Cognitive behavioural treatment programme for chronic neuropathic pain after spinal cord injury

Matagne Heutink

Cover Mike Twilhaar, Animo Reclame
Layout Renate Siebes, Proefschrift.nu
Printed by Ridderprint, Ridderkerk
ISBN 978-90-393-6181-8

Cognitive behavioural treatment programme for chronic neuropathic pain after spinal cord injury

Cognitief gedragstherapeutisch behandelprogramma voor chronische neuropathische pijn na een dwarslaesie

(met een samenvatting in het Nederlands)

Proefschrift

ter verkrijging van de graad van doctor aan de
Universiteit Utrecht op gezag van de rector magnificus,
prof.dr. G.J. van der Zwaan, ingevolge het besluit van het
college voor promoties in het openbaar te verdedigen
op woensdag 24 september 2014 des middags te 2.30 uur

door

Matagne Heutink

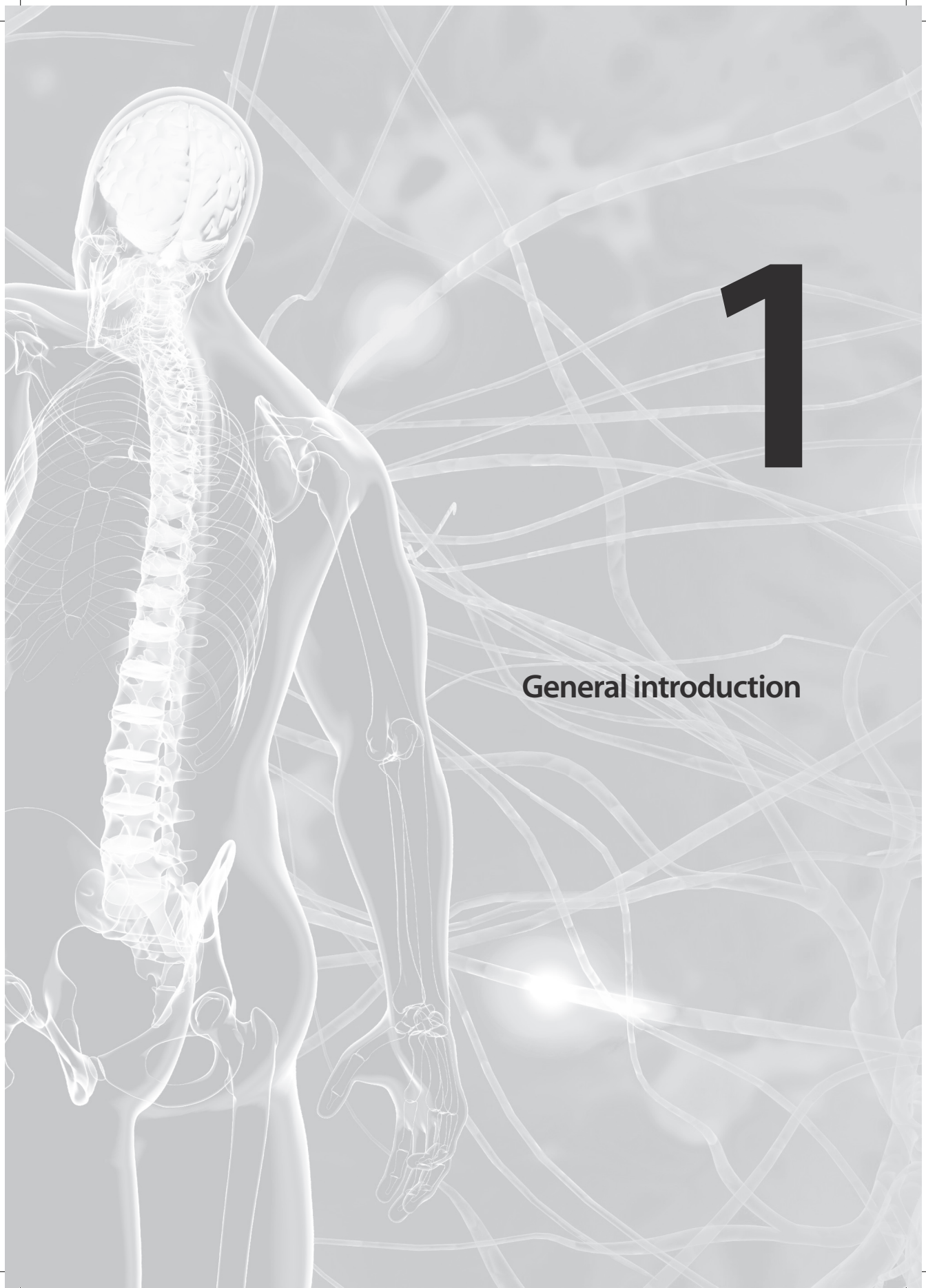
geboren op 26 april 1977
te Boxtel

Promotoren: Prof.dr. J.M.A. Visser-Meily
Prof.dr. M.W.M. Post

De in dit proefschrift beschreven studies zijn mogelijk gemaakt door financiële steun van de Stichting Wetenschappelijk Fonds De Hoogstraat te Utrecht en Pfizer (projectnummer 007-04). De studies zijn uitgevoerd binnen DALI voor PIJN (initiatief van Pfizer), een landelijk stimuleringsprogramma dat gericht was op optimalisatie van pijnzorg.

De totstandkoming van dit proefschrift werd mede mogelijk gemaakt door financiële steun van De Hoogstraat Revalidatie, de Stichting Wetenschappelijk Fonds De Hoogstraat te Utrecht en Adelante Zorggroep te Hoensbroek.

Contents



1

General introduction

Spinal cord injury

The spinal cord is a major bundle of nerves through which motor and sensory information travels to and from the brain to other parts of the body. A spinal cord lesion, also called spinal cord injury (SCI), is damage to the spinal cord resulting in a change, either temporary or permanent, of the normal motor, sensory, or autonomic function of the spinal cord.[1] SCI is characterised by the level of the lesion and is divided into tetraplegia and paraplegia. A tetraplegic lesion refers to impairment or loss of sensory and/or motor function in the cervical segments of the spinal cord, resulting in impairment of function in the arms as well as typically in the trunk, legs, and pelvic organs, i.e. including the four extremities. A paraplegic lesion refers to impairment or loss of sensory and/or motor function in the thoracic, lumbar, or sacral segments of the spinal cord, leaving arm functioning intact, but (depending on the level of injury) the trunk, legs, and pelvic organs maybe involved. Depending on the amount of damage, SCI can result in complete or incomplete loss of sensory and/or motor function below the level of the lesion.[1] SCI can be of traumatic or non-traumatic origin. The main causes of a traumatic SCI are motor vehicle collisions, falls, violence, and sport activities,[2] with falls being reported as the leading cause in the Netherlands.[3] Common non-traumatic causes are spinal degeneration, tumourous compression, vascular diseases, congenital diseases, and inflammatory conditions,[2] with the first 3 being reported as the most frequent aetiologies in the Netherlands.[4]

Epidemiology SCI

The worldwide SCI incidence varies from 10.4 to 83 per million per year, of whom one-third have tetraplegia, 50% have a complete lesion, mean age at injury is 33 years (range 16–50), and the distribution of men/women is 3.8/1.[5] A trend towards an increased incidence in the elderly is observed, likely due to non-traumatic injury and falls.[2] The incidence of a traumatic SCI surviving the acute phase has been estimated at 11.7 per million per year in the Netherlands in 2010, of whom 69% have tetraplegia, 38% have a complete lesion, mean age at injury is 62 years (range 13–96), and the distribution of men/women is 2.8/1.[3] The incidence of non-traumatic SCI is unknown in the Netherlands.[6]

An estimation of the worldwide SCI prevalence is unknown. A review identified only 5 studies on the prevalence of SCI, all of them from developed countries.[5] The prevalence of persons with SCI in the Netherlands is estimated to be about 10,000–15,000 persons.[6] Because of improvements in medical care, the average life expectancy of persons with a SCI

has considerably increased in the last decades.[7] The prevalence of SCI is therefore expected to increase.

Eight rehabilitation centres are specialised in SCI rehabilitation care in the Netherlands. Between 300 and 400 persons are admitted to a rehabilitation centre in the Netherlands as a consequence of SCI each year.[6] The proportion of persons with SCI of non-traumatic origin admitted to a rehabilitation centre has grown a majority of 54.7% in the last decade in rehabilitation centres in the Netherlands and Belgium.[8]

Impact of SCI and pain

SCI is a major life event because it usually results in reduced mobility and functional independence, impairment of social and vocational activities, as well as negative influences on the person's well-being and health.[9] The psychological impact of SCI is major. Many persons with SCI have clinical levels of depression and anxiety,[10] and their average life satisfaction is substantially below that of the general population.[11] Persons with SCI often face serious secondary health conditions, e.g. bladder and bowel disorders, pressure ulcers, neurogenic heterotopic ossification, oedema, autonomic dysregulation, spasticity, obesity, cardiovascular and respiratory problems, and in many cases pain.[6] Pain after SCI is related to poorer physical, psychological, and social functioning,[12,13] often interferes with activities (e.g. sleep, work, exercise, household),[14,15] and it negatively affects rehabilitation outcomes.[16] Moreover, SCI pain has often been reported as an important factor in reduced quality of life[16-18] and great emotional distress.[19] Pain is even found to be the most disabling condition for both daily and social activities[20] and far more disabling than the injury itself.[18,19] Furthermore, it is unlikely that the pain problem will resolve on its own[18] or will diminish over time.[13]

SCI pain

The International Association for the Study of Pain (IASP) defines pain as "an unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage".[21] Pain is a frequent complication of SCI[22,23] and most persons with chronic SCI pain report more than one pain problem.[15] Studies examining chronic pain prevalence have noted that on average, two-thirds of persons with SCI report some type of pain and around one-third rates their pain as severe,[24] but estimates vary widely from 11 to 94 percent for chronic pain and 18 to 63 percent for severe, disabling pain.[15] Part of these large variations in reported prevalence of SCI pain can be attributed to differences

in methodology, like varying time since injury (e.g. during rehabilitation versus 2 or more years post-injury), the definition of 'chronic' (e.g. 2 weeks versus 6 months), the populations being studied, and the lack of consensus regarding the classification and definitions of types of pain that occur following SCI.[25] The Spinal Cord Injury Pain Task Force of the IASP developed a taxonomy or classification of SCI pain in which SCI-related pain is classified as either nociceptive: musculoskeletal or visceral pain, or neuropathic: above-level, at-level, or below-level neuropathic pain.[26-28] Above-level neuropathic pain is less common[29] and not a direct consequence of SCI.[30]

Neuropathic SCI pain

The IASP defines neuropathic pain as "pain caused by a lesion or disease of the somatosensory nervous system".[27] Neuropathic pain is for example described as 'burning', 'shooting', 'stabbing', 'itching', 'pricking', 'tingling' sensations and pain that is 'painful cold' and 'electric shock-like'[27,31] and often accompanied by hypoesthesia (reduced touch sensation) and allodynia (pain due to a stimulus which does not normally provoke pain).[27] Despite the enmeshment of different pain types in persons with SCI, the experience of neuropathic pain is described as distinctive.[19] One of the most popular and validated questionnaires to determine neuropathic pain is the Douleur Neuropathique (neuropathic pain) 4 questions (DN4).[32] For pain to be classified as neuropathic pain with the DN4, 4 (or more) questions have to be answered with "yes" of the 10 questions about the pain characteristics (3), pain associated with symptoms in the same area (4), hypoesthesia to touch or prick in area of the pain (2), and pain caused or increased by brushing in the painful area (1).

The mechanisms underlying neuropathic SCI pain are only partly understood, and it is still unclear why some persons with SCI develop neuropathic pain and others with apparently similar injuries do not.[33] Neuropathic pain is very common five years after SCI: at-level neuropathic pain is present in about 41% and below-level neuropathic pain in about 34% of persons with SCI.[24] Neuropathic pain was found to be the most difficult pain to cope with and was recognised as severely compromising, in both adjustment after injury and quality of life.[19]

Neuropathic SCI pain treatment

The management of SCI pain appears one of the most difficult issues in the care of persons with SCI.[16] Despite impressive gains in limiting bladder, skin, cardiovascular and respiratory complications after SCI, chronic pain post-SCI has proven to be largely refractory to

medical management.[28,34,35] Particularly for chronic neuropathic SCI pain, pharmacological interventions (e.g. antidepressants, anticonvulsants, antispasmodics, anti-inflammatories, and opioid or non-opioid analgesics) are often insufficiently effective in providing significant pain relief,[16,19,30] are problematic in terms of side effects,[19] and many persons are concerned about the consequences of long-term use of these medications.[29] As a result, there is a growing interest in non-pharmacological interventions to reduce neuropathic pain post-SCI, but a lack of research to determine their effectiveness in this group.[29] The high prevalence, severity and impact of chronic neuropathic SCI pain calls for the development and evaluation of non-pharmacological interventions for this type of pain after SCI.

Psychological treatment

One of the non-pharmacological treatment options for neuropathic pain after SCI is psychological treatment. According to the biopsychosocial model of chronic pain, pain usually has an underlying biological basis, but psychosocial factors, such as attitudes and cognitions, emotions, coping responses and behaviours, and the social environment, also have a significant and sometimes profound impact on the experience, maintenance and aggravation of pain.[36-40] Psychosocial factors were even more closely associated with the experience of pain than physiological factors in persons with chronic SCI pain[41] and empirical support for this biopsychosocial perspective for understanding chronic pain in adults with physical disabilities, including persons with SCI, was found.[42] Therefore, both psychosocial and biological factors should be considered in the design of an integrated strategy to manage pain[23,39] and psychological interventions should be added to traditional biomedical interventions for neuropathic SCI pain.[30]

A main type of psychological treatment in multidisciplinary pain programmes for chronic pain in general is cognitive behavioural therapy, aimed at modifying thoughts, beliefs, and behavioural responses to pain and thereby the pain experience.[40] A Cochrane review of psychological therapies for the management of chronic pain in general (excluding headache) concluded that cognitive behavioural therapy is a useful approach to manage chronic pain.[43] Cognitive behavioural therapy, compared with treatment as usual or waiting list control conditions, had small effects on pain and disability, and moderate effects on mood and catastrophising post-treatment. At 6- to 12-months follow-up, however, the only small effect was for mood. Compared with active control conditions, cognitive behavioural therapy showed small benefits for disability and catastrophising post-treatment, but not for pain and mood. At 6- to 12-months follow-up, benefits were found for disability only.

Currently, treatment programmes based upon cognitive behavioural principles are accepted as a cornerstone of effective and relatively low-cost treatment in many chronic pain conditions, like low-back pain.[44,45] However, such programmes are rarely applied in SCI to treat chronic neuropathic pain and evidence of their effectiveness in chronic neuropathic SCI pain is lacking to date.[29,46] Only one controlled study on the effects of a cognitive behavioural pain management programme for persons with chronic neuropathic SCI pain had been published before the start of our study, and it showed positive effects on mood.[47] However, this study was not randomised and included only 27 persons in the intervention group and 11 controls. To our knowledge, no randomised controlled trials (RCTs) on cognitive behavioural interventions for chronic neuropathic SCI pain had been published at the start of our study.

In conclusion, many persons with SCI experience chronic neuropathic pain. This type of pain is a major burden and often difficult to treat. One intervention study showed the potential for comprehensive cognitive behavioural programmes for persons with chronic neuropathic SCI pain, but there was a need for RCTs to evaluate these interventions. Therefore, the CONECSI (COping with NEuropathiC Spinal cord Injury pain) trial was conducted to evaluate the effectiveness of a cognitive behavioural treatment programme for coping with chronic neuropathic SCI pain.

Aims and outline of the present thesis

The general aim of the present thesis is to describe SCI pain treatments and their effectiveness, mainly focusing on a cognitive behavioural programme for coping with chronic neuropathic SCI pain. The following research questions will be answered:

1. What are the pharmacological and non-pharmacological treatments used for chronic SCI pain and which treatments are considered most effective?
2. Does a cognitive behavioural programme result in the short-term in decreased pain intensity and pain-related disability and improved mood, participation in activities, and life satisfaction in persons with chronic neuropathic SCI pain?
3. Does a cognitive behavioural programme result in long-term improvement of pain intensity, pain disability, mood, participation in activities, and life satisfaction in persons with chronic neuropathic SCI pain?

4. Which baseline pain coping and cognitions, and change in pain coping and cognitions are associated with the pain intensity and pain-related disability outcomes of the cognitive behavioural programme?

The answers to these research questions are given in the following six chapters:

Chapter 2 gives an overview of pharmacological and non-pharmacological pain treatments used for chronic SCI pain, and describes the current treatment effectiveness in a cross-sectional study of a large Dutch population with SCI. The protocol of the CONECSI trial and the content of the multidisciplinary cognitive behavioural programme for chronic neuropathic SCI pain are described in **Chapter 3**. **Chapter 4** focuses on the short-term effectiveness of the multidisciplinary cognitive behavioural programme for coping with chronic neuropathic SCI pain in an RCT. In **Chapter 5** are the long-term outcomes of the treatment programme discussed. **Chapter 6** discusses which pain coping and cognitions, and change in pain coping and cognitions are associated with the pain intensity and pain-related disability outcomes of this cognitive behavioural intervention for chronic neuropathic SCI pain. Finally, **Chapter 7** summarises the main findings and discusses what we can conclude with respect to the effectiveness of a cognitive behavioural programme for coping with chronic neuropathic SCI pain. Moreover, this chapter discusses our programme and findings in relation to other cognitive behavioural programmes for SCI pain, cognitive behavioural programmes in other diagnoses with neuropathic pain, as well as methodological considerations, clinical implications, and directions for future research.

References

1. Kirshblum SC, Burns SP, Biering-Sørensen F, Donovan W, Graves DE, Jha A, Johansen M, Jones L, Krassioukov A, Mulcahey MJ, Schmidt-Read M, Waring W. International standards for neurological classification of spinal cord injury (revised 2011). J Spinal Cord Med 2011;34:535-546.
2. Van den Berg ME, Castellote JM, Mahillo-Fernandez I, de Pedro-Cuesta J. Incidence of spinal cord injury worldwide: a systematic review. Neuroepidemiology 2010;34:184-192.
3. Nijendijk JH, Post MW, van Asbeck FW. Epidemiology of traumatic spinal cord injuries in the Netherlands in 2010. Spinal Cord 2014;52:258-263.
4. Vervoordeldonk JJ, Post MW, New P, van Asbeck FW. Rehabilitation of patients with nontraumatic spinal cord injury in the Netherlands: etiology, length of stay, and functional outcome. Top Spinal Cord Inj Rehabil 2013;19:195-201.
5. Wyndaele M, Wyndaele JJ. Incidence, prevalence and epidemiology of spinal cord injury: what learns a worldwide literature survey? Spinal Cord 2006;44:523-529.

6. Van Asbeck FW (Ed). Handboek Dwarslaesierevalidatie [Textbook of spinal cord injury rehabilitation]. Houten: Bohn Stafleu van Loghum; 2007 (in Dutch).
7. Strauss DJ, Devivo MJ, Paculdo DR, Shavelle RM. Trends in life expectancy after spinal cord injury. Arch Phys Med Rehabil 2006;87:1079-1085.
8. Osterthun R, Post MW, van Asbeck FW, Dutch-Flemish Spinal Cord Society. Characteristics, length of stay and functional outcome of patients with spinal cord injury in Dutch and Flemish rehabilitation centres. Spinal Cord 2009;47:339-344.
9. Craig A, Tran Y, Middleton J. Psychological morbidity and spinal cord injury: a systematic review. Spinal Cord 2009;47:108-114.
10. Migliorini CE, New PW, Tonge BJ. Comparison of depression, anxiety and stress in persons with traumatic and non-traumatic post-acute spinal cord injury. Spinal Cord 2009;47:783-788.
11. Post MW, van Leeuwen CM. Psychosocial issues in spinal cord injury: a review. Spinal Cord 2012;50:382-389.
12. Rintala DH, Loubser PG, Castro J, Hart KA, Fuhrer MJ. Chronic pain in a community-based sample of men with spinal cord injury: prevalence, severity, and relationship with impairment, disability, handicap, and subjective well-being. Arch Phys Med Rehabil 1998;79:604-614.
13. Jensen MP, Kuehn CM, Amtmann D, Cardenas DD. Symptom burden in persons with spinal cord injury. Arch Phys Med Rehabil 2007;88:638-645.
14. Widerström-Noga EG, Felipe-Cuervo E, Yezierski RP. Chronic pain after spinal injury: interference with sleep and daily activities. Arch Phys Med Rehabil 2001;82:1571-1577.
15. Turner JA, Cardenas DD, Warms CA, McClellan CB. Chronic pain associated with spinal cord injuries: a community survey. Arch Phys Med Rehabil 2001;82:501-509.
16. Siddall PJ. Management of neuropathic pain following spinal cord injury: now and in the future. Spinal Cord 2009;47:352-359.
17. Wollaars MM, Post MW, van Asbeck FW, Brand N. Spinal cord injury pain: the influence of psychologic factors and impact on quality of life. Clin J Pain 2007;23:383-391.
18. Ehde DM, Jensen MP, Engel JM, Turner JA, Hoffman AJ, Cardenas DD. Chronic pain secondary to disability: a review. Clin J Pain 2003;19:3-17.
19. Henwood P, Ellis JA. Chronic neuropathic pain in spinal cord injury: the patient's perspective. Pain Res Manag 2004;9:39-45.
20. Bloemen-Vrencken JH, Post MW, Hendriks JM, De Reus EC, De Witte LP. Health problems of persons with spinal cord injury living in the Netherlands. Disabil Rehabil 2005;27:1381-1389.
21. Merskey H, Bogduk N (Eds). Classification of chronic pain: descriptions of chronic pain syndromes and definitions of pain terms. Seattle: IASP Press; 1994.
22. Dijkers M, Bryce T, Zanca J. Prevalence of chronic pain after traumatic spinal cord injury: a systematic review. J Rehabil Res Dev 2009;46:13-29.
23. Siddall PJ, Loeser JD. Pain following spinal cord injury. Spinal Cord 2001;39:63-73.

24. Siddall PJ, McClelland JM, Rutkowski SB, Cousins MJ. A longitudinal study of the prevalence and characteristics of pain in the first 5 years following spinal cord injury. Pain 2003;103:249-257.

25. Siddall PJ, Taylor DA, Cousins MJ. Classification of pain following spinal cord injury. Spinal Cord 1997;35:69-75.

26. Siddall PJ, Yezierski RP, Loeser JD. Pain following spinal cord injury: clinical features, prevalence, and taxonomy. Int Assoc for the Study Pain Newslett 2000;3:3-7.

27. Bryce TN, Biering-Sørensen F, Finnerup NB, Cardenas DD, Defrin R, Lundeberg T, Norrbrink C, Richards JS, Siddall P, Stripling T, Treede RD, Waxman SG, Widerström-Noga E, Yezierski RP, Dijkers M. International spinal cord injury pain classification: part I. Background and description. Spinal Cord 2012;50:413-417.

28. Siddall PJ, Middleton JW. A proposed algorithm for the management of pain following spinal cord injury. Spinal Cord 2006;44:67-77.

29. Mehta S, Orenczuk K, McIntyre A, Willems G, Wolfe DL, Hsieh JT, Short C, Loh E, Teasell RW, SCIRE Research Team. Neuropathic pain post spinal cord injury part 1: systematic review of physical and behavioral treatment. Top Spinal Cord Inj Rehabil 2013;19:61-77.

30. Baastrup C, Finnerup NB. Pharmacological management of neuropathic pain following spinal cord injury. CNS Drugs 2008;22:455-475.

31. Jensen TS, Gottrup H, Sindrup SH, Bach FW. The clinical picture of neuropathic pain. Eur J Pharmacol 2001;429:1-11.

32. Bouhassira D, Attal N, Alchaar H, Boureau F, Brochet B, Bruxelle J, Cunin G, Fermanian J, Ginies P, Grun-Overdyking A, Jafari-Schluep H, Lantéri-Minet M, Laurent B, Mick G, Serrie A, Valade D, Vicaut E. Comparison of pain syndromes associated with nervous or somatic lesions and development of a new neuropathic pain diagnostic questionnaire (DN4). Pain 2005;114:29-36.

33. Finnerup NB. Predictors of spinal cord injury neuropathic pain: the role of QST. Top Spinal Cord Inj Rehabil 2007;13:35-42.

34. Cardenas DD, Jensen MP. Treatments for chronic pain in persons with spinal cord injury: a survey study. J Spinal Cord Med 2006;29:109-117.

35. Finnerup NB, Jensen TS. Spinal cord injury pain--mechanisms and treatment. Eur J Neurol 2004;11:73-82.

36. Ehde DM, Jensen MP. Psychological treatments for pain management in persons with spinal cord injury: cognitive therapy and self-hypnosis training. Top Spinal Cord Inj Rehabil 2007;13:72-80.

37. Widerström-Noga EG, Finnerup NB, Siddall PJ. Biopsychosocial perspective on a mechanisms-based approach to assessment and treatment of pain following spinal cord injury. J Rehabil Res Dev 2009;46:1-12.

38. Mathew KM, Ravichandran G, May K, Morsley K. The biopsychosocial model and spinal cord injury. Spinal Cord 2001;39:644-649.

39. Widerström-Noga EG, Turk DC. Types and effectiveness of treatments used by people with chronic pain associated with spinal cord injuries: influence of pain and psychosocial characteristics. Spinal Cord 2003;41:600-609.

40. Turk DC, Swanson KS, Tunks ER. Psychological approaches in the treatment of chronic pain patients--when pills, scalpels, and needles are not enough. Can J Psychiatry 2008;53:213-223.

41. Summers JD, Rapoff MA, Varghese G, Porter K, Palmer RE. Psychosocial factors in chronic spinal cord injury pain. Pain 1991;47:183-189.

42. Jensen MP, Moore MR, Bockow TB, Ehde DM, Engel JM. Psychosocial factors and adjustment to chronic pain in persons with physical disabilities: a systematic review. Arch Phys Med Rehabil 2011;92:146-160.

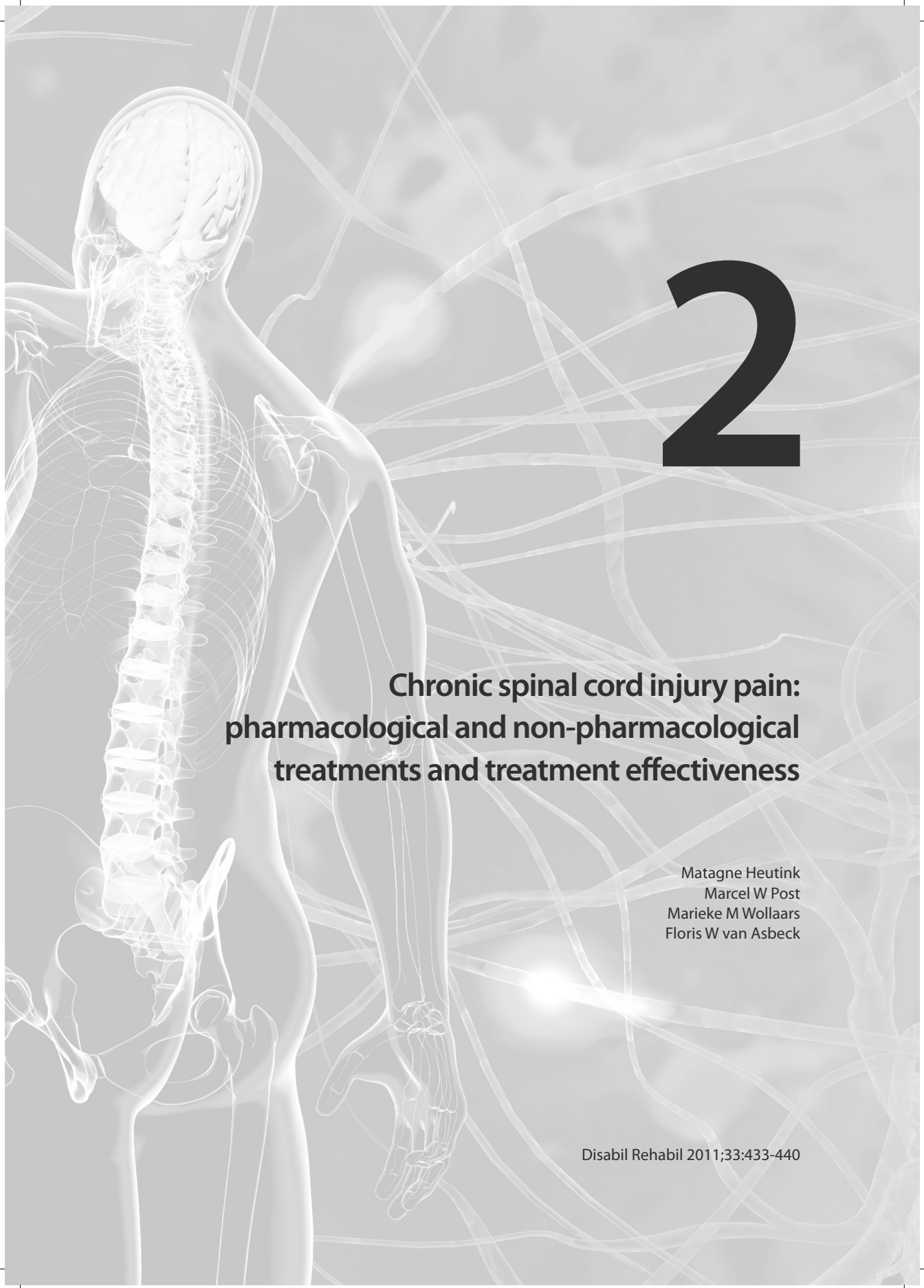
43. Williams AC, Eccleston C, Morley S. Psychological therapies for the management of chronic pain (excluding headache) in adults. Cochrane Database Syst Rev 2012;11:CD007407.

44. Hoffman BM, Papas RK, Chatkoff DK, Kerns RD. Meta-analysis of psychological interventions for chronic low back pain. Health Psychol 2007;26:1-9.

45. Lamb SE, Hansen Z, Lall R, Castelnuovo E, Withers EJ, Nichols V, Potter R, Underwood MR, Back Skills Training Trial investigators. Group cognitive behavioural treatment for low-back pain in primary care: a randomised controlled trial and cost-effectiveness analysis. Lancet 2010;375:916-923.

46. Gault D, Morel-Fatio M, Albert T, Fattal C. Chronic neuropathic pain of spinal cord injury: what is the effectiveness of psychocomportemental management? Ann Phys Rehabil Med 2009;52:167-172.

47. Norrbrink Budh C, Kowalski J, Lundeberg T. A comprehensive pain management programme comprising educational, cognitive and behavioural interventions for neuropathic pain following spinal cord injury. J Rehabil Med 2006;38:172-180.



2

Chronic spinal cord injury pain: pharmacological and non-pharmacological treatments and treatment effectiveness

Matagne Heutink
Marcel W Post
Marieke M Wollaars
Floris W van Asbeck

Disabil Rehabil 2011;33:433-440

Abstract

Purpose: To describe pharmacological and non-pharmacological pain treatments used for chronic spinal cord injury pain (CSCIP) and current treatment effectiveness in a large Dutch population with a spinal cord injury (SCI).

Method: Postal survey among 575 persons with SCI. The main outcome measures were the pain intensity score of the Chronic Pain Grade questionnaire, past and current pain treatments, and perceived effectiveness of current pain treatments.

Results: Response rate was 49% (279 persons) and 215 respondents (77.1%) had CSCIP. Most respondents with CSCIP (62.8%) reported more than one pain type, of which neuropathic pain was most frequently reported (69.3%). Of this group with CSCIP, 63.8% was currently involved in some kind of treatment, but nevertheless high levels of pain (mean 52.8 on a 0–100 scale) were reported. Massage (therapy)/relaxation (training), anticonvulsants, and non-steroidal anti-inflammatory drugs (NSAIDs) were the most often used treatments. The current treatments that were most often perceived as effective were acupuncture/magnetising, cannabis/alcohol, physiotherapy and exercise, and massage (therapy)/relaxation (training). TENS/ultrasound and antidepressants were least often perceived as effective.

Conclusions: Many SCI pain treatments have been tried. Acupuncture/magnetising, cannabis/alcohol, and physiotherapy and exercise were considered most effective. Further research is needed to establish effective SCI pain treatments.

Introduction

Pain secondary to spinal cord injury (SCI) is the most disabling secondary condition for both daily and social activities in people with SCI.[1] Approximately, two thirds of the SCI population experiences chronic SCI-related pain (CSCIP).[2-8] It has a significant impact on physical disability,[9-11] is an important contributor to poorer rehabilitation outcomes,[12] interferes with activities of daily living,[3-7,10,13-18] and has a negative influence on health,[3,13,19] and well being.[20,21] Furthermore, it strengthens feelings of sadness[21] and the risk for depression.[2-4,9,22]

Classifications of SCI pain subtypes have been described on the basis of aetiology and pathophysiology[23] and a broad distinction can be made between musculoskeletal pain, neuropathic pain, and visceral pain. First, musculoskeletal pain, caused by tissue damage of muscles, bones, or joints, is usually described as sharp or dull-aching.[23] It is mostly due to pressure or overuse, and typically located in the shoulders and carpal tunnels.[24] Second, neuropathic pain is described as burning, shooting, stabbing, or tingling, and can be located above, at, or below SCI level.[23] Third, visceral pain is described as burning, cramping, and constant but fluctuating,[25] located in the abdominal and pelvic region. Visceral pain is often poorly localised and vague, and may occur without the presence of abdominal pathology.[23]

A number of studies have mapped out pain treatments in quantity and effectiveness.[4,15,26-29] The pharmacological CSCIP treatments most frequently used were non-steroidal anti-inflammatory drugs (NSAIDs) (16–71%), paracetamol (18–70%), opioids (23–60%), and spasmolytics (17–50%).[4,15,27-29] Of the non-pharmacological treatments transcutaneous nerve stimulation (3–35%), physiotherapy (4–68%), and massage therapy (27–55%) were most often tried.[4,15,27-29] Other pharmacological and non-pharmacological treatments often reported were benzodiazepines, antidepressants, anticonvulsants, biofeedback/relaxation training, psychological treatment, acupuncture, and cannabis. Treatments reported to be most pain alleviating were opioids, anticonvulsants, benzodiazepines, heat therapy, physiotherapy, massage therapy, and cannabis.[4,15,26-29] The best available pharmacological treatments will only provide around one-third of people with a 50% reduction in their pain or have unacceptable side effects.[12] It is still ambiguous which treatment provides most pain relief. Moreover, treatment regimes may differ between countries because of cultural differences or differences in regulations of medicine and approval of new drugs.

Few studies[15,27-29] present a broad overview of pain treatment and treatment effectiveness in a large SCI population. The first aim of the present study is therefore to describe

pharmacological and non-pharmacological pain treatments used for CSCIP in a large Dutch population with SCI. The second aim is to examine the perceived effectiveness of current treatments. This will lead to a better understanding of SCI pain treatments and their effectiveness.

Methods

Subjects and procedure

In total 575 persons with SCI, who received rehabilitation treatment in Rehabilitation Centre "De Hoogstraat" between 1990 and 2005 were approached for the study. All persons above 18 years of age and living in the community were included. A questionnaire was mailed and a digital version was available for those with poor writing skills because of insufficient hand or arm functioning.

Measurement

Demographic and SCI characteristics Demographic characteristics taken into account were age, gender, marital status, highest level of education, time post-injury, cause of injury, and level and completeness of the lesion. Cause of injury was differentiated into traumatic and non-traumatic SCI, and level of lesion into paraplegia and tetraplegia. If a difference existed in sensory and motor level or between the left and right side of the body, the highest motor level of injury was taken as the level of lesion. Lesions were considered complete if they were motor complete (ASIA Impairment Scale[30] grades A or B).

Pain intensity Pain intensity was assessed using the Chronic Pain Grade (CPG) scale.[31,32] The CPG is a useful, reliable, and valid measure of severity of chronic pain.[32] Respondents were asked to rate their current pain, average pain in the past 6 months, and worst pain in the past 6 months on a 0–10 rating scale, resulting in the pain intensity score (0–100). Cronbach's α of the CPG pain intensity score is excellent (0.95).[33]

Pain type Subjects were asked to indicate their type of pain, by making one or more selections of the following: musculoskeletal pain in the back, shoulders, wrists/hands, or elsewhere; muscle spasm pain; neuropathic pain below, above, or at injury level; pain from syringomyelia; visceral pain; and non-SCI related pain. The questionnaire contained descriptions of the different types of pain with examples to help people indicate their type of pain. The duration of pain (no pain, <6 months, >6 months) was questioned to establish

the chronicity of pain. CSCIP was defined as any SCI-related pain lasting longer than 6 months.

Pain treatments Frequently used pain treatments[4-6,26-28,34] were listed in the questionnaire. Respondents were asked which pain treatment(s) they were receiving at the moment of questioning ('current' treatments), and which pain treatment(s) they had received in the past, but that they did not receive anymore ('past' treatments). Respondents were able to fill in treatments not pre-listed. Data analyses revealed that many additional treatments were included. Based on pre-coded treatments and the written responses, the following treatment categories were constructed: anticonvulsants (gabapentin, carbamazepine, clonazepam, and pregabalin), antidepressants (amitriptyline and nortrilen), opioids (morphine, codein, and tramadol), NSAIDs (diclofenac, ibuprofen, and rofecoxib), benzodiazepines (diazepam, oxazepam, and bromazepam), and other medication (e.g. paracetamol). Non-pharmacological treatments were non-invasive physical methods (Transcutaneous Electrical Nerve Stimulation (TENS) and ultrasound), invasive physical methods (injections and neurosurgical treatment), massage (therapy)/relaxation (training), psychological treatments (coping with pain training, cognitive- and/or behavioural therapy, and other psychotherapy concerning pain), acupuncture/magnetising, cannabis/alcohol, physiotherapy and exercise, and other pain treatment. If a respondent had more than one treatment in the same category (e.g. diclofenac and ibuprofen, both NSAIDs), all treatments were counted.

Current pain treatment effectiveness Participants were asked to rate the pain relief they experienced from their current pain treatment(s) on a 3-point scale: 0 (not at all), 1 (somewhat), or 2 (to a large extent). If a respondent had more than one effectiveness score in the same category (e.g. diclofenac and ibuprofen, both NSAIDs), all effectiveness scores were counted.

Analyses

Means, standard deviations, and percentages were calculated for group description and comparison. Two subgroups, of respondents with low pain and high pain intensity, were constructed by splitting the CSCIP group on the median of pain intensity (53.3). Bi-variate associations of pain intensity with other variables were determined with Somers' d. Significance level was set at $p < 0.05$. SPSS statistical program for Windows (version 16.0) was used for the analyses.

Results

Descriptives

Of the 575 questionnaires sent, 279 (49%) were completed and returned. Of all respondents, 215 (77.1%) experienced CSCIP according to our definition. All further analyses were conducted with data of the CSCIP group only. Table 2.1 describes demographic data and SCI characteristics. Age ranged from 25 to 81 years and time post-injury from 0.6 to 60 years.

Table 2.1 Demographic data and SCI characteristics of 215 respondents with CSCIP

Variable	Mean	SD
Age (years)	51.3	14.0
Time post-injury (years)	11.6	10.7
	n	%
Gender		
Men	133	61.9
Women	81	37.7
Not reported	1	0.4
Marital status		
Living alone	90	41.9
Living together	125	58.1
Educational level		
Low (primary, lower vocational, and advanced elementary)	76	35.3
High (intermediate vocational, higher general secondary, and higher vocational/academic)	138	64.2
Not reported	1	0.5
Cause of injury		
Traumatic injury	138	64.2
Non-traumatic injury	74	34.4
Unknown	3	1.4
Level of lesion		
Paraplegic lesion	127	59.1
Tetraplegic lesion	80	37.2
Not reported	8	3.7
Injury completeness		
Complete	92	42.8
Incomplete	120	55.8
Not reported	3	1.4

SCI: spinal cord injury; CSCIP: chronic spinal cord injury pain; SD: standard deviation; n: number of respondents.

Pain intensity and pain type

The pain intensity score ranged from 3.3 to 100 (Mean 52.8 and SD = 23.4). Mean pain intensity in the low pain group was 32.8 (SD = 12.9), and in the high pain group 72.5 (SD = 12.0). As seen in Table 2.2, neuropathic pain was the most frequently occurring SCI pain type, followed by musculoskeletal pain. Most persons with CSCIP reported more than one pain type (62.8%). A total of 34.9% reported to have two, 19.5% three, and 8.3% more than three pain types. In persons with one pain type (37.2%), neuropathic pain was reported most frequently. The most reported combination of pain types was musculoskeletal pain with neuropathic pain (36.7%), with or without other pain types.

Table 2.2 Total SCI pain types, locations, and pain intensity of 215 respondents with CSCIP

Type of SCI pain	n	%	Mean pain intensity	SD pain intensity
Neuropathic pain	149	69.3	56.5	22.2
Above level of injury	11	5.1	59.1	11.6
At level of injury	82	38.1	57.6	22.1
Below level of injury	111	51.6	58.3	21.8
Musculoskeletal pain	131	60.9	50.7	21.9
Back	89	41.4	51.8	21.8
Shoulders	84	39.1	51.3	20.5
Wrists/hands	61	28.4	51.0	19.1
Elsewhere	9	4.2	55.6	16.1
Pain from spasms	76	35.3	52.8	22.7
Visceral pain	65	30.2	52.6	21.8
Pain from syringomyelia	7	3.3	68.6	11.5
Other SCI pain	2	0.9	61.7	7.1

SCI: spinal cord injury; CSCIP: chronic spinal cord injury pain; n: number of respondents; SD: standard deviation.

Table 2.2 also describes the mean pain intensity for all the SCI pain types. Respondents with neuropathic pain and respondents with pain of syringomyelia, with or without any other pain type, were significantly more represented in the high pain intensity group (Somers' d = 0.303, $p < 0.001$; and Somers' d = 0.422, $p = 0.026$, respectively).

Pain treatments

Table 2.3 lists the number of reported past and current pain treatments and the number and percent of patients with CSCIP who reported ever have used or currently using these pain treatments.

Table 2.3 Number of past, current, and total pain treatments and number of respondents using these treatments

	Treatments			Respondents			
	Past	Current	Total	Past	Current	Total	
	n	n	n	n	n	n	%
Pharmacological treatments							
Anticonvulsants	48	34	82	38	32	70	33.8
Antidepressants	25	14	39	25	14	39	18.8
Opioids	32	13	45	31	13	44	21.3
NSAIDs	46	12	58	45	12	57	27.5
Benzodiazepines	32	17	49	32	17	49	23.7
Other medication	7	15	22	7	15	22	10.6
Total	190	105	295	178	103	281	135.7
Non-pharmacological treatments							
TENS/ultrasound	43	9	52	38	8	46	22.2
Injections/neurosurgery	30	11	41	25	10	35	16.9
Massage/relaxation	51	79	130	33	64	97	46.9
Psychological treatment	28	11	39	21	9	30	14.5
Acupuncture/magnetising	48	8	56	39	8	47	22.7
Cannabis/alcohol	19	12	31	18	10	28	13.5
Physiotherapy and exercise	3	18	21	3	18	21	10.1
Other treatment	7	20	27	7	18	25	12.1
Total	229	168	397	184	145	329	158.9
Grand total	419	273	692	362	248	610	294.6

Past: treatments used in the past, but not used anymore; Current: treatments used in the past and (still) using at the time of the survey; n: number; %: percentage of respondents (n = 207) who used one or more of these treatment(s).

Past and current pain treatments A total of 20% of the group had received one or more, up to 8, treatments in the past, but was currently not involved in any pain treatment. A percentage of 15.5 of the group were not involved in any pain treatment ever, at present or in the past. The total amounts, past and current together, show a more frequent use of non-pharmacological treatments (n = 397) versus pharmacological treatments (n = 295). Massage (therapy)/relaxation (training) was most often reported, followed by the anticonvulsants and the NSAIDs. Other treatments, used by more than 20% of the subjects, were the benzodiazepines, TENS/ultrasound, acupuncture/magnetising, and opioids. Cannabis use for pain relief was 24 times reported and alcohol use seven times.

Past pain treatments In the past, 91 respondents (44.0%) had tried one or more pharmacological methods (n = 190), and 89 respondents (43.0%) one or more non-pharmacological methods (n = 229). NSAIDs used were usually diclofenac (used by 20.8%), ibuprofen (1.0%) and rofecoxib (0.5%) were rarely used. Other treatments often tried in the past were acupuncture/magnetising (18.8%), TENS/ultrasound (18.4%), and the anticonvulsants (18.4%). Overall, all treatments were more frequently tried in the past but abandoned, than still used at the time of the study, except massage (therapy)/relaxation (training), physiotherapy and exercise, and other medication and treatment (see Table 2.3). Cannabis use in the past was reported 17 times and alcohol use two times.

Current pain treatments At the moment of questioning, 73 respondents (35.3%) used one or more pharmacological treatments (n = 105) and 98 respondents (47.3%) one or more non-pharmacological treatments (n = 168). In total, 63.8% of the CSCIP group used any kind of treatment: 33.3% were involved in one, 14.5% in two, and 15.9% in three or more pain treatments. A positive association was found between the number of different pain type and pain treatments (value Somers' d = 0.16, p = 0.006).

The most reported current treatment (30.9% of respondents) was massage (therapy)/relaxation (training) (see Table 2.3). The most reported pharmacological treatments (15.5% of respondents) were the anticonvulsants, of which gabapentin was reported most often (n = 22). Other treatment forms were all used by less than 10% of respondents. Cannabis use was reported seven times and alcohol use five times. Some respondents (12.6%) reported no current treatment, but high pain intensity. Ten of this group (4.9%) had never tried any pain treatment, at present or in the past. The remaining 7.8% had tried several treatments in the past.

Current pain treatment effectiveness

Effectiveness of current treatments is described in Table 2.4. Non-pharmacological treatments were overall considered to be more effective than pharmacological treatments. Of all current treatments used more than 13 times (5.1% of current treatments), between 46.9% and 68.8% of users perceived a large extent of effectiveness. Massage (therapy)/relaxation (training) and physiotherapy and exercise were rated effective to a large extent by most current users. Acupuncture/magnetising and cannabis/alcohol were not often used but were also rated very effective (83.3%). The antidepressants and TENS/ultrasound were mostly rated somewhat effective.

Table 2.4 Effectiveness of current treatment

		Not at all	Somewhat	To a large extent
	n	%	%	%
Pharmacological treatments				
Anticonvulsants	32	6.3	46.9	46.9
Antidepressants	13	15.4	46.2	38.5
Opioids	11	9.1	36.4	54.5
NSAIDs	12	–	50.0	50.0
Benzodiazepines	17	–	47.1	52.9
Other medication	13	7.7	53.8	38.5
Non-pharmacological treatments				
TENS/ultrasound	9	33.3	55.6	11.1
Injections/neurosurgery	10	30.0	30.0	40.0
Massage/relaxation	75	2.7	38.7	58.7
Psychological treatment	11	18.2	27.3	54.5
Acupuncture/magnetising	6	–	16.7	83.3
Cannabis/alcohol	12	–	16.7	83.3
Physiotherapy and exercise	16	6.3	25.0	68.8
Other treatment	18	11.1	27.8	61.1

Discussion

We examined pain treatment use and pain treatment effectiveness in a large SCI population. Results showed a diverse use of pain treatments. The most used treatments were massage (therapy)/relaxation (training), anticonvulsants, and NSAIDs. The current treatments that were most often perceived as effective were acupuncture/magnetising, cannabis/alcohol, physiotherapy and exercise, and massage (therapy)/relaxation (training).

Pain type

Another study found similar percentages of respondents with musculoskeletal pain and at-level neuropathic pain.[8] Below-level neuropathic pain in the present study occurred more frequently: 51.6% opposed to 34% found five years after SCI onset.[8] A higher percentage of respondents having more than one pain type corresponded with previous studies.[4,35] Respondents with syringomyelia and neuropathic pain experienced significantly more pain than participants with other pain types. In line with these results, Siddall et al.[8] found neuropathic pain more likely to be described as severe or excruciating.

Pain treatments

Past and current pain treatments As in a study of Norrbrink Budh and Lundeberg,[28] our results show a higher use of non-pharmacological than pharmacological pain treatments. However, other studies show the opposite.[4,26,27] The highly frequent use of massage as a CSCIP treatment is not unusual; other studies also found high percentages.[26,28,29] TENS was reported more,[4,28,29] although other studies found also lower (less than 10%) percentages.[15] The anticonvulsants, of which gabapentin was most often reported, were the most frequently reported pharmacological treatment in our study. Other researchers found varying proportions, ranging from 12 to 67%.[4,27-29] In other studies[4,5,29] more current use of opioids (14–23%), NSAIDs (6–37%), and spasmolytics (12–30%) was reported than in our study (6%, 6% and 2%, respectively). Physiotherapy[26,27,29] and paracetamol[4,27,29] were not pre-listed in our questionnaire, which could explain the lower amounts in comparison with other studies. Cannabis use in our sample is low, but comparable with that in other studies (3–5%).[4,27] It is not a registered drug for neuropathic pain in the Netherlands, which might explain its relatively low use.

Our results support the main findings of others[26,29] that many pain treatments have been tried in the past but were discontinued, probably due to lack of sufficient pain relief or to intolerable side effects. Respondents who reported no treatments in the present despite CSCIP may have tried several treatments in the past without satisfying results, perhaps leading to disbelief in successful pain relief through treatments. Other explanations may be a low pain severity, and the belief and ability to deal successfully with pain and its consequences without treatment. However, over one third of respondents without current treatment (12.6% of the total CSCIP group) reported a high pain intensity score. It is unclear why this group was not involved in any current treatment. Most respondents reported several pain types and treatments, but were not asked to specify which treatment was used for which pain type. However, current alcohol and cannabis use was only found in respondents with neuropathic pain.

Current pain treatment effectiveness

Other studies also found massage (therapy)/relaxation (training)[26-29] and physiotherapy and exercise[26,27,29] to be the most effective forms of treatment, with most users experiencing effectiveness, mostly even to a large extent. Many people find adequate pain relief from opioids.[15,27,29] However, the adverse effects of opioids such as constipation and mental instability can be disturbing, and a risk of substance tolerance is present.[15] Massage (therapy)/relaxation (training), physiotherapy and exercise are therefore preferable.

Current use of acupuncture/magnetising and alcohol/cannabis was low in our study, but rated effective to a large extent. However, other studies found acupuncture rated not to moderately effective by most people.[26,27] Finnerup et al.[5] also found alcohol to be one of the pain alleviating factors. And other studies found cannabis very effective,[27,29] and contributed to significant pain relief for some participants.[4] However, the risk of substance dependence limits recommendation for use in CSCIP.

Pregabalin, a relatively new CSCIP medication at the time of this study, was not used by enough respondents to draw valid conclusions about its effectiveness. It had possibly not been prescribed much at the moment of questioning, although results in this study indicate good effectiveness. Siddall et al.[36] found pregabalin effective in relieving central neuropathic pain. The systematic review of Tzellos et al.[37] indicates the possible efficacy of pregabalin and gabapentin in neuropathic pain following SCI. Although a clear comparison between the two drugs could not be performed, the literature data suggest that pregabalin is more efficacious than gabapentin. But the use of pregabalin is followed by more side effects than gabapentin. Some studies[28,38] suggest a combination of pharmacological and non-pharmacological treatment.

Limitations

A limitation of this study is the self-report method. Lower amounts of controversial treatments might have been reported, with consequentially an overall underrated treatment usage. Some treatments were not pre-listed in the questionnaire, which might have caused an underrating as well. The respondents had to remember all the treatments they tried in the past, which is another reason for less reliable rating. A better method would be a search of medical files, but these are not likely to be complete either and patient might receive treatments by different providers (for example primary care physician, neurologist, or physiatrist). There is also a possibility people did not indicate the right type of pain when they were asked in our mailed questionnaire. And in this study, it was not possible to relate different pain treatments to the different pain types as most people with CSCIP suffer from more than one type of pain at the same time. A final limitation is the response rate of 49%. Such a response rate is unfortunately not unusual for survey studies.[29] It is possible that people with CSCIP were more attracted to participating in this study than people without CSCIP, thereby inflating the prevalence of CSCIP and of pain treatments.

Need for further research

Further research may develop successful CSCIP treatments and improve quality of life for people with CSCIP. Siddall and Middleton[34] have proposed a SCI pain treatment algorithm, in which treatment choice should be made dependent on thorough assessment of pain contributors, generators, and type. Other factors however, such as costs, availability, side effects, drug interactions, and patient preference, may still define treatment choice and effectiveness. This may explain the large number of treatments being tried, and despite differentiation and classification, low effectiveness. There is a need for clinically controlled trials, and additionally, clear treatment guidelines for the CSCIP management.[39] A standardised and prospective registration of the history of pain treatment, pain intensity, and pain type in the SCI population is needed to compare the effectiveness of commonly applied treatment regimes.

In the numerous CSCIP treatment possibilities and attempts, one may consider another treatment approach. Associations between psychosocial factors and CSCIP have been studied, and a good possibility of involvement of coping with pain and the injury has been found in CSCIP experience.[7,11,14,40,41] Counselling[29] and cognitive intervention[42,43] were found to be CSCIP relieving, but this kind of treatment is hardly used in the SCI population. The use of cognitive strategies (such as movement imagery, visual illusions, or sensory discrimination training) to modify the neuropathic SCI pain experience is relatively new and is a possible effective approach for a certain subgroup of patients.[12] Effects and possibilities of cognitive interventions and strategies can be explored using a controlled trial methodology.

References

1. Bloemen-Vrencken JH, Post MW, Hendriks JM, de Reus EC, de Witte LP. Health problems of persons with spinal cord injury living in the Netherlands. Disabil Rehabil 2005;27:1381-1389.

2. Richards JS, Meredith RL, Nepomuceno C, Fine PR, Bennett G. Psycho-social aspects of chronic pain in spinal cord injury. Pain 1980;8:355-366.

3. Rintala DH, Loubser PG, Castro J, Hart KA, Fuhrer MJ. Chronic pain in a community-based sample of men with spinal cord injury: prevalence, severity, and relationship with impairment, disability, handicap, and subjective well-being. Arch Phys Med Rehabil 1998;79:604-614.

4. Ravenscroft A, Ahmed YS, Burnside IG. Chronic pain after SCI. A patient survey. Spinal Cord 2000;38:611-614.

5. Finnerup NB, Johannesen IL, Sindrup SH, Bach FW, Jensen TS. Pain and dysesthesia in patients with spinal cord injury: a postal survey. Spinal Cord 2001;39:256-262.

6. Turner JA, Cardenas DD, Warms CA, McClellan CB. Chronic pain associated with spinal cord injuries: a community survey. Arch Phys Med Rehabil 2001;82:501-509.

7. Turner JA, Jensen MP, Warms A, Cardenas DD. Catastrophizing is associated with pain intensity, psychological distress, and pain-related disability among individuals with chronic pain after spinal cord injury. Pain 2002;98:127-134.

8. Siddall PJ, McClelland JM, Rutkowski SB, Cousins MJ. A longitudinal study of the prevalence and characteristics of pain in the first 5 years following spinal cord injury. Pain 2003;103:249-257.

9. Haythornthwaite JA, Benrud-Larson LM. Psychological aspects of neuropathic pain. Clin J Pain 2000;16:S101-105.

10. Putzke JD, Richards JS, Dowler RN. The impact of pain in spinal cord injury: A case-control study. Rehabil Psychol 2000;45:386-401.

11. Rudy TE, Lieber SJ, Boston JR, Gourley LM, Baysal E. Psychosocial predictors of physical performance in disabled individuals with chronic pain. Clin J Pain 2003;19:18-30.

12. Siddall, PJ. Management of neuropathic pain following spinal cord injury: now and in the future. Spinal Cord 2009;47:352-359.

13. Barret H, McClelland JM, Rutkowski SB, Siddall PJ. Pain characteristics in patients admitted to hospital with complications after spinal cord injury. Arch Phys Med Rehabil 2003;84:789-795.

14. Widerström-Noga EG, Felipe-Cuervo E, Yezierski RP. Relationships among clinical characteristics of chronic pain after spinal cord injury. Arch Phys Med Rehabil 2001;82:1191-1197.

15. Widerström-Noga EG, Turk DC. Types and effectiveness of treatments used by people with chronic pain, associated with spinal cord injuries: influence of pain and psychosocial characteristics. Spinal Cord 2003;41:600-609.

16. Widerström-Noga EG, Duncan R, Turk DC. Psychosocial profiles of people with pain associated with spinal cord injury; identification and comparison with other chronic pain syndromes. Clin J Pain 2004;20:261-271.

17. Donnely C, Eng JJ. Pain following spinal cord injury: the impact on community reintegration. Spinal Cord 2005;43:278-282.

18. Jensen MP, Hoffman AJ, Cardenas DD. Chronic pain in individuals with spinal cord injury: a survey and longitudinal study. Spinal Cord 2005;43:704-712.

19. Ballinger DA, Rintala DH, Hart KA. The relation of shoulder pain and range-of-motion problems to functional limitations, disability, and perceived health of men with spinal cord injury: a multifaceted longitudinal study. Arch Phys Med Rehabil 2000;81:1575-1581.

20. Ville I, Ravaud JF, Tetrafigap Group. Subjective well-being and severe motor impairments: the Tetrafigap survey on the long-term outcome of tetraplegic spinal cord injured persons. Soc Sci Med 2001;52:369-384.

21. Widerström-Noga EG, Felipe-Cuervo E, Broton JG, Duncan RC, Yezierski RP. Perceived difficulty in dealing with consequences of spinal cord injury. Arch Phys Med Rehabil 1999;80:580-586.

22. Jensen MP, Ehde DM, Hoffman AJ, Patterson DR, Czerniecki JM, Robinson LR. Cognitions, coping and social environment predict adjustment to phantom limb pain. Pain 2002;95:133-142.

23. Bockenek WL, Stewart PJ. Pain in patients with spinal cord injury. In: Kirshblum S, Campagnolo DI, DeLisa JA (Eds). Spinal cord medicine. Philadelphia: Lippincott Williams & Wilkins; 2002: pp. 389-408.

24. Siddall PJ, Loeser JD. Pain following spinal cord injury. Spinal Cord 2001;39:63-73.

25. Siddall PJ, Yezierski RP, Loeser JD. Pain following spinal cord injury: clinical features, prevalence, and taxonomy. Int Assoc for the Study Pain Newslett 2000;3:3-7.

26. Dalyan M, Cardenas DD, Gerard B. Upper extremity pain after spinal cord injury. Spinal Cord 1999;37:191-195.

27. Warms CA, Turner JA, Marshall HM, Cardenas DD. Treatments for chronic pain associated with spinal cord injuries: many are tried, few are helpful. Clin J Pain 2002;18:154-163.

28. Norrbrink Budh C, Lundeberg T. Non-pharmacological pain-relieving therapies in individuals with spinal cord injury: a patient perspective. Complement Ther Med 2004;12:189-197.

29. Cardenas DD, Jensen MP. Treatments for chronic pain in persons with spinal cord injury: a survey study. J Spinal Cord Med 2006;29:109-117.

30. Maynard FM Jr, Bracken MB, Creasey G, Ditunno JF Jr, Donovan WH, Ducker TB, Garber SL, Marino RJ, Stover SL, Tator CH, Waters RL, Wilberger JE, Young W. International standards for neurological and functional classification of spinal cord injury. Spinal Cord 1997;35:266-274.

31. Von Korff MR, Ormel J, Keefe FJ, Dworkin SF. Grading the severity of chronic pain. Pain 1992;50:133-149.

32. Smith BH, Penny KI, Purves AM, Munro C, Wilson B, Grimshaw J, Chambers WA, Smith WC. The Chronic Pain Grade questionnaire: validation and reliability in postal research. Pain 1997;71:141-147.

33. Wollaars MM, Post MW, van Asbeck FW, Brand N. Spinal cord injury pain: the influence of psychologic factors and impact on quality of life. Clin J Pain 2007;23:383-391.

34. Siddall PJ, Middleton JW. A proposed algorithm for the management of pain following spinal cord injury. Spinal Cord 2006;44:67-77.

35. Störmer S, Gerner HJ, Gröninger W, Metzmacher K, Follinger S, Wienke C, Aldinger W, Walker N, Zimmermann M, Paeslack V. Chronic pain/dysaesthesiae in spinal cord injury patients: results of a multicentre study. Spinal Cord 1997;35:446-455.

36. Siddall PJ, Cousins MJ, Otte A, Griesing T, Chambers R, Murphy TK. Pregabalin in central neuropathic pain associated with spinal cord injury: a placebo-controlled trial. Neurology 2006;67:1792-1800.

37. Tzellos TG, Papazisis G, Amaniti E, Kouvelas D. Efficacy of pregabalin and gabapentin for neuropathic pain in spinal-cord injury: an evidence-based evaluation of the literature. Eur J Clin Pharmacol 2008;64:851-858.

38. Richards JS. Spinal cord injury pain: impact, classification, treatment trends, and implications from translational research. Rehabil Psychol 2005;50:99-102.

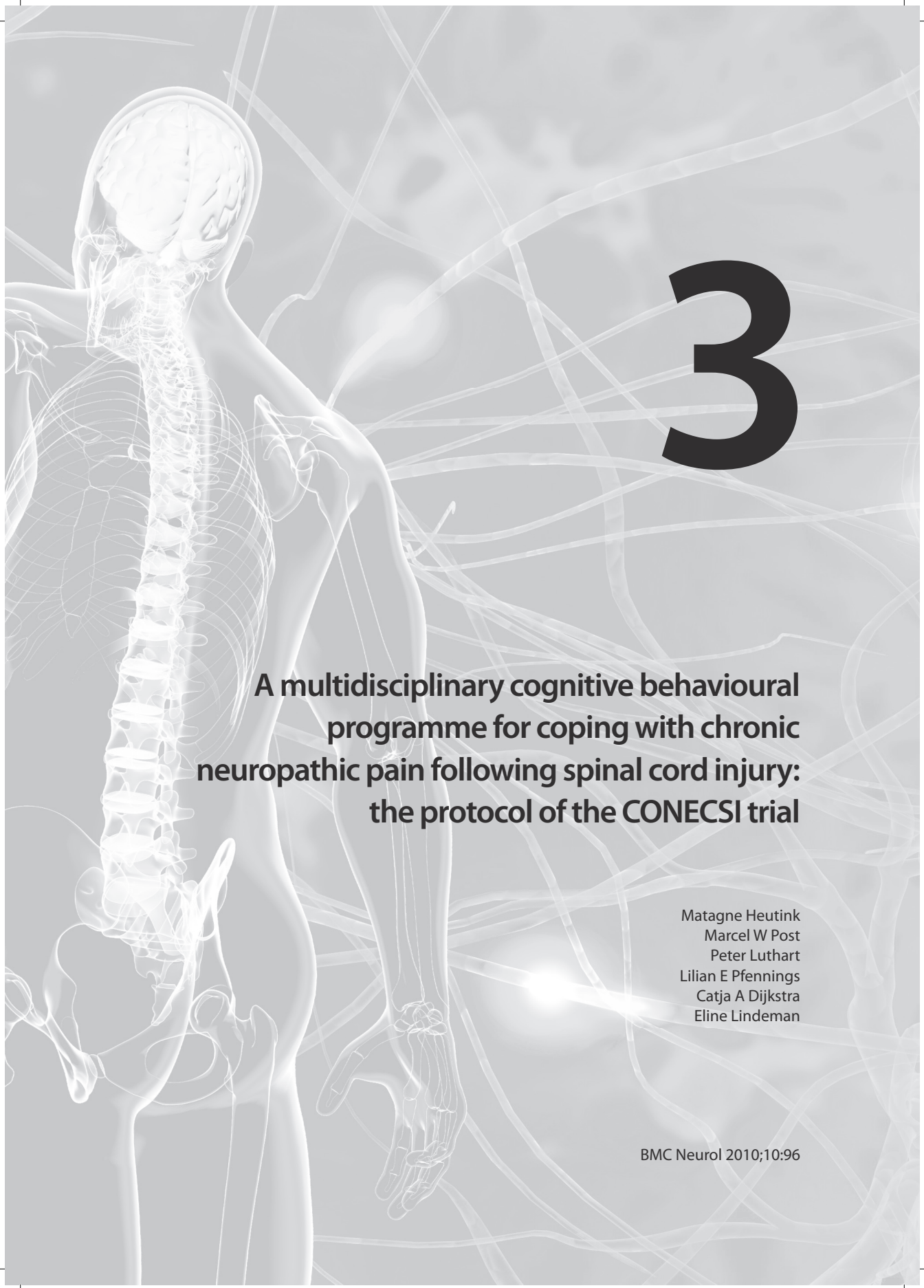
39. Sipski ML, Richards JS. Spinal cord injury rehabilitation: state of the science. Am J Phys Med Rehabil 2006;85:310-342.

40. Summers JD, Rapoff MA, Varghese G, Porter K, Palmer RE. Psychosocial factors in chronic spinal cord injury pain. Pain 1991;47:183-189.

41. Giardino ND, Jensen MP, Turner JA, Ehde DM, Cardenas DD. Social environment moderates the association between catastrophizing and pain among persons with a spinal cord injury. Pain 2003;106:19-25.

42. Ehde DM, Jensen MP. Feasibility of a cognitive restructuring intervention for treatment of chronic pain in persons with disabilities. Rehabil Psychol 2004;49:254-258.

43. Norrbrink Budh C, Kowalski J, Lundeberg T. A comprehensive pain programme comprising educational, cognitive and behavioral interventions for neuropathic pain following spinal cord injury. J Rehabil Med 2006;38:172-180.



3

A multidisciplinary cognitive behavioural programme for coping with chronic neuropathic pain following spinal cord injury: the protocol of the CONECSI trial

Matagne Heutink
Marcel W Post
Peter Luthart
Lilian E Pfennings
Catja A Dijkstra
Eline Lindeman

BMC Neurol 2010;10:96

Abstract

Background: Most people with a spinal cord injury rate neuropathic pain as one of the most difficult problems to manage and there are no medical treatments that provide satisfactory pain relief in most people. Furthermore, psychosocial factors have been considered in the maintenance and aggravation of neuropathic spinal cord injury pain. Psychological interventions to support people with spinal cord injury to deal with neuropathic pain, however, are sparse. The primary aim of the CONECSI (COping with NEuropathiC Spinal cord Injury pain) trial is to evaluate the effects of a multidisciplinary cognitive behavioural treatment programme on pain intensity and pain-related disability, and secondary on mood, participation in activities, and life satisfaction.

Methods/Design: CONECSI is a multicentre randomised controlled trial. A sample of 60 persons with chronic neuropathic spinal cord injury pain will be recruited from four rehabilitation centres and randomised to an intervention group or a waiting list control group. The control group will be invited for the programme six months after the intervention group. Main inclusion criteria are: having chronic (> 6 months) neuropathic spinal cord injury pain as the worst pain complaint and rating the pain intensity in the last week as 40 or more on a 0–100 scale. The intervention consists of educational, cognitive, and behavioural elements and encompasses 11 sessions over a 3-month period. Each meeting will be supervised by a local psychologist and physical therapist. Measurements will be performed before starting the programme/entering the control group, and at 3, 6, 9, and 12 months. Primary outcomes are pain intensity and pain-related disability (Chronic Pain Grade questionnaire). Secondary outcomes are mood (Hospital Anxiety and Depression Scale), participation in activities (Utrecht Activities List), and life satisfaction (Life Satisfaction Questionnaire). Pain coping and pain cognitions will be assessed with three questionnaires (Coping Strategy Questionnaire, Pain Coping Inventory, and Pain Cognition List).

Discussion: The CONECSI trial will reveal the effects of a multidisciplinary cognitive behavioural programme for people with chronic neuropathic spinal cord injury pain. This intervention is expected to contribute to the rehabilitation treatment possibilities for this population.

Trial registration: Dutch Trial Register NTR1580.

Background

Around 65–85% of the people with spinal cord injury (SCI) experience persistent pain and around one-third of these suffer from severe pain.[1] People with SCI consistently rate chronic SCI pain (CSCIP) as one of the most difficult problems to manage, despite the presence of other problems that interfere with daily life,[2] and it is a major impediment to effective rehabilitation.[3] Several types of pain may occur following SCI. In the taxonomy of The Spinal Cord Injury Pain Task Force of the International Association of the Study of Pain, pain types are divided into nociceptive (musculoskeletal or visceral) and neuropathic (above-level, at-level, or below-level) pain.[4] Neuropathic pain is initiated by a primary injury to the nervous system and involves abnormal sensations, such as burning, electric and shooting, and often reduced touch sensation and allodynia.[4] About one third of all people with SCI develop below-level neuropathic pain and this pain type is the most likely type of SCI pain to be described as severe or excruciating.[5]

Our understanding of the mechanisms underlying chronic neuropathic SCI pain (CNSCIP) is still incomplete[1,6,7] and, consequently, treatment often is a matter of trial-and-error.[8] Although two recent pharmacological trials showed some success in alleviating neuropathic pain,[9,10] none of the treatments that are currently available (e.g., pharmacological treatment, physical methods, surgical interventions) relieve pain in the majority of the SCI population.[1,6,11]

The biopsychosocial view provides an integrated model that incorporates mechanical and physiological processes as well as psychological and social/contextual variables that may cause and perpetuate chronic pain.[12] Strong relationships between psychosocial factors and SCI-related pain have been found.[13,14] Results of several studies indicate that pain cognitions like catastrophizing[13,15-18] and pain coping strategies like passive coping[13,17] were significant predictors of pain intensity and pain-related disability. Psychosocial factors were even stronger associated with the experience of pain than physiological factors were.[19] These findings suggest that psychological interventions may be useful to improve the quality of life of persons suffering from CNSCIP.

Cognitive behavioural therapy (CBT) aimed at modifying dysfunctional pain cognitions and coping abilities, has been shown to be effective in people with low back pain[20] and has been successfully applied as a treatment of depression and anxiety in SCI.[21] Interventions like CBT might therefore be successfully applied in SCI rehabilitation as part of a multidisciplinary programme including educational, physical, psychological, and social aspects of pain treatment.[22] Although one small controlled study showed positive effects of a comprehensive CBT programme for people with CNSCIP on mood,[8] no other evaluation studies are available,

so there is insufficient evidence for the effectiveness of cognitive behavioural interventions as a treatment of CNSCIP to date.[23] In conclusion, there is a need for randomised controlled trials (RCT) for the evaluation of cognitive behavioural interventions targeted at coping with CNSCIP.

Aims of the study

The primary aim of the CONECSI (COping with NEuropathiC Spinal cord Injury pain) trial is to evaluate the effectiveness of a multidisciplinary cognitive behavioural programme for coping with CNSCIP. The intervention is expected to result in decreased pain intensity and pain-related disability, and in higher levels of mood, participation in activities, and life satisfaction. The secondary aim is to examine post-hoc (explorative) the influence of demographic and lesion characteristics, pain coping and pain cognitions on the effect of the intervention and the relation between the change of coping and cognitions and the effectiveness of the intervention. It is expected that the intervention will be most effective in persons who show dysfunctional pain cognitions and pain coping strategies before the intervention and in persons who show a change in pain coping and pain cognitions in a positive direction. The third aim is to examine the satisfaction of the participants and trainers about the intervention and to explore which parts will be regarded as effective. This paper describes the design of the CONECSI trial and the content of the programme.

Methods/Design

Study design

A multicentre RCT will be conducted to evaluate the effects of the multidisciplinary cognitive behavioural programme for coping with CNSCIP in four Dutch rehabilitation centres. Within each participating rehabilitation centre, participants will be randomly allocated to the intervention group or to the waiting list control group. The control group will be invited for the programme after a waiting period of six months.

Ethical considerations

The Medical Ethics Committee of the University Medical Centre Utrecht and the participating rehabilitation centres have approved the study protocol. Written informed consent will be obtained from each participant. The trial is registered in the Dutch Trial Register (NTR1580).

Setting

The study will be conducted in four rehabilitation centres with a specialisation in SCI rehabilitation in different parts of the Netherlands: 'De Hoogstraat' in Utrecht; 'Het Roessingh' in Enschede; 'Rijndam' in Rotterdam, and 'Adelante Zorggroep' in Hoensbroek.

Participants and procedure

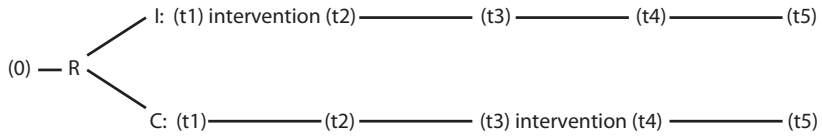
Physiatrists from the four rehabilitation centres select former patients from their centre meeting inclusion criteria 1, 2, and 3 (Table 3.1). The selected persons will be sent a questionnaire to determine if they meet the inclusion (4, 5, and 6) and exclusion (1) criteria. In an interview with the psychologist, who will conduct the programme, will be checked for the other exclusion criteria and will be asked for written consent before final inclusion in the CONECSI trial. Included participants will be randomised (stratified by rehabilitation centre) to the intervention group or the waiting list control group (Figure 3.1). Measurements will be performed in both groups before starting the programme or entering the control group (t1), at 3 (t2), 6 (t3), 9 (t4), and 12 months (t5) follow-up. The control group will be invited for the programme after the follow-up measurement at 6 months.

Table 3.1 In- and exclusion criteria

Inclusion criteria	
(1)	SCI
(2)	at least 18 years old
(3)	at least one year after discharge from first inpatient SCI rehabilitation
(4)	main pain type is neuropathic pain
(5)	duration of neuropathic pain of at least six months
(6)	pain intensity score in the previous week of at least 40 on the 0–100 numerical rating scale of the Chronic Pain Grade
Exclusion criteria	
(1)	SCI caused by metastatic tumour
(2)	previous cognitive behavioural therapy for coping with pain after SCI
(3)	inability to function in a group because of psychopathology
(4)	insufficient mastery of the Dutch language

Intervention

This multidisciplinary programme is developed for people with CNSCIP. The programme consists of ten sessions of three hours over a ten-week period and a comeback session



0:	Inclusion (interview and informed consent)
R:	Randomisation to intervention group (I) and waiting list control group (C) within each participating rehabilitation centre
t1:	Baseline measurement
t2:	Measurement at 3 months
t3:	Measurement at 6 months
t4:	Measurement at 9 months
t5:	Measurement at 12 months

Figure 3.1 Flow chart.

three weeks after the tenth session. Each meeting will be supervised by a psychologist and physiotherapist (the trainers) from the local centre. There will be a maximum of ten participants per group. The programme comprises educational, cognitive, and behavioural elements targeted at coping with CNSCIP. At the first session, participants will receive a course book containing information on all sessions, reading texts, and homework assignments. All participants will have a buddy who will attend two sessions. The buddy is the partner, family member, or a good friend of the participant. The buddy will be asked to read the course material, to help (if necessary) with the homework assignments, and the participants can discuss the intervention with the buddy. The trainers will receive the same course book as the participants, but with an extended protocol for each session. Appendix 3.1 gives an overview of the main contents of the 11 sessions.

Psycho-education Two theoretical models will be used in the programme: the BioPsycho-Social (BPS) model[24] and the Activating event-Belief-Consequence (ABC) model.[25] These two models will be explained in educational sessions and in guided group discussions using fictitious cases. These models will also be applied in sports workshops and homework assignments.

BPS model: According to the BPS model[24] biological, psychological, and social factors contribute to the experience of pain.[6] The model will be used to focus on the influence of psychosocial factors (e.g., beliefs, relationships, stress) on the experience of pain, to clarify

the relationship between biological, psychological, and social factors and pain, and to clarify the importance of maintaining a balance between capacity and load.

ABC model: Rational Emotive Behaviour Therapy (REBT)[25] proposes a 'biopsychosocial' explanation of causation, i.e., a combination of biological, psychological, and social factors are involved in the way humans feel and behave. The ABC model, an element of REBT, is used for cognitive restructuring. The ABC model is based on the fundament that most cases people do not merely get upset by adversities or situations, but also by their views and thoughts, beliefs, attitudes and self-efficacy expectations, about the world, themselves, and others. The model states that it normally is not only an activating event (A) that contributes to disturbed and dysfunctional emotional and behavioural consequences (C), but also what people believe (B) about the activating event (A). In this programme the ABC model will be used to teach participants to become aware of irrational beliefs, dysfunctional pain cognitions, and maladaptive coping, and they will be encouraged to change these thoughts and behaviours. Therefore, the trainers show the participants how to find and dispute (D) their irrational beliefs and dysfunctional pain cognitions and formulate new functional cognitions and rational coping beliefs. And subsequently judge the effect (E, arrive effective new philosophies or rational coping beliefs) of this new cognitions on emotional and behavioural consequences.[26]

Further, the trainers will be assisted by two physiatrists and a role model in three sessions (Appendix 3.1). A local physiatrist specialised in SCI rehabilitation will provide information about SCI, pain physiology and pain classification, pharmacological and non-pharmacological SCI pain treatments, and their limitations. A local physiatrist specialised in chronic pain rehabilitation will provide information about chronic pain and chronic pain rehabilitation for other pain diagnoses than SCI. The role model will tell his/her story of living with CNSCIP and how he/she was able to live his/her life despite the pain.

Workshops Four sessions will include a workshop on relaxation exercises with attention to terms for relaxation, breathing and body sensations (Appendix 3.1). The exercises target on stress reduction, restore balance, attention shift and awareness of available energy. Three sessions will include sports workshops in which exercises in circuit (power) training will be performed (Appendix 3.1). After these workshops the participants will take a closer look at their cognitions during the workshops (e.g., 'I have to work out the best I can' or 'I'm stupid because I'm not good at sports anymore') with the ABC model and at their balance between capacity and load with the BPS model.

Outcome measures

Primary outcome measure will be pain intensity and pain-related disability. Secondary outcome measures will be mood, participation in activities, and life satisfaction. All outcome measures and instrumentation are presented in Table 3.2.

Chronic Pain Grade questionnaire (CPG) The CPG[27] is used to assess pain intensity and pain-related disability. Participants have to rate their pain intensity on a 0–10 Numeric Rating Scale (NRS) for average pain, worst pain, and current pain (pain intensity score), and the degree of pain interference with daily activities, work/household activities, and recreational/social activities (pain-related disability score). The internal consistency for the pain intensity score and the pain-related disability score in an SCI population is excellent (Cronbach's alpha 0.95 and 0.94, respectively).[13] In this study, the CPG is slightly adjusted to ask for neuropathic pain in the past week instead of the past six months.

Table 3.2 Outcome measures and instrumentation

Outcome measures	Instrumentation	t1	t2	t3	t4	t5
Primary outcome measures						
Pain intensity and Pain-related disability	Chronic Pain Grade questionnaire	X	X	X	X	X
Secondary outcome measures						
Mood	Hospital Anxiety and Depression Scale	X	X	X	X	X
Participation in activities	Utrecht Activities List	X	X	X	X	X
Life satisfaction	Life Satisfaction Questionnaire	X	X	X	X	X
Pain coping and pain cognitions						
Pain coping	Coping Strategy Questionnaire	X	X	X^I		X^C
	Pain Coping Inventory	X	X	X^I		X^C
Pain cognitions	Pain Cognition List	X	X	X^I		X^C
Other measures						
Demographic variables (age, gender, marital status, and education)		X				
SCI and pain characteristics		X				
Neuropathic pain treatment		X	X	X	X	X
Functional independence	Barthel Index	X				
Satisfaction with intervention	Questionnaire on satisfaction		X^I		X^C	

X: intenvention group and waiting list control group; X^I: only intervention group; X^C: only waiting list control group.

Hospital Anxiety and Depression Scale (HADS) The HADS is a 14-item self-report measure, which is used for scoring mood. It contains two 7-item scales: one for anxiety and one for depression, both with a score range of 0–21. It is a valid and reliable measure and responsive to change.[28] Woolrich et al. report a good internal consistency within an outpatient population with SCI[29]: the Cronbach's alpha for the anxiety scale is 0.85 and for the depression scale 0.79.

Utrecht Activities List (UAL) The UAL[30,31] is a Dutch adaptation of the Craig Handicap Assessment and Rating Technique (CHART)[32] and is used to assess participation in activities. The questionnaire assesses the time spent on activities such as paid work, study, housekeeping, voluntary work, hobbies, and sports in hours per week, and the number of contacts per week with family, friends and acquaintances, and neighbours.

Life Satisfaction Questionnaire (LiSat-9) The LiSat-9[33] is used to assess life satisfaction and consists of a global item 'life as a whole' and eight domain-specific items 'activities of daily living', 'leisure', 'vocational situation', 'financial situation', 'sexual life', 'partnership relationship', 'family life', and 'contacts with friends'. These nine variables are rated on a six point scale (very dissatisfying to very satisfying), with higher scores reflecting greater satisfaction. The internal consistency of the total score (average of all item scores) is good (Cronbach's alpha of 0.80) in a Dutch SCI population.[34]

Pain coping and pain cognitions

Coping Strategy Questionnaire (CSQ) The Dutch version of the CSQ[35] is an adaptation of the CSQ developed by Rosenstiel and Keefe.[36] The main difference between the original CSQ and the Dutch adaptation is a different answering format. In the Dutch adaptation people mark 10 cm visual analogue scales with the end-points defined in the same way as on the original seven-point Likert scale, i.e., as 'never do' and 'always do'.[35] The CSQ consists of 44 items in eight subscales to assess the use of six different cognitive strategies ('Diverting Attention', 'Re-interpreting Pain Sensations', 'Coping Self-Statements', 'Ignoring Pain Sensations', 'Catastrophizing', and 'Praying/Hoping'), and one behavioural strategy ('Increasing Activity Level'). Participants also rate how effective they think they are in controlling and decreasing pain ('Perceived Effectiveness'). A high score on a particular subscale indicates a higher endorsement of the cognitive coping strategy. The internal consistency is satisfactory to good (Cronbach's alpha range from 0.67 to 0.81) and the stability is reasonable to good (stability coefficients with a time interval of eight weeks range from 0.45 to 0.86).[37]

Pain Coping Inventory (PCI) The PCI consists of 33 items with a four-point Likert scale, from 1 (rarely or never) to 4 (very often). The following six scales are distinguished, divided in active pain coping dimensions: 'Pain Transformation', 'Distraction', 'Reducing Demands', and passive pain coping dimensions: 'Retreating', 'Worrying', and 'Resting', all of which are internally reliable.[38] The internal consistency for the six scales range from 0.62 to 0.79.[39]

Pain Cognition List (PCL-2003) The PCL-2003 consists of 39 items. Each item presents a specific pain cognition statement, for example, 'My thoughts are always concentrated on the pain'. Items are scored on a five-point Likert scale, from 1 (totally disagree) to 5 (totally agree).[40] The questionnaire consists of five subscales ('Catastrophizing', 'Restrictions', 'Optimism', 'Internal Control', and 'Reliance on health care'), with varying length (from 4 to 16 items) and internal consistency (Cronbach's alpha of 0.64 to 0.88 with an average of 0.75), and with correlations between subscales ranging from 0.00 to 0.45. Validity of these subscales is supported by the meaningful pattern of correlations with other relevant constructs.[41] The stability of the PCL-2003 is satisfactory with the Pearson correlation coefficients with a time interval of two weeks ranging between 0.51 ('Reliance on health care') and 0.73 ('Catastrophizing') with an average of 0.64.[41]

Two questionnaires will be used for assessing pain coping, because they cover partly different coping styles and are additional to each other. The PCI assesses passive coping styles (like 'Resting' and 'Retreating', for example 'Slowing down when in pain'), while the CSQ assesses active coping styles (like 'Increasing Activity Level', for example 'I leave my home going to do something, like going to the movies or go shopping') and other coping styles that the PCI does not assess ('Ignoring Pain Sensations', 'Coping Self-Statements', and 'Praying/Hoping'). There is an overlap in the subscale 'Catastrophizing' between the CSQ, PCI, and PCL, but the PCL assesses people's cognitions (like 'I think fate has hit me') instead of coping ('When I'm in pain, I worry all the time about whether the pain will end').

Compliance

In order to conduct this trial in a uniform way in the four rehabilitation centres, a detailed course book and protocol for each session have been written and a one-day training for the trainers will be organized. One of the authors (MH) will monitor on a regular basis (attending two sessions of the intervention group and two sessions of the control group in each centre) whether the content of the intervention will be executed as intended in the four rehabilitation centres. Compliance will be assessed by recording the number of sessions attended.

Neuropathic pain treatments will be registered at all measurements by using a pre-coded list including common pharmacological treatments, physical methods (e.g., TENS), surgical interventions, psychological treatment, alternative treatment (e.g., acupuncture), and other treatments. Participants will be allowed to continue their current pain treatments, but will be asked to refrain, if possible, from starting, stopping, or changing pain treatment during the intervention.

Power analysis

The power analysis is based on information from a previous cross-sectional study,[13] in which a mean pain intensity score on the CPG of 64.7 (SD = 17.0) was seen in the subgroup of patients with a pain intensity score of 40 or higher. A mean difference of 16 points (corresponding to a 25% reduction from score 65 to 49) on the pain intensity score of the CPG between the intervention group and the control group in favour of the intervention group is regarded as clinically relevant. A minimum of 24 persons is required for each arm of the trial (total of 48 persons) to achieve sufficient statistical power (alpha 0.05, beta 0.90, one-tailed test). Expecting a maximum dropout rate of 20%, we aim to include 60 persons.

Data analysis

This trial with repeated measurements nested in each patient will be used to evaluate differences in effect between the intervention group and the waiting list control group. Differences between groups, with 95% confidence intervals, will be calculated for the outcome measures: CPG, HADS, UAL, and LiSat-9, by random coefficient (multilevel) analyses. The effectiveness of the intervention will be tested by analysing differences between the intervention group and the waiting list control group in the course of outcomes over time (0, 3, and 6 months, respectively t1, t2, and t3). Further, the long-term (6, 9, and 12 months, respectively t3, t4, and t5) impact of the intervention will be analysed in participants randomised to the intervention group. The impact of demographic and lesion characteristics, pain cognitions and pain coping on the effectiveness of the intervention will be studied explorative by merging data from the intervention group (t1, t2, t3) and the waiting list group (t3, t4, t5). Both the impact of differences at baseline, and changes in pain cognitions and pain coping during and after the intervention will be post-hoc analysed. Factor rehabilitation centre will be controlled for in the effectiveness analyses, but post-hoc analyses of differences between rehabilitation centres will be analysed and combined with information on compliance and patient satisfaction to identify determinants of a successful

intervention. Data will be analysed according to the intention-to-treat principle. SPSS statistical program for Windows (version 16.0) and the MLwiN program of the Centre for Multilevel Modelling, Institute of Education, University of London (version 2.02) will be used for the analyses. Significance level will be set at a p-value less than 0.05.

Discussion

This multidisciplinary cognitive behavioural programme for coping with CNSCIP will be evaluated in a multicentre trial, in which 60 persons will be randomised to an intervention group or a waiting list control group in four participating rehabilitation centres. The CONECSI trial meets the need for an RCT for the evaluation of CBT for people with CNSCIP, focusing on reduction of negative pain cognitions and promoting adaptive coping. This intervention is expected to contribute to the treatment options for people with CNSCIP, a severe problem for which existing treatments are insufficiently effective.

This study will have the advantage of a randomised control group over other studies like Norrbrink Budh et al.[8] Our use of a waiting list control group that will be invited for the intervention six months after the intervention group has potential advantages. First of all, the people who will be randomised to the waiting list group will also be given the opportunity to participate in the intervention. This prospective might minimize demoralization and enhance participation of people in the waiting list group. Second, the possible impact of pain coping and pain cognitions on the effectiveness of the intervention can be examined in twice as many people.

The intervention consists of an eclectic and multidisciplinary treatment programme: a combination of education, relaxation and activation, and using different theoretical models that are complimentary to each other. If the intervention as a whole is effective, this will make it more difficult to point out the effective elements of the intervention. But we expect to include participants with a variety of pain cognition and pain coping behaviours (for example, an active coping over-user and a passive coping, high catastrophizing under-user) and therefore we choose to present information on different topics and in different ways. The expected advantage is that, this way, each participant will find something useful in the programme.

Most people with SCI have several pain types simultaneously.[42] Therefore, including people who exclusively have CNSCIP is not an option and we decided to include people who experience CNSCIP as their main type of pain. The aim of treating persons with CBT is to

learn them to cope with CNSCIP and not primarily to lower the intensity of neuropathic pain. But we think this might have an effect on the intensity of pain as well, as was shown in persons with chronic low back pain.[20] Further, we use a pain-related disability score and not a generic disability measure to minimize the influence of the paralysis and other secondary conditions of the disability on outcomes of this trial.

If this new intervention turns out to be effective, its chances on implementation in the rehabilitation setting in the Netherlands are good. First, the multidisciplinary design fits well in a rehabilitation setting. Second, the treatment protocol will already be applied in four rehabilitation centres at that time and their experiences with this intervention will facilitate implementation in other Dutch rehabilitation centres. Third, the availability of a written treatment protocol also contributes to applicability in other rehabilitation centres by rehabilitation professionals. Finally, it is important that application of this type of interventions as part of rehabilitation outpatient treatment is eligible for reimbursement by health insurances in the Netherlands.

References

1. Siddall PJ. Management of neuropathic pain following spinal cord injury: now and in the future. Spinal Cord 2009;47:352-359.
2. Widerström-Noga EG, Felipe-Cuervo E, Broton JG, Duncan RC, Yezierski RP. Perceived difficulty in dealing with consequences of spinal cord injury. Arch Phys Med Rehabil 1999;80:580-586.
3. Siddall PJ, Loeser JD. Pain following spinal cord injury. Spinal Cord 2001;39:63-73.
4. Siddall PJ, Middleton JW. A proposed algorithm for the management of pain following spinal cord injury. Spinal Cord 2006;44:67-77.
5. Siddall PJ, McClelland JM, Rutkowski SB, Cousins MJ. A longitudinal study of the prevalence and characteristics of pain in the first 5 years following spinal cord injury. Pain 2003;103:249-257.
6. Widerström-Noga EG, Finnerup NB, Siddall PJ. Biopsychosocial perspective on a mechanisms-based approach to assessment and treatment of pain following spinal cord injury. J Rehabil Res Dev 2009;46:1-12.
7. Wrigley PJ, Press SR, Gustin SM, Macefield VG, Gandevia SC, Cousins MJ, Middleton JW, Henderson LA, Siddall PJ. Neuropathic pain and primary somatosensory cortex reorganization following spinal cord injury. Pain 2009;141:52-59.
8. Norrbrink Budh C, Kowalski J, Lundeberg T. A comprehensive pain management programme comprising educational, cognitive and behavioural interventions for neuropathic pain following spinal cord injury. J Rehabil Med 2006;38:172-180.

9. Rintala DH, Holmes SA, Courtade D, Fiess RN, Tastard LV, Loubser PG. Comparison of the effectiveness of amitriptyline and gabapentin on chronic neuropathic pain in persons with spinal cord injury. Arch Phys Med Rehabil 2007;88:1547-1560.

10. Siddall PJ, Cousins MJ, Otte A, Griesing T, Chambers R, Murphy TK. Pregabalin in central neuropathic pain associated with spinal cord injury: a placebo-controlled trial. Neurology 2006;67:1792-1800.

11. Widerström-Noga EG, Turk DC. Types and effectiveness of treatments used by people with chronic pain associated with spinal cord injuries: influence of pain and psychosocial characteristics. Spinal Cord 2003;41:600-609.

12. Turk DC, Okifuji A. Psychological factors in chronic pain: evolution and revolution. J Consult Clin Psychol 2002;70:678-690.

13. Wollaars MM, Post MW, van Asbeck FW, Brand N. Spinal cord injury pain: the influence of psychologic factors and impact on quality of life. Clin J Pain 2007;23:383-391.

14. Widerström-Noga EG, Felix ER, Cruz-Almeida Y, Turk DC. Psychosocial subgroups in persons with spinal cord injuries and chronic pain. Arch Phys Med Rehabil 2007;88:1628-1635.

15. Turner JA, Jensen MP, Warms CA, Cardenas DD. Catastrophizing is associated with pain intensity, psychological distress, and pain-related disability among individuals with chronic pain after spinal cord injury. Pain 2002;98:127-134.

16. Giardino ND, Jensen MP, Turner JA, Ehde DM, Cardenas DD. Social environment moderates the association between catastrophizing and pain among persons with a spinal cord injury. Pain 2003;106:19-25.

17. Molton IR, Stoelb BL, Jensen MP, Ehde DM, Raichle KA, Cardenas DD. Psychosocial factors and adjustment to chronic pain in spinal cord injury: replication and cross-validation. J Rehabil Res Dev 2009;46:31-42.

18. Raichle KA, Hanley M, Jensen MP, Cardenas DD. Cognitions, coping, and social environment predict adjustment to pain in spinal cord injury. J Pain 2007;8:718-729.

19. Summers JD, Rapoff MA, Varghese G, Porter K, Palmer RE. Psychosocial factors in chronic spinal cord injury pain. Pain 1991;47:183-189.

20. Ostelo RW, van Tulder MW, Vlaeyen JW, Linton SJ, Morley SJ, Assendelft WJ. Behavioural treatment for chronic low-back pain. Cochrane Database Syst Rev 2005:CD002014.

21. Kennedy P, Duff J, Evans M, Beedie A. Coping effectiveness training reduces depression and anxiety following traumatic spinal cord injuries. Br J Clin Psychol 2003;42:41-52.

22. Umlauf RL. Psychological interventions for chronic pain following spinal cord injury. Clin J Pain 1992;8:111-118.

23. Sipski ML, Richards JS. Spinal cord injury rehabilitation: state of the science. Am J Phys Med Rehabil 2006;85:310-342.

24. Engel GL. The need for a new medical model: a challenge for biomedicine. Science 1977;196:129-136.

25. Ellis A. Rational psychotherapy and individual psychology. J Individ Psychol 1957;13:38-44.

26. Ellis A. Can Rational-Emotive Behavior Therapy (REBT) and Acceptance and Commitment Therapy (ACT) resolve their differences and be integrated? J Rat-Emo Cognitive-Behav Ther 2005;23:153-168.

27. Von Korff M, Ormel J, Keefe FJ, Dworkin SF. Grading the severity of chronic pain. Pain 1992;50:133-149.

28. Zigmond AS, Snaith RP. The hospital anxiety and depression scale. Acta Psychiatr Scand 1983;67:361-370.

29. Woolrich RA, Kennedy P, Tasiemski T. A preliminary psychometric evaluation of the Hospital Anxiety and Depression Scale (HADS) in 963 people living with a spinal cord injury. Psychol Health Med 2006;11:80-90.

30. Post MW. Utrechtse Activiteiten Lijst [Utrecht Activities List]. In: Van Asbeck FWA (Ed). Handboek dwarslaesierevalidatie [Textbook of spinal cord injury rehabilitation]. Houten/ Diegum: Bohn Stafleu van Loghum; 1998: pp. 356-357 (in Dutch).

31. Schönherr MC, Groothoff JW, Mulder GA, Eisma WH. Participation and satisfaction after spinal cord injury: results of a vocational and leisure outcome study. Spinal Cord 2005;43:241-248.

32. Whiteneck GG, Charlifue SW, Gerhart KA, Overholser JD, Richardson GN. Quantifying handicap: a new measure of long-term rehabilitation outcomes. Arch Phys Med Rehabil 1992;73:519-526.

33. Fugl-Meyer AR, Brännholm I, Fugl-Meyer K. Happiness and domain-specific life satisfaction in adult northern Swedes. Clin Rehabil 1991;5:25-33.

34. Van Koppenhagen CF, Post MW, van der Woude LH, de Witte LP, van Asbeck FW, de Groot S, van den Heuvel W, Lindeman E. Changes and determinants of life satisfaction after spinal cord injury: a cohort study in the Netherlands. Arch Phys Med Rehabil 2008;89:1733-1740.

35. Spinhoven P, ter Kuile MM, Linssen AC, Gazendam B. Pain coping strategies in a Dutch population of chronic low back pain patients. Pain 1989;37:77-83.

36. Rosenstiel AK, Keefe FJ. The use of coping strategies in chronic low back pain patients: relationship to patient characteristics and current adjustment. Pain 1983;17:33-44.

37. Spinhoven P, ter Kuile MM, Linssen AC. Coping met Pijn Vragenlijst (CPV). Handleiding / experimentele versie [Coping Strategy Questionnaire (CSQ). Manual / experimental version]. Lisse: Swets & Zeitlinger; 1994 (in Dutch).

38. Kraaimaat FW, Evers AW. Pain-coping strategies in chronic pain patients: psychometric characteristics of the pain-coping inventory (PCI). Int J Behav Med 2003;10:343-363.

39. Kraaimaat FW, Bakker A, Evers AW. Pijncoping-strategieën bij chronische pijnpatiënten: De ontwikkeling van de Pijn-Coping-Inventarisatielijst (PCI) [Pain coping strategies in chronic pain patients: The development of the Pain Coping Inventory (PCI)]. Gedragstherapie 1997;3:185-201 (in Dutch).

40. Van Breukelen GJ, Vlaeyen JW. Norming clinical questionnaires with multiple regression: the Pain Cognition List. Psychol Assess 2005;17:336-344.

41. Vlaeyen JW, van Breukelen G, Nooyen-Haazen IW, Stomp-van den Berg SG, Kole-Snijders AM. Meetinstrumenten chronische pijn: deel 4. Pijn Cognitie Lijst-2003. Handleiding [Measuring instruments chronic pain: part 4. Pain Cognition List-2003. Manual]. Maastricht: Pijn Kennis Centrum; 2004 (in Dutch).

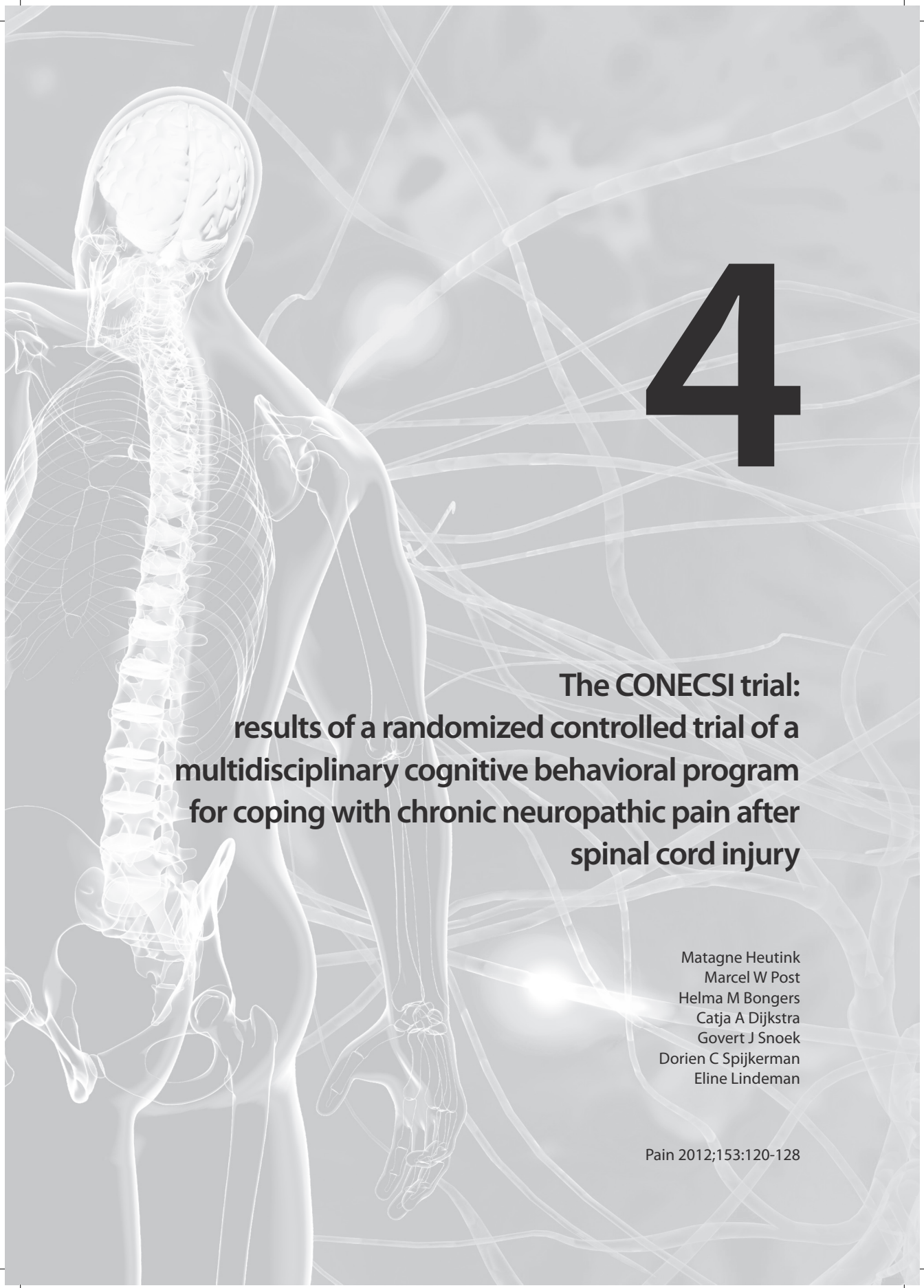
42. Turner JA, Cardenas DD. Chronic pain problems in individuals with spinal cord injuries. Semin Clin Neuropsychiatry 1999;4:186-194.

Appendix 3.1 Main contents of the 11 sessions of the cognitive behavioural programme

Sessions	Main contents
Session 1	Education BPS model Education SCI and CNSCIP Goal setting
Session 2 Buddies	Education ABC model (ABC) Education by physiatrist specialised in SCI rehabilitation
Session 3	Expansion ABC model (ABCDE) Education by physiatrist specialised in chronic pain rehabilitation Education movement and pain
Session 4	Sports workshop Evaluation sports workshop with ABC model
Session 5	Education assertiveness and communication about pain Education by role model Introduction relaxation exercises Evaluation goals
Session 6	Education pain, mood, and stress Workshop relaxation exercises Evaluation workshop relaxation exercises
Session 7	Sports workshop Evaluation sports workshop with BPS model and ABC model
Session 8 Buddies	Education social aspects and partner, family, and friends Workshop relaxation exercises Evaluation workshop relaxation exercises
Session 9	Sports workshop Evaluation sports workshop with ABC model Workshop relaxation exercises Evaluation workshop relaxation exercises
Session 10	Summary and rehearsal contents intervention Evaluation goals Application in daily life
Session 11	Summary contents intervention Evaluation intervention

BPS: BioPsychoSocial; SCI: spinal cord injury; CNSCIP: chronic neuropathic spinal cord injury pain; ABC: Activating event-Belief-Consequence.

3



4

The CONECSI trial: results of a randomized controlled trial of a multidisciplinary cognitive behavioral program for coping with chronic neuropathic pain after spinal cord injury

Matagne Heutink
Marcel W Post
Helma M Bongers
Catja A Dijkstra
Govert J Snoek
Dorien C Spijkerman
Eline Lindeman

Pain 2012;153:120-128

Abstract

Many people with spinal cord injury (SCI) rate chronic neuropathic pain as one of the most difficult problems to manage. The aim of the CONECSI (COping with NEuropathiC Spinal cord Injury pain) trial was to evaluate a multidisciplinary cognitive behavioral treatment program for persons with chronic neuropathic pain after SCI. The intervention consisted of educational, cognitive, and behavioral elements. A total of 61 people were randomized to either the intervention group or the waiting list control group in 4 Dutch rehabilitation centers. Primary outcomes were pain intensity and pain-related disability (Chronic Pain Grade questionnaire), and secondary outcomes were mood (Hospital Anxiety and Depression Scale), participation in activities (Utrecht Activities List), and life satisfaction (Life Satisfaction Questionnaire). Measurements were performed before intervention (t1), after intervention (t2), and at 3 months' follow-up. The primary statistical technique was random coefficient analysis. The analyses showed significant changes over time on both primary (t1–t2), and 2 out of 4 secondary outcomes (both t1–t2 and t1–t3). Significant intervention effects (Time * Group interactions) were found for anxiety and participation in activities, but not for the primary outcomes. Subsequent paired t-tests showed significant changes in the intervention group that were not seen in the control group: decrease of pain intensity, pain-related disability, anxiety, and increase of participation in activities. This study implies that a multidisciplinary cognitive behavioral program might have beneficial effects on people with chronic neuropathic SCI pain.

Introduction

Neuropathic pain is initiated by a primary injury to the nervous system and involves abnormal sensations, such as burning, electric and shooting, and often reduced touch sensation and allodynia.[1] After 5 years at-level neuropathic pain is present in about 41% and below-level neuropathic pain in about 34% of people with spinal cord injury (SCI).[2] Many people with SCI rate chronic neuropathic pain as one of the most difficult problems to manage. None of the treatments that are currently available (e.g., pharmacological treatment, physical methods, surgical interventions) relieve pain in the majority of the SCI population.[3-6] Chronic neuropathic pain is often associated with conditions such as depression and anxiety, and strongly affects daily functioning and overall quality of life.[7,8] According to the biopsychosocial model of chronic pain, pain usually has an underlying biological basis, but psychosocial factors, such as attitudes and cognitions, emotions, coping responses and behaviors, and the social environment, also have a significant and sometimes profound impact on the experience of pain and its effects on physical, psychological, and social functioning.[5,9]

Psychological factors have also been considered in the maintenance and aggravation of chronic neuropathic SCI pain (CNSCIP).[10,11] Therefore, it has been argued that psychological interventions should be added to traditional biomedical interventions.[8]

Psychological interventions aim to modify thoughts, beliefs, and behavioral responses to pain and thereby the pain experience.[12] Cognitive behavioral therapy (CBT) aimed at modifying dysfunctional pain cognitions and coping abilities, has been shown beneficial for chronic pain.[12-14] Coping Effectiveness Training has been successfully applied as a treatment of depression and anxiety in SCI[15] and the benefits of psychological interventions for psychological distress were generally recognized.[16] Interventions like CBT might therefore be useful for people with CNSCIP as part of a multidisciplinary program including educational, psychological, social, and physical aspects of pain treatment,[9,17] but especially for neuropathic pain following SCI, such interventions hardly exist[9,18,19] and evidence of the effectiveness of cognitive behavioral interventions as a treatment for CNSCIP is lacking to date.[20] One controlled study on the effects of a CBT program for people with CNSCIP showed positive effects on mood.[21] However, this study was not randomized and included only 27 patients in the intervention group and 11 controls. Another small non-randomized study on a pain management program for chronic SCI pain in general found a decrease of anxiety and life interference due to pain.[22] To our knowledge, no randomized trials on cognitive behavioral interventions for CNSCIP have ever been published.

In conclusion, there is a need for randomized controlled trials (RCTs) for the evaluation of cognitive behavioral interventions targeted at coping with CNSCIP. The CONECSI (COping with NEuropathiC Spinal cord Injury pain) trial was conducted to evaluate the effectiveness of such a program. This intervention was expected to result in decreased pain intensity and pain-related disability, and improved mood, participation in activities, and life satisfaction for people with CNSCIP.

Methods

Study design

This study was a multicenter RCT. Participants were randomly allocated to the intervention group or to the waiting list control group within each participating rehabilitation center.

The control group was also invited for the program after a waiting period of 6 months. Measurements were performed in both groups before starting the program or entering the control group (before intervention, t1), immediate after intervention (t2), and at 3 months' follow-up (t3). The follow-up measurement was added because we were interested in possible differences between immediate effects (immediately after the intervention) between t1 and t2, and the continued or delayed effects at follow-up (3 months after the intervention) between t1 and t3, for example because the participants have applied in practice for a while what they have learned in the intervention.

Ethical considerations

The Medical Ethics Committee of the University Medical Center Utrecht and the participating rehabilitation centers have approved the study protocol. Written informed consent was obtained from each participant. The trial is registered in the Dutch Trial Register (NTR1580).

Participants and procedure

Participants were recruited from 4 Dutch rehabilitation centers with a specialization in SCI rehabilitation. Eligible persons met the following inclusion criteria: (1) SCI; (2) at least 18 years old; (3) at least 1 year after discharge from first inpatient SCI rehabilitation; (4) main pain type neuropathic pain; (5) duration of neuropathic pain at least 6 months; and (6) pain intensity score in the previous week of at least 40 on the 0–100 numerical rating scale

of the Chronic Pain Grade. Exclusion criteria were: (1) SCI caused by metastatic tumor; (2) previous CBT for coping with pain after SCI; (3) inability to function in a group due to psychopathology; and (4) insufficient mastery of the Dutch language.

Physiatrists from the 4 rehabilitation centers selected former patients from their center meeting inclusion criteria 1, 2, and 3. The selected patients were sent a questionnaire to determine if they met the inclusion criteria 4, 5, and 6 and exclusion criterion 1. A trainer of the intervention (psychologist or nurse practitioner) checked in an interview for the other exclusion criteria before final inclusion in the CONECSI trial.

Intervention

This multidisciplinary program was developed for people with CNSCIP. The program consisted of 10 sessions of 3 hours over a 10-week period and a comeback session 3 weeks after the 10th session. Each meeting was supervised by a psychologist and a physiotherapist (the trainers) from the local center in 3 centers and by a nurse practitioner and a physiotherapist from the local center in 1 center. The program comprises educational, cognitive, and behavioral elements targeted at coping with CNSCIP. At the first session, participants received a course book containing information on all sessions, reading texts, and homework assignments. The buddy (partner, family member, or a good friend of the participant) was asked to attend 2 sessions, to read the course material, to help (if necessary) with the homework assignments, and discuss the intervention with the participant. The trainers received the same course book as the participants, but with an extended protocol for each session. Two theoretical models were used in the program: the BioPsychoSocial (BPS) model[23] and the Activating event-Belief-Consequence (ABC) model.[24] These two models were explained in educational sessions and in guided group discussions using fictitious cases. These models were applied in sports workshops and homework assignments. Table 4.1 provides an overview of the main topics of the 11 sessions. The program is described in more detail elsewhere.[25]

Measurements

Demographic characteristics, functional independence, and pain coping At the baseline measurement (t1), the demographic characteristics taken into account were age, gender, marital status, and highest level of education. The score on the Barthel Index (BI) was registered. The BI is a measure of functional independence. The Dutch self-report BI is based on Collin et al.[26] For this version, a good validity and reliability (Cronbach's α 0.87) was found in people with SCI.[27]

Table 4.1 Main contents in the 11 sessions

Sessions	Main contents
Session 1	Education BPS model Education SCI and CNSCIP Goal setting
Session 2 Buddies	Education ABC model (ABC) Education by physiatrist specialised in SCI rehabilitation
Session 3	Expansion ABC model (ABCDE) Education by physiatrist specialised in chronic pain rehabilitation Education movement and pain
Session 4	Sports workshop Evaluation sports workshop with ABC model
Session 5	Education assertiveness and communication about pain Education by role model Introduction relaxation exercises Evaluation goals
Session 6	Education pain, mood, and stress Workshop relaxation exercises Evaluation workshop relaxation exercises
Session 7	Sports workshop Evaluation sports workshop with BPS model and ABC model
Session 8 Buddies	Education social aspects and partner, family, and friends Workshop relaxation exercises Evaluation workshop relaxation exercises
Session 9	Sports workshop Evaluation sports workshop with ABC model Workshop relaxation exercises Evaluation workshop relaxation exercises
Session 10	Summary and rehearsal contents intervention Evaluation goals Application in daily life
Session 11	Summary contents intervention Evaluation intervention

BPS: BioPsychoSocial; SCI: spinal cord injury; CNSCIP: chronic neuropathic spinal cord injury pain; ABC: Activating event-Belief-Consequence.

The subscale Catastrophizing of the Coping Strategy Questionnaire (CSQ) was registered. The Dutch version of the CSQ ('Coping with Pain Questionnaire')[28] is an adaptation of the CSQ developed by Rosenstiel and Keefe.[29] The main difference between the original CSQ and the Dutch adaptation is a different answering format. In the Dutch adaptation people mark 10 cm visual analogue scales with the end-points defined in the same way as on the original 7-point Likert-type scale, that is, as 'never do' and 'always do'.[28] The Active Coping and Passive

Coping scores of the Pain Coping Inventory (PCI)[30] were also registered. The PCI consists of 33 items with a 4-point Likert scale, from 1 (rarely or never) to 4 (very often), resulting in 6 subscales. The 3 scales: 'Pain Transformation', 'Distraction', and 'Reducing Demands' were add up to the Active Coping score and the three scales: 'Retreating', 'Worrying', and 'Resting' were add up to the Passive Coping score. The satisfaction with the intervention was measured with a questionnaire in the intervention group at t2.

Lesion characteristics SCI characteristics taken into account were time post-injury, cause of injury, and level and completeness of the lesion. Neurological lesion level was defined as the highest motor level. If there was doubt about the answer of the neurological lesion level or the answer was missing, the physiatrist was asked for the neurological lesion level. Neurological levels below T1 were defined as paraplegia, and neurological levels at or above T1 were defined as tetraplegia. Cause of injury was differentiated into traumatic (traffic, work, and sports accident; fall from height; bullet, surgery, and other) and non-traumatic SCI (inflammation, tumor, and other).

Pain type and pain treatment Before inclusion, participants were asked to indicate their pain types (musculoskeletal pain; visceral pain; spasm pain; neuropathic pain below, above, or at injury level; pain from syringomyelia; and non-SCI related pain). The questionnaire contained descriptions of the different types of pain with examples to help people indicate their type of pain. Participants were also asked to answer the questions of the DN4[31] about pain characteristics, and they had to indicate whether neuropathic pain was the main pain type and was present less or longer than 6 months. The physiatrist was inquired if there was still any doubt about the presence (11.5%) or type of neuropathic pain (6.6%). Neuropathic pain treatments were registered at all measurements by using a precoded list with the categories: pharmacological treatment, physical methods (e.g., TENS), surgical interventions, psychological treatment, alternative treatment (e.g., acupuncture), and other treatments.

Primary outcome measures Primary outcomes were pain intensity and pain-related disability measured with the Chronic Pain Grade questionnaire (CPG).[32] Participants rated their pain intensity on a 0–10 numeric rating scale for average pain, worst pain, and current pain (pain intensity score), and the degree of pain interference with daily activities, work/household activities, and recreational/social activities (pain-related disability score). The internal consistency for the pain intensity score and the pain disability score in an SCI population is excellent (Cronbach's α 0.95 and 0.94, respectively).[11] In this study, the CPG has been adapted to ask for neuropathic pain ("The following questions relate to neuropathic

pain due to spinal cord injury. Please circle one number on the scale of 0 to 10") in the past week instead of the past 6 months. The CPG scores at inclusion and at t1 were combined and the mean score was used for t1. For 2 persons with a missing score of the CPG on t2 the mean score of the total group (n = 59) was imputed.

Secondary outcome measures Secondary outcomes were mood measured with the Hospital Anxiety and Depression Scale (HADS),[33] participation in activities measured with Utrecht Activities List (UAL),[34,35] and life satisfaction measured with the Life Satisfaction Questionnaire (LiSat-9).[36,37]

The HADS is a 14-item self-report measure. It contains two 7-item scales: one for anxiety and one for depression, both with a score range of 0 to 21. It is a valid and reliable measure and responsive to change.[33] Woolrich et al. report a good internal consistency within an outpatient population with SCI: the Cronbach's α for the anxiety scale is 0.85 and for the depression scale 0.79.[38]

The UAL[34,35] is a Dutch adaptation of the Craig Handicap Assessment and Rating Technique (CHART).[39] Participation in activities is assessed by the time spent on activities such as paid work, study, housekeeping, voluntary work, hobbies, and sports in hours per week.

The LiSat-9[36] consists of a global item 'life as a whole' and 8 domain-specific items 'activities of daily living', 'leisure', 'vocational situation', 'financial situation', 'sexual life', 'partnership relationship', 'family life', and 'contacts with friends'. These 9 variables are rated on a 6-point scale (very dissatisfying to very satisfying), with higher scores reflecting greater satisfaction. The internal consistency of the total score (average of all item scores) is good (Cronbach's α of 0.80) in a Dutch SCI population.[40]

Compliance

In order to conduct this trial uniformly in the 4 rehabilitation centers, a detailed course book and protocol for each session were written and a 1-day training for the trainers was organized. One of the authors (MH) monitored on a regular basis (attending 2 sessions in each center) whether the content of the intervention was executed as intended. Participant's compliance was assessed by recording the number of sessions attended. Participants were allowed to continue their current pain treatments and were asked not to change, if possible, those pain treatments during the intervention.

Statistical analyses

The power analysis was based on information from a previous cross-sectional study,[11] in which a mean pain intensity score on the CPG of 64.7 (SD = 17.0) was seen in the subgroup of patients with a pain intensity score of 40 or higher. A mean difference of 16 points (corresponding to a 25% reduction from score 65 to 49) on the pain intensity score of the CPG in favour of the intervention group was regarded as clinically relevant. To achieve sufficient statistical power a minimum of 24 patients would be required for each arm of the trial (total of 48 patients). Expecting a maximum dropout rate of 20%, we aimed to include 60 participants.

Participant's characteristics were calculated at t1 (baseline). The pain intensity score and the pain-related disability score of the CPG, the anxiety and depression score of the HADS, total participation in activities of the UAL, and the life satisfaction sum score of the LiSat-9 were calculated for the measurements t1, t2, and t3. Independent t-tests, χ^2 tests, and Mann-Whitney U tests were used to compare group baseline characteristics.

The primary statistical technique to study the effectiveness of the intervention was random coefficient analysis (multilevel analysis). The advantage of random coefficient analysis in longitudinal studies is that the number of observations per person and the temporal spacing of these observations can vary. Furthermore, this method considers dependency of repeated measures within the same person by using random intercepts and corrects for possible differences between rehabilitation centers and persons by allowing random slopes in regression coefficients.[41] The intercept or slope is fixed, unless the -2 log likelihood of the model with a random intercept or slope is significantly lower (the model is better) then the -2 log likelihood of the fixed intercept or slope model. The effectiveness of the intervention was studied with Time (t1–t2 and t1–t3), Group, and Time * Group interactions (t1–t2 and t1–t3) as the determinants. The Time variable and Time * Group interactions were entered in the model as a set of 2 dummy variables with the baseline measurement (t1) as reference. Six models were calculated to study the effectiveness of the intervention, each with 1 of the 2 primary and 4 secondary outcome measures as dependent variable.

If the random coefficient analysis revealed significant results, additional statistical testing was performed using paired t-tests to analyze change between t1 and t2 and between t1 and t3 within each group. Statistical analyses were performed by SPSS statistical program for Windows (version 16.0) and MLwiN program of the Centre for Multilevel Modelling, Institute of Education, University of London (version 2.02). All tests were 2-tailed. Because of 2 co-primary end points, the pain intensity score and the pain-related disability score of

the CPG were tested at the $p < 0.025$ significance level. Likewise, the 4 secondary outcome measures were tested at the $p < 0.0125$ significance level. Data were analyzed according to the intention-to-treat principle.

Results

Participant characteristics

A total of 61 persons were included, 31 participants were randomly allocated to the intervention group and 30 participants to the waiting list control group (see Figure 4.1 for the CONSORT flowchart of inclusion).

A comparison between the intervention group and the control group at the t1 measurement showed no significant differences regarding demographic characteristics and level of functional independence. The level of the SCI did not differ between the 2 groups, the completeness of SCI, however, differed between the 2 groups, with a higher proportion of persons with a complete SCI in the intervention group than in the control group (Table 4.2). The mean age of participants at t1 was 58.8 years (SD = 11.4). The median time between the onset of SCI and inclusion was 5.4 years (range 1.4–23.7) and the median duration of CNSCIP at inclusion was 4.5 years (range 1.3–23.7). The mean level of functional independence was 13.2 (SD = 5.8) on a scale of 0 to 20. The mean number of self-reported pain types was 2.5 (SD = 1.2). More men than women participated in this study, and the majority had a traumatic SCI, paraplegia, and lived with a spouse (Table 4.2). No significant differences in catastrophizing on the CSQ, and in active and passive coping on the PCI were found between the intervention group and control group at baseline.

Analyses of the primary outcome measures

No pre-intervention differences on the CPG scores between the intervention group and control group were found. Random coefficient analysis showed that pain intensity and pain-related disability decreased between the time period t1 and t2 (Table 4.3, Figures 4.2 and 4.3). No significant main effect for group or Time * Group interaction effects were found (Table 4.3). For pain-related disability, the interaction effect between time (period t1 and t2) and group was just outside significance. Both the decrease in pain intensity and in pain-related disability between t1 and t2 were significant in the intervention group, but not in the control group, and no significant changes between t1 and t3 were found (Table 4.4).

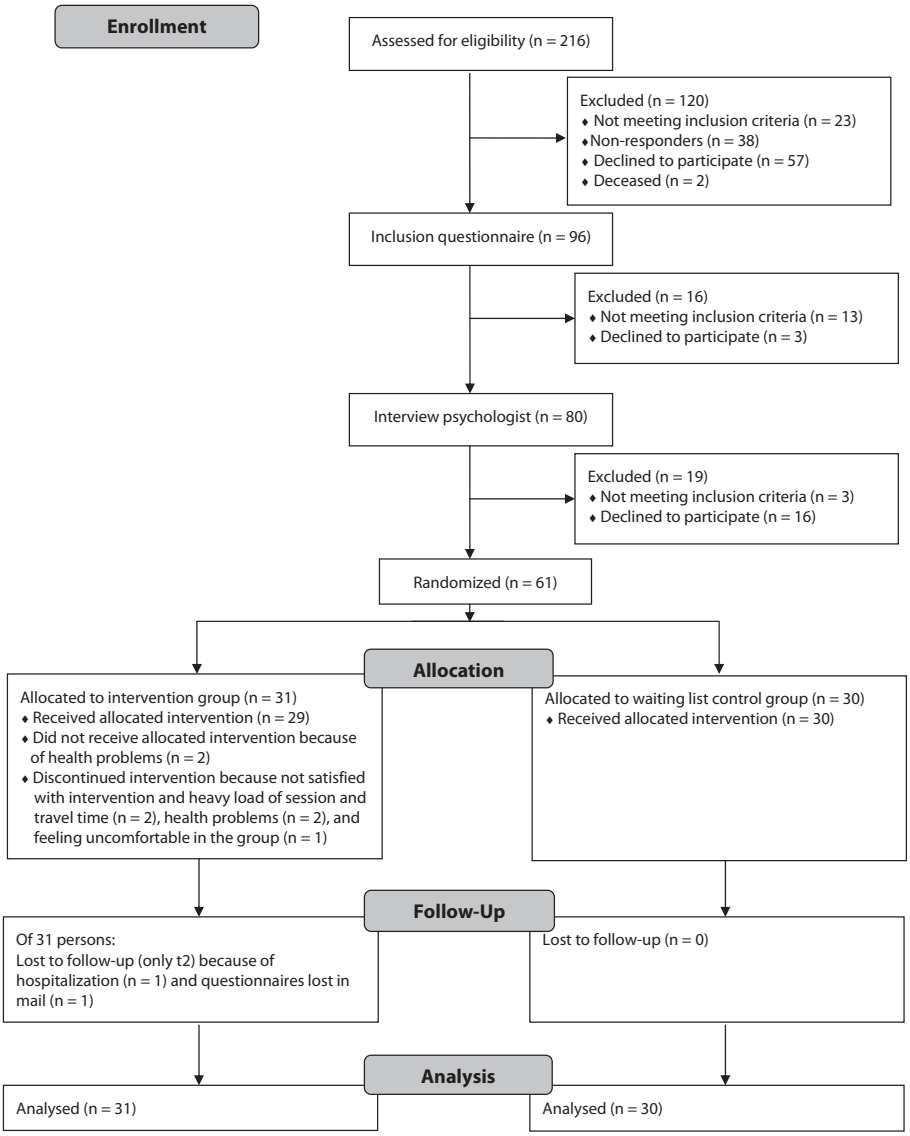


Figure 4.1 CONSORT flowchart of the recruitment and inclusion process.

Analyses of the secondary outcome measures

No significant differences in secondary outcome measures between the 2 groups were found at baseline, except for the UAL ($t(59) = -2.731$), $p = 0.008$), with a higher initial participation level in the control group ($M = 47.0$ hours a week, $SD = 21.2$) than in the intervention group ($M = 31.7$ hours a week, $SD = 22.7$).

Table 4.2 Baseline characteristics of the intervention group (n = 31) and the control group (n = 30), and independent t-tests

Characteristics	Intervention group		Control group		
	n	%	n	%	p
Gender					0.529
Male	21	67.7	18	60.0	
Female	10	32.3	12	40.0	
Marital status					0.525
Married/living with a spouse	25	80.6	26	86.7	
Not living together	6	19.3	4	13.3	
Education					0.517
Low	12	38.7	7	23.3	
Middle	12	38.7	13	43.3	
High	7	22.6	10	33.3	
Cause of injury					0.132
Traumatic	25	80.6	19	63.3	
Non-traumatic	6	19.4	11	36.7	
Lesion level					0.457
Paraplegia	20	64.5	22	73.3	
Tetraplegia	11	35.5	8	26.7	
Lesion completeness					0.010
Incomplete	15	48.4	24	80.0	
Complete	16	51.6	6	20.0	
	M	SD	M	SD	p
Pain coping and cognitions					
Catastrophizing (CSQ)	3.2	2.2	2.5	1.9	0.293
Active Coping (PCI)	24.6	4.0	26.1	4.3	0.227
Passive Coping (PCI)	42.0	9.8	42.2	11.3	0.965

n: number of respondents; p: p-value for difference between intervention group and control group; M: mean scale score on the CSQ and mean score on the PCI; SD: standard deviation; CSQ: Coping Strategy Questionnaire; PCI: Pain Coping Inventory.

The HADS depression and the LiSat-9 scores remained stable over time (Table 4.3).

The HADS anxiety score showed significant change across t1–t2 and t1–t3, and significant Time * Group interaction across t1–t2 (Table 4.3). The anxiety score of the intervention group showed a significant decrease between t1 and t2, and between t1 and t3, while the anxiety score of the control group showed no significant change (Table 4.4, Figure 4.4).

The UAL scores also changed significantly across t1–t3 with significant Time * Group interaction across t1–t2 (Table 4.4). The intervention group showed a significant increase between t1 and t2, and t1 and t3 in participation while no significant changes were found in the control group (Table 4.4, Figure 4.5).

Table 4.3 Multilevel linear regression models for the primary and secondary outcome measures: before intervention, after intervention, and at 3 months' follow-up; group comparison; and interaction effects (n = 61)

	Model for pain intensity			Model for pain-related disability		
Variables	β	SE	p	β	SE	p
Constant	69.258	1.985		47.936	4.378	
Time (t1–t2)	-4.161	1.590	0.009*	-9.903	2.805	<0.001*
Time (t1–t3)	-2.613	2.022	0.196	-9.000	4.719	0.056
Group	0.109	3.188	0.973	-1.336	6.243	0.834
Time (t1–t2) * Group	1.995	2.278	0.381	7.570	4.000	0.059
Time (t1–t3) * Group	-0.487	2.884	0.866	5.200	6.729	0.441
	Model for participation in activities			Model for anxiety		
Variables	β	SE	p	β	SE	p
Constant	31.677	3.650		6.903	0.645	
Time (t1–t2)	7.724	3.288	0.019	-1.426	0.435	0.001**
Time (t1–t3)	9.410	3.247	0.004**	-1.172	0.430	0.006**
Group	15.356	5.205	0.003**	-1.420	0.927	0.126
Time (t1–t2) * Group	-13.086	4.628	0.005**	1.667	0.613	0.007**
Time (t1–t3) * Group	-8.036	4.658	0.084	1.310	0.609	0.032
	Model for depression			Model for life satisfaction		
Variables	β	SE	p	β	SE	p
Constant	7.129	0.559		4.130	0.177	
Time (t1–t2)	-0.627	0.430	0.145	-0.201	0.143	0.159
Time (t1–t3)	-0.386	0.424	0.363	0.012	0.141	0.932
Group	-0.896	0.798	0.261	0.249	0.216	0.249
Time (t1–t2) * Group	-0.039	0.605	0.949	0.167	0.200	0.404
Time (t1–t3) * Group	-0.486	0.601	0.419	0.057	0.198	0.773

Note 1: Beta (β) stands for a non-standardised regression coefficient in multilevel analyses.
Note 2: All models had random intercepts.
Note 3: All models had fixed slopes, except for the Time (t1–t3) covariates of pain intensity and pain-related disability, group covariates of pain intensity and life satisfaction, and Time (t1–t2) * Group covariate of pain intensity.
t1: measurement 1, before intervention; t2: measurement 2, immediate after intervention; t3: measurement 3, 3 months' follow-up; SE: standard error.
* Statistically significant at $p < 0.025$.
** Statistically significant at $p < 0.0125$.

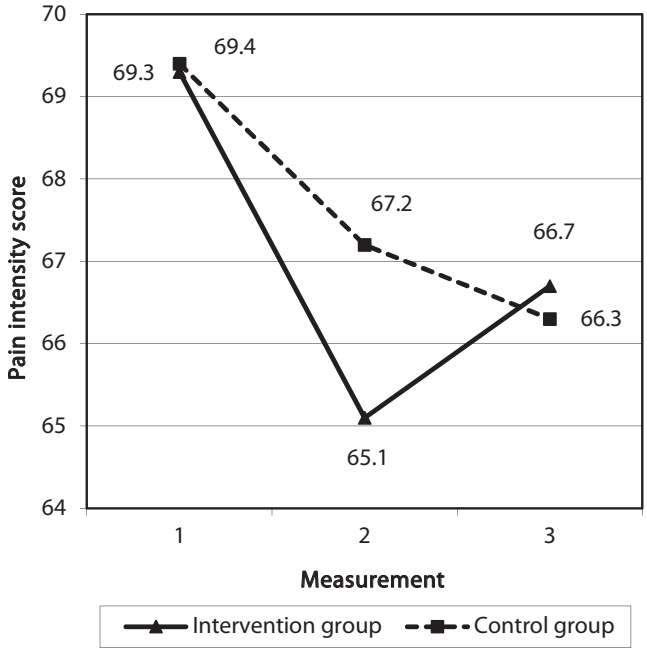


Figure 4.2 Pain intensity score of the CPG.

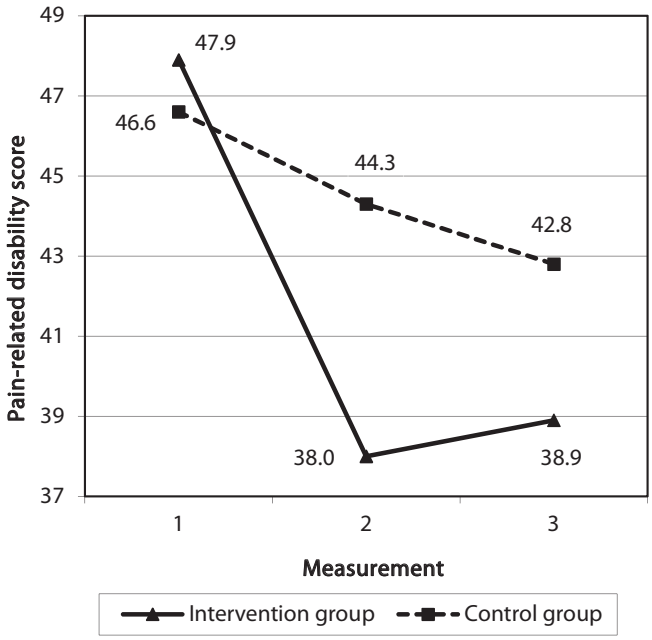


Figure 4.3 Pain-related disability score of the CPG.

Table 4.4 Mean scores and standard deviations before intervention, after intervention, and at 3 months' follow-up; independent-samples t-test at baseline; and paired t-tests for the significant results of the random coefficient analysis

Outcome measure	Before (t1)	After (t2)	Follow-up (t3)	p baseline difference	p (t1–t2)	p (t1–t3)
Pain intensity				0.949		
Intervention	69.2 (9.6)	65.2 (12.7)	66.7 (13.0)		0.019*	0.496
Control	69.4 (13.9)	67.2 (16.0)	66.3 (17.3)		0.174	0.109
Pain-related disability				0.816		
Intervention	48.0 (22.1)	38.0 (25.4)	38.9 (24.5)		0.002*	0.092
Control	46.6 (23.9)	44.2 (27.6)	42.8 (27.5)		0.389	0.336
Anxiety				0.155		
Intervention	6.9 (4.1)	5.6 (3.6)	5.9 (3.6)		0.007*	0.027*
Control	5.5 (3.4)	5.7 (3.4)	5.6 (3.6)		0.578	0.740
Participation in activities				0.008*		
Intervention	31.6 (22.7)	39.3 (20.4)	41.5 (17.5)		0.034*	0.008*
Control	47.0 (21.2)	42.6 (21.1)	51.0 (19.3)		0.127	0.900

Note: The mean scores counted in the software package SPPS differ slightly from the scores in the figures with the software package MlwiN-estimated values.
t1: measurement 1, before intervention; t2: measurement 2, immediate after intervention; t3: measurement 3, 3 months' follow-up.
* Statistically significant at $p < 0.05$.

Attendance and satisfaction

Seven people did not complete the intervention (dropout rate 11.5%). Two participants stopped before the start of the intervention, 1 participant discontinued after 1 session, and 1 participant after 2 sessions, all because of health problems not related to the intervention. The other 3 participants discontinued after 3 sessions (2 persons) and 5 sessions (1 person; for reasons see Figure 4.1). These participants were not included in the evaluation of participants' satisfaction because they did not attend at least half of the sessions. The other participants of the intervention group attended 84.2% of all sessions (M = 9.3 out of 11, SD = 1.7). Most participants (83.3%) were accompanied by their buddy in the buddy meetings. Participants generally seemed satisfied with the program. In the evaluation at t2, the intervention group rated overall satisfaction with the program with a mean of 7.6 (SD = 0.6) on a scale of 1 to 10. No participants reported the intervention to be not helpful, 45.8% somewhat helpful, 50.0% helpful, and 4.2% very helpful. Seventy-five percent of the participants reported they had developed a slightly better way to cope with the pain and 25% had developed a better way to deal with the pain. Most participants (66.7%) reported the intervention met their expectations and the participants would recommend the program to others. It was

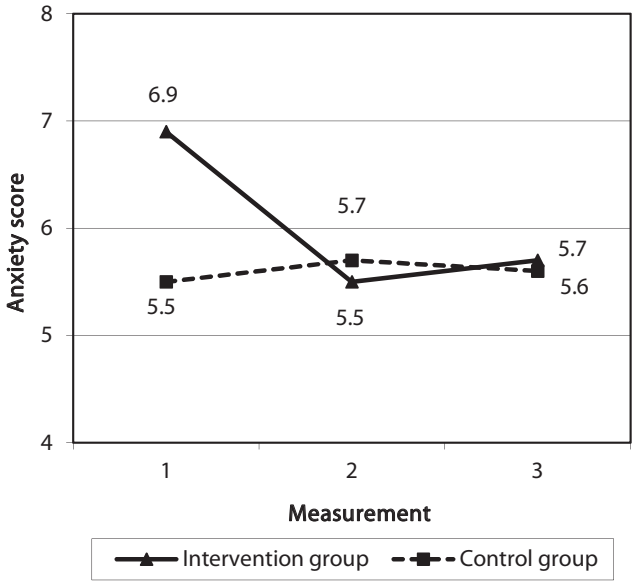


Figure 4.4 Anxiety score of the HADS.

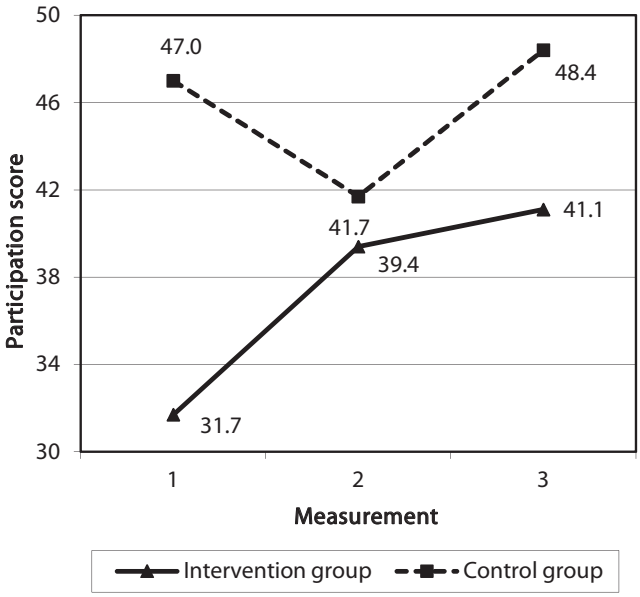


Figure 4.5 Participation score of the UAL.

suggested that it would be good to offer the program earlier after SCI. Most participants judged the total number of sessions, the frequency (once a week) and the length of each session adequate.

Discussion

To our knowledge, this is the first reported RCT to evaluate a multidisciplinary cognitive behavioral program for coping with CNSCIP. On the primary outcome measures of pain intensity and pain-related disability, no intervention effects (significant Time * Group interaction) were found, although the short-term intervention effect of pain-related disability was just outside significance, and the intervention group showed a significant decrease in pain intensity and pain-related disability between t1 and t2. On the secondary outcome measures, significant treatment effects were found for anxiety and participation in activities, showing that a multidisciplinary cognitive behavioral program for coping with CNSCIP might result in a reduction of anxiety and increase of participation in activities in people with CNSCIP.

To our knowledge, only one intervention study of a comprehensive pain management program for CNSCIP is available for comparison with our results. Norrbrink Budh et al.[21] found changes in pain intensity and life satisfaction, but these changes were similar for the treatment and the control group. Further, both anxiety and depression decreased in the intervention group, and a tendency towards better quality of sleep was seen. In the current study also a decrease of pain intensity and anxiety but no decrease of depression and life satisfaction were found, and also positive, but non-significant, changes in the control group were seen. A multidisciplinary cognitive behavioral pain management program for SCI-related chronic pain also found improvement in anxiety in the treatment group, but no significant differences with an usual care group, and no significant decrease in pain intensity and depression.[22] Other controlled studies on CBT for persons with SCI, aimed at other domains than pain, previously reported reduction in depression and anxiety,[15,42] but no significant differences between the intervention and the control groups.

Explanations of the results

This study found a small decrease of pain intensity, but no intervention effects on pain intensity. The decrease of pain intensity in the intervention group was 6%, which is far below the desired 25% that was the basis for the power analysis. One possible explanation for this

lack of effect is that the CPG might lack responsiveness in persons with SCI. However, a 0–10 point numerical rating scale, like the CPG, is recommended as the outcome measure for pain intensity after SCI.[43] Second, most persons with SCI experience multiple pain types.[3,44] However, for the same reason it was not possible to include only people who exclusively had CNSCIP, and therefore, all persons who experienced CNSCIP as their main type of pain were included. The most likely explanation of the lack of effect on pain intensity is that the biopsychosocial approach is aimed at coping with CNSCIP and not primarily aimed at pain reduction,[21] so that improvement of pain behaviors is more likely than decrease of pain itself. We therefore also used the pain-related disability score of the CPG as a primary outcome measure. A substantial (21%) and significant decrease of pain-related disability in the intervention group was seen. The intervention effect on pain-related disability was borderline significant, suggesting that a replication of this study with a slightly larger study population might reveal significant beneficial effects.

Another explanation might be that persons with chronic SCI already have developed appropriate pain cognitions and pain coping strategies. However, catastrophizing scores of participants were only slightly below those of persons with chronic low back pain or neck pain,[45] and pain coping scores were about the same as in persons with low back pain.[46,47] Nevertheless, it might be possible that the intervention is more effective in persons with high levels of catastrophizing, lower level of active coping, and higher level of passive coping at baseline.

The intervention resulted in a decrease in level of anxiety. Possibly, the intervention stimulated participants toward a more positive appraisal of their situation and, thereby, reduced feelings of anxiety. Restructuring dysfunctional cognitions according to the ABC model[24] is an important part of the intervention. Other cognitive-behavioral interventions also showed a reduction of anxiety.[15]

In line with the decrease in pain-related disability, the intervention group showed a significant increase in participation in activities. This was expected, as the intervention also focused on participation in activities.

No decrease of depression and life satisfaction were found. At baseline, depression scores were not very high and life satisfaction scores were not very low, which might explain the stability of these scores. Other CBT interventions showed a significant improvement of mood in persons with SCI whose levels of anxiety and depression were initially high.[22,42]

Finally, besides the changes in scores on the primary and secondary outcome measures in the intervention group, we see a trend in the scores of the control group in the same direction.

An explanation for the changes in scores of the control group can be the 'Hawthorne-effect', the effect on the outcome measures for being in an RCT and perspective to the upcoming program, get attention from the researchers and complete questionnaires about important aspects related to the intervention.

Limitations of the study

A possible limitation of the present study is the small number of included persons. This number should have been sufficient, given the power analyses based on a clinically relevant difference of 25% reduction of pain. In this study, however, a 6% decrease of pain intensity and a 21% reduction of pain-related disability were found. The chosen 25% decrease is nevertheless reasonable because in a group with SCI and lower limb amputation an even larger 33% decrease was found as a reasonable standard for meaningful change across chronic pain conditions.[48] A further disadvantage of a small patient group is the greater chance on baseline differences on outcome parameters despite randomization, as was found for participation. The reason for the difference in participation at the start of the intervention is unclear. However, as expected, no significant changes were found in the control group between t1–t2 and t1–t3. A final limitation might be the limited number of 33 contact hours of this intervention. An optimal dose of over 100 hours has been proposed for CBT in chronic back pain patients,[49] and other CBT programs for SCI pain comprised 45 contact hours[22] and 50 hours of treatment.[21] An intensified intervention might have shown better results, although the number and length of the sessions were judged adequate by the participants. An evaluation meeting with the trainers who executed the study neither showed a desire for a higher intensity, although it was suggested to divide the intervention over two more sessions to loosen the schedule of the sessions a bit.

Clinical implications

Although, the evidence is not unambiguous, this study implies that a multidisciplinary cognitive behavioral program might have beneficial effects on people with CNSCIP, specifically on reduction of anxiety and increase of participation, and possibly on pain-related disability. The participants would recommend the program to others, and many participants suggested that it would be good to offer the program earlier after SCI. Even small changes might be relevant to participants, considering that most people in this study had tried many different treatments for CNSCIP before entering this program. Although, education parts of this program were focused on neuropathic pain, other elements would be applicable to

people with other types of SCI-related pain as well. With relatively minor adaptations, the program could also be offered to all persons with SCI who experience chronic pain and be tested for its effectiveness in such a broader group.

Need for further research

There is a need for further research on the effectiveness of CBT-based interventions for persons with CNSCIP, with larger patient groups and earlier after, or even in, the first inpatient SCI rehabilitation period. Second, it is recommended to investigate for which patients (taken into account demographic characteristics, SCI and pain characteristics, personal characteristics, pain coping and pain cognitions at baseline) the intervention is most effective and which elements of the program are most effective. Finally, the long-term effects of the intervention need to be investigated.

Acknowledgements

The authors would like to thank the participants in this study, the participating Dutch rehabilitation centers, and the trainers in these centers: Peter Luthart and Lilian Pfennings (Rehabilitation Center De Hoogstraat, Utrecht); Conny Overdulve and Sandra Slangen (Adelante Zorggroep, Hoensbroek); Marijke Schuitemaker and Wim van de Vis (Rehabilitation Center Het Roessingh, Enschede); Lonneke Vlemmix, Hilde Schors, Jolante Sweers, and Nicole Vrijens (Rehabilitation Center Rijndam, Rotterdam).

References

1. Siddall PJ, Middleton JW. A proposed algorithm for the management of pain following spinal cord injury. Spinal Cord 2006;44:67-77.
2. Siddall PJ, McClelland JM, Rutkowski SB, Cousins MJ. A longitudinal study of the prevalence and characteristics of pain in the first 5 years following spinal cord injury. Pain 2003;103:249-257.
3. Heutink M, Post MW, Wollaars MM, van Asbeck FW. Chronic spinal cord injury pain: pharmacological and non-pharmacological treatments and treatment effectiveness. Disabil Rehabil 2011;33:433-440.
4. Siddall PJ. Management of neuropathic pain following spinal cord injury: now and in the future. Spinal Cord 2009;47:352-359.

5. Widerström-Noga EG, Finnerup NB, Siddall PJ. Biopsychosocial perspective on a mechanisms-based approach to assessment and treatment of pain following spinal cord injury. J Rehabil Res Dev 2009;46:1-12.

6. Widerström-Noga EG, Turk DC. Types and effectiveness of treatments used by people with chronic pain associated with spinal cord injuries: influence of pain and psychosocial characteristics. Spinal Cord 2003;41:600-609.

7. Harden N, Cohen M. Unmet needs in the management of neuropathic pain. J Pain Symptom Manage 2003;25:S12-17.

8. Wetering EJ, Lemmens KM, Nieboer AP, Huijsman R. Cognitive and behavioral interventions for the management of chronic neuropathic pain in adults--a systematic review. Eur J Pain 2010;14:670-681.

9. Ehde DM, Jensen MP. Psychological treatments for pain management in persons with spinal cord injury: cognitive therapy and self-hypnosis training. Top Spinal Cord Inj Rehabil 2007;13:72-80.

10. Widerström-Noga EG, Felix ER, Cruz-Almeida Y, Turk DC. Psychosocial subgroups in persons with spinal cord injuries and chronic pain. Arch Phys Med Rehabil 2007;88:1628-1635.

11. Wollaars MM, Post MW, van Asbeck FW, Brand N. Spinal cord injury pain: the influence of psychologic factors and impact on quality of life. Clin J Pain 2007;23:383-391.

12. Turk DC, Swanson KS, Tunks ER. Psychological approaches in the treatment of chronic pain patients--when pills, scalpels, and needles are not enough. Can J Psychiatry 2008;53:213-223.

13. Morley S, Eccleston C, Williams A. Systematic review and meta-analysis of randomized controlled trials of cognitive behaviour therapy and behaviour therapy for chronic pain in adults, excluding headache. Pain 1999;80:1-13.

14. Ostelo RW, van Tulder MW, Vlaeyen JW, Linton SJ, Morley SJ, Assendelft WJ. Behavioural treatment for chronic low-back pain. Cochrane Database Syst Rev 2005:CD002014.

15. Kennedy P, Duff J, Evans M, Beedie A. Coping effectiveness training reduces depression and anxiety following traumatic spinal cord injuries. Br J Clin Psychol 2003;42:41-52.

16. Dorstyn D, Mathias J, Denson L. Efficacy of cognitive behavior therapy for the management of psychological outcomes following spinal cord injury: a meta-analysis. J Health Psychol 2011;16:374-391.

17. Umlauf RL. Psychological interventions for chronic pain following spinal cord injury. Clin J Pain 1992;8:111-118.

18. Gault D, Morel-Fatio M, Albert T, Fattal C. Chronic neuropathic pain of spinal cord injury: what is the effectiveness of psychocomportemental management? Ann Phys Rehabil Med 2009;52:167-172.

19. Salle JY, Ginies P, Perrouin-Verbe B, Ventura M. Pain management: what's the more efficient model? Ann Phys Rehabil Med 2009;52:203-209.

20. Sipski ML, Richards JS. Spinal cord injury rehabilitation: state of the science. Am J Phys Med Rehabil 2006;85:310-342.

21. Norrbrink Budh C, Kowalski J, Lundeberg T. A comprehensive pain management programme comprising educational, cognitive and behavioural interventions for neuropathic pain following spinal cord injury. J Rehabil Med 2006;38:172-180.

22. Nicholson Perry K, Nicholas MK, Middleton JW. Comparison of a pain management program with usual care in a pain management center for people with spinal cord injury-related chronic pain. Clin J Pain 2010;26:206-216.

23. Engel GL. The need for a new medical model: a challenge for biomedicine. Science 1977;196:129-136.

24. Ellis A. Rational psychotherapy and individual psychology. J Individ Psychol 1957;13:38-44.

25. Heutink M, Post MW, Luthart P, Pfennings LE, Dijkstra CA, Lindeman E. A multidisciplinary cognitive behavioural programme for coping with chronic neuropathic pain following spinal cord injury: the protocol of the CONECSI trial. BMC Neurol 2010;10:96.

26. Collin C, Wade DT, Davies S, Horne V. The Barthel ADL Index: a reliability study. Int Disabil Stud 1988;10:61-63.

27. Post MW, van Asbeck FW, van Dijk AJ, Schrijvers AJ. Nederlandse interview versie van de Barthel Index onderzocht bij dwarslaesiepatiënten [Dutch interview version of the Barthel Index evaluated in patients with spinal cord injury]. Nederlands Tijdschrift voor Geneeskunde 1995;139:1376-1380 (in Dutch).

28. Spinhoven P, ter Kuile MM, Linssen AC, Gazendam B. Pain coping strategies in a Dutch population of chronic low back pain patients. Pain 1989;37:77-83.

29. Rosenstiel AK, Keefe FJ. The use of coping strategies in chronic low back pain patients: relationship to patient characteristics and current adjustment. Pain 1983;17:33-44.

30. Kraaimaat FW, Evers AW. Pain-coping strategies in chronic pain patients: psychometric characteristics of the pain-coping inventory (PCI). Int J Behav Med 2003;10:343-363.

31. Bouhassira D, Attal N, Alchaar H, Boureau F, Brochet B, Bruxelle J, Cunin G, Fermanian J, Ginies P, Grun-Overdyking A, Jafari-Schluep H, Lanteri-Minet M, Laurent B, Mick G, Serrie A, Valade D, Vicaut E. Comparison of pain syndromes associated with nervous or somatic lesions and development of a new neuropathic pain diagnostic questionnaire (DN4). Pain 2005;114:29-36.

32. Von Korff M, Ormel J, Keefe FJ, Dworkin SF. Grading the severity of chronic pain. Pain 1992;50:133-149.

33. Zigmond AS, Snaith RP. The hospital anxiety and depression scale. Acta Psychiatr Scand 1983;67:361-370.

34. Post MW. Utrechtse Activiteiten Lijst [Utrecht Activities List]. In: Van Asbeck FWA (Ed). Handboek dwarslaesierevalidatie [Textbook of spinal cord injury rehabilitation]. Houten/ Diegum: Bohn Stafleu van Loghum; 1998: pp. 356-357 (in Dutch).

35. Schönherr MC, Groothoff JW, Mulder GA, Eisma WH. Participation and satisfaction after spinal cord injury: results of a vocational and leisure outcome study. Spinal Cord 2005;43:241-248.

36. Fugl-Meyer AR, Brännholm I, Fugl-Meyer K. Happiness and domain-specific life satisfaction in adult northern Swedes. Clin Rehabil 1991;5:25-33.

37. Post MW, de Witte LP, van Asbeck FW, van Dijk AJ, Schrijvers AJ. Predictors of health status and life satisfaction in spinal cord injury. Arch Phys Med Rehabil 1998;79:395-401.

38. Woolrich RA, Kennedy P, Tasiemski T. A preliminary psychometric evaluation of the Hospital Anxiety and Depression Scale (HADS) in 963 people living with a spinal cord injury. Psychol Health Med 2006;11:80-90.

39. Whiteneck GG, Charlifue SW, Gerhart KA, Overholser JD, Richardson GN. Quantifying handicap: a new measure of long-term rehabilitation outcomes. Arch Phys Med Rehabil 1992;73:519-526.

40. Van Koppenhagen CF, Post MW, van der Woude LH, de Witte LP, van Asbeck FW, de Groot S, van den Heuvel W, Lindeman E. Changes and determinants of life satisfaction after spinal cord injury: a cohort study in the Netherlands. Arch Phys Med Rehabil 2008;89:1733-1740.

41. Twisk JW. Applied longitudinal data analysis for epidemiology. A practical guide. Cambridge: Cambridge University Press; 2003.

42. Craig AR, Hancock K, Chang E, Dickson H. Immunizing against depression and anxiety after spinal cord injury. Arch Phys Med Rehabil 1998;79:375-377.

43. Bryce TN, Budh CN, Cardenas DD, Dijkers M, Felix ER, Finnerup NB, Kennedy P, Lundeberg T, Richards JS, Rintala DH, Siddall P, Widerström-Noga E. Pain after spinal cord injury: an evidence-based review for clinical practice and research. Report of the National Institute on Disability and Rehabilitation Research Spinal Cord Injury Measures meeting. J Spinal Cord Med 2007;30:421-440.

44. Turner JA, Cardenas DD. Chronic pain problems in individuals with spinal cord injuries. Semin Clin Neuropsychiatry 1999;4:186-194.

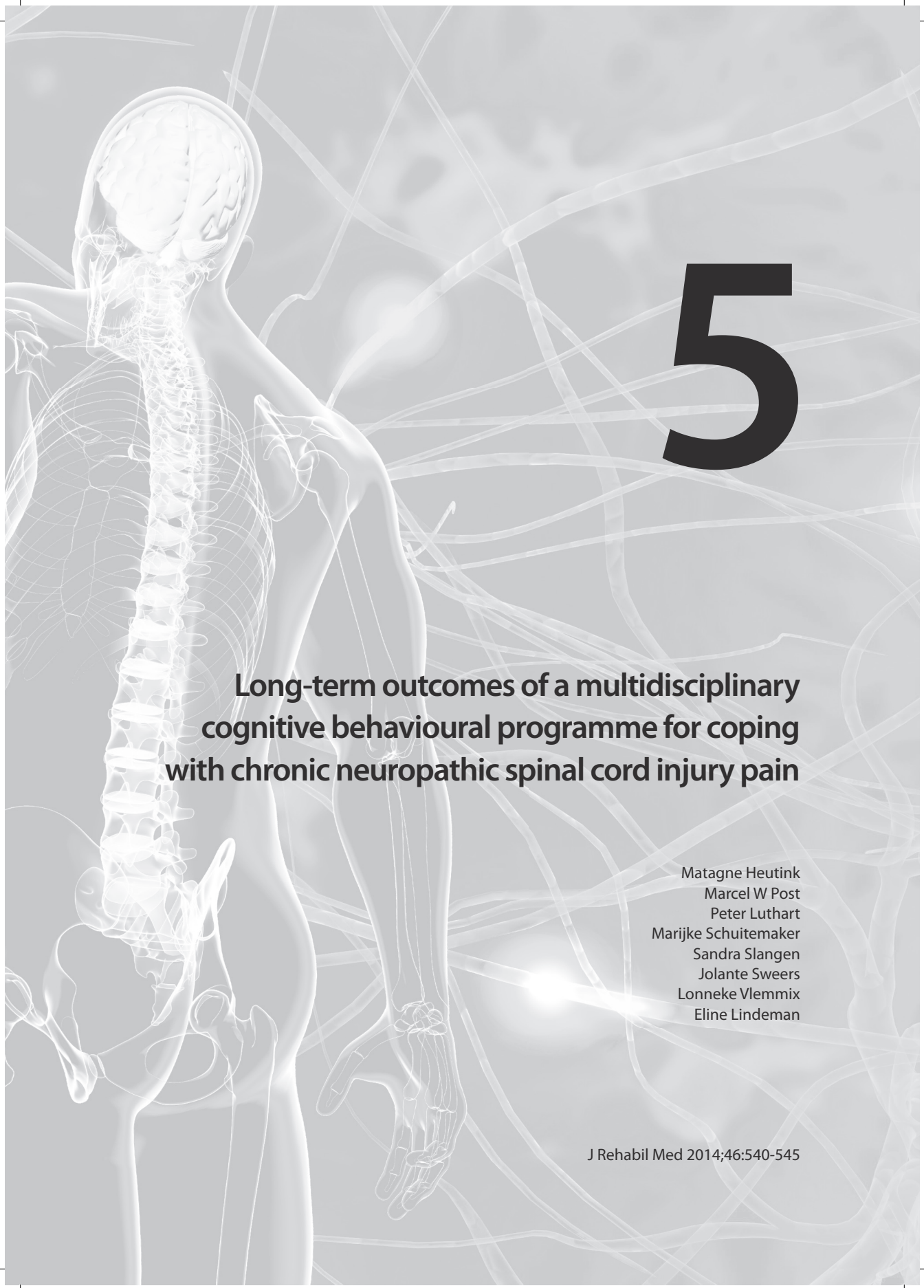
45. Spinhoven P, ter Kuile MM, Linssen AC. Coping met Pijn Vragenlijst (CPV). Handleiding / experimentele versie [Coping Strategy Questionnaire (CSQ). Manual / experimental version]. Lisse: Swets & Zeitlinger; 1994 (in Dutch).

46. Bekkering GE, Hendriks HJ, van Tulder MW, Knol DL, Simmonds MJ, Oostendorp RA, Bouter LM. Prognostic factors for low back pain in patients referred for physiotherapy: comparing outcomes and varying modeling techniques. Spine 2005;30:1881-1886.

47. Heneweer H, Aufdemkampe G, van Tulder MW, Kiers H, Stappaerts KH, Vanhees L. Psychosocial variables in patients with (sub)acute low back pain: an inception cohort in primary care physical therapy in The Netherlands. Spine 2007;32:586-592.

48. Hanley MA, Jensen MP, Ehde DM, Robinson LR, Cardenas DD, Turner JA, Smith DG. Clinically significant change in pain intensity ratings in persons with spinal cord injury or amputation. Clin J Pain 2006;22:25-31.

49. Guzman J, Esmail R, Karjalainen K, Malmivaara A, Irvin E, Bombardier C. Multidisciplinary rehabilitation for chronic low back pain: systematic review. BMJ 2001;322:1511-1516.



5

Long-term outcomes of a multidisciplinary cognitive behavioural programme for coping with chronic neuropathic spinal cord injury pain

Matagne Heutink
Marcel W Post
Peter Luthart
Marijke Schuitemaker
Sandra Slangen
Jolante Sweers
Lonneke Vlemmix
Eline Lindeman

J Rehabil Med 2014;46:540-545

Abstract

Objective: To explore the long-term outcomes of CONECSI (COping with NEuropathiC Spinal cord Injury pain), a multidisciplinary cognitive behavioural treatment programme in persons with spinal cord injury.

Design: Long-term follow-up pre-post-intervention design.

Subjects: A total of 29 subjects with a spinal cord injury and chronic neuropathic pain from 4 Dutch rehabilitation centres.

Methods: Primary outcomes were pain intensity and pain-related disability (Chronic Pain Grade questionnaire). Secondary outcomes were mood (Hospital Anxiety and Depression Scale), participation in activities (Utrecht Activities List), and life satisfaction (Life Satisfaction Questionnaire). Random coefficient analysis was used for the analyses of measurements before (t1), immediate post-intervention (t2), and 6 (t3), 9 (t4), and 12 (t5) months follow-up.

Results: The analyses showed significant improvements on pain intensity (t1–t2 and t1–t5) and pain-related disability (t1–t2, t1–t4, and t1–t5), anxiety and participation in activities (t1–t2, t1–t3, and t1–t5).

Conclusion: This exploratory study suggests that a multidisciplinary cognitive behavioural programme might have lasting improvements on pain intensity, pain-related disability, anxiety, and participation in activities in people with chronic neuropathic spinal cord injury pain and highlights the potential of such programmes.

Introduction

Spinal cord injury (SCI) is a serious condition and adjusting to the physical and psychosocial consequences of SCI is a great challenge for the person involved.[1] The prevalence of depression and anxiety is elevated in people with SCI,[2] and their average life satisfaction is substantially below that of the general population.[3] Chronic pain is one of the major consequences of SCI and affects about 70% of this population.[4] One type of pain many people with SCI have to cope with is chronic neuropathic spinal cord injury pain (CNSCIP), which strongly affects daily functioning and is associated with depression, anxiety and overall quality of life.[5-7] A review showed that no less than 40% of persons with SCI reported intense neuropathic pain.[4] Neuropathic pain is initiated by a primary injury to the nervous system and involves abnormal sensations, such as burning, electric and shooting, and often reduced touch sensation and allodynia.[8] The mechanisms underlying CNSCIP are only partly understood, and it is still unclear why some SCI patients develop neuropathic pain and others with apparently similar injuries do not.[9]

CNSCIP is difficult to treat.[10] Approaches that have been used to treat CNSCIP include pharmacological treatments (e.g., anticonvulsants, antidepressants, opioids, or non-steroidal anti-inflammatory drugs) and non-pharmacological treatments (e.g., physical methods, massage, psychological treatments, acupuncture, physiotherapy and exercise). However, to date none of these provide sufficient relief in the majority of the SCI population.[11-14] Therefore, there is a need for effective treatments for CNSCIP.

In recent years more attention has been given to psychological treatments because research showed relationships between psychological factors and maintenance and aggravation of CNSCIP.[7,15] Psychological treatment is targeted on pain cognitions, e.g., catastrophising, pain-related beliefs and coping, and social factors, to improve psychological and physical functioning in persons with chronic pain.[16] Intervention studies examining the potential for such comprehensive, cognitive behavioural therapy (CBT)-based treatment programmes to benefit persons with SCI and pain showed promising results,[17-20] in terms of changes in anxiety,[17,19,20] and depression.[17,20]

In response to these findings, the CONECSI (COping with NEuropathiC Spinal cord Injury pain) trial was conducted to evaluate the effectiveness of a comprehensive, multidisciplinary cognitive behavioural programme for coping with CNSCIP.[21] This randomised controlled trial demonstrated a short-term decrease in both primary outcome measures (pain intensity and pain-related disability) and 2 out of 4 secondary outcome measures (anxiety and participation in activities), although compared to the control group no short-term treatment

effect was found for pain intensity and only a trend was found for pain-related disability (p = 0.059).[22] However, the duration of follow-up was restricted to 3 months post-intervention. A long-term follow-up might have shown stronger favourable effects of cognitive behavioural treatment if people get more experience in applying principles learned in the programme in their daily life. To our knowledge, except from 1 study,[17] data on the long-term outcomes of cognitive behavioural interventions for CNSCIP is lacking to date.

The objective of this study was therefore to explore the long-term outcomes of the CONECSI trial. The hypothesis was that the intervention would result in a long-term decrease of pain intensity and pain-related disability, and in improvement of mood, participation in activities, and life satisfaction.

Methods

Study design

The CONECSI trial is an unblinded multicentre randomised controlled trial. Participants were randomly allocated to an immediate intervention group or to a waiting list control group within each participating rehabilitation centre. The control group was invited for the programme after a waiting period of 6 months. Measurements were performed in both groups before starting the programme (t1), immediately after intervention (t2), and at 6 months (follow-up, t3). These results have been published earlier.[22] Since a waiting-list control group was used in this trial, it was possible to perform additional follow-up measurements in the intervention group at 9 (t4) and 12 (t5) months after the start of the intervention. But these long-term measurements were not possible in the control group within the time frame of the study. Therefore only the participants in the intervention group are included in the current long-term follow-up study.

Ethical considerations

The Medical Ethics Committees of the University Medical Center Utrecht and the participating rehabilitation centres have approved the study protocol. Written informed consent was obtained from each participant. The trial is registered in the Dutch Trial Register (NTR1580).

Study population

Participants were recruited from 4 Dutch rehabilitation centres with a specialisation in SCI rehabilitation: De Hoogstraat Rehabilitation, Utrecht, Adelante Zorggroep, Hoensbroek, Rehabilitation Center Het Roessingh, Enschede, and Rijndam Rehabilitation Center, Rotterdam. Eligible persons met the following inclusion criteria: (1) SCI (determined by the physiatrists of the 4 SCI departments of the rehabilitation centres); (2) at least 18 years old; (3) at least 1 year after discharge from first inpatient SCI rehabilitation; (4) main pain type is neuropathic pain; (5) duration of neuropathic pain at least 6 months; and (6) pain intensity score in the previous week of at least 40 on the 0–100 numerical rating scale of the Chronic Pain Grade.[23] Exclusion criteria were: (1) SCI caused by metastatic tumor; (2) previous CBT for coping with pain after SCI (determined by a psychologist); (3) inability to function in a group due to psychopathology; and (4) insufficient mastery of the Dutch language.

Procedure

Physiatrists from the 4 rehabilitation centres selected former patients from their centre meeting inclusion criteria 1, 2, and 3. The selected patients were sent a questionnaire to determine if they met the inclusion criteria 4, 5, and 6 and exclusion criterion 1. A trainer of the intervention (psychologist or nurse practitioner) checked in an interview for the other exclusion criteria before final inclusion in the CONECSI trial.

Intervention

This multidisciplinary programme consists of 10 3-h sessions over a 10-week period and a comeback session 3 weeks after the tenth session. Each meeting was supervised by a psychologist and a physiotherapist (the trainers) from the local centre in 3 centres and by a nurse practitioner and a physiotherapist (the trainers) from the local centre in 1 centre. The programme comprises educational, cognitive, and behavioural elements targeted at coping with CNSCIP. Two theoretical models were used in the programme, the BioPsychoSocial (BPS) model[24] and the Activating event-Belief-Consequence (ABC) model.[25] These two models were explained in educational sessions and in guided group discussions using fictitious cases. These models were applied in sports workshops and homework assignments. Further elements of the programme were: information on SCI and CNSCIP; goal setting; information by a physiatrist specialised in SCI rehabilitation and a physiatrist specialised in chronic pain rehabilitation; information on movement and pain; information on assertiveness and communication about pain; introduction to relaxation

exercises; information on pain, mood, and stress; and information on social aspects and partner, family, and friends.

A detailed description of the study protocol and the CONECSI trial has been reported elsewhere.[21]

Instruments

Pain intensity and pain-related disability were measured with the Chronic Pain Grade questionnaire (CPG).[23] Participants rated their pain intensity on a Numeric Rating Scale for mean pain, worst pain, and current pain (pain intensity score 0–100), and the degree of pain interference with daily activities, work/household activities, and recreational/social activities (pain-related disability score 0–100). The internal consistency for the pain intensity score and the pain disability score in an SCI population was excellent (Cronbach's α 0.95 and 0.94, respectively).[7] In the present study, the CPG has been adapted to ask for neuropathic pain ("The following questions relate to neuropathic pain due to spinal cord injury") in the past week instead of the past 6 months. The mean of the CPG scores at inclusion and at t1 score was used as the baseline (t1) score.[22]

Anxiety and depression were measured with the Hospital Anxiety and Depression Scale (HADS).[26] The HADS is a 14-item self-report measure. It contains two 7-item scales: one for anxiety and one for depression, both with a score range of 0–21. It is a valid and reliable measure and responsive to change.[26] Woolrich et al.[27] reported a good internal consistency in an outpatient population with SCI, with a Cronbach's α of 0.85 for the anxiety and 0.79 for the depression scale.

Participation in activities was measured with Utrecht Activities List (UAL).[28,29] The UAL is a Dutch adaptation of the Craig Handicap Assessment and Rating Technique (CHART).[30] Participation in activities is assessed by the time spent on activities such as paid work, study, housekeeping, voluntary work, hobbies, and sports in hours per week.

Life satisfaction was measured with the Life Satisfaction Questionnaire (LiSat-9).[31,32] The LiSat-9 consists of a global item 'life as a whole' and 8 domain-specific items: 'activities of daily living', 'leisure', 'vocational situation', 'financial situation', 'sexual life', 'partnership relationship', 'family life', and 'contacts with friends'. These 9 variables are rated on a 6 point scale (very dissatisfying to very satisfying), with higher scores reflecting greater satisfaction. The internal consistency of the total score (mean of all item scores) was good (Cronbach's α of 0.80) in a Dutch SCI population.[33]

Demographic characteristics assessed at baseline were age, gender, educational level, and marital status. Functional independence was assessed with the Barthel Index (BI).[34] The Dutch translation showed good validity and reliability (Cronbach's α 0.87) in people with SCI.[35] Type of pain (musculoskeletal pain; visceral pain; spasm pain; neuropathic pain below, above, or at injury level; pain from syringomyelia; and non-SCI related pain) was assessed by self-report, as well as time post-injury, cause of injury, and level and completeness of the lesion. The questions of the DN4[36] were used to check for the presence of neuropathic pain. The DN4 questionnaire is a validated instrument with a specificity for detecting neuropathic pain of 82.9% and a sensitivity of 89.9%.[36]

Neurological lesion level was defined as the highest motor level. Completeness was distincted in motor complete (AIS grades A and B) versus motor incomplete (AIS grades C and D). Neurological levels below T1 were defined as paraplegia, neurological levels at or above T1 were defined as tetraplegia. The physiatrist was asked if there was any doubt about the patient's answer about the types of pain or the neurological lesion level, or if the answer was missing. Cause of injury was differentiated into traumatic (traffic, work, and sports accident; fall from height; surgery, and other) and non-traumatic SCI (inflammation; tumour, and other).

Statistical analyses

Participant's characteristics were calculated at t1 (baseline). The pain intensity score and the pain-related disability score of the CPG, the anxiety and depression score of the HADS, total participation in activities of the UAL, and the life satisfaction sum score of the LiSat-9 were calculated for the measurements t1 to t5. Descriptive statistics were computed using means (standard deviations (SDs)). The courses of the outcomes of the intervention over time were analysed using random coefficient analysis (multilevel analysis).[37] The hierarchy in the data of this study is the repeated measurement "time" (t1–t5) (level 1), which is grouped within the individual subjects (level 2), who are grouped in the rehabilitation centres (level 3). Six models were calculated, each with one of the outcome measures as dependent variable (pain intensity, pain-related disability score, anxiety, depression, total participation in activities, and life satisfaction) in a multilevel regression analysis and time modelled with 4 dummy variables (t1–t2, t1–t3, t1–t4, t1–t5) as the determinant. The intercept or slope is fixed, unless the -2 log likelihood of the model with a random intercept or slope is significantly lower (the model is better) then the -2 log likelihood of the fixed intercept or slope model.

Statistical analyses were performed using SPSS statistical program for Windows (version 19.0) and MLwiN program of the Centre for Multilevel Modelling, Institute of Education, University of London (version 2.25). Significance was set at a p-value less than 0.05.

Results

Participants characteristics

A total of 31 persons were randomised in the intervention group of the CONECSI trial. Two persons were excluded from the current analyses because they could not participate in the programme due to health problems before the start of the programme. The mean age of the participants at t1 was 56.5 years (SD = 12.1). The median time between the onset of SCI and inclusion was 5.4 years (range 1.9–23.7) and the median duration of CNSCIP at inclusion was 4.5 years (range 1.6–23.7). The mean Barthel Index score was 12.7 (SD = 5.8) on a 0–20 scale. The mean number of self-reported pain types was 2.5 (SD = 1.2). More men than women participated in this study, and the majority had a traumatic SCI, paraplegia, and lived with a spouse (Table 5.1).

Table 5.1 Baseline characteristics of study sample (n = 29)

Sample characteristics	n	(%)
Gender (male)	21	(72.4)
Married/living with a spouse	23	(79.3)
Level of education (low[a])	14	(48.3)
Traumatic SCI	25	(86.2)
Paraplegic level of SCI	18	(62.1)
Motor incomplete SCI	14	(48.3)
Pain type		
Musculoskeletal	15	(51.7)
Visceral	6	(20.7)
Neuropathic above	4	(13.8)
Neuropathic at level	15	(51.7)
Neuropathic below	24	(82.8)
Spasm	7	(24.1)
Syringomyelia	1	(3.4)
Other	1	(3.4)

[a] Incomplete primary education, primary school, junior secondary technical education, general secondary education (lower level).
SCI: spinal cord injury.

Course of primary and secondary outcome measures

Table 5.2 shows descriptive data of pain intensity, pain-related disability, participation in activities, anxiety, depression, and life satisfaction at each measurement time-point.

Multilevel analysis showed that pain intensity significantly decreased between the time periods t1–t2 and t1–t5, and that pain-related disability decreased between the time periods t1–t2, t1–t4 and t1–t5. The decrease in pain-related disability between t1 and t3 was just outside significance (Table 5.3, Figure 5.1).

No changes over time were found for the secondary outcome measures HADS depression and life satisfaction (Table 5.4). HADS anxiety scores significantly decreased and UAL scores significantly increased across t1–t2, t1–t3, and t1–t5 (Table 5.4 and Figure 5.2).

Discussion

The present study shows that the CONECSI intervention, a multidisciplinary cognitive behavioural programme for coping with CNSCIP, had favourable long-term outcomes on the primary outcomes pain intensity and pain-related disability, and the secondary outcomes

Table 5.2 Descriptives of pain intensity, pain-related disability, participation in activities, anxiety, depression, and life satisfaction scores at each measurement (mean and standard deviation)

Outcome	Maximum range	Actual range	t1 (n = 29)	t2 (n = 29)	t3 (n = 29)	t4 (n = 28)	t5 (n = 24)
Pain intensity	0–100	20–90	69.0 (9.8)	65.3 (12.9)	66.1 (13.2)	66.3 (12.3)	65.7 (16.6)
Pain-related disability	0–100	0–83.33	49.0 (21.2)	38.8 (25.1)	39.0 (24.7)	40.1 (23.7)	42.4 (23.9)
Participation in activities	0–no max	0–91	33.1 (22.7)	41.8 (18.8)	43.1 (16.7)	35.5 (22.5)	42.7 (17.3)
Anxiety	0–21	0–19	7.2 (4.1)	5.9 (3.6)	6.1 (3.6)	6.7 (3.4)	5.4 (3.1)
Depression	0–21	1–18	7.2 (3.6)	6.7 (4.0)	6.8 (3.1)	6.4 (3.2)	6.0 (3.8)
Life satisfaction	1–6	1–6	4.1 (1.0)	4.0 (1.1)	4.2 (0.9)	4.0 (1.1)	4.4 (0.8)

The mean scores differ slightly from the scores in the figures with MlwiN-estimated values.
t1: measurement 1, pre-intervention; t2: measurement 2, immediate post-intervention; t3: measurement 3, 6 months follow-up; t4: measurement 4, 9 months follow-up; t5: measurement 5, 12 months follow-up.

Table 5.3 Multilevel linear regression models for pain intensity and pain-related disability pre- and post-intervention, and 6, 9, and 12 months follow-up (n = 29)

	Model for pain intensity			Model for pain-related disability		
Variables	β	SE	p	β	SE	p
Constant	69.023	2.141		48.965	4.298	
Time (t1–t2)	-3.712	1.850	0.044*	-10.155	3.913	0.009*
Time (t1–t3)	-2.931	2.329	0.208	-9.999	5.104	0.050
Time (t1–t4)	-3.077	1.872	0.101	-8.697	3.960	0.028*
Time (t1–t5)	-5.520	2.004	0.006*	-8.544	4.165	0.040*

Note 1: Beta (β) stands for a non-standardised regression coefficient in multilevel analyses.
Note 2: All models had random intercepts.
Note 3: All models had fixed slopes, except for the Time (t1–t3) and (t1–t5) covariates of pain intensity and for the Time (t1–t3) covariate of pain-related disability.
t1: measurement 1, pre-intervention; t2: measurement 2, immediate post-intervention; t3: measurement 3, 6 months follow-up; t4: measurement 4, 9 months follow-up; t5: measurement 5, 12 months follow-up; SE: standard error.
* $p < 0.05$.

Table 5.4 Multilevel linear regression models for the secondary outcome measures pre- and post-intervention, and 6, 9, and 12 months follow-up (n = 29)

	Model for participation in activities			Model for anxiety		
Variables	β	SE	p	β	SE	p
Constant	33.172	3.628		7.207	0.662	
Time (t1–t2)	8.635	2.984	0.004*	-1.413	0.477	0.003*
Time (t1–t3)	9.395	2.949	0.001*	-1.256	0.471	0.008*
Time (t1–t4)	0.985	2.986	0.741	-0.357	0.477	0.453
Time (t1–t5)	8.668	3.107	0.005*	-1.427	0.497	0.004*
	Model for depression			Model for life satisfaction		
Variables	β	SE	p	β	SE	p
Constant	7.241	0.650		4.102	0.186	
Time (t1–t2)	-0.558	0.468	0.234	-0.202	0.126	0.110
Time (t1–t3)	-0.374	0.462	0.418	0.063	0.125	0.617
Time (t1–t4)	-0.644	0.468	0.168	-0.178	0.126	0.159
Time (t1–t5)	-0.821	0.487	0.091	0.196	0.132	0.139

Note 1: Beta (β) stands for a non-standardised regression coefficient in multilevel analyses.
Note 2: All models had random intercepts.
Note 3: All models had fixed slopes.
t1: measurement 1, pre-intervention; t2: measurement 2, immediate post-intervention; t3: measurement 3, 6 months follow-up; t4: measurement 4, 9 months follow-up; t5: measurement 5, 12 months follow-up; SE: standard error.
* $p < 0.05$.

anxiety and participation in activities. This study adds long-term outcomes (at 9 and 12 months) to the earlier reported short-term results of the CONECSI trial.[22]

Overall, there was no significant difference between the scores at 12 months and the scores immediately after intervention (t2; 3 months), confirming the hypothesis that improvements during the intervention would be maintained at follow-up, although the patterns of scores were variable over time for 3 of the 6 outcome variables. Only pain intensity showed a further decrease after the end of the intervention at 12 months. Although the scores changed in the right direction, no significant change over time was found for depression and life satisfaction. This is in line with the previously reported short-term results.[22]

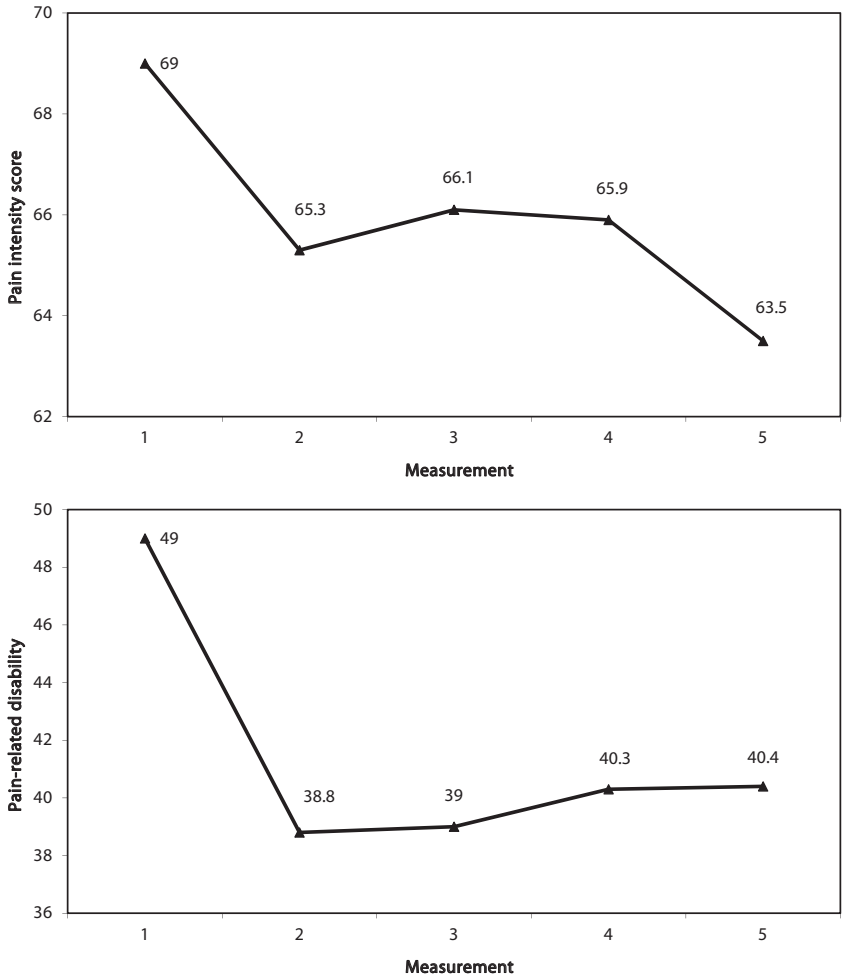


Figure 5.1 Pain intensity score and pain-related disability score of the Chronic Pain Grade questionnaire.

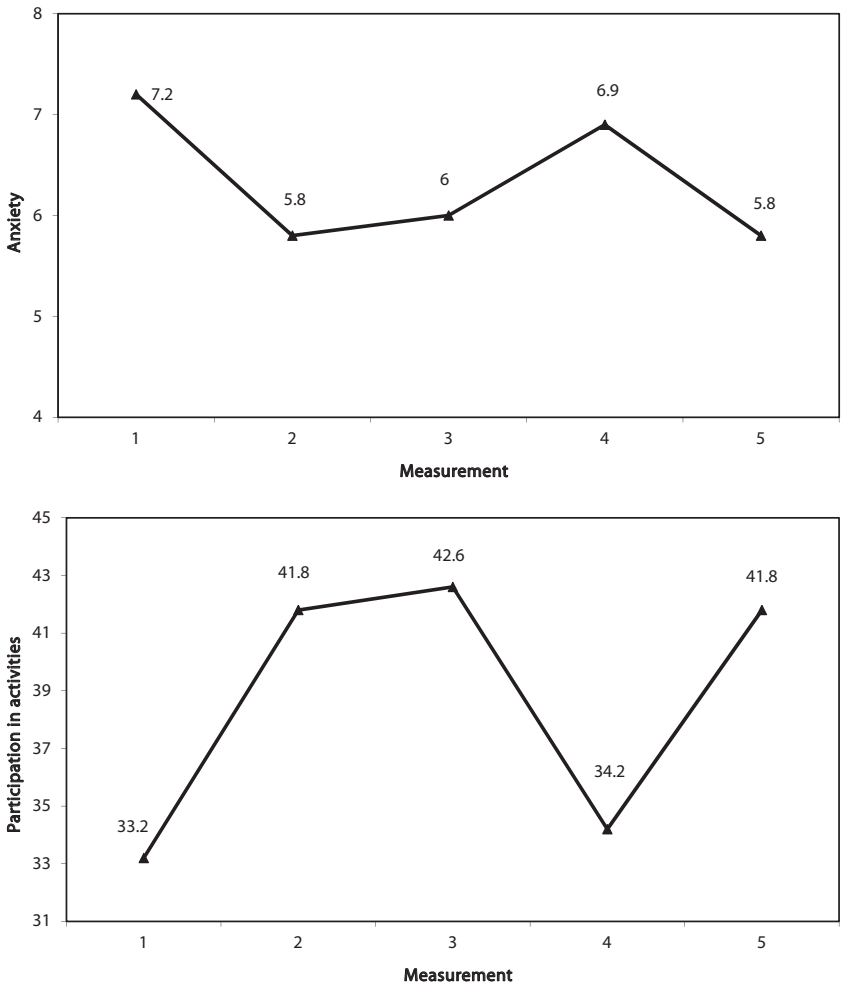


Figure 5.2 Anxiety score of the Hospital Anxiety and Depression Scale and participation in activities score of the Utrecht Activities List.

Long-term outcomes

A decrease of pain intensity is usually not a focus of cognitive behavioural programmes, but it is a common "side effect" of improvements in physical and psychological functioning.[16] In this study we indeed found a decrease in pain intensity, together with improvements in pain-related disability, anxiety, and participation in activities. This study confirms the conclusions of earlier studies[17,19] that comprehensive psychosocial pain treatment programmes are promising treatment options for persons with disabilities and pain.

Norrbrink Budh et al.[17] found also change of anxiety and no change in life satisfaction after the intervention, but in contrast to our study they found change of depression, and no change in pain intensity at 12 months follow-up. Nicholson Perry et al.[19] found a reduction in anxiety in their treatment group, and a trend towards improvement on pain intensity and depression at 1 month post-treatment, but the depression scores returned to pre-treatment levels at 9 month follow-up. Both anxiety and depressed mood were addressed in our CBT programme, but maybe it is easier to modify feelings of anxiety than depressed mood by CBT. Life satisfaction was measured using a questionnaire on satisfaction with various life domains, including satisfaction with vocational situation and sexual life, which might explain the lack of change of life satisfaction scores in this study. Dorstyn et al.[38] found in a meta-analysis that most treatment effects of CBT for the management of psychological outcomes following SCI were minimal or not sustained at follow-up. The results of the current study showed little relapse, maybe because of the booster session 3 weeks after the final group session. Nevertheless, further attention for relapse prevention is required.[19] Booster session(s) or comeback session(s) in the third to ninth month after finishing the intervention might be helpful.

Strengths and limitations of this study

This is one of the few longitudinal studies reporting long-term effect of CBT for coping with CNSCIP. The loss to follow-up was minimal and the use of random coefficient analysis allowed the inclusion of all participants in the statistical analyses. However, the sample size was small. There were only 29 persons analysed in this study, although the repeated measurements increased the statistical power of the analyses. Another limitation is, because of the use of a waiting-list control group, it was not possible to perform the current analyses in a controlled design. The results of this study should therefore be considered exploratory and in need for further confirmation. There is a need for further research utilising larger samples and longer term measurements to study the effectiveness of CBT-based interventions for CNSCIP.

Implications

Our findings highlight the potential of cognitive behavioural programmes to learn people with SCI cope with neuropathic pain. Our finding of a long-term decrease of pain intensity needs confirmation, but is encouraging and might make the CBT approach more attractive to patients. The programme focused on CNSCIP, but it can easily be adapted to include

other types of pain. CBT is easy to implement in clinical practice, since it is an accepted treatment in other diagnostic groups. More research is needed on treatment modalities, i.e. individual or group, or internet-based, and on timing, i.e. soon after persons with SCI start experiencing neuropathic pain, or only after pharmaceutical and non-pharmaceutical treatments show insufficient pain relief.

Acknowledgements

The authors would like to thank the participants in this study, the participating Dutch rehabilitation centres, and the co-trainers in these centres: Lilian Pfennings (De Hoogstraat Rehabilitation, Utrecht); Conny Overdulve (Adelante Zorggroep, Hoensbroek); Wim van de Vis (Rehabilitation Center Het Roessingh, Enschede); Hilde Schors and Nicole Vrijens (Rijndam Rehabilitation Center, Rotterdam).

References

1. Dijkers MP. Quality of life of individuals with spinal cord injury: a review of conceptualization, measurement, and research findings. J Rehabil Res Dev 2005;42:87-110.
2. Migliorini CE, New PW, Tonge BJ. Comparison of depression, anxiety and stress in persons with traumatic and non-traumatic post-acute spinal cord injury. Spinal Cord 2009;47:783-788.
3. Post MW, van Leeuwen CM. Psychosocial issues in spinal cord injury: a review. Spinal Cord 2012;50:382-389.
4. Margot-Duclot A, Tournebise H, Ventura M, Fattal C. What are the risk factors of occurence and chronicity of neuropathic pain in spinal cord injury patients? Ann Phys Rehabil Med 2009;52:111-123.
5. Harden N, Cohen M. Unmet needs in the management of neuropathic pain. J Pain Symptom Manage 2003;25:S12-17.
6. Wetering EJ, Lemmens KM, Nieboer AP, Huijsman R. Cognitive and behavioral interventions for the management of chronic neuropathic pain in adults--a systematic review. Eur J Pain 2010;14:670-681.
7. Wollaars MM, Post MW, van Asbeck FW, Brand N. Spinal cord injury pain: the influence of psychologic factors and impact on quality of life. Clin J Pain 2007;23:383-391.
8. Siddall PJ, Middleton JW. A proposed algorithm for the management of pain following spinal cord injury. Spinal Cord 2006;44:67-77.
9. Finnerup NB. Predictors of spinal cord injury neuropathic pain: The role of QST. Top Spinal Cord Inj Rehabil 2007;13:35-42.

10. DeFrates S, Cook AM. Pharmacologic treatment of neuropathic pain following spinal cord injury. Orthopedics 2011;34:203.

11. Heutink M, Post MW, Wollaars MM, van Asbeck FW. Chronic spinal cord injury pain: pharmacological and non-pharmacological treatments and treatment effectiveness. Disabil Rehabil 2011;33:433-440.

12. Siddall PJ, McClelland JM, Rutkowski SB, Cousins MJ. A longitudinal study of the prevalence and characteristics of pain in the first 5 years following spinal cord injury. Pain 2003;103:249-257.

13. Widerström-Noga EG, Finnerup NB, Siddall PJ. Biopsychosocial perspective on a mechanisms-based approach to assessment and treatment of pain following spinal cord injury. J Rehabil Res Dev 2009;46:1-12.

14. Widerström-Noga EG, Turk DC. Types and effectiveness of treatments used by people with chronic pain associated with spinal cord injuries: influence of pain and psychosocial characteristics. Spinal Cord 2003;41:600-609.

15. Widerström-Noga EG, Felix ER, Cruz-Almeida Y, Turk DC. Psychosocial subgroups in persons with spinal cord injuries and chronic pain. Arch Phys Med Rehabil 2007;88:1628-1635.

16. Jensen MP, Moore MR, Bockow TB, Ehde DM, Engel JM. Psychosocial factors and adjustment to chronic pain in persons with physical disabilities: a systematic review. Arch Phys Med Rehabil 2011;92:146-160.

17. Norrbrink Budh C, Kowalski J, Lundeberg T. A comprehensive pain management programme comprising educational, cognitive and behavioural interventions for neuropathic pain following spinal cord injury. J Rehabil Med 2006;38:172-180.

18. Ehde DM, Jensen MP. Feasibility of a cognitive restructuring intervention for treatment of chronic pain in persons with disabilities. Rehabil Psychol 2004;49:254-258.

19. Nicholson Perry K, Nicholas MK, Middleton JW. Comparison of a pain management program with usual care in a pain management center for people with spinal cord injury-related chronic pain. Clin J Pain 2010;26:206-216.

20. Kennedy P, Duff J, Evans M, Beedie A. Coping effectiveness training reduces depression and anxiety following traumatic spinal cord injuries. Br J Clin Psychol 2003;42:41-52.

21. Heutink M, Post MW, Luthart P, Pfennings LE, Dijkstra CA, Lindeman E. A multidisciplinary cognitive behavioural programme for coping with chronic neuropathic pain following spinal cord injury: the protocol of the CONECSI trial. BMC Neurol 2010;10:96.

22. Heutink M, Post MW, Bongers-Janssen HM, Dijkstra CA, Snoek GJ, Spijkerman DC, Lindeman E. The CONECSI trial: results of a randomized controlled trial of a multidisciplinary cognitive behavioral program for coping with chronic neuropathic pain after spinal cord injury. Pain 2012;153:120-128.

23. Von Korff M, Ormel J, Keefe FJ, Dworkin SF. Grading the severity of chronic pain. Pain 1992;50:133-149.

24. Engel GL. The need for a new medical model: a challenge for biomedicine. Science 1977;196:129-136.

25. Ellis A. Rational psychotherapy and individual psychology. J Individ Psychol 1957;13:38-44.

26. Zigmond AS, Snaith RP. The hospital anxiety and depression scale. Acta Psychiatr Scand 1983;67:361-370.

27. Woolrich RA, Kennedy P, Tasiemski T. A preliminary psychometric evaluation of the Hospital Anxiety and Depression Scale (HADS) in 963 people living with a spinal cord injury. Psychol Health Med 2006;11:80-90.

28. Post MW. Utrecht Activiteiten Lijst [Utrecht Activities List]. In: Van Asbeck FWA (Ed). Handboek dwarslaesierevalidatie [Textbook of spinal cord injury rehabilitation]. Houten/Diegum: Bohn Stafleu van Loghum; 1998: pp. 356-357 (in Dutch).

29. Schönherr MC, Groothoff JW, Mulder GA, Eisma WH. Participation and satisfaction after spinal cord injury: results of a vocational and leisure outcome study. Spinal Cord 2005;43:241-248.

30. Whiteneck GG, Charlifue SW, Gerhart KA, Overholser JD, Richardson GN. Quantifying handicap: a new measure of long-term rehabilitation outcomes. Arch Phys Med Rehabil 1992;73:519-526.

31. Fugl-Meyer AR, Brännholm I, Fugl-Meyer K. Happiness and domain-specific life satisfaction in adult northern Swedes. Clin Rehabil 1991;5:25-33.

32. Post MW, de Witte LP, van Asbeck FW, van Dijk AJ, Schrijvers AJ. Predictors of health status and life satisfaction in spinal cord injury. Arch Phys Med Rehabil 1998;79:395-401.

33. Van Koppenhagen CF, Post MW, van der Woude LH, de Witte LP, van Asbeck FW, de Groot S, van den Heuvel W, Lindeman E. Changes and determinants of life satisfaction after spinal cord injury: a cohort study in the Netherlands. Arch Phys Med Rehabil 2008;89:1733-1740.

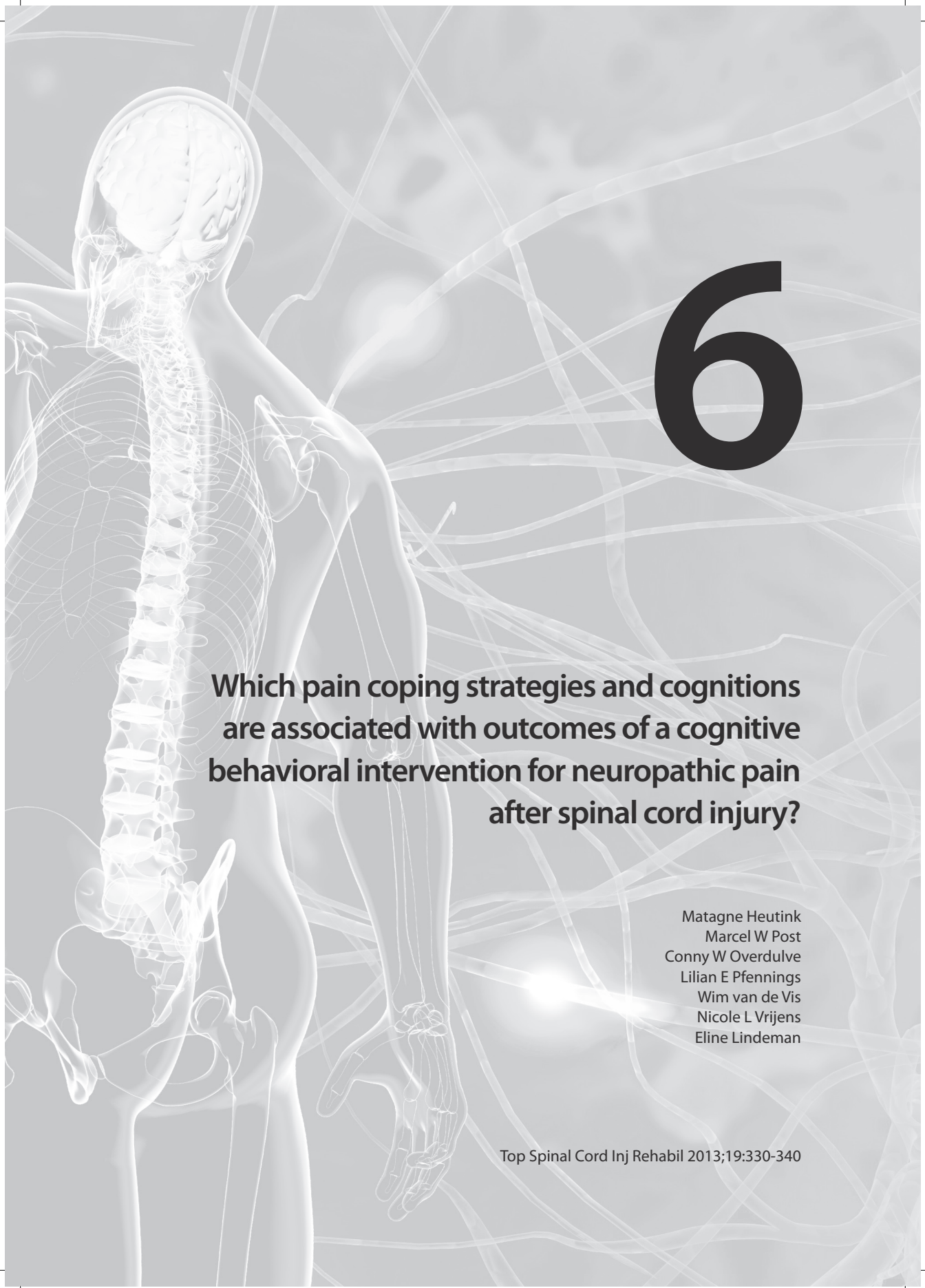
34. Collin C, Wade DT, Davies S, Horne V. The Barthel ADL Index: a reliability study. Int Disabil Stud 1988;10:61-63.

35. Post MW, van Asbeck FW, van Dijk AJ, Schrijvers AJ. Nederlandse interview versie van de Barthel Index onderzocht bij dwarslaesiepatiënten [Dutch interview version of the Barthel Index evaluated in patients with spinal cord injury]. Nederlands Tijdschrift voor Geneeskunde 1995;139:1376-1380 (in Dutch).

36. Bouhassira D, Attal N, Alchaar H, Boureau F, Brochet B, Bruxelle J, Cunin G, Fermanian J, Ginies P, Grun-Overdyking A, Jafari-Schluep H, Lantéri-Minet M, Laurent B, Mick G, Serrie A, Valade D, Vicaut E. Comparison of pain syndromes associated with nervous or somatic lesions and development of a new neuropathic pain diagnostic questionnaire (DN4). Pain 2005;114:29-36.

37. Twisk JW. Applied longitudinal data analysis for epidemiology. A practical guide. Cambridge: Cambridge University Press; 2003.

38. Dorstyn D, Mathias J, Denson L. Efficacy of cognitive behavior therapy for the management of psychological outcomes following spinal cord injury: A meta-analysis. J Health Psychol 2011;16:374-391.



6

Which pain coping strategies and cognitions are associated with outcomes of a cognitive behavioral intervention for neuropathic pain after spinal cord injury?

Matagne Heutink
Marcel W Post
Conny W Overdulve
Lilian E Pfennings
Wim van de Vis
Nicole L Vrijens
Eline Lindeman

Top Spinal Cord Inj Rehabil 2013;19:330-340

Abstract

Background: Chronic neuropathic pain is one of the most difficult problems to manage after spinal cord injury (SCI). Pain coping and pain cognitions are known to be associated with the patient's experience of neuropathic pain, but they have not been studied in the context of a cognitive behavioral treatment program for coping with neuropathic pain after SCI.

Objective: To explore associations of pain coping strategies and cognitions with pain intensity and pain-related disability and changes in pain coping strategies and cognitions with changes in pain intensity and pain-related disability.

Methods: Forty-seven persons who participated in the CONECSI (COping with NEuropathiC Spinal cord Injury pain) trial completed questionnaires before the intervention (baseline) and 3 months after of the intervention (follow-up).

Results: Compared to baseline, participants showed more favorable scores on 2 pain coping scales (Pain Transformation and Worrying), the subtotal score Active Coping, and 3 pain cognitions scales (Catastrophizing, Optimism, and Reliance on Health Care) at follow-up. Baseline Reliance on Health Care was associated with change in pain intensity and pain-related disability. Change in Catastrophizing and change in Restriction cognitions were associated with change in pain-related disability.

Conclusions: Our findings suggest that modifying pain coping strategies and cognitions by a cognitive behavioral intervention for chronic neuropathic pain after SCI may have some beneficial effects on pain intensity and pain-related disability. Further research should show how dysfunctional pain coping strategies and cognitions can be most effectively modified.

Introduction

Pain is a significant problem for many persons with spinal cord injury (SCI). About 65% to 85% of them report the presence of pain,[1,2] and around a third of them experience severe pain.[1] Chronic neuropathic SCI pain (CNSCIP) at the level of the injury is present in 41% of all persons at 5 years after SCI, and 34% of them have CNSCIP below the level of injury.[1] Pain contributes to poorer rehabilitation outcomes and reduced quality of life,[3] and it is one of the most difficult problems to manage, even in the presence of other problems that interfere with daily life.[4] The long-term prognosis for pain resolution following SCI is poor.[1,3] Many persons affected report that SCI pain persists or even worsens over time.[5]

Although biomedical changes associated with the disability itself may play a primary role in the presence and severity of pain, psychosocial factors, such as attitudes and cognitions, coping behavior, and the social environment play a major role in the perceived intensity and impact of pain in many persons with physical disabilities and chronic pain.[6-10] Psychosocial factors may perpetuate chronic pain[11] and predict pain and functioning in persons with physical disabilities.[9] Several studies identified pain cognitions such as catastrophizing[7,12-15] and pain coping strategies such as passive coping[7,14] as predictors of increased pain intensity and pain-related disability. Interventions to modify dysfunctional pain cognitions and pain coping strategies therefore hold promise for persons with CNSCIP.[7,16] In particular, cognitive behavior therapy (CBT) has been used for a variety of chronic pain problems and is expected to change patient cognitions and coping strategies.[17]

Studies to investigate the effect of CBT-based treatment programs for persons with chronic SCI pain have been rare,[18,19] especially among persons with CNSCIP.[20] Although these studies showed some promising results, in terms of changes in pain intensity and life satisfaction,[20] anxiety,[18,19,20] and depression,[18,20] they failed to prove the effectiveness of CBT as compared to placebo or usual care.

Changes in pain coping strategies and cognitions were not examined,[20] or associations with changes in the outcome measures were not studied.[18,19] Kennedy et al.[18] found no changes in coping strategies compared to controls, whereas Nicholson Perry et al.[19] found a significant improvement in pain catastrophizing in the treatment group but no differences with the usual care group.

In any case, persons vary in their response to CBT, and little is known about patient characteristics that predict or moderate the effects of CBT on patient outcomes following SCI.[17,21] A better understanding of how baseline pain coping strategies and cognitions, and

changes in such strategies and cognitions, are associated with treatment outcomes could help direct limited resources to those persons most likely to benefit, match patients with the most appropriate treatments, and tailor interventions to patient characteristics.[17]

Recently, Heutink et al.[22,23] reported the design and results of the CONECSI (COping with NEuropathiC Spinal cord Injury pain) trial, the first randomized controlled trial (RCT) on the effectiveness of a multidisciplinary CBT program for coping with CNSCIP compared with a waiting list control group. No significant intervention effects on the primary outcomes pain intensity and pain-related disability were found. There was a trend p = 0.059) toward an effect of the intervention on pain-related disability immediately after the intervention, and the intervention group showed a significant decrease in pain intensity and pain-related disability that was not found in the control group. Significant treatment effects were found on the secondary outcomes anxiety and participation in activities.[23]

The current study utilized data from the CONECSI trial to explore associations between baseline pain coping strategies and pain cognitions on the one hand and pain intensity and pain-related disability on the other, and to study the hypothesis that changes in pain coping strategies and pain cognitions are associated with outcome variables after CBT intervention. The specific aims were to explore changes in pain coping strategies and cognitions during the CBT intervention and to explore (a) associations between baseline pain coping strategies and cognitions and baseline pain and pain-related disability, (b) associations between baseline pain coping strategies and cognitions and the changes in pain and pain-related disability during the RCT, and (c) associations between changes in pain coping strategies and cognitions and changes in pain intensity and pain-related disability during the RCT. We hypothesized that more catastrophizing,[7,12-15] less active coping, and more passive coping[7,14] at baseline would be associated with higher pain intensity and pain-related disability at baseline and greater changes in pain intensity and pain-related disability. Further it was hypothesized that favorable changes in pain coping strategies and pain cognitions would be associated with favorable changes in pain intensity and pain-related disability.

Methods

Study design

In the unblinded CONECSI trial, participants were randomly allocated to the intervention group or to the waiting list control group[22] within 4 participating rehabilitation centers in different parts of the Netherlands. The control group was invited for the same intervention

after waiting for 6 months. Both groups were tested before the intervention (baseline) and 3 months after the end of the intervention (follow-up). For the purpose of this study, the data of both groups were merged, leaving a single group with pre- and post-intervention tests.

The Medical Ethics Committees of the University Medical Center Utrecht and the participating rehabilitation centers approved the study protocol. Written informed consent was obtained from each participant.

Participants and procedure

An extensive description of the study protocol has been published elsewhere.[22] Persons with SCI who had been treated at 1 of the 4 rehabilitation centers, were at least 18 years old, and had been discharged from first SCI rehabilitation at least 1 year previously were invited to participate. Other inclusion criteria were the following: the main pain type was neuropathic pain, the neuropathic pain had persisted for at least 6 months, and the neuropathic pain intensity score in the previous week was at least 40 on the 0 to 100 scale of the Chronic Pain Grade (CPG) questionnaire.[24] Exclusion criteria were the following: SCI was caused by metastatic tumor, previous CBT was received for coping with pain after SCI, inability to function in a group due to psychopathology, and insufficient command of Dutch. Data were complete for 61 persons, but 14 of them were excluded from the current analyses because they had dropped out before the start of the intervention, so that no change in pain coping strategies or pain cognitions was to be expected. Of these 14, 4 persons could not participate because of health problems not related to the intervention, and 10 persons withdrew for practical reasons (e.g., they could not get a day a week off from work to follow the course). The analyses in the current study were limited to the 47 participants who did participate in the CBT program.

Intervention

The CBT program comprises educational, cognitive, and behavioral elements targeted at coping with CNSCIP and consists of 10 three-hour sessions over a 10-week period and a follow-up session 3 weeks after the 10th session. Participants received a course book containing information on all sessions, as well as reading texts and homework assignments. The trainers received a manual and the same course book as the participants. Two theoretical models were used in the program to focus on coping and cognitions: the BioPsychoSocial (BPS) model[25] and the Activating event-Belief-Consequence (ABC) model.[26] The BPS model was used to focus on the influence of psychosocial factors on the experience of pain; to

clarify the relationship between biological, psychological, and social factors and pain; and to clarify the importance of maintaining a balance between capacity and load. The ABC model was used for cognitive restructuring. These 2 models were explained in educational sessions and in guided group discussions using fictitious cases. The models were also applied in sports workshops and homework assignments. The content of the program is described in more detail in Appendix 6.1.

Measures

Pain type Before inclusion, participants were asked to indicate their pain types (musculoskeletal pain; visceral pain; spasm pain; neuropathic pain below, above, or at injury level; pain from syringomyelia; and non–SCI-related pain). The questionnaire contained descriptions of the different types of pain with examples to help participants indicate their type. The questions of the DN4[27] were used to check for the presence of neuropathic pain. The DN4 questionnaire is a validated instrument with a specificity for detecting neuropathic pain of 82.9% and a sensitivity of 89.9%.[27] If there was any doubt about the presence of neuropathic pain, the physiatrist was asked for information. Participants were also asked to indicate for how long they had had neuropathic pain.

Pain intensity and pain-related disability Pain intensity and pain-related disability were measured with the CPG.[24] Participants rated their pain intensity on a 0 to 10 numeric rating scale (NRS) for average pain, worst pain, and current pain (pain intensity score) and rated the degree of pain interference with daily activities, work/household activities, and recreational/social activities (pain-related disability score). The internal consistency of the pain intensity score and the pain-related disability score in an SCI population is excellent (Cronbach's α of 0.95 and 0.94, respectively).[7] In the present study, the CPG was adapted to ask about neuropathic pain in the previous week instead of the previous 6 months. The CPG scores at inclusion and at baseline were combined, and the mean score was used as the baseline score.

Pain coping The Pain Coping Inventory (PCI) consists of 33 items measuring cognitive and behavioral pain coping strategies. Items are rated on a 4-point Likert scale ranging from 1 (rarely or never) to 4 (very often) in terms of the frequency with which strategies are applied when dealing with pain. Two subtotal scores can be computed: Active Coping and Passive Coping. The Active Pain Coping subtotal score consists of the scales for Pain Transformation (e.g., I pretend the pain is not there), Distraction (e.g., I look for distraction by focusing on reading, music, a television program, etc), Reducing Demands (e.g., I continue

my activities, but at a slower pace). The Passive Pain Coping subtotal score consists of the scales for Retreating (e.g., I retreat to a quiet setting), Worrying (e.g., I think the pain will get worse), and Resting (e.g., I will stop my activities). All scales are reliable,[28] and the internal consistency for the 6 scales ranges from 0.62 to 0.79.[29]

Pain cognitions The Pain Cognition List-2003 (PCL) consists of 39 items. Each item presents a specific pain cognition statement, for example, "My thoughts are always concentrated on the pain." Items are rated on a 5-point Likert scale ranging from 1 (totally disagree) to 5 (totally agree).[30] The questionnaire consists of 5 subscales: Catastrophizing (e.g., I think I was stuck by fate), Restrictions (e.g., I think the pain is a warning that I should slow down), Optimism (e.g., I think I have learned to deal with the pain), Internal Control (e.g., I know a way to reduce my pain a little), and Reliance on Health Care (e.g., I have firm confidence in medical science). The scales vary in length (from 4 to 16 items) and internal consistency (Cronbach's α of 0.64 to 0.88 with an average of 0.75), and correlations between subscales range from 0.00 to 0.45. The validity of these subscales is supported by the meaningful pattern of correlations with other relevant constructs.[31] The stability of the PCL is satisfactory with Pearson correlation coefficients across a time interval of 2 weeks ranging from 0.51 (Reliance on Health Care) to 0.73 (Catastrophizing), with an average of 0.64.[31]

Statistical analyses

Descriptive statistics were computed, using mean (SD) and when appropriate median (interquartile range [IQR]). Nonparametric statistics were used for the further analyses, because of the relatively small sample size. Wilcoxon signed-rank tests were performed to analyze changes between baseline and follow-up. Spearman correlation coefficients were used to study the 3 hypotheses (a) associations between baseline PCI and PCL scores and baseline scores of pain intensity and pain-related disability, (b) associations between baseline PCI and PCL scores and change scores (pre-post difference) for pain intensity and pain-related disability, and (c) associations between change scores (pre-post difference) for the PCI and PCL scores and change scores (pre-post difference) for pain intensity and pain-related disability. In view of the small sample size, no multivariate linear regression analyses were performed. Data analysis was performed using SPSS 16.0. Significance level was set at a p-value less than 0.05. No correction for multiple testing was performed, because of the exploratory nature of these analyses.

Results

Participant characteristics

More men than women participated in this study, and the majority had a traumatic injury, an incomplete paraplegia, and were living with a spouse (Table 6.1). The mean (SD) age of the 47 participants at the start of the study was 58.0 (11.8) years. The median time between the onset of SCI and inclusion was 7.3 years (range 1.4–23.7), and the median

Table 6.1 Baseline characteristics

Characteristics	n	%
Gender		
Male	31	66.0
Female	16	34.0
Marital status		
Married/living with a spouse	38	80.9
Living alone	9	19.1
Education		
Low[a]	15	31.9
Intermediate[b]	19	40.4
High[c]	13	27.7
Cause of injury		
Traumatic	36	76.6
Nontraumatic	11	23.4
Lesion level		
Paraplegia	31	65.9
Tetraplegia	16	34.1
Lesion completeness		
Motor incomplete	28	59.6
Motor complete	19	40.4
Pain types		
Musculoskeletal	21	44.7
Visceral	11	23.4
Neuropathic above	5	10.6
Neuropathic at level	23	48.9
Neuropathic below	40	85.1
Spasm	11	23.4
Syringomyelia	5	10.6
Other	2	4.3

[a] Incomplete primary education, primary school, junior secondary technical education.
[b] General secondary education (lower level), vocational secondary education.
[c] General secondary education (higher level), higher vocational secondary education (bachelor), master.

duration of CNSCIP at inclusion was 6.2 years (range 1.3–23.7). The mean (SD) number of self-reported pain types was 2.5 (1.4). The median (IQR) pain intensity scores and the pain-related disability scores on the CPG were 70 (58.3–76.7) and 50 (23.3–63.3) at baseline, and 70 (56.7–80.0; p = 0.104) and 40 (16.7–66.7; p = 0.088) at 3 months follow-up, respectively. None of the demographic or injury-related variables were significantly associated with the baseline scores or with the changes in pain intensity or pain-related disability.

Changes in pain coping strategies and cognitions

Favorable changes in pain coping strategies and pain cognitions between baseline and follow-up were found for 6 out of 13 scales: the Pain Transformation subscale (increase), the Active Coping subtotal score (which includes Pain Transformation) (increase), the Worrying subscale (decrease), and the subscales for Catastrophizing (decrease), Optimism (increase), and Reliance on Health Care (decrease) (Table 6.2).

Table 6.2 Median coping and cognition scores and comparison of pre- and post-intervention coping and cognition scores (n = 47)

Coping and cognition scales	Pre Median (IQR)	Post Median (IQR)	p (Pre–Post)
Pain Coping			
Active Coping	25.0 (22.0–27.0)	26.0 (23.0–29.0)	0.044*
Pain Transformation	7.0 (6.0–9.0)	8.0 (6.0–10.0)	0.038*
Distraction	11.0 (9.0–12.0)	11.0 (9.0–13.0)	0.084
Reducing Demands	6.0 (5.0–7.0)	7.0 (5.0–8.0)	0.491
Passive Coping[a]	40.0 (35.0–49.0)	40.0 (34.0–50.0)	0.182
Retreating	12.0 (10.0–16.0)	13.0 (10.0–16.0)	0.660
Worrying[a]	17.0 (14.0–22.0)	17.0 (14.0–21.0)	0.045*
Resting[a]	11.0 (9.0–14.0)	11.0 (9.0–13.0)	0.265
Pain cognitions			
Catastrophizing[a]	43.0 (32.0–52.0)	37.0 (29.0–48.0)	0.003**
Restrictions[a]	23.0 (17.0–27.0)	20.0 (18.0–25.0)	0.237
Optimism	26.0 (23.0–29.0)	27.0 (25.0–30.0)	0.015*
Internal Control	15.0 (13.0–16.0)	14.0 (12.0–18.0)	0.342
Reliance on Health Care[a]	12.0 (11.0–15.0)	12.0 (10.0–13.0)	0.002**

IQR: interquartile range.
[a] Decrease in score indicates change in a favorable direction.
* Statistically significant at the 0.05 level (2-tailed).
** Statistically significant at the 0.01 level (2-tailed).

Baseline pain coping strategies and cognitions and pain outcomes

At baseline, higher scores on 3 of the coping subscales (Retreating, Worrying, and Resting), on the subtotal score for Passive Coping, and on the cognition subscale for Catastrophizing were associated with higher baseline pain intensity. Higher baseline scores on the coping subscales for Retreating and Resting, the subtotal score for Passive Coping, and the cognition subscales for Catastrophizing and Restrictions were associated with higher baseline pain-related disability (Table 6.3).

Baseline pain coping strategies and cognitions and changes in pain outcomes

Only a higher baseline score for Reliance on Health Care was associated with a larger decrease in pain intensity and pain-related disability between baseline and follow-up (Table 6.3).

Table 6.3 Spearman correlations between baseline coping and cognitions and pain intensity and pain-related disability

Baseline coping and cognition scales	Pain intensity		Pain-related disability	
	Baseline[a]	Change[b]	Baseline[a]	Change[b]
Pain Coping				
Active Coping	0.02	0.12	0.01	0.14
Pain Transformation	0.01	-0.03	0.21	-0.02
Distraction	-0.03	0.09	-0.03	0.14
Reducing Demands	-0.01	0.13	-0.09	-0.03
Passive Coping[c]	0.48***	-0.08	0.50***	0.18
Retreating	0.35*	0.03	0.44**	0.20
Worrying[c]	0.35*	-0.19	0.29	0.03
Resting[c]	0.38**	0.01	0.40**	0.08
Pain cognitions				
Catastrophizing[c]	0.44**	-0.19	0.35*	0.04
Restrictions[c]	0.09	-0.07	0.34*	0.11
Optimism	-0.11	0.24	-0.28	0.27
Internal Control	-0.06	0.21	-0.06	0.14
Reliance on Health Care[c]	0.00	0.40**	-0.08	0.35*

[a] Baseline is preintervention.
[b] Change is the difference between baseline and follow-up scores.
[c] Decrease in score indicates change in a favorable direction.
* Association is significant at the 0.05 level (2-tailed).
** Association is significant at the 0.01 level (2-tailed).
*** Association is significant at the 0.001 level (2-tailed).

Changes in pain coping strategies and cognitions, and changes in pain outcomes

No significant correlations were found between changes in coping strategies and cognitions and changes in pain intensity. Favorable changes in the cognition subscales for Catastrophizing and Restrictions were significantly correlated with favorable changes in pain-related disability (Table 6.4).

Table 6.4 Spearman correlations between change in coping and cognitions and pain intensity and pain-related disability

Change in coping and cognition scales[a]	Change in pain intensity[a]	Change in pain-related disability[a]
Pain Coping		
Active Coping	-0.26	0.02
Pain Transformation	-0.20	0.10
Distraction	-0.24	0.02
Reducing Demands	0.15	0.15
Passive Coping[b]	-0.06	0.29
Retreating	0.02	0.13
Worrying[b]	0.02	0.28
Resting[b]	-0.15	0.19
Pain cognitions		
Catastrophizing[b]	0.24	0.39**
Restrictions[b]	0.18	0.31*
Optimism	-0.25	-0.04
Internal Control	-0.10	-0.17
Reliance on Health Care[b]	0.04	-0.01

[a] Change is the difference between baseline and follow-up scores.
[b] Decrease in score indicates change in a favorable direction.
* Association is significant at the 0.05 level (2-tailed).
** Association is significant at the 0.01 level (2-tailed).

Discussion

This study explored the associations between pain coping strategies and cognitions on the one hand and pain intensity and pain-related disability on the other hand, all measured in the context of a multidisciplinary CBT program for CNSCIP. We found baseline pain coping strategies and pain cognitions to be associated with baseline pain intensity and pain-related disability. Only the baseline cognition Reliance on Health Care was associated with changes in pain intensity and pain-related disability. Changes in the cognitions Catastrophizing and

Restrictions were associated with changes in pain-related disability. Hence, participants with a high degree of Reliance on Health Care and showing changes in Catastrophizing and Restrictions may have benefited most from the intervention.

Little is known about the way pain intensity and pain-related disability change after completion of multidisciplinary pain programs and what factors are associated with these changes.[21] In contrast with other studies of CBT in the field of SCI pain, which reported change in catastrophizing cognitions only,[18,19] we found favorable changes in 2 out of 6 coping strategies (an increase in Pain Transformation and a decrease in Worrying), as well as in 1 out of 2 coping subtotal scores (an increase in Active Coping), and 3 out of 6 pain cognitions (a decrease in Catastrophizing, an increase in Optimism, and a decrease in Reliance on Health Care). Although intensive CBT (more than 100 contact hours) has been proposed for chronic back pain patients,[32] our results suggest that pain coping strategies and cognitions in persons with CNSCIP may already change during a relatively brief CBT intervention and might be associated with (changes in) pain intensity and pain-related disability. However, other aspects of this intervention may have contributed to its outcomes, for example, advice about lifestyle and sports workshops. Changes in pain coping strategies and cognitions may also have been prompted by contacts with other persons with CNSCIP, for example, contact with peers with more pain may have caused a change of perspective.

Pain cognitions

Only the baseline score for Reliance on Health Care was associated with change in the outcome measures. This is an unexpected result, as persons who rely very much on health care have hope for pain relief but expect others to deliver the necessary effort. Their cognitions are characterized by an external locus of control, with denial of their share in the process and a focus on health care providers for help.[31] External control is usually considered negative, being associated with higher pain-related disability and poorer health and well-being.[7] Raichle et al.[15] also found that beliefs that medications are suitable for treating chronic pain, that a medical cure for one's pain exists, and that others should offer assistance in response to pain behaviors (solicitude) were negatively related to functioning. However, it is possible that participants with higher scores for Reliance on Health Care had more positive expectations of the intervention, and it has been shown that positive expectations of participants are related to favorable outcomes.[33-35] It would be interesting to assess participants' pre-existing expectations of the intervention in future studies to test directly the effects achieved through expectancy mechanisms.

Correlations were found between baseline Catastrophizing and baseline pain intensity and baseline pain-related disability. This is in agreement with cross-sectional studies among persons with SCI and persistent pain, showing strong associations between catastrophizing and pain severity,[7,12,13,36,37] physical functioning,[12,14,15,36-38] and psychological functioning.[7,12,14,15,36-38]

A decrease in catastrophizing was not associated with a decrease in pain intensity, but was associated with a decrease in pain-related disability. One longitudinal study in an SCI sample by Hanley et al.[39] found changes in catastrophizing over 6 months to be significantly associated with changes in pain-related disability. In a systematic review of chronic pain in persons with physical disabilities, Jensen et al.[9] found a moderate association of catastrophizing with pain severity but stronger associations with physical functioning. These results concur with ours. It is possible that participants in the present study who improved in terms of catastrophizing did not perceive less pain, but learned from the intervention to become more active despite their CNSCIP.

We found the cognition subscale for Restrictions at baseline to be associated with baseline pain-related disability, and we found changes in Restrictions to be associated with changes in pain-related disability. Persons with a high score on the cognition subscale for Restrictions avoid physical activity and experience little control over their situation.[31] A study by Raichle et al.[15] also found control (belief in one's own control over pain) to be significantly associated with physical and psychological functioning.

Pain coping

We found baseline scores for passive coping, but not those for active coping, to be associated with baseline pain intensity and pain-related disability. Other studies have found passive coping responses (like resting, guarding) to be associated with physical functioning.[14,15,21] Cross-sectional SCI studies found that, in addition to passive coping, the following coping strategies were associated with more favorable outcomes: acceptance (e.g., acceptance of disability, acceptance of SCI),[37,38,40] reinterpreting pain sensations, coping self-statements, ignoring pain sensations,[12] task persistence,[14,15] and relaxation and exercise.[39] Pain coping subscale scores at baseline in our study were not associated with changes in pain severity or pain-related disability during the intervention. Although some changes in coping scores during the intervention were found, these changes were not associated with change in either pain severity or pain-related disability. The reason for this is unclear and warrants further investigation.

Limitations

Due to the relatively small number of participants in this study, only correlations of at least 0.30 were statistically significant, and no multivariate statistics could be used. Twenty out of 91 correlations were significant. Although this is more than can be expected by chance alone (5/100), some of these might be attributed to chance. The results of this study should therefore be considered exploratory and require further confirmation. Nevertheless, this study has provided useful data, because little is known about change in coping strategies and cognitions in response to CBT for CNSCIP. The analyses were conducted without a control group, so do not permit causal inference. There is therefore a need for further research using larger samples with a control group to study the effectiveness of CBT-based interventions and to investigate changes in pain cognitions and pain coping strategies in relation to the treatment effects of CBT in this population.

Another limitation of this study is the self-report nature of the neuropathic pain, completeness, and level of injury variables. However, all persons were known to the physiatrist, and if there was any doubt about the presence of neuropathic pain or the injury characteristics, the physiatrist was asked for further information.

Although the instruments used in the present study were valid and commonly used in the Netherlands, other studies on SCI pain used different instruments to determine pain coping strategies and cognitions, which affects the comparability with other studies.

Clinical implications

Changes in pain coping strategies and pain cognitions in a favorable direction between baseline and follow-up were found for 6 out of 13 scales, suggesting that CBT of relatively limited intensity and duration might be useful to change pain cognitions and coping responses in persons with CNSCIP. Reliance on health care was associated with a decrease in pain intensity and pain-related disability, suggesting that this pain cognition might predict the effectiveness of CBT for CNSCIP. The results of this study suggest that CBT programs should aim at decreasing the cognitions of catastrophizing, restrictions, and reliance on health care and decreasing passive coping to maximize the effectiveness of this type of intervention.

Conclusion

This explorative study has provided new information on the effects of a CBT program on pain coping strategies and pain cognitions in persons with CNSCIP. Our results suggest that pain coping strategies and cognitions may change during a brief CBT intervention, which could have some beneficial effect on pain intensity and pain-related disability in persons with CNSCIP.

Acknowledgments

The authors would like to thank the participants in this study, the participating Dutch rehabilitation centers, and the co-trainers in these centers: Peter Luthart (De Hoogstraat Rehabilitation, Utrecht); Sandra Slangen (Adelante Zorggroep, Hoensbroek); Marijke Schuitemaker (Rehabilitation Center Het Roessingh, Enschede); Hilde Schors, Lonneke Vlemmix, and Jolante Sweers (Rehabilitation Center Rijndam, Rotterdam).

References

1. Siddall PJ, McClelland JM, Rutkowski SB, Cousins MJ. A longitudinal study of the prevalence and characteristics of pain in the first 5 years following spinal cord injury. Pain 2003;103:249-257.
2. Siddall PJ, Taylor DA, McClelland JM, Rutkowski SB, Cousins MJ. Pain report and the relationship of pain to physical factors in the first 6 months following spinal cord injury. Pain 1999;81:187-197.
3. Siddall PJ. Management of neuropathic pain following spinal cord injury: now and in the future. Spinal Cord 2009;47:352-359.
4. Widerström-Noga EG, Felipe-Cuervo E, Broton JG, Duncan RC, Yezierski RP. Perceived difficulty in dealing with consequences of spinal cord injury. Arch Phys Med Rehabil 1999;80:580-586.
5. Jensen MP, Hoffman AJ, Cardenas DD. Chronic pain in individuals with spinal cord injury: a survey and longitudinal study. Spinal Cord 2005;43:704-712.
6. Siddall PJ, Loeser JD. Pain following spinal cord injury. Spinal Cord 2001;39:63-73.
7. Wollaars MM, Post MW, van Asbeck FW, Brand N. Spinal cord injury pain: the influence of psychologic factors and impact on quality of life. Clin J Pain 2007;23:383-391.
8. Widerström-Noga EG, Felix ER, Cruz-Almeida Y, Turk DC. Psychosocial subgroups in persons with spinal cord injuries and chronic pain. Arch Phys Med Rehabil 2007;88:1628-1635.
9. Jensen MP, Moore MR, Bockow TB, Ehde DM, Engel JM. Psychosocial factors and adjustment to chronic pain in persons with physical disabilities: a systematic review. Arch Phys Med Rehabil 2011;92:146-160.

10. Haythornthwaite JA, Benrud-Larson LM. Psychological aspects of neuropathic pain. Clin J Pain 2000;16:S101-105.

11. Turk DC, Okifuji A. Psychological factors in chronic pain: evolution and revolution. J Consult Clin Psychol 2002;70:678-690.

12. Turner JA, Jensen MP, Warms CA, Cardenas DD. Catastrophizing is associated with pain intensity, psychological distress, and pain-related disability among individuals with chronic pain after spinal cord injury. Pain 2002;98:127-134.

13. Giardino ND, Jensen MP, Turner JA, Ehde DM, Cardenas DD. Social environment moderates the association between catastrophizing and pain among persons with a spinal cord injury. Pain 2003;106:19-25.

14. Molton IR, Stoelb BL, Jensen MP, Ehde DM, Raichle KA, Cardenas DD. Psychosocial factors and adjustment to chronic pain in spinal cord injury: replication and cross-validation. J Rehabil Res Dev 2009;46:31-42.

15. Raichle KA, Hanley M, Jensen MP, Cardenas DD. Cognitions, coping, and social environment predict adjustment to pain in spinal cord injury. J Pain 2007;8:718-729.

16. Ehde DM, Jensen MP. Psychological treatments for pain management in persons with spinal cord injury: cognitive therapy and self-hypnosis training. Top Spinal Cord Inj Rehabil 2007;13:72-80.

17. Turner JA, Holtzman S, Mancl L. Mediators, moderators, and predictors of therapeutic change in cognitive-behavioral therapy for chronic pain. Pain 2007;127:276-286.

18. Kennedy P, Duff J, Evans M, Beedie A. Coping effectiveness training reduces depression and anxiety following traumatic spinal cord injuries. Br J Clin Psychol 2003;42:41-52.

19. Nicholson Perry K, Nicholas MK, Middleton JW. Comparison of a pain management program with usual care in a pain management center for people with spinal cord injury-related chronic pain. Clin J Pain 2010;26:206-216.

20. Norrbrink Budh C, Kowalski J, Lundeberg T. A comprehensive pain management programme comprising educational, cognitive and behavioural interventions for neuropathic pain following spinal cord injury. J Rehabil Med 2006;38:172-180.

21. Jensen MP, Turner JA, Romano JM. Changes after multidisciplinary pain treatment in patient pain beliefs and coping are associated with concurrent changes in patient functioning. Pain 2007;131:38-47.

22. Heutink M, Post MW, Luthart P, Pfennings LE, Dijkstra CA, Lindeman E. A multidisciplinary cognitive behavioural programme for coping with chronic neuropathic pain following spinal cord injury: the protocol of the CONECSI trial. BMC Neurol 2010;10:96.

23. Heutink M, Post MW, Bongers-Janssen HM, Dijkstra CA, Snoek GJ, Spijkerman DC, Lindeman E. The CONECSI trial: results of a randomized controlled trial of a multidisciplinary cognitive behavioral program for coping with chronic neuropathic pain after spinal cord injury. Pain 2012;153:120-128.

24. Von Korff M, Ormel J, Keefe FJ, Dworkin SF. Grading the severity of chronic pain. Pain 1992;50:133-149.

25. Engel GL. The need for a new medical model: a challenge for biomedicine. Science 1977;196:129-136.

26. Ellis A. Rational psychotherapy and individual psychology. J Individ Psychol 1957;13:38-44.

27. Bouhassira D, Attal N, Alchaar H, Boureau F, Brochet B, Bruxelle J, Cunin G, Fermanian J, Ginies P, Grun-Overdyking A, Jafari-Schluep H, Lantéri-Minet M, Laurent B, Mick G, Serrie A, Valade D, Vicaut E. Comparison of pain syndromes associated with nervous or somatic lesions and development of a new neuropathic pain diagnostic questionnaire (DN4). Pain 2005;114:29-36.

28. Kraaimaat FW, Evers AW. Pain-coping strategies in chronic pain patients: psychometric characteristics of the pain-coping inventory (PCI). Int J Behav Med 2003;10:343-363.

29. Kraaimaat FW, Bakker A, Evers AW. Pijncoping-strategieën bij chronische pijnpatiënten: de ontwikkeling van de pijn-coping-inventarisatielijst (PCI) [Pain coping strategies in chronic pain patients: the development of the pain coping inventory (PCI)]. Gedragstherapie 1997;3:185-201 (in Dutch).

30. Van Breukelen GJ, Vlaeyen JW. Norming clinical questionnaires with multiple regression: the pain cognition list. Psychol Assess 2005;17:336-344.

31. Vlaeyen JW, van Breukelen G, Nooyen-Haazen IW, Stomp-van den Berg SG, Kole-Snijders AM. Meetinstrumenten chronische pijn: deel 4. Pijn Cognitie Lijst-2003. Handleiding [Measuring instruments for chronic pain: part 4. Pain Cognition List-2003. Manual]. Maastricht: Pijn Kennis Centrum; 2004 (in Dutch).

32. Guzman J, Esmail R, Karjalainen K, Malmivaara A, Irvin E, Bombardier C. Multidisciplinary rehabilitation for chronic low back pain: systematic review. BMJ 2001;322:1511-1516.

33. Mondloch MV, Cole DC, Frank JW. Does how you do depend on how you think you'll do? A systematic review of the evidence for a relation between patients' recovery expectations and health outcomes. CMAJ 2001;165:174-179.

34. Linde K, Witt CM, Streng A, Weidenhammer W, Wagenpfeil S, Brinkhaus B, Willich SN, Melchart D. The impact of patient expectations on outcomes in four randomized controlled trials of acupuncture in patients with chronic pain. Pain 2007;128:264-271.

35. Crow R, Gage H, Hampson S, Hart J, Kimber A, Thomas H. The role of expectancies in the placebo effect and their use in the delivery of health care: a systematic review. Health Technol Assess 1999;3:1-96.

36. Ullrich P, Jensen M, Loeser J, Cardenas D. Catastrophizing mediates associations between pain severity, psychological distress, and functional disability among persons with spinal cord injury. Rehabil Psychol 2007;52:390-398.

37. Nicholson Perry K, Nicholas MK, Middleton J. Spinal cord injury-related pain in rehabilitation: a cross-sectional study of relationships with cognitions, mood and physical function. Eur J Pain 2009;13:511-517.

38. Nicholson Perry K, Nicholas MK, Middleton J, Siddall P. Psychological characteristics of people with spinal cord injury-related persisting pain referred to a tertiary pain management center. J Rehabil Res Dev 2009;46:57-67.

39. Hanley MA, Raichle K, Jensen M, Cardenas DD. Pain catastrophizing and beliefs predict changes in pain interference and psychological functioning in persons with spinal cord injury. J Pain 2008;9:863-871.

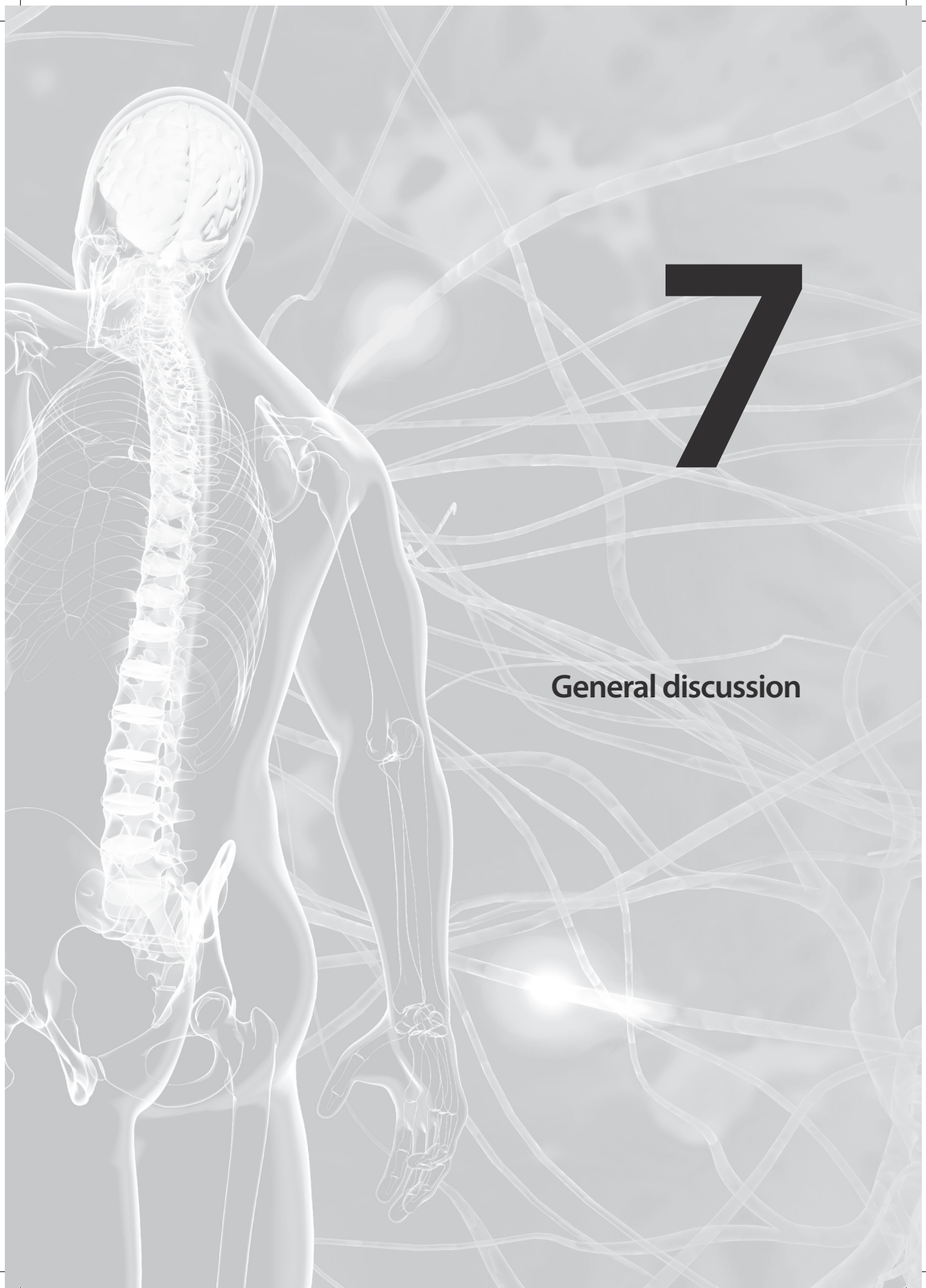
40. Summers JD, Rapoff MA, Varghese G, Porter K, Palmer RE. Psychosocial factors in chronic spinal cord injury pain. Pain 1991;47:183-189.

Appendix 6.1 Main contents of the 11 sessions of the cognitive behavioral program

Sessions	Main contents
Session 1	Information on BPS model Information on SCI and CNSCIP Goal setting
Session 2 Buddies	Information on ABC model (ABC) Information by physiatrist specialized in SCI rehabilitation
Session 3	Expansion of ABC model (ABCDE) Information by physiatrist specialized in chronic pain rehabilitation Information on movement and pain
Session 4	Sports workshop Evaluation of sports workshop with ABC model
Session 5	Information on assertiveness and communication about pain Information by role model Introduction to relaxation exercises Evaluation of goals
Session 6	Information on pain, mood, and stress Workshop on relaxation exercises Evaluation of workshop on relaxation exercises
Session 7	Sports workshop Evaluation of sports workshop with BPS model and ABC model
Session 8 Buddies	Information on social aspects and partner, family, and friends Workshop on relaxation exercises Evaluation of workshop on relaxation exercises
Session 9	Sports workshop Evaluation of sports workshop with ABC model Workshop on relaxation exercises Evaluation of workshop on relaxation exercises
Session 10	Summary and recapitulation of intervention contents Evaluation of goals Application in everyday life
Session 11	Summary of intervention contents Evaluation of intervention

BPS: BioPsychoSocial; SCI: spinal cord injury; CNSCIP: chronic neuropathic spinal cord injury pain; ABC: Activating event-Belief-Consequence.

6



7

General discussion

The general aim of this thesis was to describe spinal cord injury (SCI) pain treatments and their effectiveness, mainly focused on a cognitive behavioural programme for coping with chronic neuropathic SCI pain. This last chapter starts with an overview of the main findings of our studies. Subsequently, our treatment programme and findings in relation to other cognitive behavioural programmes for SCI pain, cognitive behavioural programmes in other diagnoses with neuropathic pain, and methodological considerations are discussed. Finally, clinical implications and directions for future research are presented.

Main findings

Neuropathic pain was the most frequently occurring pain type (69.3%) in persons with chronic SCI pain and this pain type was rated as most intense. Persons with SCI pain have often tried many pain treatments and 63.8% was involved in some kind of pain treatment at the time of the survey. Psychological treatment, like cognitive behavioural treatment, was found to relieve SCI pain, but this kind of treatment is not often used in the SCI population (**Chapter 2**). Because of the need for the evaluation of cognitive behavioural treatment for chronic neuropathic SCI pain, we composed such a treatment programme for coping with this type of pain. The CONECSI (COping with NEuropathiC Spinal cord Injury pain) randomised controlled trial (RCT) was conducted to evaluate the effectiveness of this programme. **Chapter 3** described the protocol of the CONECSI trial and the content of the cognitive behavioural programme for chronic neuropathic SCI pain. Beneficial effects in the short- and long-term were found (**Chapter 4** and **Chapter 5**). The main short-term results of the CONECSI trial were decrease of anxiety and increase of participation in activities. Besides this, a significant decrease in pain intensity and pain-related disability was also found over time, but without a significant difference between intervention and waiting list control group (**Chapter 4**). In the long-term significant decrease of pain intensity, pain-related disability, and anxiety, and a significant increase in participation in activities were found in the intervention group (**Chapter 5**). **Chapter 6** showed that participation in the programme was associated with improvements of coping strategies: pain transformation (increase), active coping (increase), and worrying (decrease), and pain cognitions: catastrophising (decrease), optimism (increase), and reliance on health care (decrease). A higher baseline score for reliance on health care was associated with a larger decrease in pain intensity and pain-related disability. A decrease of catastrophising and restriction cognitions was associated with a decrease of pain-related disability. These results support the notion that it is possible to modify dysfunctional pain coping strategies and cognitions by a cognitive

behavioural intervention, with some beneficial effects of this approach on pain intensity and pain-related disability (**Chapter 6**).

Our findings in relation to other cognitive behavioural SCI pain programmes

Cognitive behavioural programmes are the prevailing psychological treatment for many chronic pain conditions, like low back pain,[1] but have not been systematically examined in persons with chronic neuropathic SCI pain yet. At the start of our study, only one intervention study of a cognitive behavioural pain management programme for neuropathic SCI pain was available.[2] This programme was the inspiration to develop our treatment programme and to design the CONECSI trial to evaluate this programme, with measurements pre- and immediately post-treatment, and at 3, 6, and 9 months post-treatment.

The study by Norrbrink Budh et al.[2] was not randomised and included only 27 persons in the intervention group and 11 controls, with measurements pre- and post-treatment, and at 3, 6, and 12 months follow-up. Since the start of our study, only two small non-randomised studies on cognitive behavioural programmes for chronic SCI pain, not exclusively neuropathic pain, have been published.[3,4] The study by Nicholson Perry et al.[3] included 19 persons (89.5% neuropathic pain) in a multidisciplinary cognitive behavioural pain management programme, with measurements pre- and post-treatment, and at 1 and 9 months follow-up, and 17 persons (94% neuropathic pain) in the standard care group (including medical management, individual physiotherapy, and clinical psychology interventions for pain management). The study by Burns et al.[4] included 22 persons (94% neuropathic pain) in the pain programme with measurements pre- and post-treatment, and at 3 and 12 months follow-up, but lacked a control group.

All four programmes (including ours) contained essentially the same main elements: educational sessions about pain, cognitive restructuring, relaxation, and (light) exercise. The programme of Norrbrink Budh et al.[2] included in addition a body awareness training, whereas in our programme more attention was paid to education about the relationship between biological, psychological, social factors and pain (**Chapter 3**).

The following is a comparison of the results of the four studies, although this comparison is hampered by the variability in study designs and outcome measures. We found treatment effects on anxiety and participation in activities (**Chapter 4**) with lasting improvements in the intervention group 9 months post-treatment (**Chapter 5**). With respect to anxiety, Norrbrink

Budh et al.[2] showed a decreased level in the treatment group after the programme compared with baseline, and with lasting improvements at the 12-month follow-up, but no significant differences between treatment group and control group. Nicholson Perry et al.[3] showed decrease of anxiety lasting at 9 months, but also without significant difference between the groups. Burns et al.[4] did not examine effect on anxiety and used a history of major anxiety (or depressive) disorder as an exclusion criterion. With respect to participation in activities, Norrbrink Budh et al.[2] and Nicholson Perry et al.[3] did not examine participation in activities in their studies and Burns et al.[4] found no significant changes in activities.

We found no treatment effect on life satisfaction (**Chapter 4 and 5**), just like Norrbrink Budh et al.[2] and Burns et al.[4] Nicholson Perry et al.[3] did not examine life satisfaction. Although depression scores decreased, we found no treatment effect on depression (**Chapter 4 and 5**). Nicholson Perry et al.[3] neither found a decrease of depression. Burns et al.[4] did not examine depression. However, Norrbrink Budh et al.[2] found a significant decrease of depression and a treatment effect with lasting improvements at 12 months after the programme. It is not clear why Norrbrink Budh et al.[2] found positive effects on depression that were not found in the other studies. Maybe the body awareness training, that was not included in the other programmes, had a positive effect. Alternatively, it might be a finding due to chance since Norrbrink Budh et al.[2] used a small sample and analysed 17 outcome measures (increasing the likelihood of making a type I error) of which only 2 showed a significant difference between the treatment and the control group.

We also found a significant, lasting decrease of pain intensity and pain-related disability, but without treatment effect (**Chapter 4 and 5**). Nicholson Perry et al.[3] found a non-significant decrease of pain intensity, and no difference with the usual care group. However, they found an initial decrease of life interference due to pain in comparison with the usual care group, but the scores of their intervention group had returned to a level comparable to pre-treatment at the 9-month follow-up. Burns et al.[4] found no effect on pain severity. However, they did find a decrease of pain interference in daily life, not immediately after the intervention but 12 months after the intervention. Norrbrink Budh et al.[2] found no changes in pain intensity and pain unpleasantness. It appears our intervention had similar or better results on pain and pain-related disability compared to the other studies.

Finally, we showed improvements in pain coping and pain cognitions after participation in the programme. Nicholson Perry et al.[3] found, like we did, a significant improvement over time in the pain cognition catastrophising in the pain management programme group. Pain cognitions were not taken into account in the other two studies.[2,4] Nicholson Perry et al.[3] and

Burns et al.[4] did not find any change of pain coping styles during or after the intervention. Norrbrink Budh et al.[2] did not take pain coping styles into account. Nicholson Perry et al.[3] did not examine relations between changes in pain cognitions and changes in the outcomes, like we did in **Chapter 6**.

Cognitive behavioural programmes for neuropathic pain in other diagnoses

Because the lack of other cognitive behavioural programmes for chronic neuropathic SCI pain and RCTs to evaluate their effectiveness, it is interesting to take a closer look at cognitive behavioural programmes for neuropathic pain in other diagnoses. Neuropathic pain can be caused by a number of different diseases (e.g. diabetes mellitus, herpes zoster, HIV infection), medical interventions (e.g. chemotherapy, surgery), and injuries (e.g. brachial plexus avulsion).[5] However, hardly any clinical trials have examined the effectiveness of psychological treatment for neuropathic pain in other diagnostic groups.[6] To our knowledge, the only trial of cognitive behavioural treatment was a small preliminary study on HIV-related peripheral neuropathic pain.[7] In this study significant benefits of a cognitive behaviour intervention were found on distress in comparison with supportive psychotherapy. The cognitive behaviour intervention group also showed greater reductions in pain intensity and pain-related interference with functioning than the supportive psychotherapy group, but these differences were not statistically significant.

In conclusion, the use of cognitive behavioural interventions has hardly been tested in any neuropathic pain population. This emphasises the uniqueness of the CONECSI trial and the importance of subsequent research.

Methodological considerations

The cross-sectional study and the RCT reported in this thesis have several strengths. They give more insight into the effectiveness of pain treatments in persons with SCI pain, and the CONECSI trial encompass the first RCT of a cognitive behavioural programme for chronic neuropathic SCI pain with focus on both short-term as long-term outcomes, and exploration of pain coping and pain cognitions in this population. As a consequence, this study significantly contributed to the limited evidence of such programmes for persons with chronic neuropathic SCI pain. However, it has also limitations that have to be considered.

Study population

It is possible that certain types of persons are more attracted than others to participate in a cognitive behavioural treatment programme, so selection bias might have played a role in our trial (**Chapter 4, 5 and 6**). Misconceptions or social stigma associated with psychological treatment[8] might have deterred others to participate in this study. To avoid this as much as possible, we did not mention the terms psychological treatment and cognitive behavioural intervention in the invitation letters to participate in the trial. But this might still have influenced the representativeness of the study sample and thereby the degree to which the results of our study can be generalised to the whole population of persons with neuropathic SCI pain. Selection bias might also have occurred in our cross-sectional study (**Chapter 2**), since it is possible that persons with SCI pain were more attracted to participate in the study than persons without SCI pain, thereby inflating the prevalence of pain after SCI.

Study design

Unfortunately, it was not possible in the time frame of our study, in which the waiting list control group had to wait for six months before the start of the intervention, to perform long-term follow-up measurements in this control group (**Chapter 5 and 6**). Nevertheless, these studies have provided useful information, because very little is known about long-term effects of a cognitive behavioural intervention in this population and which pain cognitions and coping strategies could play an important role.

Although this study reports both short-term and long-term benefits from participation in a multidisciplinary pain programme for persons with chronic neuropathic SCI pain, the contribution of individual components of the programme remains unclear. In the ideal world, a study should compare a treatment programme that differs in only one aspect with the programme of the control group and both groups get an equal dose of attention, expectation, and information. However, this requires very strict treatment protocols and does not reflect the multi-component aspect of pain and pain treatment. Also, if two multidisciplinary programmes differ in only one aspect, small effect sizes are to be expected and consequently large samples will be needed to show significant differences.

Besides the changes in scores on the primary and secondary outcome measures in the intervention group, we see a trend in the scores of the waiting list control group in the same direction during the waiting time before the start of the programme. An explanation for these changes in scores of the waiting list control group might be the 'Hawthorne-effect': the positive effect of participating in a study. Participants have the perspective to participate

in the upcoming programme (expectation bias), get attention from the researchers and complete questionnaires about important aspects related to the intervention. It is difficult to avoid this, because persons have to give informed consent and be aware of participating in a study.

The cognitive behavioural programme

The investigated multidisciplinary cognitive behavioural programme consists of 11 sessions of 3 hours, with 33 contact hours in total. This length was mainly determined by the number of themes to be covered. Furthermore, it was a compromise between desired intensity, participant burden, and feasibility for the organisation. An interesting question is how intensive such a cognitive behavioural programme for chronic neuropathic SCI pain should be. Intensive (over 100 hours) multidisciplinary biopsychosocial rehabilitation has been proposed in persons with chronic back pain,[9] but the other published cognitive behavioural programmes for SCI pain comprised also less hours than this recommendation, namely 25 contact hours,[4] 45 contact hours,[3] and 50 hours of treatment.[2] Maybe a more intensive programme would have shown better results, although the number, frequency, and length of the sessions were judged adequate by the participants. The evaluation meeting with the trainers, who executed the study, neither showed a desire for a higher intensity, except the remark to loosen the schedule of the first sessions a bit.

In general, the trainers and participants were satisfied with the intervention. Sport workshops, peer support, information on pain, mood and stress, and information by physiatrist specialised in SCI rehabilitation were the most often appreciated elements by the participants, followed by relaxation exercises, information on assertiveness and communication about pain, information on movement and pain, and the Activating event-Belief-Consequence (ABC) model. Both participants and trainers did not agree among themselves on the relative importance of the various aspects of the programme, therefore there was no content removed from the programme after the trial.

Because the initial improvements in participation in activities and anxiety tended to regress toward baseline in scores at 6 months after the last meeting (**Chapter 5**), like the improvements at the follow-up in other programmes,[3,4] it would be useful to study the added value of more periodic relapse prevention (booster) sessions in the months after finishing the programme to reinforce learned strategies and maintain long-term efficacy.

Clinical implications

We recommend more psychological treatment in SCI rehabilitation, and more specific cognitive behavioural treatment for neuropathic SCI pain. Chronic neuropathic pain has a high impact on the lives of many persons with SCI. This multidisciplinary cognitive behavioural programme can help persons with neuropathic SCI pain by learning better coping skills and thereby minimize the impact of pain on his or her life. This programme is therefore a valuable addition to the treatment arsenal in the SCI rehabilitation. The chances on implementation of this intervention in daily practice appear to be good. First, the programme fits well in a rehabilitation setting in the Netherlands because of the multidisciplinary design and the demonstrated feasibility of the treatment protocol in four rehabilitation centres. In response to comments made by the trainers we added one session to loosen the schedule of the first few sessions, which will also contributed to the feasibility of the intervention. Second, the experiences with this intervention gained during the CONECSI trial facilitates the implementation in the participating rehabilitation centres, particularly if the same trainers are involved. Third, the availability of a written treatment protocol also contributes to applicability in other rehabilitation centres by rehabilitation professionals and facilitates implementation.[10] We wrote an extensive treatment protocol for the trainers, a course book for the participants, and presentation slides to support the trainers during the meetings, together with a checklist to facilitate the start of the programme. All this material of the treatment programme is free available and distributed though the Dutch-Flemish Society for Spinal Cord Injury Rehabilitation.

On the other hand, the time consuming nature of the treatment programme might be a barrier for implementation in the current difficult financial times and it might be a challenge to fit this group intervention in existing timetables of healthcare professionals and facilities, and also in the week schedule of the participants. Further, each meeting was supervised by two trainers, a physiotherapist and a psychologist in three centres. In particular, the needed hours of the psychologist as a trainer are expensive. In one centre, the trainer was a nurse practitioner and there were good results in this centre. However, she was very experienced in SCI, neuropathic pain and group interventions and it cannot be concluded that any nurse practitioner would be able to replace the psychologist. It remains unclear how professional degree, level of training, or expertise to deliver a cognitive behavioural programme to persons with chronic pain affect outcome. Cognitive behavioural techniques, like cognitive restructuring, require the education and experience of a psychologist and may be less appropriate for delivery by others.[1]

Another aspect is the optimal timing of this cognitive behavioural programme after SCI. The median time between the onset of SCI and inclusion was 5.4 years (range 1.4–23.7) and the median duration of neuropathic pain at inclusion was 4.5 years (range 1.3–23.7) in our study. It is possible that participants already had developed some appropriate strategies to cope with neuropathic pain themselves. Many participants reported they would rather have received the programme earlier: some participants mentioned after one year after SCI or even during rehabilitation; others also indicated earlier, but a few years after SCI. In the first months after SCI the focus is to become as independent as possible during inpatient rehabilitation and most persons will want to try the available medications for neuropathic pain first. However, it is recommended to test the effectiveness of this treatment programme in an earlier stage after SCI and onset of neuropathic SCI pain, and offer the possibility of participation in this programme and psychological support at multiple time points in rehabilitation.

Directions for future research

It would be interesting to evaluate this treatment programme in persons with other diagnoses and neuropathic pain, because the programme may easily be adjusted to a different diagnosis. However, future research should also determine the effectiveness of a tailored approach, meaning which pain treatment or combination of treatments is most effective for what type of person with chronic pain and under what circumstances.[1,11,12] It is necessary to determine which cognitive and behavioural variables mediate the effects of cognitive behavioural treatment on outcomes[1] and to identify specific treatment aspects of a cognitive behavioural programme for the best outcome, like timing of treatment, and dose and format (e.g. the use of individual versus group treatment and e-health) of the treatment. Group interventions have a number of important advantages compared to individual therapy, such as cost and time effectiveness for the facilitator and the fact that participants have the opportunity to share experiences and feel acknowledged by fellow sufferers. But some persons might fare best with the alternation of individual and group sessions. E-health (the use of information and communication technologies in health care) is increasing and internet-based cognitive behavioural therapy is shown to serve as a complement in chronic low back pain.[13] Internet-based cognitive behavioural treatment may offer advantages of accessibility and flexibility for persons with a busy schedule who appreciate a self-management approach, or a reduction of stigma that might prevent some persons from seeking psychological care.[1] On the other hand there are disadvantages of

internet-based treatment, like persons have to be very disciplined (e.g. to follow treatment assignments) and motivated.[11] Persons who feel the need for contact with fellow sufferers and thrive in a social context, might benefit more from a face-to-face group treatment programme. Furthermore, many parts of our programme (e.g. sport workshops) can not be performed in this way by e-health. But possibly e-health is a valuable addition and can replace parts of the programme (e.g. provide the information of the two physiatrist by an internet video) or play a role in the programme (e.g. digital homework assignments or booster sessions by internet modules). Future research should investigate the potential of integrating the evolving technology with face-to-face treatments,[11] and the possibility that these technologies might be able to reduce the number of face-to-face contact hours of such a comprehensive cognitive behavioural treatment programme.

In this context, it is interesting to identify modifiable predictors of treatment outcome to increase the effectiveness of treatment. Treatment credibility (how plausible, convincing, and logical the treatment seems to a person) and treatment expectancy (judged likelihood that receiving the treatment will result in therapeutic gain) have been assumed to be such modifiable predictors in rehabilitation for chronic low back pain.[14] Perceived credibility of cognitive behavioural treatment in chronic low back pain[15] and treatment expectancy of a cognitive behavioural interventions for fibromyalgia and chronic low back pain[16] significantly predicted treatment outcome. Strengthening treatment credibility and expectancy might increase the effectiveness of rehabilitation interventions[14] and it is interesting to investigated this in future research in chronic neuropathic SCI pain.

The addition of qualitative research methods, like in-depth interviews with participants from the study and analyses for main themes, can give more insight in both treatment aspects, like optimal content, timing, dose, and format of cognitive behavioural treatment for neuropathic SCI pain, as aspects of participants with neuropathic SCI pain, like perceived credibility and expectancy about the treatment and related, modifiable psychological variables (like intern control of pain and catastrophising), motivation and readiness to change, and what makes a difference to participants in the context of clinically significance.[17]

Cognitive behavioural treatment programmes for chronic pain appear to be cost-effective.[18] Finally, it is interesting to perform a cost-effectiveness study on this cognitive behavioural programme in future research to investigate whether a one-time significant time investment save costs in the long-run.

Conclusion

The studies presented in this thesis have enhanced the level of evidence on the effectiveness of cognitive behavioural programmes for persons with SCI who suffer from chronic neuropathic pain.

References

1. Ehde DM, Dillworth TM, Turner, JA. Cognitive-behavioral therapy for individuals with chronic pain: efficacy, innovations, and directions for research. Am Psychol 2014;69:153-166.
2. Norrbrink Budh C, Kowalski J, Lundeberg T. A comprehensive pain management programme comprising educational, cognitive and behavioural interventions for neuropathic pain following spinal cord injury. J Rehabil Med 2006;38:172-180.
3. Nicholson Perry K, Nicholas MK, Middleton JW. Comparison of a pain management program with usual care in a pain management center for people with spinal cord injury-related chronic pain. Clin J Pain 2010;26:206-216.
4. Burns, AS, Delparte JJ, Ballantyne EC, Boschen KA. Evaluation of an interdisciplinary program for chronic pain after spinal cord injury. PM R 2013;5:832-838.
5. O'Connor AB, Dworkin RH. Treatment of neuropathic pain: an overview of recent guidelines. Am J Med 2009;122:S22-32.
6. Turk DC, Audette J, Levy RM, Mackey SC, Stanos S. Assessment and treatment of psychosocial comorbidities in patients with neuropathic pain. Mayo Clin Proc 2010;85:S42-50.
7. Evans S, Fishman B, Spielman L, Haley A. Randomized trial of cognitive behavior therapy versus supportive psychotherapy for HIV-related peripheral neuropathic pain. Psychosomatics 2003;44:44-50.
8. Schoenberg M, Shiloh S. Hospitalised patients' views on in-ward psychological counselling. Patient Educ Couns 2002;48:123-129.
9. Guzman J, Esmail R, Karjalainen K, Malmivaara A, Irvin E, Bombardier C. Multidisciplinary rehabilitation for chronic low back pain: Systematic review. BMJ 2001;322:1511-1516.
10. Wade DT, Smeets RJ, Verbunt JA. Research in rehabilitation medicine: methodological challenges. J Clin Epidemiol 2010;63:699-704.
11. Jensen MP, Turk DC. Contributions of psychology to the understanding and treatment of people with chronic pain: why it matters to ALL psychologists. Am Psychol 2014;69:105-118.
12. Williams AC, Eccleston C, Morley S. Psychological therapies for the management of chronic pain (excluding headache) in adults. Cochrane Database Syst Rev 2012;11:CD007407.
13. Buhrman M, Nilsson-Ihrfeldt E, Jannert M, Ström L, Andersson G. Guided internet-based cognitive behavioural treatment for chronic back pain reduces pain catastrophizing: a randomized controlled trial. J Rehabil Med 2011;43:500-505.

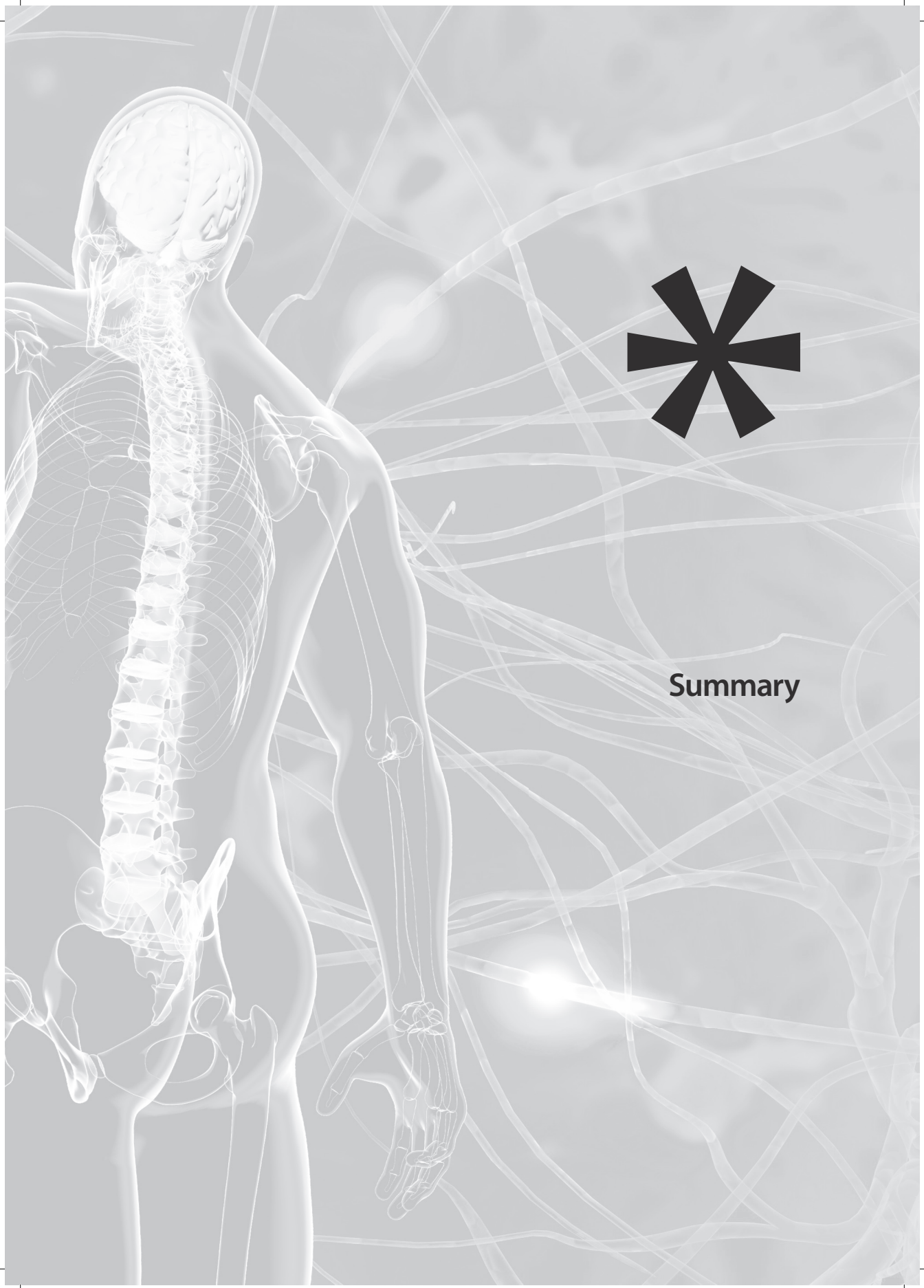
14. Smeets RJ, Beelen S, Goossens ME, Schouten EG, Knottnerus JA, Vlaeyen JW. Treatment expectancy and credibility are associated with the outcome of both physical and cognitive-behavioral treatment in chronic low back pain. Clin J Pain 2008;24:305-315.

15. Kole-Snijders AM, Vlaeyen JW, Goossens ME, Rutten-van Mölken MP, Heuts PH, van Breukelen G, van Eek H. Chronic low-back pain: what does cognitive coping skills training add to operant behavioral treatment? Results of a randomized clinical trial. J Consult Clin Psychol 1999;67:931-944.

16. Goossens ME, Vlaeyen JW, Hidding A, Kole-Snijders A, Evers SM. Treatment expectancy affects the outcome of cognitive-behavioral interventions in chronic pain. Clin J Pain 2005;21:18-26.

17. Wade D. 'But is the difference clinically significant?'. Clin Rehabil 2005;19:349-353.

18. Vlaeyen JW, Morley S. Cognitive-behavioral treatments for chronic pain: what works for whom? Clin J Pain 2005;21:1-8.



Summary

Chapter 1, the general introduction, describes the context, the aims and outline of this thesis. Persons with spinal cord injury (SCI) often face serious secondary health conditions, including neuropathic pain. Pharmacological interventions are often insufficiently effective in providing neuropathic pain relief and, consequently, there is a growing interest in non-pharmacological interventions for neuropathic pain. One of the non-pharmacological treatment options is cognitive behavioural treatment. Although this type of treatment is often applied and found to be effective in chronic pain conditions, it has not been systematically examined in persons with chronic neuropathic SCI pain yet. The general aim of this thesis is to describe SCI pain treatments and their effectiveness, mainly focusing on a cognitive behavioural treatment programme for chronic neuropathic pain after SCI.

Chapter 2 gives an overview of pharmacological and non-pharmacological pain treatments used for chronic pain after SCI, and describes the perceived effectiveness of currently used pain treatments in a cross-sectional study of a large population with SCI. The majority (77.1%) had chronic pain after SCI and more than one type of pain (62.8%), of which neuropathic pain was most frequently reported (69.3%). Persons with chronic pain after SCI have often tried many pain treatments. Although 63.8% of this group was currently involved in some kind of treatment, they still reported high levels of pain. Neuropathic pain was rated as the most severe type of pain. Massage (therapy)/relaxation (training), anticonvulsants, and non-steroidal anti-inflammatory drugs were the most often used pain treatments. The current treatments that were most often perceived as effective were acupuncture/magnetising, cannabis/alcohol, physiotherapy and exercise, and massage (therapy)/relaxation (training). Transcutaneous Electrical Nerve Stimulation/ultrasound were least often perceived as effective. Psychological treatment, like cognitive behavioural treatment, was found to be SCI pain relieving, but was rarely used. It is concluded that further research is needed to establish effective SCI pain treatments, in particular more research on non-pharmacological treatments is desired.

Because evidence of the effectiveness of cognitive behavioural interventions to relieve neuropathic SCI pain from randomised controlled trials (RCTs) was lacking, the CONECSI (COping with NEuropathiC Spinal cord Injury pain) RCT was conducted. **Chapter 3** describes the content of a cognitive behavioural programme for chronic neuropathic SCI pain and the protocol of the CONECSI trial to evaluate the effects of this programme on pain intensity and pain-related disability, and secondary on mood, participation in activities, and life satisfaction. Pain coping strategies and pain cognitions were also assessed. Persons with chronic neuropathic SCI pain were recruited from 4 rehabilitation centres and randomised to an intervention group or a waiting list control group. The control group was invited for the

programme 6 months after the intervention group. Measurements were performed before starting the programme (intervention group) or before entering the control group, and at 3, 6, 9, and 12 months. The intervention consisted of educational, cognitive, and behavioural elements and encompassed 11 sessions of 3 hours over a 3-month period. Cognitive behavioural treatment focuses on modifying irrational beliefs, and dysfunctional pain cognitions and pain coping. The BioPsychoSocial model was used for education about the relationship between biological, psychological, social factors and pain, and the importance of maintaining balance between load and capacity. The Activating event-Belief-Consequence model was used for cognitive restructuring for better coping with neuropathic pain. The two models were explained in educational sessions and in guided group discussions using fictitious cases, and applied in sports workshops and homework assignments.

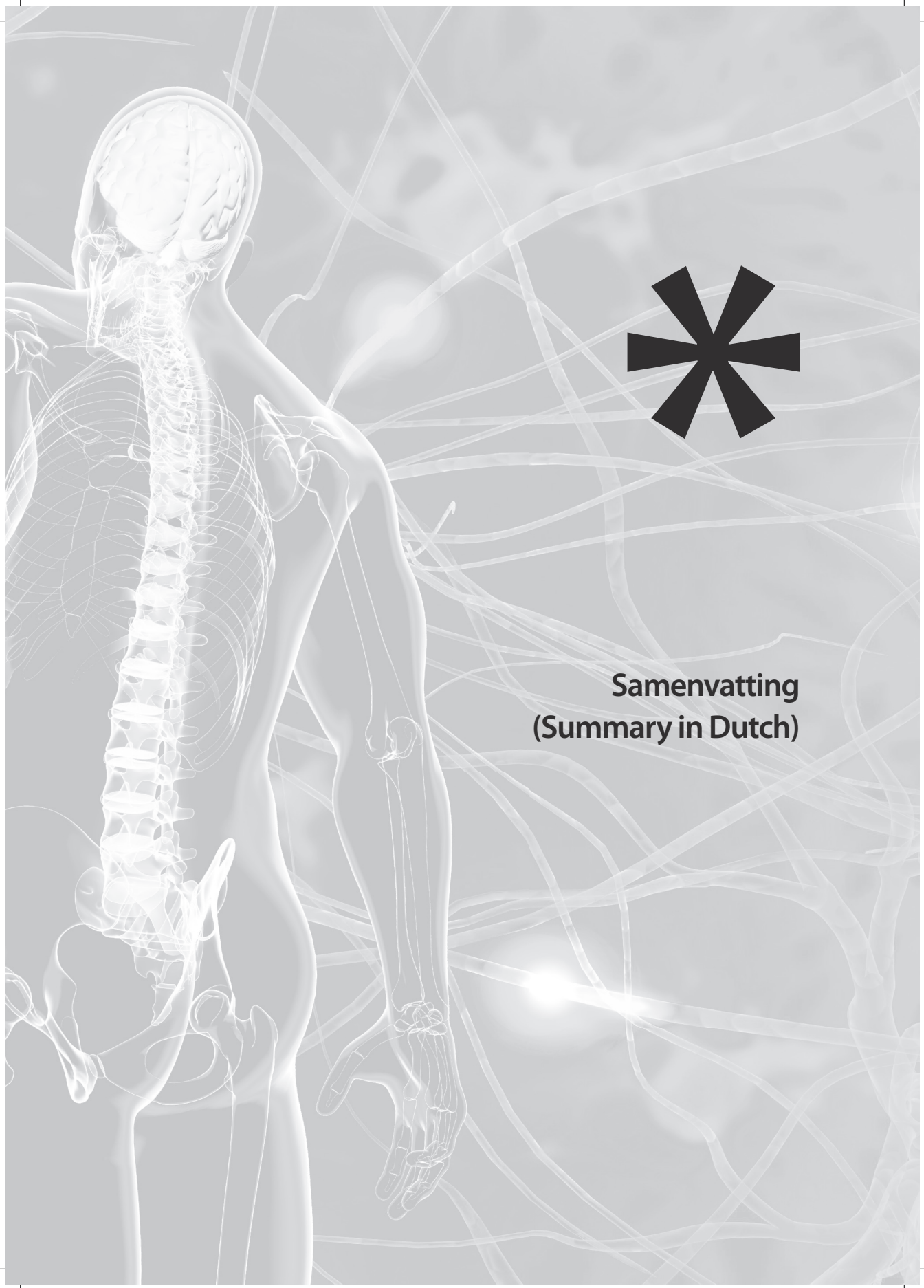
Chapter 4 describes the RCT with 31 persons randomised in the intervention group and 30 persons randomised in the waiting list control group. Measurements were performed before intervention, at 3 months (immediately after the intervention), and at 6 months (3 months' follow-up). The short-term results of the CONECSI trial were a decrease of anxiety and an increase of participation in activities. Besides this, a significant decrease in pain intensity and pain-related disability was found over time, but without a significant difference between intervention and waiting list control group. No differences were found for depression and life satisfaction. The results of this RCT imply that a cognitive behavioural programme might have beneficial effects on persons with chronic neuropathic SCI pain.

In **Chapter 5** the long-term effects of the cognitive behavioural treatment programme were explored. Because of the time frame of the study, only data from the intervention group was available for analysis. In the long-term, a significant decrease of pain intensity, pain-related disability, and anxiety, and a significant increase in participation in activities was found. Although the scores changed in the expected direction, no significant change over time was found for depression and life satisfaction. This is in line with the previously reported short-term results in **Chapter 4**. This exploratory study suggests that a cognitive behavioural programme might have lasting improvements on pain intensity, pain-related disability, anxiety, and participation in activities in persons with chronic neuropathic SCI pain and highlights the potential of such programmes.

Pain coping and pain cognitions are known to be associated with the experience of pain, but they have not been studied in the context of a cognitive behavioural treatment programme for coping with neuropathic pain after SCI. **Chapter 6** explores the associations of pain coping strategies and pain cognitions with pain intensity and pain-related disability. Also

the associations of changes in pain coping strategies and pain cognitions with changes in pain intensity and pain-related disability were examined. Participation in the programme was associated with increase in the use of the coping strategy pain transformation, more active coping, and a decrease in worrying, and also with a decrease in the pain cognitions catastrophising and reliance on health care, and an increase in optimism. More use at baseline of the coping strategies retreating, worrying, resting, and more passive coping, as well as more catastrophising were associated with higher baseline pain intensity. More use at baseline of the coping strategies retreating and resting, more passive coping, as well as more catastrophising and more perceived restrictions were associated with higher baseline pain-related disability. Persons who were more reliant on health care at baseline showed more decrease in pain intensity and pain-related disability. A decrease of catastrophising and restriction cognitions was associated with a decrease of pain-related disability. Although further research is needed, these results support the notion that it is possible to modify dysfunctional pain coping strategies and pain cognitions by a cognitive behavioural intervention, with some beneficial effects of this approach on pain intensity and pain-related disability.

Finally, **Chapter 7**, the general discussion, summarises the main findings of this thesis and discusses the results in the light of existing evidence for the effectiveness of cognitive behavioural treatment in (neuropathic) SCI pain. There are some longitudinal studies of similar cognitive behavioural programmes in SCI pain available, but these have important limitations, like small sample sizes and no randomisation. This chapter ends with methodological considerations, clinical implications, and recommendations for future research. Based on the results presented in this thesis it has been concluded that our studies enhance the level of evidence on the effectiveness of cognitive behavioural treatment for chronic neuropathic SCI pain, although more research is needed to evaluate this programme.



Samenvatting (Summary in Dutch)

Hoofdstuk 1, de algemene inleiding van dit proefschrift, beschrijft de context van het onderzoek, de onderzoeksvragen en de indeling van het proefschrift. Mensen met een dwarslaesie hebben vaak ernstige secundaire gezondheidsproblemen, waaronder neuropathische pijn. Medicamenteuze behandelingen van neuropathische pijn geven vaak onvoldoende verlichting en om die reden groeit de belangstelling voor niet-medicamenteuze behandelingen van neuropathische pijn. Eén van de niet-medicamenteuze behandelopties is een cognitief gedragstherapeutische behandeling. Ondanks dat dit type behandeling vaak wordt toegepast en effectief is bevonden bij andere vormen van chronische pijn, is het nog niet systematisch onderzocht bij mensen die last hebben van chronische neuropathische pijn na een dwarslaesie. Het algemene doel van dit proefschrift is om behandelingen voor pijn na een dwarslaesie en de effectiviteit daarvan te beschrijven, met name gericht op een cognitief gedragstherapeutisch behandelprogramma voor chronische neuropathische pijn na een dwarslaesie.

Hoofdstuk 2 geeft een overzicht van medicamenteuze en niet-medicamenteuze behandelingen die gebruikt worden bij chronische pijn na een dwarslaesie en beschrijft de ervaren effectiviteit daarvan, onderzocht in een cross-sectionele studie in een grote groep mensen met een dwarslaesie. De meerderheid van de deelnemers (77,1%) had chronische pijn ten gevolge van de dwarslaesie en had meer dan één pijntype (62,8%), waarbij neuropathische pijn het meest werd gerapporteerd (69,3%). Mensen met chronische pijn na een dwarslaesie hebben vaak veel pijnbehandelingen geprobeerd. Ondanks dat 63,8% van deze groep op het moment van het onderzoek enige vorm van pijnbehandeling had, rapporteerden zij toch veel pijn. Neuropathische pijn werd beoordeeld als het meest ernstige pijntype. Massage(therapie)/ ontspanning(soefeningen), anti-epileptica en niet-steroïdale anti-inflammatoire geneesmiddelen waren de meest gebruikte pijnbehandelingen. De op dat moment gebruikte behandelingen die het vaakst effectief werden bevonden, waren acupunctuur/ magnetiseren, cannabis/ alcohol, fysiotherapie en lichaamsbeweging, en massage(therapie)/ ontspanning(soefeningen). Transcutane Elektrische Neuro Stimulatie/ ultrageluid werden het minst vaak effectief bevonden. Psychologische behandeling, zoals cognitief gedragstherapeutische behandeling, werd wel effectief bevonden in het verlichten van pijn na dwarslaesie, maar werd zelden ingezet. Geconcludeerd wordt dat verder onderzoek nodig is om effectieve behandelingen voor pijn na een dwarslaesie vast te stellen. In het bijzonder is er meer onderzoek gewenst naar niet-medicamenteuze behandelingen.

Gerandomiseerde, gecontroleerde studies (RCT's) naar de effectiviteit van cognitief gedragstherapeutische interventies om neuropathische pijn na een dwarslaesie te verlichten, ontbreken. Om deze reden is de CONECSI (COping with NEuropathiC Spinal cord Injury

pain) trial opgezet. **Hoofdstuk 3** beschrijft de inhoud van het ontwikkelde cognitief gedragstherapeutische programma voor chronische neuropathische pijn na een dwarslaesie en het protocol van de CONECSI trial om het effect van dit programma op pijnintensiteit en pijngerelateerde beperkingen en secundair op stemming, participatie in activiteiten en tevredenheid met het eigen leven te evalueren. Daarnaast werden ook pijncopingstrategieën en pijncognities gemeten. Mensen met chronische neuropathische pijn na een dwarslaesie werden geworven in 4 revalidatiecentra en gerandomiseerd in een interventiegroep of een wachtlijst-controlegroep. De controlegroep kon het programma 6 maanden na de interventiegroep alsnog volgen. Metingen werden verricht bij aanvang van het programma voor de interventiegroep of bij aanvang van de wachttijd voor de controlegroep, en na 3, 6, 9 en 12 maanden. De interventie bestond uit educatieve, cognitieve en gedragstherapeutische elementen en omvatte 11 bijeenkomsten van 3 uur gedurende een periode van 3 maanden. Cognitief gedragstherapeutische behandeling richt zich op het aanpassen van irrationele gedachten, disfunctionele pijncognities en pijncoping. Het BioPsychoSociale model werd gebruikt voor educatie over de relatie tussen biologische, psychologische, sociale factoren en pijn en het belang van het behoud van evenwicht tussen belasting en belastbaarheid. Het 'Activating event-Belief-Consequence' model werd gebruikt voor cognitieve herstructurering om beter om te kunnen gaan met neuropathische pijn. De twee modellen werden toegelicht in educatieve sessies en begeleide groepsdiscussies met gebruik van fictieve casussen, en werden toegepast in sportworkshops en huiswerkopdrachten.

Hoofdstuk 4 beschrijft de RCT. Er werden 31 personen gerandomiseerd in de interventiegroep en 30 personen in de wachtlijst-controlegroep. De metingen vonden plaats voorafgaand aan de interventie, op 3 maanden (direct na de interventie) en na 6 maanden (3 maanden follow-up). Deze studie liet zien dat de interventie leidde tot een significante afname van angst en toename van participatie in activiteiten ten opzichte van de controlegroep. Daarnaast werd er een significante afname van pijnintensiteit en pijngerelateerde beperkingen gevonden over de tijd, maar zonder een significant verschil tussen de interventiegroep en de wachtlijst-controlegroep. Er werden geen verschillen gevonden voor depressie en tevredenheid met het eigen leven. De resultaten van deze RCT impliceren dat een cognitief gedragstherapeutisch programma gunstige effecten kan hebben op de ervaring van chronische neuropathische pijn na een dwarslaesie.

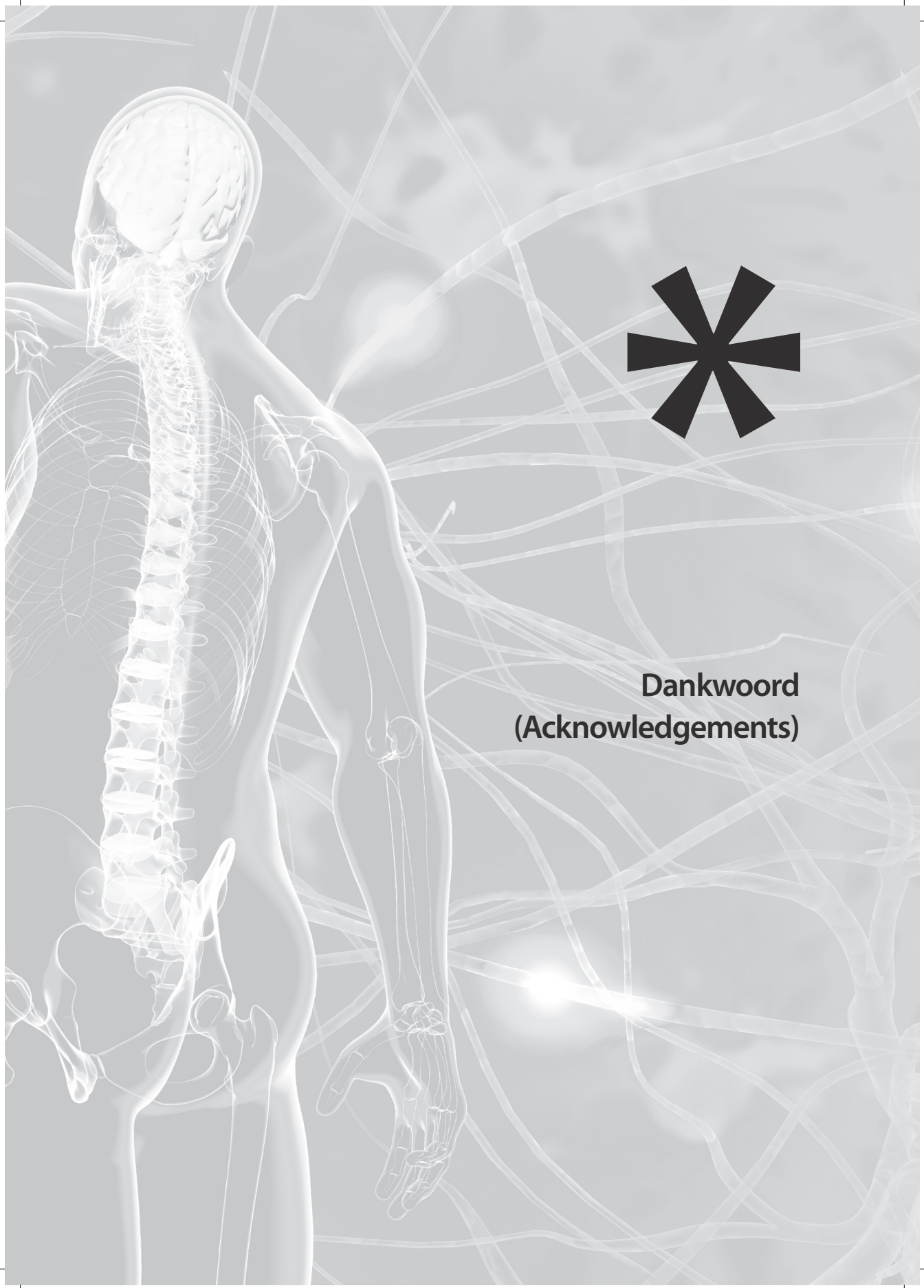
In **Hoofdstuk 5** worden de langetermijneffecten van het cognitief gedragstherapeutische behandelprogramma onderzocht. Vanwege de beperkte duur van de studie waren hiervoor alleen de gegevens van de interventiegroep beschikbaar. Op de lange termijn werd een significante afname van pijnintensiteit, pijngerelateerde beperkingen en angst, en een

significante toename van participatie in activiteiten gevonden. Hoewel de scores van depressie en tevredenheid met het eigen leven in de verwachte richting veranderden, werd er geen significante verandering over de tijd gevonden voor deze variabelen. Dit is in lijn met de eerder gerapporteerde kortetermijnresultaten in **Hoofdstuk 4**. Deze verkennende studie suggereert dat een cognitief gedragstherapeutisch behandelprogramma duurzame verbeteringen op pijnintensiteit, pijngerelateerde beperkingen, angst en participatie in activiteiten kan hebben bij mensen met chronische neuropathische pijn na een dwarslaesie en benadrukt het potentieel van dergelijke programma's.

Van pijncoping en pijncognities is bekend dat zij samenhangen met de pijnervaring, maar ze zijn niet onderzocht in het kader van een cognitief gedragstherapeutisch behandelprogramma voor het omgaan met neuropathische pijn na een dwarslaesie. **Hoofdstuk 6** bekijkt de associaties van pijncopingstrategieën en pijncognities met pijnintensiteit en pijngerelateerde beperkingen. Daarnaast werden de associaties van veranderingen in pijncopingstrategieën en pijncognities met veranderingen in pijnintensiteit en pijngerelateerde beperkingen bekeken. Deelname aan het programma was geassocieerd met een toename in het gebruik van de copingstrategie pijn transformeren, meer actieve coping, en een afname van piekeren, en ook met een afname in de pijncognities catastroferen en vertrouwen op de gezondheidszorg, en een toename van optimisme. Meer gebruik op baseline van de copingstrategieën zich terugtrekken, piekeren en rust nemen, meer passieve coping, evenals meer catastroferen waren geassocieerd met een hogere baseline pijnintensiteit. Meer gebruik op baseline van de copingstrategieën zich terugtrekken en rust nemen, meer passieve coping, evenals meer catastroferen en ervaren beperkingen waren geassocieerd met een hogere baseline score op pijngerelateerde beperkingen. Personen die op baseline meer vertrouwden op de gezondheidszorg lieten meer afname in pijnintensiteit en pijngerelateerde beperkingen zien. Een afname in catastroferende en beperkende cognities was geassocieerd met een afname van pijngerelateerde beperkingen. Hoewel verder onderzoek nodig is, ondersteunen deze resultaten de mogelijkheid om disfunctionele pijncopingstrategieën en pijncognities te veranderen middels een cognitief gedragstherapeutische interventie, met een aantal gunstige effecten van deze aanpak op pijnintensiteit en pijngerelateerde beperkingen.

Tot slot vat **Hoofdstuk 7**, de algemene discussie, de belangrijkste bevindingen van dit proefschrift samen en bespreekt het de resultaten in het licht van het bestaande bewijs voor de effectiviteit van cognitief gedragstherapeutische behandeling van (neuropathische) pijn na een dwarslaesie. Er is een aantal longitudinale studies gedaan naar gelijkwaardige cognitief gedragstherapeutische programma's voor pijn na een dwarslaesie. Deze studies hebben echter belangrijke beperkingen, zoals kleine steekproeven en geen randomisatie. Dit hoofdstuk

eindigt met methodologische overwegingen, klinische implicaties en aanbevelingen voor toekomstig onderzoek. Op basis van de resultaten beschreven in dit proefschrift wordt geconcludeerd dat onze studies het niveau van bewijs voor cognitief gedragstherapeutische behandeling van chronische neuropathische pijn na een dwarslaesie verhogen, hoewel meer onderzoek nodig is om dit programma te evalueren.



Dankwoord (Acknowledgements)

In de afgelopen jaren hebben veel mensen direct of indirect een bijdrage geleverd aan de totstandkoming van dit proefschrift. Daarom wil ik hier van de gelegenheid gebruik maken om iedereen te bedanken.

Op de eerste plaats wil ik alle mensen bedanken die vrijwillig hebben deelgenomen aan dit onderzoek. Veel dank voor jullie tijd en inzet, zonder jullie medewerking was er geen onderzoek mogelijk geweest!

Prof.dr. Post, beste Marcel, veel dank gaat uit naar jou, want zonder jou was dit proefschrift er niet gekomen. Jij, als initiator van dit onderzoek, hebt een cruciale rol gespeeld in mijn promotietraject. Je bent een onderzoeker in hart en nieren, van wie ik heel veel geleerd heb. Zeer terecht dat je onlangs bent benoemd tot hoogleraar, de eerste leerstoel dwarslaesierevalidatie! Met vragen over het onderzoek kon ik altijd bij je terecht en je was altijd bereid om me te helpen (of me zelfs letterlijk aan de hand te nemen bij het leren schaatsen op noren!). Dank voor je vertrouwen en je begeleiding.

Prof.dr. Lindeman, beste Eline, ik heb de eerste jaren dat je betrokken was bij mijn onderzoek niet gedacht dat je de afronding van mijn promotie niet mee zou maken. Als het maar enigszins ging, was je aan het werk. Je laatste woorden in ons telefonisch overleg waren dat we samen met Marcel het laatste deel van mijn promotie tot een goed einde gingen brengen, niet wetende dat het daarna snel bergafwaarts zou gaan met je gezondheid en ik je nooit meer zou spreken. Bedankt voor je begeleiding en je steun. Hoe slecht het ook met je eigen gezondheid ging, je informeerde altijd hoe het met mij was. Dat heb ik zeer gewaardeerd. Je bent zeker in mijn gedachten als ik in het Academiegebouw sta.

Prof.dr. Visser-Meily, beste Anne, ik weet nog dat ik jou in 2005 een e-mail stuurde om je succes te wensen met je promotie, niet wetende dat je later mijn promotor zou worden en ik jouw eerste promovendus. Fijn dat je zo laat in het traject in wilde springen, toen ook de vervanger van Eline, professor Wagenaar, overleed. Dank voor je begeleiding tijdens het laatste stuk van mijn promotietraject.

Dank aan de leden van de beoordelingscommissie, prof.dr. Öner, prof.dr. Bensing, prof. dr. van den Berg, prof.dr. Evers, prof.dr. Verbunt, voor het lezen en beoordelen van mijn proefschrift.

Andere personen die een belangrijke rol hebben gespeeld in de totstandkoming van dit proefschrift:

Veel dank gaat uit naar de directies van de deelnemende centra die de uitvoering van dit onderzoek mogelijk hebben gemaakt: De Hoogstraat Revalidatie te Utrecht, Revalidatie-

centrum Het Roessingh te Enschede, Adelante Zorggroep te Hoensbroek en Rijndam Revalidatiecentrum te Rotterdam. Steven, veel dank dat je me de kans hebt gegeven om te promoveren en te werken als onderzoeker bij Het Kenniscentrum.

Alle mensen van de deelnemende revalidatiecentra, die hun medewerking aan dit onderzoek hebben verleend, wil ik hiervoor hartelijk danken. Met name de revalidatieartsen, Floris, Catja, Govert, Anand, Helma, Nicole en Dorien, dank voor jullie ingezet voor de uitvoering van de CONECSI trial. De cursusleiders, Lilian, Peter, Wim, Marijke, Conny, Sandra, Hilde, Nicole, Lonneke en Jolante, hartelijk dank voor de fijne samenwerking, jullie inzet, betrokkenheid en inbreng bij dit onderzoek en het geven van de interventie aan twee groepen in jullie revalidatiecentrum. Peter en Lilian, ook hartelijk dank voor jullie werkzaamheden in de opzet van het programma, het cursusmateriaal en het onderzoek. Voormalig teammanagers van De Hoogstraat, Hans en Floris, dank voor jullie medewerking. En ook Dwarslaesie Organisatie Nederland wil ik bedanken voor het meedenken in de opzet van de CONECSI trial.

Zonder subsidie hadden de studies, die beschreven staan in dit proefschrift, niet uitgevoerd kunnen worden. Ik dank Stichting Wetenschappelijk Fonds De Hoogstraat en Pfizer voor het financieren van deze studies en De Hoogstraat Revalidatie, Stichting Wetenschappelijk Fonds De Hoogstraat en Adelante Zorggroep voor de financiële steun ten behoeve van het drukken van dit proefschrift. Verder wil ik de mensen van Pfizer, die betrokken waren bij de CONECSI trial, bedanken voor de samenwerking, de interessante werkconferenties van DALI voor PIJN, de site-visits en de coaches Gerbig, Bas en Ruud voor de begeleiding.

Roel, bedankt dat je samen met mij alle data van de CONECSI trial hebt ingevoerd. Christel, bedankt voor je hulp bij mijn statistiekvragen en de leuke congressen in binnen- en buitenland die we samen hebben bezocht. Renate, bedankt voor de fijne samenwerking, je zorgvuldigheid, snelle reacties en het nauwkeurig verzorgen van de lay-out van mijn proefschrift. Mike, heel fijn dat jij de omslag van mijn proefschrift wilde maken, veel dank daarvoor.

Junioren, Hileen, Nienke, Marloes (de gezellige etentjes en surprise-avondjes met z'n vieren houden we er ook na mijn promotie in!), Anne, Martin, Maremka, Jacinthe, Annerieke en Teuni, bedankt voor de fijne samenwerking. Leuk dat in het laatste stadium van mijn promotietraject de twee promovendi-kamers samen werden gevoegd op de nieuwe locatie. Ik wens jullie veel succes met jullie onderzoeken en promoties (weet me ook na mijn promotie te vinden als jullie vragen hebben). Ook alle andere onderzoekers die mijn pad gekruist hebben, Carlijn, Merel, Anne-Marie, Maureen, Dirk-Wouter, Hanneke, Marjolijn, Rutger, Casper, Margot, Frederique, Ingrid, Mattijs, Jurre, Michiel, Astrid, Lotte, Goos en Patricia,

bedankt voor de fijne samenwerking. Andere collega's van Het Kenniscentrum, Marjolijn, Tanja, Carin, Christel, Sacha, Olaf, Carlijn, Andrie, Nienke, Mia, Imke, Anne-Marije en verder iedereen met wie ik heb samengewerkt op Het Kenniscentrum maar niet bij naam noem, zoals oud-collega's, arts-onderzoekers, onderzoeksassistenten en de vele stagiaires, dank voor de samenwerking en de fijne werkomgeving. Ik wil ook een aantal collega's van De Hoogstraat buiten Het Kenniscentrum bedanken: Mariska, Lenneke, Arjan, Hubert, Aling, Birgit en Anne, bedankt voor het leuke contact op en buiten de werkvloer en de interesse in mijn promotieonderzoek. Naast de werkgerelateerde bijeenkomsten heb ik veel goede herinneringen aan activiteiten die ik met jullie allemaal naast het werk heb gedaan, zoals de vele wandelingen door het Wilhelminapark in de pauze, de potjes tafeltennis, het lunchen buiten De Hoogstraat, de vrijdagmiddagborrels bij Primus en De Winter, de junioren-, Kenniscentrum- en Hoogstraat-uitjes, de sportdagen van Revalidatie Nederland en de Hoogstraat ski-dagen in Werfenweng.

Ook veel dank gaat uit naar familie en vrienden die de afgelopen jaren interesse hebben getoond in mijn onderzoek en promotie, en die voor veel leuke afleiding hebben gezorgd buiten werktijd.

Een aantal mensen wil ik in het bijzonder noemen: Chantal en Wonneke (we gaan snel na mijn promotie een keer de vlieger oplaten!), Yvonne en Bas, Sandy, Susan, Mike en Natacha, Carla en Mohamed, Jan en Sandra, Thomas en Patricia, Margreet, Marjolein en Ingrid, Sandra, bedankt voor jullie steun en voor de leuke dingen die we samen hebben ondernomen.

C-girls, bedankt voor de leuke etentjes, concertbezoekjes, optredens en stedentripjes, die we samen gedaan hebben gedaan. Speciale dank voor alle Cern-energie en dat ik alles met jullie heb kunnen delen!

Henk, jij hebt mij enthousiast gemaakt voor het doen van onderzoek. Je hebt me tijdens en na mijn opleiding veel geleerd en verder geholpen, veel dank daarvoor! Hopelijk volgen er in de toekomst nog veel gezellige etentjes en komt er een moment waarop we weer samen aan hetzelfde onderzoeksproject werken.

Maartje, Veronique en Angela, leuk dat we na onze stages op Het Leijpark contact hebben gehouden. We hebben met z'n vieren heel wat meegemaakt de afgelopen jaren, al dan niet gerelateerd aan high teas (of moet ik low teas schrijven?). Snel maar weer eens eentje plannen om te bewijzen dat high teas ook positieve gevolgen kunnen hebben.

Jesse, Mayke, Fleur en Sietske, we kennen elkaar al heel lang en fijn om te merken dat, ondanks dat onze levens wat meer uit elkaar zijn gaan lopen en we elkaar niet meer zoveel

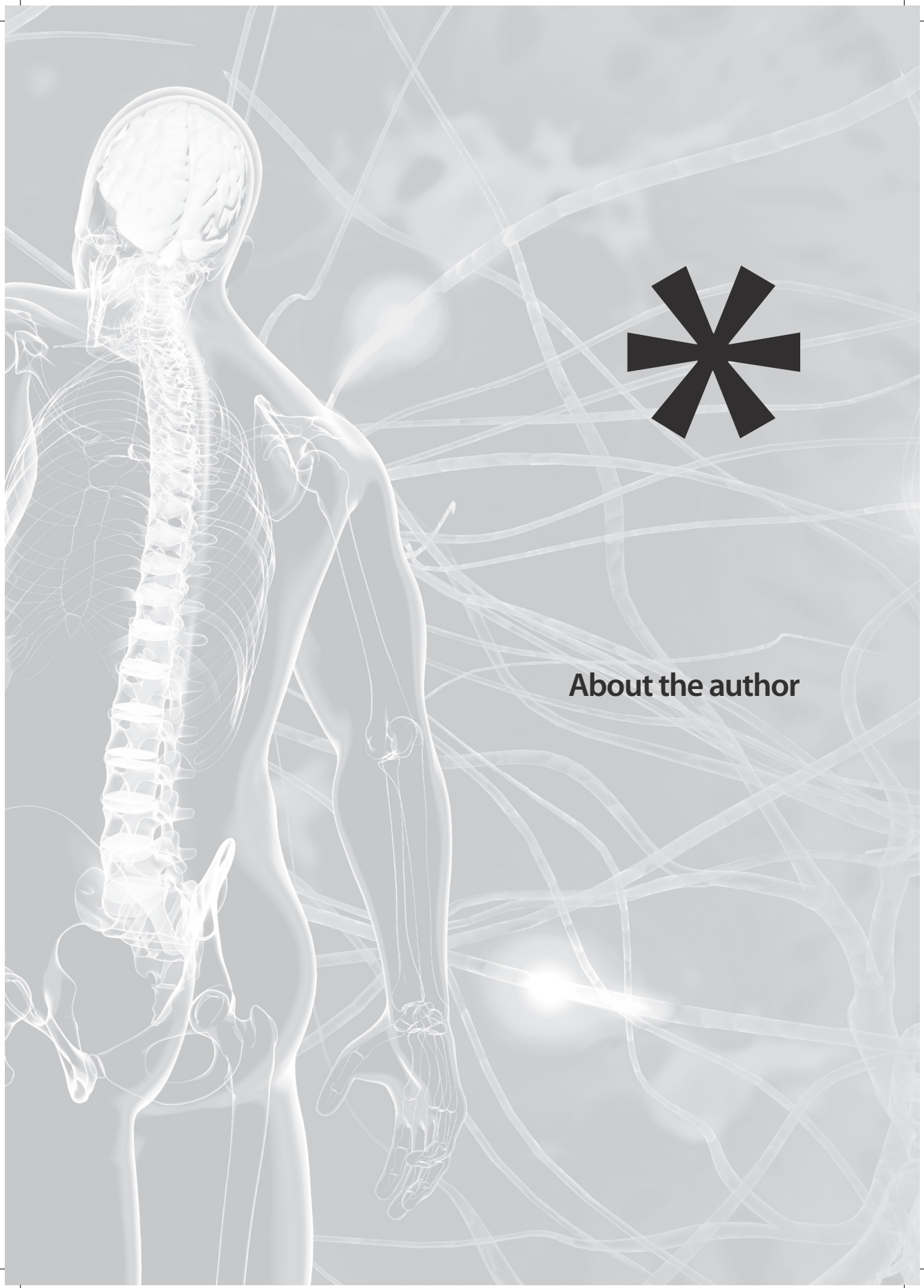
zien als vroeger, het altijd weer erg gezellig is als we elkaar zien. Jesse, speciale dank voor jou, voor de fijne gesprekken die ik met je heb gehad. Bedankt voor het luisteren en adviseren bij tegenslagen en het vieren van successen. Dat er nog maar veel drankjes bij de Verkadefabriek, op het terras tijdens Bossche activiteiten of bij elkaar thuis op de bank mogen volgen!

Rudy, bedankt voor je fijne vriendschap en voor het feit dat je altijd voor me klaar staat voor steun, afleiding of voor een luisterend oor. We zijn heel verschillend, maar vullen elkaar perfect aan. Op naar nog veel mooie dagen met onder andere strand-, theater- en concertenbezoeken, pannenkoeken en pindasoepen!

Paranimfen, we zijn ooit begonnen als collega's, maar jullie zijn in de loop van de jaren veel meer geworden dan dat. Heel fijn dat jullie tijdens de verdediging achter me staan! Bedankt voor de fijne sfeer die jullie altijd op de kamer brachten, ik heb met veel plezier met jullie samengewerkt. Bedankt ook voor het meelezen van delen van mijn proefschrift in het laatste stadium en het meedenken in de aanloop naar mijn promotie. Carlijn, heel leuk dat ik bij jouw promotie paranimf mocht zijn (kon ik de kunst alvast een beetje afkijken) en Hileen, dat ik die rol bij jou nog mag gaan vervullen. Bedankt voor jullie vriendschap, steun, en dat jullie altijd met me mee wilden denken en voor me klaar stonden in de afgelopen jaren.

Lieve Frank, Menion en Jacqueline, Hank en Joke, ik ben jullie heel dankbaar en ik ben blij dat jullie er voor me zijn! Papa en mama, een van de vele dingen die ik van jullie heb meegekregen is doorzettingsvermogen: hoeveel tegenslagen er ook op je pad komen, zet toch door! Zonder jullie was ik niet zover gekomen. Frank, bedankt voor al je hulp, steun, begrip en liefde. Ik ben er trots op dat we, toen de storm maar niet ging liggen, hebben leren dansen in de regen!

Allemaal bedankt!



About the author

Curriculum Vitae

Matagne Heutink werd geboren op 26 april 1977 te Boxtel. Na het behalen van haar gymnasiumdiploma aan het Jacob Roelandslyceum te Boxtel, is zij Psychologie gaan studeren aan de Universiteit van Tilburg (toenmalige Katholieke Universiteit Brabant). In 2004 heeft Matagne met genoegen haar doctoraaldiploma in zowel Klinische Neuropsychologie als Psychologie in de Gezondheidszorg behaald. Tijdens haar studie werkte zij onder andere als student-assistent bij de Universiteit van Tilburg en als psychologisch medewerker, projectmedewerker cognitieve revalidatie en projectmedewerker wetenschappelijk onderzoek bij Revalidatiecentrum Het Leijpark in Tilburg.

Na haar afstuderen bleef Matagne tot halverwege 2006 werkzaam als onderzoeker bij Revalidatiecentrum Het Leijpark in Tilburg binnen het onderzoek naar de effectiviteit van het behandelprogramma 'Vroege Intensieve Neurorevalidatie (VIN) van kinderen en jongeren in een vegetatieve of laagbewuste toestand na ernstig hersenletsel'. Van 2006 tot 2008 werkte zij als cognitief trainer en psychologisch medewerker bij Revalidatiecentrum Blixembosch in Eindhoven en als neuropsycholoog en neurofeedbacktherapeut bij Praktijk Neuropsychologie in Waalre. Van 2002 tot met 2009 was Matagne penningmeester in het bestuur van de Sectie Revalidatie van het Nederlands Instituut van Psychologen (NIP). Ze is sinds 2008 geregistreerd als PSYCHOLOOG NIP.

In 2008 is Matagne aangesteld als onderzoeker en halverwege 2010 als promovendus bij Kenniscentrum Revalidatiegeneeskunde De Hoogstraat in Utrecht en Universitair Medisch Centrum Utrecht. Vanaf juni 2011 heeft zij in deeltijd aan dit promotieonderzoek gewerkt en daarnaast heeft zij als onderzoeker gewerkt bij de GGZ Oost Brabant te Boekel binnen het project 'Haalbaarheid van single-case onderzoek naar het effect van medicamenteuze behandeling van gedragsmatige en emotionele gevolgen van ernstig niet-aangeboren hersenletsel' en bij De Hoogstraat Revalidatie te Utrecht binnen het project 'Arbeidsparticipatie en arbeidssatisfactie van mensen met een dwarslaesie in Nederland'.

Publications

Heutink M, Post MW, Luthart P, Schuitemaker M, Slangen S, Sweers J, Vlemmix L, Lindeman E. Long-term outcomes of a multidisciplinary cognitive behavioural programme for coping with chronic neuropathic spinal cord injury pain. J Rehabil Med 2014;46:540-545.

Heutink M, Post MW, Overdulve CW, Pfennings LE, van de Vis W, Vrijens NL, Lindeman E. Which pain coping strategies and cognitions are associated with outcomes of a cognitive behavioral intervention for neuropathic pain after spinal cord injury? Top Spinal Cord Inj Rehabil 2013;19:330-340.

Post MW, **Heutink M**. Arbeidsparticipatie en arbeidssatisfactie van mensen met een dwarslaesie in Nederland in 2012 - Resultaten van het Nederlandse deel van de studie International Labour market Integration Assessment in Spinal cord injury (ILIAS-NL). Utrecht: Kenniscentrum Revalidatiegeneeskunde Utrecht, Hersencentrum, Universitair Medisch Centrum Utrecht en De Hoogstraat Revalidatie; 2013 (in Dutch).

Heutink M, Post MW, Bongers-Janssen HM, Dijkstra CA, Snoek GJ, Spijkerman DC, Lindeman E. The CONECSI trial: results of a randomized controlled trial of a multidisciplinary cognitive behavioral program for coping with chronic neuropathic pain after spinal cord injury. Pain 2012;153:120-128.

Heutink M, Post MW, Bongers-Janssen HM, Dijkstra CA, Snoek GJ, Spijkerman DC, Lindeman E. The CONECSI trial: a randomized controlled trial of a multidisciplinary cognitive behavioral program for coping with chronic neuropathic spinal cord injury pain. Abstract in Top Spinal Cord Inj Rehabil 2011;16:29-30.

Heutink M, Post MW, Wollaars MM, van Asbeck FW. Chronic spinal cord injury pain: pharmacological and non-pharmacological treatments and treatment effectiveness. Disabil Rehabil 2011;33:433-440.

Heutink M, Post MW, Luthart P, Pfennings LE, Dijkstra CA, Lindeman E. A multidisciplinary cognitive behavioural programme for coping with chronic neuropathic pain following spinal cord injury: the protocol of the CONECSI trial. BMC Neurol 2010;10:96.

Heutink M, Eilander HJ, de Kort PL, Schouten EJ, van Heugten CM. Langdurig bewustzijnverlies na ernstig traumatisch hersenletsel bij kinderen en jongeren in Nederland: incidentie, Vroege Intensieve Neurorevalidatie en langetermijnfunctioneren. Tijdschrift voor Neuropsychologie 2007;2:10-19 (in Dutch).

Wijnen VJ, **Heutink M**, van Boxtel GJ, Eilander HJ, de Gelder B. Autonomic reactivity to sensory stimulation is related to consciousness level after severe traumatic brain injury. Clin Neurophysiol 2006;117:1794-1807.

Eilander HJ, Wijnen VJ, **Heutink M**. Wetenschappelijk eindrapport "Vroege Intensieve Neurorevalidatie (VIN) van kinderen en jongeren in een vegetatieve of laagbewuste toestand na ernstig hersenletsel". Tilburg: Revalidatiecentrum Leijpark; 2005 (in Dutch).

Wijnen V, **Heutink M**, Schuttelaars Y, Eilander H, van Boxtel G, de Gelder B. Changes in autonomic reactivity related to recovery to consciousness after severe acquired brain injury. Abstract in J Psychophysiol 2003;17:141.

Conferences and presentations

Heutink M, Post MW Resultaten CONECSI: RCT van een multidisciplinaire cognitieve gedragsinterventie voor het omgaan met chronische neuropathische pijn na een dwarslaesie. Presentation, De Hoogstraat Rehabilitation. The Netherlands, Utrecht, March 28, 2013 (in Dutch).

Eilander H, ter Mors B, Franssen F, **Heutink M**, van Heugen C, Ponds R. Feasibility of single-case study designs to evaluate neuropsychiatric treatment of behaviour disorders after severe acquired brain injury. Poster presentation, The 9th Conference of the Neuropsychological Rehabilitation. Norway, Bergen, July 2-3, 2012.

Heutink M, Post MW The CONECSI trial: an RCT of a multidisciplinary cognitive behavioral program for coping with chronic neuropathic SCI pain. Presentation, 22th ZonMw minisymposium on 'Physical strain, work capacity and mechanisms of restoration of mobility in the rehabilitation of individuals with spinal cord injury'. The Netherlands, Hoensbroek, September 23, 2011.

Heutink M, Post MW, Bongers-Janssen HM, Dijkstra CA, Snoek GJ, Spijkerman DC, Lindeman E. The CONECSI trial: a randomized controlled trial of a multidisciplinary cognitive behavioral program for coping with chronic neuropathic spinal cord injury pain. Presentation, The International Conference on Spinal Cord Medicine and Rehabilitation of the The International Spinal Cord Society (ISCoS) and The American Spinal Injury Association (ASIA), The State of the Science in SCI Rehabilitation. USA, Washington DC, June 4-8, 2011.

Post MW, **Heutink M**, Bongers-Janssen HM, Snoek GJ, Spijkerman DC, Dijkstra CA, Lindeman E. The CONECSI trial: a randomized controlled trial of a multidisciplinary cognitive behavioral program for coping with chronic neuropathic spinal cord injury pain. Presentation, The 4th European Meeting of the European Spinal Psychologists' Association (ESPA), Psychological Treatment of People with Spinal Cord Injury. Spain, Barcelona, May 5-6, 2011.

Heutink M, Post MW Multidisciplinaire cognitieve gedragsinterventie voor het omgaan met chronische neuropathische pijn na een dwarslaesie. Presentation, Nederlands-Vlaams Dwarslaesie Genootschap (NVDG), werkgroep psychologen. The Netherlands, Rotterdam, March 10, 2011 (in Dutch).

Heutink M, Post MW, Luthart P, Pfennings LE, Dijkstra CA, Lindeman E. Evaluation of a multidisciplinary cognitive behavioural programme for coping with chronic neuropathic spinal cord injury pain. Poster presentation, The RMI Symposium Brain Connections, Genes and Disease. The Netherlands, Utrecht, October 27, 2010.

Heutink M, Post MW, Luthart P, Pfennings LE, Dijkstra CA, Lindeman E. Evaluation of a multidisciplinary cognitive behavioural programme for coping with chronic neuropathic spinal cord injury pain. Poster presentation, The Neuroscience & Cognition Utrecht Research Day. The Netherlands, Utrecht, September 3, 2010.

Heutink M, Post MW. Multidisciplinaire gedragsinterventie voor het omgaan met chronische neuropathische pijn na een dwarslaesie. Presentation, 17th ZonMw minisymposium on 'Physical strain, work capacity and mechanisms of restoration of mobility in the rehabilitation of individuals with spinal cord injury'. The Netherlands, Utrecht, September 11, 2009 (in Dutch).

Heutink M, Post MW. Multidisciplinaire gedragsinterventie voor het omgaan met chronische neuropathische pijn na een dwarslaesie. Presentation, Nederlands-Vlaams Dwarslaesie Genootschap (NVDG), werkgroep psychologen. The Netherlands, Rotterdam, September 10, 2009 (in Dutch).

Heutink M, Post MW. Multidisciplinary behavioural programme for coping with chronic neuropathic pain following SCI: an ongoing randomized clinical trial in The Netherlands. Presentation, The 3rd European Meeting of the European Spinal Psychologists' Association (ESPA), Psychological Treatment of People with Spinal Cord Injury. Switzerland, Nottwil, April 23-24, 2009.

Eilander HJ, Wijnen VJ, **Heutink M**. Recovery from a prolonged unconscious state in severe brain injured young patients after early intensive neurorehabilitation. Presentation, The 8th International Neurotrauma Symposium. The Netherlands, Rotterdam, May 21-25, 2006.

Wijnen VJ, **Heutink M**, van Boxtel GJ, Eilander HJ, de Gelder B. Autonomic activity is linearly related to recovery from severe traumatic brain injury in adolescents. Presentation, The IV International Symposium on Coma and Death. Cuba, Havana, March 9-12, 2004.