due to neck pain is high. It represents one of the most costly conditions in industrialized countries, with medical, rehabilitation and lost productivity costs all running high [6,19,20].

Neck pain is the most commonly presenting musculoskeletal disorder in physiotherapy practice in The Netherlands [5]. Individuals with neck pain make up approximately 9.5% of people receiving primary care physiotherapy [5]. Given the growing impact of neck pain and the adverse effects on quality of life and ability to work as well as the social economic burden that comes with it, it is recommended that neck pain should be one of the priorities for research on prevention and therapy [12].

Definition

Neck pain can be defined as “an unpleasant sensory and emotional experience, associated with actual or potential tissue damage in the neck region, which starts at the superior nuchal line and continues down to the level of the scapular spine” [21–23]. Neck pain is a subjective, personal experience [22]. The pain can originate

from anatomical structures in the cervical region, such as zygapophyseal joints, vertebrae, muscles, ligaments, neural structures, and the intervertebral disc [24]. There is, however, no conclusive evidence regarding a specific underlying disease causing the pain. Consequently, most cases are labelled as non-specific neck pain. It develops gradually and the onset may be insidious or may be brought on by trauma. It is a complex biopsychosocial disorder with physical and psychological symptoms [24,25].

People with non-specific neck pain represent a heterogeneous population experiencing a variety of clinical symptoms. The main symptoms associated with neck pain are: pain and stiffness in cervical region, restriction of the range of motion, lower cervical strength, proprioceptive deficits, impaired postural control, headache, dizziness, and radiating pain to the occiput, nuchal muscles, shoulders and upper limbs [26–28]. Neck pain is also associated with other musculoskeletal health problems such as low back pain and headache, poorer self-rated health, and different types of mental functions such as anxiety and depression [27]. This makes neck pain a multifactorial disorder.

The Bone and Joint Decade 2000–2010 Task Force on Neck Pain divides neck pain into 4 grades [22]: Grade I neck pain and associated disorders with no signs suggestive of major pathology and no or little interference with activities of daily living; Grade II neck pain with no signs of major pathology, but interference with daily activities; Grade III neck pain with no signs of major pathology, but presence of neurologic signs of nerve compression such as decreased deep tendon reflexes, weakness, or sensory deficits; Grade IV neck pain with signs of major structural pathology.

Traumatic neck pain

Traumatic neck pain, often referred to as Whiplash-associated Disorders, is the term given to the variety of symptoms affecting the neck, that are triggered by an accident with an acceleration–deceleration mechanism such as a car accident [29]. Whiplash can be defined as “an acceleration–deceleration mechanism of energy transfer to the neck. It may be the result of rear-end or side-impact motor vehicle collisions, but can also occur during diving or other mishaps. The impact may result in bony or soft-tissue injuries (whiplash-injury), which in turn may lead to a variety of clinical manifestations called Whiplash-Associated Disorders” [29]. Traumatic neck pain is associated with a wider range of symptoms as compared to nontraumatic neck pain, including paresthesia/anesthesia in the upper

quadrant, visual and auditory disturbances, sleeping problems, and cognitive and emotional problems [27,28].

People with traumatic neck pain are regarded as a subgroup of non-specific neck pain [29–32]. However, it is unclear as to whether this distinction is relevant as some studies found no clinically relevant differences between the two neck pain conditions [33–35]. Contrary, others studies demonstrated statistically significant higher levels of pain, impairments, disability and psychological distress, and poorer prognosis in people with traumatic neck pain when compared to people with nontraumatic neck pain [36–40].

For traumatic neck pain, the most used classification system is the classification system proposed by the Quebec Task Force on Whiplash Associated Disorders [29]. This widely used classification system categorizes neck pain which occurs after a traffic collision based on the severity of signs and symptoms, into grades 0 to 4: Grade 0 WAD with no complaint about neck pain, no physical sign(s); Grade 1 WAD with neck pain, stiffness, or tenderness, with no physical sign(s); Grade 2 WAD with neck pain, stiffness, or tenderness with musculoskeletal sign(s); Grade 3 WAD with neck pain, stiffness, or tenderness with neurologic signs; Grade 4 WAD with neck fracture or dislocation.

It is suggested that different mechanisms may underlie nontraumatic neck pain and traumatic neck pain and it is encouraged to distinguish between them including different classification systems and clinical management [30,31]. However, there is a lack of high-quality evidence to determine whether people with nontraumatic neck pain and with traumatic neck pain are indeed all that different in terms of type and severity of impairments, disability, and psychological factors. This information is important for clinical management, practice guidelines and research. Should we distinguish subgroups, provide specific diagnostic and/or treatment recommendations? Or can we treat these patients as a homogeneous group and allow statistical pooling in systematic reviews?

Risk factors

There are several risk factors associated with the onset of neck pain. Most strong and consistent risk factors for developing neck pain include a previous history of neck complaints or trauma, low back pain, poor psychological health and being female [4,41,42]. There is limited evidence that old(er) age, high physically demanding jobs, low social/ work support, and smoking constitute higher risks [4,41,42].

Clinical course and prognosis

The clinical course of neck pain is generally poor [13]. About 30% of people with neck pain will develop chronic symptoms, and 50% to 85% of all people who experience neck pain at some initial point, will report neck pain 1 to 5 years later [14,43]. Because of this, a complete resolution of symptoms is less common as neck pain follows a “recurrent” or “episodic” course marked by remissions and exacerbations [7,14,44]. A study performed in general practice in The Netherlands showed that at one year, 76% of the patients felt fully recovered or much improved, even though 47% still reported neck pain [45]. About 45% of people with neck pain show a prompt decrease in pain and disability in the first 6.5 weeks, but show limited improvement after that [46].

In people with traumatic neck pain, pain and disability mainly reduce in the initial three 3 months following the accident, and approximately 50% continues to experience symptoms 1 year after the onset [44,47].

In people with neck pain the course of pain and disability is individual and variable. Poor health, prior neck pain or other musculoskeletal disorders, poor psychological health, worrying, and passive coping are associated with poor prognosis [13,14]. In people with traumatic neck pain, high pain intensity, high self-reported disability, high posttraumatic stress symptoms, strong catastrophic beliefs, and cold hyperalgesia are associated with poor recovery [13]. However, the level of certainty of these reported prognostic factors is low to moderate.

Physiotherapy assessment

Clinical examination enables the physiotherapist to assemble information about the patient's specific health problem in order to set a physiotherapeutic diagnosis, provide a prognosis, guide treatment and measure outcome [48]. To consider all various aspects of the patient's health problem, physiotherapists must select appropriate examination procedures as part of the clinical reasoning process. They are expected to choose valid, reliable and clinically applicable measurement instruments for specific purposes [48,49]. Clinically applicable measurement instruments should be inexpensive and easy to perform, both for patients and physiotherapists. Several instruments are available to measure neck-related impairments, disability and psychological factors [50–52]. However, although widely used clinically, the clinimetric properties of these instruments have often not been adequately evaluated. Advanced technologies are often not available

within a primary care physiotherapy setting due to high costs and limited applicability. Clinimetric properties of questionnaires used for assessing activity limitations and participation restrictions in people with traumatic neck pain, for example, have been studied in heterogeneous populations of people with nontraumatic neck pain [53,54]. Their content validity in a population of people with traumatic neck pain has not been established yet, while there are indications that most recommended condition specific questionnaires have limited content validity [53,54]. A possible reason is that these questionnaires omit a substantial part of the specific limitations that these patients experience [55,56]. In that case, these questionnaires would not be suitable to assess the specific limitations of people with traumatic neck pain.

Further, active cervical range of motion is the most routinely collected function by physiotherapists. Nevertheless, very few clinical tests for assessing active cervical range of motion have been evaluated in people with neck pain. Moreover, cervical kinematics are complex [57–59]. Next to planar range of motion of the primary movements (flexion-extension, lateral bending and rotation) being reduced in people with neck pain, 3-dimensional coupling between cervical rotation, lateral bending, and flexion-extension is affected as well [60,61]. Yet, no clinically applicable instrument is available to evaluate these 3-dimensional coupling motions.

The International Classification of Functioning, Disability and Health (ICF)

As neck pain is considered a multifactorial disorder, it requires insights into the functioning and disability of people with neck pain in clinically relevant domains and their interrelationship. The ICF is a classification of health and health-related domains that visualizes the interactions among the various components of functioning and health [62,63]. It is the most used classification system in physiotherapy health care, and is organized in two parts:

- *Functioning and disability.* This part consists of (1) body functions and structures, and (2) activities and participation.
- *Contextual factors.* This part consists of (1) environmental factors and (2) personal factors.

Each ICF component can be expressed in both positive and negative terms. The framework can be used for measuring health and disability at both individual and population levels [62]. However, describing the relationship between the

individual constructs of the ICF is only possible if the measurement instruments used (mostly questionnaires), measure the constructs purely, while not simultaneously measuring other constructs of health.

Aims and outline of this thesis

The general aims of this thesis were to describe the clinical characteristics of two types of neck pain presenting in primary physiotherapy care, namely nontraumatic neck pain and traumatic neck pain, and to develop and evaluate clinically applicable measurement instruments for assessing impairments and limitations in people with nontraumatic neck pain and traumatic neck pain. In this thesis two new conditioned measurement instruments are developed and evaluated. Subsequently, the clinical characteristics – in terms of type and severity of pain, impairments, disability, and mental factors – and complexity of these types of neck pain are presented and discussed.

As active cervical range of motion is the most frequently assessed impairment by physiotherapists, we conducted a large meta-analysis to quantify differences in active cervical range of motion between people with neck pain and those without neck pain, in people with nontraumatic neck pain and traumatic neck pain, and in people with acute symptoms versus those with chronic symptoms (**Chapter 2**).

Chapter 3 reports the development of a condition-specific self-assessment questionnaire for people with traumatic neck pain that measures activity limitations and participation restrictions, as defined by the ICF framework. In **chapter 4**, the measurement properties of this questionnaire in a population of people with sub-acute and chronic traumatic neck pain are described. **Chapter 5** describes the concurrent validity and interrater reliability of a new iPhone application for assessing planar active cervical range of motion and associated 3D coupling motions in people with neck pain, using an electromagnetic tracking device as a reference test. Increased knowledge about the amount of variation in measurements of active cervical range of motion within days and between days is important to interpret active cervical range of motion measurements in both clinical practice and research. Therefore, in **chapter 6**, the intraday and interday variability, and systematic change over the day of aCROM measurements in asymptomatic people is determined, using a clinically applicable iPhone application. Subsequently, **chapter 7** presents the clinical characteristics between people with nontraumatic neck pain, people with traumatic neck pain, and pain-free individuals in primary physiotherapy care. In **chapter 8**, differences in

strength of associations between pain, physical and psychological impairments on the one hand and disability on the other, are compared between people with nontraumatic neck pain and people with traumatic neck pain. Moreover, interactions between pain and physical and psychological factors contributing to disability are explored in people with nontraumatic neck pain and people with traumatic neck pain, giving insights into the complexity of these types of neck pain. Finally, in **chapter 9**, we review the main findings of this thesis, discuss the implications for practice and research, and present directions for future research.

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Chapter 2

TO WHAT DEGREE DOES ACTIVE CERVICAL RANGE OF MOTION DIFFER BETWEEN PATIENTS WITH NECK PAIN, PATIENTS WITH WHIPLASH, AND THOSE WITHOUT NECK PAIN? A SYSTEMATIC REVIEW AND META-ANALYSIS

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ABSTRACT

Objectives

To quantify differences in active cervical range of motion (aCROM) between patients with neck pain and those without neck pain, in patients with whiplash-associated disorders (WADs) and nontraumatic neck pain, and in patients with acute complaints versus those with chronic complaints.

Data Sources

Seven bibliographic databases were searched from inception to April 2015. In addition, a manual search was performed.

Study Selection

Full articles on a numerical comparison of aCROM in patients with neck pain and asymptomatic control persons of similar ages were included. Two reviewers independently selected studies and assessed risk of bias.

Data Extraction

Two reviewers extracted the data. Pooled mean differences of aCROM were calculated using a random-effects model.

Data Synthesis

The search yielded 6261 hits; 27 articles (2366 participants, 13 low risk of bias) met the inclusion criteria. The neck pain group showed less aCROM in all movement directions compared with persons without neck pain. Mean differences ranged from -7.04° (95% CI, -9.70° to -4.38°) for right lateral bending (11 studies) to -89.59° (95% CI, -131.67° to -47.51°) for total aCROM (4 studies). Patients with WADs had less aCROM than patients with nontraumatic neck pain. No conclusive differences in aCROM were found between patients with acute and patients with chronic complaints.

Conclusions

Patients with neck pain have a significantly decreased aCROM compared with persons without neck pain, and patients with WADs have less aCROM than those with nontraumatic neck pain.

INTRODUCTION

Neck pain is a common musculoskeletal disorder that causes substantial disability and burden.^{1,2} The prevalence of neck pain in western Europe was 6.5% in 2010.² Frequently mentioned symptoms are pain, discomfort, and mobility deficits.³⁻⁶

Physiotherapists and other health care providers measure active cervical range of motion (aCROM) to describe patients' health problems in terms of impairments of cervical mobility, to determine a prognosis, and to evaluate the effects of physiotherapy treatment, both in clinical and scientific settings.⁶⁻¹²

Several factors that affect the mobility of the cervical spine are described.¹³ There is conclusive evidence that age is a contributing factor to aCROM, with aCROM steadily diminishing as one grows older.¹³⁻¹⁶ There is conflicting evidence that women have greater aCROM compared with men,¹³⁻¹⁶ and Sterling et al¹⁷ found that a reduction in aCROM is more evident in patients with moderate to severe pain. It is therefore important that these factors are considered when determining whether aCROM is reduced.^{14,16}

There is evidence to suggest that reduced cervical mobility is associated with activity limitations and disability in case of neck pain and chronic whiplash-associated disorders (WADs).¹⁸⁻²⁰ Several studies^{17,21-23} have described a reduction in aCROM in patients with neck pain when compared with persons without neck pain. There is conflicting evidence to support that patients with persistent WADs have less aCROM than patients with non-traumatic neck pain.^{16,21,23,24}

Furthermore, reduced aCROM may also increase the risk of developing neck pain,⁵ and it is considered to be an unfavorable prognostic factor in the recovery of patients with neck pain.^{5,12,18,25,26}

The clinical utility of aCROM measurements, however, still remains unclear. Studies investigating differences in aCROM measurements between patients with neck pain and asymptomatic persons show conflicting results on the extent of the difference. Some authors found a large difference in aCROM,^{24,27} whereas others did not.^{28,29} The heterogeneous group of patients with neck pain, the different planes of movement, and the measurement procedures used differed between these studies, thus making it difficult to formulate definitive conclusions.¹²

To date, no previous research has systematically summarized and appraised the literature concerning the degree to which people with neck pain differ from people without neck pain, and which planes of movement are best at distinguishing patients from controls. The objective of this review was to systematically assess

the existing literature on whether and to what extent patients with neck pain differ in 2-dimensional aCROM when compared with persons without neck pain. This study aimed to reach more conclusive results by summarizing the available studies in a meta-analysis. A secondary aim was to assess differences in aCROM between patients with acute and patients with chronic neck pain, and those with nonspecific neck pain versus those with WADs.

METHODS

Study selection

Articles were included when (1) 2-dimensional aCROM was assessed in both a group of patients with either neck pain or WADs, and a group of control persons without neck pain; (2) mean age differed by no more than 10 years between groups; (3) adult patients (aged >18y) with acute or chronic neck pain grade I, II, or III according to the Task Force on Neck Pain,³⁰ or with WADs grade I, II or III according to the Quebec Task Force³¹ were included; and (4) a numerical comparison of maximal aCROM, expressed in degrees or radians with SDs, was reported. Studies using radiography, computed tomography or magnetic resonance imaging, or analyzing segmental mobility were excluded. Inclusion and exclusion criteria were set a priori. No restrictions were made on language or publication date. Non-English articles were translated independently by 2 native speakers.

Literature search

A research librarian searched the electronic databases MEDLINE (Ovid), EMBASE, Web of Science, CINAHL (EBSCOhost), Cochrane Central (Wiley), PubMed (the subset as supplied by publisher), and Google Scholar from inception to April 2015 using free text words, Medical Subject Headings (MEDLINE), and a thesaurus (EMBASE, CINAHL). Combinations were made based on neck pain (neck pain, neck injury, whiplash injury, cervicgia) and range of motion (range of motion, mobility, flexibility). A highly sensitive search was performed.³² The complete search strategy is listed in appendix 1. References from retrieved articles and related articles were also checked to retrieve additional relevant publications not captured in the electronic databases. In addition, experts were asked if they could provide any further relevant studies.

Two review authors (M.S.S., M.R.) independently selected the studies by screening the title and abstract to determine whether the study met the inclusion criteria regarding design, participants, and aCROM. We created a standardized form in Microsoft Excel to manage the records retrieved from the search. The same 2 reviewers independently assessed potentially relevant full-text articles. Disagreements were solved by discussion or through arbitration by a third review author (G.S.-P.). We listed the excluded studies and their bibliographic details with the reason for exclusion.

Data extraction and management

Two review authors (M.S.S., M.R.) independently performed the data extraction by using an electronic prepiloted data extraction form. Data extraction included general study information, characteristics of the study, characteristics of the study population, outcome assessors, measurement procedure, statistical analysis, and results. In case of insufficient information, the respective authors were e-mailed for additional data. We imported outcome results from the data extraction form into the software package Review Manager 5.3 (RevMan).

Assessment of methodological quality

Two reviewers (M.S.S., M.R.) independently performed methodological quality assessments. Since there are currently no criteria lists available with acceptable interrater reliability and construct validity for assessing risk of bias of the observational studies included in this review,^{33,34} recommendations are to customize existing quality assessment tools to design a list specifically targeting the context of the study designs of interest.^{32,34} Therefore, we created a criteria list based on the recommendations of the *Cochrane Handbook for Systematic Reviews of Interventions*,³² and on generally accepted criteria items derived from the Newcastle-Ottawa Scale and the Quality Assessment of Diagnostic Accuracy Studies-2 Scale.^{35,36} In this way we developed a tool to critically appraise potential methodological shortcomings such as selection bias, information bias, blinding, attrition bias (items 1-10), and concerns regarding applicability (items 11 and 12) (box 1). Judgments on items 1 to 10 were categorized as “low risk of bias”, “high risk of bias”, or “unclear risk of bias”.³² Criteria 11 and 12 concerning applicability were answered with either “yes”, “no”, or “unclear” when insufficient information was provided. We did not apply weights to the different items of the criteria list, and we arbitrarily considered a study to have a low risk of bias when >7 criteria (out of 10) were met. Applicability was considered sufficient if both criteria were scored as “yes”.

We resolved disagreements by consensus; if disagreement persisted, a third review author (G.S.P.) made the final decision. Results were reported in a risk of bias table. The quality criteria were first tested in multiple training sessions and discussed with experts. Consensus was achieved after several rounds of discussions among the reviewers. We prepiloted the quality assessment form and calculated the absolute and chance-corrected degree of agreement (k) between the 2 reviewers.

Box 1. Quality assessment checklist used to evaluate quality of included studies**Risk of Bias**

1. Was the case definition adequate? (*selection bias*)
2. Was the definition of controls adequate? (*selection bias*)
3. Was there an adequate description inclusion and exclusion criteria? (*selection bias*)
4. Comparability of cases and controls (*selection bias*)
5. Were the patients and controls recruited from the same source population? (*selection bias*)
6. Was a valid and reproducible test used? (*information bias*)
7. Was the measurement procedure equal for both groups? (*information bias*)
8. Was the outcome assessment blinded to the exposure status? (*blinding*)
9. Were there any systematic differences between groups in withdrawals from the study? (*Attrition Bias*)
10. Was a power analysis conducted in order to calculate the minimum number of participants?

Concerns regarding applicability

11. Was the sample of examiners used representative?
12. Representativeness of the exposed cohort

Data synthesis and meta-analysis

ACROM can be expressed as a “half-cycle” motion in 1 of the 6 primary movements (flexion, extension, right rotation, left rotation, right lateral bending, left lateral bending); as a “full-cycle” motion, which is the entire range of each primary movement plane (flexion-extension combined, right and left rotation combined, right and left lateral bending combined); or as the total cervical range of motion (each primary plane summed up).^{21,37} Numerical comparisons between groups in maximal aCROM, expressed in degrees, were used for data analysis. The meta-analysis was conducted using a random-effects model, accounting for the possibility of unexplained heterogeneity (inconsistency). To analyze heterogeneity, we used visual inspection of the point estimates in the forest plots and the overlap of confidence intervals (CIs), and the statistical I^2 test for heterogeneity. I^2 describes the percentage of total variation across studies that is ascribed to heterogeneity. A percentage of approximately 25% indicates low heterogeneity;

50%, medium heterogeneity; and 75%, high heterogeneity.³⁸ Data from individual studies were pooled with RevMan to determine a mean difference (MD) with a 95% CI for continuous data. Forest plots were used to present the point estimates and CIs for the individual studies and meta-analyses.

Subgroup analyses

Several factors may contribute to heterogeneity in aCROM outcome across studies. A priori, we specified a limited number of subgroup analyses to explore possible differences in aCROM: (1) difference between patients with WADs compared with patients with nontraumatic neck pain; and (2) acute complaints versus chronic complaints. We also planned a sensitivity analysis to determine the effect of the study quality on the MD in aCROM.

RESULTS

Search procedure

Our electronic search yielded 6257 hits. Four studies were added from hand searching. After screening of titles and abstracts, a total of 107 potentially relevant studies remained. Most studies were in English, 1 was written in French, 1 in German, and 1 in the Persian language (Farsi). After reviewing the 107 full-text articles, all but 1 of the disagreements concerning inclusion were resolved by consensus. The third reviewer excluded this article. In total, 27 articles met the inclusion criteria (fig 1) and were included.

Study designs and characteristics of population

The general characteristics of the 27 included studies are described in table 1. Included studies reported sample sizes per subgroup ranging from 7 in the study by Sjolander et al²³ to 129 in the study by Puglisi et al.³⁹ Studies were conducted in 13 different countries (Europe: Sweden 6, Germany 2, Switzerland 2, Norway 2, Spain 2, Denmark 1, Italy 1. United States 3, Australia 3, Israel 2, Canada 1, New Zealand 1, Taiwan 1).

Most of the participants in the studies were women (61.5%). The average age of the participants was late 30s to early 40s, with a range $\pm$ SD of 19.9 ± 1.9 years to 56.6 ± 3.5 years per subgroup. Four studies^{17,40-42} recruited patients with only acute complaints, while 18 studies^{20-24,27,29,39,43-51} included patients with only chronic complaints.

Measurements

A variety of tests were used for measuring aCROM; in 6 studies^{40-42,45,52,53} the cervical range of motion instrument was used, 2 studies^{24,54} used a Myrin goniometer, 1 study⁴⁹ used single inclinometers, and 1 study⁵¹ opted for a compass device. Seventeen studies used an electronic device such as the 3Space Fastrak (9 studies^{17,20-23,29,43,48,55}) or the ultrasound-based Zebris system (3 studies^{27,50,56}). Less common was the application of video systems: video-photogrammetry system,⁴⁴ Elite video system,³⁹ Spine Motion Analyzer CA-6000,⁴⁶ ProReflex system,⁴⁷ and a 6-camera motion capture system.⁵⁷

Fig 1 Flow diagram of articles reviewed.

Outcome measures

The primary outcome in our study was aCROM. All studies except one⁴⁷ described aCROM in degrees. For this study we converted radians into degrees, making pooling of the data possible.

Risk of bias in included studies

The results are presented in table 2. Thirteen^{20,22,23,29,39-41,43,45,48,49,52,54} of 27 studies were considered to have a low risk of bias ($\geq 7/10$). Most studies scored positive on usage of valid and reliable instruments (item 6) and an identical measurement procedure for all groups (item 7). Mostly, patients and controls were sufficiently comparable (item 4) and were recruited from the same source population (item 5). Researchers, however, often failed to adequately describe the characteristics of patients (63% were inadequate) and controls (30%) (items 1 and 2), and were not specific in their description of inclusion and exclusion criteria (item 3, 30%) or of the procedure of blinding (item 8, 78%). Moreover, 17 of 27 studies insufficiently described the experience and profession of the clinical examiners (item 11). Most of the studies included a representative sample of average patients with neck pain in the community (item 12, 70%). Eight studies^{17,27,41,45,49,51-53} met all criteria for external validity (items 11 + 12).

Overall, high levels of agreement between the reviewers were achieved for risk of bias assessment, resulting in a percentage of agreement of 92% (95% CI, .91-.94) and a kappa of .85 (95% CI, .79-.90) for the main analysis. Kappa values ranged from .65 (for item 1) to 1.0 (for items 7 and 10). We resolved all disagreements through consensus; consequently there was no need to consult the third reviewer for a final decision.

Differences in aCROM between patients with neck pain and persons without neck pain

Total range of motion

Four studies^{21,27,41,42} holding 507 participants (with 1 of the studies having a low risk of bias) assessed total aCROM (combination of total of flexion, extension, rotation left and right, and lateral bending left and right) between patients with neck pain and persons without neck pain. From these studies it can be concluded that a significantly decreased total aCROM is present in patients with neck pain (MD= -89.59°; 95% CI, -131.67° to -47.51°) (fig 2).

Full-cycle aCROM

Five studies^{21,29,44,53,54} with 483 participants (2 of the studies with a low risk of bias) assessed total flexion-extension. Six studies^{20,21,29,46,53,54} with 653 participants (4 of the studies with a low risk of bias) assessed total rotation. Four studies^{21,53,54,57} with 398 participants (1 of the studies with a low risk of bias) assessed lateral bending. Total flexion-extension was significantly reduced in patients compared with Controls (MD = -28.51°; 95% CI, -40.92° to -16.11°). Significant differences were also found for total rotation (MD=-29.49°; 95% CI, -39.81° to -19.16°) and total lateral bending (MD= -16.72°; 95% CI, -23.99° to -9.45°) (fig 3).

Half-cycle aCROM

Nineteen studies^{17,22,24,27,29,39-43,45,47-52,55,56} with 1649 participants (10 of the studies with a low risk of bias) evaluated half-cycle flexion and extension. Quantitative analyses showed that both flexion and extension were significantly reduced in the patient groups (flexion: MD= -11.18°; 95% CI, -13.55° to -8.80°; extension: MD= -17.48°; 95% CI, -20.95° to -14.01°). Sixteen studies^{22-24,27,29,39,40,42,43,45-48,50,51,55} with 1458 participants (8 of the studies with a low risk of bias) assessed right and left rotation. A significant reduction of active cervical rotation was found in the patient groups (right rotation: MD= -13.93°; 95% CI, -17.58° to -10.29°; left rotation: MD =-14.76°; 95% CI, -18.43° to -11.09°). Similar results were found in 11 studies^{22,27,39,40,42,43,45,49-51,55} with 1070 participants (6 of the studies with a low risk of bias) that examined right lateral bending (MD=-7.04°; 95% CI, -9.70° to -4.38°) and left lateral bending (MD=-7.78°; 95% CI, -10.15° to -5.42°) (figs 4-6). Three studies^{17,41,56} combined the mean values for right and left rotation, and for right and left lateral bending in their analysis. This led to similar results as with studies that did not.

Subgroup analyses

WADs versus nontraumatic neck pain or persons without neck pain

Three studies^{21,23,24} holding 268 participants (1 of the studies with a low risk of bias) included both a WADs group and a nontraumatic neck pain group. Fourteen studies^{17,22,27,39-48,53} included a group with only patients with WADs.

Of the 3 studies that compared patients with WADs to patients with nontraumatic neck pain, the studies by Grip²⁴ and Woodhouse²¹ and colleagues with 237 participants (both studies having a high risk of bias) found that the WADs group had significantly less aCROM than the nontraumatic neck pain group. Sjölander's

study²³ with 15 participants and a low risk of bias showed a nonsignificant result in favor of the nontraumatic neck pain group (fig 7).

The 14 studies that compared patients with WADs to persons without neck pain showed more reduction in aCROM in the WADs group for all movements, with MDs ranging from -97.52° (95% CI, -143.50° to -51.53°) for total aCROM to -8.10° (95% CI, -11.33° to -4.87°) for right lateral bending (table 3).

Thirteen studies^{20,21,23,24,29,49-52,54-57} included a “nontraumatic neck pain group” consisting of patients with nontraumatic complaints or no specification of the origin of complaints. Comparison between these patients and control persons without neck pain showed more reduction in aCROM in the neck pain group for all movements, with MDs ranging from -51.30° (95% CI, -67.31° to -35.29°) for total aCROM to -4.65° (95% CI, -6.59° to -2.71°) for right lateral bending (see table 3).

Patients with acute and chronic complaints versus persons without neck pain

No study included both an acute and chronic complaints group. Four studies^{17,40-42} with 257 participants (2 of the studies with a low risk of bias) compared a group with only acute complaints ($<6\text{wk}$) to a group of persons without neck pain, and 18 studies^{20-24,27,29,39,43-51,56} with 1653 participants (9 of the studies with a low risk of bias) compared a group with only chronic complaints to a group of persons without neck pain.

Table 4 shows the pooled MDs of the studies that included patients with acute neck pain and studies that included patients with chronic neck pain. The results vary with the different motion directions. Given these conflicting results, no clear conclusion can be drawn concerning any difference in aCROM between patients with acute and chronic complaints.

Sensitivity analyses

Substantial heterogeneity was observed among studies (see figs 2-6). Including only studies with a low risk of bias led to acceptable low or medium heterogeneity, yet slightly smaller pooled MDs (see figs 2-6). For full-cycle aCROM, the decrease of the pooled MD ranged from -8.02° (lateral bending) to -14.73° (flexion-extension), and for half-cycle aCROM from -1.47° (flexion) to -4.22° (extension).

DISCUSSION

In this systematic review and meta-analysis, including 27 studies totaling 2366 participants (women: 61.5%), the aCROM differed between patients with neck pain and controls. Largest differences in aCROM were found in full-cycle flexion-extension (MD = -28.51° ; 95% CI, -40.92° to -16.11°) and in total rotation (MD = -29.49° ; 95% CI, -39.81° to -19.16°), and for half-cycle motion in extension (MD = -17.48° ; 95% CI, -20.95° to -14.01°). Subgroups of patients with WADs appeared to have an even more reduced aCROM than groups including patients with nontraumatic neck pain. No consistent differences between patients with acute and chronic complaints could be derived from indirect comparisons.

These results are in accordance with the situation in daily physiotherapy practice, where a reduction of aCROM is usually seen as a clinical feature of patients with neck pain, and with recommendations in the literature, where assessment of cervical range of motion is accepted as being an important diagnostic and evaluative parameter.⁶ The results of the included studies are consistent. Persons without neck pain showed a larger aCROM for all movements. These differences are clinically relevant and greater than the reported measurement errors of commonly used instruments in practice and research.^{10,58,59} As such, the findings of the present study can be considered to be true differences in aCROM between patients with neck pain and those without neck pain.

At the onset of this review we hypothesized differences in aCROM between acute and chronic phases of pain, yet apparently the duration of symptoms is not an important factor for aCROM. Indeed, it is possible that other factors, such as the severity of complaints, may be more relevant. Still, only limited conclusions can be drawn about the differences in aCROM between WADs and nontraumatic neck pain, and acute versus chronic complaints, because only indirect comparisons were possible given the absence of sufficient studies that included both groups. Therefore, MDs between WADs and nontraumatic neck pain, and acute versus chronic complaints may be related to differences in the participants' characteristics, severity of complaints, symptom duration, and natural variations in the population.

Heterogeneity across studies was high. This could be explained by the heterogeneous group of patients with neck pain that was included in most retrieved studies. There were relevant differences in, for example, inclusion criteria for participants, setting, severity of symptoms, measurement procedure,

and study quality. This may have increased the heterogeneity of our study. High heterogeneity may reduce the confidence in the recommendations of this review and reduce the precision of the findings. However, analyzing only studies with a low risk of bias led to substantially less heterogeneity, mostly low or medium, and only small and nonsignificant differences in outcome, which is why we included all studies in the meta-analysis. We assume that heterogeneity may not have influenced our results substantively. Furthermore, we pooled the data of all studies because most of the studies found a greater aCROM in favor of the controls, and the relatively small CIs of pooled data indicate reasonable strength of our findings. Moreover, we used the random-effects model not to estimate one true effect, but to estimate the mean of a distribution of effects.⁶⁰ Another option to handle important heterogeneity among the studies is to divide the studies into different subgroups. However, because of the lack of detailed information about patient characteristics in most studies, the possibilities of comparing subgroups were limited. Subgroups of patients with neck pain based on signs and symptoms and neck function may demonstrate less heterogeneity and more differentiation in aCROM.

Study limitations

This review may have some limitations. Although much effort was made to retrieve all relevant studies, publication bias cannot be excluded. It is possible that studies were missed or that unpublished studies were not found. Studies showing no difference between patients and controls are more likely to remain unpublished than those reporting positive findings.⁶¹

Risk of bias was assessed using a self-developed criteria list based on validated quality assessment scales.^{32,35,36} This list as a whole has not been validated before, but individual items are generally accepted and have been carefully chosen, making all items relevant and specific to the studies' included designs.

Only 6 studies^{20,23,40,51,54,57} (4 with a low risk of bias) reported adequate blinding of the assessors. Inadequate blinding may have enlarged the difference in aCROM.

Recommendations

We were able to include a large number of studies, and the consistency of the results presented in this systematic review showed that the amount of aCROM is an important discriminating parameter in the diagnostic process in patients with neck pain. But in order to determine the clinically relevant change in aCROM in

relation to recovery (minimal clinically important change), further research should focus on the degree of increase of aCROM after therapeutic interventions and its correlation with the reduction of neck complaints, and should also determine to what extent restoring reduced aCROM contributes to health benefits. Moreover, future studies measuring aCROM in patients with cervical pain should distinguish between clinically relevant subgroups, and studies should provide detailed information about the included study population in order to determine the degree of reduced aCROM per subgroup.

CONCLUSIONS

This systematic review provides good-quality evidence that there is a significant, clinically relevant decrease in aCROM in patients with neck pain (traumatic or nontraumatic) compared with persons without neck pain. Full-cycle flexion-extension and total rotation, and half-cycle extension showed the largest differences in range of motion between patients with neck pain and those without neck pain. A traumatic origin of complaints seems to have a negative influence on aCROM.

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Table 1. Study characteristics of included studies (ranked in alphabetical order by first author)

Author (year, country)	Patient population	Healthy controls	Measurement	Results (°)	Author's conclusion
Ang (2008, Sweden)	Male helicopter pilots with neck pain at the time of testing (VAS >10 mm). Selected from 2 air force helicopter squadrons.	Male helicopter pilots. Selected from 2 air force helicopter squadrons.	Cervical measurement system (Myrin device)	Flexion-extension NP: 112.9 ±13.2 HC: 126.8 ±11.7 Axial rotation, left-right NP: 132.1 ±19.9 HC: 147.6 ±12.0 Lateral bending, left-right NP: 63.6 ±13.8 HC: 72.3 ±13.9	As compared to controls, the neck pain patients had less flexion-extension (P= 0.003) and rotation (P= 0.005)
Armstrong (2005, New Zealand)	Grade II or III WAD (QTF). Minimum of one whiplash injury, at least three months prior but no more than five years earlier. Via advertisement.	Via advertisement.	Electromagnetic motion-tracking device (Fastrak)	Flexion WAD: 50.3 ±10.7 HC: 57.3 ±8.2 Extension WAD: 54.8 ±14.7 HC: 75.6 ±13.1 Right rotation WAD: 62.5 ±11.3 HC: 73.4 ±9.3 Left rotation WAD: 64.3 ±9.9 HC: 73.5 ±9.7 Right lateral bending WAD: 37.1 ±8.1 HC: 41.4 ±7.3 Left lateral bending WAD: 37.9 ±8.8 HC: 41.6 ±8.7	Overall, when compared with the healthy group, the whiplash group displayed significantly less flexion, extension, left and right rotation RoM at most receptor sites. However, no significant differences were observed when comparing the mean maximum left and right lateral bending RoM recorded for whiplash and healthy groups
Baydal-Bertomeu (2011, Spain)	WAD grades II and III (QTF), 6 months - 1 year. Hospital Rehabilitation Unit.	Volunteers	video-photogrammetry system (Kinescan-IBV)	Total Flexion - extension WAD: 90 ±22 HC: 119 ±17	Comparison between controls and patients showed significant reductions in the range of motion in the patients
Branney (2014 Sweden)	Mechanical neck pain (reproducible by neck movement/provocation tests). Pain located within the area defined by the Neck Pain Task	Recruited from staff, students and visitors.	The CROM goniometer	Flexion: NP: 49 ±6.7 HC: 53 ±7.2 Extension: NP: 51 ±7.2 HC: 56 ±6.6	Patients had less ROM in flexion (p = 0.04) and extension (p = 0.03)

	<p>Force, Self-reported pain rating $\geq 3/11$, of at least 2 weeks duration. Recruited from a chiropractic outpatient clinic.</p> <p>N= 30 Age: 40.9 $\pm$13.1 Male / Female: 9/21</p>				
	<p>N= 29 Age: 39.7 $\pm$13.1 Male / Female: 8/21</p>				
Dall'Alba (2001, Australia)	Primary injury to the neck from motor vehicle crash, ongoing pain and disability in the neck region, time since injury 3 months to 2 years. Research Unit at the Department of Physiotherapy, The University of Queensland	Recruited by advertisement in university staff and student areas and on the radio	Electromagnetic motion-tracking device (Fastrak)	<p>Flexion WAD: 35.3 $\pm$10.8; HC: 47.2 $\pm$9.1</p> <p>Extension WAD: 36.9 $\pm$12.4; HC: 51.8 $\pm$11.0</p> <p>Right rotation WAD: 52.7 $\pm$12.6; HC: 66.8 $\pm$8.5</p> <p>Left rotation WAD: 53.6 $\pm$14.2; HC: 68.1 $\pm$8.9</p> <p>Right lateral bending WAD: 31.2 $\pm$9.5; HC: 36.8 $\pm$7.6</p> <p>Left lateral bending WAD: 30.7 $\pm$9.8; HC: 38.1 $\pm$8.3</p>	Individuals with persistent WAD have reduced primary ROM. This decrease in primary ROM is apparent even after conjunct ROM or pattern of movement is taken into account
	<p>N=114 Age: 37.2 $\pm$11.5 Male / Female: 22/93</p>				
	<p>N=89 Age: 39.2 $\pm$14.2 Male / Female: 41/48</p>				
Fernandez-Perez (2012, Spain)	Acute WAD II (QTF). Referred by primary care physician to a physical therapist.	Recruited from the general population	The CROM goniometer	<p>Flexion WAD: 39.4 $\pm$18.6 HC: 59.3 $\pm$10.7</p> <p>Extension WAD: 53.8 $\pm$22.6 HC: 83.0 $\pm$10.5</p> <p>Right rotation WAD: 44.7 $\pm$20.6 HC: 71.3 $\pm$6.7</p> <p>Left rotation WAD: 47.8 $\pm$19.8 HC: 70.2 $\pm$10.4</p> <p>Right lateral bending WAD: 35.6 $\pm$15.5 HC: 45.2 $\pm$5.5</p> <p>Left lateral bending WAD: 33.8 $\pm$11.5 HC: 44.3 $\pm$9.7</p>	Patients with acute WAD showed decreased active CROM when compared to controls for all movements ($P < .001$)
	<p>N=20 Age: 29.1 $\pm$12.2 (23.3–34.8) Male / Female: 10/10</p>				
	<p>N=20 Age: 28.7 $\pm$12.4 (22.9–34.4) Male / Female: 10/10</p>				
Grip (2007, Sweden)	a) Neck Pain group: pain lasting >3 months. Their symptoms were muscular without paraesthesia	Occasional neck or back pain was accepted as long as subjects were free from symptoms in the past 3 months.	Myrin device (gravity-reference goniometer)	<p>Flexion NP: 52.0 $\pm$17.2 WAD: 38.0 $\pm$18.4 HC: 61.4 $\pm$12.9</p> <p>Extension NP: 43.6 $\pm$18.4 WAD: 30.0 $\pm$19.4</p>	The control group reported larger ROM than the NP and WAD groups. When comparing the two groups with neck pain, the WAD group perceived their situation worse with respect to ROM in all directions

Kasch (2001, Denmark)	Presentation of whiplash-related problems at the local emergency unit within 2 days after collision.	Patients who had sustained non-sport ankle distortion. Recruited from emergency unit.	The CROM instrument	Flexion WAD: 49.3±12.6 HC: 54.1 ±10.1 Extension WAD: 63.8 ±16.0 HC: 71.6±14.0 Rotation (combined left - right) WAD: 63.5 ±13.0 HC: 71.0 ±9.8 Lateral bending (combined left - right) WAD: 40.3 ±9.1 HC: 44.0±7.0 Total CROM WAD: 320.7 ±61.2 HC: 355.8 ±42.5	The total active CROM was significantly lower in the whiplash group than in the ankle distortion group
Klein (2001, Switzerland)	Chronic WAD grades I - III, most being grade II. Symptoms lasting more than 3 months attributed to a motor vehicle accident. Referred from their general practitioners to the neurology and rheumatology departments of the hospital.	Some of the healthy control subjects had experienced neck pain in the past, but none were suffering with any symptoms at the time of the test. Mostly hospital employees (46%).	Spine Motion Analyzer CA-6000	Right rotation WAD: 41.7 ±16.9 HC: 67.8 ±10.8 Left rotation WAD: 43.1 ±16.4 HC: 69.4 ±9.8 Total Left + Right rotation WAD: 84.8 ±31.0 HC: 137.2 ±18.5	Cervical axial ROM differed significantly (p=0.0001) between the groups, with the whiplash patients showing lower values than the healthy controls
Kumbhare (2005, Canada)	Grade II WAD based on the QTF criteria. Patients treated at St Joseph's Hospital, Hamilton, Ontario, Canada.	Hospital staff, family and friends of staff, non-patient visitors, medical students and volunteers from a men's soccer club.	The CROM instrument	Flexion/Extension WAD: 92.8 ±32.1 HC: 136.5 ±20.5 Rotation WAD: 104.0 ±34.6 HC: 143.1 ±18.8 Lateral bending WAD: 64.8 ±24.9 HC: 92.0 ±17.1	Whiplash patients had lower CROM in all planes
Ohlberg (2003, Sweden)	Chronic symptoms (>3 months) and different grades of WAD (QTF). Department of	Healthy employees (e.g., office workers, nurses, physiotherapists and	ProReflex system	Flexion WAD: 41.25 ±15.47 HC: 59.01 ±9.74 Extension	Statistically significant differences were found between controls and subjects with chronic WAD

	Rehabilitation Medicine, University Hospital of Umeå, Sweden.	electricians) and students without chronic head, neck or back pain. Recruited through advertisements at the University Hospitals.		WAD: 30.94 ±12.03 HC: 49.85 ±9.17 Rotation Right WAD: 56.15 ±16.04 HC: 71.05 ±7.45 Rotation Left WAD: 55.00 ±14.32 HC: 73.91 ±7.45	
	N= 56 Age: 38.1 ±10.6 Male / Female: 29/30 (3 excluded)	N= 52 Age: 37.3 ±10.9 Male / Female: 29/27 (4 excluded)			
Osterbauer (1996, USA)	Patients who experienced a symptomatic whiplash-type neck trauma as a result of a rear-end impact. The patients were sought from the clinic and from local area chiropractors.	Matched to the characteristics of the whiplash patients. Recruited at the clinic and a local university campus.	CROM, head mounted inclinometer	Flexion Men WAD: 49 ±8 HC: 56 ±10 Woman WAD: 49 ±12 HC: 59 ±13 Extension Men WAD: 43 ±5 HC: 72 ±9 Woman WAD: 47 ±16 HC: 71 ±8 Right rotation Men WAD: 54 ±19 HC: 69 ±7 Woman WAD: 59 ±14 HC: 71 ±12 Left rotation Men WAD: 51 ±14 HC: 73 ±9 Woman WAD: 57 ±13 HC: 71 ±13 Right lateral bending Men WAD: 39 ±8 HC: 46 ±6 Woman WAD: 34 ±10 HC: 46 ±9 Left lateral bending Men WAD: 33 ±5 HC: 46 ±7 Woman WAD: 35 ±11 HC: 47 ±9 Total ROM Men WAD: 269 ±30 HC: 360 ±35 Woman WAD: 282 ±62 HC: 364 ±37	Standard ROM reasonably differentiated the injured patients from controls
Pereira (2008, Australia)	Current drivers with chronic neck pain attributed to an MVC at least 3 months post injury. Volunteers recruited from patients who attended a university whiplash research	Current drivers with no history of neck pain or trauma. Recruited from staff and students of the university and the	Electromagnetic tracker system (Fastrak)	Flexion WAD: 38.7 ±14.7 HC: 53.0 ±10.5 Extension WAD: 46.5 ±18.4 HC: 66.5 ±13.8 Right rotation WAD: 57.5 ±14.1 HC: 67.0 ±11.3 Left rotation	The subjects in the whiplash-associated disorder group had significantly less ROM in flexion, extension, and rotation left and right

	unit and through advertising in the local newspaper.	general public through advertisements.	WAD: 59.9 ±16.7 HC: 72.2 ±9.5	
	N= 30 Age: 25.6 ±5.1 Male / Female: 8/22	N= 30 Age: 33.8 ±9.4 Male / Female: 8/22		
Prushansky (2006, Israel)	WAD II - III (QTF), a minimum duration of 6 months post injury. All patients were victims of traffic accident and were all treated at the Spinal Care Unit, Sapir Medical Center near Tel Aviv. All patients were initially referred for conservative treatment.	Sample with no history of spinal disorders, peripheral or central neurologic conditions, dizziness, head ache, or back pain at the time of testing. Sample of convenience.	Zebris CMS 70P system Flexion WAD: 30.5 ±15.1 HC: 58.7 ±12.7 Extension WAD: 28.9 ±13.3 HC: 64.1 ±15.5 Right rotation WAD: 41.3 ±14.9 HC: 71.1 ±9.3 Left rotation WAD: 43.1 ±16.6 HC: 73.9 ±10.7 Right lateral bending WAD: 25.9 ±10.0 HC: 43.0 ±7.6 Left lateral bending WAD: 27.1 ±9.6 HC: 41.9 ±8.0 Total ROM WAD: 196.9 ±69.3 HC: 352.8 ±46.4	A significant reduction in CROM in chronic WAD patients relative to the controls was revealed
	N= 101 Age: 40.3 ±10.6 Male / Female: 47/54	N= 75 Age: 36.3 ±8.9 Male / Female: 16/59		
Puglisi (2004, Italy)	WAD 1 or 2 (QTF), ongoing (chronic) pain after a rear-end collision between 6 and 12 months earlier, and were awaiting some form of compensation.	Similar age and X-ray restrictions, no prior known history of neck injury and no recall of neck or dorsal spine pain in the last 12 months. Recruited by advertisement.	Elite video system Flexion WAD: 52.6 ±12.7 HC: 63.3 ±9.7 Extension WAD: 50.1 ±14.1 HC: 66.4 ±14.0 Right lateral rotation (rotation) WAD: 62.7 ±13.9 HC: 74.0 ±8.4 Left lateral rotation (rotation) WAD: 62.4 ±15.1 HC: 73.2 ±8.8 Right lateral bending WAD: 36.9 ±7.1 HC: 43.5 ±6.0 Left lateral bending WAD: 37.7 ±6.6 HC: 44.8 ±7.7	The chronic whiplash subjects showed reduced total neck range in all directions as compared to healthy subjects (p<0.001)
	N= 129 Age: 36.7 ±7.2 Male / Female: 73/56	N= 126 Age: 31.9 ±7.2 Male / Female: 69/57		
Rudolfsson (2012, Sweden)	Chronic non-traumatic neck pain (>3 months. Decreased physical functioning (>9 normalized points of the first 19 items in the Disability Arm	Recruited from?	Total Left + Right rotation NP: 115.2 ±17.0 HC: 136.2 ±15.0	In people with neck pain, extension in the upper levels and predominately flexion for the lower levels was reduced

Shoulder Hand (DASH) questionnaire) Recruited from? N= 33 Age: 47 ±10 Male / Female: 0/33 N= 102 Age: 51 ±9 Male / Female: 0/102					
Rutledge (2012, USA)	Participants with pain in the neck region (VAS ≥ 3). Volunteers were recruited from a University student, faculty and staff population, as well as from a campus clinical center. N= 19 Age: 27.5 ±13.1 Male / Female: 14/5	VAS = 0, and symmetric for right and left lateral bending diagnostic motions. Recruited from a University student, faculty and staff population, as well as from a campus clinical center. N= 22 Age: 19.9 ±1.9 Male / Female: 16/4 (2 non responders)	Six-camera motion capture system	Left + Right lateral bending NP: 77.0 ±13.5 HC: 87.4 ±12.5	Comparisons of average lateral bending ROMs between Control and Experimental groups indicated that participants in the Control group moved through a greater motion range than those in the Experimental group for both active and passive motions
Sarig Bahat (2010, Israel)	Chronic neck pain, with or without pain to the upper limb for longer than 6 weeks. Recruited from an outpatient physical therapy clinic and from the staff and student body of the University of Haifa. N= 25 Age: 39.0 ±12.7 Male / Female: 9/16	Volunteers with no history of neck pain. Recruited from an outpatient physical therapy clinic and from the staff and student body of the University of Haifa. N= 42 Age: 35.3 ±12.4 Male / Female: 11/31	Electromagnetic tracker system (Fastrak)	Flexion NP: 46.1 ±16.4 HC: 58.4 ±11.5 Extension NP: 43.1 ±15.0 HC: 44.3 ±10.9 Flexion + Extension NP: 89.2 ±25.2 HC: 102.7 ±15.7 Right rotation NP: 57.5 ±12.5 HC: 66.3 ±7.5 Left rotation NP: 59.2 ±11.0 HC: 66.7 ±6.6 Right + Left rotation NP: 116.7 ±21.1 HC: 133.1 ±12.3	Significantly reduced CROM in subjects with chronic neck pain as compared with the asymptomatic subjects
Schops (1997, Germany)	Chronic neck pain. Recruited from? N= 20	Volunteers without neck complaints. Recruited from? N= 20	Inclinometer and compass	Flexion NP: 70.2 ±6.1 HC: 74.7 ±4.2 Extension NP: 56.5 ±6.3 HC: 71.3 ±4.6 Right rotation	The mean values of the range of motion in the three planes were significantly smaller for the patients than for the volunteers

Shahidi (2012, USA)	<p>Age: 37 (21-55) Male / Female: 8/12</p> <p>Age: 33 (23-61) Male / Female: 10/10</p> <p>NP: 76.3 ±6.5 HC: 81.8±4.6 Left rotation NP: 67.4 ±5.4 HC: 80.1 ±5.5 Right lateral bending NP: 40.5 ±4.3 HC: 45.9 ±3.8 Left lateral bending NP: 40.0 ±4.7 HC: 45.5 ±3.8</p>	Gravity inclinometer	<p>No history of neck pain in the last year, and a NDI score of less than 5 points. Recruited from university medical campus and surrounding community</p> <p>N= 20 Age: 34.0 ±9.9 Male / Female: 10/9</p>	<p>Flexion NP: 48 ±11 HC: 61 ±8 Extension NP: 56 ±13 HC: 70 ±14 Right lateral bending NP: 39 ±7 HC: 42 ±7 Left lateral bending NP: 39 ±7 HC: 46 ±8</p>	The neck pain group had significantly less ROM in all directions compared to the healthy group ($P \leq 0.012$), with the exception of side bending toward the right ($P = 0.511$)
Sjölander (2008, Sweden)	<p>Neck pain lasting >6 months. Insidious neck pain or WAD grade II or III (QTF). Patients who entered a rehabilitation programme.</p> <p>a: NP N=8 b: WAD N=7 Age: NP 40 ±9; WAD: 45 ±11 Male / Female: NP 0/8; WAD 2/5</p>	Electromagnetic tracker system (Fastrak)	<p>Absence of current, previous or repeated periods of neck pain. Recruited from the local community.</p> <p>N= 16 Age: 41 ±9 Male / Female: 3/13</p>	<p>Right rotation NP: 57.9 ±11.0 WAD: 67.7 ±12.1 HC: 70.7 ±11.2 Left rotation NP: 61.8 ±10.9 WAD: 68.5 ±12.9 HC: 71.9 ±13.0</p>	Although the patients on average showed less ROM than the controls, there were no statistically significant differences between the three groups
Sterling (2004, Australia)	<p>Acute WAD grade II or III (QTF). Three subgroups were identified based on the Neck Disability Index: a: mild, b: moderate, or c: severe pain and disability. Recruited within 1 month of injury via hospital accident and emergency departments,</p>	Electromagnetic tracker system (Fastrak)	<p>Never experienced any previous pain or trauma to the cervical spine, head, or upper quadrant. Recruited from the general community via print media.</p>	<p>Flexion WAD Mild symptoms: 38.0 ±2 WAD Moderate symptoms: 34.5 ±2 WAD Severe symptoms: 32.1 ±4 HC: 45.4 ±2.5 Extension WAD Mild symptoms: 47.4 ±2 WAD Moderate symptoms: 36.4 ±2 WAD Severe symptoms: 31 ±4.3</p>	All three whiplash groups demonstrated less range of movement in all directions when compared to controls ($p < 0.01$) with the severe group also showing less range of extension and left rotation than the other two whiplash groups ($p < 0.01$)

	primary care practices (medical and physiotherapy), and from advertisement.				HC: 57.7 ±3 Combined Right-Left rotation WAD Mild symptoms: 53.5 ±1.7 WAD Moderate symptoms: 50.2 ±1.9 WAD Severe symptoms: 45.3 ±3.5 HC: 60.5 ±2.3 Combined Right-Left lateral bending WAD Mild symptoms: 28 ±1.3 WAD Moderate symptoms: 27.2 ±1.4 WAD Severe symptoms: 26.2 ±2.6 HC: 34.1 ±1.7	
	a: Mild N=36 b: Moderate N=32 c: Severe N= 12 Age: Mild: 30.1 ±13.2 Moderate: 34.1 ±14.3 Severe: 39.5 ±14.8 Male / Female: Mild: 17/19 Moderate: 4/28 Severe: 3/9	N= 20 Age: 39.5 ±14.6 Male / Female: 9/11	Zebris CMS 70 system	Healthy full-time employees who were recruited through advertisements in the daily press in the Frankfurt area. N= 18 Age: 56.6 ±3.5 Male / Female: 8/10	Flexion NP: 40.7 ±19.9 HC: 56.3 ±8.5 Extension NP: 44.3 ±18.4 HC: 67.3 ±10.2 Right rotation NP: 56.6 ±18.9 HC: 68.6 ±6.5 Left rotation NP: 52.6 ±17.3 HC: 75.3 ±7.5 Right lateral bending NP: 27.8 ±14.0 HC: 35.1 ±6.8 Left lateral bending NP: 25.4 ±9.6 HC: 35.0 ±6.6	There were significant differences found in the group of patients with chronic neck pain compared with the control subjects for flexion or extension, axial rotation and lateral bending movement amplitudes (p <0.05) except for lateral bending to the right
Wirth (2014, Switzerland)	Neck pain including WAD. Recruited from various medical practices and from a sports club. N= 19 Age 46.5 ±9.9 Male / Female: 7/12	Recruited from various medical practices and from a sports club. N= 19 Age: 46.6 ±10.5 Male / Female: 7/12	Zebris CMS10 system		Flexion NP: 57.0 ±13.6 HC: 67.0 ±10.6 Extension NP: 56.0 ±16.1 HC: 65.8 ±11.1 Mean of R+L rotation NP: 67.7 ±7.4 HC: 74.6 ±10.3 Mean of R+L lateral bending NP: 38.4 ±8.5 HC: 39.7 ±9.8	Cervical mobility was significantly reduced in all movements, apart from lateral bending
Woudhouse (2008, Norway)	a: Neck pain group: Pain >6 months and <10 years.	No previous or current neck pain or history of neck trauma.	Electromagnetic tracker system (Fastrak)	Flexion + Extension NP: 114.0 ±20.0 WAD: 81.8 ±34.6 HC: 134.0 ±20.7	The WAD group had significantly less ROM than the two control groups in all three	

Recruited from local PT's and GP's. b: Persistent WAD I and II (QTF) for >6 months (WAD group). Symptom duration of 6 months -10 years and onset of symptoms within 48 hours after the accident. Referred to the National Center for Spinal Disorders St Olav's Hospital.	Recruited from different workplaces and educational institutions. N= 57 Age: 38.2 ±10.9 Male / Female: 29/28 a: NP N=57 b: WAD N= 56 Age: NP: 43.7 ±12.6 WAD: 38.2 ±10.8 Male / Female: NP: 19/38; WAD: 22/34	Left + Right rotation NP: 133.1 ±18.6 WAD: 106.2 ±34.7 HC: 151.7 ±13.5 Left + Right lateral bending NP: 72.2 ±13.2 WAD: 60.9 ±18.8 HC: 84.9 ±13.8 Total ROM NP: 319.4 ±44.1 WAD: 248.9 ±83.7 HC: 370.7 ±43.1	primary motion planes as well as total cervical ROM
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WAD = Whiplash associated disorders; QTF = Quebec Task Force; HC = healthy controls;

* Data are mean (SD) unless otherwise indicated

	Definition of cases	Definition of controls	Inclusion and exclusion criteria	Comparability of cases and controls	Source population	Test validity and reproducibility	Measurement procedure	Blinding of outcome assessment	Differences in withdrawals	Power analysis	Representativeness of the examiners	Representativeness of the sample
Ang 2008	+	+	+	+	+	+	+	+	+	+	?	-
Armstrong 2005	+	+	+	+	+	+	+	?	?	?	?	+
Baydal-Bertomeu 2011	-	+	+	?	?	+	+	?	+	+	?	-
Branney 2014	+	+	+	+	+	+	+	-	+	+	+	+
Dall'Alba 2001	+	+	+	-	+	+	+	?	+	?	?	-
Fernandez-Perez 2012	+	+	+	+	+	+	+	+	+	+	?	+
Grip 2007	-	+	+	+	+	+	+	?	-	?	?	+
Grip 2007a	-	+	+	+	+	+	+	?	-	?	?	+
Grip 2007b	-	+	+	+	+	+	+	?	-	?	?	+
Guo 2012	-	-	+	?	?	+	+	?	+	?	?	-
Kaale 2007	+	+	-	+	+	+	+	?	+	?	+	+
Kasch 2001	-	+	+	+	+	+	+	-	+	?	+	+
Klein 2001	+	-	-	+	?	+	+	?	?	?	?	+
Kumbhare 2005	+	-	+	?	+	+	+	?	-	+	+	+
Ohberg 2003	-	+	-	+	+	+	+	?	+	?	?	+
Osterbauer 1996	-	+	-	+	+	+	+	-	+	?	?	-
Osterbauer 1996a	-	+	-	+	+	+	+	-	+	?	?	-
Osterbauer 1996b	-	+	-	+	+	+	+	-	+	?	?	-
Pereira 2008	-	+	+	+	+	+	+	?	+	?	?	+
Prushansky 2006	+	-	+	-	?	+	+	?	?	?	+	+
Puglisi 2004	-	+	+	+	+	+	+	?	+	?	?	-
Rudolfsson 2012	+	-	+	+	?	+	+	+	+	?	+	-
Rutledge 2012	-	-	-	+	+	+	+	+	-	?	+	-
Sarig Bahat 2010	-	+	+	+	+	+	+	?	+	?	?	+
Schops 1997	-	-	-	+	?	+	+	+	+	?	+	+
Shahidi 2012	-	+	+	+	+	+	+	-	+	?	+	+
Sjolander 2008	+	+	+	+	+	+	+	+	+	?	?	+
Sjolander 2008a	+	+	+	+	+	+	+	+	+	?	?	+
Sjolander 2008b	+	+	+	+	+	+	+	+	+	?	?	+
Sterling 2004	-	+	+	-	+	+	+	?	?	?	+	+
Sterling 2004a	-	+	+	-	+	+	+	?	?	?	+	+
Sterling 2004b	-	+	+	-	+	+	+	?	?	?	+	+
Sterling 2004c	-	+	+	-	+	+	+	?	?	?	+	+
Vogt 2007	-	+	+	+	+	+	+	?	?	?	?	+
Wirth 2014	-	-	-	+	+	+	+	?	?	?	?	+
Woodhouse 2008	-	+	-	+	+	+	+	-	+	?	?	+
Woodhouse 2008a	-	+	-	+	+	+	+	-	+	?	?	+
Woodhouse 2008b	-	+	-	+	+	+	+	-	+	?	?	+

Table 2. Quality assessment of included studies

Table 3. Pooled Mean Differences of WAD groups compared to Healthy Controls and non-traumatic neck pain groups compared to Healthy Controls

Movement	#Studies	WAD/HC (n)	MD(°) (95% CI) (WAD – HC)	#Studies	NP/HC (n)	MD(°) (95% CI) (NP – HC)
Total aCROM	4	227/223	-97.52 [-143.50, -51.53]	1	57/57	-51.30 [-67.31, -35.29]
Full Cycle Flexion-extension	3	157/157	-35.26 [-44.41, -26.11]	3	102/124	-16.28 [-21.01, -11.54]
Full Cycle Rotation	3	173/176	-45.36 [-52.86, -37.85]	4	204/157	-18.68 [-22.28, -15.09]
Full Cycle Lateral Bending	2	127/128	-25.37 [-29.97, -20.77]	3	96/104	-11.35 [-15.09, -7.61]
Flexion	12	732/579	-12.55 [-15.43, -9.66]	8	176/186	-7.93 [-11.11, -4.75]
Extension	12	732/579	-20.65 [-24.69, -16.61]	8	176/186	-10.95 [-15.68, -6.22]
Right Rotation	12	665/583	-15.82 [-20.34, -11.29]	6	117/133	-8.36 [-10.85, -5.88]
Left Rotation	12	665/583	-16.56 [-21.08, -12.03]	6	117/133	-10.47 [-15.37, -5.57]
Right Lateral bending	7	504/414	-8.10 [-11.33, -4.87]	4	82/71	-4.65 [-6.59, -2.71]
Left Lateral bending	7	504/414	-9.24 [-12.78, -5.70]	4	82/71	-5.85 [-8.28, -3.41]

aCROM = active Cervical Range of Motion, #Studies = number of studies, N = number of patients, WAD = whiplash associated disorders, MD = mean difference, 95% CI = 95% confidence interval, NP = nonspecific neck pain

Table 4. Pooled Mean Differences of Acute neck pain groups compared to Healthy Controls and Chronic neck pain groups compared to Healthy Controls

Movement	#Studies	Acute/HC (n)	MD(°) (95% CI) Acute - HC	#Studies	Chronic/HC (n)	MD(°) (95% CI) Chronic - HC
Total aCROM	2	70/91	-63.31 [-78.69, -47.93]	2	214/132	-112.30 [-123.91, -100.70]
Full Cycle Flexion-extension	0	0	-	3	168/128	-28.46 [-43.68, -13.25]
Full Cycle Rotation	2	120/60	-10.11 [-13.71, -6.51]	4	286/180	-30.36 [-43.32, -17.40]
Full Cycle Lateral Bending	2	120/60	-6.55 [-7.70, -5.41]	2	132/79	-15.74 [-23.79, -7.69]
Flexion	4	170/131	-9.96 [-12.65, -7.27]	13	682/567	-12.97 [-16.72, -9.21]
Extension	4	170/131	-20.84 [-27.20, -14.48]	13	682/567	-17.33 [-21.65, -13.00]
Right Rotation	2	50/71	-17.91 [-28.28, -7.54]	13	705/592	-13.73 [-17.88, -9.58]
Left Rotation	2	50/71	-17.91 [-23.80, -12.02]	13	705/592	-15.01 [-18.98, -11.04]
Right Lateral bending	2	50/71	-10.31 [-13.89, -6.73]	8	509/400	-6.75 [-10.01, -3.50]
Left Lateral bending	2	50/71	-11.97 [-15.28, -8.66]	8	509/400	-7.21 [-9.99, -4.43]

aCROM = active Cervical Range of Motion, #Studies = number of studies, N = number of patients, WAD = whiplash associated disorders, MD = mean difference, 95% CI = 95% confidence interval, NP = nonspecific neck pain

Figure 2. Neck Pain patients versus Healthy Controls: total aCROM.

Abbreviations: df, degrees of freedom; IV, inverse variance; RoB, risk of bias

Figure 3. Neck Pain patients versus Healthy Controls: Full Cycle aCROM. Above: total flexion-extension, middle: total rotation, below: total lateral bending. Abbreviations: df, degrees of freedom; IV, inverse variance; RoB, risk of bias

Figure continues

Figure 4. Neck Pain patients versus Healthy Controls: Half Cycle aCROM, flexion and extension.

Abbreviations: df, degrees of freedom; IV, inverse variance; RoB, risk of bias.

1.3.3 Extension

Armstrong 2005	54.8	14.7	23	75.6	13.1	23	4.1%	-20.80 [-28.85, -12.75]
Branney 2014	51	7.2	29	56	6.6	30	4.9%	-5.00 [-8.53, -1.47]
Dall'Alba 2001	36.9	12.4	114	51.8	11	89	5.0%	-14.90 [-18.13, -11.67]
Fernandez-Perez 2012	53.8	22.6	20	83	10.5	20	3.4%	-29.20 [-40.12, -18.28]
Grip 2007a	43.6	18.4	21	59.3	13.8	12	3.4%	-15.70 [-26.79, -4.61]
Grip 2007b	30	19.4	22	59.3	13.8	12	3.4%	-29.30 [-40.56, -18.04]
Guo 2012	70.8	15.4	27	79.4	11.7	13	3.9%	-8.60 [-17.21, 0.01]
Kaale 2007	65	18.7	87	75	16.7	29	4.2%	-10.00 [-17.24, -2.76]
Kasch 2001	63.8	16	40	71.6	14	40	4.4%	-7.80 [-14.39, -1.21]
Ohberg 2003	30.94	12.03	56	49.85	9.17	52	4.9%	-18.91 [-22.93, -14.89]
Osterbauer 1996a	43	5	5	72	9	15	4.4%	-29.00 [-35.32, -22.68]
Osterbauer 1996b	47	16	25	71	8	36	4.3%	-24.00 [-30.79, -17.21]
Pereira 2008	46.5	18.4	30	66.5	13.8	30	4.0%	-20.00 [-28.23, -11.77]
Prushansky 2006	28.9	13.3	101	64.1	15.5	75	4.8%	-35.20 [-39.56, -30.84]
Puglisi 2004	50.1	14.1	129	66.4	14	126	4.9%	-16.30 [-19.75, -12.85]
Sarig Bahat 2010	43.1	15	25	44.3	10.9	42	4.3%	-1.20 [-7.94, 5.54]
Schops 1997	56.5	6.3	20	71.3	4.6	20	4.9%	-14.80 [-18.22, -11.38]
Shahidi 2012	56	13	19	70	14	20	4.0%	-14.00 [-22.47, -5.53]
Sterling 2004a	47.4	2	36	57.7	3	7	5.1%	-10.30 [-12.62, -7.98]
Sterling 2004b	36.4	2	32	57.7	3	7	5.1%	-21.30 [-23.63, -18.97]
Sterling 2004c	31	4.3	12	57.7	3	6	4.9%	-26.70 [-30.12, -23.28]
Vogt 2007	44.3	18.4	16	67.3	10.2	18	3.6%	-23.00 [-33.17, -12.83]
Wirth 2014	56	16.1	19	65.8	11.1	19	3.9%	-9.80 [-18.59, -1.01]
Subtotal (95% CI)			908			741	100.0%	-17.48 [-20.95, -14.01]

Heterogeneity: $\tau^2 = 60.34$; $\chi^2 = 260.66$, $df = 22$ ($P < 0.00001$); $I^2 = 92\%$

Test for overall effect: $Z = 9.87$ ($P < 0.00001$)

1.3.4 Extension Low RoB

Armstrong 2005	54.8	14.7	23	75.6	13.1	23	9.0%	-20.80 [-28.85, -12.75]
Branney 2014	51	7.2	29	56	6.6	30	12.1%	-5.00 [-8.53, -1.47]
Dall'Alba 2001	36.9	12.4	114	51.8	11	89	12.2%	-14.90 [-18.13, -11.67]
Fernandez-Perez 2012	53.8	22.6	20	83	10.5	20	7.2%	-29.20 [-40.12, -18.28]
Kaale 2007	65	18.7	87	75	16.7	29	9.6%	-10.00 [-17.24, -2.76]
Kasch 2001	63.8	16	40	71.6	14	40	10.1%	-7.80 [-14.39, -1.21]
Pereira 2008	46.5	18.4	30	66.5	13.8	30	8.9%	-20.00 [-28.23, -11.77]
Puglisi 2004	50.1	14.1	129	66.4	14	126	12.1%	-16.30 [-19.75, -12.85]
Sarig Bahat 2010	43.1	15	25	44.3	10.9	42	10.0%	-1.20 [-7.94, 5.54]
Shahidi 2012	56	13	19	70	14	20	8.8%	-14.00 [-22.47, -5.53]
Subtotal (95% CI)			516			449	100.0%	-13.26 [-17.60, -8.91]

Heterogeneity: $\tau^2 = 37.39$; $\chi^2 = 53.71$, $df = 9$ ($P < 0.00001$); $I^2 = 83\%$

Test for overall effect: $Z = 5.98$ ($P < 0.00001$)

Figure 4 continued. Neck Pain patients versus Healthy Controls: Half Cycle aCROM, flexion and extension. Abbreviations: df, degrees of freedom; IV, inverse variance; RoB, risk of bias.

Figure 5. Neck Pain patients versus Healthy Controls:

Half Cycle aCROM, rotation. Abbreviations: df, degrees of freedom; IV, inverse variance; RoB, risk of bias.

Figure 6. Neck Pain patients versus Healthy Controls: Half Cycle aCROM, lateral bending. Abbreviations: df, degrees of freedom; IV, inverse variance; RoB, risk of bias.

Figure 7. WAD versus non-traumatic neck pain.

Abbreviation: IV, inverse variance.

APPENDIX 1

The search terms from MEDLINE (Ovid), EMBASE, Web-of-science, CINAHL (EBSCOhost), Cochrane Central (Wiley), PubMed (the subset as supplied by publisher) and Google Scholar electronic database searches are found below.

Embase.com

('neck pain'/de OR 'whiplash injury'/de OR 'neck injury'/de OR (((neck OR Cervical) NEAR/3 (pain* OR ache OR hyperextens* OR injur*)) OR neckache* OR Cervicalgia* OR Cervicodynia* OR whiplash*):ab,ti) AND ('range of motion'/de OR 'joint mobility'/de OR ((range* NEAR/3 (motion* OR movement*)) OR mobilit* OR (neck* NEAR/3 (flexib* OR inflexib* OR movement* OR dysfunction* OR function*)) OR rom):ab,ti) NOT ([animals]/lim NOT [humans]/lim) NOT ([Conference Abstract]/lim OR [Conference Paper]/lim OR [Note]/lim OR [Letter]/lim OR [Editorial]/lim OR [Erratum]/lim)

Medline (Ovid)

("neck pain"/ OR exp "neck injuries"/ OR (((neck OR Cervical) ADJ3 (pain* OR ache OR hyperextens* OR injur*)) OR neckache* OR Cervicalgia* OR Cervicodynia* OR whiplash*).ab,ti.) AND ("Range of Motion, Articular"/ OR ((range* ADJ3 (motion* OR movement*)) OR mobilit* OR (neck* ADJ3 (flexib* OR inflexib* OR movement* OR dysfunction* OR function*)) OR rom).ab,ti.) NOT (exp animals/ NOT humans/) NOT (case reports OR comment OR editorial OR letter OR congresses OR news OR practice guideline).pt.

Cochrane Central (Wiley)

(((((neck OR Cervical) NEAR/3 (pain* OR ache OR hyperextens* OR injur*)) OR neckache* OR Cervicalgia* OR Cervicodynia* OR whiplash*):ab,ti) AND (((range* NEAR/3 (motion* OR movement*)) OR mobilit* OR (neck* NEAR/3 (flexib* OR inflexib* OR movement* OR dysfunction* OR function*)) OR rom):ab,ti)

Web-of-science

TS=((((neck OR Cervical) NEAR/3 (pain* OR ache OR hyperextens* OR injur*)) OR neckache* OR Cervicalgia* OR Cervicodynia* OR whiplash*)) AND (((range* NEAR/3 (motion* OR movement*)) OR mobilit* OR (neck* NEAR/3 (flexib* OR inflexib* OR movement* OR dysfunction* OR function*)) OR rom)))

CINAHL (EBSCOhost)

(MH "neck pain+" OR MH "neck injuries+" OR (((neck OR Cervical) N3 (pain* OR ache OR hyperextens* OR injur*)) OR neckache* OR Cervicalgia* OR Cervicodynia* OR whiplash*)) AND (MH "Range of Motion+" OR ((range* N3 (motion* OR movement*)) OR mobil* OR (neck* N3 (flexib* OR inflexib* OR movement* OR dysfunction* OR function*)) OR rom)) NOT (MH animals+ NOT MH humans+) NOT PT (case reports OR comment OR editorial OR letter OR congresses OR news OR practice guideline)

PubMed (publisher)

("neck pain"[mh] OR "neck injuries"[mh] OR (((neck OR Cervical) AND (pain*[tiab] OR ache OR hyperextens*[tiab] OR injur*[tiab])) OR neckache*[tiab] OR Cervicalgia*[tiab] OR Cervicodynia*[tiab] OR whiplash*[tiab])) AND ("Range of Motion, Articular"[mh] OR ((range*[tiab] AND (motion*[tiab] OR movement*[tiab])) OR mobil*[tiab] OR (neck*[tiab] AND (flexib*[tiab] OR inflexib*[tiab] OR movement*[tiab] OR dysfunction*[tiab] OR function*[tiab])) OR rom)) NOT (animals[mh] NOT humans[mh]) NOT (case reports[pt] OR comment[pt] OR editorial[pt] OR letter[pt] OR congresses[pt] OR news[pt] OR practice guideline[pt]) AND publisher[sb])

Google Scholar

"neck|Cervical pain|ache"|neckache|whiplash "range of motion"|mobility|
"neck flexibility|inflexibility|movement|dysfunction|function"

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Chapter 3

MEASUREMENT OF CLINICALLY RELEVANT FUNCTIONAL HEALTH PERCEPTIONS IN PATIENTS WITH WHIPLASH-ASSOCIATED DISORDERS: THE DEVELOPMENT OF THE WHIPLASH SPECIFIC ACTIVITY AND PARTICIPATION LIST (WAL)

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ABSTRACT

Purpose

The International Classification of Human Functioning Disability and Health (ICF) provides insight into functional health status in patients with whiplash-associated disorders (WAD). In the assessment of functional limitations in patients with WAD, there are several condition-specific questionnaires available. Estimation of the true relationship between the separate constructs of the ICF is only possible if the items of the salient questionnaires measure exactly the constructs of interest, while not simultaneously measuring other constructs of the model. This study aimed to develop a condition specific and clinically relevant and usable instrument for patients with WAD that measures activity limitations and participation restrictions, as defined by the ICF framework.

Methods

Item generation consisted of (1) a semi-structured interview which was conducted among 69 WAD patients; (2) a Delphi study involving 13 health professionals experienced in the assessment of patients with WAD; (3) a literature search for items from self-assessment questionnaires for neck pain.

Results

A 35-item condition-specific self-assessment questionnaire for patients with WAD was developed. This new questionnaire measures purely activity limitations and participation restrictions according to the ICF and is based on patients' opinions and expert opinions.

Conclusion

The whiplash activity and participation list tends to measure clinically relevant activity limitations and participation restrictions in WAD patients.

INTRODUCTION

A fair proportion of patients with whiplash-associated disorders (WAD) experience chronic complaints [1], which may interfere with patients' jobs, everyday activities, and leisure time pursuits [2]. WAD literature traditionally tends to focus on measurement of impairments, especially of pain, cervical muscle function, and cervical range of motion [3]. Evidence, however, shows that other factors, such as personal factors, activity limitations, and participation restrictions, are equally important [4].

The use of a theoretical framework for functioning and health can be helpful in the clinical decision making and provides insight into functional health status in patients with WAD [5]. The International Classification of Human Functioning Disability and Health (ICF) seems to provide such a framework [5]. The ICF contains three main constructs (components) of health: body functions and structures, activity and participation and their respective opposites. Personal factors and environmental factors are additional components of the ICF [5].

Understanding of functioning and health in WAD requires an appreciation of the interrelationship between clinically relevant constructs and additional components [6]. Estimation of the true relationship between separate constructs is only possible if the items of the salient questionnaires measure the constructs purely, while not simultaneously measuring other constructs of the model [6]. A self-administrated questionnaire is favorable in both health assessment and outcome measurement [7]. A self-administrated, condition-specific instrument should meet the following criteria: (1) the instrument is developed within a conceptual framework, in this case the ICF; (2) the instrument measures exactly the constructs of interest; (3) the measure should have a good content and construct validity; (4) the instrument is clinically applicable and acceptable, both for patients and health professionals [8].

In the assessment of patients with WAD, there are several condition-specific questionnaires available [9-15]. While the clinometric properties of these questionnaires have been studied in patients with neck pain, their validity, specifically for assessing activity and participation in WAD, has not been established. Existing questionnaires do not meet the above-mentioned second and third criteria, as they measure multiple domains of the ICF and do not include a representative spectrum of restrictions deemed to be important by WAD patients [7]. The neck disability index (NDI), for instance, contains four items on

impairments, and six items on activity/participation [16]. The whiplash disability questionnaire (WDQ) contains four functions, six items on activity/participation, and three items on personal factors. All existing questionnaires on WAD provide a sum score. The sum scores are based on the sub scores in several domains of the ICF. Estimation of the true relationship between separate constructs is only possible if the items of the salient questionnaires measure the constructs purely, while not simultaneously measuring other constructs of the model. It is, therefore, difficult to make inferences on the relationship between impairments, activity limitations/participation restrictions and personal factors.

As a consequence, there has, until now, been no condition-specific instrument available by which activity limitations and participation restrictions could be assessed in patients with WAD, without contamination of other ICF domains [2]. Until now, clinicians and researchers had to rely on generic disability measures, which are often not sufficiently comprehensive and have less discriminative capacities [15]. Therefore, there is a need for a condition-specific questionnaire that measures solely the constructs of activity and participation.

When developing a condition-specific questionnaire, the development process should be based both on patients' perceptions, experiences, and opinions, as well as on experts' opinions and experiences [8]. This study aimed to develop a condition specific and clinically relevant and usable instrument for patients with WAD that measures activity limitations and participation restrictions, as defined by the ICF framework.

METHODS

A new condition-specific self-assessment questionnaire for patients with WAD, measuring activity and participation (WAL), was developed. Item collection was established via three main separate yet interlinked procedures: (1) items were obtained via patients with WAD; (2) items were obtained via experts in the field of diagnosis and treatment of patients with WAD; and (3) selected items were compared with existing questionnaires aiming to measure perceptions of activity limitations and/or participation restrictions in patients with neck pain or WAD. Items obtained from the patients were combined with the items obtained via the experts, and compared with items from the existing questionnaires. In order to assess the feasibility of the WAL, we performed a pilot study (Fig. 1).

Fig. 1 Design of the study.

WAD whiplash-associated disorders, MACTAR McMaster Toronto arthritis patient preference disability questionnaire, ICF International Classification of Functioning, Disability and Health, WAL the whiplash activity and participation list

Procedure 1: Item collection via patients with WAD

Patients with sub-acute or chronic WAD provided demographic data, and completed the NDI and the Neck Bournemouth Questionnaire (NBQ). NDI and NBQ scores were compared with existing databases.

For the development of the new questionnaire, items were selected based on replies to the adapted version of the McMaster Toronto arthritis patient preference disability questionnaire (MACTAR) [17]. The MACTAR is a semi-structured interview. It provides insight into problems of physical function that really matter to patients [18]. It was used in all patients as part of a routine assessment in clinical practice, in rehabilitation centres and in primary care settings. Each patient was asked to identify a maximum of ten of the most problematic activities or participation restrictions that he/she experienced due to WAD. Subsequently, the patients were asked to rate the activities according to their importance, and to score the level of limitation of each item on a numerical rating scale from 0 to 10. Inclusion of patients was continued until saturation of new items was established. All patients signed for informed consent.

After completion of item collection, all items obtained by the MACTAR that did not match with the definitions of activity limitations or participation restriction, were removed from the list. The remaining items were then linked to the ICF according to established linking rules of Cieza et al. [19], by three of the authors (MSch, MSt, PS) during a consensus meeting. A priori, the decision was made to include all items mentioned by 10% or more of all participating patients.

The study was approved by the Medical Ethics Committee of the University Medical Center Utrecht.

Procedure 2: Item collection via experts

Using an electronic mail survey [20], a three-round Delphi study was conducted among physiotherapists, physiatrists, and occupational therapists. Prior to the first round of the Delphi study, seven physiotherapists and two occupational therapists who were not involved in the Delphi study, made a selection of all ICF activities and participation items that could be relevant for patients with WAD. These experts in the field of WAD were asked to rate all chosen items on a 5-point Likert scale. All items scored with a median Likert score of 3 or higher, were included and used in the first Delphi round (153 items).

Potential participants of the Delphi study were recruited via the professional networks of the authors. Inclusion criteria were (1) professional background as a

physiatrist, physiotherapist, or occupational therapist; (2) more than 5 years of clinical experience in the diagnosis and treatment of WAD patients; (3) treatment of at least three new WAD patients a month; and (4) ability and willingness to respond to the electronic mail survey for each Delphi round within 4 weeks.

Round 1 of the Delphi study

In the first round, the aims of the Delphi study were explained and information on the experts was collected. The experts were asked to select items that were relevant for measuring activities and participation WAD patients. Selection was done by clicking on "yes, this item is relevant", or clicking on "no, this item is not relevant". The number of items to be selected was not a priori limited. Experts were given the opportunity to suggest additional items. The level of in- and exclusion for round 2 was a priori set at 80% agreement among the experts, i.e., items were included in the list.

Round 2 of the Delphi study

In the second round, the experts were provided with their own scores and the group score. The scores were coded for the sake of anonymity. Experts were then asked to assess the importance of the selected items of round 1. A 7-point Likert scale (1, not important; 7, most important) was used. In addition, participants were given the opportunity to select a maximum of ten items out of the residual items which did not reach agreement in the first round (<80%) to still add to the third round. Prior to round 2, it was decided that items were to be included when more than 80% of the experts scored 5 or more on the Likert scale.

Round 3 of the Delphi study

The coded scores and the group scores (median and range) and the concept item list, were sent to the experts, who were asked to indicate, using "yes" or "no", which of the items absolutely had to be included in the questionnaire. There was no restriction as to number of items that could be selected, but experts were asked to weight their decisions based on the following criteria: (1) all questionnaire items must be relevant for measuring problem perceptions in activities and participation by WAD patients; (2) the questionnaire must be applicable in clinical practice, especially in respect to decision making; (3) the questionnaire must be clinically relevant and acceptable, for both patients and professionals. In addition, the experts were asked to indicate which scoring system would be the most appropriate for the items specifically and for the questionnaire as a whole.

Procedure 3: Literature search

Results of both item collections (via patients and experts) were compared with those of existing questionnaires that include items that assess activity limitations and/or participation restrictions. This comparison was done to examine whether there were items lacking in the concept questionnaire (WAL). A literature search was performed in MedLine, Cinahl, and EMBASE.

Pilot study

In a pilot study, patients were asked to complete the WAL and to appreciate the questionnaire. Patients rated the individual items in terms of relevance to their daily living and clarity using a 0-10 scale, and their appreciation on the whole questionnaire, with respect to wording, clarity, and layout.

RESULTS

Procedure 1

A total of 69 patients participated in the study, 53 women (77%) and 16 men (23%), median age 42 (range 21-68) (Table 1). Mean and range of the NDI scores of our sample were slightly higher than reference data [21-23]. Mean range of the NBQ scores were not different from reference data [24] (Table 1). In all collected MACTAR registration forms, 178 activity limitations or participation restrictions were mentioned. After grouping, rewording, and linking to the ICF, 132 items remained. Of these 132 items, all items suggested by 10% or more of all participating patients were included in the concept list (Table 2). Items with nearly a 10% score were only included if they scored high with the experts.

Procedure 2

Twenty-five experts were invited to participate and 18 responded to the invitation, 13 of which met the inclusion criteria and agreed to participate (8 physical therapists, 3 occupational therapists, and 2 physiatrists). The characteristics of the experts are described in Table 3. All experts participated in all three rounds of the Delphi study.

Based on the 80% agreement level, 65 items were included during the first round of the Delphi study. No extra items were added to the list by the participants. As a result of round 2, 23 items were included. In round 3, one item was chosen by <20% of the experts and was consequently removed, leaving 22 items.

Four suggestions for the questionnaire-scoring system were put forward. Five experts suggested a 5-point Likert scale, three experts suggested a 7-point Likert scale, and four suggested an 11-point Likert scale while one suggested a visual analogue scale (VAS).

Procedure 3

Thirteen self-administered questionnaires that are used for the clinical assessment or evaluation of patients with neck pain, were found. Eight questionnaires [the extended Aberdeen spine pain scale (APS) [9], NBQ [10], cervical spine outcome questionnaire (CSOQ) [11], Copenhagen neck functional disability scale (CNFDS) [25], current perceived health 42 profile (CPH42) [12], NDI [13], sickness impact profile (SIP) [14], and the WDQ] [15], were selected by three authors (MSch, MSt, PS), based on full text articles, and after judging the original

questionnaires. This selection was compared with the selection of self-assessment questionnaires by Nordin et al. [7]. Two questionnaires reported by Nordin et al. (problem elicitation technique, and VAS) were not included in this study, because they do not contain a fixed number of activity limitations and/or participation restrictions. In a consensus meeting with three of the authors (MSch, MSt, PS), questionnaire items which seem to measure activity limitations and/or participation restrictions, were selected, using the ICF linking rules [23].

Twenty-six activity limitations and/or participation restrictions were selected from literature and compared with the results of procedure 1 and procedure 2 (see Table 2). Compared to this list of activity limitations and participation restrictions suggested by experts and by patients, no new items were found in the screened eight questionnaires.

The items suggested by patients (28) and experts (22) were combined and reworded in the language that was applied by the patients. Double items (15) were removed. This resulted in a 35-item questionnaire. A scoring system for the WAL was chosen based on suggestions of the experts, and was also based on the ICF severity score (0, no problem; 1, mild problem; 2, moderate problem; 3, severe problem; 4, complete problem).

Pilot study

Thirteen patients with WAD were invited to participate. Ten (77%) agreed to participate (8 women, 2 men). Median age was 42.5 (range 27-60). The questionnaire was found to be relevant and understandable to these patients. The mean score for relevance of the individual items was 7.3 out of 10 (range 5.1-9.2), and the mean score for comprehensibility of the separate items was 8.7 (range 7.7-9.2). The mean score of the general appreciation of the total questionnaire was 7.8 (range 6-10), and the score for the instruction of the questionnaire was 6.8 (range 4.0-8.0). The mean appreciation of the scoring method was 7.3 (range 2-10). In response to the results of the pilot study, some minor revisions were made in the wording of the items and the instruction.

Table 1 Demographic data patients with WAD, time since injury, and type of injury (*N* = 69)

	Women (<i>N</i> = 53) 76.8%	Men (<i>N</i> = 16) 23.2%
Age (years): median (range)	40 (22-64)	49.5 (21-68)
Time since collision (months) (median, range)	12 (2-120)	9 (3-42)
Type of injury (n)		
Rear end collision	42	12
Front collision	2	1
Side impact collision	2	
Sport injury	2	
Other injury	5	3
Total NDI score		
Mean (SD)	26.9 (7.9)	23.4 (8.9)
Range	10-47	8-39
Total NBQ score		
Mean (SD)	46.3 (13.8)	40.5 (14.3)
Range	12-69	20-61

WAD whiplash-associated disorders, *NDI* neck disability index, *NBQ* neck Bournemouth questionnaire, *SD* standard deviation

Table 2 Activity limitations (AL) and participation restrictions (PR) suggested by patients ($N = 69$) (after removing <10% suggestions patients) and experts ($N = 13$); compared with questionnaires for the evaluation of function and disability in patients with cervical pain

Activity limitation/ participation restriction	ICF code	Number of patients (%) who suggested the AL/PR	Number of experts (%) who suggested the AL/PR	Questionnaires which contain suggested items by experts and patients
<i>Learning and applying knowledge</i>				
Focusing attention	d160		12 (92.3)	SIP
Reading	d166	22 (31.9)	10 (76.9)	CNFDS; NDI; NBQ; NPNPQ;
Solving complex problems	d1751		6 (46.2)	
<i>General tasks and demands</i>				
Undertaking multiple tasks	d220	10 (14.5)	10 (76.9)	CPH42
Handling stress and other psychological demands	d240		12 (92.3)	
<i>Communication</i>				
Conversation	d350	10 (14.5)		
Using writing machines	d3601	13 (18.8)		SIP
<i>Mobility</i>				
Bending	d4105	7 (10.1)	3 (23.1)	CPH42; SIP
Maintaining a sitting position	d4153	15 (21.7)		SIP
Transferring oneself, unspecified ^a	d4209		11 (84.6)	
Lifting and carrying objects	d430	36 (52.2)	9 (69.2)	CNFDS; NDI; NBQ; NPNPQ; SIP
Reaching	d4452	13 (18.8)		CPH42
Catching	d4455		1 (7.7)	
Walking and moving	d450	12 (17.4)		SIP
Moving around	d455		3 (23.1)	
Climbing	d4551	11 (16.0)		CPH42; SIP
Running	d4552	12 (17.4)	5 (38.5)	
Jumping	d4553		5 (38.5)	
Using transportation	d470	8 (11.6)		
Driving human-powered transportation	d4750	14 (20.3)		
Driving motorized vehicles	d4751	34 (49.3)	9 (69.2)	NDI; NPNPQ; WDQ; EABPS; NBQ
<i>Self-care</i>				
Washing oneself	d510			NDI; WDQ; SIP
Caring for body parts	d520		9 (69.2)	CNFDS
Dressing	d540	13 (18.8)		CNFDS; NDI; WDQ; SIP; NBQ
<i>Domestic life</i>				
Shopping	d6200	11 (16.0)	8 (61.5)	
Household tasks	d630	14 (20.3)	3 (23.1)	SIP
Doing housework	d640		11 (84.6)	SIP; NBQ
Cleaning living area	d6402	24 (34.8)		SIP
Using household appliances	d6403	28 (40.6)		
Disposing of garbage	d6405	8 (11.6)		
<i>Interpersonal interactions and relationships</i>				
Basic interpersonal interactions	d710	8 (11.6)		
Complex interpersonal interactions	d720	11 (16.0)		CNFDS; NPNPQ; WDQ; NBQ
Family relationships	d760	12 (17.4)		
Sexual relationships	d7702	11 (16.0)	7 (53.8)	EABPS; SIP

Table continues

Table 2 continued.

Activity limitation/ participation restriction	ICF code	Number of patients (%) who suggested the AL/PR	Number of experts (%) who suggested the AL/PR	Questionnaires which contain suggested items by experts and patients
<i>Major life areas</i>				
School education	d820		6 (46.2)	WDQ
Remunerative employment	d850	27 (39.1)	13 (100)	NDI; NPNPQ; WDQ; EABPS: NBO
<i>Community, social and civic life</i>				
Recreation and leisure	d920		12 (92.3)	EABPS; SIP; NBQ
Community life	d910		6 (46.2)	
Sports	d9201	56 (81.2)	9 (69.2)	WDQ
Arts and culture	d9202	7 (10.1)		SIP
Socializing	d9205	7 (10.1)		CPH42; SIP; NBQ

MACTAR McMaster Toronto arthritis patient preference disability questionnaire, *CNFDS* Copenhagen neck functional disability scale; *CPH42* current perceived health 42 profile, *EABPS* extended Aberdeen back pain scale, *NDI* neck disability index, *NBQ* neck Bournemouth questionnaire, *NPNPQ* Northwick Park neck pain questionnaire, *SIP* sickness impact profile, *WDQ* whiplash disability questionnaire, *ICF* The International Classification of Human Functioning Disability and Health

^a Suggested by the experts: rotating the head and the cervical spine while sitting in a car

Table 3 Characteristics of the experts (*N* = 13)

Mean age in years (range)	43.1 (29-54)
Profession	
Physical therapist	8
Occupational therapist	3
Physiatrist	2
Work setting (number)	
Rehabilitation center	8
Primary care setting	5
Mean of number of WAD patients per month (range)	5.2 (3-10)
<i>WAD</i> whiplash-associated disorders	

DISCUSSION

The aim of this study was to develop a condition-specific questionnaire for WAD patients, which measures activity limitations and participation restrictions, in accordance with the ICF.

In literature, there is an on-going discussion concerning the use of diagnostic instruments that are based on a theoretical framework, i.e., the ICF. Existing questionnaires for WAD are not primarily based on the ICF. If a questionnaire is not primarily based on the ICF, it is difficult to compare components of functioning and health, like "impairments" versus "activity limitations". This new questionnaire has a potential advantage over existing questionnaires because the instrument is developed within the conceptual framework, and measures the constructs of the framework purely.

The WAL was based on expert opinions, patients' experiences, and on existing questionnaires, meaning that it has face and content validity. The WAL is condition-specific, is framed within the ICF and is clinically applicable (brevity and simplicity). The ICF allows clinicians to comprehensively describe and categorize functioning and disability in a systematic and standardized way. An instrument that uses the ICF as conceptual framework allows clinicians to assess severity of the health problem and facilitates clinical reasoning [5].

Our study does have its limitations though. Firstly, all the patients who participated in this study followed regular treatment programs for physiotherapy, which might have been of influence on the suggested items they provided. And, it is unknown whether the sample in our study is representative for the population of patients with WAD, as we did not check clinical characteristics of patients who refused to participate in our study. There is possibly some selection bias. In addition, patients' suggestions were based on MACTAR data. Clinicians should be adequately trained on how to use the MACTAR correctly. In our study, we did not check the level of experience with the MACTAR. A different use of the MACTAR can lead to biased data. The MACTAR is normally used and validated for other types of conditions. A parallel study, however, showed the appropriateness of the methodology [22, 23]. Furthermore, we used a Delphi method to obtain expert opinions. The success of such a strategy rests on characteristics of the experts. We did not use sound criteria for the assessment of the participants' level of expertise. In the Delphi study, the cut-off point for including items was set at 10%, which is arbitrary. To our knowledge no methodological criteria are available concerning decision

making in item selection in relation to the development of a questionnaire. It is worthwhile to develop such criteria.

Prior to using the WAL in practice it has to be validated in a representative sample of the population. Therefore, the next steps in the development of this instrument are to evaluate its reproducibility and construct validity, both cross-sectional and longitudinal, and to estimate the minimally important change score.

In conclusion, the WAL measures activity limitations and participation restrictions in patients with WAD. The WAL is a first step in the development of a clinically applicable instrument. Further research must focus on the validation and generalizability of this questionnaire.

APPENDIX

Concept version of the whiplash activity and participation list (WAL)

Focusing attention
Reading
Solving complex problems
Undertaking multiple tasks
Handling stress
Conversing
Using a desktop computer or laptop
Bending over
Maintaining a sitting position
Looking over the shoulder
Lifting and carrying objects
Prolonged walking
Running
Overhead work
Using public transportation (bus, train or subway)
Cycling
Driving a motor vehicle (such as an automobile or motorcycle)
Wash or shower
Caring for body parts (such as face, teeth) or washing hair
To dress or undress
Shopping
Preparing meals (slicing, cooking, etc.,)
Doing housework (cleaning, washing clothes, window cleaning, etc.,)
Using household appliances (such as vacuum cleaners)
Gardening
Interacting with people (such as friends, co-workers, partner, and children)
Maintaining relationships
Sexuality
Following education or training
Work (maintaining a job, performing volunteer work)
Engaging in recreational activity (such as sightseeing or visiting an amusement park)
Engaging in social activities (such as organized religious ceremonies, political events or activities in a social club)
Sports
Going out (such as going to the theater, cinema or museum)
Visiting friends or relatives

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Chapter 4

VALIDATION OF A NEW QUESTIONNAIRE TO ASSESS THE IMPACT OF WHIPLASH ASSOCIATED DISORDERS: THE WHIPLASH ACTIVITY AND PARTICIPATION LIST (WAL)

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ABSTRACT

Background

Valid questionnaires for measuring functional limitations in patients with Whiplash Associated Disorders (WAD) are lacking, since existing measures are not suitable for addressing the specific limitations of these patients and because of cross contamination between theoretical constructs.

Objective

The objective of this study was to evaluate dimensionality, test-retest reliability, measurement error, construct validity, and responsiveness of a new condition-specific questionnaire for WAD as well as to estimate the minimally important change score.

Methods

Patients with WAD grade I or II were recruited from physical therapy practices and rehabilitation centers. Dimensionality was examined by internal consistency analysis (Cronbach's alpha) and factor analysis. Test-retest reliability was estimated by intraclass correlations and measurement error was calculated by the minimal detectable change (MDC) scores. Construct validity was investigated by testing predefined hypotheses on correlations of the WAL scores with generic health measures and by using the known group method. Responsiveness was expressed as the minimal clinically important change (MCIC) score.

Results

73 patients (53 women) were included. Cronbach's alpha was high (0.95) and unidimensionality was plausible because factor analysis showed 40.3% variance explained by one dominant factor, which was more than 4.5 times larger than the second largest factor. Test-retest reliability was excellent (0.92, 95%CI 0.87-0.95). Construct validity was supported by 14 out of 15 confirmed hypotheses and the WAL showed statistically significant differences between known groups. The MDC was 16 points while the MCIC was 18 points.

Conclusions

In conclusion, the present study suggests that the WAL has adequate measurement properties, but additional research is needed.

INTRODUCTION

The incidence of Whiplash Associated Disorders (WAD) in Western countries has increased over the past 30 years. The estimated yearly incidence is at least 300 per 100,000 inhabitants (Carroll et al., 2006; Holm et al., 2008). Prognosis after whiplash injury is generally good, although a proportion of all patients will develop persistent disorders (Carroll et al., 2008; Sterling et al., 2006).

Biomechanical factors seem to have little influence on the prognosis (Richter et al., 2004; Sterner and Gerdle, 2004; Holm et al., 2008). In the assessment of the impact of WAD, it is therefore considered useful to describe patients' activities and participation level (Scholten-Peeters et al., 2002; Pinfold et al., 2004; Sterling, 2006).

The International Classification of Functioning, Disability and Health (ICF) describes constructs of health: impairment, activity limitation and participation restriction (WHO, 2001). The ICF research branch does not differentiate activity limitation from participation restriction. These constructs are influenced by environmental factors as well as personal factors (Steiner et al., 2002; WHO, 2001). When measuring ICF constructs, it is important that the measures only assess the constructs of interest and avoid cross contamination between constructs (Pollard et al., 2009).

To our knowledge, no valid measures exist for quantifying activities and participation in WAD since existing measures do not make a clear distinction between ICF constructs. Measures of activity limitation and participation restriction are contaminated with items that also measure impairments in body functions and structures (Nordin et al., 2008; Schmitt et al., 2013). Moreover, adequate content validity is a primary concern (Terwee et al., 2011; Schellingerhout et al., 2012). The present condition-specific questionnaires have potential limitations in their applicability to patients with WAD, because problems commonly identified in these patients, are frequently omitted (Hoving et al., 2003; Pinfold et al., 2004). Item generation did not involve a population with WAD nor did it seek input from clinical experts (Hoving et al., 2003; Nordin et al., 2008). We found that out of the 40 of limitations most important to patients with WAD, the most frequently used Neck Disability Index (NDI) covers only 6 items (Schmitt et al., 2013). Hoving et al. also concluded that the NDI and Northwick Park neck pain Questionnaire (NPQ) not fully reflect the full spectrum of disabilities judged to be important by patients with WAD. Only three of the most important problems were

included in the NDI and only four were included in the NPQ (Hoving et al., 2003). Existing scales may, therefore, not be suitable for the specific limitations of patients with WAD.

In the absence of a condition-specific disability outcome measure for whiplash, generic disability measures are available (Guyatt & Rennie, 2008; Pinfold et al., 2004). However, generic measures tend to measure more than one construct and cover the concerning areas superficially, which may limit the discriminative capacity (Guyatt & Rennie, 2008; Nordin et al., 2008).

We developed a new questionnaire that specifically focuses on activity limitations and participation restrictions in patients with WAD: the Whiplash Activity and participation List (WAL) (Schmitt et al., 2013). The WAL is a self-assessment instrument that is based on the ICF framework. The WAL is primarily developed for the Dutch Language, and consists of 35 activity and participation items, scored on a 5-point scale according to the ICF (WHO, 2001). A sum score is calculated ranging from 0 (no limitations) to 140 (extremely limited). The development process of the WAL consisted of separate but interlinked sub-studies for devising the items, including focus groups with experts in WAD care (Delphi study) and with patients with sub-acute or chronic WAD. All items were linked to the ICF construct activity and participation using the ICF linking rules (Cieza et al., 2002). After the items were devised, the WAL was adapted, based on the results of a pilot study (n=10). The aim of this study was to validate the WAL by assessing its measurement properties in a population of sub-acute and chronic patients with WAD.

METHODS

Participants

Recruitment of patients took place between November 2010 and July 2011 and was done via primary care physical therapists and rehabilitation centers in the Netherlands. Patients were included if they were suffering from WAD grade I or II (Spitzer et al., 1995) with symptom duration exceeding six weeks, if they were between 18 and 65 years of age, and were able to understand the Dutch written language. Potential patients received detailed information concerning the aims and procedures of the study. The study was approved by the Medical Ethics Committee of the Academic Medical Centre, University of Amsterdam, the Netherlands.

Measurements

At baseline, demographic details and disease-specific information were collected. Global level of activity limitations and participation restrictions were both assessed on a 5-point scale (*not at all, slightly, moderately, quite a bit, extremely*). Pain severity was scored on a 6-point scale (*none, very mild, mild, moderate, severe, very severe*). Subsequently, patients completed the WAL, the Short Form-36 (SF36) (Brazier et al., 1992) and the the Sickness Impact Profile (SIP68) (Post et al., 1996). To investigate whether the functional limitations of our sample were representative for the population of patients with WAD, the Neck Disability Index (NDI) was administered, and compared with existing data-bases (Hoving et al., 2003; Kaale et al., 2005). The physical therapist was instructed to check whether the patient had fully completed the questionnaires.

The SF36 is a generic Quality of Life scale that is validated for the Dutch language (Aronson et al., 1998). It contains 36 items describing eight dimensions: physical functioning (PF), social functioning (SF), role limitations due to physical health problems (RP), role limitations due to emotional problems (RE), general mental health (MH), vitality (VT), bodily pain (BP) and general health perceptions (GHP). The SIP68 is a recommended generic health status measure for research in rehabilitation medicine (Post et al., 1996). It consists of 68 items, measuring individuals' perceptions regarding "sickness impact" on daily activities, behaviors and feelings which are divided over 6 subscales: somatic autonomy (SA), mobility control (MC), psychological autonomy and communication (PAC), social behavior (SB), emotional stability (ES) and mobility range (MR). The NDI is a 10-item

questionnaire for assessment of physical disability of subjects with neck pain, and has adequate psychometric properties (Hoving et al., 2003; Pool et al., 2007; Schellingerhout et al., 2012).

After a time interval of 1-2 months, the same items and questionnaires, except for the SF36 and the SIP68, were administered. In addition to this, patients rated their neck complaints on a 5-point transition scale (*much improved, slightly improved, not changed, slightly worsened, much worsened*). Ratings on the transition scale were re-scored as improved ('much improved' was indicated as meaningfully improved), versus relatively stable ('slightly improved', 'not changed' and 'slightly worsened') versus worsened ('much worsened').

Item reduction

A qualitative item reduction was performed during the development of the preliminary WAL (Schmitt et al., 2013). For the present study, we performed a quantitative item reduction of the WAL after baseline administration. Individual items which were scored as "no problem" by more than 50% of the participants, and/or which had a mean score of <1 , and/or on which more than 10% of patients failed to respond, and/or which had a low item-rest correlation ($r < 0.20$) were considered to be potentially irrelevant for the population under study. In a consensus meeting, three researchers (MSS, MAS, CS) determined which of the individual items that were identified as being potentially irrelevant, could be removed. The resulting reduced WAL was subsequently further examined for its measurement properties.

Internal consistency

It was assumed that both activity and participation items form a common construct of health. We used Cronbach's alpha as a measure of internal consistency of the WAL. Alpha should be higher than >0.80 for comparisons at group level (Portney and Watkins, 2000; Streiner and Norman, 2008). Additionally, item-rest correlations were examined. Item-rest correlation >0.40 for all items would support unidimensionality.

In addition, a factor analysis was performed to examine whether individual items load onto a single underlying dimension. Factor extraction was performed using the Kaiser criterion (eigenvalues >1.0) and by graphing the eigenvalues in a scree-plot. Subsequently, a forced two-factor exploratory factor analysis was conducted to examine whether a two factor model was appropriate.

Test-retest reliability

Test-retest reliability of the final WAL was determined using the part of the study population that did not report change in their condition on the transition scale at follow-up (Guyatt & Rennie, 2008). Test-retest reliability was expressed using intraclass correlations (case 3, single measure). An Intraclass Correlation Coefficient (ICC) of at least 0.70 was considered to indicate acceptable reliability.

Measurement error

To assess the measurement error of the WAL, the minimal detectable change (MDC) was calculated using the 95% limit cut-off point. The MDC was estimated using the 95% upper limit of the distribution of the patients who rated themselves as 'not changed' on the transition index. The 95% limits cut-off point was calculated as the mean change + $1.645 * SD_{\text{change}}$ whereby 1.645 corresponds to 5% upper limit (one-tailed) (de Vet et al., 2007).

Construct validity

The construct validity of the final WAL was examined by correlating the WAL scores with the domain scores of the SF36 en SIP68. We evaluated both convergence with similar domains and divergence with dissimilar domains of the SF36 en SIP68. We formulated 15 predefined hypotheses (Table 1). Construct validity was considered acceptable if at least 75% of the hypotheses were confirmed. Spearman's rank correlations were used to express convergence and divergence validity.

Supporting construct validity is also provided when a test can discriminate between individuals who are known to differ in disease severity (clinical validity) (Portney & Watkins, 2000). We therefore examined whether the WAL could discriminate between patients with different levels of activity and participation as assessed on the global indices of functioning (cross-sectional known group method). To test the differences between the known groups, the Kruskal – Wallis test was used.

Table 1

Hypotheses for construct validity of the Whiplash Activity and participation List (WAL)

General

The overall WAL score has a positive correlation of at least 0.5 with the pain score

The overall WAL score has a positive correlation of at least 0.6 with the indicated activity limitations

The overall WAL score has a positive correlation of at least 0.6 with the indicated participation restrictions

SF36

The correlation between the overall WAL score and the physical functioning subscale of the SF36 is at least 0.6

The correlation between the overall WAL score and the social functioning subscale of the SF36 is at least 0.6

The correlation between the overall WAL score and the bodily pain subscale of the SF36 is at least 0.5

The total score of the WAL has at least 0.1 stronger correlation with the physical functioning subscale of the SF36 than with the general mental health component of the SF36

The total score of the WAL has a least 0.1 stronger correlation with the role limitations due to physical health problems subscale of the SF36 than with:

- the role limitations due to emotional problems subscale of the SF36
 - the vitality subscale of the SF36
-

SIP68

There is a positive correlation between the overall WAL score and the psychological autonomy and communication subscale of the SIP68 of at least 0.5

There is a positive correlation between the overall WAL score and the social behavior subscale of the SIP68 of at least 0.5

The score on the WAL should have at least 0.1 higher correlation with the social behavior subscale than with the emotional stability subscale

The score on the WAL should have at least 0.1 higher correlation with the psychological autonomy and communication subscale than with the mobility control subscale

The total score of the WAL has a low correlation (<0.3) with the somatic autonomy subscale of the SIP68.

The score on the WAL should have at least 0.1 higher correlation with the mobility range subscale than with the somatic autonomy subscale

SF36 = Short Form-36

SIP68 = Sickness Impact Profile

Responsiveness

The minimal clinically important change (MCIC) was estimated by calculating the mean change score and the optimal cut-off point of the Receiver Operating Characteristics (ROC) curve. The 5-point transition index was used in both MCIC methods as an external criterion for change, distinguishing the mentioned three subgroups: improved, relatively stable and worsened. The mean change score was calculated by subtracting follow-up scores from the baseline scores. Positive scores stand for improvement. The ROC curve method was used as a diagnostic test to distinguish between subjects who are 'improved' versus subjects who are 'not improved' on the transition scale. The ROC curve plots sensitivity (true positive rate) against the 1 – specificity (false positive rate) for each possible cut-off change score of the WAL. The optimal cut-off point of the ROC curve was chosen where the percentage of misclassifications is lowest, i.e. where the sum of sensitivity and specificity had the highest value.

The ROC curve method was also used to quantify the responsiveness of the WAL. We considered an area under the curve (AUC) of at least 0.70 to be adequate to detect real changes in patients (Terwee et al., 2011).

Floor and ceiling effects

In the evaluation of floor and ceiling effects of the WAL, we hypothesized that no more than 15% of the respondents should have the minimum or maximum score of the WAL at baseline.

Statistical analyses were performed using SPSS version 17.0. A sample size of at least 50 patients was deemed sufficient for the assessment of construct validity and reproducibility (Streiner & Norman, 2008; Terwee et al., 2007).

RESULTS

Item reduction

Ten items were identified to be potentially irrelevant. All of these items, however, showed good variance and high item-rest correlations, ranging from 0.49 to 0.70. As a result of that, in combination with the priority given by the patients to the concerning items during the item generation procedure and the items proposed by Hoving et al. (Hoving et al., 2003), it was decided that no further items should be removed. The final version of the WAL consists of 35 items on activity and participation (Appendix A and B).

Seventy-three patients were included and they all completed the questionnaires at baseline. Patients' characteristics are shown in Table 2. The NDI scores of our sample were in accordance with reference data (Hoving et al., 2003; Kaale et al., 2005). At follow-up assessment, 63 patients replied to the questionnaires (response rate 86%). Six patients indicated in advance not to be prepared to fill out the questionnaire a second time.

Internal consistency

Cronbach's alpha for internal consistency reliability was 0.95, indicating a high homogeneity of all items. All items showed item-rest correlations >0.40 (median 0.61; range 0.49-0.70) supporting unidimensionality of the WAL. The inter-item correlation matrix showed correlations ranging from 0.47 to 0.82.

Factor analysis showed that seven factors had an eigenvalue greater than 1. The first dominant factor explained 40.3% of total variance. The second factor explained another 9% of the variance. The remaining factors each explained between 6.6% and 3.0% of the total variance of 72%. A forced two-factor model showed that four items loaded approximately equally (<0.09 difference in correlation) on both factors and only one item ('Sports') loaded 0.62 on the second factor and 0.49 on the first dominant factor. Given the, in comparison, large first factor, the steepness of the scree plot and the high factor loadings we considered the scale to measure a single dimension.

Test-retest reliability

For test-retest reliability calculation, scores from 53 participants who did not report change in their condition on the transition index were used. Reliability was excellent (ICC 0.92, 95% CI 0.87-0.95).

Table 2

Characteristics of patients with scores on the WAL, NDI, pain scale and transition scale (n=73)

Characteristics	Value	
Age (yr) (mean $\pm$ SD)	43.3 $\pm$ 13.0	
Female (%)	72.6	
Time since injury (months) (median, IQR)	13.3 (6.8 – 75.2)	
	Baseline	Follow-up (n=63)
WAL score (median, IQR)*	43.0 (28.0 – 61.5)	40.0 (22.0 – 58.0)
NDI score (median, IQR) [†]	20.0 (13.5 – 25.0)	19.0 (12.0 – 23.0)
Pain score (n,%)		
None	2 (2.7)	
very mild	18 (24.7)	
mild	36 (49.4)	
moderate	14 (19.2)	
severe	3 (4.1)	
very severe	0 (0)	
Transition scale (n,%)		
much improved		8 (12.7)
slightly improved		15 (23.8)
not changed		34 (54.0)
slightly worsened		4 (6.3)
much worsened		2 (3.2)

WAL = Whiplash Activity and participation List; NDI = Neck Disability Index; SD = Standard deviation;
IQR = interquartile range

* WAL score: 35 items ranging from 0 to 4 points; worst possible score = 140 points

[†]NDI score: 10 items ranging from 0 to 5 points; worst possible score = 50 points

Measurement error

There was no systematic error on retesting, mean score difference was zero (SD 9.5). The MDC based on the 95% limit cut-off was $1.645 * 9.5 = 15.6$ points. No tendency toward unequal spreading of the data was visible.

Construct validity

Correlations concerning the hypotheses for construct validity are shown in Table 3. The correlations for the convergent validity hypotheses were all higher than the threshold (range 0.50e0.67) while the correlations for the divergent validity hypothesis did not exceed the threshold (range 0.27e0.41). Concerning the global indices of functioning, the overall WAL score had a 0.59 correlation with the pain score, a 0.70 correlation with the global scale for activity limitations, and a 0.63 correlation with the global scale for participation restrictions. Of the 15 predefined hypotheses, 14 (93.3%) were confirmed. Only the assumed correlation of the WAL with the social behavior subscale was less than 0.1 higher than with the emotional stability subscale.

Table 4 shows the differences in mean WAL score between the severity groups. The results of the Kruskal-Wallis test showed statistically significant different WAL scores between known groups ($p < 0.001$) which supports the clinical validity.

Responsiveness

The WAL change score of the 'improved' patients was larger than the MDC, the mean change score was 17.38 (SD 11.93). The MCIC determined by the optimal cut-off point of the ROC curve was 3.5 points. Corresponding sensitivity was 87.5% and specificity was 66.0%.

The ability of the WAL to distinguish between patients who were improved and those who were not improved was adequate: the AUC was 0.88 (95% CI 0.77e0.99).

Floor and ceiling effects

No floor and ceiling effects were found at baseline and follow-up assessment, because none of the patients reported the worst or the best possible score on both test occasions.

Table 3

Correlations concerning the hypotheses for construct validity (n = 73)

SF subdomain	WAL total	SIP subdomain	WAL total
SF36 PF	0.644 *	SIP68 MR	0.426
SF36 SF	0.668 *	SIP68 SA	0.267 *
SF36 RP	0.567 *	SIP68 MC	0.395 *
SF36 RE	0.337 *	SIP68 PAC	0.501 *
SF36 MH	0.373 *	SIP68 SB	0.534 *
SF36 VT	0.414 *	SIP68 ES	0.445
SF36 BP	0.655 *		
SF36 GHP	0.374		
Pain score (NDI)	0.587 *		
Global level of activity	0.700 *		
Global level of participation	0.631 *		

* Correlations confirming the hypotheses displayed in Table 1.

WAL = Whiplash Activity and participation List; SF36 = Short Form-36 (PF = physical functioning; SF = social functioning; RP = role limitations due to physical health problems; RE = role limitations due to emotional problems; MH = mental health; VT = vitality; BP = bodily pain; GHP = general health perceptions). SIP68 = Sickness Impact Profile (MR = mobility range; SA = somatic autonomy; MC = mobility control; PAC = psychological autonomy and communication; SB = social behavior; ES = emotional stability). NDI = Neck Disability Index

Table 4

Known groups method (n = 73)

Activity problems	n	Mean score* (SD)	Median* (range)	Participation problems	n	Mean score* (SD)	Median* (range)
no	3	4.67 (2.08)	4 (3-7)	no	6	22.33 (15.58)	19.5 (3-44)
mild	25	29.00 (14.5)	27 (6-62)	mild	11	24.91 (12.02)	26 (7-43)
moderate	29	52.55 (17.16)	46 (24-98)	moderate	35	44.17 (19.80)	44 (4-83)
severe	12	60.58 (22.85)	59 (23-105)	severe	17	60.00 (18.45)	61 (24-98)
very severe	4	84.50 (26.86)	74.5 (65-124)	very severe	4	88.50 (31.08)	85 (60-124)

SD = Standard deviation

*p<0.001

DISCUSSION

We investigated the measurement properties of a new condition-specific questionnaire for patients with WAD in a Dutch population. We found that the WAL has good psychometric properties, and scores at least as good as other widely used neck questionnaires, yet has better content and construct validity. We attempted to improve the methodological flaws of previous studies, including adequate methods for internal consistency, construct validity, responsiveness, and statistical methods (Pollard et al., 2009; Schellingerhout et al., 2012; Schmitt et al., 2013)

The internal consistency of the scale was high (0.95). An alpha value higher than 0.90 may suggest redundant items in the scale. However, the correlation matrix showed that the highest correlation between two items was 0.82, which does not support redundancy.

One dominant factor was extracted within the factor analysis, but a forced-two way factor analysis showed that a two factor model was also appropriate. We justified the acceptance of the one factor model, based on the fact that the item-rest correlations all exceeded 0.40, based on the high variance ratio, and on the fact that only 4 of the 35 items loaded equally on both factors and only one item loaded on the second factor. Additionally, no theoretical construct behind the two factors was recognized. However, the analysis was intended to be exploratory and should be interpreted cautiously because of the small sample size.

The WAL was able to discriminate in scores between patients with different levels of functional limitations, and was able to detect real change in health status: the MDC (16/140) was lower than the observed mean change score of 'improved' patients. The mean change score of 17.38 points is just beyond this random measurement error. The MCIC was 3.5 points as estimated by the optimal cut-off point of the ROC curve. As for the estimation of cut-off points for clinically relevant change, there is no consensus as to which method is to be preferred and these differences in results between the two methods often occur (de Vet et al., 2007; Pool et al., 2007). The ROC cut-off point of 3.5 has an advantage because in this method both false positive and false negative classifications are equally taken into account and it therefore better reflects the clinically important change (Pool et al., 2007). A substantial part of the scores of the clinically improved patients, however, will fall within the measurement error of the 'unchanged' patients. We therefore used the more conservative 95% limit cut-off method, which is more appropriate

to estimate what constitutes a clinically relevant change on the WAL. Furthermore, in case of higher baseline scores, a larger MCIC makes more sense because score changes are nonlinear at the extremes of a scale with smaller score differences indicating more change in health (de Vet et al., 2007; Pool et al., 2007). We recommend a decrease of 18 or more points to be considered clinically relevant improvement.

A remark of our study is that we used a retrospective transition scale as external criterion. There is debate about the validity of the transition scale for estimation of the MCIC. Norman et al. (1997) found that transition ratings were positively correlated with a subject's current health status but uncorrelated with the baseline score. These findings indicate that patients may not be able to recall their primary health status and the 1-2 month time interval between measurements could be considered to be quite large (Norman et al., 1997; Fletcher et al., 2010). We therefore used a conservative strategy by only indicating 'much improved' to be meaningfully improved and 'slightly improved' as unchanged, in order to distinguish between stable and changed patients. And although a transition scale is not a perfect criterion standard, Terwee et al. state that it currently should be considered the best criterion available (Terwee et al., 2011).

Validation of a questionnaire is difficult and its methodology is a matter of debate because of the absence of a gold standard or an ideal similar questionnaire. We therefore correlated the WAL with subscales of two different and widely used reliable and valid instruments for musculoskeletal disorders. Additionally we used the known group method.

It has been argued that there is no need to develop new neck-specific questionnaires until existing questionnaires have been adequately assessed (Terwee et al., 2011; Schellingerhout et al., 2012). The reason behind this is that the methodological quality of the studies for measurement properties is often poor and therefore the measurement properties as such are uncertain, especially as far as the relevance and comprehensiveness of the items is concerned. The potential low content validity of currently used questionnaires consequently calls for high quality studies, evaluating the measurement properties of these questionnaires (Terwee et al., 2011; Schellingerhout et al., 2012). We concur with this statement, which is why we have extensively assessed the content validity of the questionnaires currently used in WAD patients, prior to the development of the WAL. We concluded that the defects in the content and underlying constructs

of the existing questionnaires warranted the development of a new questionnaire for WAD patients (Schmitt et al., 2013).

In further research, the psychometric properties of the WAL should be tested against other currently used measures in a similar population of WAD patients. We recommend to test the WAL in different and larger populations of WAD patients as well as to evaluate the ability of the WAL to distinguish between other clinically relevant health statuses like employment status or general health status. In conclusion, the results of the present study suggest that the WAL has adequate measurement properties for the assessment of activity limitations and participation restrictions in patients with WAD. In addition to other known neck questionnaires, it has good content validity and fits well within the theoretical framework of the ICF for clinical reasoning. The WAL is a promising instrument for describing the state of health in primary care settings and rehabilitation centers, but additional research is needed.

APPENDIX A

The Whiplash Activity and participation List (WAL)

1	Focusing attention
2	Reading
3	Solving complex problems
4	Undertaking multiple tasks
5	Handling stress
6	Conversing
7	Using a desktop computer or laptop
8	Bending over
9	Maintaining a sitting position
10	Looking over the shoulder
11	Lifting and carrying objects
12	Prolonged walking
13	Running
14	Overhead work
15	Using public transportation (bus, train or subway)
16	Cycling
17	Driving a motor vehicle (such as an automobile or motorcycle)
18	Wash or shower
19	Caring for body parts (such as face, teeth) or washing hair
20	To dress or undress
21	Shopping
22	Preparing meals (slicing, cooking etc.)
23	Doing housework (cleaning, washing clothes, window cleaning, etc)
24	Using household appliances (such as vacuum cleaners)
25	Gardening
26	Interacting with people (such as friends, co-workers, partner and children)
27	Maintaining relationships
28	Sexuality
29	Following education or training
30	Work (maintaining a job, performing volunteer work)
31	Engaging in recreational activity (such as sightseeing or visiting an amusement park)
32	Engaging in social activities (such as organized religious ceremonies, political events or activities in a social club)
33	Sports
34	Going out (such as going to the theatre, cinema or museum)
35	Visiting friends or relatives

APPENDIX B

WHIPLASH ACTIVITEITEN EN PARTICIPATIE LIJST (WAL)

M.S. Stenneberg, M.A. Schmitt, 2014

In deze lijst worden 35 activiteiten beschreven.

Wilt u aangeven in welke mate u in het dagelijks leven wordt belemmerd bij het uitvoeren van deze activiteiten *vanwege* uw neklachten.

Het gaat over de *afgelopen* week.

Voor elke activiteit is er een schaal van 0 tot 4. Wilt u bij iedere activiteit één antwoord kiezen en het daarbij behorende cijfer omcirkelen.

Kies het antwoord dat het beste bij u past.

Als u een activiteit de afgelopen week niet heeft uitgevoerd, dan kruist u bij deze activiteit de eerste optie aan: score 0.

Voorbeeld:

In welke mate had u de afgelopen week problemen met:

Lezen	0 Geen probleem	1 Lichte problemen	2 Matige problemen	3 Ernstige problemen	4 Onmogelijk om te doen of uit te voeren
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In welke mate had u de afgelopen week problemen met:

		0 Geen probleem	1 Lichte problemen	2 Matige problemen	3 Ernstige problemen	4 Onmogelijk om te doen of uit te voeren
1	Concentreren	0	1	2	3	4
2	Lezen	0	1	2	3	4
3	Oplossen van moeilijke problemen	0	1	2	3	4
4	Uitvoeren van verschillende taken tegelijkertijd	0	1	2	3	4
5	Omgaan met stress	0	1	2	3	4
6	Het voeren van gesprekken	0	1	2	3	4
7	Werken met een pc of laptop	0	1	2	3	4
8	Bukken of vooroverbuigen	0	1	2	3	4
9	Lang in een zelfde houding zitten	0	1	2	3	4
10	Omkijken	0	1	2	3	4
11	Tillen en dragen van voorwerpen	0	1	2	3	4
12	Langdurig lopen of wandelen	0	1	2	3	4
13	Hardlopen	0	1	2	3	4
14	Boven het hoofd werken	0	1	2	3	4
15	Reizen met een bus, tram of trein	0	1	2	3	4
16	Fietsen	0	1	2	3	4
17	Autorijden of rijden op een motorfiets	0	1	2	3	4
18	Wassen en/ of douchen	0	1	2	3	4
19	Lichaamsverzorging, zoals opmaken, tanden poetsen, haren wassen	0	1	2	3	4
20	Uzelf aan kleden en / of uitkleden	0	1	2	3	4
21	Winkelen en/ of boodschappen doen	0	1	2	3	4
22	Eten klaarmaken (snijden, koken etc)	0	1	2	3	4
23	Huishouden doen (schoonmaken, de was doen, ramen zemen, etc)	0	1	2	3	4
24	Het gebruik van huishoudelijke apparaten (zoals stofzuigen)	0	1	2	3	4
25	Tuinieren	0	1	2	3	4
26	Het omgaan met anderen, zoals collega's, vrienden, partner, kinderen	0	1	2	3	4
27	Het onderhouden van relaties	0	1	2	3	4
28	Seksualiteit	0	1	2	3	4
29	Het volgen van een studie, opleiding of cursus	0	1	2	3	4
30	Werken (betaald werk of vrijwilligerswerk)	0	1	2	3	4
31	Het meedoen aan recreatieve activiteiten, zoals een dagje uit, bezoek aan een pretpark	0	1	2	3	4
32	Het deelnemen aan sociale activiteiten (zoals kerkbezoek, politieke bijeenkomsten, activiteiten binnen een club)	0	1	2	3	4
33	Het beoefenen van sport	0	1	2	3	4
34	Uitgaan (zoals het bezoek aan een theater, bioscoop of museum)	0	1	2	3	4
35	Op visite gaan	0	1	2	3	4

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Chapter 5

CONCURRENT VALIDITY AND INTERRATER RELIABILITY OF A NEW SMARTPHONE APPLICATION TO ASSESS 3D ACTIVE CERVICAL RANGE OF MOTION IN PATIENTS WITH NECK PAIN

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ABSTRACT

Background

There is a lack of valid, reliable, and feasible instruments for measuring planar active cervical range of motion (aCROM) and associated 3D coupling motions in patients with neck pain. Smartphones have advanced sensors and appear to be suitable for these measurements.

Objectives

To estimate the concurrent validity and interrater reliability of a new iPhone application for assessing planar aCROM and associated 3D coupling motions in patients with neck pain, using an electromagnetic tracking device as a reference test.

Design

Cross-sectional study

Methods

Two samples of neck pain patients were recruited; 30 patients for the validity study and 26 patients for the reliability study. Validity was estimated using intraclass correlation coefficients (ICCs), and by calculating 95% limits of agreement (LoA). To estimate interrater reliability, ICCs were calculated. Cervical 3D coupling motions were analyzed by calculating the cross-correlation coefficients and ratio between the main motions and coupled motions for both instruments.

Results

ICCs for concurrent validity and interrater reliability ranged from 0.90 to 0.99. The width of the 95% LoA ranged from about 5 degrees for right lateral bending to 11 degrees for total rotation. No significant differences were found between both devices for associated coupling motion analysis.

Conclusions

The iPhone application appears to be a useful discriminative tool for the measurement of planar aCROM and associated coupling motions in patients with neck pain. It fulfills the need for a valid, reliable, and feasible instrument in clinical practice and research. Therapists and researchers should consider measurement error when interpreting scores.

INTRODUCTION

Assessment of active cervical range of motion (aCROM) is an important part of the clinical examination of impairments in patients with neck pain (Childs et al., 2008; de Koning et al., 2008; Williams et al., 2010). Nearly all patients with neck pain show a decreased aCROM compared to healthy subjects (Stenneberg et al., 2016). Current physical therapy guidelines recommend to assess and restore cervical range of motion in order to reduce neck complaints (Bier et al., 2016; Childs et al., 2008).

It is essential for physical therapists to use valid measurement instruments with high levels of interrater reliability when obtaining quantitative data on the amount of aCROM in patients (Kottner et al., 2011; Streiner and Norman, 2008; de Vet et al., 2006). Instruments such as visual estimation, single inclinometers, and the cervical range of motion device (CROM-device), of which the latter two possess adequate measurement properties, are available to measure aCROM (Antonaci et al., 2000; Jordan, 2000; de Koning et al., 2008; Williams et al., 2010). All of these instruments, however, only provide measurements of primary planar movements, which may not be sufficient to register intricate cervical movements (Jordan, 2000).

Cervical kinematics are complex (Antonaci et al., 2000; Bogduk and Mercer, 2000; Cook et al., 2006; Feipel et al., 1999) and are likely to change as a consequence of neck pain (Guo et al., 2012; Woodhouse and Vasseljen, 2008). Not only is planar range of motion of the primary movements (flexion-extension, lateral bending and rotation) reduced in neck pain patients compared to healthy controls, but also 3D coupling between cervical rotation, lateral bending, and flexion-extension is affected as well (Guo et al., 2012; Woodhouse and Vasseljen, 2008). To evaluate these 3D coupling motions, instruments should be capable of measuring these adequately (Guo et al., 2012).

To measure both the planar movements of the cervical spine and the associated coupled motions, devices such as electromagnetic tracking systems and optical motion systems are available and accepted as a gold standard (Antonaci et al., 2000; Nafis et al., 2006; Williams et al., 2010). These systems, however, are expensive, difficult to use, and the interpretability of data is complex. Therefore, these are not ideally suited for use in clinical practice.

To date, there is a growing tendency to use smartphone applications for assessment of range of motion in different joints (Ferriero et al., 2011; Kolber et al., 2013; Ockendon and Gilbert, 2012; Shin et al., 2012). Current smartphones have high-quality built-in microelectromechanical systems (MEMS) such as gyroscopes and accelerometers. Using integrated gyroscopic data allows assessment of 3D neck orientation (Jasiewicz et al., 2007; Luinge, 1999; Theobald et al., 2012).

Smartphone applications for measuring planar aCROM are clinically applicable and have moderate to good measurement properties (Guidetti et al., 2016; Kolber et al., 2013; Quek et al., 2014; Tousignant-Laflamme et al., 2013). However, all these studies included healthy participants, which limits generalizability of their results. The study sample should reflect the population of interest because reliability is highly dependent on the distribution of aCROM in the population (de Vet et al., 2011).

Moreover, the reliability and validity of the reference tests, i.e., goniometers and inclinometers, which are used in most of these studies are questionable, therewith biasing claimed measurement properties of the applications (Guidetti et al., 2016; Kolber et al., 2013; Tousignant-Laflamme et al., 2013). Furthermore, to the best of our knowledge, none of the existing applications are able to conduct analysis of the 3D coupling motions of the cervical spine.

Due to lack of valid, reliable, and feasible instruments for measuring planar aCROM and associated coupling motions, we developed a new iPhone-application. The aim of this study was to estimate the concurrent validity and interrater reliability of this new iPhone-application to assess aCROM in patients with nonspecific neck pain presenting in primary care physical therapy, using an electromagnetic tracking device as a reference test.

MATERIALS AND METHODS

Design

A cross-sectional study in which concurrent validity and interrater reliability of the Apple iPhone 4s was estimated using two samples of neck pain patients, with one sample being used for the validity study and the other for the reliability study. An electromagnetic tracking device (Polhemus Liberty) was used as a reference test.

Participants

Patients from both samples were recruited from five primary care physical therapy practices in the Netherlands. Patients were considered eligible for inclusion when above 18 years, with neck pain existing for at least one week and defined as grade I or II by the Neck Pain Task Force (Guzman et al., 2009). Participants had to be able to read the Dutch language adequately in order to be able to complete the questionnaires. Patients with previous surgery of the cervical spine or suffering from neurological, visual or vestibular disorders, cervical radiculopathy, or dizziness were excluded.

All participants signed an informed consent. The Central Committee on Research Involving Human Subjects (CCMO, The Hague, The Netherlands) approved the study protocol (non-WMO statement).

Raters

Two manual therapists (HB, MS) with more than 10 years of clinical experience performed all measurements according to a rigidly standardized protocol. Both raters attended two four-hour training sessions to become familiar with use of the measurement devices and standardization of the examination procedures.

Self-Reported Measures

All patients completed a questionnaire to register demographics (age, sex, height and weight, and duration of symptoms) and scored their pain intensity on a 10-point Numeric Pain Rating Scale (NPRS) and completed the Neck Disability Index (NDI) (Vernon and Mior, 1991). The NPRS and NDI are widely used and reliable and valid measurement tools to assess pain and disability in individuals with neck pain (MacDermid et al., 2009; Pool et al., 2007; Schellingerhout et al., 2012).

Instruments

iPhone application

An Apple iPhone 4s device with the operation system IOS 7.0 (Apple Inc. Cupertino, California, United States) was used. To measure 3D aCROM, we developed an application called '3D range of motion'. This application uses the built-in gyroscope and accelerometer function of the smartphone as angular measurement tools. A CMMotionManager class developed by Apple Inc. converts raw and processed accelerometer, gyroscope, and magnetometer data into range of motion along three axes: x , y , and z axis. The iPhone application records data at a sampling rate of 50 Hz.

Polhemus Liberty

The Polhemus Liberty (Polhemus, Colchester, Vermont, United States), a three-dimensional electromagnetic tracking device, was used as a reference test. The Polhemus Liberty is a valid and reliable instrument for measuring spinal range of motion when compared to a Vicon motion capture system (Kaliarntas et al., 2009). The Polhemus Liberty detects angular motions with an accuracy of 0.3° (Nafis et al., 2006). Tracking data were obtained at a sampling rate of 240 Hz. The data of the Polhemus Liberty were processed using Mathcad software (version 14).

Measurement procedures

Preliminary validation

Prior to the main study, preliminary validation tests were carried out to investigate to which extent the iPhone displays the same angle as the Polhemus Liberty. For this purpose, a wooden phantom (frame) was used with a lever arm that could move left and right to 75° , with 15° stages (Figure 1). The transmitter of the Polhemus Liberty was placed horizontally, squared, and centered. The measurement devices were placed on the phantom, with 25 centimeters interspace to avoid interference between the two devices. Before each test, both measurement devices were calibrated in the zero-position. One of the researchers (MS) performed the movements with the arm of the phantom, while simultaneously capturing data from the two devices.

The arm of the phantom was moved into respectively 15° , 30° , 45° , 60° , and 75° and then back to the starting point, in five different measurement cycles. For each angle, three trials were carried out in succession. Movements to the left and right were recorded separately. This test procedure was repeated for each of the three

axes (x, y, and z axis), by changing the orientation of the iPhone in the concerning position.

Mean differences between the Polhemus Liberty and the iPhone were found to be very small, ranging from 0.02° (95%CI -0.15° – 0.19°) for flexion-extension trial 2, to 0.75° (95%CI 0.56° – 0.94°) for rotation trial 3 (Table 1). Because these differences were substantially smaller than those of the instruments currently used in practice (Fletcher and Bandy, 2008; Law and Chiu, 2013), we considered our results sufficient to proceed with the main study.

Figure 1. Preliminary validation study setup and specific positions of the equipment

Table 1. Preliminary validation on wooden phantom (frame): differences between Polhemus Liberty and iPhone

	Difference Polhemus - iPhone Flexion-extension (x axis)		Difference Polhemus - iPhone Rotation (y axis)		Difference Polhemus - iPhone Lateral bending (z axis)	
	MD (95%CI)	LoA	MD (95%CI)	LoA	MD (95%CI)	LoA
Trial 1+2+3 (deg)	0.06 (-0.36 – 0.16)	-0.46 – 0.58	0.48 (0.37 – 0.59)	-0.12 – 1.08	0.10 (-0.01 – 0.21)	-0.49 – 0.69
Trial 1 (deg)	-0.05 (-0.15 – 0.04)	-0.31 – 0.20	0.23 (0.13 – 0.34)	-0.06 – 0.52	0.03 (-0.13 – 0.20)	-0.42 – 0.49
Trial 2 (deg)	0.02 (-0.15 – 0.19)	-0.46 – 0.49	0.46 (0.30 – 0.62)	0.01 – 0.91	0.09 (-0.12 – 0.30)	-0.50 – 0.67
Trial 3 (deg)	0.22 (-0.00 – 0.45)	-0.41 – 0.85	0.75 (0.56 – 0.94)	0.23 – 1.27	0.17 (-0.10 – 0.43)	-0.56 – 0.90

deg = degrees; MD = mean difference; 95%CI = 95% confidence interval; LoA = limits of agreement

Measurement procedures main study

ACROM was assessed with participants sitting on a plastic chair with a backrest and a seat height of 50 centimeters. Their feet were resting on the ground, knees and hips flexed 90°, hands on thighs, and with neutral head-neck position. The iPhone was fastened securely on the forehead by means of an iPhone holder with a rigid Velcro strap (Figure 2).

Prior to the assessment, there was an introduction to the procedure and participants performed all movements to become familiar with the measurements. Participants were asked to move their head three times, fluently, and as far as possible without provoking pain, and avoiding any movements of the shoulder girdle and thorax. The raters supervised to ensure that the movements were executed properly and followed the relevant axis.

Participants performed a full cycle flexion-extension, left and right rotation, and lateral bending, each three times and with a 10-second interval between each cycle. To eliminate a potential influence of a fixed movement sequence on the aCROM measurements, the movement sequence (flexion-extension, lateral bending, rotation) was randomized per participant.

Data were recorded directly on the iPhone and sent to the outcome assessor after the measurements were performed. Raters and patients were blinded to their own and each others' results.

Figure 2. Positioning of the iPhone and sensor of the Polhemus Liberty.

Concurrent validity

To avoid interference between the iPhone and Polhemus Liberty, the sensor of the Polhemus Liberty was mounted on a Plexiglas holder which was attached to the strap that was holding the iPhone in place against the forehead (Figure 2). The transmitter of the Polhemus Liberty was placed horizontally, squared, and centered in front of the subject, leveled with the sternum.

Before each measurement, the two instruments were calibrated simultaneously by the two raters. After three seconds, the Polhemus Liberty and the iPhone simultaneously registered the aCROM.

Interrater reliability

To assess interrater reliability, both raters performed the test sequence at an interval of 15 minutes, under the same conditions and in the same room. Based on clinical experience, we assumed this time frame to be long enough to allow for adequate recovery following cervical range of motion testing, and short enough to prevent changes in mobility and major variation due to rater or patient-related factors.

Sample size

To detect a minimal intraclass correlation coefficient (ICC) of 0.75 ($\alpha=0.05$, $1-\beta=0.80$), with an expected ICC of 0.9, and two measurements per participant with a 10% drop-out rate, a total of 26 participants was required for the reliability study (Walter et al., 1998). For the validity study, an equal sample was considered adequate, based on previous studies evaluating validity of cervical range of motion measures (Assink et al., 2008; Guidetti et al., 2016; Quek et al., 2014; Tousignant-Laflamme et al., 2013).

Statistical analysis

Statistical analyses were performed using SPSS version 24.0 (SPSS Inc., Chicago, Illinois, USA). For both the validity study and the reliability study, the mean scores of the three repetitions were calculated and used for data analysis.

Concurrent validity

Validity was estimated using the intraclass correlation coefficient, two-way mixed effects model (ICC 3,1) to measure the association between scores from the two

devices, and by calculating the limits of agreement ($LoA = 1.96 \times SD$) (Bland and Altman, 1986; de Vet et al., 2011). ICCs of 0.75 and higher were considered to be acceptable for a useful measurement instrument (de Vet et al., 2011).

Coupled motion was defined as an association of one cervical motion (rotation or lateral bending) about an axis with another motion about a second axis. 3D coupling motion analysis consisted of two analysis methods (Cattrysse et al., 2012, 2008, 2006). First, to characterize the relationship between the main motion (e.g. rotation) and the coupled motion (e.g. lateral bending), cross-correlation coefficients between the main motion and the coupled motion were calculated for both instruments. A positive cross-correlation expresses an ipsilateral coupling pattern, a negative correlation a contralateral one (Cattrysse et al., 2012, 2008, 2006). Second, the ratio between the main and coupled motion was compared between both devices. The ratio expresses the amount of coupled motion relative to the main motion component (Cattrysse et al., 2012, 2008, 2006). The ratio was calculated as the standard deviation of the main motion divided by the standard deviation of the coupled motion ($ratio = SD \text{ main motion} / SD \text{ coupled motion}$) (Cattrysse et al., 2012, 2008, 2006). Data on cross correlation and ratio were visually checked for normality with histograms, Q-Q plots and boxplots, and tested with the Shapiro-Wilk test.

Differences in cross-correlation and ratio between the Polhemus Liberty and the iPhone were analyzed using the Wilcoxon signed-ranks test in case of not normally distributed data. The level of significance for all tests was set as $\alpha=0.05$.

Interrater reliability

In order to estimate interrater reliability, intraclass correlation coefficients, for two-way random effects models (ICC 2,1), were calculated. Since in clinical practice different physical therapists perform the measurements, systematic differences between raters were considered to be part of the measurement error. ICC values of at least 0.70 were considered to be a minimal requirement for interrater reliability (de Vet et al., 2011).

Further, limits of agreement (LoA) were calculated to determine the absolute measurement error between the two raters (Bland and Altman, 1986; de Vet et al., 2011).

RESULTS

Patient characteristics

Thirty patients (63.3% female) with a mean age of 53.4 years (SD 12.2) participated in the validity study, and the reliability sample consisted of 26 patients (73.1% female) with a mean age of 45.2 years (SD 15.3) (Table 2). Some observations were not available for all subjects because of technical iPhone and/or Polhemus Liberty errors (Tables 3-5).

Table 2. Characteristics of the patients

Characteristics	Validity study (n=30)	Reliability study (n=26)
Age (years) (mean, SD)	53.4 ± 12.2	45.2 ± 15.3
Female	19 (63.3%)	19 (73.1%)
BMI (mean, SD)	25.6 ± 3.6	24.7 ± 3.7
Duration of symptoms		
0-3 months	6 (20.0%)	7 (26.9%)
3-6 months	6 (20.0%)	3 (11.5%)
6-12 months	3 (10.0%)	4 (15.4%)
>1 year	15 (50.0%)	12 (46.2%)
NRS (median, range)		
Average	5.5 (0-8)	5.0 (2-8)
Minimal	2.5 (0-6)	3.0 (0-7)
Maximal	8.0 (1-10)	8.0 (3-9)
NDI score (median, range)	9.5 (1-26)	11.4 (3-27)

SD = standard deviation; BMI = Body Mass Index; NRS = Numeric Rating Scale (ranging from 0 to 10 points. Maximum pain = 10 points); NDI = Neck Disability Index (10 items ranging from 0 to 5 points. Maximal disability = 50 points)

Concurrent validity

Planar movements

Mean differences of half cycle movements between the iPhone and Polhemus Liberty ranged from 0.7° (SD 1.4°) for left lateral bending to 4.3° (SD 1.9°) for right rotation. For full cycle movements, mean differences ranged from 1.5° (SD 2.4°) for lateral bending to 8.4° (SD 2.9°) for rotation (Table 3).

All ICC values exceeded 0.90, indicating excellent validity. The limits of agreement ranged from -1.85° to 3.43° for right lateral bending to 2.70° to 14.14° for total rotation (Table 3), indicating that the difference between the measurements are expected to vary within this range.

Table 3. Validity of the iPhone compared to the Polhemus Liberty (planar movements and mean scores on both devices, expressed in degrees)

	iPhone Mean (SD) (deg)	Polhemus Mean (SD) (deg)	MD (SD) (deg)	ICC (95%CI)	LoA (deg)
<i>Total Flexion-extension (n=27)</i>	100.7 (15.1)	96.5 (14.0)	4.1 (2.4)	0.95 (0.15 - 0.99)	-0.62 – 8.82
Flexion (n=27)	49.5 (8.5)	46.2 (7.9)	3.3 (1.5)	0.91 (-0.01 - 0.98)	-0.21 – 6.35
Extension (n=27)	51.1 (12.4)	50.3 (11.8)	0.8 (1.7)	0.99 (0.97 - 0.99)	-2.56 – 4.15
<i>Total Rotation (n=29)</i>	128.9 (22.4)	120.5 (20.6)	8.4 (2.9)	0.92 (-0.01 - 0.98)	2.70 – 14.14
Right (n=29)	63.7 (10.8)	59.4 (9.8)	4.3 (1.9)	0.91 (-0.01 - 0.98)	0.58 – 8.08
Left (n=29)	65.2 (14.1)	61.1 (13.1)	4.1 (1.8)	0.95 (0.04 - 0.99)	0.60 – 7.61
<i>Total lateral bending (n=30)</i>	61.4 (18.2)	59.9 (16.7)	1.5 (2.4)	0.99 (0.96 - 0.99)	-3.24 – 6.32
Right (n=30)	29.6 (8.8)	28.9 (8.2)	0.8 (1.3)	0.98 (0.95 - 0.99)	-1.85 – 3.43
Left (n=30)	31.7 (10.1)	31.0 (9.4)	0.7 (1.4)	0.99 (0.97 - 0.99)	-2.11 – 3.61

SD = standard deviation; MD = Mean difference; deg = degrees; ICC = intraclass correlation coefficient; 95%CI = 95% confidence interval; LoA = Limits of agreement

3D coupling motions

For both rotation and lateral bending as main motions, there were no significant differences in cross-correlation coefficients between the iPhone and the Polhemus Liberty (Table 4). In addition, for both rotation as well as for lateral bending as the primary movement no significant differences were found for the ratio analyses between both devices (Table 4).

Table 4. Three-dimensional range of motion: cross-correlation and ratio

	<i>Polhemus</i> Mean (SD)	<i>iPhone</i> Mean (SD)	<i>p</i> value
<i>Rotation (n = 29)</i>			
Cross correlation	0.55 (0.44)	0.57 (0.46)	0.230
Ratio	12.29 (5.05)	11.38 (7.11)	0.214
<i>Lateral bending (n = 30)</i>			
Cross correlation	-0.91 (0.19)	-0.91 (0.14)	0.271
Ratio	2.03 (1.19)	2.10 (1.44)	0.629

SD = standard deviation

Interrater reliability

ICCs for interrater reliability were 0.90 (95%CI 0.78 - 0.95) for flexion-extension, 0.92 (95%CI 0.82 - 0.97) for rotation, and 0.96 (95%CI 0.90 - 0.98) for lateral bending (Table 5). The limits of agreement, indicating the measurement error between the two independent raters, were -11.97° to 15.19° for flexion-extension, -10.06° to 13.82° for rotation, and -10.95° to 9.93° for lateral bending (Table 5).

Table 5. Interrater reliability and mean scores of rater 1 and 2

	Rater 1 Mean (SD) (deg)	Rater 2 Mean (SD) (deg)	ICC (95%CI)	LoA (deg)
Flexion-extension (<i>n</i> =24)	101.88 (14.47)	101.49 (16.19)	0.90 (0.78 - 0.95)	-11.97 - 15.19
Rotation (<i>n</i> =21)	117.09 (21.87)	115.08 (18.74)	0.96 (0.90 - 0.98)	-10.06 - 13.82
Lateral bending (<i>n</i> =23)	54.73 (12.03)	54.34 (13.83)	0.92 (0.82 - 0.97)	-10.95 - 9.93

SD = standard deviation; deg = degrees; 95%CI = 95% confidence interval; LoA = Limits of agreement

DISCUSSION

This study demonstrates high concurrent validity and interrater reliability of a new iPhone application to measure planar aCROM and associated coupling motions in patients with neck pain. Based on the observed ICC values (≥ 0.90) for concurrent validity and interrater reliability, the iPhone application appears to be a suitable instrument for discriminative purposes in the clinical decision making process in neck pain patients. However, 95% LoA between the measurement devices were wide implying relatively large absolute measurement error.

There are no comparable smartphone studies assessing the aCROM in neck pain patients. Previous smartphone studies assessing aCROM in healthy subjects found moderate to good concurrent validity for flexion-extension and lateral bending, and poor to moderate validity for rotation (Quek et al., 2014; Tousignant-Laflamme et al., 2013). Another study comparing an iPhone 5c with an inclinometer in healthy subjects demonstrated excellent validity and reliability, with ICCs above 0.99 and 95% LoA of 1.5 to 3.0 degrees (Guidetti et al., 2016).

Results of previous studies that assessed concurrent validity and reliability of iPhone applications for the measurement range of motion of joints other than the cervical spine, are in line with our results. Good to excellent validity and reliability were reported for lumbar spine, shoulder, ankle, and knee range of motion measurements (Jones et al., 2014; Kolber et al., 2013; Mitchell et al., 2014; Pourahmadi et al., 2016; Romero Morales et al., 2017), which strengthens legitimacy of the use of iPhone applications for range of motion measurement in physical therapy.

There have been no studies with smartphones being used for the examination of coupled motions. Analysis of the results of our preliminary validity study show only small, negligible differences (ranging from 0.02° to 0.75°) around all three axes between the iPhone and the Polhemus Liberty, which supports the conclusion that the iPhone adequately registers data on all axes.

We found substantial differences between the results of the measurements performed on patients compared to those from our preliminary validation on the wooden frame. A possible reason for this is that there may have been a magnetic field interference between the MEMS-sensors and the Polhemus Liberty during the measurements on patients. Sensors of the devices were placed further apart

on the wooden frame than they were on patients. MEMS sensors are very sensitive to electromagnetic fields (Roetenberg et al., 2005). Influence of electromagnetic fields on the results of smartphone measurements have been reported before (Quek et al., 2014). A study in which there was no such influence showed noticeably higher validity and smaller measurement errors (Guidetti et al., 2016). Another reason for the discrepancies might be that, despite our fixation, movement of the sensor of the Polhemus Liberty relative to the iPhone and head was still possible, which could also have negatively influenced intertrial variability. As a consequence, the results of the present study could have underestimated validity and reliability of the iPhone application.

It is important for clinicians to appreciate the width of the 95% LoA between the measurement devices. The estimated 95% LoA indicate that differences up to 2.6 degrees for lateral flexion and up to 5.7 degrees for total rotation may exist when using these devices interchangeably. Absolute measurement errors indicate that change scores should exceed 10.4 degrees for total lateral bending to 13.6 degrees for total flexion-extension to detect changes that are not due to intertrial variability or to measurement error. This magnitude of measurement error is reported at least as large in other studies that assessed validity and reliability of aCROM measures (Assink et al., 2005; Fletcher and Bandy, 2008; Jordan, 2000; Quek et al., 2014). In order to reduce the measurement error in practice, it is recommended to make sure that raters are well trained, measurements are taken under controlled conditions and the average of multiple measurements is used to determine aCROM (Streiner and Norman, 2008).

It is important that the measurement error found between raters is smaller than the minimal clinically important difference in order to discriminate in scores between neck pain patients and those without neck pain. A recent study showed that at group level differences between patients with neck pain and healthy persons are larger, with a minimum of 7.0 degrees for half cycle movements (lateral bending) and 16.7 degrees for full cycle movements (total lateral bending) (Stenneberg et al., 2016). Therefore, the clinically important difference seems to be beyond the measurement error found in the present study. We suggest further investigation of the clinically important difference in neck pain patients compared to individuals without neck pain to consider the clinical relevance of aCROM measurements.

Limitations

High ICCs could be explained by high variability in aCROM between patients (de Vet et al., 2006). However, the variability in aCROM among patients of our sample was comparable to that of other clinically representative samples (Stenneberg et al., 2016). We therefore assume our ICC values to be a valid estimate of reliability.

Further, intrarater reliability was not assessed in this study. In clinical settings, therapists evaluate aCROM over time which is why an estimation of the intrarater reliability could be required as well. However, examining intrarater reliability seems to be of less importance when an instrument shows good interrater reliability, (Streiner and Norman, 2008) as intrarater reliability usually tends to be higher than interrater reliability (Streiner and Norman, 2008).

Last, the technology used in smartphones innovates rapidly. New smartphones contain more advanced sensors and software. It is expected that newer and better technology will lead to more accurate measurements. The clinimetric properties of these new smartphones will need continuous validity and reliability evaluation.

CONCLUSIONS

Our results suggest that the new iPhone application is a useful diagnostic tool for the measurement of planar aCROM and associated coupling motions in patients with neck pain. As such it fulfills the need for a valid, reliable, and feasible instrument for measuring aCROM in clinical practice and research. Therapists and researchers should, however, consider measurement error when interpreting scores.

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Chapter 6

ACTIVE CERVICAL RANGE OF MOTION VARIES SUBSTANTIALLY WITHIN AND BETWEEN DAYS IN HEALTHY PARTICIPANTS

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Chapter 7

CLINICAL CHARACTERISTICS DIFFER BETWEEN PATIENTS WITH NON-TRAUMATIC NECK PAIN, PATIENTS WITH WHIPLASH-ASSOCIATED DISORDERS, AND PAIN-FREE INDIVIDUALS

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Chapter 8

DISABILITY IS DETERMINED BY MORE COMPLEX INTERACTIONS BETWEEN PAIN, AND PHYSICAL AND PSYCHOLOGICAL IMPAIRMENTS IN PEOPLE WITH TRAUMATIC VERSUS NON-TRAUMATIC NECK PAIN

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Chapter 9

GENERAL DISCUSSION

The general aims of this thesis were to describe the clinical impact of two types of neck pain presenting in primary physiotherapy care, namely nontraumatic neck pain and traumatic neck pain, and to develop and evaluate clinically applicable measurement instruments for assessing impairments and limitations in people with nontraumatic neck pain and traumatic neck pain. The main findings, weaknesses, and implications for daily physiotherapy care and future research are discussed in this chapter.

Cervical mobility

Our research shows that reduced active cervical range of motion (aCROM) is one of the main physical impairments associated with neck pain (Chapters 2 and 7) and aCROM is associated with activity limitations and disability in both nontraumatic neck pain and traumatic neck pain (Chapter 8) [1]. The results of our systematic review show that people with neck pain have a significant and clinically relevant decrease in aCROM compared to people without neck pain. Patients with traumatic neck pain are more restricted in their cervical movements than patients with nontraumatic neck pain. This is in line with previous research showing that patients with neck pain move their neck in a more rigid way than healthy individuals who move in a more variable way [2]. Flexion-extension, total rotation, and extension are the most discriminating movements between people with neck pain and those without neck pain (Chapter 2).

In clinical practice, aCROM is the most routinely collected function by physiotherapists and other health care providers as an integral part of the physical examination process [3]. ACROM is assessed to determine diagnosis and prognosis, guide treatment, and evaluate treatment effects. ACROM measurements are also used in research as a diagnostic tool to describe physical functioning and to assess the usefulness of various treatment interventions. There is evidence for the diagnostic and predictive value of aCROM [3]. Mobilization techniques have a positive effect on aCROM, although the evidence is limited [3–5].

While the importance of aCROM has been acknowledged by our studies, there is debate as to whether aCROM should be used as a diagnostic and evaluative parameter (or both). The clinical utility of aCROM measurements is complex for several reasons, which are discussed below.

First, the size of measurement error of commonly used instruments is relatively large in relation to clinically relevant differences (Chapters 2, 5 and 7). Obtained changes in aCROM, for example, due to natural course or treatment, may be important to the patients, but are mostly small and within the measurement error of the instruments used. As a result, the extent to which aCROM is limited or changes over time is difficult to objectify with instruments currently used in physiotherapy practice. With that being said, aCROM still is an important factor, even if it cannot be measured precisely enough, and it should not deter us from further investigation into the cause and role of reduced aCROM in individuals.

Second, there are significant differences in normal range of motion between healthy individuals. These differences are even larger in those with neck pain (Chapter 2). Because of this variation in aCROM between individuals, and an overlap in aCROM between people with and without impaired cervical mobility, there is no clear cut-off point to consider aCROM as limited or not. The interpretation of the obtained aCROM scores remains subjective to the assessor. Differences *between* individuals are thus not easily interpreted and limit diagnostic value of aCROM. Differences *within* individuals over time, can be detected more precisely, as intrarater reliability usually tends to be higher than interrater reliability [6]. However, we found in chapter 6 that the natural variation of aCROM is considerable and this should be taken into account when interpreting aCROM scores. Since clinicians usually evaluate aCROM over a longer period of time, e.g., when evaluating the course of neck pain or effectiveness of treatment, we recommend that studies on intrarater reliability should select an appropriate (i.e. longer) time period to be reflective for use in daily practice. Moreover, physiotherapists should not only take measurement error but also the variability of aCROM measurements into account when interpreting the results. Based on the previous considerations, we recommend not to indicate aCROM measurements as 'limited' or 'not-limited', but rather in degrees.

Another reason to discuss the clinical utility of aCROM measurements, is the complexity of cervical kinematics. Next to planar aCROM of the primary movements (flexion-extension, rotation and lateral bending) being reduced, 3-dimensional coupling between movement directions also tends to be affected in neck pain patients [7-9]. Commonly used instruments in research and clinical practice are not capable of measuring multi-planar movements adequately and therefore, we also assessed aCROM only in one planar movement. Measuring only

single plane movements, however, is simplifying human cervical kinematics and may not be sufficient to analyse intricate cervical movements as they are affected by movement in other planes. Assessing coupled and 3-dimensional kinematics can probably provide more insights into the mechanisms behind the movement disorders and recognizing kinematic differences between patients is important for clinicians to identify possible movement risk factors that may be targeted with treatment [7]. We therefore recommend that future research on differences between subgroups and individual patients should include all 3 axes of movement and coupled movements. To support this in future research, we developed a measuring instrument that is valid, applicable, and can also measure around 3 axes simultaneously (Chapter 5).

Furthermore, the upper and lower parts of the cervical spine differ substantially from a kinematic perspective [10,11]. In addition, specific diagnoses, such as cervicogenic headache, are associated with dysfunctions in the upper cervical segments, and other symptoms may have their source in the lower cervical segments (Jull et al., 2007). Thus, objective measures of cervical range of motion that can separate movement of the upper and lower cervical spine can be valuable for characterization of patients, diagnostics and treatment evaluation [1].

When objective measurements appear to have limitations, subjective measurements might have some additional value. Self-assessment tools to evaluate elbow, shoulder and knee range of motion, and mouth opening have been shown to be useful measurements for range of motion [12–15], and recently, two studies found that subjective assessment of aCROM is promising, showing moderate to high levels of agreement between patient-reported measurements and the physiotherapists' measurements [16,17]. Considering that the patient's perception of mobility may offer an important insight into the extent to which mobility restrictions are a problem for an individual, and including patient's perception in goal setting, may improve the diagnostic process and treatment success. We recommend that future studies investigate whether combining objective and subjective aCROM measurements leads to a more comprehensive assessment of patients with neck pain.

It is important to emphasize that neck pain and reduced cervical mobility are symptoms and not diseases. They can be manifestations of many different underlying impairments, disorders and pathologies. When patients visit

physiotherapy clinics to seek treatment for their neck pain, an important element of the diagnostic process of the physiotherapist is focused on identifying impairments or pathologies that are related to the symptoms presented. This would involve pathologies that may cause the symptom of neck pain, either through referred pain or direct involvement of the cervical spine. For example, we cannot identify patients who have neck pain due to hypofunction of the facet joints, meniscoid entrapment, fattening of the musculature, disc degeneration, etc. We therefore assume that a symptom-based approach to diagnosis and treatment is the better than a patho-anatomical-based approach for physiotherapists. Although restricted aCROM is an important factor that contributes to disability and may thus be an important treatment goal, we recommend not to use aCROM as a single test or individual prognostic factor, but to regard it as a part of a multivariable, hypothesis-based decision-making process.

As stated earlier in this thesis, neck pain is a biopsychosocial condition (Chapters 1, 7 and 8). Most studies examining explanatory factors for the degree of disability or investigating prognostic factors for persistent pain, mainly include psychosocial factors, physical activities, general health and symptoms like pain intensity in their studies and models [18,19]. In such studies, most variables are collected using questionnaires. Physical impairments such as neck mobility or muscle function are often omitted and there are only very few low quality studies that investigated impairments as a predictor of long term symptoms [18,19]. As a consequence, a vicious circle forms and the physical disorders are less often included in future models. While the importance of psychological factors is indisputable and supported by evidence [20,21], it does not exclude the possibility that physical impairments play an important role in the cause and course of neck pain. We have to prevent that the management advocated for patients with neck pain focuses only on psychosocial aspects with little or no regard to biological features [22], which is not in line with the approach of clinicians in daily practice and the results of the studies presented in this thesis which actually show that aCROM is an important factor.

Measuring functional limitations

In this thesis, we used the Neck Disability Index (NDI) as a measure of disability in several studies. The NDI is the most frequently used questionnaire in clinical

practice to measure disability and has shown to be a reliable and responsive measure of change for nontraumatic neck pain and traumatic neck pain [23–26]. However, in chapters 3 and 7 we addressed limitations in the applicability for patients with nontraumatic neck pain and traumatic neck pain, because the problems commonly identified in these patients, are frequently omitted. We found that the NDI does not fully reflect the spectrum of disabilities judged to be important by patients. Moreover, in chapter 7 we show that patients with nontraumatic neck pain and traumatic neck pain experience different types of activity limitations and participation restrictions. It is, therefore, not suitable for the specific limitations of individual patients and for identifying differences between subgroups of patients with neck pain presenting in physiotherapy clinics. These findings are in line with other studies [27,28]. We recommend including assessments of patient-specific activity restrictions in future research and clinical practice.

Traumatic neck pain versus nontraumatic neck

People with traumatic neck pain and nontraumatic neck pain differ in several aspects. In Chapter 7, we found that differences in physical impairments, such as aCROM and muscle strength, differ more than psychological factors between people with nontraumatic neck pain and traumatic neck pain. People with traumatic neck pain score significantly higher on activity limitations, participation restrictions, and disability, and both groups experience different types of activity limitations and participation restrictions. Further, in traumatic neck pain more clinical characteristics interact in their contribution to disability (Chapter 8). We therefore concluded that traumatic neck pain is a more severe and more complex condition than nontraumatic neck pain and should be considered a separate subgroup of neck pain (Chapter 7 and 8). However, in our studies we included a pragmatic selection of physical functions and psychological factors which are routinely collected in clinical practice and recommended in clinical guidelines. This could limit our conclusion. Nevertheless, our findings might be an underestimation of the differences between nontraumatic neck pain and traumatic neck pain, as other studies showed even more factors that differentiated between nontraumatic neck pain and traumatic neck pain, such as muscle degeneration, physiological distress, joint position error, and cognitive performance such as memory and concentration symptoms [29–33]. These factors were not measured in the thesis. This strengthens our conclusion that

nontraumatic neck pain and traumatic neck pain are separate subgroups, and as a consequence, we believe a different approach is required in both clinical practice and research.

Individualized physiotherapy

Prognosis of acute neck pain is generally good, although a proportion of all patients will develop chronic neck pain [34]. In the management of neck pain, no particular diagnostic strategy, intervention or outcome measure will be suitable for an entire population in preventing the transition from acute to chronic pain. It is important to realize that 20 to 30% of neck pain patients recover within 6 to 12 weeks, and about 15 to 20% inevitably shows a prolonged recovery, regardless of any physical therapy intervention [35,36]. Yet, we assume that the large middle proportion of 50-65% will benefit from individualized physiotherapy. There are various clinical characteristics to explain why some individuals, but not others, develop persistent problems [18,34,37]. In research, patients are usually assigned to different subgroups or interventions, not taking into account the individual clinical characteristics or preferences. These methodological principles limit the accuracy of the outcomes of research [38]. It is, therefore, recommended in research to provide personalized treatment, targeted to the individual problems identified in the diagnostic process. In clinical practice, it is possible to apply a hypothesis-based, individual clinical reasoning and decision-making process, including outcomes of history taking and physical examination, preferences, expectations, cultural biases, beliefs, and experiences of both physiotherapist and patient. When assessing patient specific characteristics, we advocate the use of a classification system, such as the International Classification of Functioning, Disability and Health (ICF), to make a clear distinction between constructs of health. When clinicians use questionnaires, it is important that these only assess the constructs of interest and avoid cross contamination between constructs.

Complexity of neck pain

In Chapter 8, we demonstrated that the degree of complexity differs between nontraumatic and traumatic neck pain, and that different factors interact and affect disability differently. The degree of complexity of neck pain lies in the unpredictable and nonlinear relationship between different variable components that interact and in how they affect disability [39,40]. In complex conditions and unpredictable professional contexts, existing scientific evidence may not apply. To deal with this clinical uncertainty, education on Master level is required. Master level educated physiotherapists are able to apply an expert level of evidence-based clinical reasoning and decision making. Highly educated therapists are able to recognize the multifactorial and multidimensional nature of neck pain, as well as the complex interactions between factors that contribute to the functional consequences of neck pain [41]. The frame of reference must be the unique individual patient. Unique neck pain related characteristics include not only the severity and duration of symptoms, the level of disability, or separate impairments as pain intensity, cervical range of motion, intersegmental mobility or cervical muscle strength, but also the psyche, social environment and capabilities of the individual patient. Master level physiotherapists can incorporate these factors in their decision-making and work together with the patients to reveal the causes of neck pain and find tailored interventions. This will lead to more personalized medicine.

Conclusion

In conclusion, nontraumatic neck pain and traumatic neck pain are both complex health conditions that differ in several dimensions. We also found that a variability in clinical characteristics and symptoms exists between individuals with and without neck pain, and that these clinical features vary over time. The degree of pain-related disability is associated with both physical and psychosocial factors, illustrating the multi-dimensional nature of neck pain. Reduced aCROM is an important feature of neck pain and plays an important role in the functioning of people with neck pain.

We recommend to consider nontraumatic neck pain and traumatic neck pain as separate subgroups of neck pain, in both clinical settings and research. We hope that our findings and recommendations will contribute to a better understanding of the complexity of neck pain, and that the development and evaluation of condition-specific measurement instruments will allow clinicians to better

evaluate patient specific diagnosis, and to perform tailored treatment in individuals with neck pain. Our methodological recommendations may guide researchers in the development and conduct of future research that considers individual patient characteristics and subgroup-specific differences.

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Chapter 10

IMPACT PARAGRAPH

This chapter provides a reflection on the relevance and innovativeness of the findings described in this thesis. It presents the scientific and social impact of the results from the perspective of patients, health care professionals and researchers.

Relevance

Neck pain is one of the most prevalent and burdensome health problems, with substantial impact on the patient's own and public health [1,2]. Neck pain is also regarded as one of the most disabling health conditions worldwide [1,3]. The economic burden related to neck pain is increasing and costs are rising, mainly due to work absenteeism and usage of health care services. People with neck pain form a highly diverse population presenting differences in patho-anatomic changes, biomechanical behaviour of the spine, physical impairments, activity limitations, personal factors, and societal factors. Despite its high prevalence in and high impact on society, knowledge gaps on the topic of neck pain remain.

The studies conducted in this thesis address a number of important research priorities. First, it is considered important to identify clinical features and clinically important subgroups within the heterogeneous population of people with neck pain in order to contribute to a specific diagnosis, and to identify which treatment leads to a better outcome for specific individuals with neck pain [4]. A special focus lies on the subgroup of traumatic neck pain.

Further, the studies conducted in this thesis answer the need for the development and evaluation of diagnostic instruments for differential diagnosis of neck pain. This includes investigating the measurement properties of these instruments [4]. Finally, the studies carried out in this thesis meet the recommendations to ensure that the methods used, and the research data generated, are clinically applicable and easy to use to ensure that research evidence is used to guide neck pain management [4].

Innovativeness

The innovative elements of this thesis involve: 1) the development and validation of new, clinically applicable measurement instruments; 2) the implementation of a new method to evaluate the clinimetric properties of measurement instruments to reliably quantify cervical range of motion, and 3) a new approach to gain a better understanding of the differences in complexity between nontraumatic neck pain and traumatic neck pain.

Despite aCROM being the most routinely collected measure of function by physiotherapists, valid and applicable instruments to assess 3-dimensional aCROM are lacking. To that purpose, a valid and new, condition specific and clinically usable iPhone application that measures planar aCROM and associated 3-dimensional coupling motions, was developed.

Another innovation is the development of a new condition-specific and clinically relevant questionnaire, tailored specifically to people with traumatic neck pain, which measures activity limitations and participation restrictions, as defined by the ICF framework [5]. Using this questionnaire allows clinicians to address the specific limitations of people with traumatic neck pain, while not simultaneously measuring other theoretical constructs of health.

Assessment of neck mobility is used for diagnostic purposes and for evaluating effects of therapy, both in clinical settings and in scientific research. ACROM values normally vary between measurement sessions due to biological variation, even if the patient's neck pain related symptoms remain stable over time. Traditionally, research on measurement error of instruments measuring cervical mobility is carried out with two testers and two measuring moments in short succession. In clinical practice, however, measurements are often carried out with more days in between assessments, which is why we introduced a study method performing measurements 3 times a day, on 7 days, spread over a period of 3 weeks, to explore intraday and interday variability of aCROM in order to better interpret aCROM measurements in clinical practice and research.

People with traumatic neck pain demonstrate higher levels of disability compared to people with nontraumatic neck pain and different mechanisms seem to be at the root of nontraumatic neck pain and traumatic neck pain. Both groups are assumed to represent complex health conditions. The degree of complexity lies in the unpredictable and nonlinear relationship between clinical features, in how they interact, and in how they affect disability. In order to explore differences between nontraumatic neck pain and traumatic neck pain, however, previous studies mostly limited themselves to reporting differences in clinical characteristics between the conditions. This, however, appears to be insufficient in terms of accounting for account for the complex, multifactorial nature of the health problems. To the best of our knowledge, we were the first to explore interactions between potential factors explaining disability in people with nontraumatic neck pain as compared to those with traumatic neck pain.

Scientific impact

The findings presented in this thesis contribute to the body of scientific knowledge about the characteristics of people with neck pain and the different subgroups. Measurement tools and knowledge about differences between subgroups and distinctive clinical features of people with neck pain are used in the design of new research and in the interpretation of available evidence. Acquired knowledge is implemented in the educational program and research programs of SOMT University of Physiotherapy, and in addition, the newly developed questionnaire and the iPhone application have been used in research programs of different universities. The developed measurement instruments (questionnaire and ROM application) for example, have been used by researchers and students in two large scientific projects, in which the specific limitations of subgroups of people with neck pain were assessed.

Conducting the research for this thesis also contributed to the research collaboration between SOMT University of Physiotherapy and the Vrije Universiteit in Brussels, resulting in joint research and educational activities. Moreover, interprofessional collaboration was achieved in other research projects in which researchers from different departments and universities took part. Finally, studies in this thesis explore new methodological challenges to investigate the measurement properties of instruments, and to understand the complexity of neck pain. As such, they offer insights into the challenges of different methodologies used in our practice-based research.

Societal impact

The higher goal of this thesis was to improve care for people with neck pain in primary health care. With the results of this thesis, health care professionals can now use specific diagnostic and evaluative instruments that are clinically applicable and easy to interpret. The knowledge gained in this thesis is being used by physiotherapists and students at SOMT University of Physiotherapy for diagnostic, prognostic and therapeutic purposes. The results of our research provide insights into relevant clinical characteristics of people with neck pain that can be used to set a patient-specific diagnosis and can be used to direct treatment decisions. This may lead to better outcomes for specific individuals with neck pain and could reduce the burden of neck pain for the individual patient and society,

assuming that better treatment outcomes are likely when the treatment is tailored to individual-specific characteristics.

Activities and further dissemination

All research articles have been submitted or published in international peer-reviewed journals. Summaries of the findings have been presented on several conferences and symposia, where we shared our acquired knowledge with other researchers and professionals. Furthermore, research results are also shared with other universities, for instance through means of providing capita selecta, as was the case at the Vrije Universiteit in Brussels. To spread our knowledge in the most efficient way among clinicians in The Netherlands, we published summaries in Dutch in the 'Fysiopraxis', the journal of the Royal Dutch Society for Physical Therapy (KNGF), and have used social media to highlight and disseminate our results. Our work has already been cited by other authors for over 100 times.

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Addendum

SUMMARY IN ENGLISH

Neck pain

Most people will develop some degree of neck pain (NP) during their lifetime. Acute NP mostly resolves within days or weeks. A substantial proportion of people, however, report persistent problems, and recurrence rates are high. NP may cause considerable personal problems due to pain, disability, reduced quality of life and decreased ability to work and as such constitutes a social economic burden. NP is the most commonly presenting musculoskeletal disorder in primary care physiotherapy in The Netherlands.

Traumatic NP, often referred to as Whiplash-associated Disorders, is regarded as a subgroup of nonspecific neck pain. However, evidence as to whether this subgroup is actually different in terms of type and severity of impairments in physical or mental functions and disability, is lacking. If we know whether, and to what extent, these differ from each other, it provides us an indication whether a different approach in clinical practice and research is needed for nontraumatic NP and traumatic NP.

Both, nontraumatic and traumatic NP, are considered to be complex biopsychosocial disorders with physical and psychological symptoms. As a consequence, the assessment of neck pain requires an appreciation of the interrelationship between biological factors, impairments in physical and mental functions, and disability. In order to consider these aspects, physiotherapists must select appropriate measurement instruments. However, appropriate instruments are not always clinically applicable, or the clinimetric properties of commonly used instruments are insufficient.

The general aims of this thesis were to describe the clinical characteristics of two types of neck pain presenting in primary physiotherapy care, namely nontraumatic neck pain and traumatic neck pain, and to develop and evaluate clinically applicable measurement instruments for assessing impairments and limitations in people with nontraumatic neck pain and traumatic neck pain. Therefore, in this thesis, two new measurement instruments were developed and evaluated. Subsequently, the clinical characteristics, in terms of type and severity of pain, impairments in physical or mental functions and disability, and the complexity of these types of neck pain are presented and discussed, contributing to a more thorough understanding of differences between subgroups of neck pain, the impact of neck pain on the daily functioning of people, and the use of accurate diagnostic instruments in primary care physiotherapy.

Summary of main findings

Evidence shows that reduced cervical mobility is associated with disability in case of neck pain, and can be considered a negative prognostic factor in the recovery of patients with neck pain. The systematic review and meta-analysis presented in **chapter 2**, yielded 6257 hits, and quantified differences in active cervical range of motion (aCROM) between people with neck pain and those without neck pain, in people with nontraumatic NP and traumatic NP, and in people with acute complaints versus those with chronic complaints. The review contained twenty-seven articles, including 2366 participants.

The neck pain group showed significant less aCROM in all movement directions compared with persons without neck pain. Flexion-extension (MD= -28.51°; 95% CI, -40.92° to -16.11°), total rotation (MD= -29.49°; 95% CI, -39.81° to -19.16°), and extension (MD=-17.48°; 95% CI, -20.95° to -14.01°) showed the largest differences. People with traumatic NP had less aCROM than those with nontraumatic NP. Furthermore, the meta-analysis showed that no conclusive differences in aCROM were found between people with acute and people with chronic complaints.

Based on the good-quality evidence of this systematic review, we believe that people with neck pain have significant, clinically relevant decreased aCROM, and a traumatic origin of complaints seems to have a negative influence on aCROM. The duration of symptoms, however, is not an important factor for aCROM.

In the assessment of people with traumatic NP, there are several condition-specific questionnaires available to assess activity limitations and participation restrictions. These questionnaires, however, partially lack adequate content and construct validity because they do not address the specific limitations of these patients and do not measure the constructs purely, while not simultaneously measuring other theoretical constructs. **Chapter 3** describes the development of a new condition-specific and clinically relevant instrument for people with traumatic NP that measures activity limitations and participation restrictions, as defined by the ICF framework. Item generation consisted of (1) a semi-structured interview based on the use of the McMaster-Toronto Arthritis Patient Preference Disability Questionnaire (MACTAR), which was conducted among 69 people with traumatic NP; (2) a Delphi study involving 13 health professionals experienced in the assessment of people with traumatic NP; (3) a literature search for items from self-assessment questionnaires for neck pain. As a result, the 35-item Whiplash Activity and Participation List (WAL) was developed which purely measures

clinically relevant activity limitations and participation restrictions according to the ICF, and is based on patients' opinions and expert opinions.

Chapter 4 describes the validation process of the WAL. A total of 73 people with traumatic NP grade I or II were recruited from physical therapy practices and rehabilitation centres. At baseline, participants completed the WAL, the Short Form-36 and the Sickness Impact Profile. Further, the global level of activity limitation and participation restriction were both assessed on a 5-point scale, and pain severity was scored on a 6-point scale. After a time interval of 1-2 months, the participants were reassessed, and in addition, they rated their neck complaints on a 5-point transition scale.

First, a quantitative item reduction was performed after baseline administration. Subsequently, dimensionality was examined by internal consistency analysis (Cronbach's alpha) and factor analysis. Test-retest reliability was estimated by intraclass correlations and measurement error was calculated by the minimal detectable change (MDC) scores. Construct validity was investigated by testing predefined hypotheses on correlations of the WAL scores with generic health measures and by using the known group method. Responsiveness was expressed as the minimal clinically important change (MCIC) score.

Cronbach's alpha was high (0.95) and unidimensionality was plausible because factor analysis showed 40.3% variance explained by one dominant factor, which was more than 4.5 times larger than the second largest factor. Test-retest reliability was excellent (0.92, 95%CI 0.87-0.95). Construct validity was supported by confirmed hypotheses and the WAL showed statistically significant differences between known groups. MDC was 16 points while the MCIC was 18 points. We therefore consider the WAL as a valid and reliable questionnaire to assess activity limitations and participation restrictions.

There is a lack of valid, reliable, and applicable instruments for measuring planar aCROM and associated 3D coupling motions in people with neck pain. Modern smartphones have advanced sensors and appear to be suitable for these measurements. **Chapter 5** describes the concurrent validity and interrater reliability of the developed iPhone application '3D range of motion' for assessing planar aCROM and associated 3-dimensional coupling motions in people with neck pain. An electromagnetic tracking device (Polhemus Liberty) was used as a reference test. First, preliminary validation tests were carried out on a wooden

phantom (frame) to investigate to which extent the iPhone displays the same angle as the reference test. Mean differences between the reference test and the iPhone were found to be very small, ranging from 0.02° (95%CI -0.15° – 0.19°) for flexion-extension to 0.75° (95%CI 0.56° – 0.94°) for rotation. Subsequently, two samples of people with neck pain were recruited; 30 people for the validity study and 26 people for the reliability study. Intraclass correlation coefficients for concurrent validity and interrater reliability were high, ranging from 0.90 to 0.99. However, 95% limits of agreement (LoA) between the measurement devices were wide, ranging from about 5° for right lateral bending to 11° for total rotation, implying relatively large absolute measurement error between the two raters. Cervical 3-dimensional coupling motions were analysed by calculating the cross-correlation coefficients and ratio between the main motions and coupled motions for both instruments. No significant differences were found between both devices for associated coupling motion analysis. We considered the iPhone application to be a useful discriminative tool for the measurement of planar aCROM and associated coupling motions in people with neck pain. It fulfils the need for a valid, reliable, and feasible instrument in clinical practice and research. Therapists and researchers should, however, consider measurement error when interpreting scores in individual patients.

Estimates of aCROM are influenced by variables such as biologic variation within individuals, the complexity of cervical kinematics, initial head and body position, presence of pain, and psychological state. As several of these factors can vary over the course of the day as well as between days, it is assumed that aCROM may vary accordingly. However, information concerning to which extent aCROM normally varies between measurement sessions, and whether there is a systematic change over the day, is lacking. **Chapter 6** describes the intraday and interday variability, and systematic change over the day of aCROM measurements in asymptomatic people using a clinically applicable iPhone application, in order to better interpret aCROM measurements in clinical practice and research. A prospective observational study was performed, including sixteen adults without neck pain. ACROM was estimated using the iPhone application '3D range of motion'. Measurements were performed 3 times a day, on 7 days, spread over a period of 3 weeks. Mean values of aCROM were calculated and intraday and interday variability was estimated by calculating LoA. The results showed substantial intraday and interday variability of aCROM measurements, with LoA for intraday

variability ranging from $\pm 12.1^\circ$ for left rotation to $\pm 15.5^\circ$ for total rotation. For interday variability the LoA ranged from $\pm 14.2^\circ$ for right rotation to $\pm 20.1^\circ$ for total rotation. No trend towards an increased or decreased aCROM was observed throughout the course of the day. When interpreting aCROM values, clinicians should consider the degree of variation in aCROM measurements over time.

Evidence regarding to what extent people with nontraumatic NP, those with traumatic NP, and pain-free individuals differ regarding type and severity of impairments, disability, and psychological factors, is lacking. In **Chapter 7** we compared the clinical characteristics between people with nontraumatic NP, those having traumatic NP, and pain-free individuals in primary care physiotherapy. In addition, we assessed the differences in these characteristics between people with acute nontraumatic NP versus acute traumatic NP and between people with chronic nontraumatic NP versus chronic traumatic NP. A cross-sectional study was performed including 168 people with traumatic NP, 336 matched patients with nontraumatic NP, and 336 pain-free individuals. Patients with traumatic NP scored significantly worse on pain intensity ($p < 0.01$), had a wider distribution of their neck pain ($p = 0.02$), more restricted cervical flexion-extension (-11.9°) and rotation (-12.4°), less muscle endurance (-25.0 seconds), and more disability ($+14.0\%$), compared to patients with nontraumatic NP. More patients with traumatic NP reported low back pain ($+9.5\%$) and headache ($+12.2\%$) as musculoskeletal comorbidities. As for anxiety, depression, and stress, no differences between the groups were observed ($p > 0.05$). Both patient groups scored significantly worse than pain-free individuals on all characteristics. Patients with nontraumatic NP and traumatic NP experienced different types of activity limitations and participation restrictions.

We concluded that traumatic NP is a more severe condition than nontraumatic NP and should be considered a separate subgroup of neck pain. A different approach in clinical practice and research is required for nontraumatic NP and traumatic NP and synthesis of results in systematic reviews.

Explaining differences in levels of disability between nontraumatic NP and traumatic NP based on single associations between clinical characteristics and disability is likely to be an oversimplification. Complexity between nontraumatic NP and traumatic NP is supposed to differ as several characteristics may interact and affect disability. In **Chapter 8** we therefore compared the associations

between pain, and physical and psychological impairments on the one hand and disability on the other, in people with nontraumatic NP versus those with traumatic NP, and explored interactions between these variables contributing to disability. In a cross-sectional study, we included 2653 people with nontraumatic NP and 249 people with traumatic NP. Multivariable linear regression analyses were performed to explore which characteristics contributed to disability in both patient groups. To assess whether interactions between characteristics differed between groups, three-way interactions were tested. For nontraumatic NP, multivariable analyses revealed that sex ($B=-0.784$), pain intensity ($B=-1.154$), aCROM ($B=-0.030$), neck flexor endurance ($B=-0.019$), anxiety ($B=2.371$), depression ($B=4.137$), and comorbid headache ($B=2.515$) were associated with disability, whereas comorbid low back pain was not. For traumatic NP, pain intensity ($B=1.441$), aCROM ($B=-0.081$), depression ($B=6.228$), comorbid headache ($B=1.923$), and comorbid low back pain ($B=1.809$) were related to disability, while sex, neck flexor endurance, and anxiety were not. Three-way interaction testing revealed that pain, sex, aCROM, and comorbid headache interacted with disability in traumatic NP but not in nontraumatic NP.

We concluded that clinical characteristics were differently associated with disability in people with nontraumatic NP compared to people with traumatic NP. Traumatic NP is a more complex condition as more characteristics interact, and the contribution of individual characteristics to disability is less straightforward. Clinicians and researchers should be aware of complexity in neck pain and avoid reductionist clinical reasoning and research approaches.

Addendum

SAMENVATTING IN HET NEDERLANDS

Nekpijn

Ongeveer 70% van de volwassenen zal ooit in zijn leven een episode van nekpijn doormaken. Acute nekpijn verdwijnt meestal binnen enkele dagen of weken en gaat in de meeste gevallen niet of nauwelijks gepaard met beperkingen in dagelijks functioneren. Een aanzienlijk deel van de mensen ervaart echter aanhoudende klachten en 50 tot 85% van de mensen met nekpijn heeft na een jaar nog steeds of opnieuw nekpijn. Nekpijn kan aanzienlijke persoonlijke problemen met zich meebrengen zoals pijn en bewegingsbeperkingen, beperkingen in functioneren, verminderde kwaliteit van leven en een verminderd vermogen om te werken. Het vormt daardoor ook een substantieel sociaal- economisch probleem. Nekpijn is na lage rugpijn de meest voorkomende musculoskeletale aandoening die gezien wordt in de eerstelijns fysiotherapie in Nederland.

Traumagerelateerde nekpijn, vaak aangeduid als 'whiplash associated disorder', wordt beschouwd als een subgroep van specifieke nekpijn. Er ontbreekt echter wetenschappelijk bewijs of deze subgroep daadwerkelijk verschilt wat betreft ernst van pijn, fysieke of mentale functiebeperkingen en de impact op het functioneren in het dagelijks leven. Als we weten of, en in welke mate, traumagerelateerde nekpijn afwijkt van niet-traumagerelateerde nekpijn, geeft dit ons een indicatie of er een verschillende aanpak nodig is in de klinische praktijk en bij de opzet en interpretatie van wetenschappelijk onderzoek.

Zowel traumagerelateerde als niet-traumagerelateerde nekpijn worden beschouwd als complexe aandoeningen waarbij interacterende biologische, psychologische en sociale factoren een belangrijke rol spelen bij het ontstaan en voortbestaan van de klachten. Daarom vereist het diagnostisch proces bij mensen met nekpijn een analyse van de interactie tussen deze factoren. Om met deze factoren rekening te kunnen houden en op waarde te kunnen schatten, moeten fysiotherapeuten geschikte meetinstrumenten gebruiken. Deze meetinstrumenten zijn echter niet altijd klinisch toepasbaar en ook de klinimetrische eigenschappen van deze meetinstrumenten zijn vaak onvoldoende.

Het algemene doel van dit proefschrift is het beschrijven van de klinische impact die traumagerelateerde nekpijn en niet-traumagerelateerde nekpijn hebben op het functioneren in het dagelijks leven. Daarvoor worden in dit proefschrift twee nieuwe meetinstrumenten ontwikkeld en geëvalueerd. Vervolgens worden de klinische kenmerken, in termen van ernst van de pijn, fysieke en mentale functiebeperkingen, de impact op het functioneren, en de complexiteit van beide

subgroepen van nekpijn gepresenteerd en besproken. Dit draagt bij aan een beter begrip van verschillen tussen deze subgroepen, de impact van nekpijn op het dagelijks functioneren, en het gebruik van adequate diagnostische meetinstrumenten in de eerstelijns fysiotherapie.

Samenvatting van de belangrijkste bevindingen

Literatuur suggereert dat verminderde cervicale mobiliteit geassocieerd is met beperkingen in het dagelijks functioneren bij mensen met nekpijn, en beschouwd kan worden als een negatieve prognostische factor in het herstel van mensen met nekpijn. De systematische review en meta-analyse, gepresenteerd in **hoofdstuk 2**, kwantificeerde de verschillen in actieve cervicale range of motion (aCROM) tussen mensen met nekpijn en mensen zonder nekpijn, tussen mensen met traumagerelateerde nekpijn en niet-traumagerelateerde nekpijn, en tussen mensen met acute klachten versus mensen met chronische klachten. De literatuurstudie leverde 6257 hits op en bevatte 27 artikelen, met in totaal 2366 deelnemers. De nekpijngroep vertoonde significant minder aCROM in alle bewegingsrichtingen vergeleken met mensen zonder nekpijn. Flexie-extensie (MD= -28.51° ; 95%CI, -40.92° tot -16.11°), totale rotatie (MD= -29.49° ; 95%CI, -39.81° tot -19.16°), en extensie (MD= -17.48° ; 95%CI, -20.95° tot -14.01°) vertoonden de grootste verschillen. Mensen met traumagerelateerde nekpijn hadden minder aCROM dan mensen met niet-traumagerelateerde nekpijn. Verder bleek uit de meta-analyse dat er geen klinisch relevante verschillen in aCROM zijn tussen mensen met acute klachten en mensen met chronische klachten. Geconcludeerd kan worden dat mensen met nekpijn een significant en klinisch relevant verminderde aCROM hebben ten opzichte van mensen zonder nekpijn, en dat een traumatische oorsprong van de klachten een negatieve invloed lijkt te hebben op aCROM. De duur van de klachten bleek geen belangrijke factor voor aCROM.

Voor het diagnostisch proces bij mensen met traumagerelateerde nekpijn zijn er verschillende aandoeningspecifieke vragenlijsten beschikbaar om beperkingen in dagelijkse activiteiten en participatieproblemen in kaart te brengen. Deze vragenlijsten missen echter deels adequate inhouds- en constructvaliditeit omdat ze niet ingaan op de specifieke beperkingen van deze patiëntgroep en constructen niet zuiver meten omdat ze ook items bevatten die andere theoretische constructen meten. **Hoofdstuk 3** beschrijft de ontwikkeling van een nieuwe

aandoenings specifieke en klinisch relevante vragenlijst voor mensen met traumagerelateerde nekpijn die beperkingen in dagelijkse activiteiten en participatieproblemen meet, volgens de definities van The International Classification of Functioning, Disability and Health (ICF). Het genereren van items bestond uit (1) een semigestructureerd interview gebaseerd op het gebruik van de McMaster-Toronto Arthritis Patient Preference Disability Questionnaire (MACTAR), dat werd afgenomen bij 69 mensen met traumagerelateerde nekpijn; (2) een Delphi-studie waar 13 zorgprofessionals bij betrokken waren die expertise hadden op het gebied van traumagerelateerde nekpijn; (3) een literatuuronderzoek naar items uit bestaande vragenlijsten voor nekpijn. Als resultaat werd de 35-item Whiplash Activiteit- en Participatielijst (WAL) ontwikkeld (score range 0–140), die louter klinisch relevante beperkingen in dagelijkse activiteiten en participatieproblemen meet volgens de ICF, en gebaseerd is op de meningen van patiënten en meningen van deskundigen.

Hoofdstuk 4 beschrijft het validatieproces van de WAL. In totaal werden 73 mensen met traumagerelateerde nekpijn graad I of II gerekruteerd uit eerstelijns fysiotherapiepraktijken en revalidatiecentra. Op baseline vulden de deelnemers de WAL en de generieke gezondheidsvragenlijsten Short Form-36 en Sickness Impact Profile in. Verder werden het ervaren niveau van beperkingen in dagelijkse activiteiten en participatieproblemen beoordeeld op een 5-puntsschaal, en werd de ernst van de pijn gescoord op een 6-puntsschaal. Na een tijdsinterval van 1–2 maanden werden de deelnemers opnieuw bevraagd, en aanvullend beoordeelden ze de eventuele verandering van hun nekpijn op een 5-punts transitie-schaal. Eerst werd een kwantitatieve itemreductie uitgevoerd na de baseline meting. Vervolgens werd de dimensionaliteit van de lijst onderzocht met een interne consistentieanalyse (Cronbach's alpha) en een factoranalyse. De test-hertest betrouwbaarheid werd geschat door intraclass correlatie coëfficiënt, en de meetfout werd berekend door de minimaal detecteerbare verandering (MDC). De constructvaliditeit werd onderzocht door het testen van vooraf opgestelde hypothesen over correlaties van de WAL-scores met generieke gezondheidsvragenlijsten en door het gebruik van de 'know-group' methode. De responsiviteit werd uitgedrukt als de minimaal klinisch relevante veranderscore (MCIC).

Cronbach's alpha was hoog (0,95) en unidimensionaliteit was plausibel omdat uit de factoranalyse bleek dat 40,3% van de variantie verklaard werd door één dominante factor, die meer dan 4,5 keer groter was dan de op één na grootste

factor. De test-hertest betrouwbaarheid was uitstekend (0,92, 95%CI 0,87 tot 0,95). De constructvaliditeit werd ondersteund door de bevestigde hypothesen en de WAL toonde statistisch significante verschillen tussen groepen die klinisch relevant verschillen in de mate van beperkingen in activiteiten en participatieproblemen. De MDC was 16 punten terwijl de MCIC 18 punten was. Wij beschouwen de WAL daarom als een valide en betrouwbare vragenlijst om beperkingen in dagelijkse activiteiten en participatieproblemen te meten.

Er is een gebrek aan valide, betrouwbare en klinisch toepasbare meetinstrumenten voor het meten van unidimensionele en 3-dimensionele cervicale bewegingen bij mensen met nekpijn. Moderne smartphones hebben geavanceerde sensoren die geschikt lijken te zijn voor deze metingen.

Hoofdstuk 5 beschrijft de concurrente validiteit en interbeoordelaarsbetrouwbaarheid van de zelf ontwikkelde iPhone applicatie '3D range of motion' voor het meten van cervicale bewegingen en de koppeling tussen de 3 bewegingscomponenten, te weten flexie- extensie, axiale rotatie en lateroflexie bij mensen met nekpijn. Elektromagnetische registratieapparatuur (Polhemus Liberty) werd gebruikt als referentietest. Eerst werden, ter kalibratie, validatietests uitgevoerd op een houten frame om te onderzoeken in hoeverre de iPhone dezelfde hoek weergeeft als de referentietest. De gemiddelde verschillen tussen de referentietest en de iPhone bleken zeer klein te zijn, variërend van 0,02° (95%CI -0,15° tot 0,19°) voor flexie-extensie tot 0,75° (95%CI 0,56° tot 0,94°) voor axiale rotatie. Vervolgens werden twee steekproeven van mensen met nekpijn gerekruteerd; 30 mensen voor de validiteitsstudie en 26 mensen voor de betrouwbaarheidsstudie. Intraclass correlatie coëfficiënten voor concurrente validiteit en interbeoordelaarsbetrouwbaarheid waren hoog, variërend van 0,90 tot 0,99. De 95% limits of agreement (LoA) tussen de meetinstrumenten bleken echter breed, variërend van ongeveer 5° voor rechts lateroflexie tot 11° voor totale axiale rotatie, wat een relatief grote absolute meetfout tussen de twee beoordelaars impliceert. Cervicale 3-dimensionele gekoppelde bewegingen werden geanalyseerd door berekening van de kruiscorrelatiecoëfficiënten en de verhouding tussen de hoofdbewegingen en de gekoppelde bewegingen voor beide instrumenten. Er werden geen significante verschillen gevonden tussen beide apparaten voor de analyse van deze gekoppelde bewegingen.

Op basis van deze resultaten beschouwden wij de iPhone applicatie als een geschikt instrument voor de meting van aCROM met de bijbehorende gekoppelde bewegingen bij mensen met nekpijn. Het voorziet in de behoefte aan een valide,

betrouwbaar en klinisch toepasbaar meetinstrument in de dagelijkse praktijk en in wetenschappelijk onderzoek. Therapeuten en onderzoekers moeten echter wel rekening houden met de meetfout wanneer zij de scores interpreteren voor individuele patiënten.

Metingen van aCROM worden beïnvloed door variabelen zoals biologische variatie binnen individuen, de complexiteit van de cervicale kinematica, de hoofd- en lichaamspositie, de aanwezigheid van pijn, en de psychosociale situatie. Aangezien verschillende van deze factoren in de loop van de dag en van dag tot dag kunnen variëren, wordt aangenomen dat aCROM eveneens kan variëren. Het ontbreekt echter aan informatie over de mate waarin aCROM van nature varieert tussen meetsessies en of er sprake is van een systematische verandering in de loop van de dag. **Hoofdstuk 6** beschrijft de mate van variabiliteit van aCROM binnen en tussen dagen, alsook de systematische verandering over de dag. Hiervoor werd een prospectieve observationele studie met zestien volwassenen zonder neklachten uitgevoerd. ACROM werd geregistreerd met behulp van de iPhone applicatie '3D range of motion'. Metingen werden 3 keer per dag uitgevoerd, op 7 verschillende dagen, verspreid over een periode van 3 weken. De gemiddelde waarden van aCROM werden berekend, en de variabiliteit binnen en tussen dagen werd bepaald door de LoA te berekenen. De resultaten toonden aanzienlijke variabiliteit binnen en tussen dagen, met LoA's voor variabiliteit binnen dagen variërend van $\pm 12.1^\circ$ voor links rotatie tot $\pm 15.5^\circ$ voor de totale rotatie. Voor variabiliteit tussen dagen varieerde de LoA van $\pm 14.2^\circ$ voor rechts rotatie tot $\pm 20.1^\circ$ voor totale rotatie. In het verloop van de dag werden geen trends naar een toename of afname van aCROM waargenomen. Clinici wordt aanbevolen om bij de interpretatie van aCROM-waarden rekening te houden met de mate van variatie in aCROM-metingen in de loop van de tijd.

Evidentie over de mate waarin mensen met traumagerelateerde nekpijn verschillen van mensen met niet-traumagerelateerde nekpijn op het gebied van pijnintensiteit, functiestoornissen, beperkingen in functioneren en psychologische factoren, ontbreekt. In **hoofdstuk 7** vergeleken we daarom de klinische kenmerken tussen mensen met traumagerelateerde nekpijn, mensen met niet-traumagerelateerde nekpijn, en mensen zonder nekpijn in de eerstelijns fysiotherapie praktijk. Daarnaast hebben we de verschillen in klinische kenmerken onderzocht tussen mensen met acute traumagerelateerde nekpijn ten opzichte van mensen met acute niet-traumagerelateerde nekpijn en tussen mensen met

chronische traumagerelateerde nekpijn ten opzichte van mensen met chronische niet-traumagerelateerde nekpijn. Hiervoor werd een cross-sectionele studie uitgevoerd met 168 mensen met traumagerelateerde nekpijn, 336 gematchte personen met niet-traumagerelateerde nekpijn, en 336 pijnvrije personen. Personen met traumagerelateerde nekpijn scoorden significant hoger op pijnintensiteit ($p < 0,01$), hadden meer wijdverspreide pijn ($p = 0,02$), meer beperkte cervicale flexie-extensie ($-11,9^\circ$) en rotatie ($-12,4^\circ$), minder spieruithoudingsvermogen ($-25,0$ seconden), en meer beperkingen in functioneren ($+14,0\%$), vergeleken met patiënten met niet-traumagerelateerde nekpijn. Meer personen met traumagerelateerde nekpijn rapporteerden lage rugpijn ($+9,5\%$) en hoofdpijn ($+12,2\%$) als comorbiditeit. Er werden geen verschillen tussen de groepen waargenomen ($p > 0,05$) voor angst, depressie en stress. Beide patiëntengroepen scoorden significant slechter dan pijnvrije mensen op alle kenmerken. Beide patiëntengroepen bleken bovendien van elkaar te verschillen in type beperkingen in dagelijkse activiteiten en participatieproblemen. Wij concludeerden dat de gevolgen voor het fysiek functioneren ernstiger zijn voor personen met traumagerelateerde nekpijn dan voor personen met niet-traumagerelateerde nekpijn en dat hier sprake is van klinisch verschillende subgroepen van aspecifieke nekpijn. Op basis hiervan is het aan te bevelen om een specifiek klinisch redeneerproces te hanteren in de klinische praktijk, bij de opzet van onderzoek en bij de synthese van resultaten in systematische reviews.

Het verklaren van verschillen in niveaus van dagelijks functioneren tussen traumagerelateerde nekpijn en niet-traumagerelateerde nekpijn op basis van enkelvoudige associaties tussen klinische kenmerken en beperkingen in dagelijks functioneren is waarschijnlijk een simplificatie van de werkelijkheid. Er wordt verondersteld dat traumagerelateerde nekpijn en niet-traumagerelateerde nekpijn verschillen in complexiteit, omdat verschillende klinische kenmerken anders met elkaar interacteren en daarmee de beperkingen in functioneren op verschillende wijze kunnen beïnvloeden. In **hoofdstuk 8** vergeleken we daarom voor beide patiëntengroepen de associaties tussen pijn en lichamelijke en psychische symptomen enerzijds en beperkingen in functioneren anderzijds, en onderzochten we interacties tussen deze variabelen in de bijdrage aan beperkingen in functioneren. In een cross-sectionele studie includeerden wij 2653 mensen met niet-traumagerelateerde nekpijn en 249 mensen met traumagerelateerde nekpijn.

Er zijn multivariabele lineaire regressieanalyses uitgevoerd om te onderzoeken welke kenmerken bijdragen aan de beperkingen in functioneren van beide patiëntgroepen. Om te beoordelen of interacties tussen kenmerken verschillen tussen de groepen, werden drieweg interactie-analyses uitgevoerd. Voor niet-traumagerelateerde nekpijn toonden multivariabele analyses aan dat geslacht ($B=-0,784$), pijnintensiteit ($B=-1,154$), aCROM ($B=-0,030$), uithoudingsvermogen van de nekflexoren ($B=-0,019$), angst ($B=2,371$), depressie ($B=4,137$), en comorbiditeit hoofdpijn ($B=2,515$) geassocieerd zijn met beperkingen in functioneren, terwijl comorbiditeit lage rugpijn dat niet is. Voor traumagerelateerde nekpijn bleken pijnintensiteit ($B=1,441$), aCROM ($B=-0,081$), depressie ($B=6,228$), comorbiditeit hoofdpijn ($B=1,923$), en comorbiditeit lage rugpijn ($B=1,809$) geassocieerd met beperkingen in functioneren, terwijl geslacht, uithoudingsvermogen van de nekflexoren en angst, dat niet zijn. Drieweg interactie-effecten lieten zien dat pijn, geslacht, aCROM, en comorbiditeit hoofdpijn samenhangen met beperkingen in functioneren bij traumagerelateerde nekpijn maar juist niet bij niet-traumagerelateerde nekpijn.

Wij concludeerden dat klinische kenmerken anders geassocieerd zijn met beperkingen in functioneren bij mensen met traumagerelateerde nekpijn in vergelijking met mensen met niet-traumagerelateerde nekpijn. Traumagerelateerde nekpijn lijkt een complexere aandoening omdat er meer kenmerken op elkaar inwerken, en de bijdrage van individuele kenmerken aan beperkingen in functioneren minder eenduidig is. Clinici en onderzoekers moeten zich bewust zijn van de complexiteit van nekpijn, en reductionistische klinisch redeneerprocessen in de praktijk en bij de opzet van de methodologie van onderzoek vermijden.

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Addendum

CURRICULUM VITAE
LIST OF PUBLICATIONS

CURRICULUM VITAE

Martijn Stenneberg werd geboren in Amsterdam op 29 december 1977 en groeide op in Amstelveen samen met zijn ouders, broer en zus. Hij behaalde zijn vwo-diploma in 1996 aan het Hermann Wesselink College te Amstelveen. In hetzelfde jaar begon hij de studie bewegingswetenschappen aan de Vrije Universiteit te Amsterdam. Hij kwam er echter al snel achter dat wetenschappelijk onderzoek doen *toch echt niets voor hem was* en besloot de opleiding fysiotherapie te gaan volgen aan de Hogeschool van Amsterdam (1998-2002). Voor zijn afstudeeropdracht werkte hij als junior onderzoeker mee aan een onderzoek van de Vrije Universiteit te Amsterdam naar de effecten van loopsnelheid en oefentherapie op de romprotatie van Parkinsonpatiënten tijdens het lopen, wat leidde tot zijn eerste publicatie als co-auteur in een Nederlands tijdschrift. Na zijn afstuderen werkte hij van 2002 tot 2013 met veel plezier bij de particuliere praktijk voor fysiotherapie en manuele therapie Praktijk Deen te Heemstede. Hier raakte hij geïnteresseerd in het vak van manueeltherapeut. In 2006 startte hij dan ook met de masteropleiding manuele therapie aan de destijds genaamde Stichting Opleiding Manuele Therapie (SOMT) te Amersfoort. Hij volgde het excellententracé en studeerde in 2008 cum laude af. Tijdens het laatste jaar van zijn studie begon hij al met lesgeven als tutor bij SOMT onder de inspirerende leiding van Maarten Schmitt. Hij startte vervolgens in 2009 met de Master Evidence Based Practice in Health Care aan de Universiteit van Amsterdam, om daar in 2011 af te studeren als klinisch epidemioloog. Zijn afstudeerwerk resulteerde in zijn eerste internationale publicatie. In 2013 werd hij benoemd als hoofd van de masteropleiding manuele therapie van SOMT. In datzelfde jaar is hij ook gestart met zijn promotieonderzoek aan de Universiteit van Maastricht in samenwerking met SOMT. Sinds april 2021 is hij aangesteld als titularis 'evidence-based klinisch redeneren in de manuele therapie' aan de Vrije Universiteit Brussel. Naast deze werkzaamheden is hij thesiscoördinator en lid van de curriculumcommissie en toetscommissie van de universitaire opleiding fysiotherapie & revalidatiewetenschappen van SOMT. Martijn was verder bestuurslid en medeoprichter van de alumnivereniging van SOMT, 'VASTUS'.

Martijn woont samen met Jolijn Jonker en hun drie kinderen, Finn, Loet en Bent, in Amstelveen.

LIST OF PUBLICATIONS

Publications in international peer-reviewed journals

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AWARD

Beste Posterpresentatie. Concurrent validity and interrater reliability of a new smartphone application to assess 3D active cervical range of motion in patients with neck pain. Symposium Thoracic Park, Nederlandse Vereniging voor Manuele Therapie, 7 april 2018

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