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Systematic review

Joint position sense error in people with neck pain: A systematic review

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ABSTRACT

Background: Several studies in recent decades have examined the relationship between proprioceptive deficits and neck pain. However, there is no uniform conclusion on the relationship between the two. Clinically, proprioception is evaluated using the Joint Position Sense Error (JPSE), which reflects a person's ability to accurately return his head to a predefined target after a cervical movement.
Objectives: We focused to differentiate between JPSE in people with neck pain compared to healthy controls.
Study design: Systematic review according to the PRISMA guidelines.
Method: Our data sources were Embase, Medline OvidSP, Web of Science, Cochrane Central, CINAHL and Pubmed Publisher. To be included, studies had to compare JPSE of the neck (O) in people with neck pain (P) with JPSE of the neck in healthy controls (C).
Results/findings: Fourteen studies were included. Four studies reported that participants with traumatic neck pain had a significantly higher JPSE than healthy controls. Of the eight studies involving people with non-traumatic neck pain, four reported significant differences between the groups. The JPSE did not vary between neck-pain groups.
Conclusions: Current literature shows the JPSE to be a relevant measure when it is used correctly. All studies which calculated the JPSE over at least six trials showed a significantly increased JPSE in the neck pain group. This strongly suggests that 'number of repetitions' is a major element in correctly performing the JPSE test.

1. Introduction

The primary measure to clinically operationalize cervical proprioception is the Joint Position Sense Error (JPSE) (Armstrong et al., 2008; Strimpakos, 2011). Joint position sense, an individual's ability to reproduce and perceive previous predetermined positions or ranges of motion of a joint, is a major component of proprioception. The error people make whilst reproducing the predefined position is defined as the JPSE. Recently, several studies on the relation between neck pain and JPSE have been published (Woodhouse and Vasseljen, 2008; Cheng et al., 2010; Chen and Treleaven, 2013).

Cervical proprioception is the sense of position of the head or neck in space, describing the complex interaction between afferent and efferent receptors to monitor the position and movement

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(Newcomer et al., 2000). In the cervical spine, this sense has its neurological basis in muscle spindles (Proske and Gandevia, 2012) and, to a lesser extent, in tendon organs (Golgi receptors) (Hogervorst and Brand, 1998), cutaneous receptors, and joint receptors (McCloskey, 1978; Grigg, 1994; Lephart et al., 1997; Proske et al., 2000). The cervical muscles provide information to (Bolton et al., 1998) and receive information from the central nervous system (Kalaska, 1994; Hellström et al., 2005). Afferent information from the cervical muscles converges in the vestibular nuclei, where the head movement-related information from the visual and vestibular system also converges (Corneil et al., 2002). Malmström et al. (2009) showed that accurate head-on-trunk orientation can be achieved without vestibular information. This suggests that proprioceptive information of the cervical spine is important for head-on-trunk orientation. The cervical JPSE is assessed by testing the ability of a blindfolded participant to accurately relocate their head to the trunk relative to a predefined target (often the neutral position of the head) after a cervical movement. Other examples of joint regions in which JPSE has been used for testing proprioception are the shoulder (Anderson and Wee, 2011), the knee (van der Esch et al., 2013), and the ankle (Nakasa et al., 2008).

People with neck pain originating from trauma and people whose neck pain has developed more gradually both seem to have a higher JPSE than people without neck pain (Feipel et al., 2006; Cheng et al., 2010). This implies that an increase in JPSE may not be caused solely by soft tissue damage or neurological impairments following trauma (Revel et al., 1991; Sterling et al., 2003).

Narrative reviews of the literature on cervical JPSE have been published (Armstrong et al., 2008, Strimpakos, 2011). Both reviews give conflicting conclusions concerning the presence of a higher JPSE in people with neck pain. The present study is a comprehensive, systematic overview according the PRISMA guidelines of the literature. It presents the data of the JPSE of the cervical spine caused by neck pain of traumatic and non-traumatic origin in comparison of the JPSE in healthy controls.

2. Methods

The PRISMA guidelines (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) (Moher et al., 2009) were used in this systematic literature review to report the method of literature search, appraisal, and presentation of evidence.

2.1. Eligibility criteria

To be included in this systematic review, studies had to report on joint position sense error of the neck (O); and, include participants with neck pain (P), compared to healthy controls (C). It is important to compare the JPSE of people with neck pain with the JPSE of healthy controls because it is assumed that a higher JPSE test reflects aberrant afferent input from the neck (Revel et al., 1991; Heikkilä and Wenngren, 1998; Treleaven et al., 2003; Malmström et al., 2009). Therefor a reference score form healthy controls is a necessity.

2.2. Information sources and search parameters

In order to be as comprehensive as possible, the following databases were searched on December 17th 2014:Embase, Medline OvidSP, Web of Science, Cochrane Central, CINAHL and Pubmed Publisher. Keywords were derived from the research question and transformed to associated "Emtree" terms and free-text words. For Embase, the following Emtree terms were used: sensorimotor integration, sensorimotor function, somatosensory system, somatosensory cortex, balance impairment, motor control, proprioception, body equilibrium, eye movement, proprioceptive feedback, cornea reflex, neck pain, and whiplash injury.

The free-text words were as follows: deep sensitivity, kinesthe*, propriorecep*, propriocep*, kinesio NEXT/1 percept*, cornea*, eye* OR ocular OR cervicoocul* NEAR/3 reflex*, movement*,body, musculoskelet*, postural, NEAR/3 balanc*, equilibr*, sway, control, joint position, head position, neck position, NEAR/3 error*, sense*, reproduc*, abilit*, inaccura*, accura*, replicat*, head NEAR/3 steadiness, balance NEAR/3 impair*, difficult*, neck, cervic* NEAR/6 pain*, hyperextension*, ache, neckache*, Cervicalgia*, Cervicodynia*, whiplash. In addition, Medline, OvidSP, Web of Science, Cochrane Central, CINAHL and Pubmed Publisher were similarly searched with their own thesaurus used for indexing studies and free entries, in order to be as comprehensive as possible.

2.3. Study selection

In order to be included, studies had to meet the following criteria: (1) Participants in the study had to be over 18 years old; (2) Participants had to suffer from neck pain; (3) The outcome measures in the study had to be the JPSE; (4) Control subjects had to be healthy individuals; and (5) The study had to be written in English. Initially, the search results were screened based on title and abstract. The studies that fulfilled all inclusion criteria were evaluated in full-text, and included in the systematic review.

2.4. Data items and collection

Information was extracted from the included studies and presented in three evidence tables (Tables 1–3). This information is presented in the evidence table regarding (1) study, (2) sample size, (3) characteristics of the participants, (4) JPSE testing instrument, (5) JPSE testing protocol, and (6) results. Data extraction was executed by author JV and checked by author LV.

2.5. Risk of bias in individual studies

The validity and risk of bias of the remaining studies was checked by using the "Methodology Checklist 4: Case-control studies" version 2.0, provided by the Scottish Intercollegiate Guidelines Network (SIGN) (www.sign.ac.uk). The SIGN-group develops evidence-based clinical practice guidelines in order to translate new knowledge into clinical action. One aspect of the work of this group is the development of critical appraisal checklists. Studies were scored on a clearly focused research question, on the description of the internal validity: *i.e.* the selection of subjects; exclusion of selection bias; clear definition of outcomes; blinding of assessors; reliable assessment of exposure; identification of potential confounders; and provision of confidence intervals. For the studies, the grading score has been set from "Low quality" (0), to "Acceptable" (+), to "High quality" (++). In the present review, only studies graded as "Acceptable" (+) or "High quality" (++) were included. This criterion was set a priori.

Methodological quality of the included studies was assessed blindly and independently by authors JV and LV. After both researchers had appraised the selected studies, results were compared and any differences discussed after screening the studies a second time. In the event of disagreement a third opinion was provided by author GK.

2.6. Summary measures

The principal outcome measure of this review was the JPSE, which was the main issue to be researched in the included studies. In 9 of the 14 included studies, JPSE was defined as "the ability to

Table 1
Evidence table of the included studies which researched participants with traumatic neck pain.

Reference	Sample	Inclusion criteria	JPSE testing instrument	JPSE testing protocol	Results
Armstrong et al. (2005)	23 WAD (8M, 15W) 41.2 ± 11.9 23 CON (10M, 13W) 33.9 ± 12.1	WAD: grade 2 or 3 not <3 Mo or >5Yr CON: Healthy WAD and CON matched; age and gender	3 Space Fastrak (model 3SF0002, Polhemus, Navigation Science Division, Kaiser Aerospace,Colchester, Vermont, USA) 1 sensor centered on the forehead, 1 sensor on the spinous of c3 and 1 sensor on the spinous of T1	Flexion, extension, rotation. Mean of 3 trials Self-selected neutral position Self-selected pace	WAD: Global abs. error ± SD (°), all movements combined Neutral = 3.55 ± 1.72, Mid-range = 2.97 ± 1.15 CON: Global abs. error ± SD (°), all movements combined Neutral = 3.25 ± 2.32, Mid-range = 2.43 ± 0.62
Woodhouse and Vasseljen (2008)	56 WAD (34W, 22M) 57 CON (28W, 29M) 38.2 ± 10.9	WAD: grade 1 or 2 not <6 Mo or >10Yr CON: Healthy	3 Space Fastrak (Polhemus, Inc, Colchester, Vermont, USA) 1 sensor centered on the forehead, 1 sensor placed on the upper part of the wooden backrest above the subject's head.	Rotation Mean of 4 trials Absolute error (largest error was selected) Selected neutral position Self-selected pace. Examiner was not blinded	WAD: Global abs. error ± SD (°) Left-right rotation = 3.35 ± 1.6 CON: Global abs. error ± SD (°) Left-right rotation = 2.86 ± 1.2
Treleaven et al. (2003)	76 WAD Dizzy (71% W) 39.11(1.3SE) 26 WAD Not Dizzy (73%W) 40.23 (1,9 SE) 102 WAD combined 44 CON (66% W) 34.1(1.8 SE)	WAD Dizzy: grade 2 or 3 not < 3Mo with dizziness WAD Not Dizzy: grade 2 or 3 not < 3Mo without dizziness WAD combined: grade 2 or 3 CON: Healthy	3 Space Fastrak (Polhemus, Inc, Colchester, Vermont, USA) 1 sensor centered on the forehead, 1 sensor on the spinous of C7	Rotation, extension Mean of 3 trials Absolute errors Selected neutral position Self-selected pace	WAD Dizzy: Global abs. error ± SE (°) Right rotation = 4.5 ± 0.3, Left rotation = 3.9 ± 0.3, Extension = 3.5 ± 0.3 WAD Not Dizzy: Global abs. error ± SE (°) Right rotation = 2.9 ± 0.4, Left rotation = 2.8 ± 0.4, Extension = 3.5 ± 0.3 WAD Combined: Global abs. error ± SE (°) Right rotation = 4.1 ± 1.6*, Left rotation = 3.6 ± 0.3*, Extension = 2.4 ± 0.3* CON: Global abs. error ± SE (°) Right rotation = 2.5 ± 0.2, Left rotation = 2.0 ± 0.2, Extension = 2.4 ± 0.3
Heikkilä and Wenngren. (1998)	27 WAD (13W,14M) 39 CON (24W, 15M)	WAD Dizzy: grade 2 or 3 CON: Healthy	Laser pointer	Rotation, flexion/ extension Mean of 10 trials maximal end range	WAD: Global abs. error ± SD (cm) Right rotation = 4.05 ± 3.4*, Left rotation = 3.36 ± 2.41*, Flexion = 4.14 ± 3.75*, Extension = 3.80 ± 2.,86* CON: Global abs. error ± SD (cm) Right rotation = 2.85 ± 2.00 Left rotation = 2.73 ± 1.78, Flexion = 2.57 ± 2.08, Extension = 2.85 ± 1.88
Kristjansson et al. (2003)	22 WAD (11W, 11M) 33.4 ± 10.6 21 CON (11W, 10M) 26.9 ± 6.4	WAD Whiplash injury not < 3 Mo or > 48Mo, no previous history of NP CON: Healthy	3 Space Fastrak (Polhemus, Inc, Colchester, Vermont, USA) 1 sensor centered on the forehead, 1 sensor on the spinous of C7	Rotation Mean of 3 trials Error in degrees Full active rotation within comfortable limits Selected neutral position	Trauma: Global abs. error ± SD (°) Rotation = 4.14 ± 1.58* CON: Global abs. error ± SD (°) Rotation = 2.48 ± 1.12
Sterling et al. (2003)	19 Moderate/severe pain and disability after WAD (84%W) 41.3 ± 13.6 22 Mild pain and disability after WAD (64%W) 34.7 ± 12.6 25 Recovered after WAD (60%W) 33.5 ± 10.2 20 CON (60%W) 40.1 ± 13.6	Mod/Sev pain and dis: WAD grade 2 or 3, NDI score of >30 after 3 Mo post injury Mild pain and dis: WAD grade 2 or 3, NDI score of 10–28 after 3 Mo post injury Recovered: WAD grade 2 or 3, NDI score of <8 after 3 Mo post injury CON: Healthy	3 Space Fastrak (Polhemus, Inc, Colchester, Vermont, USA) sensor placement unclear	Rotation, extension Mean of 3 trials Absolute errors Selected neutral position Movement within comfortable limits	Moderate/severe pain and disability after WAD: Marginal means ± SEM (°) Left rotation = 3.2 ± 0.3, Right rotation = 4.8* ± 0.3, Extension = 4.1 ± 0.3 Mild pain and disability after WAD: Marginal means ± SEM (°) Left rotation = 2.7 ± 0.2, Right rotation = 2.7 ± 0.3, Extension = 3.4 ± 0.3 Recovered after WAD: Marginal means ± SEM (°) Left rotation = 3.0 ± 0.2, Right rotation = 3.6 ± 0.3, Extension = 3.3 ± 0.3

Table 1 (*continued*)

Reference	Sample	Inclusion criteria	JPSE testing instrument	JPSE testing protocol	Results
					CON: Marginal means ± SEM (°) Left rotation = 2.6 ± 0.3, Right rotation = 2.7 ± 0.3, Extension = 2.8 ± 0.3
Feipel et al. (2006)	29 WAD (62%W) 37 ± 14 26 CON (54%W) 35 ± 11	WAD: Grade 1, 2 or 3 CON: Healthy	Electrogoniometer (CA 6000 Spine Motion Analyzer, O.S.I., Union City, CA)mounted using a harness on the spinous process of the first thoracic vertebra and a helmet on top of the head	Rotation, flexion, extension, lateral bending. Mean of 4 trials	WAD: Global abs. error ± SD (°) Rotation = 1.1 ± 1.1 Flexion/extension = 3.5 ± 2.4 Lateral bending = 0.8 ± 0.6 CON: abs. error ± SD (°) Rotation = 0.6 ± 0.5 Flexion/extension = 2.1 ± 2.0 Lateral bending = 0.4 ± 0.3
Grip et al. (2007)	22 WAD (17W, 5M) 49 ± 15 24 CON (16W, 8M) 50 ± 18	WAD: grade 1, 2 not <3 Mo of symptoms Con: Healthy	ProReflex System(Qualisys Medical AB, Gothenburg, Sweden) 13 markers were placed on the head and upper torso	Rotation, flexion, extension Mean of 5 trials Error in degrees Self-selected neutral position Self-selected pace Moving back to neutral only with closed eyes	WAD: Global abs. error ± SD (°) Right rotation = 3.7 ± 1.9. Left rotation = 4.0 ± 2.1 Flexion 3.4 ± 1.6, Extension = 3.5 ± 1.8 CON: Global abs. error ± SD (°) Right rotation = 3.1 ± 1.3, Left rotation = 3.5 ± 1.3 Flexion 2.9 ± 0.9, Extension = 2.7 ± 1.0
Sjolander et al. (2008)	7 WAD (2M, 5W) 45 ± 11 16 CON (3M, 13W) 40 ± 9	WAD: grade 2 or 3 not < 6 Mo CON: Healthy	3 Space Fastrak (Polhemus, Inc, Colchester, Vermont, USA) 1 sensor centered on the forehead, 1 sensor on the spinous of T1	Rotation Mean of trials unclear Move the head as fast and far as possible Self-selected neutral position Patients were standing. Examiner was blinded	WAD: Over/under shooting ± SD (°) Right rotation = −0.4 ± 0.8, Left rotation = 0.7 ± 1.1 CON: Over/under shooting ± SD (°) Right rotation = 0.1 ± 0.5, Left rotation = 0.1 ± 0.6

JPSE; Joint position sense error, WAD = Whiplash associated disorder; WAD grade 1 = neck complaints of pain, stiffness or tenderness only, but no physical signs are noted by the examining physician; WAD grade 2 = neck complaints and musculoskeletal signs as decreased range of motion and point tenderness in the neck; WAD grade 3 includes additional signs (decreased or absent deep tendon reflexes, weakness, and sensory deficits); CON: Healthy controls; SD = Standard Deviation; SE = Standard Error, SEM = Standard Error of the Mean; M = Men; W = Women; Abs. = Absolute; Mod = Moderate; Sev = Severe; NDI = Neck Disability Index; Yr = Year; Mo=Month* = indicates statistically significant differences between groups.

reposition the head to the starting position after a maximal active movement of the head in a vertical or horizontal plane with occluded vision" (Revel et al., 1991; Treleaven et al., 2003; Sterling et al., 2003; Armstrong et al., 2005; Feipel et al., 2006; Sjolander et al., 2008; Woodhouse and Vasseljen, 2008; Cheng et al., 2010; Uthaikhup et al., 2012; Chen and Treleaven, 2013). The outcome measure was given in degrees or centimeters.

3. Results

3.1. Study selection

A total of 1163 studies were identified. As shown in Fig. 1, 14 studies remained after two screening phases.

3.2. Study characteristics

The characteristics of the data that were extracted from the included studies (study, sample size, characteristics of the participants, JPSE testing instrument, JPSE testing protocol, and results) are presented in Tables 1–3. In nine out of 14 included studies JPSE was assessed in participants with traumatic neck pain (Heikkilä and Wenngren, 1998; Kristjansson et al., 2003; Sterling et al., 2003; Treleaven et al., 2003; Armstrong et al., 2005; Feipel et al., 2006; Grip et al., 2007; Sjolander et al., 2008; Woodhouse and Vasseljen, 2008). Seven of those nine studies used the classification of the Quebec Task Force on Whiplash-Associated Disorders (WAD) (Spitzer et al., 1995; Rydevik et al., 2008). In this classification system, WAD grade 1 corresponds to complaints of neck pain, stiffness or tenderness only without physical signs that are noted by an examining physician; WAD grade 2 corresponds to complaints of neck pain and musculoskeletal signs, such as a decreased range of motion and point tenderness in the neck; and WAD grade 3 includes additional signs (decreased or absent deep tendon reflexes, weakness, and sensory deficits). Of these nine studies, four also included a group of people with non-traumatic neck pain (Kristjansson et al., 2003; Grip et al., 2007; Sjolander et al., 2008; Woodhouse and Vasseljen, 2008). The studies that reported both on participants with traumatic and with non-traumatic neck pain are presented in Tables 1 and 2 Another study, described in Table 3, had a combined group consisting of both participants with traumatic and idiopathic neck pain (Chen and Treleaven, 2013).

3.3. Risk of bias

Thirty-six of the included studies remained after the first screening. These 36 studies fulfilled all of the inclusion criteria, based on title and abstract. After the first full-text reading, two researchers agreed on twelve studies. On two studies, the researchers disagreed regarding the validity of the measurement protocol. Another study was subject of discussion with regard to the outcome measure. After a second reading and comparison of the differences, the researchers reached consensus for the three studies. Both conflicting studies regarding the validity of the

Table 2
Evidence table of the included studies which researched participants with non-traumatic neck pain.

Reference	Sample	Inclusion criteria	JPSE testing instrument	JPSE testing protocol	Results
Rix and Bagust (2001)	11 Non Traumatic NP (6M,5W) 41.1 ± 13.3 11 CON (5M,6W) 39.3 ± 10.3	Non traumatic NP: Continuous neck pain > 7Wk, No history of cervical injury or WAD CON: Healthy Non traumatic neck pain and CON matched; age and gender	Laser pointer	Flexion, extension, rotation. Mean of 10 trials Absolute errors Self-selected neutral position Self-selected paceExaminer was not blinded	Non Traumatic NP: Global abs. error median (°) Right rotation = 6.1, Left rotation = 3.7, Flexion = 5.7*, Extension = 4.3 CON: Global abs. error median (°) Right rotation = 6.0, Left rotation = 4.0, Flexion = 4.2, Extension = 5.1
Kristanjansson et al. (2003)	20 Non Traumatic NP (9W, 11M) 30.0 ± 9.1 21 CON (11W, 10M) 26.9 ± 6.4	Non Traumatic NP: Insidious onset of neck pain not < 3 Mo or > 48Mo CON: Healthy	3 Space Fastrak (Polhemus, Inc, Colchester, Vermont, USA) 1 sensor centered on the forehead, 1 sensor on the spinous of C7	Rotation Mean of 3 trials Error in degrees Full active rotation within comfortable limits Selected neutral position	Non traumatic NP: Global abs. error ± SD (°) Rotation = 3.33 ± 1.42* CON: Global abs. error ± SD (°) Rotation = 2.48 ± 1.12
Woodhouse and Vasseljen (2008)	57 Chronic NP (38W, 19M) 43.7 ± 12.6 57 CON (28W, 29M) 38.2 ± 10.9	Chronic NP: not <6 Mo or >10 Yr CON: Healthy	3 Space Fastrak (Polhemus, Inc, Colchester, Vermont, USA) 1 sensor centered on the forehead, 1 sensor placed on the upper part of the wooden backrest above the subject's head.	Rotation Mean of 4 trials Absolute error (largest error was selected) Selected neutral position Self-selected pace Examiner was not blinded	Chronic NP: Global abs. error ± SD (°) Rotation = 3.17 ± 1.1 CON: Global abs. error ± SD (°) Rotation = 2.86 ± 1.2
Sjolander et al. (2008)	9 Insidious NP (0M, 9W) 40 ± 9 16 CON (3M, 13W) 40 ± 9	Insidious NP: Idiopathic neck pain of insidious onset CON: Healthy	3 Space Fastrak (Polhemus, Inc, Colchester, Vermont, USA) 1 sensor centered on the forehead, 1 sensor on the spinous of T1	Rotation Mean of trials unclear Move the head as fast and far as possible Self-selected neutral position Participants were standing. Examiner was blinded	Insidious NP: Over/under shooting ± SD (°) Right rotation = 0.4 ± 0.5, Left rotation = 0.4 ± 0.8 CON: Over/under shooting ± SD (°) Right rotation = 0.1 ± 0.5, Left rotation = 0.1 ± 0.6
Grip et al. (2007)	21 NP (14W, 7M) 49 ± 16 24 CON (16W, 8M) 50 ± 18	NP: not <3 Mo Neck Pain Con: Healthy	ProReflex System (Qualisys Medical AB, Gothenburg, Sweden) 13 markers were placed on the head and upper torso	Rotation, flexion, extension Mean of 5 trials Error in degrees Self-selected neutral position Self-selected pace Only moving back to neutral with closed eyes	NP: Global abs. error ± SD (°) Right rotation = 3.7 ± 1.6, Left rotation = 3.6 ± 3.0 Flexion 2.8 ± 1.2, Extension = 2.9 ± 1.3 CON: Global abs. error ± SD (°) Right rotation = 3.1 ± 1.3, Left rotation = 3.5 ± 1.3 Flexion 2.9 ± 0.9, Extension = 2.7 ± 1.0
Revel et al. (1991)	30 Chronic NP (10M, 20W) 45 (25−73) 30 CON (10M, 20W) 44 (21−72)	Chronic NP: Cervical pain. CON: Healthy	Laser pointer	Flexion, extension, rotation. Mean of 10 trials Absolute errors Self-selected neutral position Self-selected pace. Examiner was not blinded	Chronic NP: Global abs. error ± SD (°) Left-right rotation = 6.11 ± 1.59*, Flexion-extension = 5.47 ± 1.75* CON: Global abs. error ± SD (°) Left-right rotation = 3.50 ± 0.82, Flexion-extension = 3.37 ± 0.73
Cheng et al. (2010)	12 Non traumatic NP (6W,6M) 25.4 ± 2.1 12 CON (7W,5M) 24.9 ± 1.8	Non traumatic NP: not <2 Yr Neck Pain CON: Healthy Non traumatic neck pain and CON matched; age	Electrogoniometer CXTLA02, Crossbow, Inc., San Jose, CA, USA	Flexion, extension Mean of 3 trials Constant error Selected neutral position Self-selected pace	Non traumatic NP: Constant error ± SD (°) Flexion = 7.1 ± 3.5*, Extension = 6.3 ± 4.7* CON: Constant error ± SD (°) Flexion = 3.5 ± 1.8, Extension = 4.2 ± 3.3
Uthaikhup et al. (2012)	20 NP (70%W) 73.2 ± 6.2 20 CON (60%W) 69.6 ± 4.2	NP: >3 Mo neck pain, at least a score 10 or more on the NDI and older than 65Yr CON: Healthy and older than 65Yr	3 Space Fastrak (Polhemus, Inc, Colchester, Vermont, USA) 1 sensor centered on the forehead, 1 sensor on the spinous of C7	Rotation, extension Mean of 3 trials Absolute error Selected neutral position Self-selected pace	NP: Global abs. error ± SD (°) Right rotation = 5.5 ± 3.1, Left rotation = 5.1 ± 4.0, Extension = 5.2 ± 3.4 CON: Global abs. error ± SD (°) Right rotation = 4.2 ± 2.2, Left rotation = 2.8 ± 1.8, Extension = 3.6 ± 2.4

JPSE; Joint position sense error, NP = Neck Pain; CON: Healthy controls; SD = Standard Deviation; M = Men; W = Women; Abs. = Absolute; NDI = Neck Disability Index; Yr = Year; Mo=Month; * = indicates statistically significant differences between groups.

Table 3
Evidence table of the included study with a combined group of participants with traumatic and non-traumatic neck pain.

Reference	Sample	Inclusion criteria	JPSE testing instrument	JPSE testing protocol	Results
Chen and Treleaven (2013)	25 NP (10M, 15W) 39.4 ± 12.8 26 CON (11M, 12W) 31.0 ± 11.9	NP: chronic NP of traumatic or idiopatic origin, not <3 Mo Neck Pain and at least a score 10 or more on the NDI CON: Healthy Av. age of the NP group significantly differs from the CON group (p = 0.02)	3 Space Fastrak (Polhemus, Inc, Colchester, Vermont, USA) 1 sensor centered on the forehead, 1 sensor on the spinous of C7 and one sensor at the mid sternum region Laser Pointer	Rotation Mean of 6 trials Absolute errors Examiner was not blinded "Conventional JPS (conv)" in this protocol the subjects actively move their heads. "JPS trunk to head rotation (tors)" in this protocol the subjects actively move their trunk whilst their head does not move.	NP: Global abs. error ± SD (°) Conv R Fastr 2.79 ± 1.3, Conv L Fastr 3.00 ± 1.1, Conv RL Fastr 2.89 ± 0.9 Conv R Laser 3.08 ± 1.3, Conv L Laser 3.25 ± 1.1, Conv RL Laser 3.21 ± 0.9* Tors R Fastr 2.66 ± 1.3, Tors L Fastr 2.81 ± 0.8*, Tors RL Fastr 2.74 ± 0.7* Tors R Laser 3.31 ± 1.4, Tors L Laser 3.07 ± 1.1*, Tors RL Laser 3.19 ± 1.0* CON: Global abs. error ± SD (°) Conv R Fastr 2.36 ± 0.9, Conv L Fastr 2.83 ± 1.0, Conv RL Fastr 2.58 ± 0.8 Conv R Laser 2.37 ± 1.0, Conv L Laser 2.77 ± 1.1, Conv RL Laser 2.57 ± 0.8 Tors R Fastr 2.17 ± 0.9, Tors L Fastr 2.22 ± 0.9, Tors RL Fastr 2.19, 0.7 Tors R Laser 2.81 ± 1.2, Tors L Laser 2.56 ± 1.0, Tors RL Laser 2.69 ± 0.8

JPSE; Joint position sense error, NP = Neck Pain; CON: Healthy controls; SD = Standard Deviation; M = Men; W = Women; Abs. = Absolute; NDI = Neck Disability Index; Yr = Year; Mo = Month; Fast = Fastrak* = indicates statistically significant differences between groups.

measurement protocol were included. The study that was subject of discussion with regard to the outcome measure was excluded, resulting in 14 included studies.

Methodological quality of all of the included studies was "acceptable" (+) according to the SIGN criteria checklist. This implies some weaknesses in the study, with an associated risk of bias. Most of the studies lost points on "sample size" or "not blinding the assessor".

3.4. Outcome measures

The included studies in this review used JPSE as an outcome measure to reflect proprioception of the cervical spine. The JPSE was described in angular units (degrees) or centimeters to measure the error.

3.5. Traumatic neck-pain

As shown in Table 1, four studies (Heikkilä and Wenngren, 1998; Kristjansson et al., 2003; Sterling et al., 2003; Treleaven et al., 2003) reported that participants with traumatic neck pain had a significantly higher JPSE than healthy controls. Of these four studies, Sterling et al. (2003) reported a significant difference compared to healthy controls on rotation to the right. Rotation to the left and extension were not significantly different from the healthy controls. In the studies of Kristjansson et al. (2003) (rotation), and Heikkilä and Wenngren (1998) (rotation and flexion-extension), all the investigated directions of movement regarding the JPSE were significantly higher in participants with traumatic neck pain. In the study of Treleaven et al. (2003), JPSE in all the investigated directions of movement (right rotation, left rotation, and extension) was significantly higher compared to healthy controls, but only after results from the two different neck pain groups were pooled. Five studies that included participants with neck-pain of a traumatic origin did not show a significantly altered JPSE compared to healthy controls (Armstrong et al., 2005; Feipel et al., 2006; Grip et al., 2007; Sjolander et al., 2008; Woodhouse and Vasseljen, 2008).

3.6. Non-traumatic neck pain

Eight studies were included, involving participants with non-traumatic neck pain (Revel et al., 1991; Rix and Bagust, 2001; Kristjansson et al., 2003; Grip et al., 2007; Sjolander et al., 2008; Woodhouse and Vasseljen, 2008; Cheng et al., 2010; Uthaikhup et al., 2012) as can be seen in Table 2. Of these eight studies, four (Revel et al., 1991; Rix and Bagust, 2001; Kristjansson et al., 2003; Cheng et al., 2010) reported a significantly higher JPSE in people with non-traumatic neck pain than in controls. Joint position sense error in the investigated directions of movement was significantly higher in the studies of Kristjansson et al. (2003) (rotation), Revel et al. (1991) (rotation and flexion-extension) and Cheng et al. (2010) (flexion and extension). For the study of Rix and Bagust (2001), this was not the case. In this study only, the flexion movement was significantly higher than in healthy controls. With respect to right rotation, left rotation and extension, JPSE in participants with neck pain was not significantly different. The studies of Woodhouse and Vasseljen (2008), Sjolander et al. (2008), Grip et al. (2007), and Uthaikhup et al. (2012), did not report any significant differences in JPSE between participants with non-traumatic neck pain and healthy controls.

3.7. Combined group consisting of traumatic and non-traumatic neck pains

As shown in Table 3, Chen and Treleaven (2013) included participants with chronic neck pain with either a traumatic or idiopathic origin. This study used a laser pointer as well as the "Fastrak™" instrument to measure the JPSE. The authors also used two different measurement protocols for measuring the JPSE. In the conventional protocol, participants were asked to actively rotate their heads (left or right) as far as was comfortable, and then had to return to the starting position as accurately as possible. In the alternative protocol, participants had to actively rotate the trunk (instead of the head) and return to the starting position. The chest sensor and the chest laser were used to obtain data on trunk rotation error. As can be seen in Table 3, for the conventional measurement protocol only the pooled JPSE (left/right rotation) significantly differed between participants with neck pain and controls when measured with the laser pointer. The JPSE measured with the "Fastrak™" did not show any significant differences when measured with the conventional protocol.

For the trunk-to-head measurement protocol, left rotation and the pooled left/right rotation significantly differed from the healthy controls. This held for the laser pointer measurement instrument as

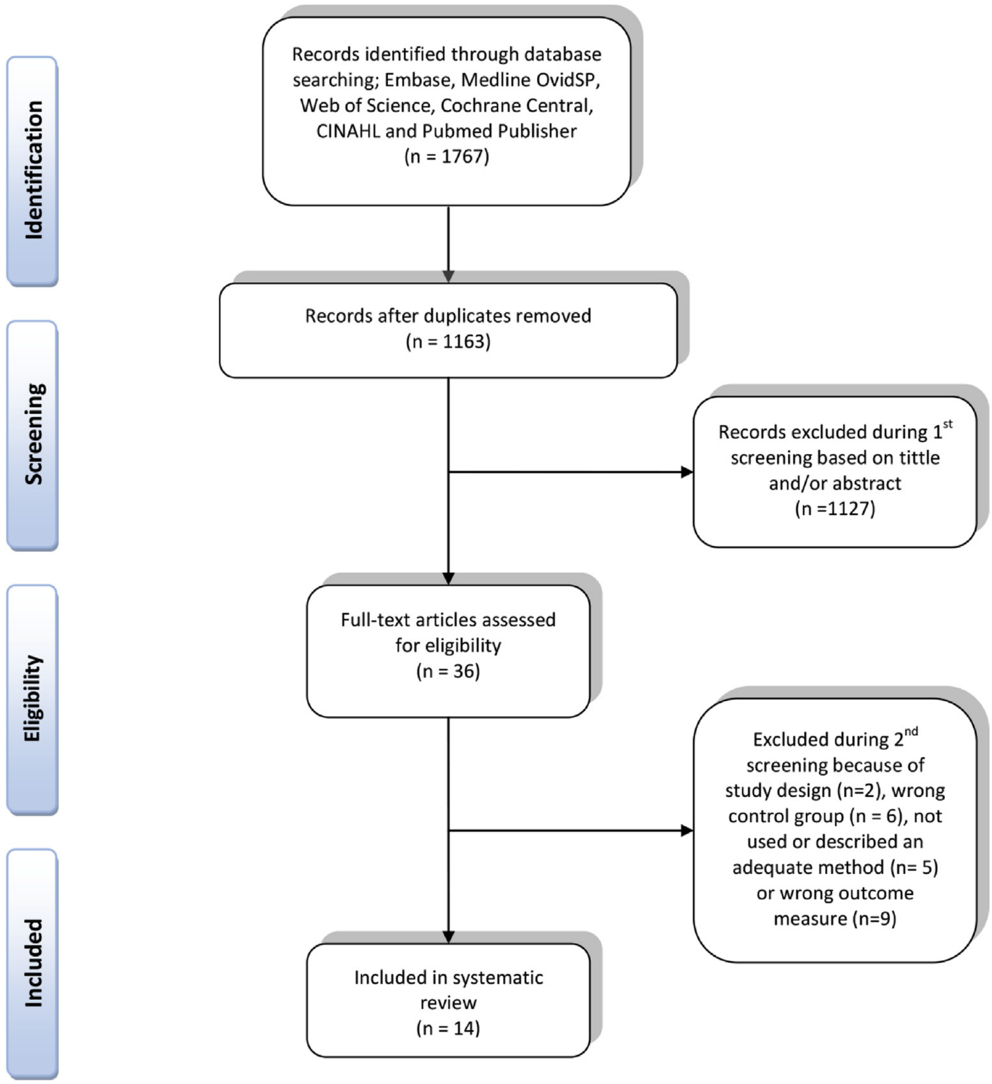

Fig. 1. Flowchart of the literature search and the selection of the studies.

well as for the "Fastrak™" measurement instrument. Rotation to the right was not significantly altered, regardless of measurement instrument or protocol.

4. Discussion

The main goal of this systematic review was to differentiate between JPSE of the cervical spine in participants with neck pain of a traumatic or a non-traumatic origin, compared to healthy controls. The results of this review suggest that when the JPSE is measured over 6 trials or more, the JPSE is generally higher in the neck pain group than in the control group.

Various factors might influence the outcome of the JPSE measurement. The first is the influence of the vestibular system. As the peripheral and central vestibular systems provide and integrate information essential for establishing the position of the head in space, they indirectly influence the head-to-body position sense. Deficits in any of the vestibular mediated pathways may thus affect JPSE (Treleaven, 2008; Chen and Treleaven, 2013). However, Pinsault et al. (2008) and Malmström et al. (2009) did not find an increased JPSE in people with vestibular loss when compared to healthy controls. Because the vestibulum is particularly sensitive to fast, jerky head movements (Day and Fitzpatrick, 2005), the velocity of head motion during measurement of the JPSE is important. When participants move their heads faster than 2.1°/s, cervical input decreases and vestibular input increases (Kelders et al., 2003). Thus, the faster the head moves, the more JPSE represents vestibular afferention rather than cervical afferention. It is not clear whether all the included studies tried to rule out as much afferention from the vestibulum as possible, by having the subjects move slowly. A study by Chen and Treleaven (2013) showed interestingly that trunk-to-head rotation, excluding input of the vestibulum, gave different results compared to the conventional measurement protocol of head-to-trunk rotation. However, as the differences were small, this measurement protocol should be examined further to see whether possible vestibular input plays a role in the conventional measurement protocol.

A second factor that may affect the conclusion is the anatomy of the cervical spine. Large quantities of muscle spindles in the cervical spine muscles provide (Bolton et al., 1998) and receive information from the central nervous system (Kalaska, 1994; Hellström et al., 2005). In the cervical spine, the information from muscles (muscle afferention) is a dominant source of information (Hogervorst and Brand, 1998; Proske and Gandevia, 2012). A study using Magnetic Resonance Imaging has shown a widespread presence of fatty infiltrates in the neck muscles of people with persisting moderate to severe levels of pain following a whiplash injury (Elliott et al., 2011). This implies that the intensity of the perceived pain may influence proprioception. For the traumatic group, the duration of complaints or severity of the WAD did not seem to influence JPSE significantly. In the non-traumatic group, there was no correlation between the duration of the neck pain and an altered JPSE. Likewise, the intensity of perceived pain, which was described in all studies, did not seem to influence the JPSE. In some of the included studies, relatively low perceived pain levels were correlated with significantly altered JPSE, and *vice versa*.

A third factor is the variety of measurement devices used. Some researchers used a laser pointer, where others used either the electromagnetic tracking system 3 Space “Fastrak™” (Polhemus Inc, USA), a ProReflex System (Qualisys Medical AB, Gothenburg, Sweden), or different types of electrogoniometers. This made it difficult to compare the various study results. The 3 Space “Fastrak™” was the most commonly used instrument, employed in eight out of fourteen studies (Kristjansson et al., 2003; Sterling et al., 2003; Treleaven et al., 2003; Armstrong et al., 2005; Sjolander et al., 2008; Woodhouse and Vasseljen, 2008; Uthaikhup et al., 2012; Chen and Treleaven, 2013). The “Fastrak™” system is an electromagnetic measuring instrument that tracks the positions of sensors relative to a source in three dimensions. Previously Jordan et al. (2000). demonstrated that it is a reliable and valid measurement system with an accuracy of up to $\pm 0.2^{\circ}$. The nine studies using it produced contrasting results regarding the JPSE in people with neck pain.

The sensor placement is another possible source of measurement bias. Not all studies used the same placement, or described the placement of the sensors precisely. This inconsistency could have consequences for the validity of the measurements and the ability to compare the different study results.

The laser method, which is also commonly used to assess the JPSE (four out of fourteen studies), has a good test-retest reliability and a strong correlation with an ultrasound technique for measuring JPSE (Roren et al., 2009). It is remarkable that all four studies (Revel et al., 1991; Heikkilä and Wenngren, 1998; Rix and Bagust, 2001; Chen and Treleaven, 2013) using a laser pointer showed significantly higher cervical joint reposition errors in people with neck pain than in controls. However, in none of these four studies were the examiners blinded for “controls” or “participants with neck-pain”. The results in these studies may, therefore, have been influenced by expectation bias. Revel et al. (1991) compared the inter-observer reliability of the laser pointer instrument in 11 controls. This test showed no significant difference between the examiners.

The application of various testing protocols and data-analysis software is a fourth possible factor influencing the conclusion. Swait et al. (2007) reported that at least six trials were needed to optimize the stability and reliability of the cervical JPSE measurement. Nonetheless, in only four of the fourteen studies did the researchers use six or more trials to calculate the mean JPSE. All four studies (Revel et al., 1991; Heikkilä and Wenngren, 1998; Rix and Bagust, 2001; Chen and Treleaven, 2013), in which the mean JPSE was calculated over six or more trials, showed significantly higher joint position errors in people with neck pain than in controls. These studies used a laser pointer as a JPSE testing device. An explanation for this could lie in the applied statistics. It might be that the vulnerability to outliers is less when the mean JPSE is calculated over more trials hereby reducing the standard error of the mean. This stresses the importance of calculating the joint position error over at least six trials. Further research needs to be performed on the effect on learning curves in the presence of pain and/or after (traumatic) damage to the joints of the cervical spine.

The studies with an electronic testing device used custom-made analysis software (Kristjansson et al., 2003; Sterling et al., 2003; Treleaven et al., 2003; Armstrong et al., 2005; Feipel et al., 2006; Sjolander et al., 2008; Woodhouse and Vasseljen, 2008; Uthaikhup et al., 2012; Chen and Treleaven, 2013). As only a general description of the algorithms of this software was given, the reproducibility of these experiments are low. Absence of the presentation of raw data in most studies is in line with the previous point. Only when both the data-analysis protocol and the (raw) data are presented, readers can interpret results and conclusions of the studies. Another threat to the validity and reliability of the included studies is the small number of participants that some of them included (Rix and Bagust, 2001; Sjolander et al., 2008; Cheng et al., 2010). However, studies which have included a relative high number of participants do not show other or more robust results than the studies with a smaller amount of participants.

In general, data cannot be compared without harmonization of testing protocols and data analysis systems. Therefore, it was not possible to conduct a meta-analysis of the included studies. This pooling of data would help to resolve the problem of the small number of participants included in some of the studies. Besides improving the current study designs, it is also important to correlate JPSE with other specific variables (i.e. age, gender, location of perceived pain, anxiety levels, perceived disability, and cervical range of motion).

5. Conclusion

In general, the results of the included studies give an equivocal answer to the question of whether the JPSE is higher in people with cervical spine lesions caused by trauma and/or non-traumatic neck complaints than in controls. The JPSE is overall higher in the neck pain group when measured over at least 6 trials.

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