

# Perceived pain extent is associated with disability, depression and self-efficacy in individuals with whiplash associated disorders

Falla, Deborah; Peolsson, Anneli; Peterson, Gunnel; Landén Ludvigsson, Maria; Soldini, Emiliano; Schneebeli, Alessandro; Barbero, Marco

DOI:  
[10.1002/ejp.873](https://doi.org/10.1002/ejp.873)

License:  
None: All rights reserved

Document Version  
Peer reviewed version

Citation for published version (Harvard):  
Falla, D, Peolsson, A, Peterson, G, Landén Ludvigsson, M, Soldini, E, Schneebeli, A & Barbero, M 2016, 'Perceived pain extent is associated with disability, depression and self-efficacy in individuals with whiplash associated disorders', *European Journal of Pain*. <https://doi.org/10.1002/ejp.873>

[Link to publication on Research at Birmingham portal](#)

## Publisher Rights Statement:

Checked for eligibility: 30/06/2016. This is the peer reviewed version of the following article: "Falla, D., Peolsson, A., Peterson, G., Ludvigsson, M.L., Soldini, E., Schneebeli, A. and Barbero, M. (2016), Perceived pain extent is associated with disability, depression and self-efficacy in individuals with whiplash-associated disorders. *European Journal of Pain*", which has been published in final form at 10.1002/ejp.873. This article may be used for non-commercial purposes in accordance with Wiley Terms and Conditions for Self-Archiving.

## General rights

Unless a licence is specified above, all rights (including copyright and moral rights) in this document are retained by the authors and/or the copyright holders. The express permission of the copyright holder must be obtained for any use of this material other than for purposes permitted by law.

- Users may freely distribute the URL that is used to identify this publication.
- Users may download and/or print one copy of the publication from the University of Birmingham research portal for the purpose of private study or non-commercial research.
- User may use extracts from the document in line with the concept of 'fair dealing' under the Copyright, Designs and Patents Act 1988 (?)
- Users may not further distribute the material nor use it for the purposes of commercial gain.

Where a licence is displayed above, please note the terms and conditions of the licence govern your use of this document.

When citing, please reference the published version.

## Take down policy

While the University of Birmingham exercises care and attention in making items available there are rare occasions when an item has been uploaded in error or has been deemed to be commercially or otherwise sensitive.

If you believe that this is the case for this document, please contact [UBIRA@lists.bham.ac.uk](mailto:UBIRA@lists.bham.ac.uk) providing details and we will remove access to the work immediately and investigate.

# **PERCEIVED PAIN EXTENT IS ASSOCIATED WITH DISABILITY, DEPRESSION AND SELF-EFFICACY IN INDIVIDUALS WITH WHIPLASH ASSOCIATED DISORDERS**

Deborah Falla<sup>1,2</sup>, Anneli Peolsson<sup>3</sup>, Gunnel Peterson<sup>3,4</sup>, Maria Landén Ludvigsson<sup>3,5</sup>, Emiliano Soldini<sup>6</sup>, Alessandro Schneebeli<sup>7</sup>, Marco Barbero<sup>7</sup>

1. Pain Clinic, Center for Anesthesiology, Emergency and Intensive Care Medicine, University Hospital Göttingen, Göttingen, Germany.
2. Institute for Neurorehabilitation Systems, Bernstein Center for Computational Neuroscience, University Medical Center Göttingen, Georg-August University, Göttingen, Germany
3. Department of Medical and Health Sciences, Division of Physiotherapy, Linköping University, Linköping, Sweden
4. Centre for Clinical Research Sörmland, Uppsala University, Uppsala, Sweden
5. Rehab Väst, County Council of Östergötland, Motala, Sweden
6. Department of Business Economics, Health and Social Care, University of Applied Sciences and Arts of Southern Switzerland (SUPSI), Manno, Switzerland.
7. Rehabilitation Research Laboratory, Department of Business Economics, Health and Social Care, University of Applied Sciences and Arts of Southern Switzerland, Manno, Switzerland

**Running Head:** Pain extent in whiplash

**Category:** Original Article

**Declaration:** The authors declare no conflict of interest. Not supported by external funding.

## **What this study adds:**

- Women with chronic WAD, those with unsettled insurance claims and those with poorer financial status perceive more widespread pain.
- When controlling for these factors, larger pain areas remain associated with perceived pain and disability, depression and self-efficacy.
- The pain drawing is useful to support psychological screening in people with chronic WAD.
- 

## **Address for correspondence:**

Prof. Dr. Deborah Falla

Center for Anesthesiology, Emergency and Intensive Care Medicine,  
University Hospital Göttingen,

Robert-Koch-Str. 40,

37075, Göttingen,

Germany

Tel: +49 (0) 551 / 3920109

Fax: + 49 (0) 551 / 3920110

Email: [deborah.falla@bccn.uni-goettingen.de](mailto:deborah.falla@bccn.uni-goettingen.de)

## **ABSTRACT**

**Background:** Completion of a pain drawing is a familiar task in those presenting with whiplash associated disorders (WAD). Some people report pain almost over their entire body. Yet the reasons for larger pain extent has not been fully explored.

**Methods:** A novel method was applied to quantify pain extent from the pain drawings of 205 individuals with chronic WAD. Pain extent was evaluated in relation to sex, age, educational level, insurance status and financial status. Multiple linear regression analysis was used to verify whether pain extent was associated with other health indicators including perceived pain and disability, health-related quality of life, pain catastrophizing, anxiety, depression and self-efficacy.

**Results:** Pain extent was influenced by sex ( $\chi^2$ :10.392,  $p<0.001$ ) with larger pain extent in women compared to men ( $7.88\pm 7.66\%$  vs  $5.40\pm 6.44\%$ ). People with unsettled insurance claims ( $\chi^2$ : 7.500,  $p<0.05$ ) and those with a worse financial situation ( $\chi^2$ :12.223,  $p<0.01$ ) also had larger pain extent. Multiple linear regression models revealed that, when accounting for age, sex, education, insurance status, financial status and neck pain intensity, pain extent remained associated with perceived disability ( $p<0.01$ ), depression ( $p<0.05$ ), and self-efficacy ( $p<0.001$ ).

**Conclusion:** By utilizing a novel method for pain extent quantification this study shows that widespread pain is associated with a number of factors including perceived disability, depression and self-efficacy in individuals with chronic WAD. Widespread pain should alert the clinician to consider more specific psychological screening, particularly for depression and self-efficacy, in patients with WAD.

**Keywords:** whiplash associated disorders, chronic pain, widespread pain, pain drawing

### **What's already known about this topic?**

- Completion of a pain drawing is a familiar task in those presenting with whiplash associated disorders (WAD).
- Some report widespread pain affecting almost their entire body.
- No studies have evaluated the relation of pain extent to perceived pain and disability, financial status, general health and psychological status in individuals with chronic WAD.

### **What does this study add?**

- Women with chronic WAD, those with unsettled insurance claims and those with poorer financial status perceive more widespread pain.
- When controlling for these factors, larger pain areas remain associated with perceived pain and disability, depression and self-efficacy.
- The pain drawing is useful to support psychological screening in people with chronic WAD.

## INTRODUCTION

Whiplash associated disorders (WAD) are a complex, multifaceted disorder associated with significant social and economic costs (Carroll et al., 2008). Over fifty percent of individuals will continue to report symptoms six months after the initial injury (Carroll et al., 2008).

People with chronic WAD report a myriad of symptoms besides pain including stiffness, dizziness, headache, sleep disturbances, visual impairments, nausea and difficulty swallowing (Crutebo et al., 2010; Sterling 2011). In addition, the physical and psychological features associated with chronic WAD are multiple. Motor disturbances include altered muscle behavior and impaired movement (Dall'Alba et al., 2001; Peterson et al., 2015; Schomacher et al., 2012; Woodhouse and Vasseljen 2008) and psychological factors include varying levels of distress, anxiety and depression (Kongsted et al., 2008; Sterling et al., 2003; Williamson et al., 2008). Many people with chronic WAD show widespread pain which may reflect central sensitization (Curatolo and Sterling 2011; Van Oosterwijck et al., 2013). Several studies have documented widespread hypersensitivity (Curatolo et al., 2001; Lemming et al., 2012) and hyperexcitable spinal cord reflexes (Banic et al., 2004; Lim et al., 2011) in people with chronic WAD. Mechanisms for post injury central hyperexcitability may be related to a number of mechanisms but regardless of the mechanism, these changes have the potential to amplify the individual's pain experience and can contribute to persistent pain (Curatolo et al., 2004; Curatolo and Sterling 2011).

Widespread pain can be observed from the patient's pain drawing. The patient is asked to report their pain by coloring a template representing the human body in different views. Some studies suggest that the pain drawing can be a useful indicator of pain severity (George et al., 2007) or psychological status (Hayashi et al., 2015), yet other studies show limited value of the

pain drawing in predicting a patient's psychological profile (Barbero et al., 2015; Carnes et al., 2006). Such contradiction is likely attributed to the different patient populations examined where some may not have significant psychological disturbances (Barbero et al., 2015) but the discrepancy may also be related to the method of analysis of the pain drawing. Most methods described rely on an operator to classify the pain drawing as "organic" or "nonorganic" and use scoring systems which have not been validated (Dahl et al., 2001) or have low sensitivity (Pande et al., 2005) such as the penalty point system (Ransford et al., 1976).

In chronic WAD, the patients perceived pain extent could potentially be influenced by a number of factors including their level of pain catastrophizing and level of depression (e.g. larger pain areas in those with a tendency to magnify the threat value of pain and to feel helpless in the context of pain) or even their compensation or financial status (e.g. larger pain area if currently in the process of litigation).

Pain extent quantification should be investigated as a potential useful tool to broaden our understanding of the presentation of individuals with chronic WAD as this may assist in diagnosis and treatment decision making. Here we analyze pain extent in a cohort of individuals with chronic WAD and evaluate the relation between pain extent and a number of variables including perceived pain and disability, financial status, general health and psychological status.

## **METHODS**

### **Participants**

The participants included in this observational study formed part of a sample of patients included in a randomized controlled trial (Ludvigsson et al., 2014). The pain drawing analysis was not included in the published trial and is unique to this study. A total of 205 individuals

with chronic WAD participated in the study, including 133 (65%) women and 72 (35%) men with a mean age of 40.1 years (range, 18 to 63 years, SD 11.4 years).

Inclusion criteria were a whiplash injury in the preceding 6 to 36 months that was nominated as the cause of current symptoms, classification as WAD grade 2 or 3 (Spitzer et al., 1996), aged between 18 and 63 years, Neck Disability Index (NDI) score of at least 10/50 points (Vernon and Mior 1991), and an average neck pain intensity the preceding week  $>20/100$  mm on a Visual Analogue Scale (VAS) (0=no pain, 100=worst imaginable pain) (Carlsson 1983). Participants were excluded if they reported signs of traumatic brain injury in connection to the whiplash injury, previous neck trauma, myelopathy, spinal infection, previous neck surgery, malignant disease, severe psychiatric disorders, neurological diseases or drug abuse. Participants were also excluded if they reported a more dominant pain elsewhere in the body or if they reported having neck pain which led to  $>1$  month's work absence in the preceding year before the whiplash injury. Insufficient competence of the Swedish language was also an exclusion criteria.

Potential participants were identified from health care registers of six Swedish counties, including primary health care centers, specialist orthopedic clinics, and hospital outpatient services. A four-step process was applied to screen participants for eligibility as illustrated in Figure S1. In summary this procedure involved: (1) an initial letter, including a prepaid return envelope, was sent to 7950 participants which detailed the study information, inclusion/exclusion criteria and included a visual analogue scale (VAS) measure of neck pain intensity for the preceding week; (2) a telephone interview; (3) review of medical records if there was any ambiguity regarding the patients' medical history; (4) physical examination by an experienced physiotherapist (mean of 18 years' experience) to classify the disorder as either a WAD grade 2

(neck pain and clinical findings) or WAD grade 3 (addition of neurological signs) (Spitzer et al., 1996). The presence of neurological signs was confirmed if the patient described arm pain or paresthesia without other known causes, together with at least two positive examination findings indicating neurological deficit in the same dermatome/myotome (Bertilson et al., 2007). Figure S1 indicates the reasons for exclusion at each step including the further exclusion of 11 participants since their pain drawings could not be processed (poor shading of the pain area).

Participants were recruited between February 2011 and May 2012 and all participants received verbal and written information about the study. Informed written consent was obtained from all participants. The study was approved by the Regional Ethics Committee of Linköping, Sweden and was conducted according to the Declaration of Helsinki.

### *Questionnaires*

Patients completed a generic questionnaire which detailed their sex, age, time since the injury occurred, previous history of neck pain, previous neck trauma, cause of the whiplash injury, whether or not they were a smoker, education level, use of analgesics, employment status, marital status, financial status, and whether or not they had made an insurance claim related to their whiplash injury. Moreover, patients completed a symptom frequency questionnaire where they were asked to rate the frequency of neck pain, arm pain, headache, paresthesia, stiffness, difficulties lifting their arms, dizziness, sleep disturbances, visual impairment, hearing impairment, difficulties swallowing, nausea, concentration difficulties and jaw problems on a five point scale: never, from time to time, every day, several times a day, constantly.



## *Pain*

Pain was measured using a VAS anchored by 0=no pain, and 100=worst imaginable pain (Carlsson 1983). Patients were asked to complete a VAS for their average pain intensity, and when their neck pain was at its worst and its best. In addition, pain bothersomeness was quantified, recorded for the preceding 24 hours with the VAS anchored by 0=not bothersome at all and 100=extremely bothersome. Pain bothersomeness has been shown to be a useful measure in individuals with WAD (Stewart et al., 2007).

## *Disability*

The NDI (Vernon and Mior 1991) was used to assess pain-related disability specifically related to neck pain. The NDI consists of 10 items grading neck disability from 0 (no activity limitations) to 5 (major activity limitations) with a total maximum score of 50 points which is then expressed as a percentage (0-100 %) with a higher score representing a higher level of disability. The NDI is widely used, and is a reliable (intraclass correlation coefficient up to 0.98) and valid measurement of disability in neck pain disorders (MacDermid et al., 2009). The Pain Disability Index (PDI) was also used which assess the degree to which chronic pain interferes with various daily activities. Seven categories are coded from 0=no disability to 10= the activities in which you would normally be involved have been totally disrupted or prevented by pain. The reliability and validity of the PDI has also been established (Tait et al., 1990).

## *General Health and Psychological Aspects*

Health-related quality of life was quantified using the EuroQol Five Dimension Scale (EQ-5D, 243 possible health states converted to a single index value  $-0.594$  to 1, where 1 is

perfect health) and EuroQol VAS (0–100 representing worst to best imaginable health state respectively) (Brooks 1996).

The participants' confidence in their ability to perform activities despite their pain was evaluated using the Self-Efficacy Scale (SES). SES is a reliable instrument in WAD populations consisting of 20 different physical and psychosocial activity items (from 0=not confident at all, to 10=very confident), thus generating a total score from 0 to 200 (Kall 2009). The Pain Catastrophizing Scale (score range 0–52) provides an indication of individuals who ruminate, magnify or feel helpless about controlling their pain (Sullivan et al., 1995). A higher score reflects greater negative pain-related thoughts, greater emotional distress, and greater pain intensity. The Hospital Anxiety and Depression Scales (HADS) were used to detect depression and anxiety and their role in the manifestation of somatic symptoms. There are 7 items, which produce a cumulative score of 0–21 for the anxiety (HADS-A) and depression (HADS-D) subscales with a higher score indicative of greater anxiety and depression (Bjellanda et al., 2002). The 11-item Tampa Scale of Kinesiophobia (score range 11–44) evaluated fear of movement with higher scores indicating greater fear of movement (Roelofs et al., 2007).

#### *Work-related factors*

Work stress was measured with the Effort-Reward Imbalance Scale (Siegrist et al., 2004). It is comprised of different dimensions scored on a 4-point Likert scale and an overall Effort-Reward Imbalance score. A value close to zero indicates a favorable condition (relatively low effort, relatively high reward), whereas values beyond 1.0 indicate a high amount of effort spent that is not met by the rewards received or expected in turn (Siegrist et al., 2004).

### *Pain drawings*

All participants were requested to complete a pain drawing indicating their pain on two body charts, one reporting a front view of the body and one a dorsal view (the body chart templates are illustrated in Figure 2). Body charts were printed on A4 sheet and patients were instructed to color, using a pencil, every part of the body where they perceived pain, independently from the type and the severity of pain.

Afterwards all pain drawings were digitalized using commercial scanner and imported in an image analysis software (Inkscape version 0.48). Two operators superimposed the imported body charts with a transparent digital body charts with the same size and characteristics. Pain drawings were then encircled and copied on the digital body charts using the region of interest tool/function. The described procedure to digitalize pain drawings was previously described and its reliability was confirmed (Barbero et al., 2015). This novel method for pain extent quantification is automated and not dependent on interpretation by an operator.

The automatic computation of pain extent and pain location was performed with custom software developed with Matlab® as described previously (Barbero et al., 2015). The software generates the number of shaded pixels from the pain drawing and exports this data which is defined as pain extent. Only pixels colored inside the body chart perimeter was considered. Pain extent for each subject was reported as the sum of the pixels in the frontal and in the dorsal body chart, and expressed as the percentage of total body chart area (i.e. a total of 381151 pixels, ventral: 191823 pixels, dorsal: 189328). In addition, pain frequency maps were generated which consisted of all of the pain drawings superimposed and analyzed simultaneously solely for the purpose of illustrating where pain is most frequently perceived across the entire cohort. This was

performed for the dorsal and ventral view. The custom software also performs a pain location analysis according to the body region affected as proposed by Margolis et al. (Margolis et al., 1986). The presence of the pain in a body region was confirmed when the pain drawing involved at least 10% of the body region area or where the number of pixels was greater than 60. The features extracted from the pain drawings are summarized below:

- 1) *Pain extent*: Each individual pain drawing was analyzed to quantify the pain extent. Pain extent for each participant was reported as the sum of the pixels in the frontal and in the dorsal body chart, and expressed as the percentage of total body chart area. Pain extent for each participant was retained for statistical analysis.
- 2) *Pain frequency maps*: The pain drawings from all participants were superimposed to illustrate where the subjects most frequently report pain. A colour grid indicates the percentage of individuals that report pain in a specific area.
- 3) *Pain location*: The body chart was divided into regions and the percentage of participants reporting pain in specific defined body regions is presented.

## **Statistical analysis**

Descriptive statistics were used to describe the participant characteristics, their symptom characteristics and their overall health status considering pain, work, disability, general health and psychological factors. Pain frequency maps and the pain location analysis were also generated for descriptive purposes only to illustrate the most frequently painful regions across the entire sample.

The Kruskal-Wallis test was used to verify if the value of pain extent significantly changed according to sex, age, educational level, insurance status and financial status at the

bivariate level. We opted for the Kruskal-Wallis test instead of the ANOVA because of the non-normality of the pain extent distribution. Then, multiple linear regression analysis was used to verify whether pain extent (i.e. the independent variable of interest) is significantly associated with the other health indicators considered (i.e. the dependent variables of the regression models) when accounting for sex, age, educational level, insurance status, financial status and pain intensity (i.e. the independent control variables). Data were analysed with SPSS Version 22.0 (IBM Corp., Armonk, NY, USA). Statistical significance was set at  $p < 0.05$ .

## **RESULTS**

Table S1 presents the characteristics of the sample including their sex, age, time since the injury occurred, previous history of neck pain, previous neck trauma, cause of the whiplash injury, WAD grade, whether or not they were a smoker, education level, use of analgesics, employment status, marital status, financial status. The majority were women, attributed their WAD to a motor vehicle accident and had WAD grade 2. Table S2 presents the symptom characteristics of the study sample and Table 3 their general health status.

The Kruskal-Wallis test (Table 4) indicated that the variable pain extent was influenced by the sex of the participant ( $p < 0.01$ ). The mean score of pain extent was higher for women compared to men ( $7.88 \pm 7.66$  vs  $5.40 \pm 6.44$ ). The insurance status was also a factor influencing pain extent ( $p < 0.05$ ); people who had not settled their insurance claim yet showed the highest score on pain extent ( $10.44 \pm 11.30$ , which is more than twice the pain extent score of people who did not make an insurance claim). Finally, pain extent scores were higher in those with a worse financial situation ( $p < 0.01$ ); people with a very good financial situation had a mean score of

4.64±4.68 versus 9.40±7.82 for people with a rather bad financial situation. Age and education level did not have a significant influence on pain extent.

Multiple linear regression models (Tables 5 and 6) showed that, when accounting for all control variables (age, sex, education, insurance status, financial status and pain intensity), there was a statistically significant association between pain extent and the NDI score ( $p < 0.01$ ). A one point increase in pain extent was associated with an average increase of 0.304 points on the NDI (95% Confidence Interval: 0.102 – 0.506). The HADS depression score was significantly associated with pain extent ( $p < 0.05$ ); a one point increase in pain extent was associated with an average increase of 0.076 points on the HADS depression score (95% Confidence Interval: 0.001 – 0.151). A one point increase in pain extent was associated with an average decrease of 0.945 points on the SES score ( $p < 0.01$ ; 95% Confidence Interval: -0.288 – -1.602) and a one point increase in pain extent was associated with an average increase of 0.333 points on the PDI score ( $p < 0.01$ ; 95% Confidence Interval: 0.091 – 0.575). Pain extent was not associated with the EQ-5D health score, the HADS anxiety score, the TSK score, or the PCS score. Two regression models were fitted for the VAS of pain bothersomeness, one with and one without the variable of VAS for neck pain intensity. The pain extent variable was no more statistically significant when we introduced VAS for neck pain intensity in the model, indicating that if we control for pain intensity pain extent is not significantly associated with the VAS pain bothersomeness score.

Figure 2 illustrates the pain frequency maps for the full sample of participants included in the study whereas Figure 3 illustrates the perceived painful regions of the body.

## **DISCUSSION**

The patients evaluated in this study were representative of people with chronic WAD. They displayed a wide range of symptoms including neck pain, headache, paresthesia, arm pain, dizziness and visual and hearing impairments. The majority were women with an average age of forty years and most had WAD grade 2. Eighty percent were involved in the process of litigation or had already settled their insurance claim. The scores on the various questionnaires of general and psychological health indicated that their health was not optimal.

The individual pain drawings showed large variability between participants yet collectively, as seen from the pain frequency maps presented in Figure 2, their pain covered almost the entire body. Nevertheless, both the ventral and dorsal pain frequency maps clearly indicate the typical pain pattern for people with WAD with the neck and shoulder regions most frequently affected. The pain location analysis (Figure 3) further illustrated that the neck, shoulder region and head were the most frequently affected regions. In contrast, substantially less people reported pain in their lower limbs or abdomen region, although these regions were painful for some.

We show that pain extent in people with chronic WAD is influenced by sex, insurance claim status, and financial status. In the observed cohort, woman reported a significantly higher pain extent with respect to men, and patients with a poor financial situation and unsettled compensation reported higher pain extent values. When accounting for age, sex, education, insurance status, financial status and pain intensity, pain extent remained significantly associated with perceived pain and disability (NDI and PDI score), depression (HADS depression score), and self-efficacy (SES score). Overall these results indicate that the pain drawing may be useful

to support psychological screening in people with chronic WAD. Body charts which contain large pain extent or show that many body regions are affected should alert the clinician to further specific psychological assessment.

There remains uncertainty of the value of pain drawings as an indicator of the psychological status of the patient. A recent systematic review (Carnes et al., 2006) identified only three studies out of nineteen studies where a definite association between pain drawings and psychological state was identified. Six articles found a positive association between pain drawing scores and psychological data, with the authors suggesting possible clinical utility whereas the remaining ten studies concluded that the statistical association between the pain drawing and the psychological test was too weak to use the pain drawing as a diagnostic psychological screening tool. The vast majority of these studies were conducted in people with chronic low back pain and many suffered from poor study sampling and size (Carnes et al., 2006). Moreover the majority of these studies used methods to extract and define the pain area which were dependent on scoring from an evaluator thus methods which are unreliable (Carnes et al., 2006).

Surprisingly the evaluation of pain drawings in people with chronic WAD is almost non-existent. Yet psychological distress (including affective disturbances, anxiety, and depression) is common in individuals with chronic WAD (Kasch et al., 2013; Sterling et al., 2003; Williamson et al., 2008) thus it is an appropriate population to evaluate the relation between pain extent psychological features. Vernon and colleagues (Vernon et al., 2010) found that the pain area (evaluated using a grid system and modified “Ransford scores”) showed small but significant correlations with NDI and “pain severity” scores in people with chronic WAD. The authors concluded that partial support was obtained for the use of the pain drawing in evaluating the



“nonorganic pain behavior of chronic WAD patients” (Vernon et al., 2010). It should be noted that the only factors examined in this study were age, sex, NDI score, VAS for pain intensity and TSK score.

In a more recent study, the incidence of widespread pain was evaluated in a sample of people six weeks after motor vehicle collision (McLean et al., 2014). Subjects were divided into those engaged in litigation ("litigants") and those not engaged in litigation ("nonlitigants"). This study adopted a method whereby pain extent was assessed in 19 discrete body regions evaluated in the regional pain scale and in the head region and individuals reporting  $\geq 7$  bodily regions of pain were defined as having widespread pain. Compared to individuals who were not engaged in litigation, study participants who hired a lawyer had a greater extent of pain.

The current study also showed that pain extent is influenced by the insurance claim status of the patient. Moreover, their financial status was also associated to their pain extent. That is, people who had not settled their insurance claim yet and/or had a worse financial situation showed the highest scores of pain extent. Compensation seeking has long been believed by some to be a dominant factor in complaints of persistent pain after a whiplash injury (McLean et al., 2014) and the ongoing debate regarding the role of compensation has led to WAD being described as "one of the most controversial conditions in medicine" (Carette 1994; McLean et al., 2014). However, when accounting for age, insurance status and financial status together with other factors including pain intensity, sex and education, pain extent remained significantly associated with perceived pain and disability, depression, and self-efficacy. Thus regardless of the patient's litigation or financial status, widespread pain can still provide an indication of

higher perceived pain and disability and psychological status (specifically depression and self-efficacy).

An association between pain catastrophizing and pain extent was not observed. In contrast, Hayashi et al. (Hayashi et al., 2015) indicated that the presence of “organic” versus “non-organic” pain (evaluated from the pain drawing) was significantly associated with pain catastrophizing in people with neck-shoulder pain. In the current study pain catastrophizing was however associated with higher neck pain intensity (VAS). Likewise, pain-related fear was associated with higher neck pain intensity (VAS) but not pain extent. The EQ-5D score, a measure of health-related quality of life, was not associated to pain extent. This questionnaire includes different dimensions which evaluate mobility, self-care, usual activities, pain/discomfort and anxiety/depression thus it may be that the overall score is too general to be associated to pain extent. Specific subscores e.g. for anxiety/depression may be more relevant to consider. However this was captured in the HADS questionnaire. We cannot rule out that the level of general health measured by different questionnaires, e.g. the Short Form Health Survey (SF-36), is not associated to pain extent. The SF-36 score has been associated with pain extent in people with chronic low back pain (Dahl et al., 2001).

Pain bothersomeness has been shown to be a useful measure in individuals with WAD (Stewart et al., 2007). Although pain bothersomeness was significantly associated with pain extent, when we controlled for their average pain intensity, pain extent was no longer significantly associated with pain bothersomeness.

### *Clinical considerations*

There is clear evidence that psychological factors are associated with increased risk of developing chronic pain and these may include depression, anxiety, pain catastrophizing and fear avoidance beliefs (Chou and Shekelle 2010; Sullivan et al., 2011; Thomas et al., 1999).

Psychological status is an important factor for the prognosis of patients with WAD (Carstensen et al., 2008; Myrtveit et al., 2013; Sterling et al., 2005; Williamson et al., 2015) and psychological factors can mediate physiological changes and function in chronic WAD (Carriere et al., 2015; Vernon et al., 2013). The current results suggest that widespread pain drawn on a body chart may alert the clinician to consider more specific psychological screening, particularly for depression and self-efficacy, in patients with WAD to evaluate the need of targeted interventions that address psychological health.

### *Methodological Considerations*

A strength of this study is that the pain drawings were analyzed without any intervention from an investigator, which likely improves the accuracy and the validity of analysis. The software used to estimate pain extent and location eliminates estimation errors (i.e. software is a deterministic system in which no randomness is involved) which potentially occur with visual-subjective scoring methods.

We studied a large subject sample, with variation of sex, age, education level, financial status, insurance claim status as well as variation of symptom characteristics and psychological status. Chronic WAD is a complex disorder and this can be appreciated by reviewing the symptoms reported in the current cohort. Pain is just one of the many symptoms that people with

chronic WAD report. Thus the results can likely be generalized to wider populations of people with chronic WAD. However, we have only studied a Swedish population which may limit the generalization of findings to other cultures. Based on reports of limited evidence of chronic WAD in some countries such as Lithuania (Obelieniene et al., 1999) and Singapore (Balla 1982), some suggested that chronic WAD is a culturally constructed illness behavior (Ferrari 2003). However, recent work showed the same findings of motor dysfunction, sensory hypersensitivity, and impaired conditioned pain modulation in Singaporeans with chronic WAD which seems to contradict the notion of chronic WAD being culturally dependent (Ng et al., 2014).

We examined a set of specific questionnaires which were included in the original study design of this project (Ludvigsson et al., 2014). It cannot be excluded that scores on other questionnaires such as the Impact of Events Scale (Horowitz et al., 1979) which measures the level of post-traumatic stress reaction (a prognostic factor of poor outcome in WAD (Kongsted et al., 2008; Sterling et al., 2005)) or the Central Sensitization Inventory (Mayer et al., 2012) could show an even stronger relation to pain extent in chronic WAD. Quantitative sensory tests were not included in this study. However it may be relevant in future studies to evaluate the association between pain extent and quantitative measures of central sensitization e.g. local and widespread thermal or mechanical hyperalgesia.

### *Conclusion*

Women with chronic WAD, those with unsettled insurance claims and those with poorer financial status perceive more widespread pain. When controlling for these factors, larger pain areas remained associated with perceived pain and disability, depression and self-efficacy. The pain drawing may be useful to support psychological screening in people with chronic WAD.

**Declaration:** The authors declare no conflict of interest. Not supported by external funding.

**Contributors:** DF, AP, MB contributed to the conception and design of the study. GP and MLL collected the data. AS, MB and DF analysed the data. ES performed the statistical analysis of the data. DF wrote the first draft of the paper. All authors contributed to the interpretation of findings, revising the manuscript for important intellectual content, and approved the final version to be published. All authors had full access to all of the data (including statistical reports and tables) in the study and can take responsibility for the integrity of the data and the accuracy of the data analysis.

## REFERENCES

- Balla JI. The late whiplash syndrome: a study of an illness in Australia and Singapore. *Cult Med Psychiatry* 1982;6: 191-210.
- Banic B, Petersen-Felix S, Andersen OK, Radanov BP, Villiger PM, Arendt-Nielsen L, Curatolo M. Evidence for spinal cord hypersensitivity in chronic pain after whiplash injury and in fibromyalgia. *Pain* 2004;107: 7-15.
- Barbero M, Moresi F, Leoni D, Gatti R, Egloff M, Falla D. Test-retest reliability of pain extent and pain location using a novel method for pain drawing analysis. *Eur J Pain* 2015;19: 1129-1138.
- Bertilson B, Grunnesjo M, Johansson SE, Strender LE. Pain drawing in the assessment of neurogenic pain and dysfunction in the neck/shoulder region: inter-examiner reliability and concordance with clinical examination. . *Pain Med* 2007;8: 134-146.
- Bjellanda I, Dahlb A, Haugc T, Neckelmann D. The validity of the Hospital Anxiety and Depression Scale, an updated literature review. . *J Psychosom Res* 2002;52: 69-77.
- Brooks R. EuroQol: the current state of play. *Health Policy* 1996;37: 53-72.
- Carette S. Whiplash injury and chronic neck pain. *N Engl J Med* 1994;330: 1083-1084.
- Carlsson AM. Assessment of chronic pain. I. Aspects of the reliability and validity of the visual analogue scale. *Pain* 1983;16: 87-101.
- Carnes D, Ashby D, Underwood M. A systematic review of pain drawing literature: should pain drawings be used for psychologic screening? *Clin J Pain* 2006;22: 449-457.
- Carriere JS, Thibault P, Milioto M, Sullivan MJ. Expectancies Mediate the Relations Among Pain Catastrophizing, Fear of Movement, and Return to Work Outcomes After Whiplash Injury. *J Pain* 2015;16: 1280-1287.

- Carroll LJ, Holm LW, Hogg-Johnson S, Cote P, Cassidy JD, Haldeman S, Nordin M, Hurwitz EL, Carragee EJ, van der Velde G, Peloso PM, Guzman J. Course and prognostic factors for neck pain in whiplash-associated disorders (WAD): results of the Bone and Joint Decade 2000-2010 Task Force on Neck Pain and Its Associated Disorders. *Spine (Phila Pa 1976)* 2008;33: S83-92.
- Carstensen TB, Frosthalm L, Oernboel E, Kongsted A, Kasch H, Jensen TS, Fink P. Post-trauma ratings of pre-collision pain and psychological distress predict poor outcome following acute whiplash trauma: a 12-month follow-up study. *Pain* 2008;139: 248-259.
- Chou R and Shekelle P. Will this patient develop persistent disabling low back pain? *JAMA* 2010;303: 1295-1302.
- Crutebo S, Nilsson C, Skillgate E, Holm LW. The course of symptoms for whiplash-associated disorders in Sweden: 6-month followup study. *J Rheumatol* 2010;37: 1527-1533.
- Curatolo M, Arendt-Nielsen L, Petersen-Felix S. Evidence, mechanisms, and clinical implications of central hypersensitivity in chronic pain after whiplash injury. *Clin J Pain* 2004;20: 469-476.
- Curatolo M, Petersen-Felix S, Arendt-Nielsen L, Giani C, Zbinden AM, Radanov BP. Central hypersensitivity in chronic pain after whiplash injury. *Clin J Pain* 2001;17: 306-315.
- Curatolo M and Sterling M. Pain-processing mechanisms in whiplash associated disorders. In: *Whiplash: evidence base for clinical practice*. Australia: Elsevier; 2011.
- Dahl B, Gehrchen PM, Kiaer T, Blyme P, Tøndevold E, Bendix T. Nonorganic pain drawings are associated with low psychological scores on the preoperative SF-36 questionnaire in patients with chronic low back pain. *Eur Spine J* 2001;10: 211-214.

Dall'Alba P, Sterling M, Treleaven J, Edwards S, Jull G. Cervical range of motion discriminates between asymptomatic persons and those with whiplash. *Spine* 2001;26: 2090-2094.

Ferrari R. Myths of whiplash. *Surgeon* 2003;1: 99-103.

George SZ, Bialosky JE, Wittmer VT, Robinson ME. Sex differences in pain drawing area for individuals with chronic musculoskeletal pain. *J Orthop Sports Phys Ther* 2007;37: 115-121.

Hayashi K, Arai YC, Morimoto A, Aono S, Yoshimoto T, Nishihara M, Osuga T, Inoue S, Ushida T. Associations between pain drawing and psychological characteristics of different body region pains. *Pain Pract* 2015;15: 300-307.

Horowitz M, Wilner N, Alvarez W. Impact of Event Scale: a measure of subjective stress. *Psychosom Med. Psychosom Med* 1979;41: 209-218.

Kall LB. Psychological determinants of quality of life in patients with whiplash associated disorders, a prospective study. *Disabil Rehabil* 2009;31: 227-236.

Kasch H, Kongsted A, Qerama E, Bach FW, Bendix T, Jensen TS. A new stratified risk assessment tool for whiplash injuries developed from a prospective observational study. *BMJ OPen* 2013;3: e002050.

Kongsted A, Bendix T, Qerama E, Kasch H, Bach FW, Korsholm L, Jensen TS. Acute stress response and recovery after whiplash injuries. A one-year prospective study. *Eur J Pain* 2008;12: 455-463.

Lemming D, Graven-Nielsen T, Sørensen J, Arendt-Nielsen L, Gerdle B. Widespread pain hypersensitivity and facilitated temporal summation of deep tissue pain in whiplash associated disorder: an explorative study of women. *J Rehabil Med* 2012;44: 648-657.



- Lim E, Sterling M, Stone A, Vicenzino B. Central hyperexcitability as measured with nociceptive flexor reflex threshold in chronic musculoskeletal pain: a systematic review. *Pain* 2011;152: 1811-1820.
- Ludvigsson ML, Peterson G, O'Leary S, Dedering Å, Peolsson A. The Effect of Neck-specific Exercise With, or Without a Behavioral Approach, on Pain, Disability, and Self-Efficacy in Chronic Whiplash-associated Disorders: A Randomized Clinical Trial. *Clin J Pain* 2014;31: 294-303.
- MacDermid JC, Walton DM, Avery S, Blanchard A, Etruw E, McAlpine C, Goldsmith CH. Measurement properties of the neck disability index: a systematic review. *J Orthop Sports Phys Ther* 2009;39: 400-417.
- Margolis RB, Tait RC, Krause SJ. A rating system for use with patient pain drawings. *Pain* 1986;24: 57-65.
- Mayer TG, Neblett R, Cohen H, Howard KJ, Choi YH, Williams MJ, Perez Y, Gatchel RJ. The development and psychometric validation of the central sensitization inventory. *Pain Pract* 2012;12: 276-285.
- McLean SA, Ulirsch JC, Slade GD, Soward AC, Swor RA, Peak DA, Jones JS, Rathlev NK, Lee DC, Domeier RM, Hendry PL, Bortsov AV, Bair E. Incidence and predictors of neck and widespread pain after motor vehicle collision among US litigants and nonlitigants. *Pain* 2014;155: 309-321.
- Myrtveit SM, Wilhelmsen I, Petrie KJ, Skogen JC, Sivertsen B. What characterizes individuals developing chronic whiplash?: The Nord-Trøndelag Health Study (HUNT). *J Psychosom Res* 2013;74: 393-400.

- Ng TS, Pedler A, Vicenzino B, Sterling M. Less efficacious conditioned pain modulation and sensory hypersensitivity in chronic whiplash-associated disorders in Singapore. *Clin J Pain* 2014;30: 436-442.
- Obelieniene D, Schrader H, Bovim G, Miseviciene I, Sand T. Pain after whiplash: a prospective controlled inception cohort study. *J Neurol Neurosurg Psychiatry* 1999;66: 279-283.
- Pande KC, Tripathi S, Kanoi R. Limited clinical utility of pain drawing in assessing patients with low back pain. *J Spinal Disord Tech* 2005;18: 160-162.
- Peterson G, Nilsson D, Trygg J, Falla D, Dederling A, Wallman T, Peolsson A. Novel insights into neck muscle coordination in individuals with whiplash-associated disorders. *Sci Rep* 2015;5: 15289.
- Ransford AO, Cairns DC, Mooney V. The pain drawing as an aid to the psychologic evaluation of patients with low-back pain. *Spine* 1976;1: 127-134.
- Roelofs J, Sluiter JK, Frings-Dresen MH, Goosens M, Thibault P, Boersma K. . 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: 181-190.
- Schomacher J, Farina D, Lindstroem R, Falla D. Chronic trauma-induced neck pain impairs the neural control of the deep semispinalis cervicis muscle. *Clin Neurophysiol* 2012;123: 1403-1408.
- Siegrist J, Starke D, Chandola T, Godin I, Marmot M, Niedhammer I, Peter R. The measurement of effort-reward imbalance at work: European comparisons. *Soc Sci Med* 2004;58: 1483-1499.

- Spitzer WO, Skovrom ML, Salmi LR, Cassidy JD, Duranceau J, Suissa S. Scientific monograph of the Quebec Task Force on Whiplash Associated Disorders: Redefining "whiplash" and its management. *Spine* 1996;20: 2-73.
- Sterling M. Clinical presentation of whiplash associated disorders. In: *Whiplash: evidence base for clinical practice*. Australia: Elsevier; 2011.
- Sterling M, Jull G, Vicenzino B, Kenardy J, Darnell R. Physical and psychological factors predict outcome following whiplash injury. *Pain* 2005;114: 141-148.
- Sterling M, Kenardy J, Jull G, Vicenzino B. The development of psychological changes following whiplash injury. *Pain* 2003;106: 481-489.
- Stewart M, Maher CG, Refshauge KM, Bogduk N, Nicholas M. Responsiveness of pain and disability measures for chronic whiplash. *Spine (Phila Pa 1976)* 2007;32: 580-585.
- Sullivan JL, Bishop SR, Pivik J. The pain catastrophizing scale: development and validation. *Psychological Assessment* 1995;7: 524-532.
- Sullivan MJ, Adams H, Martel MO, Scott W, Wideman T. Catastrophizing and perceived injustice: risk factors for the transition to chronicity after whiplash injury. *Spine (Phila Pa 1976)* 2011;36: S244-249.
- Tait RC, Chibnall JT, Krause S. The Pain Disability Index: psychometric properties. *Pain* 1990;40: 171-182.
- Thomas E, Silman AJ, Croft PR, Papageorgiou AC, Jayson MI, Macfarlane GJ. Predicting who develops chronic low back pain in primary care: a prospective study. *BMJ* 1999;318: 1662-1667.
- Van Oosterwijck J, Nijs J, Meeus M, Paul L. Evidence for central sensitization in chronic whiplash: a systematic literature review. *Eur J Pain* 2013;17: 299-312.

- Vernon H, Guerriero R, Kavanaugh S, Soave D, Moreton J. Psychological factors in the use of the neck disability index in chronic whiplash patients. *Spine (Phila Pa 1976)* 2010;35: E16-21.
- Vernon H, Guerriero R, Kavanaugh S, Soave D, Puhl A. Self-rated disability, fear-avoidance beliefs, nonorganic pain behaviors are important mediators of ranges of active motion in chronic whiplash patients. *Disabil Rehabil* 2013;35: 1954-1960.
- Vernon H and Mior S. The Neck Disability Index: A study of reliability and validity. *J Manipulative Physiol Ther* 1991;14: 409-415.
- Williamson E, Williams M, Gates S, Lamb SE. A systematic literature review of psychological factors and the development of late whiplash syndrome. *Pain* 2008;135: 20-30.
- Williamson E, Williams MA, Gates S, Lamb SE. Risk factors for chronic disability in a cohort of patients with acute whiplash associated disorders seeking physiotherapy treatment for persisting symptoms. *Physiotherapy* 2015;101: 34-43.
- Woodhouse A and Vasseljen O. Altered motor control patterns in whiplash and chronic neck pain. *BMC Musculoskelet Disord* 2008;9: 90.

## TABLE LEGENDS

**Table S1.** Characteristics of the participants. n=205 unless otherwise stated.

**Table S2:** Symptom characteristics. Results are reported as number of participants (%).

**Table 3:** Overall health status considering pain, work, disability, general health and psychological factors.

**Table 4:** Results of the Kruskal-Wallis test to verify if pain extent significantly changes according to sex, age, educational level, insurance status and financial status at the bivariate level. \*\*\*p<0.001, \*\*p<0.01, \*p<0.05

**Table 5:** Result of the multiple linear regression model. Coefficient (standard error). Pain extent was the independent variable of interest. \*\*\*p<0.001, \*\*p<0.01, \*p<0.05

**Table 6:** Result of the multiple linear regression model. Coefficient (standard error). Pain extent was the independent variable of interest \*\*\*p<0.001, \*\*p<0.01, \*p<0.05

## FIGURE LEGENDS

**Figure S1:** Participant flow through study. \*Whiplash injury in the preceding 6 to 36 months, reported to be the onset of current symptoms, excluding unconsciousness/loss of memory in connection to the whiplash injury, previous neck trauma with unresolved symptoms, previous neck surgery, ongoing malignant disease, severe psychiatric disorders, drug abuse, difficulties understanding the Swedish language. NDI indicates Neck Disability Index; VAS, Visual Analogue Scale; WAD, whiplash-associated disorders.

**Figure 2:** Pain frequency maps generated by superimposing the pain drawings of all patients included in the study (n=205). Pain frequency maps have been generated for both the dorsal and ventral view. The color grid indicates both the number and the percentage of individuals that reported pain in that specific area. Dark red represents the most frequently reported area of pain.

**Figure 3:** Pain location analysis which shows the percentage of individuals (n=205) reporting pain in a specific body region. The regions of the body have been color coded as displayed on the left side of the figure. The presence of the pain in a body region was confirmed when the pain drawing involved at least 10% of the body region area or where the number of pixels was greater than 60.