Cervico-cephalalgiaphobia: a subtype of phobia in patients with cervicogenic headache and neck pain? A pilot study

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Objectives: The term ‘cephalalgiaphobia’ was introduced in the mid-1980s and defined as fear of migraine (attacks). We hypothesized that a specific subtype of cephalalgiaphobia affects patients with cervicogenic headache (CEH). This study aimed to: (1) define the term ‘cervico-cephalalgiaphobia’; (2) develop a set of indicators for phobia relevant to patients with CEH; and (3) apply this set to a practice test in order to estimate the frequency of cervico-cephalalgiaphobia in the Dutch primary care practice of manual physical therapy.

Methods: A systematic approach was used to develop a definition and potential indicators for cervico-cephalalgiaphobia. An expert group appraised the definition and the set of indicators (score per indicator: never; sometimes; often/always). An invitation to participate in the practice test was sent to Dutch manual physical therapy practices ($n=56$) representing 134 manual physical therapists (MPTs). The cut-off point for percentages of scores for coverage of the indicators was set at $\geq 60\%$.

Results: The expert group agreed with the proposed definition of cervico-cephalalgiaphobia. A set of eight indicators for cervico-cephalalgiaphobia was selected from 10 initial indicators. Thirty-six MPTs provided data from 46 patients diagnosed with CEH. The coverage of ‘often/always’ was substantial for the indicators, ‘Short-term positive results in previous manual physical therapeutic treatment’, ‘Shorter interval between treatment sessions’, ‘Fear of “locked facet joints” of the neck’, ‘More frequent manipulation’, and ‘Fear of increase in headaches’. Coverage was also substantial for ‘never’ regarding ‘Long-term positive results in previous manual physical therapeutic treatment’. ‘Confirmation of “locked facet joints” of the cervical spine by MPT as a cause for increase of CEH’ scored ‘often/always’ in all patients. Coverage for ‘Increased use of medication with insufficient effect’ was substantial, scoring as ‘sometimes’ in 39 (84.8%) patients.

Discussion: Cervico-cephalalgiaphobia was defined and a set of eight indicators formulated based on the literature and clinical expertise. The practice test provides valuable information on the frequency of indicators for cervico-cephalalgiaphobia in the Dutch manual physical therapy practice, suggesting that cervico-cephalalgiaphobia is common in patients with CEH.

Keywords: Cervicogenic headache, Neck pain, Phobia, Chronic pain, Indicators, Manual physical therapy

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Introduction

The term ‘cephalgiaphobia’ is derived from Greek cephalic (head), algia (pain) and phobia (a persistent, irrational fear of a specific object, activity, or situation that leads to a compelling desire to avoid it),¹ and refers to a fear of headache or fear of the next headache (attack), analogous to kinesiophobia (fear of movement), claustrophobia (fear of closed spaces) and agoraphobia (fear of open spaces). The term was originally introduced by Dr. Harvey Featherstone in the mid-1980s.² In 2008, a study reviewed relevant articles published on specific phobias during the previous 2 years, including two officially recognised specific phobias (fear of spiders and blood, injection/injury) and two ‘unofficial’ specific phobias (fever phobia and cephalgiaphobia).³ Peres *et al.* (headache clinic in Brazil)⁴ used the term ‘cephalgiaphobia’ to describe 12 migraine patients with phobic-avoidant behaviour (overuse of medication) related to their headache attacks, classifying the behaviour as a possible specific illness phobia. In a recent pilot study by Giannini *et al.* (headache clinic in Italy),⁵ moderate–high migraine frequency was associated with higher risk of cephalgiaphobia. Chronic migraineurs with medication overuse scored higher for cephalgiaphobia than those without medication overuse. The authors concluded that cephalgiaphobia may represent a high-frequency feature and might play a role in chronicization of migraine.

Giannini *et al.*⁵ provided the following definition: ‘Cephalgiaphobia is the fear of having a headache attack during a pain-free period that may induce patients to use analgesic in the absence of pain to prevent headache and to improve their performances’. Based on 30-years’ experience (RABO) of treating patients for cervicogenic headache (CEH), dizziness (CED) and tinnitus (CET),^{6–8} many practical lessons were learned in the assessment and treatment of patients; for instance: (1) many patients exhibit fear of cervical mobility impairment (locked facet joints) during a period of mild headache; and (2) patients’ attitudes and beliefs about headache, dizziness and tinnitus are often derived from those of the manual physical therapist (MPT). This reciprocal association can be mutually reinforcing, as is the case for CEH. The respective behaviours of patients and MPTs may increase rather than reduce headache and MPTs disability, similar to reported effects in patients with lower back pain.^{9–13} To date, cephalgiaphobia has not been described or studied in patients with CEH. Therefore, the question arises as to whether a comparable phobia, termed ‘cervico-cephalgiaphobia’, is present in patients with CEH. Recognition of the existence and understanding the indicators of cervico-cephalgiaphobia could be relevant to the assessments made by MPTs when indicating treatment for patients with CEH.

Although CEH is still controversial, there is now a tendency to accept CEH as a separate headache entity^{14–17} and nociception of the neuromusculoskeletal structures of the cervical region as a generator for headache.^{18,19} The

International Headache Society (IHS) classifies CEH as a secondary headache that has its nociceptive source in the neck and head.²⁰ Cervicogenic headache is typically perceived in one or more segmentally related areas of the neck, head and/or face (C1–C4), and of the supra-orbital nerve (VI). The prevalence of CEH in the general population ranges from 0.7 to 17.8%, depending on the diagnostic criteria.^{21–24} Using Dutch longitudinal physical therapy data ($n=9346$) from 2009, the prevalence of CEH was estimated to be 0.95%.²⁵

The proposed mechanisms underlying the development and progression of CEH are based on the convergence of nociceptive inputs of different cervical structures at the caudal part of the spinal tract nucleus of the trigeminal nerve.^{26–30} The assumption is that central sensitisation is driven by both bottom-up sensitisers (i.e. nociception and dysfunction of the upper cervical facet joints) and top-down sensitisers (i.e. fear of headache). It is reasonable to assume that both types of sensitisers need to be addressed during the diagnostic and therapeutic process of manual physical therapy. Several studies examining the underlying mechanisms of manual therapy have indicated that the effects are, at least in part, centrally mediated.^{31–33} Understanding these underlying mechanisms is the first step towards more effective treatments, one of which is manual physical therapy.^{34–36}

In the Netherlands, general practitioners (GPs) regularly refer patients with CEH and tension-type headache for manual physical therapy as the second treatment option after medication.^{37,38}

The focus of manual physical therapy in patients with CEH is treatment of musculoskeletal impairments, static posture (forward head position), restricted range of motion, dysfunction of the motion segments of the joints of the cervical spine and of the temporomandibular joint, palpably painful upper cervical joints, impaired muscle function (tone, endurance, tenderness and trigger points), asymmetry in skin-fold tenderness and cervical dyskinesia-sensory sense.³⁹ Systematic reviews of the effectiveness of spinal manipulative therapy in patients with CEH, based on only a few well-designed randomised controlled trials (RCTs), concluded that evidence is limited, results are mixed, and any effects are mostly short-term.^{36,40–44}

Evidence from systematic reviews of the rehabilitation and manual physical therapy of patients with chronic musculoskeletal pain indicates that the combination of psychological and biomedical factors are more important for treatment outcomes in terms of functional recovery than biomedical factors alone.^{45–48} Among these psychological factors, fear (including fear of movement, ‘kinesiophobia’) has repeatedly been identified as an important factor contributing to chronic pain.^{49–53} Fear of headache may also be such a factor in patients with CEH.

The aims of this study were: (1) to define the term ‘cervico-cephalgiaphobia’; (2) to develop a set of indicators for neck-headache type of phobia relevant to patients

with CEH; and (3) to put this set of indicators to the test in practices in order to estimate the frequency of cervico-cephalgiaphobia among CEH patients in primary care manual physical therapy.

Methods

Design

The study consisted of two phases: (1) definition of cervico-cephalgiaphobia and development of indicators of cervico-cephalgiaphobia; and (2) a cross-sectional study in manual physical therapy practice in which this set of indicators was tested. The Medical Ethics Committee of Radboud University Medical Centre Nijmegen, The Netherlands, stated that ethical approval was not necessary for the present study.

Phase 1: definition and development of indicators of cervico-cephalgiaphobia

Based on existing definitions for cephalgiaphobia,^{4,5} the following definition of cervico-cephalgiaphobia was formulated: 'Cervico-cephalgiaphobia is a persistent excessive or unreasonable fear of experiencing unilateral or unilaterally dominant headache in combination with neck pain associated with impairments of the cervical muscles and spinal joints and aggravated by head and neck movement or postures'.

Criteria for cervico-cephalgiaphobia were formulated on the basis of our combined clinical expertise and identified with the aid of the literature on phobia in relationship with musculoskeletal disorders.⁵⁴⁻⁶² Systematic reviews of the screening, assessment and management of patients with headache and neck pain,^{39,63-65} segmental innervation of the neck and head structures⁶⁶ and core sets for musculoskeletal disorders of the International Classification of Functioning, Disability and Health (ICF) were also consulted.⁶⁷ Criteria were extracted by two members of the research team (RABO and WD) and, where necessary, differences were discussed with a member of the project group until consensus was reached. The criteria for cervico-cephalgiaphobia were transformed into indicators by determining percentages of patients for whom an indicator was applicable (for example, the percentage of patients for whom 'the interval between treatment sessions had shortened'). In order to ensure content validity of the indicator set, a number of experts from different disciplines (manual physical therapy [$n=5$], psychology [$n=1$], neurology [$n=1$], quality of care [$n=2$] and implementation science [$n=1$]) were invited to participate, including authors of RCTs ($n=2$) and systematic reviews ($n=3$) in the field of manual physical therapy and headache, and clinical specialists with expertise in patients with headache ($n=5$) identified by the project group. None of the members of the expert group participated in the practice test. The definition and the indicator set for cervico-cephalgiaphobia were presented to this expert group for clinical relevance and content validity.

Phase 2: pilot practice test

Setting and participants

Based on 10 postcode areas in the southern part of the Netherlands, an invitation to participate in the study was sent to all primary care physiotherapy practices ($n=56$), of which 46 (82.1%) indicated willingness to participate (Fig. 1). MPTs from 39 practices (84.8%) participated in a regional information session that outlined the purpose and content of the study and the expected contribution. Each participating practice formally consented to participate. Study enrolment finally included 134 MPTs from 27 (69.2%) participating practices. These MPTs were asked to collect data on the characteristics of patients with headache meeting the criteria for CEH.^{36,68-71} Patients had to meet the following inclusion criteria: (1) aged between 18 and 50 years; (2) experiencing headaches for longer than 6 months; (3) unilateral or unilaterally dominant headache without side shift; (4) headache associated with ipsilateral neck, shoulder, or arm pain; (5) pain beginning in the neck; (6) headache aggravated by neck movement or postures; (7) pressure pain over the upper cervical/occipital region; (8) associated with restricted neck range of motion; (9) headache episodes of varying duration and (10) previously treated with manual physical therapy.

All patients were informed about the design and purpose of the study and they gave permission for anonymous use of data. Patients were excluded when their history, signs and symptoms suggested another type of headache or flagged a potential non-benign cause (including previous surgery of the neck), or when evidence was present for a specific pathologic condition such as, progressive headache, malignancy, neurologic disease, fracture, herniated disc or systemic rheumatic disease.

Data collection

Data were collected over a period of 6 months. The diagnostic criteria for CEH were established at the first appointment, and CEH was diagnosed according to the diagnostic criteria of Sjaastad *et al.*^{70,71} History taking formed the basis of the data collection.

Data analysis

The indicators for cervico-cephalgiaphobia were scored as 'never', 'sometimes', 'often/always'. Descriptive statistics were computed (mean, standard deviation, median and range [minimum and maximum]) and percentages calculated, with the number of times an indicator was met as the numerator and the number of patients assessed as the denominator. Based on our studies describing indicator adherence, the cut-off point for substantial coverage for every indicator was set at 60%.⁷²

The content validity was expressed as a ratio. Content validity ratio (CVR) values ranged between -1 (perfect disagreement) and +1 (perfect agreement).^{73,74} Content validity ratio values above 0 indicated that over half of panel members agreed on an item as 'essential' (<50%).

Figure 1 Flowchart of manual physical therapy practices' and participant manual physical therapists' (MPTs) responses and reasons for non-response and dropouts.

Results

Phase 1: indicator development

The expert group agreed with the proposed definition of cervico-cephalalgia phobia (CVR=1.0 [perfect agreement]). In total, 10 phobia-related criteria for patients with CEH in manual physical therapy practice were extracted from the literature. After evaluation by the expert group, this number was reduced to eight items.

Two items were excluded: fear of increased headache combined with dizziness and fear of unsteadiness and falling (disagreement). The eight remaining criteria showed CVRs of above 0 (>50% agreement), and four criteria showed a score of 1.0 (perfect agreement). The eight criteria were then phrased, applied as indicators and subsequently added as eight questions during history taking (Appendix).

Table 1 presents the set and the CVRs of the eight indicators for cervico-cephalgiaphobia in patients with CEH in manual physical therapy practice.

Phase 2: practice test

Response rates

Of the 134 MPTs, 64 (47.8%) screened their patient population for cases with CEH as defined by diagnostic criteria of Sjaastad *et al.*^{70,71} Thirty-six (56.3%) submitted data to the practice test (Fig. 1). Of the 112 patients with headache, 48 were diagnosed with CEH. Two patients were excluded from the analysis due to incomplete data sets, thus leaving 46 patients in the study.

Participating MPTs and patients

The mean age of participating MPTs ($n=36$) was 36.6 years (SD=7.8; median 35.5; range 27 [min 23; max 50]), of whom 47.2% were female. All participants had postgraduate level education in manual physical therapy (Educational Institute for Manual Therapy, Amersfoort, the Netherlands or Vrije Universiteit Brussel, Brussels, Belgium). The range of practice experience with patients with headache was 5–18 years.

The mean age of patients ($n=46$) was 36.1 years (SD=5.2; median 36; range 36 [min 21; max 48]), of them 73.9% were female. Patients reported having neck pain and headache for a mean of 1.8 years (SD=1.0; median 2; range 3.5 [min 0.5; max 4]) before entering the study.

Indicators for cervico-cephalgiaphobia

Frequencies and percentages of scores for the eight indicators of cervico-cephalgiaphobia are presented in Table 1. The coverage of ‘often/always’ was substantial for six indicators, as was ‘never’ for coverage of ‘Long-term good results in previous manual physical therapeutic treatment’. ‘Confirmation of “locked” facet joints of the neck by the MPT as a cause for increase of headache’ scored ‘often/always’ in all patients. ‘Increased use of medication with insufficient effect’ scored ‘often/always’ in four (8.7%) patients, while it was substantially covered by ‘sometimes’

in 39 (84.8%) patients. The median score for all indicators was ‘often/always’.

Discussion

Presentation of main findings

We used a systematic approach in defining cervico-cephalgiaphobia and in the development and piloting of a set of indicators reflecting the fear of headache that may be associated with CEH. Having defined cervico-cephalgiaphobia and formulated a consensus-based set of eight indicators with sufficient content validity, the results showed that patients with CEH often experienced fear of headache in combination with fear of locked facet joints of the neck. The MPTs confirmed dysfunction (locked facet joints) of the cervical spine in all cases. Based on these findings, we hypothesise that patients and MPTs were unaware of their reciprocal influence on chronification of CEH and poorer treatment outcome, in terms of long term functional recovery. To the best of our knowledge, this is the first study of fear of headache in patients with CEH.

Indicator set for cervico-cephalgiaphobia

It is difficult to compare our set of eight indicators for cervico-cephalgiaphobia with the set of four questions on medication use of patients with migraine in the study by Giannini *et al.*⁵ These authors concluded that ‘chronic migraineurs with medication overuse had higher scores or cephalgiaphobia than those without medication overuse’. We agree with Peres *et al.*⁴ who hypothesised that ‘cephalgiaphobia may decrease the threshold for initiating analgesic consumption behaviour, leading to acute medication overuse’. Despite the differences between the two studies, our study points to a use of medication with insufficient effect by patients with CEH of around 80%. The possible association between the intensity of CEH and cervico-cephalgiaphobia could be explained by the same hypothesis.

Indicators covering the use of manual physical therapy included the interval between treatments and frequency of manipulation of the neck, as well as short-term and

Table 1 Content validity values, frequency and percentage scores for eight indicators of cervico-cephalgiaphobia in patients with cervicogenic headache (CHE) ($n=46$)

Indicators cervico-cephalgiaphobia	CVR	Never n (%)	Sometimes n (%)	Often/always n (%)
Increased use of medication with insufficient effect	0.6	3 (6.5)	39 (84.8)	4 (8.7)
Short-term positive results in previous manual physical therapeutic treatment	0.8	14 (30.4)	32 (69.6)	
Long-term positive results in previous manual physical therapeutic treatment	1.0	36 (78.2)	5 (10.9)	5 (10.9)
Shorter interval between treatments	0.8	12 (26.1)	34 (73.9)	
Fear of increase in headaches	1.0	8 (17.4)	38 (82.6)	
Fear of ‘locked’ facet joints of the neck	1.0	15 (32.6)	31 (67.4)	
Confirmation of ‘locked’ facet joints of the neck by manual physical therapist (MPT) as a cause for increase in headache	1.0		46 (100)	
More frequent manipulation of the neck	0.8	9 (19.6)	37 (80.4)	

CVR: Content validity ratio (–1: perfect disagreement; +1: perfect agreement; 0: level of 50% agreement of content validity).

*Operational definition of locked facet joint: impairment of joint mobility (decreased movement of facet joint) and pain, diagnosed via a hands-on examination by a physical therapist specialised in spinal manual therapy

long-term results of previous manual physical therapy treatment. These indicators point to a subtype of phobia that accords with the consensus-based definition of cervico-cephalgiaphobia in terms of a shorter interval between treatment sessions and an increase of frequency of manipulation of the neck.

Based on our earlier implementation studies, we set the response cut-off point for indicators at $\geq 60\%$.⁷² This indicator percentage was arbitrarily designated as 'substantial'. The response percentages scores 'often/always' for almost all indicators or 'never' for indicator 'Long-term good results in previous manual physical therapeutic treatment' were around 70% or higher. Increasing the cut-off point to $\geq 70\%$ could therefore be considered in subsequent studies.

The conceptual construct of cervico-cephalgiaphobia is a very important point of discussion. It concerns the conceptual question of whether this type of phobia is exclusively patient-oriented. It could be hypothesised that the MPT may also experience attitudes and beliefs about phobic headache and pain cognitions that subsequently initiate more frequent treatment sessions and shortening of the time interval between sessions. This therapist-oriented phobia may cause an iatrogenic worsening of the phobia in patients and may consequently negatively affect the course of cervico-cephalgiaphobia over time in spite of frequent manual therapy. This implies that the indicators under study should have the capacity to discriminate between patient-oriented and therapist-oriented cervico-cephalgiaphobia. This aspect of the conceptual construct of phobia may be part of future studies in cervico-cephalgiaphobia.

Cervico-cephalgiaphobia

The exact nature of the possible association between worsening of CEH and cervico-cephalgiaphobia, as observed here, remains to be revealed. One plausible hypothesis is that cervico-cephalgiaphobia, like kinesiophobia and other phobias, directly increases the intensity and frequency of CEH by (further) increasing the sensitivity of the central nervous system, making it a potential factor involved in 'cognitive emotional sensitisation'.⁷⁵ We used an indicator set for cervico-cephalgiaphobia to provide only an indirect indication of the level of sensitivity of the central nervous system. Many studies in populations with chronic musculoskeletal pain and headache have utilised Quantitative Sensory Testing (QST) to measure thermal, electrical and mechanical pain thresholds.^{76,77} The additional use of QST measurement (e.g. pressure algometry) in a follow-up study is well worth considering.

Another hypothesis is that worsening of the intensity and frequency of CEH may be iatrogenically provoked by an increased frequency of manual physical therapy treatment sessions in combination with ineffectiveness of increased use of medication. In agree with the hypothesis,

Peres *et al.*⁴ and Giannini *et al.*⁵ about the decrease of threshold for initiating the analgesic consumption behaviour leading to acute medication overuse, our hypothesis is that cervico-cephalgiaphobia may lead to a behaviour of increased use of manual physical therapy, resulting in overuse of treatment sessions involving manipulation of the neck. However, further work is required to support this hypothesis.

CEH and recognition of sensitisation of the nervous system

One can speculate about an association between cervico-cephalgiaphobia and the hyperexcitability of the central and peripheral nervous systems (central and peripheral sensitisation). Over the past decade, a number of studies have investigated central sensitisation of the nervous system (including the sympathetic nervous system) in patients with different types of headache such as tension-type headache,^{78–80} cluster headache^{81,82} and migraine.^{83–85} It is plausible that peripheral and central sensitisation also play a role in the development of CEH. The results of a hypothesis-generating study suggest that rostral neuraxial spread of central sensitisation, probably to the trigeminal spinal nucleus, plays a major role in the development of CEH.⁸⁶ In this state of sensitization, the responsiveness of central neurons to input from polymodal nociceptors of segmentally innervated cervical and pericranial muscles and cervical zygapophysial joints are augmented, resulting in a pathophysiological vicious circle of hypersensitivity of all the cervical tissues and in an increase of CEH.

Practical guidelines for the recognition and treatment of central sensitization in patients with musculoskeletal disorders are available, however, many issues remain.^{87–90} For example, how should clinicians apply the science and evidence regarding central sensitization of the nervous system to CEH when the patients and the MPTs are convinced that the recurrent impairments of cervical motion segments and cervical flexor and extensor muscles are the only cause of chronification of CEH? Despite the many articles published on the recognition and treatment of central sensitisation in groups with chronic musculoskeletal pain other than headache in relation to manual physical therapy, there are indications that many MPTs perform screening for (chronic) musculoskeletal pain mainly on the basis of biomedical and somatic features, and feel unprepared to treat patients with chronic pain patients by exploring the psychosocial factors that influence recovery.^{91–93} Clinical management of patients with CEH needs to extend beyond the anatomical structures related to movement (facet joints) and expand beyond the confines of neuromusculoskeletal and movement-related function (mobility of facet joint functions).^{36,45} It will be a major challenge to develop, implement, and evaluate practical clinical guidelines that integrate with the science and evidence of peripheral and central sensitisation in patients with CEH.

Limitations of the study

First, investigation of cervico-cephalgiaphobia was limited to eight questions asked during history taking. This may have restricted the potential for gathering data on cervico-cephalgiaphobia.

Second, this study is an initial step in the development process of indicators that need to be further tested for reliability and validity in addition to being sensitive to change, acceptable, feasible, and communicable. Our set of indicators was based on the literature and clinical expertise in consensus with a small expert group. Consequently, it has only face and content validity. A next approach may be to reformulate the indicators taking into account specific criteria for phobia described by DSM-V.¹ We suggest additional semi-structured interviews with an international multidisciplinary expert group, practicing MPTs, patients, psychiatrists and psychologists. Of the various methods used to develop indicators, the most frequently applied are the Delphi technique and the RAND method.⁹⁴

Third, although the sample of MPTs was comparable to the national average (mean age 42years; female 56%; $n=17.802$)⁹⁵ and the sample of patients with CEH was comparable to participants in the national Primary Care Physiotherapy Database (mean age 46years; female 75%; $n=80$),²⁵ the relatively low response rates of this study may have introduced selection bias and limited the external validity of the study results.

Lastly, this study took place within primary care structures of the Dutch healthcare system. This may imply that the external validity of the study findings is limited to the Netherlands. Given the importance of the findings for therapists treating patients with CEH anywhere in the world and to enhance the external validity of this primary set of indicators, there is a need for further worldwide research on larger populations, taking into consideration cultural adaptation. The aforementioned results, summarised and discussed, will provide a general view of cervico-cephalgiaphobia, and further associated discussion will be very valuable in the formulation of clinical guidelines.

Conclusions

Cervico-cephalgiaphobia, or fear of headache, in patients with CEH was proposed as a concept and operationalized through the formulation of a consensus-based set of eight indicators. Testing in Dutch manual physical therapy practices suggested that cervico-cephalgiaphobia is common in patients with CEH.

The authors have the impression that in regard to CEH, there is currently a gap between patients and MPTs on the one side and the application of science and available evidence regarding phobia and neurophysiological mechanisms of nociception and pain on the other.

When present in patients with CEH, cervico-cephalgiaphobia must be recognised and treated in order to reduce chronification of headache, limitations to activity

and restrictions to participation. Practical guidelines for the recognition of peripheral and central sensitisation, education programmes in pain physiology and self-reflection models are available for both patients and MPTs, and might be usefully applied to decrease cervico-cephalgiaphobia in CEH patients. Future studies in this area are warranted.

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Disclaimer Statements

Contributors RABO developed the idea, concept, and design of the study, carried out data collection, statistical analysis, interpretation of data and study supervision, and drafted the manuscript. HE was involved in study design, statistical analysis, interpretation of data, and critical review. EM was involved in study design and critical review, and helped draft the manuscript. NR was involved in study design, critical review, and drafting of the manuscript. EvT was involved in study design, support of statistical analysis, interpretation of data and critical review, and helped draft the manuscript. HS helped in developing the idea, concept, study design, and in critical review. JN was involved in study design, interpretation of data and critical review, and helped draft the manuscript. WD helped develop the idea, concept, and design of the study, and was involved in data collection. He sadly passed away on February 4, 2008. All living authors read and approved the final manuscript.

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References

- 1 American Psychiatric Association. Diagnostic and statistical manual of mental disorders. DSM-5. 5th edn. Washington, DC 2013.
- 2 Saper JR. Pearls from an inpatient headache unit. *Headache*. 2008;48(6):820–7.
- 3 Pull CB. Recent trends in the study of specific phobias. *Curr Opin Psychiatry*. 2008;21(1):43–50.
- 4 Peres MF, Mercante JP, Guendler VZ, Corchs F, Bernik MA, Zukerman E, et al. Cephalgiaphobia: a possible specific phobia of illness. *J Headache Pain*. 2007;8(1):56–9.
- 5 Giannini G, Zanigni S, Grimaldi D, Melotti R, Pierangeli G, Cortelli P, et al. Cephalgiaphobia as a feature of high-frequency migraine: a pilot study. *J Headache Pain*. 2013;14:49.
- 6 Oostendorp RAB. Functionele Vertebrobasilaire Insufficiëntie (Vertebrobasilar Insufficiency). Academic Thesis. Nijmegen, The Netherlands: Catholic University Nijmegen; 1988.
- 7 Oostendorp RAB, van Eupen AAJM, Elvers JWH. Aspects of sympathetic system regulation in patients with cervicogenic vertigo.

- Proceedings 5th International Conference International Federation of Orthopaedic Manipulative Therapists. Vail, CO. 1992.
- 8 Oostendorp R, van Eupen AAJM, Elvers JWH, Bernards J. Effects of restrained cervical mobility on involuntary eye movements. *J Man Manip Ther.* 1993;1(4):148–53.
  - 9 Armstrong TG. The use of reassurance. *Lancet.* 1946;2(6423):480–2.
  - 10 Bishop A, Thomas E, Foster NE. Health care practitioners' attitudes and beliefs about low back pain: a systematic search and critical review of available measurement tools. *Pain.* 2007;132(1–2):91–101.
  - 11 Bishop A, Foster NE, Thomas E, Hay EM. How does the self-reported clinical management of patients with low back pain relate to the attitudes and beliefs of health care practitioners? A survey of UK general practitioners and physiotherapists. *Pain.* 2008;135(1–2):187–95.
  - 12 Ostelo RW, Vlaeyen JW. Attitudes and beliefs of health care providers: extending the fear-avoidance model. *Pain.* 2008;135(1–2):3–4.
  - 13 Linton SJ, McCracken LM, Vlaeyen JW. Reassurance: help or hinder in the treatment of pain. *Pain.* 2008;134(1–2):5–8.
  - 14 Antonaci F, Bono G, Mauri M, Drottning M, Buscone S. Concepts leading to the definition of the term cervicogenic headache: a historical overview. *J Headache Pain.* 2005;6(6):462–6.
  - 15 Haldeman S, Dagenais S. Cervicogenic headaches: a critical review. *Spine J.* 2001;1(1):31–46.
  - 16 Hülse M, Seifert K. Cervicogenic head and neck pain. *HNO.* 2001;53(9):804–9.
  - 17 Gallagher RM. Cervicogenic headache. *Expert Rev Neurother.* 2007;7(10):1279–83.
  - 18 Becker WJ. Cervicogenic headache: evidence that the neck is a pain generator. *Headache.* 2010;50(4):699–705.
  - 19 Chua NH, Wilder-Smith O, Vissers K. The neck: a pain generator for the head. *Pain Manag.* 2012;2(3):191–4.
  - 20 Headache Classification Subcommittee of the International Headache Society. The International Classification of Headache Disorders: 2nd edition. *Cephalalgia.* 2004;24(Suppl 1):9–160.
  - 21 Sjaastad O, Bakkeiteig LS. Prevalence of cervicogenic headache: Vågå study of headache epidemiology. *Acta Neurol Scand.* 2008;117(3):173–80.
  - 22 Nilsson N. The prevalence of cervicogenic headache in a random population sample of 20–59 year olds. *Spine.* 1995;20(17):1884–8.
  - 23 Anthony M. Cervicogenic headache: prevalence and response to local steroid therapy. *Clin Exp Rheumatol.* 2000;18(2 Suppl 19):S59–S64.
  - 24 Van Suijlekom HA, Antonaci F. Cervicogenic headache. In: Martiletti P, Steiner TJ, editors. *Handbook of headache practical management.* Milan: Springer; 2011; pp. 472 – 482.
  - 25 Barten D, Swinkels I, Veenhof C. Patiëntkarakteristieken, het gezondheidsprobleem en het fysiotherapeutische behandelproces bij patiënten met hoofd-, hals- en nekklachten (Characteristics, health problems and physical therapy treatment process in patients with head and neck complaints). In: Calders P, Geraets JJXR, Nijs J, Veenhof C, van Wegen EEH, van Wilgen CP, editors. *Jaarboek fysiotherapie – kinesiotherapie 2012.* Houten: Bohn Stafleu van Loghum; 2011; pp. 162–174.
  - 26 Escolar J. The afferent connections of the 1st, 2nd, and 3rd cervical nerves in the cat; an analysis by Marchi and Rasdolsky methods. *J Comp Neurol.* 1948;89(2):79–92.
  - 27 Kerr FW. Structural relation of the trigeminal spinal tract to upper cervical roots and the solitary nucleus in the cat. *Exp Neurol.* 1961;4:134–48.
  - 28 Kerr FW. Central relationships of trigeminal and cervical primary afferents in the spinal cord and medulla. *Brain Res.* 1972;43(2):561–72.
  - 29 Bogduk N. The anatomical basis for cervicogenic headache. *J Manipulative Physiol Ther.* 1992;15(1):67–70.
  - 30 Chua NHL, Suijlekom HV, Wilder-Smith OH, Vissers KCP. Understanding cervicogenic headache. *Anesth Pain Med.* 2012;2(1):3–4.
  - 31 Oosterhof J, Wilder-Smith OH, Oostendorp RA, Crul BJ. Different mechanisms for the short-term effects of real versus sham transcutaneous electrical nerve stimulation (TENS) in patients with chronic pain: a pilot study. *J Pain Palliat Care Pharmacother.* 2012;26(1):5–12.
  - 32 Meeus M, Hermans L, Ickmans K, Struyf F, Van Cauwenbergh D, Bronckaerts L, et al. Endogenous pain modulation in response to exercise in patients with rheumatoid arthritis, patients with chronic fatigue syndrome and comorbid fibromyalgia, and healthy controls: a double-blind randomized controlled trial. *Pain Pract.* 2015;15(2):98–106.
  - 33 Chu J, Allen DD, Pawlowsky S, Smoot B. Peripheral response to cervical or thoracic spinal manual therapy: an evidence-based review with meta analysis. *J Man Manip Ther.* 2014;22(4):220–9.
  - 34 Bodes-Pardo G, Pecos-Martin D, Gallego-Izquierdo T, Salom-Moreno J, Fernandez-de-Las-Penas C, Ortega-Santiago R. Manual treatment for cervicogenic headache and active trigger point in the sternocleidomastoid muscle: a pilot randomized clinical trial. *J Manipulative Physiol Ther.* 2013;36:403–11.
  - 35 Fernandez-de-Las-Penas C, Alonso-Blanco C, Cuadrado ML, Pareja JA. Spinal manipulative therapy in the management of cervicogenic headache. *Headache.* 2005;45:1260–3.
  - 36 Fernandez-de-Las-Penas C, Courtney CA. Clinical reasoning for manual therapy management of tension type and cervicogenic headache. *J Man Manip Ther.* 2014;22:44–50.
  - 37 Kroese MEAL, de Vet HCW, Scholten RJPM. Inventarisatie van effectonderzoek naar regelmatig toegepaste fysiotherapeutische behandelingen bij chronisch benigne pijn (Inventory of effectiveness on frequently used physiotherapy treatments in patients with chronic benign pain). *Ned Tijdschr Fysiother.* 2002;112(2):42–9.
  - 38 Dekker F, Van Duijn NP, Ongerling JEP, Bartelink MEL, Boelman L, Burgers JS, et al. NHG-Standaard Hoofdpijn (derde herziening) (Standard Headache of Dutch College of General Practitioners). *Huisarts Wet.* 2014;57(1):20–31.
  - 39 Zito G, Jull G, Story I. Clinical tests of musculoskeletal dysfunction in the diagnosis of cervicogenic headache. *Man Ther.* 2006;11(2):118–29.
  - 40 Gross A, Miller J, D'Sylva J, Burnie SJ, Goldsmith CH, Graham N, et al. Manipulation or mobilisation for neck pain. *Cochrane Database Syst Rev.* 2010; 20(1):CD004249.
  - 41 Racicki S, Gerwin S, Diclaudio S, Reinmann S, Donaldson M. Conservative physical therapy management for the treatment of cervicogenic headache: a systematic review. *J Man Manip Ther.* 2013;21(2):113–24.
  - 42 Chaibi A, Russell MB. Manual therapies for cervicogenic headache: a systematic review. *J Headache Pain.* 2012;13(5):351–9.
  - 43 Posadzki P, Ernst E. Spinal manipulations for cervicogenic headaches: a systematic review of randomized clinical trials. *Headache.* 2011;51(7):1132–9.
  - 44 Jull G, Trott P, Potter H, Zito G, Niere K, Shirley D, et al. A randomized controlled trial of exercise and manipulative therapy for cervicogenic headache. *Spine (Phila Pa 1976).* 2002;27(17):1835–43.
  - 45 Nijs J, Roussel N, Paul van Wilgen C, Koke A, Smeets R. Thinking beyond muscles and joints: therapists' and patients' attitudes and beliefs regarding chronic musculoskeletal pain are key to applying effective treatment. *Man Ther.* 2013;18(2):96–102.
  - 46 Butler CC, Evans M, Greaves D, Simpson S. Medically unexplained symptoms: the biopsychosocial model found wanting. *J R Soc Med.* 2004;97(5):219–22.
  - 47 Jull GA, Stanton WR. Predictors of responsiveness to physiotherapy management of cervicogenic headache. *Cephalalgia.* 2005;25:101–8.
  - 48 Hall T, Briffa K, Hopper D. Clinical evaluation of cervicogenic headache: a clinical perspective. *J Man Manip Ther.* 2008;16:73–80.
  - 49 Buer N, Linton SJ. Fear-avoidance beliefs and catastrophizing: occurrence and risk factor in back pain and ADL in the general population. *Pain.* 2002;99:485–91.
  - 50 Picavet HS, Vlaeyen JW, Schouten JS. Pain catastrophizing and kinesiophobia: predictors of chronic low back pain. *Am J Epidemiol.* 2002;156:1028–34.
  - 51 Crombez G, Vlaeyen JW, Heuts PH, Lysens R. Pain-related fear is more disabling than pain itself: evidence on the role of pain-related fear in chronic back pain disability. *Pain.* 1999;80(1–2):329–39.
  - 52 Leeuw M, Goossens ME, Linton SJ, Crombez G, Boersma K, Vlaeyen JW. The fear-avoidance model of musculoskeletal pain: current state of scientific evidence. *J Behav Med.* 2007;30(1):77–94.
  - 53 Wideman TH, Asmundson GG, Smeets RJ, Zautra AJ, Simmonds MJ, Sullivan MJ, et al. Rethinking the fear avoidance model: toward a multidimensional framework of pain-related disability. *Pain.* 2013;154(11):2262–5.
  - 54 Swinkels-Meewisse IE, Roelofs J, Verbeek AL, Oostendorp RA, Vlaeyen JW. Fear of movement/(re)injury, disability and participation in acute low back pain. *Pain.* 2003;105(1–2):371–9.
  - 55 Swinkels-Meewisse IE, Roelofs J, Schouten EG, Verbeek AL, Oostendorp RA, Vlaeyen JW. Fear of movement/(re)injury predicting chronic disabling low back pain: a prospective inception cohort study. *Spine (Phila Pa 1976).* 2006;31(6):658–64.
  - 56 Roelofs J, Sluiter JK, Frings-Dresen MH, Goossens M, Thibault P, Boersma K, et al. Fear of movement and (re)injury in chronic musculoskeletal pain: evidence for an invariant two-factor model of the Tampa Scale for Kinesiophobia across pain diagnoses and Dutch, Swedish, and Canadian samples. *Pain.* 2007;131(1–2):181–90.
  - 57 Ramprasas M, Shenoy DS, Sandhu JS, Sankara N. The influence of kinesiophobia on trunk muscle voluntary responses with pre-

- programmed reactions during perturbation in patients with chronic low back pain. *J Bodyw Mov Ther.* **2011**;15(4):485–95.
- 58 George SZ, Calley D, Valencia C, Beneciuk JM. Clinical investigation of pain-related fear and pain catastrophizing for patients with low back pain. *Clin J Pain.* **2011**;27(2):108–15.
  - 59 Visscher CM, Ohrbach R, van Wijk AJ, Wilkosz M, Naeije M. The Tampa Scale for Kinesiophobia for temporomandibular disorders (TSK-TMD). *Pain.* **2010**;150(3):492–500.
  - 60 Nijs J, Meeus M, Heins M, Knoop H, Moorkens G, Bleijenberg G. Kinesiophobia, catastrophizing and anticipated symptoms before stair climbing in chronic fatigue syndrome: an experimental study. *Disabil Rehabil.* **2012**;34(15):1299–305.
  - 61 Suhr J, Spickard B. Pain-related fear is associated with cognitive task avoidance: exploration of the cogniphobia construct in a recurrent headache sample. *Clin Neuropsychol.* **2012**;26(7):1128–41.
  - 62 Martins IP, Gouveia RG, Parreira E. Kinesiophobia in migraine. *J Pain.* **2006**;7(6):445–51.
  - 63 De Hertogh WJ, Vaes PH, Vijverman V, De Cordt A, Duquet W. The clinical examination of neck pain patients: the validity of a group of tests. *Man Ther.* **2007**;12(1):50–5.
  - 64 De Hertogh W, Vaes P, Duquet W. The validity of the manual examination in the assessment of patients with neck pain. *Spine J.* **2007**;17(7):628–9.
  - 65 De Koning CH, van den Heuvel SP, Staal JB, Smits-Engelsman BC, Hendriks EJ. Clinimetric evaluation of active range of motion measures in patients with non-specific neck pain: a systematic review. *Eur Spine J.* **2008**;17(7):905–21.
  - 66 Hansen K, Schliack H. Segmentale Innervation. Ihre Bedeutung für Klinik und Praxis (Segmental Innervation). Stuttgart: Georg Thieme Verlag; **1962**.
  - 67 WHO. International classification of functioning, disability and health. Geneva: WHO; **2001**.
  - 68 Sjaastad O, Fredriksen TA. Cervicogenic headache: criteria, classification and epidemiology. *Clin Exp Rheumatol.* **2000**;18(2 Suppl 19):S3–S6.
  - 69 Antonaci F, Fredriksen TA, Sjaastad O. Cervicogenic headache: clinical presentation, diagnostic criteria, and differential diagnosis. *Curr Pain Headache Rep.* **2001**;5(4):387–92.
  - 70 Sjaastad O, Fredriksen TA, Pfaffenrath V. Cervicogenic headache: diagnostic criteria. The Cervicogenic Headache International Study Group. *Headache.* **1998**;38(6):442–5.
  - 71 Sjaastad O, Fredriksen TA, Pfaffenrath V. Cervicogenic headache: diagnostic criteria. *Headache.* **1990**;30(11):725–6.
  - 72 Oostendorp RA, Rutten GM, Dommerholt J, Nijhuis-van der Sanden MW, Harting J. Guideline-based development and practice test of quality indicators for physiotherapy care in patients with neck pain. *J Eval Clin Pract.* **2013**;19(6):1044–53.
  - 73 Lawshe CH. Quantitative approach to content validity. *Pers Psychol.* **1975**;28(4):563–75.
  - 74 Ayre C, Scally AJ. Critical values for Lawshe's content validity ratio: revisiting the original methods of calculation. *Meas Eval Couns Dev.* **2014**;47(1):79–86.
  - 75 Brosschot JF. Cognitive-emotional sensitization and somatic health complaints. *Scand J Psychol.* **2002**;43(2):113–21.
  - 76 Curatolo M, Arendt-Nielsen L. Central hypersensitivity in chronic musculoskeletal pain. *Phys Med Rehabil Clin N Am.* **2015**;26(2):175–84.
  - 77 Fernandez-de-las-Penas C. Clinical evidence of generalised mechanical hypersensitivity in local musculoskeletal pain syndromes and headaches. Aalborg: Center for Sensory-Motor Interaction (SMI), Department of Health Science and Technology, Aalborg University; **2012**.
  - 78 Bezov D, Ashina S, Jensen R, Bendtsen L. Pain perception studies in tension-type headache. *Headache.* **2011**;51(2):262–71.
  - 79 Bendtsen L. Central sensitization in tension-type headache – possible pathophysiological mechanisms. *Cephalalgia.* **2000**;20(5):486–508.
  - 80 Filatova E, Latysheva N, Kurenkov A. Evidence of persistent central sensitization in chronic headaches: a multi-method study. *J Headache Pain.* **2008**;9(5):295–300.
  - 81 Chua NH, Vissers KC, Wilder-Smith OH. Quantitative sensory testing may predict response to sphenopalatine ganglion pulsed radiofrequency treatment in cluster headaches: a case series. *Pain Pract.* **2011**;11(5):439–45.
  - 82 Fernandez-de-Las-Penas C, Ortega-Santiago R, Cuadrado ML, Lopez-de-Silanes C, Pareja JA. Bilateral widespread mechanical pain hypersensitivity as sign of central sensitization in patients with cluster headache. *Headache.* **2011**;51(3):384–91.
  - 83 Burstein R, Levy D, Jakubowski M. Effects of sensitization of trigeminovascular neurons to triptan therapy during migraine. *Rev Neurol (Paris).* **2005**;161(6–7):658–60.
  - 84 Jakubowski M, Levy D, Gloor-Aryeh I, Collins B, Bajwa Z, Burstein R. Terminating migraine with allodynia and ongoing central sensitization using parenteral administration of COX1/COX2 inhibitors. *Headache.* **2005**;45(7):850–61.
  - 85 de Tommaso M, Delussi M, Vecchio E, Scirucchio V, Invitto S, Livrea P. Sleep features and central sensitization symptoms in primary headache patients. *J Headache Pain.* **2014**;15(1):64.
  - 86 Chua NH, van Suijlekom HA, Vissers KC, Arendt-Nielsen L, Wilder-Smith OH. Differences in sensory processing between chronic cervical zygapophysial joint pain patients with and without cervicogenic headache. *Cephalalgia.* **2011**;31(8):953–63.
  - 87 Nijs J, Paul van Wilgen C, Van Oosterwijck J, van Ittersum M, Meeus M. How to explain central sensitization to patients with 'unexplained' chronic musculoskeletal pain: practice guidelines. *Man Ther.* **2011**;16(5):413–8.
  - 88 van Wilgen P, Beetsma A, Neels H, Roussel N, Nijs J. Physical therapists should integrate illness perceptions in their assessment in patients with chronic musculoskeletal pain; a qualitative analysis. *Man Ther.* **2014**;19(3):229–34.
  - 89 Carlson K, Carlson N. An overview of the management of persistent musculoskeletal pain. *Ther Adv Musculoskelet Dis.* **2011**;3(2):91–9.
  - 90 Nijs J, Van Houdenhove B, Oostendorp RA. Recognition of central sensitization in patients with musculoskeletal pain: application of pain neurophysiology in manual therapy practice. *Man Ther.* **2010**;15(2):135–41.
  - 91 Oostendorp Rob AB, Elvers H, Mikołajewska E, Laekeman M, van Trijffel E, Samwel H, et al. Manual physical therapists' use of biopsychosocial history taking in the management of patients with back or neck pain in clinical practice. *ScientificWorldJourn.* **2015**;2015:170463.
  - 92 Synnott A, O'Keeffe M, Bunzli S, Dankaerts W, O'Sullivan P, O'Sullivan K. Physiotherapists may stigmatise or feel unprepared to treat people with low back pain and psychosocial factors that influence recovery: a systematic review. *J Physiother.* **2015**;61(2):68–76.
  - 93 Singla M, Jones M, Edwards I, Kumar S. Physiotherapists' assessment of patients' psychosocial status: are we standing on thin ice? A qualitative descriptive study. *Man Ther.* **2015**;20(2):328–34.
  - 94 Braspenning J, Campbell S, Grol R. Measuring changes in patient care: development and use of indicators. In: Grol R, Wensing M, Eccles M, editors. Improving patient care: the implementation of change in clinical practice. Edinburgh: Elsevier Butterworth Heinemann; **2005**; pp. 222–234.
  - 95 Van Hassel DTP, Kenens RJ. Beroepen in de gezondheidszorg. Cijfers uit de registratie van fysiotherapeuten. (Health professions. Registration of physical therapists) Peildatum 1 januari 2012 (reference date January 1, 2012). Utrecht: NIVEL; **2013**.

Appendix

History taking – Questionnaire Cervicocephalalgia phobia	Never	Sometimes	Often/always
1. Have you used medication due to fears of headache but without effect?			
2. Have you experienced any short-term positive effects of previous MPT* treatments?			
3. Have you experienced any long-term positive effects of previous MPT* treatments?			
4. Have you noticed that the interval between treatments has become shorter?			
5. Are you afraid that your headaches will increase?			
6. Are you afraid of 'locked facet joints'*** in the neck?			
7. Did the manual therapist confirm 'locked facet joints'*** in the neck as the cause of your headache?			
8. Has the frequency of manipulation increased?			
*MPT: manual physical therapy. **Operational definition of locked facet joint: impairment of joint mobility (decreased movement of facet joint) and pain, diagnosed via a hands-on examination by a physical therapist specialised in spinal manual therapy.			