Complaints of the Arm, Neck and/or Shoulder

A new approach to its terminology and classification: the CANS model

Bionka M.A. Huisstede

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Klachten in de arm-, nek- en/of schouderregio
Een nieuwe benadering van terminologie en indeling:
Het CANS model

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“Tu, pro tua sapientia, debebis optare optima, cogitare difficillima, ferre quacumque erunt”

(Cicero, Epistulae 9.17.3)

Voor mijn ouders

Contents

Chapter 1

Introduction

Musculoskeletal disorders of the upper extremity and neck are extremely common and one of the major causes of disability, sickness absence and health care usage worldwide. Although these conditions are not life-threaten they cause the patient considerable discomfort and are a financial burden on society.
The absence of unambigious terminology and classification in musculoskeletal upper-extremity disorders hampers communication between health care professionals and makes it difficult to compare results of clinical research. The aim of this thesis is to contribute to the accomplishment of uniformity in this field.

History of terminology

The term musculoskeletal refers to conditions that involve the nerves, tendons, muscles, and supporting structures of the body.[1] Various names are given to upper-extremity musculoskeletal disorders depending on the country of origin: For example, cervicobrachial syndrome in Japan, repetitive strain injury (RSI) in Australia, cumulative trauma disorders (CTDs) of the upper extremity in North America, and work-related upper-extremity musculoskeletal disorders (WRUMD) in the USA.[2] The oldest description of upper-extremity musculoskeletal disorders can be found in the revised edition of the classic book 'De Morbis Artificum Diatrib' ("Diseases of workers") written by Bernardino Ramazzini in 1713.[3] Ramazzini recognized that problems of the upper extremity can arise from standing and sitting postures, repetitive motions, and mental stress: "I have noticed bakers with swelled hands, and painful, too; in fact the hands of all such workers become much thickened by the constant pressure of kneading the dough." "The maladies that affect the clerks arise from three causes; first, constant sitting; secondly, incessant movement of the hand and always in the same direction; and thirdly, the strain on the mind..." "The incessant driving of the pen over paper causes intense fatigue of the hand and the whole arm because of the continuous strain ... on the muscles and tendons".[4]

Prevalence and incidence of upper-extremity disorders

Although upper-extremity musculoskeletal disorders represent a large proportion of illnesses worldwide, it is difficult to precisely estimate their actual occurrence. Most studies that reported incidence or prevalence rates refer to the working population only. For example, 41% of the newspaper employees in the USA[5] and almost 20% of the newspaper employees in Canada[6] reported upper-extremity musculoskeletal disorders during the previous year. In Europe, the 'European Foundation for the improvement of living and working conditions' studied differences in the occurrence of musculoskeletal upper-extremity disorders in the

working population in 15 European countries.[7] Highest prevalence rates in the working population in Europe were found in the Scandinavian countries (33-54% arm-shoulder complaints; 17-25% arm complaints). Also in Greece a high prevalence (27%) of arm pain was reported. The prevalence rates found for the Netherlands in the same study were 21% neck-shoulder complaints and 9% arm complaints. In the Netherlands, the 12-month prevalence of upper-extremity disorders increased from 26% to 28% between 2000 and 2002.[8] Other studies in several countries also report on a rapidly increasing occurrence of work-related musculoskeletal disorders of the upper extremities.[9]

It is thus clear that upper-extremity disorders affect many people all over the world. However, to establish whether there is a perceptible increase in these problems, a review of the scientific literature is required.

Classification

The epidemiological issues and treatment options with regard to disorders of the upper extremity and neck have been well explored. However, interpretation of the outcomes and comparison of the results of the different studies is seriously hampered by the use of different case definitions.[10] The multiplicity of approaches to the terminology and classification also confuses clinicians and paramedical staff. Moreover, it is difficult to optimize policy-making and disease treatment in the absence of unambiguous communication.

In 1997 in the United Kingdom, Harrington and colleagues[11] achieved consensus on diagnostic criteria of eight specific upper-extremity disorders and one disorder designed as non-specific. The results from a Delphi process were used for a final workshop in which consensus was achieved. Following the consensus statement achieved in this workshop, a structured examination schedule was developed for diagnosing musculoskeletal disorders of the upper limb. This so-called Southampton examination schedule was tested in a hospital setting by nurses and physicians trained in the examination schedule. The protocol was found to be repeatable, had the benefit of face validity and construct validity, and showed acceptable diagnostic accuracy.[12]

Sluiter et al.[13] produced a criteria document for evaluating the work-relatedness of upper-extremity musculoskeletal disorders'. The resulting document (102 pages) described the signs and symptoms of 11 specific musculoskeletal disorders. Non-specific upper-extremity musculoskeletal disorder is mentioned as a twelfth diagnosis. A final workshop, consisting of experts in these disorders, was held to define the consensus criteria.

Until now, various attempts have been made to achieve consensus on the diagnostic criteria of upper-extremity disorders[10], but none of the proposed diagnostic systems has provided a complete overview of musculoskeletal upper-extremity disorders. Moreover, they did not produce a workable classification tool that can be used in daily practice in an easy way (i.e.

no special training and/or no substantial time needed to perform) by both researchers and health professionals.
Therefore, a new classification system is required that includes all upper-extremity conditions, and that can be considered in the larger context of mono- and multidisciplinary communication regarding upper-extremity musculoskeletal disorders.

Effectiveness of treatment of specific musculoskeletal upper-extremity disorders

Upper-extremity musculoskeletal disorders are generally divided into specific and non-specific complaints. A specific disorder can be seen as an entity that is recognizable by unique characteristics, which can be established by case history, physical examination, imaging and/or laboratory testing. A non-specific complaint is seen as a diagnosis by a process of exclusion.
For some specific disorders, such as carpal tunnel syndrome and lateral epicondylitis, randomized clinical trials (RCTs), controlled clinical trails (CCTs), or systematic reviews on the effectiveness of therapeutic interventions are available. Various non-surgical interventions such as medication (NSAIDs, paracetamol etc.), steroid injection and immobilization, as well as surgical intervention, have been studied in controlled clinical trials.[14]
For other (low-incidence) disorders, (such as Guyon canal syndrome and radial tunnel syndrome), no controlled studies but only observational studies are available. So, systematic reviews based on RCTs and CCTs are not available. Therefore, it may be useful to systematically review all observational studies that have investigated the effectiveness of interventions for these conditions. Although the internal validity of observational studies is generally inferior to that of controlled trials, these studies may provide valuable tendencies for the efficacy of treatment options and may guide future research, for example in the design of new RCTs.

Measurement of disability of the entire upper extremity and neck

Many questionnaires have been developed to evaluate the impact and course of disorders of the upper extremity and neck. According to kinesiologic and biomechanical theories, the upper extremity acts as a single functional unit.[15] One of the questionnaires used to assess disability of the arm, shoulder and hand is the Disabilities of the Arm, Shoulder and Hand (DASH) questionnaire. This questionnaire was developed as an outcome measure that conceptualizes the upper extremity as a single functional unit. It has been assessed regarding its reliability, validity and responsiveness in a variety of arm disorders.[14,16-19] The DASH questionnaire is now available in several languages and its use is growing rapidly.[20]

Because neck complaints are common in patients with upper-extremity disorders, a valid and responsiveness questionnaire designed for the whole upper extremity, including the neck, would be useful and practical in upper-extremity research. Maybe, the DASH can contribute to this statement. Therefore, studies are needed to evaluate the validity and responsiveness of the DASH questionnaire in persons with complaints in the whole upper extremity, including the neck.

Outline of this thesis

Chapter 2 of this thesis presents an overview of the scientific literature reporting the worldwide incidence and prevalence rates of musculoskeletal disorders of the upper extremity and neck, in order to establish the range of these estimates in various countries and to determine whether the rates are increasing over time. Comparison of such studies is hampered by the absence of a universally accepted terminology and classification of these complaints. Therefore, we initiated a project with the aim to achieve multidisciplinary consensus on this topic.

The results of the Delphi consensus strategy that we used to achieve the consensus are described in **chapter 3.** In order to contribute to systematic overviews on the effectiveness of interventions for specific upper-extremity disorders in the absence of controlled trials, **chapter 4 and 5** present our systematic reviews of observational studies on interventions for treating the radial tunnel syndrome and the posterior interosseus nerve syndrome.

Chapter 6 focuses on the validity and responsiveness of the Disabilities of the Arm, Shoulder and Hand (DASH) questionnaire for patients with complaints of the whole upper extremity, including the neck.

Based on the results of chapter 3 (i.e. the consensus on the terminology and classification), in **chapter 7** we studied the prevalence and socio-demographic and health characteristics, and use of health care related to disorders of the upper extremity and neck as defined by our experts in the Delphi consensus strategy.

Chapter 8 discusses the findings of the previous chapters and recommendations are made for future research. Finally, an English and Dutch summary of the work in this thesis is given.

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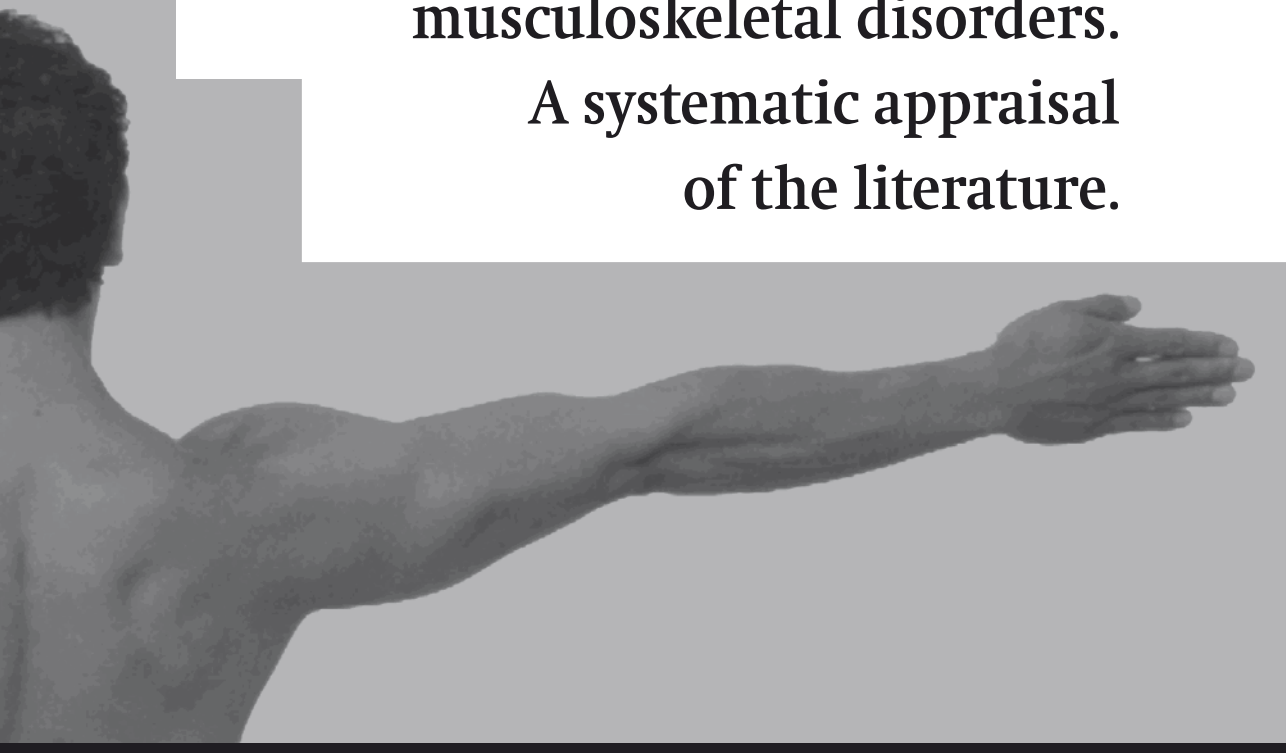
Chapter 2

Incidence and prevalence of upper-extremity musculoskeletal disorders. A systematic appraisal of the literature.



Huisstede BM, Bierma-Zeinstra SM, Koes BW, Verhaar JA.

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Abstract

Background: A systematic appraisal of the worldwide incidence and prevalence rates of upper-extremity disorders (UEDs) available in scientific literature was executed to gauge the range of these estimates in various countries and to determine whether the rates are increasing in time.

Methods: Studies that recruited at least 500 people, collected data by using questionnaires, interviews and/or physical examinations, and reported incidence or prevalence rates of the whole upper extremity including neck, were included.

Results: No studies were found with regard to the incidence of UEDs and 13 studies that reported prevalence rates of UEDs were included. The point prevalence ranged from 1.6-53%; the 12-months prevalence ranged from 2.3-41%. One study reported on the lifetime prevalence (29%). We did not find evidence of a clear increasing or decreasing pattern over time. The case definitions for UEDs used in the studies, differed enormously. Therefore, it was not possible to pool the data.

Conclusions: There are substantial differences in reported prevalence rates on UEDs. Main reason for this is the absence of a universally accepted way of labelling or defining UEDs. If we want to make progress in this field, the first requirement is to agree on unambiguous terminology and classification of EUDs.

Introduction

Upper-extremity disorders (UEDs) are a major problem in modern society. Besides the impact on patients themselves, the disorders also form a huge economic burden due to costs for sick leave and health care. UEDs affect people all over the world. In the early 1980's in Australia Hocking[1] even reported an epidemic of a disorder he called RSI (repetitive strain injury). Numerous other terms have been used to indicate UEDs such as cumulative trauma disorders, physical overuse syndrome, and occupational cervicobrachial disorders. UEDs comprise various clinically defined (e.g. carpal tunnel syndrome) and undefined conditions of muscles, tendons, or nerves in the upper extremity due to multiple factors. Not only occupational use of the upper limbs, but also psychosocial work characteristics such as high job stress[2], high job demand[3], non-work-related stress[2] and personal characteristics such as coping[4] can cause UEDs. Most UEDs are manifested by pain, discomfort, or tingling in the upper extremity.[5]

In medical literature authors repeatedly suggested that during the last decade's data are reported to indicate the extent, and in some cases increase of UEDs over time in Australia, Canada, the USA, France, The Netherlands, and elsewhere.[1,2,6-11] For example, based on workers' compensation claims Silverstein et al.[12] reported a dramatic increase of UEDs since the early 1980s in the USA affecting workers in virtually every industry. In 1981, 28,6% of the allowed workers' compensation claims in New York State concerned UEDs and by 1986, these numbers were increased by 10,2%.[13] In 1989 the total U.S. workers compensation costs for UEDs was estimated to be $563 million.[14] Also in the early 1990s UEDs have dramatically increased in incidence according the data from the U.S. Bureau of Labor Statistics, 1998a.[15] In Ontario, Canada, UEDs constituted up to 24% of lost-time workers compensation claims in 1992.[16] In 2000/01, one in ten Canadians aged 20 or older reported an UED that was serious enough to limit their normal activities in the previous 12 months.[17] In 2000 the Health Council of the Netherlands reported that if no distinction is made on the basis of duration or seriousness, the prevalence of UEDs in the Netherlands was between 20 and 40 percent.[18]

A systematic appraisal of worldwide incidence and prevalence studies may permit us to gauge the range of incidence and prevalence of UEDs in various countries and, where possible, to pool data. It provides the basis for determining whether these estimates of UEDs are increasing over time. The data are also needed to estimate the size of study populations for experimental and preventive trials. Therefore, a systematic appraisal of the worldwide incidence and prevalence rates of UEDs reported in available studies will be presented here.

Methods

Literature search

Studies were identified by searches of the computerized bibliography database Medline (1966 to June 2004). All the keywords mentioned for UEDs in relevant articles were used in the literature search, such as repetitive strain injury (RSI), upper-extremity disorders (UED), work-related musculoskeletal disorders (WMSD), and cumulative trauma disorders (CTD). In order to identify relevant studies for this review, these keywords were combined with the terms "prevalence" and "incidence" in the title or abstract. On the basis of title and abstract articles were excluded in which prevalence and UEDs were no issue. Full texts of the remaining articles were assessed on eligibility.

Eligibility of studies

Studies were eligible for inclusion if (1) at least 500 people were included in the study; (2) incidence or prevalence rates of UEDs were reported for the whole upper-extremity region including neck and (3) data were collected by using questionnaires, interviews and/or physical examinations. When incidence or prevalence rates were only presented for neck, shoulder, elbow or hand separately, the study was excluded. Studies based on administrative data such as data from workers' compensation claims or from registrations of occupational health services were excluded because these studies may represent changes in administrative policy and economical matters rather than the actual incidence or prevalence.
Studies that recruited persons from the open population as well as from a selected population (working, non-working, primary care, secondary care, etc.) were included. Only studies written in English, French, German, and Dutch were considered.

Data extraction

Relevant data were collected from eligible studies on standardized forms concerning incidence or prevalence rates, the used term, definition for UEDs, the year of measurement, the setting and the country in which the study was carried out.

Pooling of data

Before data can be pooled from different studies, the homogeneity of the data should be taken into account. The minimum criteria for data pooling in this systematic appraisal were the use of similar case definitions of UEDs, homogeneity of the study population, and the use of similar types of incidence or prevalence rates.

Results

Study selection

The search strategy resulted in a total of 523 studies. After the first eligibility screening, based on title and abstract, 206 potentially relevant articles were identified. Reviewing full text articles, 47 studies reporting incidence or prevalence of UEDs consisting of a population of 500 cases or more were found. Of these, 13 studies met the inclusion criteria. They all reported prevalence rates. No studies were found with regard to the incidence of UEDs that met the inclusion criteria. One study[19] was found that studied the prevalence in nurses by asking the following question: "Do you suffer regularly from arm or neck complaints?" Because 'regularly' is not defined we decided to exclude this study.

Study characteristics

The 13 studies included in this review are presented in Table 1, together with their relevant characteristics. All studies were published between 1987-2003; the data of the studies were collected between 1983-1998. Six studies were executed in the USA. In Canada two studies were carried out. The other studies were from Australia, England, Italy, The Netherlands and Sweden. The majority of the studies (seven) focused on a working population that was expected to be at high risk for UEDs, whereas two studies focused on a low risk working population. Two studies concerned students and the other two studies were carried out in the general population.

The studies reported different types of prevalence rates, i.e., point prevalence (six studies), 12-month prevalence (six studies) and lifetime prevalence (one study). The occurrence of UEDs was assessed either through questionnaires (eight studies), a telephone interview (one study), a questionnaire and clinical examination (two studies), or an interview and a clinical examination (two studies).

Case definition of UEDs

A diversity of terms and case definitions for UEDs were used (Table 2). Three of the six studies reporting point prevalence rates[20-22] did not present any definition of UEDs. Ehrmann Feldman et al.[23] defined UEDs as 'having substantial neck and upper limb pain' and Fry[24] described the 'overuse (injury) syndrome' as 'those changes brought about in the muscle and joint ligaments from excessive use, causing pain, loss of function, and almost always demonstrable tenderness in the affected structure'. McCormack et al.[25] used in addition a (specified) physical examination to define UEDs.

One of the six studies reporting 12-months prevalence rates did not give a definition of UEDs. The authors of this study[26] reported about neck and upper-extremity symptoms without any specification. Hales et al.[27] defined cases of UEDs using a symptom questionnaire and physical examination. Morse et al.[28] defined UEDs as 'pain or discomfort of the hand,

Table I Characteristics of the study populations

Study	Year of data collection	Country	Study population	Number studied	Response rate	Age (years)	Females
Studies reporting point prevalence							
Fry et al. 1987	1985	Australia	Music population (7 performing music schools)	1249	-	-	55%
McCormack et al. 1990	-	USA	Textile workers	1) 2047 2) 895	91% 94%	33.0-38.1 -	75.8% -
Feldman et al. 2002	1995	Canada	High school students	502	62%	13.8 (0.1)	47.4%
Palmer et al. 2001	1997-98	Great Britain	Non-manual occupations	4889	58%	16-64	53.4%
Picavet et al. 2003	1998	The Netherlands	Open population	3664	46.9%	>25	49.6%
Katz et al. 2000	1998	USA	College students	1544	96%	-	45.5%
Studies reporting 12-months prevalence							
Dimberg et al. 1989	1983	Sweden	Engineering industry	2814	96%	>10	13.6%
Bernard et al. 1994	-	USA	Newspaper employees	973	93%	39.2 (10.5)	59.3%
Hales et al. 1994	-	USA	Tele-communication employees	518	-	37.5 (9.8)	-
Polanyi et al. 1997	1995	Canada	Newspaper employees	1007	84%	42 (9.4)	44%
Batevi et al. 1998	-	Italy	Kindergarten teachers and traffic policemen	749	-	15-35 (42%) >35 (58%)	15-35 (55.9%) >35 (60.1%)
Morse et al. 2003	1996	USA	Connecticut workers	3200	78%	Working age	-
Studies reporting lifetime prevalence							
Stockstill et al. 1993	1991	USA	Dentists	1016	98%	-	-

arm, shoulder, or neck for one continuous week or 20 days total over the previous 12 months (= chronic pain)'. The other three studies[16,29,30] also reported a specified definition of UEDs. The terms they used refer to musculoskeletal disorders located in the upper extremity. In the case definitions they specified the duration of the complaints within the last 12 months and the sensation the patients must have beside pain such as discomfort or paraesthesia. In addition, Batavi et al.[30] and Bernard et al.[29] excluded UEDs caused by an acute trauma. Bernard et al.[29] made the case definition even more specific by labeling work-relatedness of the disorder caused by the current job and the seriousness of the disorder.
Stockstill et al.[31], reporting lifetime prevalence, used the term 'upper-extremity neuropathy' and defined the conditions as 'altered sensation in hands or arms, forearms, cervical area or neck'.

Pooling data

The first requirement to enable pooling of data is the use of similar case definitions of UEDs across studies. The case definitions used in the 13 included studies, as illustrated above, differed enormously. None of the studies used the same or a similar description of UEDs. Therefore, it is not possible to pool the data and the results will be described.

Prevalence rates of UEDs

Point prevalence

Point prevalence ranged from 1.6-53.0%. The point prevalence rates of self-reported complaints in the working population and students were higher (30.0-53.0%) than the point prevalence rates acquired by physical examinations (range 9.3-26.9). The highest point prevalence rates were reported in the USA within textile workers and students, 47% and 53% respectively[25,32], although McCormack et al.[25] reported a lower point prevalence rate of 26.9% after physical examination of the positive cases according the results of the questionnaire. In the Dutch open population the lowest point prevalence rates were measured. The rate in male and females being similar (2%).[22] A prevalence rate of 30% was reported in the late 1990's in people with non-manual occupations in England.[21]

12-months prevalence

The 12-months prevalence ranged from 2.3-41,0%. Dimberg et al.[26] reported a 12-months prevalence rate (23%) of self-reported complaints in the early 1980s in aircraft engineers, consisting of 86% males. The 12-months prevalence rate of self-reported complaints in newspaper employees in the early 1990s in the USA and Canada was 41% and 19.8%, respectively.[16,29] The study population of the newspaper employees in the USA included more females than the study population in Canada (59% and 44%, respectively). Morse et al.[28] reported a 12-months prevalence rate of 11.7% in workers in Connecticut (USA), in 1996. The 12-months prevalence rate of complaints collected by using a questionnaire and a physical examination in high-risk

Table 2 Overview of terminology and prevalence rates

Country, year of da ta collect ion (reference)	Term	Definition	Measurement tool	Prevalence (%)		
				Total	Men	Women
Point prevalence						
Australia, 1985 (Fry et al. 1887)	Overuse (injury) syndrome	Those changes brought about in the muscles and joint ligaments form excessive use, causing pain, loss of function, and almost always demonstrable tenderness in the affected structures	Interview and examination (most) of the effected cases	9.3	3.2	6.1
USA, - (McCormack et al. 1990)	Upper extremity disorders	1) Current problems in the upper extremity 2) ICD.9CM used to code diagnosis. Excluded osteoarthritis, previous trauma unrelated to present employment, and rheumatic diseases	1) Questionnaire	47	-	-
			2) Physical examination of the positive cases	26.9	-	-
Canada, 1995 (Feldman et al. 2002)	Neck and upper limb pain	Having substantial neck and upper limb pain at inception	Questionnaire	31.9	-	-
Great Britain, 1997-98 (Palmer et al. 2001)	Upper limb symptoms (inclusive neck)	-	Questionnaire	30.0	-	-
The Netherlands, 1998 (Picavet et al. 2003)	RSI Repetitive strain injury	-	Questionnaire	1,9	2.0	1.9
USA, 1998 (Katz et al. 2000)	Upper extremity musculo-skeletal disorders	-	Questionnaire	53.0	21.2	31.8
12-months prevalence						
Sweden, 1983 (Dimberg et al. 1989)	NES Neck and upper extremity symptoms	-	Questionnaire	23.0	-	-
USA, 1991 (Bernard et al. 1994)	Musculo-skeletal disorders of the upper extremity	Pain, numbness, tingling, aching, stiffness, or burning in neck, shoulder, hand, or wrist and all of the following criteria applied: 1) no previous accident or sudden injury that was work-related 2) symptoms began after the current job was started 3) symptoms lasted for more than one week or occurred at least once a month within the last year 4) symptoms were reported as “moderate” or “worse” of a five-point intensity scale.	Questionnaire	41.0	-	-

USA, - (Hales et al. 1994)	Work-related UE disorders Upper extremity disorders	Pain, aching, stiffness, burning, tingling, or numbness Symptoms occurred within the past year No previous accident or trauma within the past year Symptoms began after employment within the company Symptoms occurred on the current job Symptoms lasted for more than 1 week, or occurred at least once a month Positive findings on the symptomatic joint area (criteria defined for various medical conditions)	Questionnaire and physical examination	22.0	2.8	17.2
Canada, 1995 (Polanyi et al. 1997)	Upper limb (neck, shoulder and arm) work-related musculo-skeletal disorders (WMSDs)	Those who experienced moderate, severe, or unbearable pain or discomfort either once per month or for longer than a week over the past year	Questionnaire	19.8	7.7	12.1
Italy, - (Batevi et al. 1998)	WMSDs work-related musculo-skeletal disorders of the upper limbs	'Anamnestic cases': pain or paraesthesia present for at least 1 week during the previous 12 months, or appearing at least once a month, and not subsequent to acute trauma	Anamnestic	1.9 age 15-35 7.2 age >35 9.1 total age >15	0.8 age 15-35 2.7 age >35 3.5 total ag e >15	1.1 age 15-35 4.5 age >35 5.6 total age >15
		'Clinical examination' (cases out of the positive anamnestic cases): no definition	Clinical examinations	0.5 age 15-35 3.4 age >35 3.9 total age >15	no data age 15-35 0.8 age > 35 0.8 total age >15	0.5 age >35 2.6 age >35 3.1 total age >15
USA, 1996 (Morse et al. 2003)	UEMSD Upper extremity musculo-skeletal disorders	Pain or discomfort of the hand, arm, shoulder, or neck for one continuous week or 20 days total over the previous 12 months (= chronic pain)	Telephone survey	11.7	-	-
Lifetime prevalence						
USA, 1991 (Stockstill et al. 1993)	Upper extremity neuro-pathy	Altered sensation in hands or arms, forearms, cervical area or neck	Questionnaire	29	-	-

telecommunication employees in the USA was 22%.[27] In a population in Italy that is not occupational exposed to tasks implying repetitive and/or forced movements of the upper limbs Batevi et al.[30] reported that the 'anamnestic cases' of UEDs occurred in about 2% of persons aged 15 to 35 years; in persons aged 35 years and older the prevalence rate increased to more than 7%. After clinical examination of the positive anamnestic cases, however, the prevalence rates of both age groups decreased to 0.5% and 3.4% respectively.

Lifetime prevalence

In just one study[31] the lifetime prevalence was estimated. In this study in dentists the lifetime prevalence was estimated to be 29%.

Discussion

In this systematic appraisal worldwide incidence and prevalence rates for UEDs available in scientific literature were collected. No studies were found with regard to the incidence of UEDs that met the inclusion criteria. The estimates of the prevalence rates varied enormously across the 13 included studies. The point prevalence ranged from 1.6-53% and the 12-months prevalence ranged from 2.3-41%. One study reported on the lifetime prevalence (29%). Only Picavet et al.[22] studied the prevalence in an open population. The low point prevalence they reported can not be compared with the other studies available, because they all studied a specific (working) population. In addition, Picavet et al.[22] reported on the occurrence of 'RSI', while the occurrence of an epicondylitis (around 11%) and a tendonitis or capsulitis (for the whole body they reported a prevalence rate around 16%) were reported separately and therefore not included in 'RSI'.

In this study studies were included that reported incidence and prevalence rates of the whole upper extremity. Studies, which reported incidence or prevalence rates on different regions of the upper extremity separately, but give no estimates for the whole upper extremity, were excluded. Reviews on the prevalence rates of a specific disorder or complaints in one region of the upper extremity have been reported elsewhere. For example, the estimates of the occurrence of the carpal tunnel syndrome in different occupational groups was studied by Hagbert et al.[33] and varied between 0.6 and 61%. Luime et al.[34] reported on prevalence rates of shoulder pain in the open population: the point-prevalence ranged from 7 - 27% and the 12-months prevalence ranged from 8.4 – 20%.

In general, higher prevalence rates of UEDs were found in women than in men and the estimates of self-reported complaints were higher than those acquired by using (in addition) physical examinations. No evidence of a clear increasing or decreasing pattern over time was found. Although period prevalence can be more biased then point prevalence because of incomplete response or due to recall bias[35], 'firm' conclusions can not been drawn because of the diversity of terms and definitions of UEDs used in the included studies.

To describe the conditions a variation of terms such as 'pain', 'disorders', 'complaints', 'syndrome', 'symptoms', and 'injury' are used in the literature. Because of the different meanings of the terms, it is important to give sound arguments when using certain terms. For example, it you want to describe specific and non-specific cases, using the term disorder is not very clear, because a 'disorder' indicates a specific disease, which can be diagnosed by fixed criteria. All terms used for UEDs in the included studies, except those used by Picavet et al.[22] and Fry et al.[24] indicated the location of the condition. In our opinion, it is practical and functional to use the localization of the conditions in the term.

Although the term used for UEDs is important because of the perception it causes and the clarity of the medical condition, the definition is even more important. This is not only the case for researchers when they want to compare data of different studies, but also for medical and paramedical staff, so they can speak in an unambiguous way or 'language'. This unambiguous 'language' has to make sure that physicians and other healthcare workers have in mind and speak about the same condition when they discuss the subject or, for example to evaluate the (multidisciplinary) treatment of one of their patients. The case definitions used in the included studies varied enormously. Although studies reporting prevalence rates for UEDs were not included and this appraisal was limited to studies which included 500 cases or more and studies of which the data were published in scientific literature, the diversity of case definitions and classification of UEDs that was found was substantial. This is a general problem and reported in literature by many authors before.[36-39]

The diversity in terms and case definitions of EUDs in the included 13 studies prevented any meaningful pooling of data. Drawing comparisons between countries, different working population and assessment of changes in time within a population or country could therefore not be carried out in a quantitative manner.

Different questionnaires and tests used for the physical examinations were presented in the studies; little was said about the validity and reliability of the measurement tools. Developing criteria for classification or diagnosis would be easy if gold-standard diagnostic tests would be available. Unfortunately, no criterion standard for any of the upper extremity soft tissue musculoskeletal conditions is available.[37]

If we want to make progress in this field, the first requirement is to agree on unambiguous terminology and classification of EUDs. Physicians and other healthcare workers dealing with patients with these conditions should be involved in such a project. Studies of classification criteria suggest that expert clinicians can more accurately identify cases than most history, physical examination, or laboratory parameters.[40] Furthermore, involving all key disciplines dealing with patients with UEDs will make implementation of the results more successful. Therefore, a multidisciplinary project on national or international level in which all key disciplines cooperate with the intention to achieve multidisciplinary consensus on terminology and classification of UEDs is recommended. When they have agreed about an 'unambiguous'

language, the next step is to achieve consensus about valid diagnostic criteria for UEDs and to study the best (multidisciplinary) prevention and/or treatment.

Conclusions

No studies were found with regard to incidence rates of UEDs and there are substantial differences in reported prevalence rates on UEDs. One of the main reasons for this is the absence of a universally accepted way of labelling or defining UEDs. Health professionals and policy makers should be aware of this problem when they estimate the occurrence of the conditions in populations and the necessary demand and related costs for health care.

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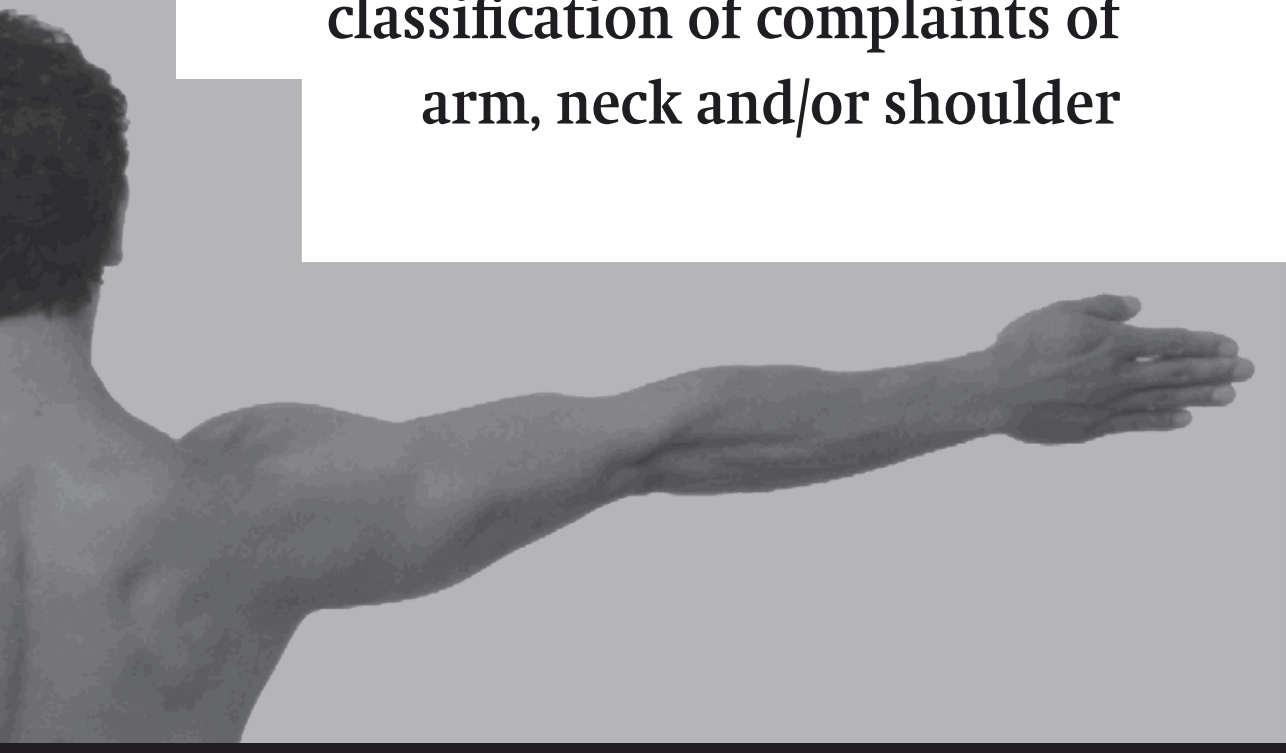
Chapter 3

Multidisciplinary consensus on terminology and classification of complaints of arm, neck and/or shoulder



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Abstract

Background: There is no universally accepted way of labelling or defining upper-extremity musculoskeletal disorders. A variety of names are used and many different classification systems have been introduced. The aim of this study was to agree on an 'unambiguous language' concerning the terminology and classification that can be used by all relevant medical and paramedical disciplines in the Netherlands.

Methods: A Delphi consensus strategy was initiated. The outcomes of a multidisciplinary conference were used as a starting point. In total, 47 experts in the field of upper-extremity musculoskeletal disorders were delegated by 11 medical and paramedical professional associations to form the expert-panel for the Delphi consensus strategy. Each Delphi round consisted of a questionnaire, an analysis and a feedback report.

Results: After three Delphi rounds, consensus was achieved. The experts reported the consensus in a model. This so-called CANS model describes term, definition and classification of complaints of arm, neck and/or shoulder (CANS) and helps professionals to classify patients unambiguously. CANS is defined as "Musculoskeletal complaints of arm, neck and/or shoulder not caused by acute trauma or by any systemic disease". The experts classified 23 disorders as specific CANS, because they were judged as diagnosable disorders. All other complaints were called non-specific CANS. In addition, the experts defined 'alert symptoms' on the top of the model.

Conclusions: The use of the CANS model can increase accurate and meaningful communication amongst healthcare workers, and may also have a positive influence on the quality of scientific research, by enabling comparison of data of different studies.

Introduction

Multidisciplinary consensus on terminology and classification of upper-extremity musculoskeletal disorders is a first requirement for accurate and meaningful communication amongst clinicians. Universal classification of these conditions of the upper limb and neck is necessary to assess prognosis and options for treatment[1,2], to study the natural course of the conditions and to compare research findings across geographic regions and time periods within different (working) populations.
In a systematic appraisal of worldwide prevalence rates[3], substantial differences in reported prevalence rates of upper-extremity disorders were found. Point prevalence estimates ranged from 1.6-53% and the 12-months prevalence estimates ranged from 2.3-41%. It was concluded that one of the main reasons for the differences found in this latter study is the absence of a universally accepted taxonomy for upper-extremity musculoskeletal disorders.

A variety of terms for upper-extremity musculoskeletal disorders are used in different countries all over the world, including repetitive strain injury (RSI), upper-extremity cumulative trauma disorder (UECTD) and work-related upper-limb disorder (WRULD). Many different classification systems have been introduced. Van Eerd et al.[4] found 27 different classification systems for the working population. The systems differed in the disorders they included, the labels used to identify the disorders, and the criteria used to describe the disorders.
Two sets of consensus criteria for upper-extremity disorders were recently proposed in the United Kingdom[5] and in Europe[6]. Both Harrington et al.[5] and Sluiter et al.[6] gave criteria for a limited number of upper-extremity disorders only. Despite their efforts, implementation of these criteria would have been easier if the experts, chosen by the researchers in both studies, would have been key persons chosen by representatives of the persons who have to work with the criteria in practice.
Until now, none of the proposed classification systems have resulted in a complete overview in which (in principal) all musculoskeletal upper-extremity disorders are evaluated and discussed for inclusion. Moreover, they did not produce a workable classification tool that can be used in daily practice in an easy way (i.e. no special training and/or no substantial time needed to perform) by both researchers and health professionals.

Therefore, we concluded that there is a need for a classification system on musculoskeletal upper-extremity disorders that 1) could be generally accepted and used by all disciplines, 2) can support the diagnosis and classification of (in principal) all upper-extremity conditions and, 3) is reported as a practical tool.

Our first aim is to achieve consensus in the Netherlands, with a further intention to use the results of this study to eventually achieve international consensus. The decisions made

Table 1 Participating disciplines

PARTICIPATING DISCIPLINES
On behalf of the professional associations
general practitioners
physical and rehabilitation medicine specialists
occupational physicians
orthopaedic surgeons
rheumatologists
neurologists
physical therapists
exercise therapists Cesar
exercise therapists Mensendieck
occupational therapists
psychologists

regarding classification were of course based on the international literature. To make implementation of the results of the project more feasible, we invited 11 medical and paramedical associations to assign delegates to participate in this consensus project (Table 1).
An unambiguous classification system that is accepted by all professionals involved may increase multidisciplinary cooperation and have a positive influence on the performance of studies and also allow data to be compared. This paper presents the results of the Delphi consensus strategy used to achieve consensus and the resulting model.

Methods

The staff team

The staff team initiated and executed the Delphi consensus strategy. All three staff team members have an epidemiological as well as a clinical background. The epidemiologist/physician, the occupational health physician/psychologist and the health scientist/physiotherapist were responsible for the construction of the questionnaires, the analysis of the responses and the formulation of feedback. The staff team first initiated an invitational conference; the outcomes of this conference were used for the design of the first questionnaire of the Delphi consensus strategy.

Invitational conference

A multidisciplinary invitational conference (December 2002) was the starting point of the project. A total of 19 representatives of 10 of the 11 different medical and paramedical professional associations concerned with treatment of patients with upper-extremity disorders were present. Only one psychologist representing one national association was lacking. Structured group communication techniques were used at the conference to exchange ideas

and expertise on the subject. The outcomes of the conference were used for further research to achieve the consensus.

Terminology

In the Netherlands the term 'RSI' is often used for symptoms of the arm or neck without a clear diagnosis. However, more than 90% of the participants of the conference were of the opinion that 'RSI' is an unclear and confusing name for these ailments. During the conference, the staff team offered the participants a list of 14 Dutch and English terms used for upper-extremity musculoskeletal disorders that are frequently used in scientific literature and medical textbooks. The participants selected seven terms from this list and added one other term to it. The resulting eight terms were proposed in the Delphi-I questionnaire.

Definition

During the invitational conference, it became clear that the participants gave priority to a general and broad definition of upper-extremity disorders rather than a narrowly described definition. It should include 'complaints of pain', 'localized in the arm, neck and/or shoulder' and 'no trauma involved'. Possibly 'no systemic disease involved' could be included; 'related to the musculoskeletal system' could be added to indicate that only musculoskeletal disorders should be considered. The participants chose not to mention the suspected aetiology of complaints in the definition.

Classification and model

Complaints meeting the general definition should be divided into diagnosable and non-diagnosable disorders. A diagnosable disorder should be defined as one with discernible characteristics, which can be diagnosed in a reproducible way. The diagnosis can be made through case history, physical examination, imaging and laboratory testing. It is important to realise that when a disorder is diagnosable, it does not necessarily mean that treatment is available.

During the conference, two models were initially discussed for the classification of patients (Figure 1a and 1b). In model 1a the diagnosable and non-diagnosable disorders are two defined groups. Model 1b is largely similar to model 1a; however, the group 'diagnosable disorders' was subdivided into separate disorders, which have to be mentioned and approached individually. The staff team decided to present both models in the Delphi-I questionnaire.

Delphi consensus strategy

Of all consensus techniques available, we chose the Delphi consensus strategy as our preferred method. In this method an expert-panel is asked to answer questions concerning the subject. Then, through repeated feedback of the answers in several rounds involving all participants, the researchers try to develop consensus on opinions.[7] The advantage of this method is that

it is a written, anonymous method[8] in which the opinions of the experts are combined whilst bias through institutional role, status, or dominant personality is avoided.[9]

Selection of participants

The boards of the 11 relevant medical and paramedical associations in the Netherlands were asked to delegate a maximum of six experts each in the field of upper-extremity disorders, who were willing to participate in the expert panel.

Procedure

In the questionnaires of each Delphi round we asked questions about term, definition and classification of complaints of arm, neck and/or shoulder. We used structured questions with the answer formats 'agree/don't agree/don't know' or 'yes/no/don't know'. For classification of the different complaints, the possible answers were diagnosable/non-diagnosable/no opinion. We invited the expert-panel to give an explanation for their choices. After each round a feedback report was made to inform the expert-panel about the answers and argumentations of the other experts. On the basis of the answers and arguments of the experts, the staff team decided which questions would appear in the next questionnaire. Staff team decisions were presented and justified in the feedback report.

Delphi-I questionnaire

The Delphi-I questionnaire was constructed using the outcomes of the invitational conference. The questionnaire of Delphi-I consisted of two parts. Part A contained questions concerning items for which 70% or more participants of the conference agreed on, and part B concentrated on the conflicting items. Items which were only discussed in small groups and not plenary, were also included in part B. Separately, one question was included about the cut-off point for consensus concerning the whole Delphi survey.

Delphi-II and III questionnaires

The questionnaires of Delphi-II and Delphi-III were constructed using the results of Delphi-I and Delphi-II, respectively. The remarks of the expert panel were incorporated in the questionnaire of the next round. In this way we collected and reported the opinions of the expert panel in each round in order to achieve consensus.

Analysis

The analysis of the responses from the Delphi rounds was both qualitative and quantitative. Qualitatively, two staff members independently analysed the answers of the expert-panel; they compared the results of their analysis. Quantitatively, we reported for each question on how many participants gave which answers. Also, percentages were given of the positive and negative answers.

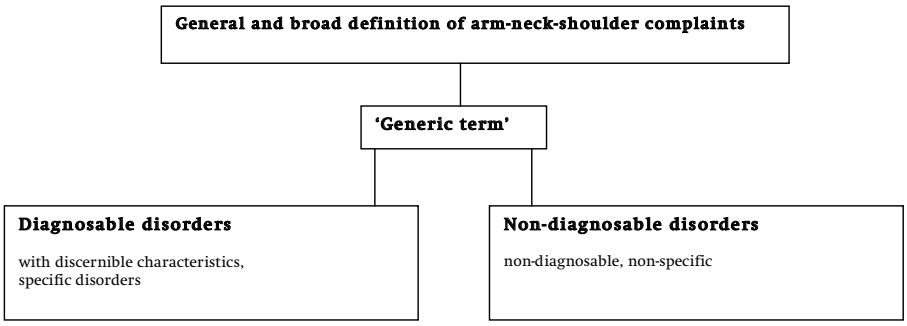


Figure 1a Model 1a

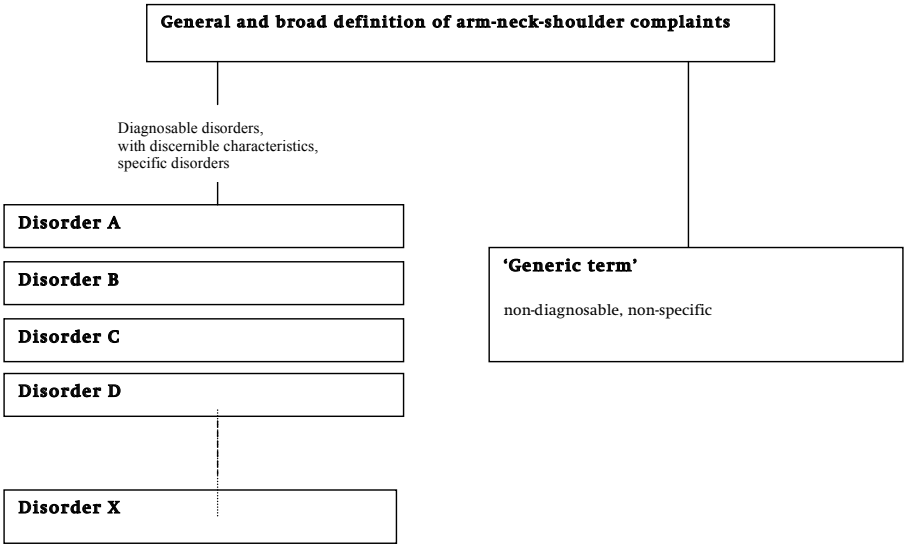


Figure 1b Model 1b

Results

Expert-panel

From January till March 2003, the 11 medical and paramedical professional associations selected 47 experts to form the expert-panel for the Delphi consensus strategy. Experts from all 11 disciplines participated in the survey as delegates for their respective professional associations. Three experts, all Mensendieck exercise therapists, ended their participation during the process. Two of them only returned the Delphi-I questionnaire, the third did not return any of the questionnaires. Of the 46 experts, 44 (96%) returned the Delphi-I questionnaire; 40

Table 2 Classification of complaints

	S	N-S	Ex	
Neck region				
Cervical disc hernia	x			
Radiating neck complaints		x		
Tension neck syndrome		x		
Shoulder region				
Frozen shoulder	x			
Instability of the shoulder	x			
Labral lesion of the glenoid	x			
Rotator cuff tears	x			
Bursitis of the shoulder				They can only be discerned as a group. Consensus achieved about the term 'subacromial impingement syndrome' for these disorders and classified as specific.
Rotator cuff syndrome				
Tendinitis of the m.infraspinatus	x			
Tendinitis of the m.subscapularis				
Tendinitis of the m.supraspinatus				
Suprascapular nerve compression	x			
Elbow region				
Bursitis of the elbow	x			
Instability of the elbow	x			
Lateral epicondylitis	x			
Medial epicondylitis	x			
Tendinitis of the biceps tendon	x			
Forearm, wrist and hand region				
Carpal tunnel syndrome	x			
Cubital tunnel syndrome	x			
De Quervain's disease	x			
Dupuytren disease	x			
Guyon canal syndrome	x			
Hand-arm vibration syndrome		x		No consensus about classification. Therefore, non-specific CANS
Oarsman's wrist	x			
Radial tunnel syndrome	x			
Raynaud's phenomenon	x			
Tendinitis of the wrist/forearm			x	Mention the specific tendon involved
Trigger finger	x			
Not specific one region				
Bechterew disease			x	Rheumatic disease, added as 'alert symptom' in the CANS model
Complex regional pain syndrome	x			
Fibromyalgia			x	Rheumatic disease, added as 'alert symptom' in the CANS model
Local arthritis (not RA) in a joint of upper extremity	x			
Lung tumor			x	No musculoskeletal disorder, added as 'alert symptom' in the CANS model
Osteoarthritis			x	Rheumatic disease, added as 'alert symptom' in the CANS model
Rheumatoid arthritis			x	Rheumatic disease, added as 'alert symptom' in the CANS model
Thoracic outlet syndrome		x		No consensus about classification. Therefore, non-specific CANS

S: specific

N-S: non-specific

Ex: excluded from CANS

(87%) and 43 (93%) returned the Delphi-II and Delphi-III questionnaires, respectively. The most common reason for non-response was 'lack of time'. The final results of the Delphi consensus strategy, - that is, the consensus model - were presented in October 2004.

Cut-off point for consensus

In the Delphi-I questionnaire a cut-off point of 70% agreement was accepted: Consensus was reached when 70% or more of the experts gave the same answer to a question.

Term

In Delphi-I, it became clear that almost all experts (93%) gave priority to dispose of the term RSI. Although the term RSI has played an important role in recognising the extent of the problem, the term has led to negative associations concerning patients dealing with these problems. It was considered to be an umbrella term. Furthermore, the term was judged unclear and confusing: an injury is not always involved, and besides 'repetitive strain', also 'static burden' may generate the complaints.

In the Delphi-I questionnaire the expert-panel was asked to rank the eight terms on the list composed of the outcomes of the invitational conference and to place their preferred name on the top. In this round they could also bring up other (new) terms. The staff team decided that the five terms, which scored 70% of the votes in Delphi-I, complemented with another term given by one of the experts, would be used for the Delphi-II questionnaire. This list involved three English and three Dutch terms. In Delphi-II the expert-panel was asked to divide six points among both the English and the Dutch terms, separately. Elsewhere in Delphi-II the experts were asked whether an English or a Dutch term should be used. In Delphi-II, consensus was reached about bringing into use an English term: **CANS - Complaints of Arm, Neck and/or Shoulder.**

Definition

In Delphi-I, the experts agreed to bring into use a general and broad definition. During Delphi-I and Delphi-II, all of the items mentioned in the conference were adopted, with a few minor changes. The expert-panel decided to change 'complaints of pain' into 'complaints' because pain and also other sensations, such as tingling, can be involved.

Traumata such as fractures and ruptures needed to be excluded from the definition. However, micro-traumata can be involved in CANS. Therefore, the word 'acute' was added to the element concerning the presence of traumata and became 'no acute trauma involved'.

In conclusion, 'complaints', 'localised in the arm, neck and/or shoulder', 'no acute trauma involved', 'no systematic disease involved' and 'related to the musculoskeletal system' were included in the definition. In Delphi-III, consensus was achieved on the following definition of CANS: **"Musculoskeletal complaints of arm, neck and/or shoulder not caused by acute trauma or by any systemic disease".**

Classification and model

Number of disorders classified

The staff team constructed a list of 29 disorders of the upper extremity based on textbooks and scientific literature. The experts added eight other disorders during Delphi-I. In total, the expert-panel discussed 37 diagnoses that met the definition of CANS and classified these as diagnosable or non-diagnosable. During this process, six disorders were excluded from this list for various reasons (Table 2). Finally, 23 disorders were classified as diagnosable and four as non-diagnosable.

Diagnoses excluded from classification

In Delphi-III, the experts decided to exclude the diagnosis 'tendonitis of the wrist/forearm' from the list; this term was considered too general and specific disorders, such as De Quervain's disease, were already part of the list. The experts also decided to exclude the diagnoses 'lung tumour' and 'cardiac diseases'. Although these diseases can cause problems in the upper extremity, they are not related to the musculoskeletal system. Because the experts achieved consensus on excluding systemic diseases from CANS, they decided to delete rheumatic diseases from the list after Delphi-II. Although osteoarthritis is not a systemic disease, it was included within rheumatic diseases.

Shoulder complaints

In Delphi-I, a well-known clinical problem concerning musculoskeletal disorders of the shoulder, such as tendonitis and bursitis, emerged; they are difficult to differentiate but can be identified as a group. Therefore, some of the experts pleaded for the introduction of a generic term for these disorders, so that they can be classified as diagnosable. This idea was presented and adopted in Delphi-II. In Delphi-III, consensus was achieved to use the term 'subacromial impingement syndrome' for the disorder that includes the rotator cuff syndrome, tendonitis of the m.infraspinatus, m.supraspinatus and m.subscapularis, and bursitis in the shoulder area.

Non-diagnosable disorders

In Delphi-II consensus was achieved on the classification of the 'tension neck syndrome' and 'radiating neck complaints' (or 'radiculopathy without a herniated disc') as non-diagnosable.

In Delphi-III the experts decided that disorders for which no consensus about classification was achieved during the three Delphi rounds would be classified as non-diagnosable, until more information becomes available about diagnostic criteria for the disorder. This was the case for the 'thoracic outlet syndrome' and the 'hand-arm vibration syndrome'.

Alert Symptoms

It is generally known that a physician has to be aware of so-called 'alert symptoms' while making a diagnosis. For example, symptoms may appear to be a result of complaints in the upper extremity, but are in fact caused by serious conditions such as angina pectoris. Diseases such as rheumatoid arthritis and osteoarthritis also need to be identified. To make sure that the symptoms of these disorders get the attention they need, the expert-panel decided in Delphi-II to add 'alert symptoms' at the top of the final model.

The CANS model and the flow chart

In Delphi-I, consensus was achieved to use model 1b (Figure 1b) for the classification of patients. In Delphi-III the experts achieved consensus to use the terms **'specific CANS' and 'non-specific CANS'** instead of 'diagnosable CANS' and 'non-diagnosable CANS'. The whole model will be called **the CANS model**. A flow chart has been developed to help the doctor or paramedical therapist to classify the patient using the CANS model (Figure 2). When complaints meet the definition of CANS, the clinician has to investigate whether or not one of the 23 disorders mentioned as specific CANS is present. If present, the diagnosis will be mentioned by its specific label, such as 'carpal tunnel syndrome' or 'lateral epicondylitis'. If not present, the complaints will be diagnosed as 'non-specific CANS'.

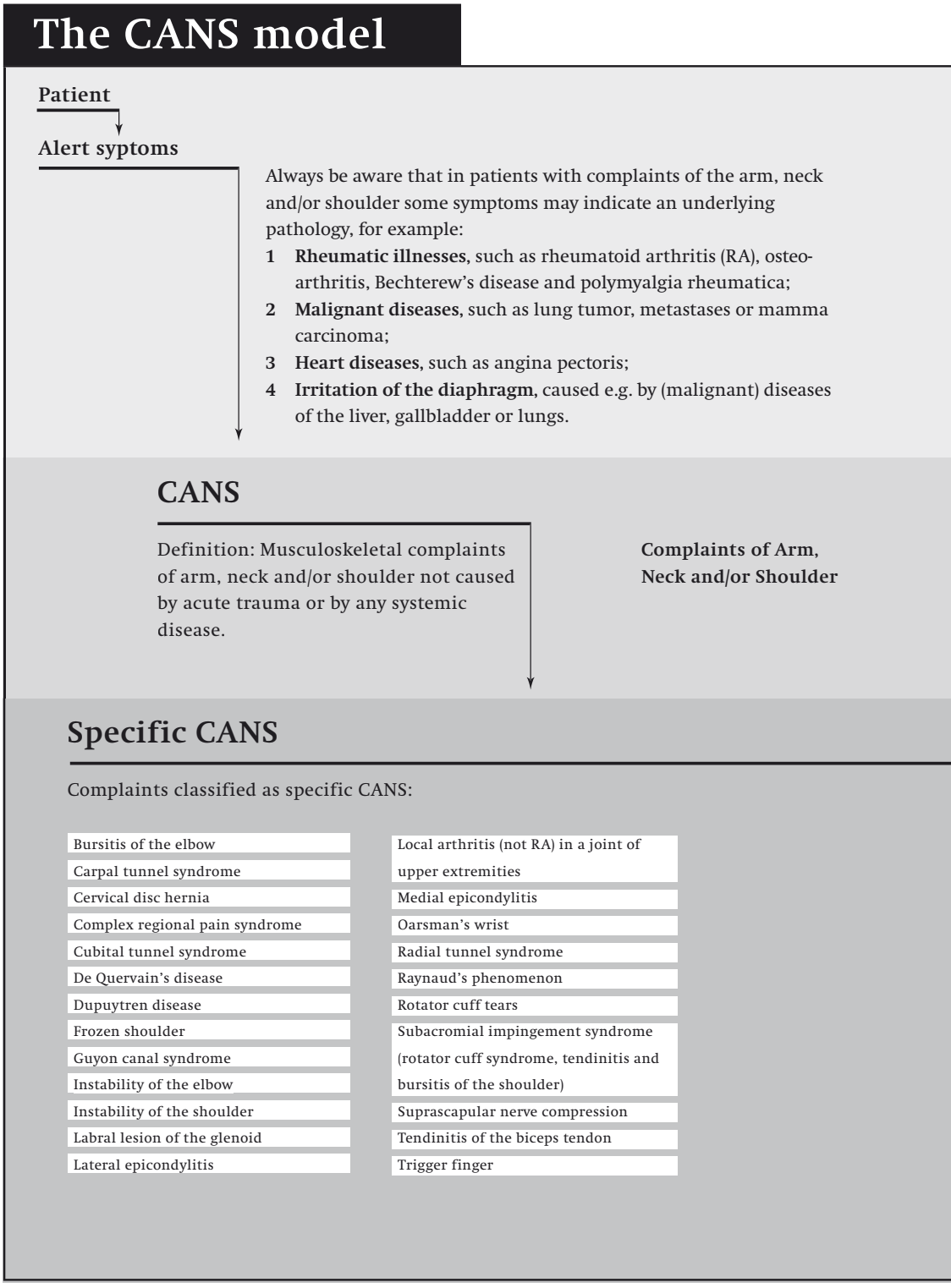
Discussion

The aim of the Delphi consensus strategy was to decide on an 'unambiguous language' concerning the terminology and classification of complaints of the arm, neck and/or shoulder for all relevant medical and paramedical disciplines in the Netherlands involved in the treatment of patients with these complaints. After three Delphi rounds, multidisciplinary consensus was achieved and reported in the CANS model. As far as we know, this is the first time a multidisciplinary classification system on a national level has been developed in which all relevant medical and paramedical professions dealing with the treatment of patients with CANS were involved and in which (in principal) all musculoskeletal upper-extremity disorders were evaluated and discussed for inclusion.

Williams and Webb[10] observed weaknesses in the Delphi consensus strategies, including 1) limited descriptions of experts' characteristics, 2) imprecise definitions for consensus and, 3) low response rates.

In a consensus procedure, there is a risk of bias in the selection of participants. In the present Delphi consensus strategy, 11 medical and paramedical associations selected the expert-panel. In this way, the expert-panel consisted of professionals with various medical and paramedical backgrounds, all seen as experts on upper-extremity disorders within their

Figure 2 The CANS model



Flow chart

1

Step 1. Are alert symptoms involved?
The complaints of the arm, neck and/or shoulder can be caused by the symptoms of an underlying (malignant) pathology.
When there is an alert symptom, then this has to be treated. If there is not, go to step 2.

2

Step 2. Is there a possibility of an acute trauma or systemic disease?
If so, one does NOT call it CANS. Otherwise, it does concern CANS and go to step 3.

3

Step 3. Is there a possibility of specific CANS?
You can find the 23 complaints designated as specific CANS in the model. These disorders will be mentioned by their specific name.
Treat these complaints as is usual for your profession.

4

Step 4. Is there a possibility of non-specific CANS?
If the complaints cannot be diagnosed as one of the disorders mentioned in the list of specific CANS, one calls these complaints 'non-specific CANS'. Treat these complaints as is usual for your profession.

Non-specific CANS

Complaints that are NOT one of the disorders classified as specific CANS.

own discipline. In a decision-making group heterogeneity can lead to a better performance than homogeneity in terms of considering all relevant aspects of the topic.[11] Furthermore, it has been shown that doctors willing to participate in an expert-panel are representative for their colleagues.[12]
To avoid an imprecise definition for consensus, the experts discussed the cut-off point for consensus and decided in Delphi-I that consensus would be defined as 70% or more agreement.
To maintain rigour when using the Delphi method, a 70% minimum response rate should be achieved.[13] We were privileged with high response rates in all three Delphi rounds; an average of 92% (range 87-96%) of the participants returned the questionnaires.

The experts achieved consensus about excluding systemic diseases, such as rheumatic diseases, from CANS and decided to add them as 'alert symptoms' on the top of the model. Although osteoarthritis is not a systemic disease, it was included within the group of rheumatic diseases.
Local arthritis (not rheumatoid arthritis) in a joint of the upper extremity is classified as one of the 23 specific disorders. An inflammation of the AC joint is an example of such a local arthritis. In the Delphi consensus strategy the experts did not discuss 'local osteoarthritis in a joint of the upper extremity'. A joint can degenerate as a result of overuse, such as osteoarthritis of the AC joint as a result of sports such as tennis or swimming. We cannot change the results of the consensus, but we see the absence of this specific disorder as a limitation of our model.

One of the oldest classifications systems used is the ICD. The ICD is used in many countries for general epidemiological and many health-management purposes. It is used to classify diseases and other health problems recorded on many types of health and vital records, including death certificates and hospital records. Buchbinder et al.[14] studied the ICD-9 for soft-tissue disorders of the neck and upper limb; they examined the overall accuracy of identifying soft-tissue disorders of these conditions and studied whether the codes themselves, on an individual basis, accurately reflected the underlying problems as documented in the medical records. They found poor agreement between the diagnostic labels recorded in the medical records and the ICD codes, suggesting that many of the terms are being used interchangeably.
To date, the 'RSI' report by the Health Council of the Netherlands[15] and the so-called SALTSA report 'Criteria document for evaluating the work-relatedness of upper-extremity musculoskeletal disorder'[6] were considered the state-of-the-art in the Netherlands. Many professional organisations and researchers used these reports as a starting point to develop their own terminology and classification system. This way, over and over again, new terms and classification systems have been generated; this problem occurs not only in the Neth-

erlands but also in other countries. Use of the CANS model can help solve this problem, but we realise that different implementation projects and strategies will be needed before all professionals accept the model. We have already launched projects to implement the CANS model in daily practice. A national conference on upper-extremity musculoskeletal disorders was organised for researchers, clinicians and paramedical health professionals in which the CANS model was revealed. The results of our study were also presented at other congresses and meetings. The Dutch media were very interested in our work; they published on CANS and reported that consensus was achieved. Nowadays, the CANS model is taught in the professional training and retraining of healthcare professionals. However, despite all our efforts to implement the CANS model, and the fact that the CANS model is already used in practice by many professionals, more time and more projects are needed before the model is fully accepted in the Netherlands.

The factor 'work-relatedness' is not mentioned in the CANS model. Ergonomic workloads such as repetitive and forceful motion, work organisational factors and psychosocial work factors have definitely been implied as a cause of CANS. Currently, many experts are of the opinion that a single common pathway that links exposure in the workplace resulting in CANS cannot be identified.[16] Work-relatedness is not a decision-making factor for including or excluding patients in the CANS model. The model does more justice to reality, as activities at work as well as activities in daily living, such as housekeeping, sports, hobbies and stress at home, can influence the complaints.

Although few data are available on the validity and repeatability of the diagnostic tests of upper-extremity disorders[5], the expert panel of the Delphi consensus strategy achieved consensus to label 23 diagnoses as specific CANS. We did not develop consensus on the diagnostic criteria for these disorders, because the aim of this project was to agree on an 'unambiguous language'. However, the results of this study are just a starting point for the use of consensus terminology. The CANS model should be re-evaluated after testing it in clinical practice. Moreover, further development of consensus regarding the diagnostic criteria of all specific disorders is needed; this will make the CANS model even more practical.

Because the criteria specified for diagnoses of specific disorders vary among different classification systems[4], we recommend (inter)national multidisciplinary cooperation to describe these criteria in which key persons - researchers and paramedical and medical professionals - cooperate.

Conclusion

The participants in this Delphi survey achieved multidisciplinary consensus on the terminology and classification of complaints of the arm, neck and/or shoulder, and reported their result in the CANS model. Adoption of this model can be the first step towards an unambiguous, multidisciplinary accepted classification system for these conditions. Studies on diagnostic criteria and validation studies for both the classification system and the diagnostic criteria are needed to further refine this work.

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Selection participants Delphi consensus strategy: Dutch College of General Practitioners, Dutch Orthopaedic Society, Royal Dutch Society for Physical Therapy, The Netherlands Society of Occupational Medicine, The Netherlands Society of Physical and Rehabilitation Medicine, The Netherlands Society of Neurology, Dutch Society of Exercise therapists Cesar and Mensendieck, Dutch Society for Rheumatology, Dutch Association of Occupational Therapy and the Dutch Professional Association of Psychologists.

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Their participation in this project does not necessarily mean that they fully agree with the final achieved consensus. The CANS model is the result of a 'communis opinio'.

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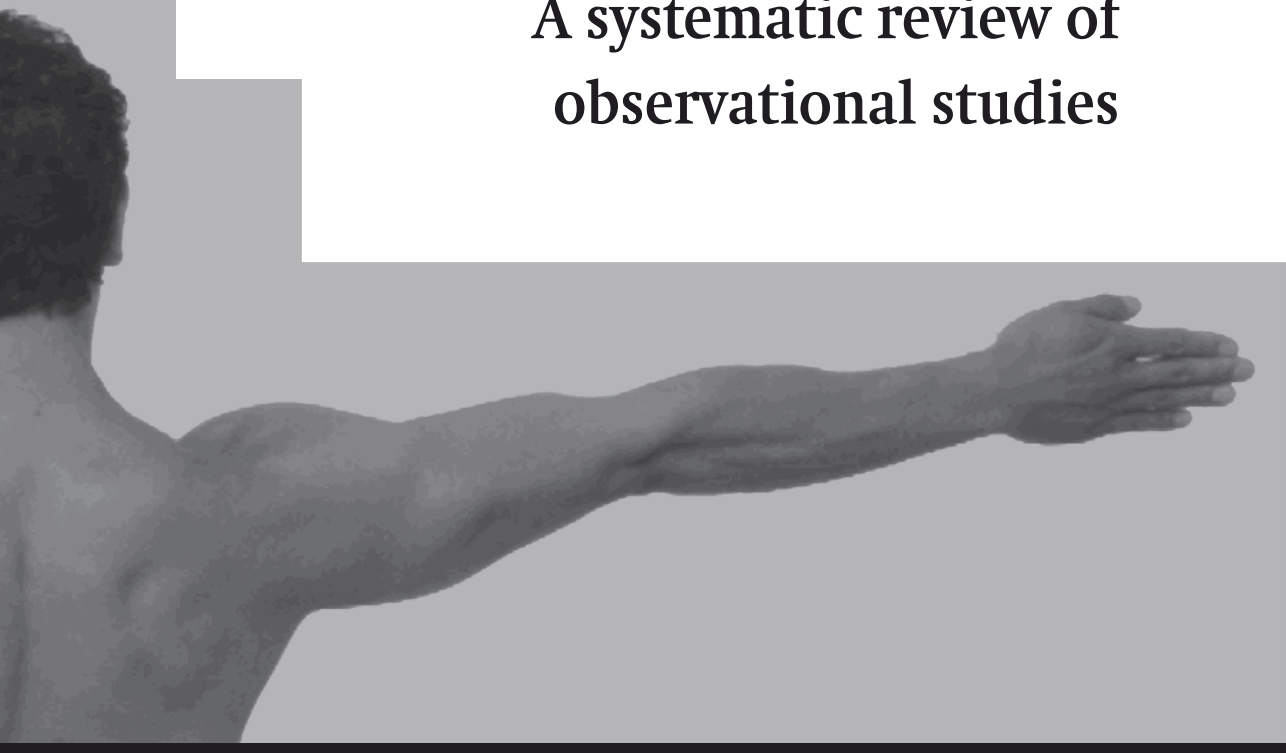
Chapter 4

Interventions for treating the radial tunnel syndrome. A systematic review of observational studies



Huisstede BM, Miedema HS, van Opstal T, de Ronde MT, Kuiper JI, Verhaar JA, Koes BW

Submitted

Abstract

Background: For some disorders, such as the radial tunnel syndrome (RTS), no randomized controlled trials or controlled clinical trails are available. To gain insight in the effectiveness of conservative and surgical interventions for treating RTS, we systematically reviewed all available observational studies on treatment of RTS. Although the validity of case series is inferior to controlled trials, they may provide valuable data about the efficacy of treatment options.

Methods: A literature search and additional reference checking resulted in 21 eligible case series for this review. Based on previous checklists we constructed a new quality assessment and rating system to analyse the included case series. The methodological quality was assessed, and data extraction was performed. Studies with less than 50% of the maximum points on the methodological quality assessment were considered inadequate and excluded from the analysis. To summarise the results according to the rating system for the strength of the scientific evidence, we introduced four levels: 1) tendency; 2) slight tendency; 3) conflicting tendency; and 4) no tendency.

Results: After the methodological quality assessment, six higher quality studies were included in the final analysis. They all reported on surgical decompression of the PIN.

Conclusions: There is a strong tendency that surgical decompression of the posterior interosseus nerve may be effective in patients with RTS. The effectiveness of conservative treatments for RTS is unknown because, for most treatments, no studies were available. Additional high-quality controlled studies are needed to assess the level of 'conclusive evidence' for surgical treatment, and also to evaluate conservative treatments for RTS. For this, we recommend a multi-center randomised clinical trial. Due to the lack of a clear protocol for diagnosing RTS, a reliable and valid diagnostic tool needs to be developed.

Introduction

For some disorders, such as the radial tunnel syndrome (RTS), no randomized controlled trials (RCTs) and controlled clinical trials (CCTs) about the effectiveness of interventions can be indentified using the criteria published by Dickersin et al.[1] Only case series of the effectiveness for treating the RTS are available. Although the validity of case series is inferior to controlled trials, they may give valuable tendencies for the efficacy of treatment options.
RTS is one of the syndromes associated with the radial nerve in the forearm. Jalovaara[2] reported an approximately 3.5% frequency of RTS as the dominating etiopathogenetic element in tennis elbow.

Proximal to the supinator arch the radial nerve divides into a superficial branch, the sensory superficial radial nerve, and a deep motor branch, the posterior interosseus nerve (PIN).[3] The PIN passes the radial tunnel. In this area of the forearm, the radial nerve is associated with two different syndromes: radial tunnel syndrome (RTS) and posterior interosseus nerve syndrome (PINS).[4,5] In the literature, a clear distinction is not always made between these two syndromes. Some clinical findings can be found in both syndromes: pain in the forearm and marked weakness of extensors of fingers or wrist. It is generally accepted that PINS is caused by continuous or intermittent compression of the PIN in the radial tunnel.[6] However, there are contradictory ideas about the cause of RTS. Some attribute the RTS to compression of the PIN in the radial tunnel or consider that the syndrome results from intermittent and dynamic compression of the nerve in the proximal part of the forearm associated with repeated pronation and supination[7], whereas others reject these hypothesis.[8,9] Little is know about the natural history of RTS.

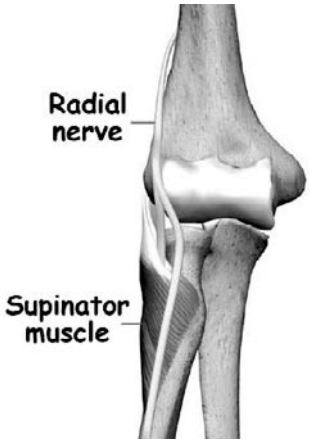


Figure 1 The radial tunnel
(www.muziekenzorg.nl)

The interventions such as release of the superficial part of the supinator muscle, carried out for both syndromes overlap to a large extent. However, since the aetiology might be different, we decided to look at the syndromes separately. We used the following descriptions of the syndromes:
The clinical presentation of **PINS** is characterized by the loss of motor function or even complete palsy of one or more muscles innervated by the PIN. The patient with PINS may have pain, but this is not the main symptom.
The hallmark of **RTS** is pain over the radial proximal forearm with little or no motor weakness. Use of the upper extremity aggravates the pain. Nocturnal symptoms, which awaken the patient, are often present. Pain at the lateral epicondyle is also common, making it difficult to distinguish RTS from a 'tennis elbow'. Motor weakness, if present, can be explained as a result of the pain. Tenderness over the radial tunnel is an important criterion in establishing the diagnosis RTS.[4] Pain during resisted middle finger extension and during resisted forearm supination with a fully extended elbow can also be found in patients with RTS. EMG findings are either absent or inadequate. Therapeutic interventions for patients with RTS include surgical decompression of the PIN, physiotherapy, steroid injection and immobilisation.

Seperately, we wrote a systematic review about the effectiveness of interventions of the PINS.[10]
In the present review we systematically reviewed the available observational studies on the effectiveness of conservative and surgical interventions for treating the RTS.

Materials and Methods

Literature Search

To identify relevant publications Medline (1966 to April 2004), Embase (1980 to April 2004), the Cochrane Library (1993 to April 2004), Pedro (up to April 2004), Cinahl (1982 to April 2004) and CENTRAL (up to April 2004) was searched. All the keywords related to the treatment of RTS were included, such as: "radial tunnel syndrome", "supinator syndrome", "posterior interosseous nerve syndrome", "PINS", "RTS", "radial nerve compression", "treatment", "therapy", "surgical" and "conservative". The complete search strategy is available on request. One reviewer (MTMR) executed the citation tracking.

A study was included if it met all of the following criteria: (1) an intervention for treating the RTS was included; (2) the study population consisted of patients aged 18 years or older diagnosed with RTS in one or both arms; (3) pain over the radial proximal forearm is described as a hallmark; and (4) at least 5 patients were included in the study. Only English, German, French and Dutch articles were considered.

Studies were excluded if the study population concerned patients with an acute trauma, poly-neuropathies, RTS as secondary consequence of diseases e.g. rheumatic syndromes and diabetes, tumors, neurological diseases and hereditary neuropathy. Studies in which part of the study population met our criteria were included if the results for this subpopulation were presented separately.

Study selection

Two reviewers (BMAH and MTMR) independently applied the inclusion criteria to select potential relevant studies from the title, abstracts, and keywords of the references retrieved by the literature search. A consensus method was used to solve disagreements concerning inclusion of studies and a third reviewer (HSM) was consulted if disagreement persisted.

Assessment of methodological quality

Because the current quality assessments are specifically developed for RCTs, a new quality assessment list for the included case series was constructed (Table 1. Appendix 1). shows the operationalisation of the methodological quality assessment. The criteria were adapted from van Tulder et al.[11], Lievense et al.[12] and Borghouts et al.[13] and modified to cover the topic of this review. The list (19 items) consists of five topics: study population, interventions, study design, outcome measurements, and analysis.

Two reviewers (BMAH and TvO) independently scored the quality of each study. A consensus method was used to solve disagreements and a third reviewer (HSM) was consulted if disagreements persisted. Each item was scored as positive ("+"), negative ("-") or unclear ("?") using the operationalisation of the criteria list for the methodological quality assessment of observational studies (Appendix 1). Each quality item was given one point when the reviewer scored a "+".

Data extraction

Two reviewers (BMAH and TvO) independently collected data on the study population, interventions used, study design, outcome measurements, and data analysis.

Data analysis

We used a rating system to analyse the case series on methodological quality. Because no RCTs or CCT s were found, the conclusions of the present review cannot be seen as 'conclusive evidence' for the efficacy of treatments. Therefore, we used the term 'tendency' to emphasise that case series cannot supply strong evidence for or against the efficacy of interventions. Studies with less than 50% of the points on the methodological quality assessment were considered inadequate and excluded from the final analysis. The quality of a case serie was considered to be high when the score on the methodological assessment was 50% or more. To summarise the results according to the rating system for the strength of the scientific

evidence, we introduced four levels: 1) Tendency: providing that generally consistent findings in multiple high-quality case series were found; 2) Slight tendency: one high-quality case series was found; 3) Conflicting tendency: inconsistent or contradictory findings in multiple high-quality case series were found; and 4) No tendency: no high-quality case series were available. Division into these levels of tendencies is arbitrary.

Table 1 Methodological quality assessment for the evaluation of the effectiveness of interventions for treating radial tunnel syndrome.

Criteria		Score
Study population		
1	Specified eligibility criteria	+ / - / ?
2	Sufficient description of baseline characteristics	+ / - / ?
3	Cases ≥ 50	+ / - / ?
Interventions		
4	Adequate description of the used intervention	
	a) Description of the type of intervention	+ / - / ?
	b) Description of the application technique	+ / - / ?
5	Description of co-interventions	
Study design		
6	Prospective study design	+ / - / ?
7	Loss to follow-up/drop-outs	
	a) Information is given about loss to follow-up/drop-outs	+ / - / ?
	b) Item 7a positive and less than 10% loss to follow-up/drop-outs	+ / - / ?
	c) Item 7a positive and information is given about completers versus loss to follow up/drop-outs	+ / - / ?
8	Timing follow-up measurements	
	a) A short-term follow-up measurement was performed	+ / - / ?
	b) A long-term follow-up measurement was performed	+ / - / ?
Outcome measurements		
9	The outcome measurements are relevant	+ / - / ?
10	Description of adverse effects	+ / - / ?
11	The interval between (different) measurements was identical for all patients	+ / - / ?
12	Standardized or valid measurements	+ / - / ?
13	Data presentation of most important outcome measurements	+ / - / ?
Analysis		
14	An intention-to-treat analysis was used if necessary	+ / - / ?
15	Appropriate analysis techniques were used	+ / - / ?

+: Positive item

-: Negative item

?: unclear/information unknown

Results

Study selection

In total 282 potentially relevant abstracts were found after searching the 5 databases: 276 in Medline, 6 in Embase and none in Central, Cinahl or Pedro; 70 seemed to be relevant for our review. Reviewing the full text resulted in the inclusion of 16 articles. Citation tracking resulted in the identification of another 26 relevant abstracts and 5 articles could be included. In total, 21 articles were included in this review.

Study characteristics

All 21 included studies evaluated the efficacy of surgical treatment (decompression of the PIN). One study also reported on conservative treatments (all cases underwent corticosteroid injection, one case had additional physiotherapy).

Methodological quality assessment

Table 2 presents the results of the quality assessment score of each study. Only six articles scored 50% or more (≥ 10 points) of the maximum attainable score and were considered to be of high quality. The ratings of the most important criteria of these six articles are examined below.

Study population

All six articles described the symptoms and diagnostic criteria of the RTS. Descriptions of the baseline characteristics were sufficient. Two studies[14,15] included more than 50 cases of RTS.

Interventions

Surgical decompression was adequately described in all of the six studies. Co-interventions were mentioned in one study.[16]

Study design

Only Werner[15] used a prospective study design. All studies reported on loss to follow-up. One study[17] had more than 10% loss to follow-up and two studies[17,18] gave no information about completers versus loss to follow-up. In two studies[15,17] the follow-up period exceeded 12 months or more.

Outcome measurements

Most articles used relevant, standardised or validated outcome measures. Four studies used the criteria of Roles and Maudsley[19]; their results were classified into four groups (excellent, good, fair and poor) and contained aspects of pain, activity and movement. Hagert et al.[14]

Table 2 Scores on the methodological quality assessment.

Treatment RTS		Study population			Interventions			Study design							Outcome measurements				Analysis		Score
Author	*Year*	*1*	*2*	*3*	*4a*	*4b*	*5*	*6*	*7a*	*7b*	*7c*	*8a*	*8b*	*9*	*10*	*11*	*12*	*13*	*14*	*15*	*Max 19*
Werner	1979	+	+	+	+	+	-	+	+	+	+	+	+	+	+	+	+	+	+	+	**18**
Hagert et al.	1977	+	+	+	+	+	-	+	+	+	+	-	-	+	+	-	+	-	+	-	**13**
Roles et al.	1972	+	+	-	+	+	?	-	+	+	+	-	-	+	-	-	+	+	+	-	**11**
Jebson et al.	1997	+	+	-	+	+	-	-	+	-	-	-	+	+	+	-	+	+	-	-	**10**
Lister et al.	1979	+	+	-	+	+	+	?	+	+	+	-	-	-	+	-	-	-	+	-	**10**
De Smet et al.	1999	+	+	-	+	+	-	-	+	+	-	?	?	+	-	-	+	+	-	+	**10**
Border of 50% score on the methodological quality assessment																					
Kalb et al.	1999 2000	+	-	+	+	+	-	-	+	-	-	-	-	+	+	-	+	+	-	-	9
Lawrence et al.	1995	+	+	-	+	+	-	-	+	-	-	-	+	-	+	-	+	+	-	-	9
Raimbeau et al.	1990	+	+	-	+	+	-	-	+	+	-	-	-	+	-	-	+	+	-	-	9
Ritts et al.	1987	+	+	-	+	+	-	-	+	+	-	-	-	+	-	-	+	+	-	-	9
Sotereanos et al.	1999	+	+	-	+	+	-	?	+	-	-	-	+	+	-	-	+	+	-	-	9
Atroshi et al.	1995	+	+	-	+	+	-	-	-	-	-	-	+	+	+	-	?	+	-	-	8
Feldmeier et al.	1981	+	+	-	+	+	-	?	+	+	-	-	-	+	-	-	-	+	-	-	8
Sarhadi et al.	1998	+	+	-	+	+	+	?	-	-	-	+	-	-	+	?	-	-	-	-	7
Vollinger et al.	1998	+	-	-	+	+	-	-	-	-	-	-	-	+	+	-	+	+	-	-	7
Younge et al.	1994	+	+	-	+	+	-	-	-	-	-	?	?	+	+	-	?	+	-	-	7
Alnot et al.	1993	+	-	-	+	+	-	-	-	-	-	-	-	+	?	-	+	+	-	-	6
Hong et al.	1989	+	+	-	+	+	-	?	-	-	-	+	-	-	-	-	+	-	-	-	6
Moss et al.	1983	+	+	-	+	+	-	-	-	-	-	-	-	-	+	-	-	+	-	-	6
Minami et al.	1992	+	+	-	+	+	-	?	-	-	-	-	-	-	-	-	-	+	-	-	5
Makai et al.	1989	+	-	-	+	+	-	?	-	?	?	-	+	-	?	-	?	-	?	-	4

+: Positive item / -: Negative item / ?: unclear/information unknown

Table 3 Criteria according to Roles and Maudsley, and Hagert et al.

Results	Description of Roles and Maudsley	Description of Hagert et al.
Excellent	• no pain • full movement • full activity	• complete and persisting relief of all complaints
Good	• occasional discomfort • full movement • full activity	• considerable improvement • relief of all complaints except a slight, subsiding pain following exertion periodically • pain is not impairing the ability to continue working • no pain at night
Fair	• some discomfort after prolonged activity	• improvement, but periods of pain following exertion • periodically compromising the capacity of work • periodically pain at night
Poor	• pain limiting activities	• no improvement • pain as preoperatively

also classified their results in four groups, which are comparable with the classification of Roles and Maudsley[19] (Table 3). Lister et al.[16] restricted their study to relief of pain only (Table 4). Three studies[16-18] also reported patients' satisfaction with treatment (Table 4). Four studies described adverse effects. Only Werner[15] applied identical intervals between assessments. Four studies reported frequencies or percentages of the main outcome measures.

Analysis

Four articles used an intention to treat analysis. Two studies[14,15] used appropriate analysis techniques.

Data extraction

Table 5 presents the data extraction of the six high-quality studies. Data extraction of the low-quality articles is available on request.

Table 4 Improvement (%) in the study population, measured by the different criteria lists.

Author	Year	No. of patients	Criteria Roles & Maudsley or Hagert et al.	Lister et al.	Patients' satisfaction
De Smet	1999	22	75%		40%
Jebson	1997	33	67%		83%
Lister	1979	20		95%	83%
Werner	1979	90	81%		
Hagert	1977	50	84%		
Roles	1972	38	92%		

Table 5 Characteristics of the six high-quality studies.

Author, Year of publication	Score Quality	Study population	Case definition	Intervention
Hagert et al. 1979 Retrospective N=48 (50 cases)	12	Age (y): male: 47.0 (19-55) female:40.8 (20-59) Sex : male: 20; female: 28 Duration complaints: ± 2 years (6 months-10 yrs) Extremity operated on: right arm: 35 cases left arm: 15 cases	Compression of the PIN at the level of the arcade of Frohse, which causes pain without pareses at the level of the proximal forearm.	Decompression of the PIN Operation technique: Dissection of all possible compressing structures
Jebson et al. 1997 Retrospective N=31 (33 cases)	10	Age (y): mean: 37 (17-61) Sex : male: 7; female: 16 Duration complaints: 15 months (2-30 months) Extremity operated on: RD: 17 LD: 2 RND: 1 LND: 4	RTS is caused by compression of the PIN consisting of forearm pain without motor weakness.	Surgical decompression of the PIN. Operation technique: brachioradialis muscle splitting approach.
Lister et al. 1979 Retrospective N=18 (20 cases)	10	Age (y): 40.25 (18-58) Sex : male: 9; female: 9 Duration complaints: 21.1 months (3 months-5 yrs) Extremity operated on: dominant: 15 extremities non-dominant: 5 extremities	RTS is a compression of the PIN, which primarily causes pain at the extensor mass just below the elbow. Pain- related weakness of the grip might be present.	Surgical decompression of the PIN Operation technique: Anterior approach followed by dissection of all compressing elements.
Roles et al. 1972 Retrospective N=36 (38 cases)	11	Age (y): 44,6 (80% 30-50) Sex : male: 20 (56%); female: 16 (44%) Duration complaints: 32 months (3-168 months) Extremity operated on: RD: 28 LD: 1 LND: 5 L+R: 2 D: 89%	RTS is an entrapment neuropathy of the radial nerve and/or its branches at the elbow and it is a possible explanation for the complaints of resistant lateral epicondylitis.	Surgical decompression of the PIN. Operation technique: Anterior approach to divide the edge of the ECRB

Outcome Measures	Results	Conclusion authors
Classification into four groups including the following outcomes (Table 5): - relief of complaints - pain - work	- excellent: 33/50 - good: 9/50 - fair: 6/50 - poor: 2/50 Follow-up: mean: 2 years range: 1 months- 3 years Lost to follow-up: 2	- if the complaints do not subside spontaneously, relief can be expected in the majority of patients by decompression of the PIN - RTS and lateral epicondylitis are two different entities
Criteria included outcome measurements of Roles & Maudsley (Table 5) and Ritts et al.: Pain, ROM elbow, return to work,- recreational activity, patients satisfaction with treatment, additional surgery Criteria by Rittset al.: - excellent: minimal or no discomfort, return to work and recreational activity, feel functional improvement after the surgery - good: feel improvement but still moderate pain limited in physical capacity to use the extremity at work or recreation - poor: moderate or severe discomfort, inability to work or recreation because of forearm pain need of additional surgery	Classification of Roles and Maudsley: - excellent: 8/23 (38%) - good: 7/23 (29%) - fair: 3/23 (13%) - poor: 5/23 (21%) Classification of Ritts et al.: - good: 16/23 (71%) - fair: 3/23 (13%) - poor: 4/23 (17%) Return to work: - same occupation: 16/23 (71%) - change occupation: 5/23 (22%) - change occupation for unrelated reasons: 2/23 (9%) Patients' satisfaction with results: 19/23 (83%) Follow-up: mean 97 months Lost to follow up: 8	- a significant percentage of patients with RTS are helped by surgery but complete pain relief and return to occupational and recreational activities is unpredictable - the cause of RTS is controversial
- relief of complaints (pain) - satisfaction with the procedure	- complete relief symptoms: 19/20 - no complete relief symptoms: 1/20 - dissatisfaction because of scar: 3 patients Follow-up: mean: 29.8 months range: 9 months-4 years and 6 months No loss to follow-up.	RTS and epicondylitis lateralis are two different disorders. Radial tunnel release by dividing the four compressing factors is a sufficient intervention. RTS can be diagnosed by four symptoms (Lister et al. page. 58)
Classification into four groups by Roles and Maudsley (table 5) including the following outcomes: - pain - movement - activity	- excellent: 18/38 (47%) - good: 17/38 (45%) - fair: 2/83 (5%) - poor: 1/38 (3%) Follow-up: - mean: 29 months - range: 1-192 months	- depending on the severity, duration and site of compression of the PIN in the radial tunnel, there may be a range of clinical syndromes from simple tennis elbow to irreversible paralysis.

Table 5 continued

Author, Year of publication	Score Quality	Study population	Case definition	Intervention
De Smet et al. 1999 Retrospective N=21 (22 cases)	10	Age (y): mean: 40.4 (19-53) Sex: male: 10; female: 11 Duration complaints: 10.6 months (1 month- 3 yrs) Extremity operated on: right arm: 12 left arm: 8 bilateral: 1	RTS is a compression syndrome of the radial nerve. Lateral elbow pain is the main symptom, but motor weakness may be present.	Decompression of the PIN by radial tunnel release Operation technique: Anterolateral approach followed by transsection of potentially compressing structures.
Werner 1979 Prospective N=85 (90 cases)	18	Age (y): male: 47(± 9 years) female:41 (± 9 years) Sex : male: 37; female: 48 Duration complaints: 6-12 months: 3 patients 12-24 months: 53 patients >24 months: 29 patients Extremity operated on: D: 65 ND: 11	RTS is an entrapment of the PIN at the elbow and causes pain at the lateral aspect of the elbow and the proximal part of the forearm, with or without motor weakness.	Surgical decompression of the PIN Operation technique: Posterior lateral approach.

Outcome Measures	Results	Conclusion authors
Classification into four groups by Roles and Maudsley (table X) including the following outcomes: - pain - movement - activity - weakness - patients' satisfaction	- all symptoms disappeared in 4/19 patients, - decrease of symptoms in 11/19 patients - 4/19 patients showed no change in symptoms, - 1/19 patient got more symptoms - 75% showed good and excellent results -VAS decreased from 6.7 to 3.6 - patients' satisfaction: high (>8) for 8 elbows and insufficient (<8) for 11 elbows (one not evaluated) Follow-up: unknown Lost to follow-up: 2	- surgical treatment is the preferred treatment when conservative treatments fail; - patient satisfaction is associated with a shorter duration of complaints before operation; - associated lateral epicondylar release seems to be essential for a satisfactory result - the cause of RTS is controversial
Classification into four groups comparable with Roles and Maudsley and Hagert (Table 5) including the following outcomes: - pain - movement - activity Measurements of 5 diagnostic tests by follow-up	- for results of the 5 outcome measures (table 3 page 20 of this study) - normalisation of grip strength: 64/81 (79%) against 22/81 (27%) preoperatively Definitions comparable to Roles and Maudsley criteria + Hagert et al criteria: - excellent: 64/90 (71%) - good: 9/90 (10%) - fair: 7/90 (8%) - poor: 10/90 (11%) Follow-up: - 1.5 months - 6 months - 12 months - 24 months Lost to follow up: 3	- decompression of the PIN gives good results by RTS - lateral elbow pain may be caused by dynamic compression of the nerve where it enters through the supinator muscle - the diagnostic tests for RTS are unreliable

Effectiveness of interventions

Conservative treatment

No articles presenting data on conservative treatment of RTS could be included. Therefore, no tendency was found for the effectiveness of conservative treatments.

Surgical treatment

All six studies evaluated the effectiveness of surgical decompression of the PIN. Table 4 gives the improvement percentage in the studied populations for the different criteria lists. The effectiveness of the surgical treatment ranged from 67-92% when the criteria of Roles and Maudsley[19] or Hagert et al.[14] were used. Lister et al.[16] reported pain relief in 95% of their patients. Patients' satisfaction ranged from 40-83%. Therefore, there is a tendency (level 1) that surgical decompression of the PIN is effective in patients with RTS.

Discussion

This study systematically reviewed all available observational studies on the treatment of RTS and showed that there is a tendency for the effectiveness of surgical decompression of the PIN in patients with RTS. The use of observational studies was introduced due to a lack of published RCTs and CCTs. When no controlled studies are available and the results of case series are systematically evaluated, this can be a useful method to summarize and compare case series and to identify methodological flaws.[20] We used the term 'tendency' to underline the relative weakness of the 'conclusive evidence' for efficacy of treatments relative to conclusions based on controlled trials.

None of the six included high-quality studies reported the effectiveness of conservative treatment of RTS. In the low-quality study of Sarhadi et al.[21] (score 7 on the methodological quality assessment) all patients first had conservative treatment by means of corticosteroid injection (25 cases) and physiotherapy (one case). Sarhadi et al.[21] reported 9 failures of conservative treatment. Thus, 16 patients (64%) were treated successfully by a conservative intervention. However, because of its low quality, this study was excluded from this review.

The decision to have a cut-off point of 50% is arbitrary. Had we used a cut-off point of 60%, two high-quality studies with generally consistent conclusions would still have remained, meaning that the conclusion of this review would have stayed the same. In 13 of the 15 low-quality studies the effectiveness of the surgical treatment ranged from 64-100%. This range is the same as the six articles identified using the cut-off point of 50%. Only Atroshi et al.[22] and Kalb et al.[23] reported a lower effectiveness of 35% and 58%, respectively, using the criteria of Roles and Maudsley.[19]

There are some limitations to this review and its conclusions. Of the 6 studies included in the analysis, 5 used a retrospective study design. Exclusion bias could have been introduced by including only those cases that were available for follow-up.
The patient selection of the included studies can be influenced by a difference in diagnostic criteria. Several authors mentioned the contradictory findings regarding the diagnosis of RTS. Werner[15] and Atroshi et al.[22] reported that the symptoms and signs used as diagnostic criteria for RTS are not reliable. Ritts et al.[24] mentioned a diagnostic grey zone that appears to exist for diagnosing RTS. The lack of a clear protocol for diagnosing RTS and the absence of reliable and valid diagnostics tests may have caused differences in patient selection and the results of the treatment.

Although much research has been done to determine the cause of the pain in patients with RTS, no precise anatomical pathology was found. Pain is the most common complaint in patients with RTS. When the duration of the pain is longer than 3 months, the symptoms can be interpreted as a chronic pain syndrome. In chronic complaints it is necessary to establish whether psychological and social factors exert an influence on the recovery. Focusing on the anatomic cause of RTS only, may lead to an inappropriate classification of patients with RTS and a sub-optimal treatment.[25]

Because the natural history of RTS is unknown, the precise role of surgery remains to be established. No attempt was made in the case series to consider the placebo effect of surgery.[26] Therefore RCTs are necessary. Because of the low incidence of RTS, we recommend a multi-center RCT.

This systematic review shows that there is a tendency for the effectiveness of surgical decompression of the PIN in patients with RTS. The effectiveness of conservative treatment of RTS is unknown because, for most treatments, no studies were identified. Additional high-quality controlled studies are needed to assess the level of 'conclusive evidence' for this treatment. There is also a need for more research into conservative treatment of RTS, including physiotherapy and steroid injection. Due to the lack of a clear protocol for diagnosing RTS, it is advised to develop a reliable and valid diagnostic protocol for RTS.

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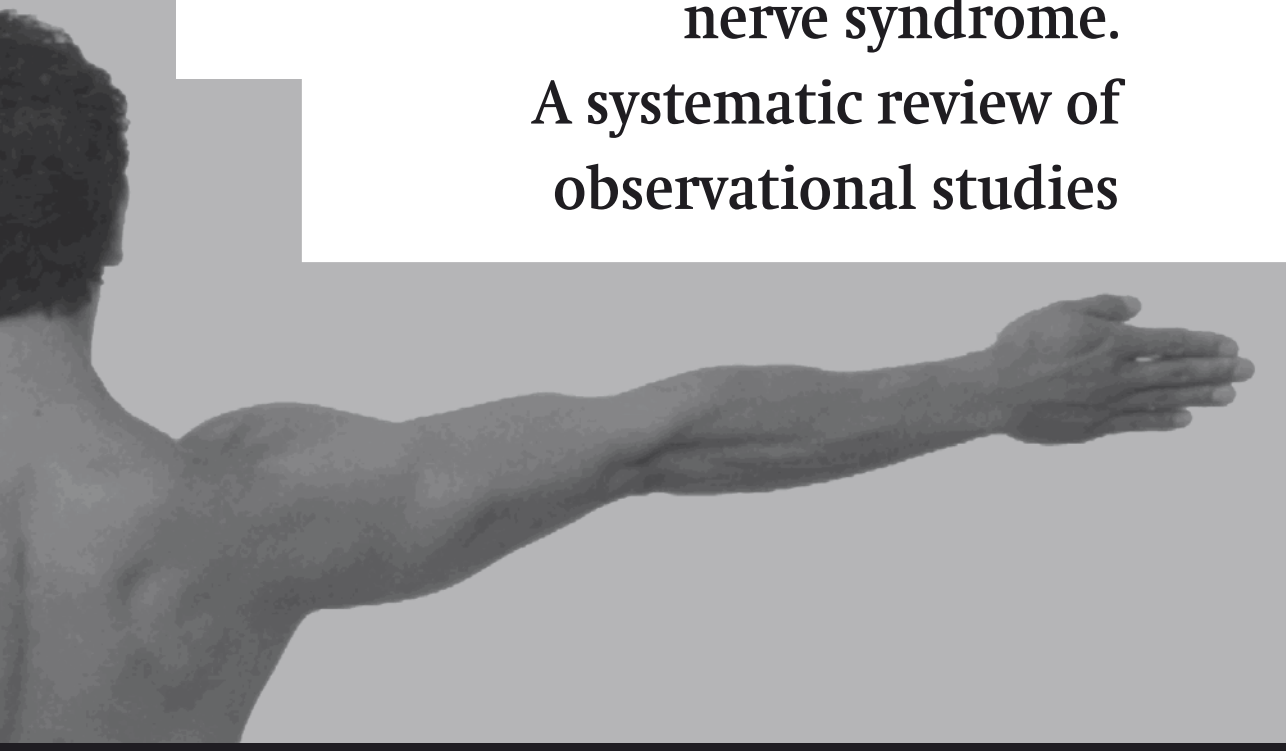
Appendix 1

Criteria
Study population
1 Positive if the symptoms and diagnostic criteria of the disorder were described
2 Positive if at least 4 of the following 6 items were reported at baseline: a) age (mean and standard deviation or CI) b) sex (number and/or percentage) c) outcome measures. Values for pain and functional status (mean and standard deviation or CI) d) affected extremity (dominant- non dominant) e) duration of symptoms (mean and standard deviation or CI) f) job description
3 Positive if the total number of cases was ≥ 50
Interventions
4 Adequate description of the used intervention a) Positive if the type of intervention was described b) Positive if the application technique was described
5 Positive if co-interventions were avoided in the design
Study design
6 Positive if the study design was prospective
7 Loss to follow-up a) Positive if information was given about loss to follow-up/drop-outs b) Positive if 7a) is positive and there was less than 10% loss to follow-up/drop-outs c) Positive if 7a) is positive and sociodemographic/clinical information (e.g. age, sex, type of complaints/ disabilities/ participation problems or prognostic factors) was presented for completers and those lost to follow-up/drop-outs at the main moment of outcome measurement, or no drop-outs/loss to follow-up. It is important that sociodemographic and clinical information was given for completers and follow-up/drop-outs to compare reasons for patients being lost. Reasons have to be unrelated to the outcome of recovery (complaints and disabilities). Loss to follow-up/drop-outs: all patients of the assembled cohort minus the number of patients at the moment of health status measurement for the main outcome measure, divided by all patients of the assembled cohort.
8 Timing follow-up measurements a) Positive if outcome assessment occurred at the end of the intervention period within 12 months after the intervention b) Positive if outcome assessment occurred > 12 months after the intervention period.
Outcome measurements
9 Positive if at least 3 of the following 6 items were used as outcome measures: pain, overall improvement, functional status (muscle strength, range of motion), medical consumption, disability (lost days of work or return to work), satisfaction with treatment
10 Each event should be described and correctly attributed to the allocated treatment: If it was explicitly reported that 'no adverse effects have occurred' or the adverse effects were described, this item is positive.
11 The interval between the intervention and the (different) measurements was identical for all individual patients. To score positive this item must be mentioned and described.
12 Positive if one or more of the main outcome measures were reported in a standardized or valid way (for example by means of a questionnaire, a diary or an objective outcome measure such as registration of lost days of work or medication use in the patient chart of general practitioners).
13 Positive if frequencies, or percentages of mean (and standard deviation/CI), or median (and Interquartile Range) were reported for one or more of the main outcome measures for the most important follow-up measurements.
Analysis
14 Positive if 7a) was positive and all patients were reported/analyzed for the most important outcome moments of effect measurement (minus missing values) irrespective of non-compliance and co-interventions.
15 Positive if both point estimates and measures of variance are presented. Point estimates are: means, medians, modes etc. Measures of variance are: standard deviations, 95% confidence intervals, etc

Chapter 5

Interventions for treating the posterior interosseus nerve syndrome. A systematic review of observational studies



Huisstede BM, Miedema HS, van Opstal T, de Ronde MT, Kuiper JI, Verhaar JA, Koes BW.

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Abstract

Background: For the posterior interosseus nerve syndrome (PINS), no randomized controlled trials or controlled clinical trials about the effectiveness of interventions are available; only case series can be found. Although the validity of case series is inferior to controlled trials, they may provide valuable data about the efficacy of treatment options. Therefore, we systematically reviewed all available observational studies on treatment of PINS.

Methods: A literature search and additional reference checking was done. On the basis of previous checklists, we constructed a quality assessment and rating system to analyse the included case series. Studies with less than 50% of the maximum points on the methodological quality assessment were excluded from the analysis.

Results: The results are summarised according to a rating system for the strength of the scientific evidence. Six eligible case series for this review were found. After the data extraction and methodological quality assessment, two higher quality studies that evaluated the effectiveness of surgical decompression of the PIN were included in the final analysis.

Conclusions: There is a tendency for the effectiveness of surgical decompression of the PIN in patients with PINS. The effectiveness of a conservative treatment for PINS is unknown because no higher quality studies are available. Additional high-quality controlled studies are needed to assess the level of 'conclusive evidence' for surgical treatment. There is also a need for high-quality controlled trials into the effectiveness of conservative treatments for PINS.

Introduction

Compressive neuropathies are common conditions of the upper extremity, with involvement of the ulnar and median nerves. Radial nerve involvement, such as the posterior interosseus nerve syndrome (PINS), is a less encountered condition. When searching for information in scientific literature about the effectiveness of interventions of the PINS, only case series can be found; no controlled studies are available. Although the validity of case series is inferior to controlled trials, they may give valuable tendencies for the efficacy of treatment options. Therefore, in this study we systematic reviewed all available case series about the effectiveness of the interventions of PINS.

Proximal to the supinator arch, the radial nerve divides into a superficial branch, the sensory superficial radial nerve and a deep motor branch, the (PIN).[1] The PIN passes the radial tunnel. The radial tunnel is defined as a space created by structures surrounding the radial nerve and the PIN that expands through the proximal forearm over a length of 5 cm starting at the humeroradial joint and past the proximal edge of the supinator muscle.[1] In the proximal region of the forearm, the radial nerve is associated with two different syndromes: radial tunnel syndrome (RTS) and PINS.[2,3]

In the literature, a clear distinction is not always made between these two syndromes. Some clinical findings can be found in both syndromes: pain in the forearm and marked weakness of extensors of fingers or wrist. It is generally accepted that PINS is caused by continuous or intermittent compression of the PIN in the radial tunnel.[4] There are contradictory ideas about the cause of RTS. Some attribute the RTS to compression of the PIN in the radial tunnel or consider that the syndrome results from intermittent and dynamic compression of the nerve in the proximal part of the forearm associated with repeated pronation and supination[5], whereas others reject these hypotheses.[6,7] Verhaar and Spaans[6] suggested that the symptoms of RTS may be caused by a lesion in the supinator muscle or in the septum between the extensor carpi radialis brevis and the extensor digitorum muscle.

The interventions, such as release of the superficial part of the supinator muscle, carried out for both syndromes, overlap to a large extent. However, since the aetiology might be different, we decided to look at the syndromes separately. We used the following descriptions of the syndromes:
The hallmark of **RTS** is pain over the radial proximal forearm with little or no motor weakness. Motor weakness, if present, can be explained as a result of the pain. Tenderness over the radial tunnel is an important criterion in establishing the diagnosis RTS.[2]
The patient with **PINS** shows loss of motor function or even complete palsy of one or more muscles innervated by the PIN. Patients with PINS may have pain, but this is not the main

symptom. The clinical presentation of the PINS is characterized by the loss of function due to variable degrees of weakness involving ulnar wrist extension, finger and thumb extension. When the compression in the radial tunnel causes a complete palsy, wrist dorsiflexion is possible but only in radial direction. When all the extensors of the fingers are paralysed, extension of the fingers or thumb is not possible at their metacarpophalangeal joints, but extension of the interphalangeal joint initiated by the intrinsic muscles remains possible. Paralysis or pareses may be limited to one or two fingers. Careful clinical and electrophysiological examination is important and essential for a reliable diagnosis.[8,9] An electromyogram (EMG) can establish the topography of the lesion and the severity of the muscular denervation.[10]

Cravens and Kline[9] reviewed 170 patients with radial nerve disorders. Of these, 19% were diagnosed with PINS. Kalb et al.[11] reported less than 10% PINS in 111 patients with problems related to the radial nerve in the proximal forearm. Vrieling et al.[10] reported that in 25% of patients the PINS is caused by a trauma, in 15% the syndrome is iatrogenic, and that in the remaining 60%, the disorder develops spontaneously. The 'spontaneous' PINS is most frequently caused by an entrapment at the level of the arcade of Frohse, the part where the nerve enters the supinator muscle.[8-10] It is most likely caused by a combination of anatomical anomalies and combined with repetitive pronation-supination movements of the forearm.[10-12] PIN entrapment is more likely to occur when the arcade of Frohse is fibrous and thickened.

In this systematic review, we concentrate on the effectiveness of interventions of the PINS. A systematic review about the effectiveness of the interventions of the RTS can be found elsewhere.[13]
Therapeutic interventions for patients with PINS include surgical decompression of the PIN, physiotherapy, steroid injection and immobilisation. The primary objective of this review is to systematically review the efficacy of conservative and surgical interventions for PINS.

Materials and methods

Literature Search

To identify relevant publications, we searched the following databases: Medline (1966 to April 2004), Embase (1980 to April 2004), the Cochrane Library (1993 to April 2004), Pedro (up to April 2004), Cinahl (1982 to April 2004) and CENTRAL (up to April 2004). All the keywords related to the treatment of PINS in relevant articles were included in the literature search, such as: 'posterior interosseus syndrome', 'supinator syndrome', 'radial tunnel syndrome, 'PINS', 'radial nerve compression', 'treatment', 'therapy', 'surgical' and 'conservative'. The complete search strategy is available upon request. One reviewer (MTMR) checked the references of all retrieved articles to identify additional studies on the topic (i.e., citation tracking).

A study was eligible for inclusion if it met all of the following criteria: (1) loss of motor function of one or more muscles innervated by the PIN is described; (2) an intervention for treating the PINS was done; (3) the study population consisted of patients aged 18 years or older, and (4) at least five patients were included in the study. All languages were included.

PINS due to an acute trauma, poly-neuropathies, PINS as secondary consequence of disease e.g., rheumatic syndromes, diabetes or tumours, neurological diseases and hereditary neuropathy were excluded. Studies in which part of the study population met our inclusion criteria were included if the results for this subpopulation were presented separately.

Study selection

Two reviewers (BMAH and MTMR) independently applied the inclusion criteria to select potential relevant studies from the titles, abstracts, and keywords of the references retrieved by the literature search. A consensus method was used to solve disagreements concerning inclusion of studies, and a third reviewer (HSM) was consulted if disagreement persisted.

Assessment of methodological quality

Because the current quality assessments are specifically developed for randomised controlled trials (RCTs), a new quality assessment list for the included case series was constructed (Table 1). The Appendix summarises the operationalisation of the criteria. The criteria were adapted from Borghouts et al. (1998), Lievense et al. (2001) and Van Tulder et al. (2003)[14-16] and modified to cover the topic of this review. The list consists of five topics: study population, interventions, study design, outcome measurements, and analysis. The five topics consist of a total of 19 items. Items of both internal and external validity were included.
Two reviewers (BMAH and TvO) independently scored the quality of each study. A consensus method was used to solve disagreements, and a third reviewer (HSM) was consulted if disagreements persisted. Each quality item was scored as positive ('+'), negative ('-') or unclear ('?') and was given one point when the reviewer scored a '+'. The maximum overall score was 19 points.

Data extraction

Two reviewers (BMAH and TvO) independently extracted the data. Information was collected on the study population, interventions used, study design, outcome measurements and data analysis.

Data analysis

We used a rating system to analyse the included case series on methodological quality. Because no randomized clinical trials (RCTs) or controlled clinical trials (CCTs) were found, the conclusions of this review cannot be seen as 'conclusive evidence' for the efficacy of

Table 1 Methodological quality assessment for the evaluation of the effectiveness of interventions for treating posterior interosseus nerve syndrome.

Criteria		Score
Study population		
1	Specified eligibility criteria	+ / - / ?
2	Sufficient description of baseline characteristics	+ / - / ?
3	Cases ≥ 50	+ / - / ?
Interventions		
4	Adequate description of the used intervention	
	a) Description of the type of intervention	+ / - / ?
	b) Description of the application technique	+ / - / ?
5	Description of co-interventions	
Study design		
6	Prospective study design	+ / - / ?
7	Loss to follow-up/drop-outs	
	a) Information is given about loss to follow-up/drop-outs	+ / - / ?
	b) Item 7a positive and less than 10% loss to follow-up/drop-outs	+ / - / ?
	c) Item 7a positive and information is given about completers versus Loss to follow up/drop-outs	+ / - / ?
8	Timing follow-up measurements	
	a) A short-term follow-up measurement was performed	+ / - / ?
	b) A long-term follow-up measurement was performed	+ / - / ?
Outcome measurements		
9	The outcome measurements are relevant	+ / - / ?
10	Description of adverse effects	+ / - / ?
11	The interval between (different) measurements was identical for all patients	+ / - / ?
12	Standardized or valid measurements	+ / - / ?
13	Data presentation of most important outcome measurements	+ / - / ?
Analysis		
14	An intention-to-treat analysis was used if necessary	+ / - / ?
15	Appropriate analysis techniques were used	+ / - / ?

+: Positive item

-: Negative item

?: Unclear/information unknown

treatments. In this rating system, the term 'tendency' was used to emphasise that case series cannot supply strong evidence for or against the efficacy of interventions. Studies with less than 50% of the total score on the methodological quality assessment were defined as inadequate and excluded from the final analysis. The quality of the case series was considered to be higher when the score on the methodological assessment was 50% or more.

To summarise the results according to the rating system for the strength of the scientific evidence, four levels are used: (1) Tendency: providing that generally consistent findings in multiple higher quality case series were found; (2) Slight tendency: one higher quality case serie was found; (3) Conflicting tendency: inconsistent or contradictory findings in multiple higher quality case series were found; and (4) No tendency: no higher quality case series were available. Division into these levels of tendencies is arbitrary.

Results

Study selection

In total, 282 potentially relevant abstracts were found after searching the five databases: 276 in Medline, 6 in Embase and none in Central, Cinahl or Pedro. On the basis of the inclusion criteria, 70 of these abstracts seemed to be relevant for our review. Citation tracking resulted in identification of another six articles. Reviewing the full text of the 76 articles resulted in inclusion of six articles.

Study characteristics

Surgical treatment was evaluated in all six included studies. One study[8] also evaluated the efficacy of a not further specified conservative treatment, and another study[17] reported a patient group who did not receive any treatment.

Methodological quality assessment

The results of the quality assessment score of the six studies are shown in Table 2. Only two articles[10,11] scored 50% or more (≥ 10 points) of the maximum attainable score and were considered to be of higher quality for inclusion in this review. The ratings of the most important criteria of these two articles are examined below.

Study population

Both Vrieling et al.[10] and Kalb et al.[11] reported the eligibility criteria and made a sufficient description of baseline characteristics. Vrieling et al.[10] studied 14 patients, and Kalb et al.[11] studied 110 patients. From both populations, eight and nine patients, respectively, were diagnosed as having PINS.

Interventions

The description of the surgical decompression was adequately described in both articles. Co-interventions were not mentioned.

Study design

Both studies used a retrospective study design. Vrieling et al.[10] reported no loss to follow-up. Kalb et al.[11] lost one patient during follow-up and did not present information about completers versus loss to follow-up. Kalb et al.[11] performed a long-term follow-up. Follow-up time presented by Vrieling et al.[10] varied from 1 month to 12 years.

Outcome measurements

Both studies used relevant, standardised or validated outcome measures, described adverse effects and presented data of the most important outcome measurements. In both studies,

Table 2 Scores on the methodological quality assessment.

Treatment RTS		Study population			Interventions					Study design						Outcome measurements					Analysis		Score
Author	*Year*	*1*	*2*	*3*	*4a*	*4b*	*5*	*6*	*7a*	*7b*	*7c*	*8a*	*8b*	*9*	*10*	*11*	*12*	*13*	*14*	*15*			*Max 19*
Vrieling et al.	1998	+	+	-	+	+	-	-	+	+	+	-	+	+	+	-	+	+	+	-			**13**
Kalb et al.	2000	+	+	-	+	+	-	-	+	-	-	-	+	+	+	-	+	+	-	-			**10**
Border of 50% score on the methodological quality assessment																							
Cravens et al.	1990	+	+	-	+	+	-	-	-	-	-	?	?	-	-	?	-	+	-	-			6
Hashizume et al.	1996	+	+	-	+	+	-	-	-	-	-	?	?	-	-	?	-	+	-	-			5
Jürgens et al.	1987	+	-	-	+	-	-	-	+	-	+	-	-	-	-	-	-	+	?	-			5
Privat et al.	1979	+	-	-	+	-	?	-	-	-	-	?	?	-	-	?	-	-	-	-			2

+: Positive item

-: Negative item

?: Unclear/information unknown

the interval between the surgical treatment and the follow-up measurement was not identical for all patients.

Analysis

No appropriate analysis techniques were used in both studies, and no statistical measurements were presented.

Data extraction

Table 3 presents the data extraction (study population, case definition, intervention, outcome measures, results and the authors conclusion) of the six studies that met the inclusion criteria.

Effectiveness of interventions

Conservative treatment

None of the two included studies reported on the effectiveness of conservative treatment of PINS. One low-quality study[8] mentioned conservative treatment. Of the four patients who were treated conservatively, three recovered. No information was given about the type of conservative treatment.

Surgical treatment

Vrieling et al.[10] and Kalb et al.[11] evaluated the effectiveness of surgical decompression of the PIN. Vrieling et al.[10] reported excellent or good results on muscle strength and pain in six of the eight patients. Two patients had poor results. Kalb et al.[11] reported full recovery of the paresis in six out of the eight patients available at follow-up; four of these patients state that they would undergo the operation again knowing the postoperative results.

On the basis of the two included studies, there is a tendency (level 1) that surgical decompression of the PIN is effective in patients with PINS.

Discussion

We systematically reviewed all available observational studies for the treatment of the PINS.

Although the distinction in PINS and RTS may be confusing to some because they contribute the syndromes to entrapment of the same nerve within the same region, we decided to separate them in order to avoid drawing conclusions about the effectiveness of interventions of RTS or PINS based on possible differences in aetiology.

Table 3 Characteristics of the six studies.

Author, Year of publication	Score Quality	N	Study population	Case definition	Intervention
Vrieling et al. 1998 retrospective	13	N=8 (of the reported 14 cases)	Age (y): 52 (24-84) Sex: Male: 4 Female: 4 Duration of complaints: 3,9 months (2-12 months) Extremity operated on: RD: 6 patients LND: 2 patients	PIN is an entrapment neuropathy of the PIN at the location where the nerve passes through the radial tunnel which causes pareses in the muscles innervated by the PIN	Of the 8 non traumatic cases: Surgical decompression of the PIN Operation technique: Anterior approach
Kalb et al. 2000 retrospective	10	N= 9 (out of 110 patients)	Age (y): Mean: 41 range: unknown Sex: Male: 5 Female: 4 Duration complaints: Unknown Extremity operated on: Unknown	The supinator syndrome contains two disorders with different clinical manifestations, which are both caused by compression of the PIN at the supinatorloge: 1) RTS. With pain/ tenderness as the main symptom, 2) PIN-syndrome: with paresis as the main symptom.	Surgical decompression of the PIN by cutting the superficial origin of the supinator muscle with consecutive relaxation of Frohse´s arcade. Operation technique: anterior approach by Wilhelm's denervation procedure
Cravens et al. 1990 retrospective	6	N=14 (out of 32 cases)	Of the 32 cases: Age (y): Men: 29 (12-72) Women: 32.8 (19-49) Sex: Male: 19 Female: 11 Duration complaints: Less than 1 year Extremity operated on: Right side: 22 Left side: 10 26 patients (28 cases) underwent operation from which 14 patients had an entrapment.	Entrapment at the level of the supinator muscle or arcade of Frohse causing isolated weakness of muscles innervated by the PIN. Diffuse forearm pain may be associated symptoms.	Surgical exploration of the PIN Operation technique: anterior approach followed by dissection of vessels and a neurolysis.

Outcome Measures	Results	Conclusion authors
Classification into four groups: Excellent: A grade-5 motor power existed in all the affected muscles and/or a complete and persisting relief of pain was experienced Good: If the patient had at least grade-4 muscle power and/or slight elbow pain following heavy use of the arm Fair: The patient had at least grade-3 muscle power and/or pain provoked by moderate exertion, thus limiting the function of the involved limb Poor: There was very little or no improvement compared to the pre-operative status.	Excellent: 1 Good: 5 Fair: - Poor: 2 Follow up: Mean: 5 years (1 month -12 years) No loss to follow up	For patients with PIN paresis the period of observation should be six to eight months, for patients with a full paralysis it is not possible to make a recommendation concerning treatment from this series.
- pain - ROM elbow - daily activity - return to work - patients satisfaction with results	-Recovery of the paresis: full recovery: 4 largely recovery: 2 no recovery: 2 - 4 of 7 patients noticed a subjective improvement of complaints - mean DASH-score: 19 - 4 of the 8 patients would undergo the operation again knowing the postoperative results. Follow-up: - Mean: 55 months (range: 19-72 months) Loss to follow-up: 1 patient	-Surgical decompression of the PIN is recommended if incomplete palsy worsens or if complete palsy persists for more than 12 weeks. After 24 months no effect can be expected by surgical decompression.
-Recovery of strength of extensor carpi ulnaris (ECU), extensor communis (EC) and extensor pollicus longus (EPL) 0:No ECU, EC, or EPL, muscle function; 1:Trace or against gravity of ECU only, absent EC and EPL muscle function; 2:Recovery of ECU, absent or trace only of EC and/or EPL muscle; 3:Recovery of ECU, some EC, weak or absent EPL muscle function; 4: Recovery of moderate strength of EC and EPL, full strength in ECU muscle function 5: Recovery of full strength of EPL, EC, and ECU muscle function	Preoperative grade versus postoperative grade: 0/5 - 5/5: 3 patients 0/5 - 4/5: 1 patient 0/5 - 3/5: 1 patient 1/5 - 5/5: 1 patient 2/5 - 4/5: 3 patients 3/5 - 4/5: 1 patient 3/5 - 5/5: 4 patients 13 of 14 patients recovered either to Grade 4/5 (5 patients) or 5/5 (8 patients). One recovered to Grade 3/5.	Patients who do not have a return of function either clinically or with electrical testing after 3 months are candidates for surgical decompression. In the present series, neurolysis or surgical repair utilizing direct suture or interfascicular grafts provided good to excellent results in almost all cases.

Table 3 continued

Author, Year of publication	Score Quality	N	Study population	Case definition	Intervention
Hashizume et al. 1996 retrospective	5	N=21 (out of 31 cases)	Age (y): 40.3 (17-71) Sex: Male: 7 Female: 14 Duration complaints before operation: Mean: 8,8 weeks Range: 2-18 weeks Duration complaints before conservative treatment: Unknown Extremity operated on: Right side: 14 Left side: 17 Dominant: 17 (55%) 15 of the 31 patients had a pure entrapment of the PIN (without ganglion or neuralgic amyotrophy).	Non-traumatic paralysis of the PIN is caused by entrapment by the edge of the supinator.	Surgical decompression of the PIN by 17 patients with an entrapment of the PIN. Operation technique: exploration of the PIN by the anterior approach described by Henry. 4 of the 21 patients with an entrapment of the PIN were treated conservatively
Jürgens et al. 1987 retrospective	5	N=9 (out of 20 cases)	Age (y): 47,55 (27-73) Sex: male: 3 female: 6 Duration complaints 41 months (12 days – 16 years) Extremity operated on: unknown	The supinator syndrome can be caused by an entrapment of the deep branch of the radial nerve.	Surgical decompression of the PIN: 5 patients. Operation technique: unknown No treatment: 3 patients Treatment unknown: 1 patient
Privat et al. 1979 retrospective	2	N= 7 (out of 16 cases)	Age (y): unknown. Sex: unknown. From the 16 patients, 7 patients had a pure entrapment of the PIN.	Radial nerve compression by the radial tunnel which causes paralysis of one or more muscles innervated by the PIN	Surgical decompression of the PIN. Surgical technique: Unknown.

Outcome Measures	Results	Conclusion authors
Recovery of strength of the muscles innervated by the PIN.	Of the 17 patients with a PIN entrapment and treated by surgical decompression 16 recovered. 1 patient which had a surgical decompression had a poor result Of the 4 patients who were treated conservatively, 3 recovered. Period till total recovery (of the 19 recovered patients): Mean: 4.5 months Range: 2-8 months Follow-up: Unknown.	Patients who do not have a return of function either clinically or with electrical testing after 3 months are candidates for surgical exploration.
Recovery of strength of the muscles innervated by the PIN.	Surgical decompression: 3 out of 5 patients recovered No treatment: 1 out of 3 patients recovered Follow up: 19 months (1 month - 3 years) Loss to follow up: 1 patient	Operative results in idiopathic cases are poor; it seems to be purposeless to operate patients who have a paresis for a long time. An indication for operative treatment is present only in the case of complete transsection by trauma.
Not mentioned.	3 out of the 7 patients showed a clinical improvement after surgical decompression Follow-up: Unknown	A non-traumatic entrapment of the PIN generally responds well on a surgical treatment.

The use of observational studies in this review was due to a lack of published RCTs and CCTs. Systematic reviewing observational studies can be a useful method to summarize and compare case series and to identify methodological flaws.[18] On the basis of previous lists, a methodological quality assessment list was constructed for case series. The scores on the quality assessment of the six studies included in this review ranged from 10 to 68%. The term 'tendency' was introduced to underline the relative weakness of the 'conclusive evidence' for efficacy of treatments relative to conclusions based on controlled trials.
Only those two studies[10,11] with more than 50% of the score were considered to be of higher methodological quality for inclusion in this review. They both reported recovery rates of 75% for surgical treatment. Therefore, we concluded that there is tendency that surgical decompression of the PIN may be effective for treating PINS. As well as the two higher quality studies, the four lower quality studies[8,9,17,19] reported recovery rates of 93, 94, 60 and 43%, respectively, on surgical decompression of the PIN.
There were no studies of higher quality available evaluating conservative treatments. Hence, unfortunately, no information is available to draw a firm conclusion regarding the effectiveness of conservative treatment.
The decision to have a cut-off point of 50% is arbitrary. Had we used a cut-off point of 60%, just one higher quality study remained and the conclusion should be changed in 'slight tendency' for effectiveness of surgical decompression of the PIN.

Diagnosing PINS may be difficult. In our opinion electrophysiological examination should always be done to confirm the diagnosis. Reduced conduction velocity and EMG abnormalities may be found in the mm.extensor digitorum. In five of the six included studies, electrophysiological examinations were done; in four studies, including the two studies of higher quality, the findings of these examinations were used to confirm the diagnose (Table 4).
There is no agreement about treatment in the literature. Vrieling et al.[10] concluded that patients with PIN paresis should be observated for at least 6-8 months before surgical decompression is indicated for patients with a complete paralysis. Kalb et al.[11] recommended surgical decompression of the PIN if incomplete palsy increases or if complete palsy persists for more than 12 weeks. According to Kalb et al.[11], after 18 months of PIN palsy, surgical treatment is disputable. Other authors[17,20] also concluded that the results of the operations depended on the duration of the symptoms; a long-existing paralysis made reinnervation less likely to occur. Hence, timing of surgical intervention is an issue and under debate.

The presence of several forms of bias limits this review and its conclusions. Although publication bias was avoided by screening the reference lists of the included studies, it is possible that some relevant publications other then RCTs, CCTs and observational studies may have been missed. A second limitation is that our search was restricted to studies published in indexed journals (i.e., MEDLINE, EMBASE, CENTRAL, CINAHL and PEDRO), so that unpublished

studies and non-indexed journals were discarded.[21] Due to the fact that the included studies have a retrospective study design, exclusion bias could have been present because of inclusion of only those cases that were available for follow-up. It was difficult to compare the results of the studies because of multiple variables and differences in follow-up periods.

Because of the low incidence of PINS, it is complicated to evaluate the effectiveness of treatments of the disorder. The cases we found were all extracted from a larger and broader study population. Using the rating system, the results of our systematic review shows that there is a tendency that surgical decompression of the PIN may be effective in patients with PINS. The performance of high-quality RCTs can be recommended to provide conclusive evidence. The effectiveness of conservative treatment of PINS is unknown because no higher quality studies could be identified. Further research is necessary to assess the effectiveness of conservative treatments for PINS. Because of the low incidence of PINS, multi-center randomised clinical trials can be recommended.

Table 4 Electrophysiological examinations in the six studies

Study	Electrophysiological examination	
	Performed?	Used to diagnose PINS?
*Higher quality studies**		
Vrieling et al. 1998	+	+
Kalb et al. 2000	+	+
*Lower quality studies**		
Cravens et al. 1990	+	-
Hashizume et al. 1996	+	+
Jürgens et al. 1987	+	+
Privat et al. 1979	-	-

* in order of score on the quality assessment

+: yes

- : no

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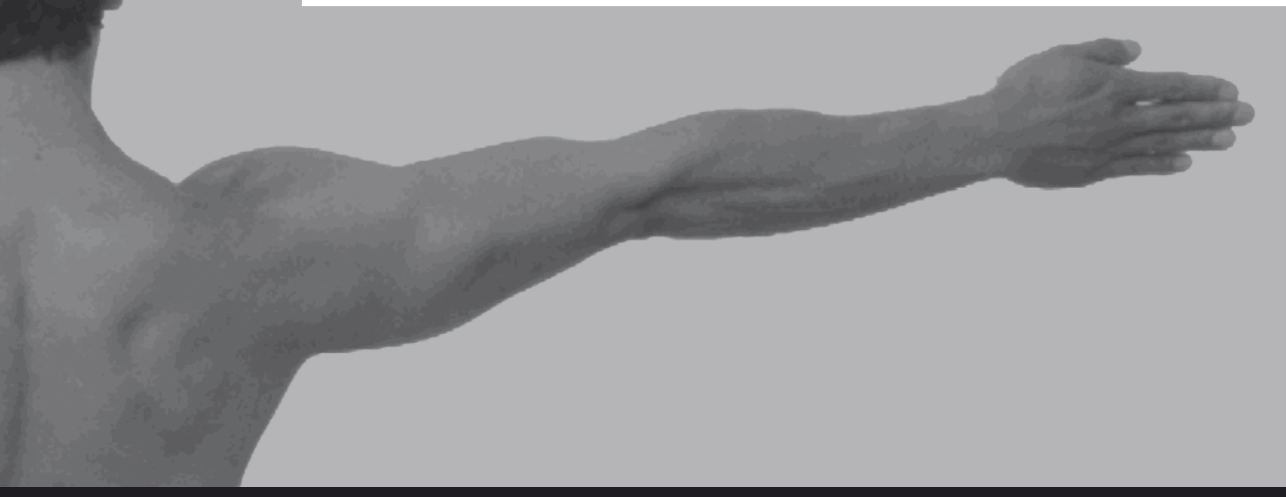
Appendix Operationalisation of the methodological quality assessment.

Criteria
Study population
1 Positive if the symptoms and diagnostic criteria of the disorder were described
2 Positive if at least 4 of the following 6 items were reported at baseline: g) age (mean and standard deviation or CI) h) sex (number and/or percentage) i) outcome measures. Values for pain and functional status (mean and standard deviation or CI) j) affected extremity (dominant- non dominant) k) duration of symptoms (mean and standard deviation or CI) l) job description
3 Positive if the total number of cases was ≥ 50
Interventions
4 Adequate description of the used intervention a) Positive if the type of intervention was described b) Positive if the application technique was described
5 Positive if co-interventions were avoided in the design
Study design
6 Positive if the study design was prospective
7 Loss to follow-up a) Positive if information was given about loss to follow-up/drop-outs b) Positive if 7a) is positive and there was less than 10% loss to follow-up/drop-outs c) Positive if 7a) is positive and sociodemographic/clinical information (e.g. age, sex, type of complaints/ disabilities/ participation problems or prognostic factors) was presented for completers and those lost to follow-up/drop-outs at the main moment of outcome measurement, or no drop-outs/loss to follow-up. It is important that sociodemographic and clinical information was given for completers and follow-up/drop-outs to compare reasons for patients being lost. Reasons have to be unrelated to the outcome of recovery (complaints and disabilities). Loss to follow-up/drop-outs: all patients of the assembled cohort minus the number of patients at the moment of health status measurement for the main outcome measure, divided by all patients of the assembled cohort.
8 Timing follow-up measurements a) Positive if outcome assessment occurred at the end of the intervention period within 12 months after the intervention b) Positive if outcome assessment occurred > 12 months after the intervention period.
Outcome measurements
9 Positive if at least 3 of the following 6 items were used as outcome measures: pain, overall improvement, functional status (muscle strength, range of motion), medical consumption, disability (lost days of work or return to work), satisfaction with treatment
10 Each event should be described and correctly attributed to the allocated treatment: If it was explicitly reported that 'no adverse effects have occurred' or the adverse effects were described, this item is positive.
11 The interval between the intervention and the (different) measurements was identical for all individual patients. To score positive this item must be mentioned and described.
12 Positive if one or more of the main outcome measures were reported in a standardized or valid way (for example by means of a questionnaire, a diary or an objective outcome measure such as registration of lost days of work or medication use in the patient- chart of general practitioners).
13 Positive if frequencies, or percentages of mean (and standard deviation/CI), or median (and Interquartile Range) were reported for one or more of the main outcome measures for the most important follow-up measurements.
Analysis
14 Positive if 7a) was positive and all patients were reported/analyzed for the most important outcome moments of effect measurement (minus missing values) irrespective of non-compliance and co-interventions.
15 Positive if both point estimates and measures of variance are presented. Point estimates are: means, medians, modes etc. Measures of variance are: standard deviations, 95% confidence intervals, etc

Chapter 6

Is the disability of arm, shoulder and hand questionnaire (DASH) also valid and responsive in patients with neck complaints?



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Submitted

Abstract

Background: The DASH has shown to be a valid and responsive questionnaire to evaluate disability in patients with arm, shoulder and hand complaints. However, patients with shoulder, arm, or hand complaints frequently report neck complaints as well. A valid and responsive questionnaire designed for the whole upper extremity including the neck would be very useful and practical in upper-extremity research. Therefore, in this study we evaluated whether the DASH is not only a valid and responsive instrument to measure patients with arm, shoulder and hand complaints, but also to evaluate patients with neck complaints.

Methods: 679 patients visiting their general practitioner with a new episode of non-traumatic complaints of the neck and upper extremity were evaluated by use of questionnaires at baseline and at 6-months follow-up. Four (sub)groups (most complaints in arm-shoulder-hand, arm-shoulder-hand-neck, neck-shoulder, and neck) were formulated. Disability (DASH), general health (SF-12 (physical and mental component)), severity, and persistence of complaints were assessed. Construct validity and responsiveness were studied by testing 14 predefined hypotheses based on correlations, responsive ratios, and floor and ceiling effects.

Results: Correlations between the DASH and the other measures within the four (sub)groups at baseline (construct validity) and for the change scores at 6-months follow-up (responsiveness) were found adequate; responsiveness ratios in all of the four (sub)groups were classified as good. No floor and ceiling effects were found. All hypotheses could be accepted.

Conclusions: This study has shown sufficient validity and responsiveness of the DASH for use in patients with non-traumatic neck complaints.

Introduction

Many questionnaires for upper-extremity musculoskeletal disorders have been developed to evaluate functional status or the impact of health on performance in daily living.
In 1999, Davis et al.[1] concluded that questionnaires designed for the whole extremity could provide a more practical and still valid measure of upper-extremity disability. Items in existing specific upper-extremity questionnaires may also be relevant to other regions and conditions. This finding is in agreement with kinesiologic and biomechanic theories that the upper extremity acts as a single functional unit.[1] Moreover, use of one questionnaire in research on upper-extremity disorders makes it easier to compare the results of various scientific studies.

The Disabilities of the Arm, Shoulder and Hand questionnaire (DASH), developed as a regional outcome measure that conceptualizes the upper extremity as a single functional unit[2] addressed this viewpoint. It measures the impact on function of a variety of musculoskeletal disorders of the upper extremity and can be used for patients with any single or multiple disorders in arm, shoulder and hand.[3]
After this self-reported 30-item questionnaire was developed, many authors reported positive results with regard to the validity, reliability and/or responsiveness in patients with upper-extremity conditions: e.g. in a specific region such as the elbow[4] or the shoulder[5], for a specific disease such as the carpal tunnel syndrome[6] or a specific trauma such as the Colles' Fracture[7], and also for a variety of conditions in the upper extremity.[3,8-10] In brief, the DASH can be seen as an useful instrument to assess disorders affecting the arm, shoulder and hand whereby function and symptoms can be assessed in one combined scale.
However, patients with shoulder, arm, or hand complaints frequently report neck complaints as well. For example, in a study that reported data on patients with chronic conditions in the neck and upper extremity in the Dutch open population, 53.7% reported complaints of the neck and also in other regions of the upper extremity and 19% reported singularly neck problems.[11]

A valid and responsive questionnaire designed for the whole upper extremity including the neck would be very useful and practical in upper-extremity research. The DASH has shown to be a reproducible, valid and responsive measurement for the arm, shoulder and hand region and may contribute to this statement if the instrument is also a valid and responsive instrument for use in patients with complaints in the neck. Therefore, in this study we evaluated the validity and responsiveness of the DASH in patients with non-traumatic neck complaints.

Method

Design

The study population consisted of participants of an observational prospective cohort study of patients with a new episode of musculoskeletal complaints in the arm, neck and/or shoulder presented in Dutch general practices from September 2001 to December 2002. In total, 36 general practitioners (GPs) from 21 practices recruited the patients. An episode was considered 'new' if the patient had not presented to the GP with the same complaint during the preceding 6 months. At baseline and at 6-months follow-up, data were collected using self-administered questionnaires. Informed written consent was obtained from all participants. The Medical Ethics Committee of the Erasmus Medical Center approved the study.

Study population

Patients who visited their GP with musculoskeletal complaints of the arm, neck and/or shoulder were eligible for inclusion if they met the following criteria: (1) a new complaint or new episode of complaints of the neck, shoulder, upper arm, elbow, forearm, wrist of hand; (2) age 18-64 years, and (3) adequate understanding of the Dutch language. Patients with complaints due to an acute trauma, amputation, prosthesis, malignancy, a hereditary defect, previously diagnosed existing systemic and/or generalised neurological disorder were excluded. Patients were also excluded when they reported to be recovered at the time of filling in the baseline questionnaire.

Measurements

Procedures

On the initial visit to the GP the patients received study information, the informed consent form and the first questionnaire. The GP sent a fax, regarding the age, gender, diagnosis and expected prognosis of the complaint, to the investigators. After the researchers had received the baseline questionnaire and the inform consent within eight weeks, inclusion criteria were verified in the computerised medical records of the GP by an independent researcher, the patient could be included in the cohort. Six months later the follow-up questionnaire was sent from the research center.

Questionnaires

The baseline questionnaire contained questions on patient characteristics (age, gender, work participation and educational level) and characteristics concerning the duration and location of the complaints.

At baseline and at 6-months follow-up the patients reported functional limitations, general health, and the severity of the complaints during the previous week. Functional limitations

of the arm, neck and/or shoulder were measured using the DASH. As pointed out previously, the DASH items are designed to measure arm, shoulder and hand functional disability. Each item was scored on a 5-point Likert scale. Response scores were summed and transferred to a score ranging from 0 (no disability) to 100 (completely disabled).

General health was measured using the SF-12. The SF-12 contains a physical component summary scale (PCS-12) and a mental component summary scale (MCS-12), ranging from 0 to 100. Higher scores represent better health.[12] The categories of the first question on the SF-12 were recoded: 'good' and 'poor' were used for 'excellent' / 'very good' and 'fair' / 'poor' respectively.

The severity of the complaints was scored on an 11-point numerical rating scale, ranging from 0 (no complaints) to 10 (unbearable complaints).

At 6-months follow-up the patients scored the persistence (compared to baseline) of their complaints on a 7-point ordinal scale, ranging from 1 (worse than ever) to 7 (completely recovered).

Statistic analyses

To assess the validity and responsiveness of the DASH for non-traumatic complaints of the upper extremity and neck we first formulated four (sub)groups: 1) most complaints in the arm, shoulder and/or hand (A-S-H); 2) most complaints in the arm, shoulder, hand and/or neck (A-S-H + N); 3) most complaints in the neck and/or shoulder (N-S); and 4) most complaints in the neck (N). We formulated 14 hypotheses to evaluate the construct validity and the responsiveness of the DASH for each (sub)group. These hypotheses are described below.

Construct validity

Construct validity refers to the extent to which scores on a particular instrument relate to other measures in a manner that is consistent with theoretically-derived hypotheses concerning the constructs that are measured.[13] Construct validity is assessed if no 'gold standard' for measuring the domain of interest is available. No gold standard is available for measuring the disability of neck and upper-extremity disorders.

For each subgroup the Pearson's correlations for the total scores of the DASH, and the physical and mental component of the SF-12 were examined at baseline. It was hypothesized that all the correlations were positive and that the correlations were higher with the physical health component scores than with the mental component of the SF-12 (Figure 1, hypotheses 1 to 3). Furthermore, we hypothesized that the Pearson's correlations of the DASH with the physical component of the SF-12 was above 0.5 (Figure 1, hypothesis 4). Moreover, we hypothesized that the DASH and the severity of the complaints during the previous week had a positive correlation (Figure 1, hypothesis 5). Because the DASH was developed and tested to measure complaints in the arm, shoulder and hand, we also hypothesized that the correlations of the subgroups would not differ more than 15% (i.e. on a scale from 0 to 1

(-1 to 0) ± 0.15) among the correlations found for those affected by arm, shoulder and hand complaints (Figure 1, hypothesis 6). Construct validity was considered sufficient when the results were in correspondence with the hypotheses 1 to 6 (Figure 1).

Floor and ceiling effects

The presence of floor and ceiling effects may influence the responsiveness of an instrument to detect clinically relevant change.[14] To evaluate the floor and ceilings effects of the DASH, we hypothesized that no more than 30% of the respondents in each (sub)group should have the maximum or minimum score on the DASH (Figure 1, hypothesis 7).[15]

Responsiveness

Responsiveness refers to an instrument's ability to detect important change over time in the concept being measured.[16] There is no consensus on the optimal method to assess responsiveness. First, we choose to study the measurement error of the DASH within the four (sub)groups. Therefore, we defined responsiveness as the ability of the DASH to discriminate between clinically stable and improved subjects as suggested by Guyatt et al.[17] The Guyatt Responsiveness Index is defined as the ratio of the average change in patients identified as improved divided by the standard deviation of the change in patients identified as remaining stable:

$$\textbf{responsiveness ratio} = \frac{\text{mean change score in clinically improved patients}}{\text{variability (SD) of change scores in clinically stable patients}}$$

In the absence of a 'gold standard' as an external criterion for clinical stability, we used the self-reported 7-point ordinal scale for persistence of complaints (at 6 months compared to baseline) to identify clinically improved and stable patients. The scores 'much improved' or 'completely recovered' were considered as recovered. The scores 'little improvement', 'no change' and 'slightly worse' were considered as stable. We used the average change in score on the DASH after 6 months compared to the score at baseline.

If the responsiveness ratio is larger than 1, the mean change score in clinically improved patients exceeds the measurement error and the instrument may be considered to be responsive, to an extent that is proportional to the magnitude of the responsiveness ratio (Figure 1, hypothesis 8).[18]

Second, we assessed the Pearsons's correlation of the change scores to investigate whether this instrument really measures changes within the patients. Therefore, similar to the hypotheses for the construct validity, we formulated hypotheses for the change scores (Figure 1, hypotheses 9 to 14), i.e. the 6-months follow-up score minus the baseline score, because we expected similar correlation coefficients on the change scores of the DASH, SF-12 (physical and mental component) and the severity of the complaints during the previous week.

Responsiveness was considered sufficient when the results were in correspondence with hypotheses 8 to 14 (Figure 1).

Figure 1 Hypotheses for the validation of the DASH for each (sub)group

Hypotheses	
Construct validity	
1.	The total score of the DASH has a positive correlation with the physical component of the SF-12
2.	The total score of the DASH has a positive correlation with the mental component of the SF-12
3.	The total score of the DASH has a stronger correlation with the physical component of the SF-12 than with the mental component of the SF-12
4.	The correlation between the DASH score and the physical component of the SF-12 is at least 0.5
5.	The DASH score correlates has a positive correlation with the severity of the complaints during the previous week
6.	The correlation between the DASH score in the ASH group and the score in the other subgroup does not differ more than 15% (± 0.15)
Floor and ceiling effects	
7.	No more than 30% of the respondents should have the maximum or minimum score on the DASH
Responsiveness	
8.	The Guyatt's responsiveness ratio is larger than 1
9.	The change score on the DASH has a positive correlation with the change score on the physical component of the SF-12
10.	The change score on the DASH has a positive correlation with the change score on the mental component of the SF-12
11.	The correlation between the change score on the DASH and the change score on the physical component of the SF-12 is stronger than the correlation between the change score on the DASH and the change score on the mental component of the SF-12
12.	The correlation between the change score on the DASH and the change score on the physical component of the SF-12 is at least 0.5
13.	There is a positive correlation between the change on the DASH and the change on severity of the complaints during the last week
14.	The correlation between the change score on the DASH in the ASH group and the change score in the other subgroup does not differ more than 15% (± 0.15)

Results

Patient characteristics

In total 798 patients met our inclusion criteria and 682 (85.5%) entered the cohort after they returned the completed questionnaire and informed consent. Of these, 679 patients, 280 men and 399 women, with a mean age of 44.4 years (range, 18-64 years, SD=11.4) completed the DASH questionnaire and responded to a sufficient number of items for their scores to be calculated. Complaints in the neck-shoulder region were the most reported (72%). Table 1 presents the characteristics of the four (sub)groups of patients.

Table 1 Patient characteristics at baseline

	A-S-H	A-S-H+N	N-S	N
	n=588	n=679	n=489	n=213
Men, n (%)	252 (42.9)	280 (41.2)	204 (41.7)	68 (31.9)
Age, mean (SD)	44.9 (11.0)	44.4 (11.4)	44.0 (11.6)	42.3 (12.0)
Duration of current complaints, n (%)*				
0-3 weeks	153 (26.0)	179 (26.4)	136 (27.8)	49 (23.0)
3-6 weeks	141 (24.0)	162 (23.9)	116 (23.7)	53 (24.9)
6 weeks – 3 months	110 (18.7)	115 (16.9)	70 (14.3)	20 (9.4)
3-6 months	42 (7.1)	49 (7.2)	30 (6.1)	16 (7.5)
> 6 months	141 (24.0)	173 (25.5)	136 (27.8)	75 (35.2)
Recurrent complaints, n (%)	151 (25.7)	189 (27.8)	156 (31.9)	85 (39.9)
Work participation, n (%)				
Employed**	457 (77.7)	531 (78.2)	385 (78.7)	170 (79.8)
Sick leave***				
None	352 (77.0)	402 (75.7)	280 (72.7)	113 (66.5)
Once	73 (16.0)	90 (16.9)	74 (19.2)	37 (21.8)
2-5 times	23 (5.0)	29 (5.5)	24 (6.2)	17 (10.0)
> 5 times	6 (1.3)	6 (1.1)	4 (1.0)	2 (1.1)
missing	3 (0.7)	4 (0.8)	3 (0.8)	1 (0.6)
Education level, n (%)*				
Elementary school	35 (6.0)	40 (5.9)	33 (6.7)	14 (6.6)
Secondary school	389 (66.2)	448 (66.0)	328 (67.1)	141 (66.3)
Higher education/university	163 (23.1)	190 (0.1)	127 (26.0)	58 (27.2)

Abbreviations: SD: standard deviation; n: number; n.a: not applicable; A-S-H: most complaints in the arm, shoulder and/or hand; A-S-H+N: most complaints in the arm, shoulder, hand and/or neck; N-S: most complaints in the neck and/or shoulder; N: most complaints in the neck

* One missing

** Employed defined as having a paid job

*** Sick leave (during the last 6 months) due to upper-extremity complaints of those with a paid job

Descriptive statistics of the DASH

Mean scores including the standard deviations (SD) for the DASH at baseline, at 6-months follow-up and the change scores within the four (sub)groups are presented in Table 2.

Construct validity

The Pearson's correlations used to evaluate the construct validity are shown in Table 3. Correlations of the DASH scores within each (sub)group was the highest with the physical component of the SF-12. The correlations with the mental component of the SF-12 were relatively low. The DASH scores correlated moderately with the severity of the complaints

Table 2 Descriptive statistics of the DASH within the four (sub)groups

	Baseline			6-months follow-up			Change scores*		
	mean	SD	n	mean	SD	n	mean	SD	n
A-S-H	37.6	19.1	588	19.3	18.4	527	18.5	18.7	526
A-S-H+N	36.8	18.9	679	18.9	18.3	610	18.0	18.5	609
N-S	37.5	19.4	489	20.1	18.7	440	17.5	18.9	439
N	37.4	18.5	213	21.7	19.3	189	15.5	17.5	189

Abbreviations: SD: standard deviation; n: number; A-S-H: most complaints in the arm, shoulder and/or hand; A-S-H+N: most complaints in the arm, shoulder, hand and/or neck; N-S: most complaints in the neck and/or shoulder; N: most complaints in the neck.

* Change scores calculated as 6-months follow-up scores minus baseline scores

Table 3 Pearson correlations between the DASH scores and the other measurements (SF-12 scores and severity), at baseline

	SF-12 physical component	SF-12 mental component	Severity**
A-S-H	.624*	.147*	.551*
A-S-H+N	.612*	.156*	.524*
N-S	.626*	.194*	.501*
N	.617*	.266*	.442*
± 15% with respect to the score in the A-S-H group ***	.474 ⇔ .774	-.003 ⇔ .297	.401 ⇔ .701

Abbreviations: A-S-H: most complaints in the arm, shoulder and/or hand; A-S-H+N: most complaints in the arm, shoulder, hand and/or neck; N-S: most complaints in the neck and/or shoulder; N: most complaints in the neck.

* Correlations significant at the .01 level (1-tailed)

** Severity of the complaints during the last week

*** i.e. ± .15

during the previous week. Overall, all hypotheses formulated to study the construct validity (hypotheses 1 to 6) can be accepted. Therefore, it can be concluded that the construct validity of the DASH is sufficient to study patients with non-traumatic neck complaints.

Floor or ceiling effects

No maximum and minimal scores were found in one of the (sub)groups. Therefore, there was no evidence of any floor or ceiling effect for the DASH score in the four (sub)groups and hypothesis 7 can be accepted.

Responsiveness

The responsiveness ratios of the four (sub)groups are reported in Table 4. The responsiveness ratios in all of the four (sub)groups were over 1 and can be classified as adequate, consequently, hypothesis 8 can be accepted.

Table 5 gives the Pearson's correlations of the change scores of the four (sub)groups. The correlations of the change scores of the DASH with the change scores of the physical component of the SF-12 (range .520-.563) and with the change scores of the severity of the

complaints during the previous week (range .591-.631) were high. The correlations with the change scores of the mental component of the SF-12 were very low (range .032 to .130). Overall, all the hypotheses (8 to 14) that we postulated for these correlations can be accepted and we can conclude that the responsiveness of the DASH is sufficient to study patients with neck complaints.

Table 4 Responsiveness ratios

	Change score of the DASH		Responsiveness Ratio*
	mean	SD	
A-S-H			
Improved (n=283)	27.38	17.47	27.4 / 13.6 = 2.01
Stable (n=225)	8.53	13.60	
A-S-H+N			
Improved (n=328)	26.67	17.15	26.7 / 14.0 = 1.91
Stable (n=262)	8.28	13.97	
N-S			
Improved (n=226)	27.27	17.82	27.2 / 13.4 = 2.04
Stable (n=199)	7.66	13.36	
N			
Improved (n=85)	25.96	15.33	26.0 / 14.1 = 1.85
Stable (n=97)	7.83	14.06	

Abbreviations: SD: standard deviation; n: number; A-S-H: most complaints in the arm, shoulder and/or hand; A-S-H+N: most complaints in the arm, shoulder, hand and/or neck; N-S: most complaints in the neck and/or shoulder; N: most complaints in the neck.

* Responsiveness ratio formulated as the mean change score in improved patients divided by the variability (SD) of the change score in stable patients

Table 5 Pearson correlations of the change scores (6-months follow-up minus baseline) of the DASH and the other measurements

	SF-12 physical component	SF-12 mental component	Severity***
A-S-H	.539**	.032	.631**
A-S-H+N	.536**	.054	.611**
S-N	.563**	.071	.612**
N	.520**	.130*	.591**
15% marge**** A-S-H	.389 ⇔ .689	-.118 ⇔ .182	.481 ⇔ .781

Abbreviations: A-S-H: most complaints in the arm, shoulder and/or hand; A-S-H+N: most complaints in the arm, shoulder, hand and/or neck; N-S: most complaints in the neck and/or shoulder; N: most complaints in the neck.

* Correlations significant at the .01 level (1-tailed)

** Correlations significant at the .05 level (1-tailed)

*** Severity of the complaints during the last week

**** i.e. ± .15

Discussion

Validity, reproducibility and responsiveness of the DASH has been found adequate for measuring disability in patients with complaints in the arm, shoulder and hand. In this paper we evaluated whether the DASH is also valid and responsive in patients with neck complaints. We formulated 4 (sub)groups (most complaints in arm-shoulder-hand, arm-shoulder-hand-neck, neck-shoulder, and neck) and evaluated the validity and responsiveness of each (sub)group by testing 14 predefined hypotheses. Finally, we could accept all hypotheses. Therefore, we conclude that the DASH is not only a valid and responsive instrument to measure arm, shoulder and hand complaints, but also has shown sufficient validity and responsiveness in patients with non-traumatic neck complaints. As far as we know, this is the first study in which the DASH is evaluated for patients with neck complaints.

Four (sub)groups (most complaints in arm-shoulder-hand, arm-shoulder-hand-neck, neck-shoulder, and neck) were formulated. We compared the results of the group with complaints in the arm-shoulder-hand region with the results of the three other subgroups, because the validity and responsiveness of the DASH for patients with complaints in the arm-shoulder-hand is known to be acceptable. Because we stated that one questionnaire for the measurement of disability of the whole upper extremity including the neck is needed, we compared the results of the group with most complaints in the arm-shoulder-hand with those with complaints in the arm-shoulder-hand-neck. Because it is difficult to discriminate between neck and shoulder complaints, we compared not only a subgroup of patients who reported most complaints in the neck, but also a subgroup of patients who reported most complaints in the neck or the shoulder with the group with complaints in the arm-shoulder-hand.

One of the questionnaires developed to measure disability of patients with neck complaints is the Neck Disability Index (NDI). The NDI is a one-dimensional questionnaire specifically measuring physical aspects of neck pain.[19,20] Good results on validity and reproducibility of the NDI have been reported for different groups of patients.[20,21] Vos et al.[22] studied the responsiveness of the NDI in patients with acute neck pain in general practice; in that study the NDI scored a responsiveness ratio of 1.82 after one week. This is comparable with our results of the DASH for patients with incident neck complaints recruited in general practice. Future research to compare the DASH and the NDI in patients with neck complaints is recommended.

Responsiveness of the DASH had been evaluated and reported in several ways. Most studies reported a standardized response mean (SRM).[3,6,23-25] The SRM is the mean change in scores from baseline to follow-up divided by the SD of changes. The SRM can be used to study the effect of interventions. In our cohort study we followed patients with upper-extremity

disorders who visited their GP over time. Some patients were treated and others were not. Because of this, we may expect more variation in disability of the upper extremity over time. Therefore, calculating the SRM in our study was not the best option because the denominator in the formula to calculate the SRM will be high and, as a result of this, the SRM will be lower.

In our study, Pearson correlations at baseline and for the change scores between the DASH and the SF-12 mental component are higher if the neck is involved. This was also the case if the DASH scores were compared to the severity scores. For the subgroup of those with most complaints in the neck the correlations are the highest. A possible explanation for this may be the association between mental stress and neck complaints.[26,27] This association may be higher than in those with most pain in other regions of the upper extremity and may also have its impact on severity.

This study has several limitations. First, the hypotheses we formulated are arbitrary, although they were formulated in advance. Second, a minority of our patients reported most complaints in the arm or hand, whereas a majority of patients reported most complaints in the shoulder and neck. In this way, the results of those with neck and shoulder pain influenced the results in the four (sub)groups the most. Third, our study was limited by the fact that we could not compare our results with available 'neck questionnaires', such as the NDI. Furthermore, in this study we included patients who visited their GP with a new complaint or new episode of complaints of the arm, neck and/or shoulder. Course, prognosis and disability in first-time consulters can differ from those with chronic neck complaints, and may also differ from the selected population of patients with neck pain presented in secondary care settings. Therefore, the validity and responsiveness of the DASH for patients with chronic neck complaints may differ from our results.

In conclusion, in research on upper-extremity disorders, a valid and responsive questionnaire should be available that is suitable to assess the entire region, including the neck. Thereby, different studies on the impact, the prognosis of upper-extremity and neck disorders or its treatment can be compared more easily. The results of this study can contribute to this statement, because this study has shown sufficient validity and responsiveness of the DASH for use in patients with non-traumatic neck complaints.
However, more studies are needed to compare the DASH with other neck-specific questionnaires and to assess reliability, validity and responsiveness of the DASH to measure disability in patients with different types of neck complaints.

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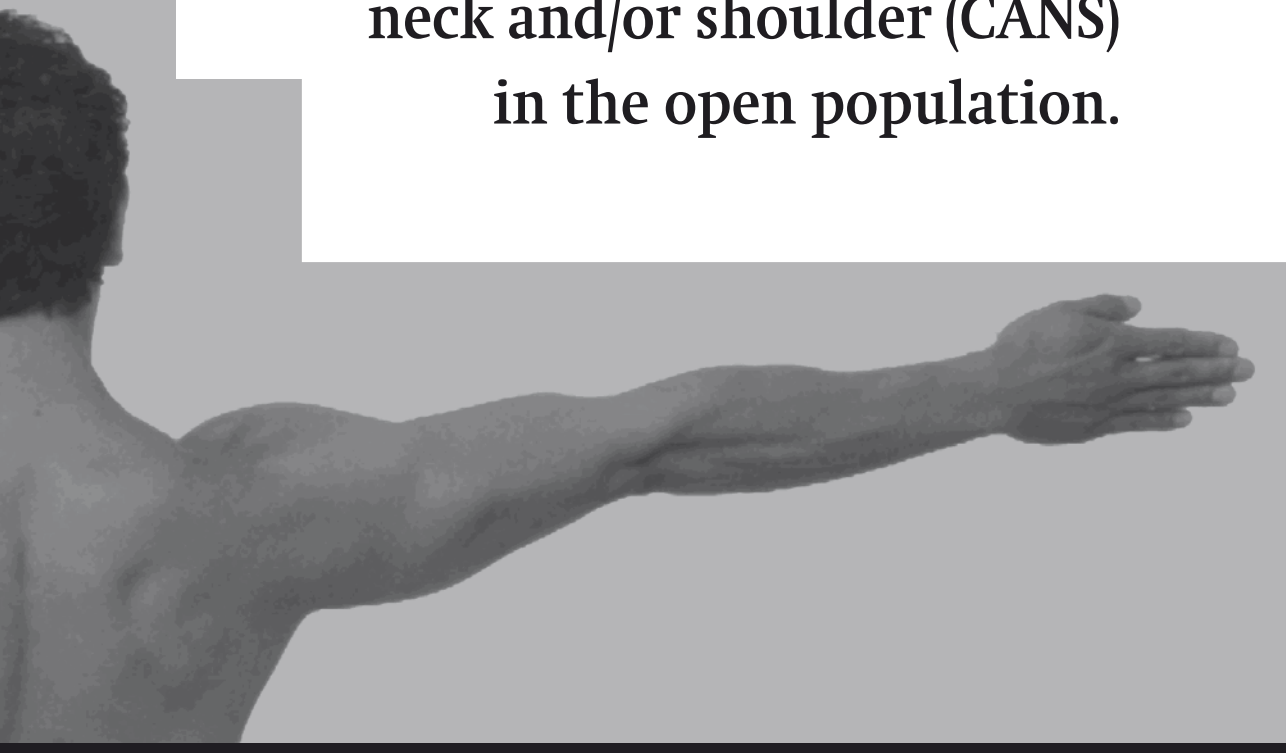
Chapter 7

Prevalence and characteristics of complaints of the arm, neck and/or shoulder (CANS) in the open population.



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Submitted

Abstract

Background: Accurate prevalence rates and consequences of CANS are needed to serve as a basis for etiologic studies and healthcare evaluation. Therefore, we studied the prevalence of UEDs (disorders of the upper extremity and neck as a total) and CANS (Complaints of the Arm, Neck and/or Shoulder not caused by acute trauma or any systemic disease) as defined in the CANS model in the open population and assessed socio-demographic and health characteristics of chronic complaints.

Methods: Data were obtained from the DMC3-study, a Dutch questionnaire survey on musculoskeletal conditions (>25 years, n = 3664). Data on four anatomic sites were assessed: neck, shoulder, elbow and wrist. Various health characteristics were measured including the SF-36. Rectangle diagrams were used to illustrate co-occurrence of pain in the four anatomic sites.

Results: The 12-months prevalence of CANS was 36.8%, the point prevalence was 26.4%, and 19.0% reported chronic CANS. Women, persons aged 45-64 years, with the lowest education level, and those not working were the most affected. Within those with UEDs, around 25% of cases was caused by an acute trauma or by some systemic disease. Of those with chronic CANS, 58% reported use of healthcare. Healthcare users scored worse on general health, limitations in daily living, pain, and sickness absence than non-healthcare users; over 43% reported complaints in more than one anatomic site.

Conclusions: UEDs and CANS frequently occur in the open population. Excluding acute traumata and systemic diseases reduced the prevalence of CANS and resulted in a relatively healthier population. A compound definition of CANS seems indicated because of the large overlap of affected anatomic sites.

Introduction

Besides personal distress and suffering, upper-extremity musculoskeletal disorders have a high economic and social impact worldwide. It is difficult to estimate the precise extent of the problem, because reported prevalence rates differ substantially between studies. For upper-extremity musculoskeletal disorders the reported prevalence rates range from 1.6-53% and the 12-months prevalence rates range from 2.3-41% depending on the setting, definition and classification used.[1] Different terminology and classification systems have impeded critical scientific interchange within the medical community.

To support and compare scientific research and to increase multidisciplinary cooperation within healthcare, a universal and multidisciplinary accepted classification system for upper-extremity musculoskeletal disorders, the CANS (complaints of the arm, neck and/or shoulder) model, was developed[2] by performing a Delphi consensus strategy. All relevant medical and paramedical disciplines dealing with the treatment of patients with CANS and their national professional associations in the Netherlands were involved in this project. The results of the consensus project, the CANS model, describes the terminology and classification of CANS. CANS was defined as "Musculoskeletal complaints of arm, neck and/or shoulder not caused by acute trauma or by any systemic disease". It was the first time that an expert-panel in this field, which included 11 medical and paramedical disciplines cooperated on a national level, and entered into debate on the terminology and classification of a large number of conditions of the upper extremity. In total they discussed 37 conditions that met the definition of CANS. Finally, within CANS, 23 conditions were classified as specific disorders. All other conditions were labelled as non-specific CANS.

Accurate prevalence estimates of CANS are needed to serve as a basis for etiologic studies and healthcare evaluation, and to assess the consequences of the complaints in the open population. To provide insight into the prevalence of CANS according to this new proposed model we re-analyzed data from a population-based study to address the following questions:

(1) Among those with chronic CANS, what is the prevalence of specific and non-specific CANS in total and based on socio-demographic characteristics?
(2) Among those with chronic CANS, who makes contact with healthcare and who does not?
(3) Among those with chronic CANS, is there a relationship between the health and sickness absence characteristics and the use/non-use of healthcare?
(4) What amount of overlap of pain locations can be visualized within chronic CANS?

Methods

To provide insight in the prevalence rates and consequences of CANS we used baseline data of a large population-based study, the DMC_3-study (Dutch population-based Musculoskeletal Complaints and Consequences Cohort study).

The DMC_3-study

The DMC_3-study is a national health survey on musculoskeletal conditions using postal questionnaires. An age-gender stratified sample of 8,000 persons of the Dutch population aged 25 years and older was approached for this study and 3,664 persons (46.9%) returned the questionnaire. Of the respondents, 50.9% were women and 47.0%, 34.6% and 18.4% were aged 25-44, 45-64 and 65+ years respectively. The study was carried out in 1998-1999 by the National Institute of Public Health and the Environmental in co-operation with Statistics Netherland. The exact methods of this survey have been described extensively elsewhere.[3]

The questionnaire

The questionnaire of the DMC_3-study consisted of general questions and health questions about musculoskeletal conditions in five anatomic areas: 1) neck, shoulder or higher part of the back; 2) elbow or wrist/hand; 3) the lower part of the back; 4) hip or knee; and 5) ankle and foot. Each anatomic area included one, two or three anatomic sites of which data were collected separately. The questions about each area started with the following question: "Did you have pain in <anatomical area> during the past 12 months?" With a positive reply, the person was asked to answer all the questions concerning the relevant area focusing on: whether or not the pain still exists, duration and severity of the pain, consequences of the pain for healthcare utilization, general health during the 12 months before the survey including contacts with general practitioner (GP), medical specialist or physiotherapist, work status and sickness absence. The working population was defined as respondents who reported to have a paid job.

General health was measured using the SF-36. The SF-36 contains 36 items from which total scores on 8 subscales can be calculated. The total score ranges from 0 to 100. Higher scores represent better health.[4]

Study population

For this survey we used the baseline data regarding upper-extremity complaints. These data included four anatomic sites: neck, shoulder, elbow, and wrist/hand. We studied persons with musculoskeletal upper-extremity disorders and neck as a total (UEDs) as well as according to the definition of CANS in the CANS model. In order to study CANS we excluded patients with complaints in these areas caused by an acute trauma (such as a fracture), or a systemic disease (such as rheumatoid arthritis).

Statistical analysis

The following prevalence estimates were calculated for UEDs and as defined in the CANS model: 12-months prevalence (complaints during the last 12 months), point prevalence (pain at the moment of investigation), and prevalence of chronic pain (pain at baseline and lasting more than 3 months in the last 12 months). In order to present estimations for the Dutch population, weighting factors were used to make the distribution by age and gender equal to that found in the Netherlands in 1998.

Frequencies (percentages) of those with chronic complaints were used to examine socio-demographic characteristics and health characteristics in persons with UEDs and in persons with CANS. In the same way, we examined differences in health characteristics in healthcare users compared to non-healthcare users in patients with chronic CANS.

Of those with chronic CANS prevalence rates of the neck, shoulder, elbow and wrist/hand regions were studied separately. Rectangle diagrams (Venn diagrams) were used to illustrate the co-occurrence of pain in the four anatomic sites. Rectangles are drawn with an area proportional to the frequency of categories, and the rectangles are positioned to overlap each other so that the areas of overlap are in proportion to the joint frequencies of the characteristics.[5] The overlap in anatomic sites of complaints in those with CANS at the moment of the study and in those with CANS during the last year was also studied.

Results

Prevalence rates

Table 1 reports prevalence rates of UEDs as well as prevalence rates of CANS according its definition in the CANS model (i.e. excluding acute traumata and systemic diseases). Estimates show that almost half (48%) of the Dutch population aged 25 years and older reported UEDs in the last 12 months, and 36.8% reported CANS.

From the total sample of the responders 36.6% reported current UEDs while 27.2% of those with UEDs met the criteria for chronic pain; for CANS these percentages were 26.4% and 19.0%, respectively. Excluding those with systemic diseases and complaints due to an acute trauma (as instructed by the CANS model) resulted in a lowering of the CANS prevalence by 8-11 percentage points. In other words, within those with musculoskeletal UEDs in the open Dutch population, 24-30% of cases was caused by an acute trauma or by some systemic disease.

Socio-demographic characteristics

Differences in socio-demographic characteristics between those with chronic UEDs and those with chronic CANS are given in Table 2.

Table 1 Self-reported prevalence rates of complaints in the upper extremity and neck (number and % of total population) in total (UEDs) and according to the CANS model, DMC3-study, n=3664.

	UEDs		Systemic disease among those with complaints of upper extremity and neck		Acute trauma mentioned as cause of complaints of upper extremity and neck		CANS Systemic disease and/or acute trauma excluded	
	n	% of total population	n	% of total population	n	% of total population	n	% of total population
12-months prevalence, number (%)	1762	(48.1)	274	(7.5)	203	(5.5)	1349	(36.8)
Point prevalence, number (%)	1341	(36.6)	254	(6.9)	177	(4.8)	969	(26.4)
Prevalence of chronic complaints, number (%)	996	(27.2)	210	(5.7)	137	(3.7)	697	(19.0)

Table 2 Socio-demographic characteristics of persons with chronic complaints of the upper extremity and neck in total (UEDs, n=996) and according to the CANS model (n=697), the DMC3-study (n=3664).

		Chronic UEDs		Chronic CANS			
		Total n=996		Total n=697		Healthcare users** n=404	
		n	% of total population	n	% of total population	n	% of total population
Gender, number (%)	Men	373	(10.2)	283	(7.7)	149	(4.1)
	Women	623	(17.0)	413	(11.3)	255	(7.0)
Age in years, number (%)	25-44	256	(7.0)	200	(5.5)	110	(3.0)
	45-64	450	(12.3)	318	(8.7)	193	(5.3)
	65+	290	(7.9)	186	(5.1)	101	(2.8)
Work status*, number (%)	Working men	184	(5.0)	149	(4.1)	78	(2.1)
	Non-working men	71	(1.9)	54	(1.5)	34	(0.9)
	Working women	195	(5.3)	145	(4.0)	83	(2.3)
	Non-working women	256	(7.0)	170	(4.6)	108	(2.9)
Education, number (%)	Lowest	196	(5.3)	137	(3.7)	95	(2.6)
	Low	405	(11.1)	283	(7.7)	165	(4.5)
	High	221	(6.0)	153	(4.2)	88	(2.4)
	Highest	173	(4.7)	123	(3.4)	56	(1.5)

* Excluding those aged 65 years and over

** Healthcare user = contact with general practitioner, medical specialist or physiotherapist

Chronic UEDs as well as chronic CANS were more common in women, persons aged 45-64 years, the non-working population, and in those with a lower education level.
Of those with chronic CANS, 58% (n=404) reported use of healthcare in the last 12 months due to CANS. The socio-demographic characteristics of these persons are listed in Table 2. More women than men (7.0% versus 4.1% of the total open population) reported use of healthcare due to chronic CANS. Use of healthcare within those with chronic CANS is higher in the non-working population than in the working population. We found no gender differences in the non-working population. However, among the working population with chronic CANS, about 5% more women than men were healthcare users due to CANS.

Health characteristics and healthcare utilization

Those with chronic CANS scored higher (i.e. had better generic health) on eight dimensions of the SF-36 than those with UEDs (Table 3). Thus, the reported general health became better by excluding systemic diseases and acute traumas.
Table 3 shows different healthcare characteristics separately for the group of healthcare users and non-healthcare users with chronic CANS.
Of those with chronic CANS who sought medical care in the past 12 months, 81.1% visited their GP and more than half contacted a medical specialist (58.8%) or physiotherapist (54.1%). Healthcare users reported more continuous severe, mild pain and recurrent severe pain than non-healthcare users who reported more recurrent mild pain. Healthcare users reported more limitation in daily life due to chronic CANS (48.9%) than non-healthcare users (8.5%). Furthermore, the healthcare users reported more sickness absence due to chronic CANS than healthcare users; 90.7% of non-healthcare users reported no sickness absence at all due to CANS. Of the healthcare users 37.2% reported sickness absence due to CANS (versus 9.3% for non-healthcare users) and 12.4% reported sickness absence for more than 4 weeks.

CANS specified for anatomic sites

As shown in the rectangle diagram of chronic CANS (Figure 1) complaints of the shoulder (56.1%) were most common, followed by complaints of the neck (53.7%). The shoulder and neck complaints were for 19.9% and 19.1%, respectively, reported as single-sited complaints. Combined neck-shoulder complaints were presented in 23.0% of the persons.
In total, 56.8%, 29.3%, 6.7% and 3.7% of the persons with chronic CANS had complaints in 1, 2, 3 or 4 anatomic sites, respectively (Table 4). Table 4 shows that also in those with CANS at the moment of the study and in those with CANS during the last year a substantial part (46.0% and 55.4%, respectively) reported complaints in more than one anatomic site.

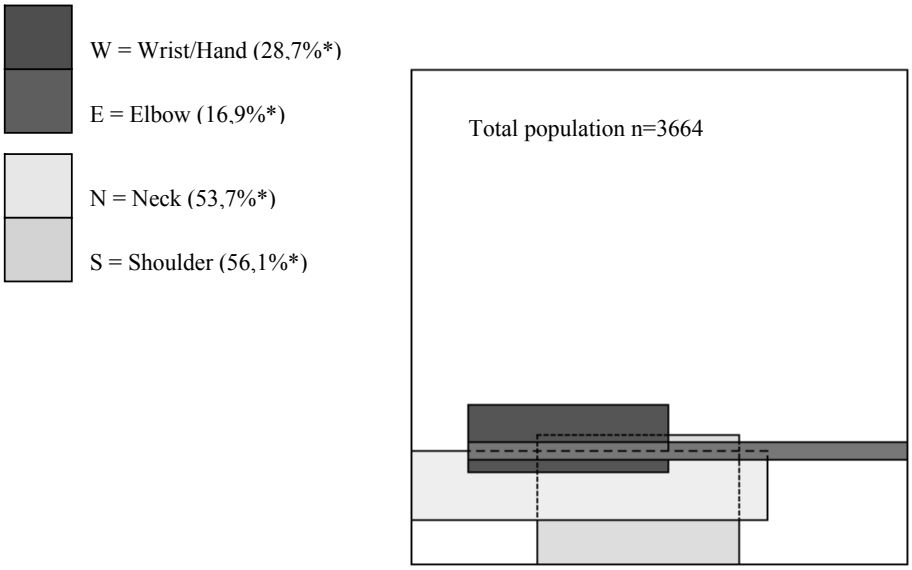
Table 3 Health characteristics of persons with chronic complaints of the upper extremity and neck in total (UEDs, n=996) and according the CANS model (n=697) and characteristics of healthcare users versus non healthcare users due to chronic CANS, the DMC3-study (n=3664).

		Chronic UEDs	Chronic CANS		
		Total n=996	Total n=697	Healthcare users* n=404	Non healthcare users n=293
General health, SF-36 (mean (sd))	Physical functioning	69.9 (26.6)	74.0 (25.8)	69.5 (27.1)	80.1 (22.5)
	Role-physical	59.5 (42.6)	64.9 (41.4)	56.2 (43.0)	76.4 (35.9)
	Bodily pain	66.5 (22.9)	70.1 (22.3)	63.9 (23.0)	78.7 (18.0)
	General health	59.8 (20.7)	61.5 (20.2)	57.9 (21.1)	66.5 (18.1)
	Vitality	58.2 (20.7)	59.9 (20.7)	57.0 (21.4)	63.9 (19.0)
	Social functioning	76.1 (25.7)	78.1 (25.4)	74.4 (26.7)	83.1 (22.7)
	Role-emotional	80.6 (35.9)	82.0 (35.0)	80.1 (37.2)	84.6 (31.9)
	Mental health	72.4 (18.5)	72.7 (18.7)	70.5 (19.2)	75.7 (17.5)
Utilization of healthcare (%)**	General practitioner	50.2	47.5	81.1	-
	Medical specialist	40.4	35.0	58.8	-
	Physiotherapist	36.5	31.8	54.1	-
Course of pain (%)	Continuous severe pain	5.4	3.9	6.5	0.4
	Continuous mild pain	38.2	36.4	39.1	32.8
	Recurrent severe pain	12.7	12.8	15.8	8.7
	Recurrent mild pain	36.4	39.4	30.1	52.3
	Non-recurrent	1.3	1.3	1.5	1.1
	Complaint varies	6.0	6.1	7.0	4.9
Limitation in daily life due to CANS (%)		38.3	32.1	48.9	8.5
		(n =358)	(n =282)	(n =153)	(n =129)
Work leave due to CANS (%)***	Never	72.4	75.5	62.8	90.7
	Less than 1 week	7.8	5.7	8.5	2.3
	1-4 weeks	7.5	6.0	11.1	0.0
	More than 4 weeks	7.8	7.5	12.4	1.6
	Not applicable	4.5	5.3	5.2	5.4

* healthcare user = contact with general practitioner, medical specialist or physiotherapist

** utilization of healthcare during the last year

*** 1 year; among those with paid work and age 25-64 years



* Percentages of persons with chronic CANS

Figure 1 Venn diagram of chronic CANS (n=697) in the DMC3-study

Table 4 Overlap of complaints in anatomic sites in persons with CANS.

		CANS		
		Point prevalence* n = 969 (no missings)	12-months prevalence** n = 1349 (no missings)	Prevalence of chronic complaints*** n = 697 (24 missings)
Complaints in:	one anatomic site, number (%)	523 (54.0)	601 (44.6)	396 (56.8)
	two anatomic sites, number (%)	322 (33.2)	516 (38.3)	204 (29.3)
	three anatomic sites, number (%)	82 (8.5)	142 (10.5)	47 (6.7)
	four anatomic sites, number (%)	42 (4.3)	90 (6.7)	26 (3.7)

* CANS at the moment of the study

** CANS during the last 12 months

*** CANS at baseline and lasting more than 3 months in the last 12 months

Discussion

This study estimates that in about 25% of the total population with UEDs, the complaint is due to an acute trauma or some systemic disease, resulting in an estimation of the prevalence of (specific and non-specific) CANS of 36.8% (12-months prevalence), 26.4% (point prevalence) and 19.0% (chronic CANS). Excluding those with acute trauma and systemic diseases implies excluding those with a relatively worse health status. Women, persons aged 45-64 years, those with the lowest education level and the non-working population were most affected by CANS or UEDs. In addition, this study shows that those with CANS who had contact with healthcare in the past year had relatively more severe pain, more disabilities, and more often sickness absence. Among those with CANS, almost half (>43%) reported complaints in more than one anatomical site.

A few methodological issues need to be discussed. First, the DMC_3-study was designed to provide insight into the prevalence of musculoskeletal health problems of different anatomic sites and was not specifically designed to study CANS according to the CANS model.
Because of this, the data of the DMC_3-study do not allow us to distinguish between specific and non-specific complaints in as much detail as described in the CANS model. Bongers reported in September 2003 that for two thirds of the persons with complaints in the arm, neck or shoulder no specific diagnosis could be made.[6]
Secondly, collecting data based on patient self-report and patient recall has certain drawbacks.[7,8] For example, sickness absence may be underreported due to socially desirable answers. However, for pain this method is the only source of information.
Furthermore, the DMC_3-study had a relatively high non-response rate (46.9%). In addition, there may be some over-reporting of those who sought medical care, because this has been found before in postal surveys.[9,10] Based on general characteristics from the population register, respondents and non-respondents did not differ significantly.[11] A specific study among the non-responders showed that the DMC_3-study gives a slight overestimation of pain prevalence.[3]

Bot et al.[12] studied the prevalence of neck and upper-extremity musculoskeletal disorders in general practice in the Netherlands. ICPC codes were used to provide insight in the occurrence of UEDs. In the Netherlands almost everyone has their own GP who is generally seen before being referred to a specialist or physiotherapist. Therefore, prevalence rates similar to ours could be expected. Bot et al.[12] reported that approximately 8% of all registered consulted their GP at least once for neck or upper-extremity symptoms during the last year. We found that 11% (n=404) of the total population used healthcare and 81.1% of these visited their GP. Thus, in the DMC_3-study 8.9% of the people with CANS visited their GP. Therefore, as expected, similar prevalence rates were found.

Our study showed that only 58% of those with chronic CANS used healthcare and that these healthcare users reported more pain, worse general health, more limitations in daily living, and more sickness absence due to CANS. Researchers should be aware of this selection bias when they recruit patients within healthcare.

Prevalence of CANS was higher in the non-working population than in the working population. Most studies suggest that the working population is at risk because of biomechanical risk factors at work, such as repetitiveness[13,14] and posture[15], or due to psychosocial work characteristics and increased stress symptoms such as high job demands and lack of control or social support.[14,16-18] The same factors, as well as other mechanisms, may play a role in the non-working population. For example, some persons might not work due to their complaints.[19] More research is needed to study the impact of this phenomenon and the role of risk factors in the non-working population.

Gender differences in those with chronic CANS were found in the working and the non-working population; women are more often affected than men. The differences in prevalence rates between the working and non-working population were higher among men.

Women are known to be more at risk for upper-extremity musculoskeletal disorders than men[20,21], but we can only speculate why the difference in prevalence rates for the working and non-working population is higher in men than in women. Perhaps the "healthy workers survivor effect" plays a role[19] and occurs more often in men than in women, or perhaps there is a stronger selection of healthy men than women into the workforce.[22]

Neck-shoulder complaints were the most frequently reported in combination with complaints in other upper-extremity sites. Andersson et al.[23], who studied chronic pain in the open Swedish population, also reported the combination with neck-shoulder pain as most frequent. Of our subjects suffering from CANS, a substantial part reported complaints in more than one anatomic area. Therefore, we believe that prevalence rates and studies for each anatomic area separately provide only a fragmented picture of complaints in the upper extremity.

In conclusion, UEDs as well as CANS are frequently occurring conditions in both the working and non-working population aged 25 years and older. Women, persons aged 45-64 years, with the lowest education level, and not working were the most affected. By excluding those with an acute trauma or any systemic disease, the population affected by upper-extremity complaints becomes more specific. Prevalence rates decreased with 23.5% (12-months), 27.9% (point) and 30.1% (chronic); those with a more delicate health, with more limitations in daily life, experiencing more continuous pain and reporting more sickness absence due to the complaints were filtered out.

A substantial part of the persons with chronic CANS (58%) are healthcare users. Particularly those who reported more pain, worse general health, more limitations in daily living, and

more sickness absence due to CANS, sought medical treatment. Many (> 43%) of those with CANS reported complaints in more than one anatomic site. More research is needed on epidemiology, treatment and prevention of CANS in both the working and the non-working population.

Acknowledgement

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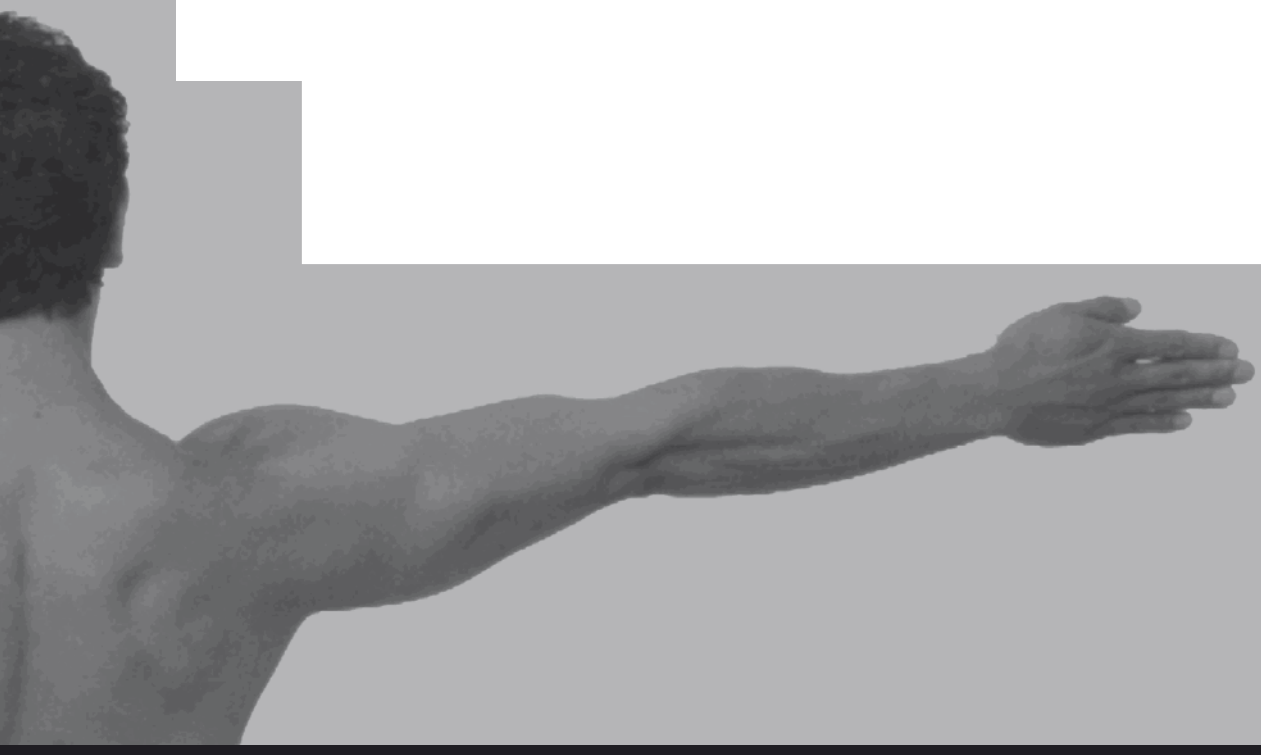
Chapter 8

General discussion



Different approaches regarding the terminology and classification have proven to be a major problem in the prevention, treatment and research on musculoskeletal disorders in general and upper-extremity disorders in particular. In this thesis we described the development of a new approach for the terminology and classification of musculoskeletal upper-extremity disorders, i.e. the CANS model.

History of classification and diagnosing

Diagnosis (from the Greek words dia = discriminate and gnosis = knowledge) is the process of identifying a disease by its signs, symptoms and results of various diagnostic procedures.
The term 'diagnostic criteria' encompasses the combination of symptoms obtained from the patient's history, physical examination and laboratory and/or imaging test results, which allows the physician to ascertain the diagnosis of the respective disease.
Classification can be described as a systematic arrangement into classes or groups.
The Greek philosopher Aristotle (384-322 B.C.E.) was the great classifier[1] and established one of the earliest classification schemes. Aristotle believed that the complexity of life could be divided into a natural order based on dichotomies, or polar opposites. For example, Aristotle divided animals into those with blood and those without blood, a classification that roughly corresponds to the division between vertebrates and invertebrates used in contemporary classification schemes. Sir George Knibbs, an Australian statistician, credited Francois Bossier de Lacrois (1706-1777), better known as Sauvages, with the first attempt to classify disease systematically.[2]

Classification and diagnosing of musculoskeletal upper-extremity disorders

A new approach for classification of upper-extremity disorders: the CANS model

In 2003 Van Eerd et al.[3] reviewed the scientific literature on diagnostic criteria for musculoskeletal upper-extremity disorder and found 27 different systems of labelling patients with upper-extremity disorders, no two of which were the same. Differences were found with regard to the criteria by which individual disorders were specified, the names by which they were identified, and the range of diagnoses that were distinguished. The absence of a universally accepted way of labelling or defining these conditions is one of the main reasons why substantial differences in prevalence rates have been reported (chapter 2 of this thesis).
We initiated a project with the aim to create a classification system for musculoskeletal upper-extremity disorders that could: 1) be generally accepted and multidisciplinary used, 2) classify in principal all upper-extremity conditions, and 3) is reported as a practical tool.

The project did not aim to create another set of diagnostic criteria for upper-extremity disorders, but to decide on an 'unambiguous language'. Unambiguous language is important to increase accurate and meaningful communication, to teach and share experiences of health care workers in a useful way, to compare information from various studies, and to stimulate appropriate decision making. After two years of intensive cooperation with experts in the field we succeeded in achieving consensus on the terminology and classification of upper-extremity disorders: the CANS model was born (chapter 2 of this thesis). CANS (complaints of the arm, neck and/or shoulder) is not a diagnosis in itself, but refers to a complexity of musculoskeletal symptoms in the upper extremity not caused by an acute trauma or by a systemic disease. The consensus was achieved by means of an invitational conference, followed by a Delphi consensus strategy – and not the other way around – so that the advantages of the Delphi procedure (anonymous and no bias through status, or dominant personality) were optimally applied.

As mentioned in the Introduction (chapter 1), in the project of both Harrington et al.[4] and Sluiter et al.[5] consensus on the diagnostic criteria for upper-extremity disorders was achieved in a final workshop which took place after a literature study and/or a Delphi consensus strategy. A Delphi consensus strategy is a proven method when no conclusive evidence is found in literature and experts' opinion is needed to achieve consensus.[6-8] It is difficult to ascertain in which way the final workshop in the consensus projects of Harrington et al.[4] and Sluiter et al.[5] contributed to the final criteria; the advantages offered by the Delphi procedure may have been reduced in this way. In the present consensus project, we started with a workshop which included invited experts from the various disciplines dealing with the subject on which consensus had to be achieved. The purpose of this workshop was to exchange ideas and expertise on the subject, and to create a starting point for the Delphi consensus strategy. In the final step (the Delphi consensus strategy) the topics on which consensus was achieved were discussed, and the results of this procedure were reported as the consensus.

Implementation of the CANS model

From the start of the consensus project we included all medical and paramedical disciplines involved in the treatment of patients with upper-extremity musculoskeletal disorders. Even if the classification is to be used in a research project in which no detailed clinical diagnoses are planned, there is still need for clinical approval to ensure understanding and relevance of the results. All experts in our consensus project were medical and paramedical health care professionals considered to be key persons in the field of upper-extremity disorders by their own professional discipline. In this way we created a number of 'ambassadors' who may facilitate the implementation of the CANS model in daily practice in the Netherlands.

In the Netherlands the CANS model is already included in the training and the post-graduate courses of medical and paramedical professionals. In fact, many professionals

are already familiar with the CANS model and its use in daily practice. Despite this, more time and specific implementation projects are needed to enable the model to become more widely accepted in the Netherlands. For example, an attempt should be made to introduce the model in (updates) of clinical guidelines on upper-extremity disorders for health care professionals.

Other projects on classification and diagnosing of upper-extremity disorders

During the two-years period in which the CANS model was developed, research groups in other countries also initiated projects with regard to the diagnosing of musculoskeletal upper-extremity disorders; as done by others[4,5] before them, they developed diagnostic criteria for a number of these conditions.

As mentioned earlier (chapter 1, the introduction), Harrington et al.[4] achieved consensus on eight specific upper-extremity disorders and one disorder called non-specific. Based on the findings of a literature review, in 2003 Walker-Bone et al.[9] added to the Harrington list diagnostic criteria for acromioclavicular joint dysfunction, subacromial bursitis and, osteoarthritis of the distal interphalangeal joints and the thumb.

Also in 2003, Helliwelll et al.[10] established core variables for diagnosing upper-extremity disorders based on multivariate modeling rather than on a consensus statement. In this way, variables are selected statistically by their ability to discriminate between different diagnoses. Table 1 presents the criteria of the Harrington list, the Sluiter list and the Helliwell list for the 23 specific disorders of the CANS model. The similarity between the criteria found by Helliwell et al.[10], the consensus approach of Harrington et al.[4], and the literature/consensus approach of Sluiter et al.[5] is striking. Future collaboration between the Dutch research groups and the research group of Harrington et al.[4] seems a reasonable next step to achieve international consensus on the classification and diagnosing of upper-extremity disorders. The CANS model and the results listed in Table 1 can be used as a meaningful starting point to initiate a new international Delphi consensus strategy.

Lack of a gold standard for diagnostics

A major problem in upper-extremity musculoskeletal research is the lack of an independent gold standard to diagnose the disorders[11], particularly when the pathology underlying a disorder is unknown or can not be reliably diagnosed.[12]

Epidemiologists have embraced the concept of diseases as objective natural phenomena that can be observed, classified and investigated, and most epidemiological textbooks include sections on assessment of the ‘accuracy’ of diagnostic tests. Test accuracy studies need a well-defined illness definition, and a clear-cut diagnostic gold standard or reference standard. However, in clinical reality illness definitions are often vague or a mere description

Table 1 Diagnostic criteria of the disorders mentioned in the CANS model according to the Harrington list, the Sluiter list and the Helliwell list

CANS model		Harrington list 4	Sluiter list 5 (symptoms and time rule)	Helliwell list 10
Hand/wrist				
1	Trigger finger	Not included	Not included	Not included
2	Raynaud's phenomenon		Peripheral neuropathy associated with exposure to hand-arm vibration: At least intermittent numbness in the fingers, with or without tingling AND History of exposure to hand-arm vibration preceding symptoms AND Symptoms present now or on at least 4 days during the last 7 days OR symptoms present on at least 4 days during at least 1 week in the last 12 months	
3	Dupuytren disease	Not included	Not included	Not included
4	De Quervain's disease	Pain over the radial styloid and tender swelling of the first extensor compartment and either pain reproduced by resisted thumb extension or positive Finkelstein test	Intermittent pain or tenderness localized over the radial side of the wrist; either may radiate proximally to the forearm or distally to the thumb AND Symptoms present now or on at least 4 days during the last 7 days OR symptoms present on at least 4 days during at least 1 week in the last 12 months	Tenosynovitis: pain on movement of tendon or swelling of tendon sheath or triggering/locking/nodule on tendon located in finger flexor or extensor tendon, or thumb flexor, extensor or abductor tendon. Absence of pain in the neck or neurological symptoms in the median nerve distribution
5	Carpal tunnel syndrome	Pain or paresthesias or sensory loss in the median nerve distribution and one of Tinel's test positive, Phalen's test positive, noctural exacerbation of symptoms, motor loss with wasting of abductor pollicis brevis, abnormal nerve conduction time	Intermittent paresthesias or pain in at least 2 of digits I, II, or III; either may be present at night as well (allowing pain in the palm, wrist, or radiation proximal to the wrist) AND Symptoms present now or on at least 4 days during the last 7 days OR symptoms present on at least 4 days during at least 1 week in the last 12 months	Paresthesias or numbness in median nerve distribution, diminished power related to a peripheral nerve at the wrist. Absence of depressions, early morning stiffness, finger joint pain or swelling, rest pain and neurological symptoms in the ulnar nerve distribution
6	Guyon canal syndrome	Tenosynovitis of the wrist: Pain on movement localized to the tendon sheaths of the wrist and reproduction of pain by resisted active movement	Intermittent paresthesias in the palmar ulnar nerve distribution of the hand, distal to the wrist OR pain in the ulnar innervated area of the hand; the pain may radiate to the forearm AND Symptoms present now or on at least 4 days during the last 7 days OR symptoms present on at least 4 days during at least 1 week in the last 12 months	Tenosynovitis: Pain on movement of tendon or swelling of tendon sheath or triggering/locking/nodule on tendon located in finger flexor or extensor tendon, or thumb flexor, extensor or abductor tendon. Absence of pain in the neck or neurological symptoms in the median nerve distribution
7	Oarsman's wrist	Tenosynovitis of the wrist: Pain on movement localized to the tendon sheaths of the wrist and reproduction of pain by resisted active movement	Tenosynovitis of the forearm-wrist region: Intermittent pain-ache in the ventral or dorsal forearm or wrist region AND Symptoms now or on at least 4 days during the last 7 days OR symptoms present on at least 4 days during at least 1 week in the last 12 months	Tenosynovitis: Pain on movement of tendon or swelling of tendon sheath or triggering/locking/nodule on tendon located in finger flexor or extensor tendon, or thumb flexor, extensor or abductor tendon. Absence of pain in the neck or neurological symptoms in the median nerve distribution
Elbow				
8	Radial tunnel syndrome	Not included	Pain in the lateral elbow region or forearm muscle mass of the wrist extensors-supinator or weakness on extending the wrist and fingers AND symptoms present now or on at least 4 days during the last 7 days OR symptoms have been present on at least 4 days during at least 1 week in the last 12 months	Not included

9	Lateral epicondylitis	Epicondylar pain and epicondylar tenderness and pain on resisted extension of the wrist	Lateral epicondylitis: At least intermittent, activity-dependent pain directly located around the lateral epicondyle AND Symptoms present now or on at least 4 days during the last 7 days OR symptoms present on at least 4 days during at least 1 week in the last 12 months	Pain or tenderness or pain on loading relevant muscle at lateral epicondyle. Absence of shoulder pain.
10	Medial epicondylitis	Epicondylar pain and epicondylar tenderness and pain on resisted flexion of the wrist	Medial epicondylitis: At least intermittent, activity-dependent pain directly located around the medical epicondyle AND Symptoms present now or on at least 4 days during the last 7 days OR symptoms present on at least 4 days during at least 1 week in the last 12 months	Not included
11	Cubital tunnel syndrome		At least intermittent paresthesias in the 4th or 5th digit or both OR on the ulnar border of the forearm, wrist, or hand AND symptoms present now or on at least 4 days during the last 7 days OR symptoms present on at least 4 days during at least 1 week in the last 12 months	Not included
12	Bursitis of the elbow	Not included	Not included	Not included
13	Instability of the elbow	Not included	Not included	Not included
Shoulder and neck				
14	Tendinitis of the biceps tendon	Bicipital tendonitis: history of anterior shoulder pain and pain on resisted active flexion or supination of the forearm	Not included	Not included
15	Frozen shoulder	Shoulder capsulitis: history of pain in the deltoid area and equal restriction of active and passive glenohumeral movement with capsular pattern (external rotation>abduction>internal rotation)	Not included	Not included
16	Instability of the shoulder	Not included	Not included	Not included
17	Labral lesion of the glenoid	Not included	Not included	Not included
18	Rotator cuff tears	Not included	Not included	Not included
19	Subacromial impingement syndrome (rotator cuff syndrome, tendonitis and bursitis of the shoulder)	Rotator cuff tendonitis: history of pain in the deltoid region and pain on resisted active movement (abduction-supraspinatus, external rotation-infraspinatus, internal rotation-subscapularis)	Rotator cuff syndrome: At least intermittent pain in the shoulder region without paresthesias worsened by active elevation movement of the upper arm as in scratching the upper back AND Symptoms present now or on at least 4 days during the last 7 days OR symptoms present on at least 4 days during at least 1 week in the last 12 months	Shoulder tendonitis: limitation of abduction of the shoulder, painful arc on abduction of the shoulder, pain in the shoulder, sleep disturbance. Absence of a finger joint pain or swelling, pain in hand or wrist, dropping things or clumsiness, and difficulty with writing.
20	Suprascapular nerve compression	Not included	Not included	Not included
21	Cervical disc hernia	Not included	Not included	Not included
General				
22	Local arthritis (not RA) in a joint of upper extremities	Not included	Not included	
23	Complex regional pain syndrome	Not included	Not included	Not included
Non-specific CANS		Non-specific diffuse forearm pain: pain in the forearm in the absence of a specific diagnosis or pathology (sometimes includes loss of function, weakness, cramp, muscle tenderness, allodynia, slowing of fine movements)		Non-specific upper limb disorder: presence of pain in hand or wrist, pain in neck, discomfort and/or pain, weakness of arms or hands, dropping things or clumsiness. Absence of a painful arc of the shoulder, pain at lateral epicondyle on loading muscle, finger joint pain or swelling, sleep disturbance and fibromyalgia tender points.
Others		-	Radiating neck complaints	Fibromyalgia, inflammatory arthritis

of a set of manifestations, mostly clinical signs and symptoms.[13] The lack of consensus on a disorder's definition may impede a valid evaluation of diagnostic technology in test accuracy studies.[13] Moreover, using non-valid diagnostic tools to diagnose a disorder in research projects may lead to circular reasoning and therefore to overestimation of the diagnostic properties of a test.[11]

To overcome the problems caused by the absence of a gold standard, different approaches are used to formulate diagnostic criteria for upper-extremity disorders.

Approach based on: expert opinions

The first approach is based on 'experts' gold standard'. The studies of Harrington et al.[4] and Sluiter et al.[5] are examples of the use of expert opinions to formulate the diagnostic criteria.

Approach based on: statistical techniques

Another approach is based on the use of statistical techniques, employed by Helliwell et al.[10] who established core variables of the most common disorders seen in population samples. Consecutive new cases seen in clinical practice in five different centers were evaluated with multivariate modeling.

Approach based on: probabilities

A third approach is characterized by the identification of prognostic factors that can be translated into probabilities. In this approach diagnoses are not necessarily viewed as labels for disease processes, but more generally as a useful method for classifying people for the purpose of prevention or managing illness.[12] With this perspective the value of a case definition lies in its practical utility in distinguishing groups of people whose complaints share the same types of determinants of a certain outcome (including response to treatment).

Specific and non-specific CANS are characterized by their multifactorial etiology and prognosis. CANS arise from a complex interplay of pathological, physiological, psychosocial and/or cultural influences. In complaints with a multifactorial etiology and prognosis, like CANS, conceptual models to capture the multiplicity of the disease and to translate it to practical use can be proven by using the concept of probability.[14]

For example, Feleus et al.[15] studied patients with a new episode of CANS in general practice to identify prognostic factors for non-recovery at 6 months; 46% of the patients reported persistent complaints at 6 months. Complaint characteristics (long duration of the complaint before consultation, recurrent complaint, musculoskeletal co-morbidity, and complaints mainly located at wrist or hand) as well as psychosocial characteristics (more somatization, experienced less social support) were found to be predictors of non-recovery after 6 months. Having a specific diagnosis was also associated with recovery. Bot et al.[16] studied a multidimensional battery of predictors of short and long-term outcome of patients with

neck or shoulder symptoms in general practice. Similar to the results of Feleus et al.[15], they found that the characteristics of symptoms (duration, history of symptoms) as well as several psychological factors, were related to the short-term and long-term outcome.
Clinicians are used to basing their therapeutic decisions on the clinical picture or on a clear diagnosis. However, diagnosing is not an aim itself, but is relevant in as far as it has implications for prognosis and treatment.[17] The results of the studies of Feleus et al. 2006[15] and Bot et al. 2005[16] may help general practitioners to provide patients with more accurate information on their prognoses. Knowledge on these prognostic factors should be translated into probabilities of persistent CANS. This may guide health care professionals in their prediction of prognosis and treatment choice for the individual patient.
Moreover, when we can differentiate groups of CANS patients with respect to probabilities we can then initiate research to investigate the most effective interventions for each of these (sub)groups of patients.

Some issues with regard to these approaches need to be addressed. The process of validation spans many years and includes many studies. In fact, for criteria to be most robust several validation exercises should be carried out in a range of different patient populations.[18] Furthermore, with advances in technology on the disorders, the criteria will need to be updated. Therefore, this will be an ongoing process.
For example, establishing consensus among clinical experts on the terminology and classification (as done in our consensus project) does not necessarily ensure validity of the CANS model. As clinical experience evolves, the opinions of experts may also change.[19] The CANS model should then be re-evaluated according these new insights. Furthermore, the consensus achieved among clinical opinion makers should be seen as a starting point to establishing an 'unambigious' classification of musculoskeletal upper-extremity disorders whose validity should then be tested.

Prevalence of CANS

Prevalence rates are important to estimate the burden and impact of a disease. Trends in the occurrence and consequences of CANS may help researchers to initiate new research projects, and can also play an important role in the planning of health care strategies. Therefore, to provide insight into the prevalence and consequences of CANS according to the CANS model, we re-analyzed data from a population-based study (DMC_3-study, n=3664, chapter 7 of this thesis). We concentrated on chronic CANS because chronic complaints have the strongest impact on the patients and on health care due to the high costs related to disability, the use of health care, and to sick leave.

Excluding acute traumata and systemic diseases reduced the prevalence of CANS by about 25% and resulted in a relatively healthier population. Almost half of the patients with chronic CANS reported complaints in more than one anatomic site.
Walker-Bone et al.[18,20] reported that extensive pain in the neck and upper limbs tend to cluster and that many of their patients reported pain at three or more of the seven anatomic regions that they had formulated in the neck and upper limbs. This result is similar to our findings in the general Dutch population; for example, combined neck-shoulder complaints were present in 23% of the persons and over 43% reported complaints in more than one anatomic site. Therefore, to enable comparison of different research projects, the same valid measurement tools should be used in studies on the upper extremity and the neck. We have shown that the DASH (disability of arm-shoulder-hand) questionnaire has sufficient validity and responsiveness for use in patients with non-traumatic neck complaints (chapter 6 of this thesis) and may be used for this purpose.

Use of health care in patients with CANS

Of those with chronic CANS, 58% reported use of health care during the last year (chapter 7 of this thesis). Particularly those who reported more pain, worse general health, more limitations in daily living, and more sickness absence due to CANS, sought medical treatment.
In a Norwegian population-based study Hagen et al.[21] studied the use of health care in patients with musculoskeletal pain. In this study those with self-reported inflammatory rheumatic diagnoses were excluded. Of those who experienced non-inflammatory musculoskeletal pain during the last 12 months, 45% consulted their general practitioner. The results of this study suggest that consultation for the complaints was associated with greater mental distress; the association between mental distress and consulting was stronger for women than for men. This compares well with the study of Macfarlane et al.[22], who also concluded that consultation for chronic widespread pain was associated with a significant increase in psychological disturbance in women but not in men. In a cohort study (n=1347)[23] of Swedish middle-aged male farmers and rural non-farmers, 62.9% of the men reported current neck and/or low back pain during the last year prior to the baseline survey. Only 15.7% of these men had had at least one primary care consultation because of these conditions, and about 7% had been on sick leave and very few had been hospitalized or granted a disability pension owing to the conditions. Because the cohort included many farmers (a group known to have relatively low morbidity and use of health care in relation to reported complaints[24]) and consisted of men, the percentage that used health care is lower than can be expected in the general population.

Effectiveness of the treatment of CANS

The literature provides some evidence for the effectiveness of interventions used to treat specific and non-specific upper-extremity complaints.

Specific complaints

The CANS model lists 23 specific upper-extremity musculoskeletal disorders. Various therapeutic interventions for these conditions have been described in literature, including immobilization, physiotherapy, steroid injections, and surgical treatment. In the literature, it is unclear what information is available with regard to the effectiveness of interventions for the 23 specific upper-extremity disorders in the CANS model. Investigating the current state-of-the-art regarding the evidence for or against the most commonly used interventions for these disorders could be valuable for clinicians and researchers and help clinicians to choose the most appropriate intervention available. Moreover, such an overview will address the following questions: 1) What gaps exist in scientific knowledge with regard to interventions for specific musculoskeletal disorders of the upper extremity? and 2) Regarding to what extent is the outcome of the effectiveness of each intervention for the 23 specific disorders similar to each other?

To fill the gaps with regard to evidence-based information on the effectiveness of the low-incidence disorders and due to the absence of RCTs and CCTs, it may be useful to systematically review the available observational studies for these disorders separately, as we did for the radial tunnel syndrome and the posterior interosseus nerve syndrome (chapters 4 and 5 of this thesis). Such reviews may provide valuable data about the efficacy of treatment options and offer directions for future research.

Non-specific CANS

Verhagen et al.[25] systematically reviewed the literature with regard to the effectiveness of ergonomic and physiotherapeutic interventions for non-specific upper extremity work-related disorders in adults. A total of 21 interventions were included of which 17 studied persons with chronic non-specific neck or shoulder complaints or non-specific upper-extremity disorders. Over 25 interventions were evaluated. Five subgroups of interventions were determined: exercises, manual therapy, massage, ergonomics, and energized splint. The quality of the studies was generally poor. Limited evidence in favor of exercises was found compared to massage, and conflicting evidence was found in favor of exercises compared to no treatment. There was limited evidence for adding breaks during computer work, for massage as an add-on treatment to manual therapy, and for manual therapy as an add-on treatment to exercises.

The CANS model can be the first step towards an unambiguous, multidisciplinary accepted classification system for specific and non-specific upper-extremity disorders. This is the first requirement for accurate and meaningful communication amongst clinicians and researchers. Only when the majority of health care professionals and researchers speak 'the same language' the best treatment options and prevention strategies for each individual with musculoskeletal upper-extremity disorders can be identified and studied in the future.

Recommendations for future research

CANS model

Studies on diagnostic criteria and validation studies for both the classification system and the diagnostic criteria are needed to further refine the CANS model. For this, international multidisciplinary cooperation is recommended. Key persons (medical and paramedical professionals) should collaborate in this process. Furthermore, studies, focusing on the probability of persistent CANS should be conducted because knowledge on the probability of outcomes of persistent CANS may guide health care professionals in their prediction of prognosis and choice of treatment.

The number of occurrences of CANS is higher in the non-working population than in the working population aged 25 years and older, but those not working are affected most often. Therefore, research is needed on epidemiology, treatment and prevention of CANS in both the working and the non-working population.

Measurement of CANS

Almost half of those with chronic CANS reported complaints in more than one anatomic site. Therefore, a valid and responsive instrument that can measure disability in the whole upper-extremity (including the neck) was needed. We concluded that the DASH has shown sufficient validity and responsiveness for use in patients with non-traumatic neck complaints.

Although the DASH has been explored in several validation studies for patients with several arm-shoulder-hand complaints, more research is needed to confirm our findings with regard to the use of the DASH in patients with neck complaints. Studies on the minimal clinical important change (MCID) of the DASH are also needed in different (sub)groups of patients with CANS, because they will provide insight into the smallest meaningful change score that can be detected with the DASH.

Treatment of CANS

In the CANS model, consensus was achieved on 23 disorders that are considered to be specific. An overview is needed to provide insight to the current state-of-the-art with regard to

the effectiveness of the interventions for these specific disorders of the upper extremity. This overview will help health professionals to choose the most appropriate interventions available, and also guide future research by revealing similarities between interventions; moreover, any lack of knowledge on the effectiveness of the interventions will become apparent. Finally, in the absence of RCTs and CCTs, as seen in low-incident specific CANS, systematic reviews of observational studies should be performed to reveal possible tendencies with regard to the effectiveness of these disorders.

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Summary

Musculoskeletal disorders of the upper-extremity and neck are extremely common and one of the major causes of disability, sickness absence and health care use all around the world. The conditions do not threaten life, but they result in considerable discomfort for the patient and financial burden to society. Various names are given to musculoskeletal disorders of the upper extremity and various project have been conducted to achieve consensus on diagnostic criteria. However, a complete overview of all musculoskeletal neck and upper-extremity disorders coupled with a classification system that can be multidisciplinary approved was still missing. This thesis reported on the development of a new approach for terminology and classification of these disorders: CANS (complaints of the arm, neck and/or shoulder) and the CANS model.

Chapter 2 describes the results of a systematic appraisal of the worldwide incidence and prevalence rates of upper-extremity disorders (UEDs) available in scientific literature. The aim of this study was to gauge the range of these estimates in various countries and to determine whether the rates are increasing in time. Studies that recruited at least 500 people, collected data by using questionnaires, interviews and/or physical examinations, and reported incidence or prevalence rates of the whole upper extremity including the neck, were included. No studies were found with regard to the incidence of UEDs and 13 studies that reported prevalence rates of UEDs were included. The point prevalence ranged from 1.6-53%; the 12-months prevalence ranged from 2.3-41%. One study reported on the lifetime prevalence (29%). We did not find evidence of a clear increasing or decreasing pattern over time. The case definitions for UEDs used in the studies, differed enormously. Therefore, it was not possible to pool the data. We concluded that there are substantial differences in reported prevalence rates on UEDs. Main reason for this is the absence of a universally accepted way of labelling or defining UEDs. If we want to make progress in this field, the first requirement is to agree on unambiguous terminology and classification of EUDs.
Therefore, we initiated a multidisciplinary consensus project on terminology and classification of complaints of arm, neck and/or shoulder.

Chapter 3 reports on the results of this consensus project. The aim of this study was to agree on an 'unambiguous language' concerning the terminology and classification that can be used by all relevant medical and paramedical disciplines in the Netherlands. A Delphi consensus strategy was initiated. The outcomes of a multidisciplinary conference were used as starting point. In total 47 experts in the field of upper-extremity musculoskeletal disorders were delegated by 11 medical and paramedical professional associations to form the expert-panel for the Delphi consensus strategy. Each Delphi round consisted of a questionnaire, an analysis and a feedback report. After three Delphi rounds consensus was achieved. The experts reported the consensus in a model. This so-called CANS model describes term, definition and classification of complaints of arm, neck and/or shoulder

(CANS) and helps professionals to classify patients unambiguously. CANS is defined as "Musculoskeletal complaints of arm, neck and/or shoulder not caused by acute trauma or by any systemic disease". The experts classified 23 disorders as specific CANS, because they were judged as diagnosable disorders. All other complaints were called non-specific CANS. In addition, the experts defined 'alert symptoms' on the top of the model. The use of the CANS model can increase accurate and meaningful communication amongst healthcare workers, and may also have a positive influence on the quality of scientific research, by enabling data of different studies to be compared.

Chapter 4 describes the results of a systematic review executed to evaluate the effectiveness of conservative and surgical interventions for treating the radial tunnel syndrome (RTS). RTS is one of the 23 specific disorders in the CANS model. No RCTs (randomized clinical trials) or CCTs (controlled clinical trials) are available for RTS. Therefore, we systematically reviewed all available observational studies on treatment of RTS. Although the validity of case series is inferior to controlled trials, they may provide valuable data about the efficacy of interventions. A literature search and additional reference checking was done. On the basis of previous checklists, we constructed a quality assessment and rating system to analyse the included case series. Studies with less than 50% of the maximum points on the methodological quality assessment were excluded from the analysis. The results were summarised according to a rating system for the strength of the scientific evidence.
The literature search and additional reference check resulted in 21 eligible case series for this review. The methodological quality and data extraction was performed. Six higher quality articles were included in the final analysis. They all reported on surgical decompression of the posterior interosseus nerve (PIN). We concluded that there is a tendency for the effectiveness of surgical decompression of the posterior interosseus nerve in patients with RTS. The effectiveness of conservative treatments for RTS is unknown because, for most treatments, no studies were available. Additional high-quality controlled studies are needed to assess the level of 'conclusive evidence' for surgical treatment. There is also a need for high-quality controlled trials into the effectiveness of conservative treatments for RTS.

Chapter 5 presents a systematically review of all available observational studies on treatment of the posterior interosseus nerve syndrome (PINS). Also for the PINS, no randomized controlled trials or controlled clinical trials about the effectiveness of interventions are available; only case series can be found. To study the effectiveness of treatment of the PINS, we used the same method as described in the systematic review about the effectiveness of treatment of RTS in chapter 4.
For PINS, six eligible case series for this review were found. After the data extraction and methodological quality assessment, two higher quality studies that evaluated the effectiveness of surgical decompression of the PIN were included in the final analysis. We

concluded that there is a tendency for the effectiveness of surgical decompression of the PIN in patients with PINS. The effectiveness of a conservative treatment for PINS is unknown because no higher quality studies are available. Additional high-quality controlled studies are needed to assess the level of 'conclusive evidence' for surgical treatment, and also to evaluate conservative treatments for PINS.

In **chapter 6** a prospective cohort was used to study whether the DASH (disability of the arm, shoulder and hand) questionnaire is not only a valid and responsive instrument to measure patients with arm, shoulder and hand complaints, but also to evaluate patients with neck complaints. The DASH has shown to be a valid and responsive questionnaire to evaluate disability in patients with arm, shoulder and hand complaints. However, patients with arm, shoulder, or hand complaints frequently report neck complaints as well. Therefore, a valid and responsive questionnaire designed for the whole upper extremity including the neck would be very useful and practical in upper-extremity research. In total, 679 patients visiting their general practitioner with a new episode of non-traumatic complaints of the neck and upper extremity were evaluated by use of questionnaires at baseline and at 6-months follow-up. Four (sub)groups (most complaints in arm-shoulder-hand, arm-shoulder-hand-neck, neck-shoulder, and neck) were formulated. Disability (DASH), general health (SF-12 (physical and mental component)), severity, and persistence of complaints were assessed. Construct validity and responsiveness were studied by testing 14 predefined hypotheses based on correlations, responsive ratios, and floor and ceiling effects.
Correlations between the DASH and the other measures within the four (sub)groups at baseline (construct validity) and for the change scores at 6-months follow-up (responsiveness) were found adequate; responsiveness ratios in all of the four (sub)groups were classified as sufficient. No floor and ceiling effects were found. All hypotheses could be accepted. So, this study demonstrated sufficient validity and responsiveness of the DASH for use in patients with non-traumatic neck complaints.

In **Chapter 7** the results were presented of a study that evaluated the prevalence rates of UEDs (disorders of the upper extremity and neck as a total) and CANS (Complaints of the Arm, Neck and/or Shoulder not caused by acute trauma or any systemic disease) as defined in the CANS model and the socio-demographic and health characteristics of chronic complaints in the open population. Data were obtained from the DMC_3-study, a Dutch questionnaire survey on musculoskeletal conditions (>25 years, n = 3664). Data on four anatomic sites were assessed: neck, shoulder, elbow and wrist. Various health characteristics were measured including the SF-36. Rectangle diagrams were used to illustrate co-occurrence of pain in the four anatomic sites. The 12-months prevalence of CANS was 36.8%, the point prevalence was 26.4%, and 19.0% reported chronic CANS. Women, persons aged 45-64 years, with the lowest education level, and those not working were the most affected. Within those with

UEDs, around 25% of cases was caused by an acute trauma or by some systemic disease. Of those with chronic CANS, 58% reported use of healthcare. Healthcare users scored worse on general health, limitations in daily living, pain, and sickness absence than non-healthcare users; over 43% reported complaints in more than one anatomic site. We concluded that UEDs and CANS frequently occur in the open population. Excluding acute traumata and systemic diseases reduced the prevalence of CANS and resulted in a relatively healthier population. A compound definition of CANS seems indicated because of the large overlap of affected anatomic sites.

Chapter 8 reflects on the findings in this thesis and gives recommendation for future research. International multidisciplinary cooperation is recommended to refine the CANS model. Studies on diagnostic criteria and validation studies for both the classification system and the diagnostic criteria are needed. Furthermore, also studies, which concentrated on the probabilities of persistent CANS, should be conducted, because the knowledge of the probability of clinical outcomes of persistent CANS may guide health care professionals in their prediction of prognosis and treatment choice.
CANS are frequently occurring conditions in both the working and non-working population aged 25 years and older, but those not working were most affected. Therefore, research is needed on epidemiology, treatment and prevention of CANS in the working as well as in the non-working population.

Samenvatting

Klachten van het bewegingsapparaat in de arm-, nek- en schouderregio vormen een veel voorkomend gezondheidsprobleem. Over de hele wereld worden mensen geconfronteerd met de problematiek die deze klachten met zich meebrengt. De klachten zijn weliswaar niet levensbedreigend, maar ze resulteren in leed en ongemak bij de patiënt en zijn omgeving en leiden tot toenemende kosten voor de maatschappij door ziekteverzuim en gebruik van de gezondheidszorg.
Verschillende namen zijn in omloop voor deze aandoeningen in de arm-, nek- en schouderregio. Er zijn meerdere projecten uitgevoerd om consensus te bereiken over diagnostische criteria voor verschillende klachtencomplexen. Echter, het ontbreekt aan een compleet overzicht van alle aandoeningen van het bewegingsapparaat in de arm, nek en schouderregio gekoppeld aan een indeling in specifieke en a-specifieke aandoeningen, die multidisciplinair bruikbaar en acceptabel is. Dit proefschrift springt in op deze leemte en handelt over de totstandkoming van een nieuwe benadering van terminologie en indeling van klachten in de arm, nek en schouderregio: CANS (complaints of the arm, neck and/or shoulder) en het CANS model.

In **Hoofdstuk 2** worden de resultaten beschreven van een systematische evaluatie van de wetenschappelijke literatuur over de incidentie (het aantal nieuwe mensen dat de klachten heeft gekregen) en prevalentie (het vóórkomen van de klachten in een bepaalde populatie in een bepaalde periode) van klachten in de arm-, nek- en schouderregio. Centrale vragen daarbij waren: Wat is de omvang van de klachten in verschillende landen? En stijgen (of dalen) de klachten in de loop van de tijd? Studies over de incidentie en/of prevalentie van deze klachten zijn meegenomen in deze evaluatie als in de desbetreffende studie ten minste 500 patiënten waren geëvalueerd en de resultaten verkregen waren middels vragenlijsten, interviews en/of lichamelijk onderzoek. Verder moesten incidentie en prevalentie cijfers gerapporteerd zijn over de gehele arm-, nek- en schouderregio. Uiteindelijk zijn er geen studies gevonden over de incidentie. Wel zijn er 13 studies gevonden over de prevalentie. Uit de prevalentie studies is gebleken dat het aantal mensen dat klachten in de arm-, nek- en schouderregio aangaf op het moment van meten varieerde van 1,6 – 53%. Het aantal mensen dat deze klachten in het afgelopen jaar had gehad varieerde van 2,3-41%. Eén van de studies gaf een ‘lifetime’ prevalentie cijfer (29%). Er is geen bewijs gevonden voor een duidelijke toename of afname van de klachten in de loop van de tijd. Door het gebruik van verschillende definities voor klachten in de arm-, nek- en schouderregio in de geïncludeerde studies, is het niet mogelijk geweest de data samen te voegen. De eindconclusie is dat arm-, nek- en schouderklachten in veel verschillende landen voorkomen en dat er substantiële verschillen bestaan in gerapporteerde prevalentie cijfers. Dit wordt hoofdzakelijk veroorzaakt door de afwezigheid van een algemeen aanvaarde manier van benoemen en indelen van deze klachten. Om vooruitgang te boeken op dit terrein, is het een eerste vereiste om te komen tot eenduidige terminologie en indeling van arm-, nek- en schouderklachten.

Dat is dan ook de reden geweest om het multidisciplinaire consensustraject over terminologie en indeling van deze klachten te initiëren.

In **Hoofdstuk 3** wordt verslag gedaan van de uitvoering en de resultaten van dit consensustraject. De term RSI (repetitive strain injury) was aan vervanging toe, omdat deze term in de praktijk een groot aantal nadelen heeft. Ten eerste heeft RSI voor patiënten een negatieve lading. Daarbij schept de term verwarring: het gaat veelal niet om een 'injury'. Bovendien kan niet alleen 'repetitive strain', maar ook statische belasting de klachten veroorzaken. Naast RSI worden in Nederland, maar ook daarbuiten nog vele andere termen gebruikt voor arm-, nek- en/of schouderklachten en zijn vele definities en indelingen in omloop. Het heeft geleid tot spraakverwarring onder zowel behandelaars als patiënten. Het spreken van dezelfde taal is een eerste vereiste voor goede samenwerking. Maar ook voor het vergelijken van wetenschappelijk onderzoek is eenduidig taalgebruik van belang. Daarom is het multidisciplinaire consensustraject over terminologie en indeling van deze klachten ingezet. Een panel van experts, bestaande uit 47 afgevaardigden van elf medische- en paramedische beroepsorganisaties heeft zich gebogen over een nieuwe naam, definitie en indeling van arm-, nek- en/of schouderklachten. Het uiteindelijke resultaat moest voor alle beroepsgroepen bruikbaar zijn.
Allereerst heeft een multidisciplinaire werkconferentie plaatsgevonden. De uitkomsten hiervan zijn vervolgens gebruikt als startpunt voor een Delphi-onderzoek. Elke Delphi-ronde bestond uit een vragenlijst, een analyse en een feedbackrapport. Na drie Delphi-rondes werd consensus bereikt. Het resultaat is weergegeven in het zogenaamde CANS model: Er werd overeengekomen de klachtengroep voortaan te omschrijven als CANS (Complaints of Arm, Neck and/or Shoulder). Volgens de daarbij opgestelde definitie zijn dit klachten van het bewegingsapparaat in arm, nek en/of schouder, die niet veroorzaakt worden door een acuut trauma, zoals een botbreuk, of een systemische aandoening, zoals reumatoïde artritis. Het CANS model verdeelt de klachten in specifieke en a-specifieke CANS. Het panel classificeerde 23 aandoeningen als specifieke CANS, omdat ze beoordeeld werden als diagnosticeerbare aandoeningen. Elke andere aandoening wordt a-specifieke CANS genoemd. Verder definieerden de experts 'alert symptomen' - zoals het hebben van pijn in de linkerarm bij hartproblematiek - als eerste stap in het model.
Het CANS model helpt medici en paramedici om patiënten eenduidig te classificeren. Door het gebruik van dezelfde terminologie en indeling zullen behandelaars elkaar beter begrijpen, verbetert de multidisciplinaire samenwerking en zullen onderzoeksresultaten beter vergelijkbaar worden. Ook de patiënt zal hier de voordelen van ervaren: door betere communicatie kan sneller de juiste behandeling worden ingezet. De consensus is hiertoe de eerste stap.

Hoofdstuk 4 beschrijft de resultaten van de systematische evaluatie van de wetenschappelijke literatuur (systematische review) over de effectiviteit van conservatieve en chirurgische behandelingen bij het Radiaal tunnel syndroom (RTS). RTS is één van de 23 specifieke aan-

doeningen uit het CANS model. Van het RTS zijn geen gerandomiseerde gecontroleerde klinische onderzoeken (RCTs) of niet-gerandomiseerde gecontroleerde klinische onderzoeken (CCTs) beschikbaar. Daarom zijn voor deze studie alle beschikbare observationele studies die de behandeling van het RTS onderzoeken, op systematische wijze bekeken en beoordeeld. Hoewel de validiteit van beschrijvingen van behandelingen van patiënten series (case series) ondergeschikt is aan (gerandomiseerde) gecontroleerde trials, kunnen ze mogelijk toch waardevolle informatie verschaffen over de effectiviteit van behandelingen.
Allereerst is de wetenschappelijke literatuur doorzocht en is een aanvullende referentie check uitgevoerd. Gebaseerd op eerdere scoringslijsten die de kwaliteit van studies evalueren, is een eigen scoringslijst samengesteld die de kwaliteit van de geïncludeerde artikelen moest beoordelen. Studies met minder dan 50% van de maximale score op deze lijst, zijn geexcludeerd. Voor de uiteindelijke conclusies is gebruik gemaakt van een classificatiesysteem voor de sterkte van het wetenschappelijk bewijs dat gebaseerd is op de kwaliteitsscore van de verschillende studies. Het literatuuronderzoek en de referentie check resulteerden in 21 geschikte studies. Na de beoordeling van de kwaliteit van de studies, bleven er zes studies over die meegenomen zijn in de uiteindelijke analyses. Uiteindelijk concludeerden we dat er een tendens is dat chirurgische decompressie van de posterior interosseus zenuw effectief is bij patiënten met het RTS. Omdat er geen studies gevonden zijn die de mate van effectiviteit van conservatieve behandelingen van het RTS onderzoeken, kon hierover geen conclusie getrokken worden. Er is dan ook behoefte aan kwalitatief hoogwaardig onderzoek op dit terrein. Verder zijn ook (gerandomiseerde) klinische trials van hoge kwaliteit nodig om de tendens die wij hebben gevonden verder te bestuderen.

Hoofdstuk 5 presenteert een systematische review over de effectiviteit van behandelingen voor het posterior interosseus nerve syndrome (PINS). Ook voor het PINS zijn geen RCTs en CCTs voorhanden. Daarom is ook hier gebruik gemaakt van de resultaten die beschreven staan in observationele studies. Voor deze systematische review, gebaseerd op case series, is dezelfde methode gebruikt als hierboven is beschreven voor het RTS (hoofdstuk 4).
Op basis van het literatuuronderzoek en de referentie check zijn zes studies geïncludeerd die de effectiviteit van behandelingen van het PINS bestuderen. Na de data extractie en kwaliteitsbeoordeling, bleven twee studies van hoge kwaliteit over, die beiden de effectiviteit van chirurgische decompressie evalueren. Op basis van deze studies is geconcludeerd dat er een tendens is dat chirurgische decompressie van de posterior interosseus zenuw effectief is bij patiënten met het PINS. Over de effectiviteit van conservatieve behandeling bij het PINS kan vooralsnog geen uitspraak gedaan worden, omdat geen studies van hoogwaardige kwaliteit gevonden zijn. Onderzoek op dit gebied is dan ook gewenst. Verder moet ook hier geconcludeerd worden dat (gerandomiseerde) gecontroleerde klinische trials van hoogwaardige kwaliteit nodig zijn om het overtuigende bewijs te leveren voor de effectiviteit van chirurgisch ingrijpen bij het PINS.

In **hoofdstuk 6** wordt bestudeerd of de DASH (Disability of the arm, shoulder and hand) vragenlijst valide (meet het instrument wat het moet meten?) en responsief (kan het instrument veranderingen in de loop van de tijd meten?) is voor patiënten met nekklachten. De DASH wordt gezien als een valide en responsief meetinstrument om patiënten met arm-, schouder of handklachten te evalueren. Echter, de ervaring leert dat mensen met klachten in deze regio's vaak ook nekklachten hebben. Daarom zou het voor wetenschappelijk onderzoek nuttig en praktisch zijn om een vragenlijst beschikbaar te hebben die voor de hele bovenste extremiteit, inclusief de nek, te gebruiken is. Om de validiteit en responsiviteit van de DASH bij nekklachten te bestuderen is gebruik gemaakt van een prospectief cohort. Het cohort (n=679) bestond uit patiënten die hun huisarts bezochten met een nieuwe episode van niet traumatische nekklachten. De patiënten hebben verschillende vragenlijsten ingevuld bij aanvang van de studie en zes maanden later. Er zijn vragen gesteld over hun beperkingen (DASH), algemene gezondheid (SF-12 (fysieke en mentale component)), de ernst van de klachten en het aanhouden van de klachten na zes maanden.
Voor het onderzoek zijn vier (sub)groepen geformeerd met respectievelijk de meeste klachten in de volgende regio's: 1) arm-schouder-hand, 2) arm-schouder-hand-nek, 3) nek-schouder en 4) nek. Er zijn vooraf 14 hypotheses geformuleerd om de zogenaamde construct validiteit en de responsiviteit te beoordelen. De hypotheses hadden betrekking op de correlaties tussen uitkomsten op de verschillende vragenlijsten, de responsiviteitsratio's en de 'floor' en 'ceiling' effecten binnen de verschillende (sub)groepen. Zowel de correlaties tussen de DASH en de verschillende meetinstrumenten bij baseline (construct validiteit) als die van de veranderingsscores na zes maanden follow-up (responsiviteit) voor alle (sub)groepen waren adequaat. De responsiviteitsratio's in alle (sub)groepen waren eveneens goed. Verder zijn geen 'floor' en 'ceiling' effecten aangetroffen. Daarom konden alle hypothesen worden geaccepteerd en kan geconcludeerd worden dat de DASH voldoende valide en responsief is om te gebruiken bij onderzoek naar patiënten met niet traumatische nekklachten.

In **hoofdstuk 7** worden de resultaten gepresenteerd van een onderzoek naar de omvang van CANS en de gezondheidskarakteristieken van mensen met deze klachten in de open Nederlandse populatie. Doel van het onderzoek was om het effect van het gebruik van de definitie van CANS (dus het uitsluiten van acute traumata en systemische ziektes) te achterhalen op de prevalentiecijfers en de gezondheidskarakteristieken. Ook wilden we weten hoe vaak mensen met chronische CANS een medicus of paramedicus bezoeken in verband met deze klachten en wat de overlap van de klachten is in de verschillende anatomische regio's. Om deze vragen te beantwoorden, is gebruik gemaakt van een onderzoek dat uitgevoerd is in de open Nederlandse populatie (DMC_3-study, n=3664, leeftijd > 25 jaar). In dit onderzoek hebben de deelnemers schriftelijk allerlei vragen beantwoord over eventuele klachten van het bewegingsapparaat, hun algemene gezondheid (SF-36) en het bezoeken van medicus of paramedicus in verband met de klachten. Er is gebruik gemaakt van de vragen uit dit

onderzoek die betrekking hebben op de nek-, schouder-, elleboog- en pols/handregio. Zogenaamde 'rectangle diagrammen' zijn gebruikt om de overlap van de pijn in de verschillende anatomische regio's te illustreren.
Uit de analyses van de data blijkt dat 36,8% van de mensen de laatste 12 maanden CANS had gehad, dat 26,4% van de mensen op het moment van meting klachten had en dat 19% van de mensen getroffen was door chronische CANS. Het gebruik van de definitie van CANS (uitsluiten acute traumata en systemische ziekten) reduceert de groep met klachten met zo'n 25%. Klachten komen het meest voor bij vrouwen, mensen in de leeftijdsgroep van 45-64 jaar, mensen met een lage opleiding en niet-werkenden. Van de mensen met chronische CANS, zoekt 58% medische hulp. Deze laatste groep van zorggebruikers vertoont een slechtere algemene gezondheid, heeft meer pijn en beperkingen en scoort hoger als het gaat om ziekteverzuim dan de mensen die geen gebruik maakten van de gezondheidszorg. Verder blijkt dat meer dan 43% van de mensen met chronische CANS pijn heeft in meer dan één anatomische regio. We concludeerden dat klachten van het bewegingsapparaat in de bovenste extremiteit inclusief de nek en CANS veel voorkomt in de open Nederlandse populatie. Het uitsluiten van de mensen met klachten ten gevolge van een acuut trauma dan wel een systemische ziekte resulteerde in een relatief gezondere populatie. Vanwege de grote overlap van klachten in de aangedane anatomische regio's lijkt een brede definitie van CANS geïndiceerd.

Hoofdstuk 8 bevat een reflectie van de resultaten van dit proefschrift en geeft aanbevelingen voor vervolg onderzoek om het CANS model verder te optimaliseren. Er wordt aanbevolen om hiervoor internationaal te gaan samenwerken. Toekomstig onderzoek zou zich moeten richten op het formuleren en evalueren van diagnostische criteria van de in het CANS model genoemde specifieke aandoeningen en op het valideren van het CANS model als geheel. Ook zouden studies uitgevoerd moeten worden naar modellen die de kans op het ontstaan van persisterende CANS voorspellen. De kennis hierover kan medische professionals helpen bij het geven van een prognose en het instellen van de juiste behandeling.
CANS komt veelvuldig voor bij zowel werkende als niet-werkende mensen van 25 jaar en ouder, maar de niet-werkende populatie is het meest getroffen. Daarom is verder onderzoek van zowel de werkende als de niet-werkende populatie nodig naar de epidemiologie, behandeling en preventie van CANS.

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Curriculum Vitae

Bionka Huisstede is op 14 oktober 1969 geboren in Goor, gemeente Hof van Twente. Na het gymnasium in Hengelo, ging zij in 1989 Fysiotherapie studeren aan de Hogeschool van Enschede. Tijdens haar stage op de afdeling Fysiotherapie van het Streekziekenhuis Midden Twente te Hengelo heeft zij niet alleen veel geleerd over het vak fysiotherapie, maar heeft ze ook kennis gemaakt met het doen van wetenschappelijk onderzoek door het opzetten en uitvoeren van een onderzoek naar de effectiviteit van drie fysiotherapeutische behandelmethoden bij enkeldistorsies. Geënthousiastmeerd door deze ervaring, is Bionka in 1993 Gezondheidswetenschappen, afstudeerrichting Bewegingswetenschappen aan de Universiteit van Maastricht gaan studeren. Twee jaren later, in 1995, rondde ze deze studie af met een onderzoek naar de validiteit en betrouwbaarheid van echografie als meetinstrument ter bepaling van de omvang van de musculus quadriceps. Dit onderzoek heeft zij uitgevoerd bij de afdeling Traumatologie van het Academische Ziekenhuis Maastricht onder begeleiding van prof.dr. Jouwert Stapert. Naast haar opleiding werkte Bionka als fysiotherapeut in de Praktijk voor Fysiotherapie Phillippens te Maastricht. Hier is zij ook na haar studie nog enige tijd blijven werken.

In mei 1996 is Bionka naar Groningen verhuisd om als Rayon Manager te gaan werken bij Glaxo Wellcome (nu Glaxo Smith Kline). In juli 1999 is zij in dienst getreden bij de Reumapatiëntenbond in Amersfoort. Zij heeft hier het project 'kwaliteitscriteria voor groepsoefentherapie voor mensen met Reumatoïde Artritis en de ziekte van Bechterew' geleid en heeft zich daarnaast onder andere bezig gehouden met collectieve belangenbehartiging voor patiënten. In juni 2000 is zij bij het Integraal Kankercentrum Rotterdam gaan werken, waar zij onder andere mee heeft gewerkt aan projecten over palliatieve zorg en Herstel & Balans.

Sinds februari 2002 werkt Bionka bij het Erasmus MC in Rotterdam. Zij heeft hier een duobaan. Zij is aangesteld als coördinator van het onderzoeksinstituut MUSC (Musculoskeletal Science Center) en heeft tot februari 2005 als beleids- en wetenschappelijk medewerker bij het Kenniscentrum AKB (Arbeid en Klachten Bewegingsapparaat) gewerkt. Bij het Kenniscentrum AKB heeft Bionka het multidisciplinaire consensustraject over terminologie en indeling van klachten van arm, nek en schouder geleid. Het CANS model is het resultaat van dit traject. In de loop van het traject is het idee ontstaan om hier een promotie van te maken.
Bionka is in november 2003 met haar promotie gestart bij het Kenniscentrum AKB. Vanaf februari 2005 heeft zij gedurende twee jaar één dag per week bij de afdelingen Huisartsgeneeskunde en Orthopedie aan haar promotie gewerkt. De andere vier dagen is zij blijven werken voor MUSC.

Vanaf februari 2007 werkt Bionka drie dagen per week als coördinator van MUSC en zet zij twee dagen per week met recent verworven subsidie haar onderzoek naar CANS voort bij de afdeling Huisartsgeneeskunde.