

# Is kinesiophobia and pain catastrophising at baseline associated with chronic pain and disability in whiplash-associated disorders?

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**RUNNING TITLE: KINESIOPHOBIA AND CATASTROPHIZING IN WHIPLASH ASSOCIATED DISORDERS**

**Is kinesiophobia and pain catastrophizing at baseline associated with chronic pain and disability in whiplash associated disorders? A systematic review**

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## **DISCLOSURES**

Javier Martinez-Calderon, PhD student at University of Malaga, is supported by the University of Malaga through a pre-doctoral grant. All authors state they have no conflicts of interest to declare. All authors have made a substantial scientific contribution to the study and are thoroughly familiar with the primary data. All authors have read the complete manuscript and take responsibility for its content and completeness.

## **ABSTRACT**

**Background** Kinesiophobia and pain catastrophizing may be associated with patients' transition from having acute to chronic pain following a whiplash injury.

**Objective** To systematically review and critically appraise the literature to determine whether kinesiophobia and pain catastrophizing are associated with greater likelihood of patients developing chronic pain and disability following a whiplash injury.

**Design** A systematic review of the literature

**Data sources** Electronic searches of PubMed, AMED, CINAHL, PsycINFO, and PubPsych, and grey literature were undertaken from inception to September 2017.

**Eligibility criteria for selecting studies** Study selection was based on longitudinal studies evaluating how kinesiophobia and/or pain catastrophizing at baseline are associated with pain intensity, disability, or both after a whiplash injury.

**Results** We included 14 longitudinal studies that described 12 independent cohorts with a total sample of 2,733 participants with whiplash associated disorder. Kinesiophobia at baseline was not associated with pain intensity over time (three studies). Whether kinesiophobia at baseline

was associated with disability was unclear as results were conflicting (six studies). There were also conflicting results when we examined the association between pain catastrophizing and both pain intensity (five studies) and disability (eight studies).

**Summary/Conclusions** Kinesiophobia at baseline was not associated with pain intensity over time. There were conflicting results were for the remaining analyses. The size of the associations were small. The overall quality of the evidence was very low.

**Trial registration number** CRD42016053864

**Key Words:** pain; whiplash injury; fear; psychological factors.

#### WHAT IS ALREADY KNOWN

- There is inconsistent evidence on how kinesiophobia and pain catastrophizing are associated with the course of whiplash associated disorders.
- A systematic review which updates current knowledge is necessary.

#### WHAT ARE THE NEW FINDINGS?

- Baseline kinesiophobia is not associated with pain intensity over time.

- There were conflicting results regarding the remaining associations.
- The overall quality of the evidence was very low and further research is warranted.

## **INTRODUCTION**

Whiplash-associated disorders (WAD) are a highly prevalent and costly condition [1]. It is the most common injury associated with a motor vehicle accident and affects up to 83% of the population [2–4]. WAD may also occur following falls or other mishaps [5]. The incidence of WAD is increasing, and is estimated at 300 per 100,000 inhabitants in the Western world [6]. As a result, WAD is associated with massive economic, personal, and emotional consequences, not only for individuals with WAD, but also for family members, health providers, medical-legal systems, and third-party payers [7].

People with WAD present with a wide range of symptoms such as pain, disability, and mood disorders [4,5]. Clinical prediction rules have been developed in order to assist clinical decision making for the best management of WAD [8–10]. Nevertheless, recovery rates remain static, and approximately 20% to 30% of those affected continue to report symptoms after twelve months [10–12]. The presentation of WAD can be complex and the mechanisms that contribute to the maintenance of pain and non-recovery are still not entirely clear. Many factors such as physical, biological, cognitive, behavioural, social, and occupational are often associated with poor prognosis of this entity [13,14]. Among the

biopsychosocial factors found to contribute to the development and maintenance of chronicity in WAD, maladaptive psychological factors (e.g., fear) can play a key role [13,15,16].

Kinesiophobia and pain catastrophizing are two psychological factors that have received significant empirical attention over the last two decades [17–22]. Kinesiophobia is defined as an excessive, irrational, and debilitating fear to carry out a specific movement or activity, due to a feeling of vulnerability of sustaining a painful injury or re-injury [23]. Pain catastrophizing has been conceptualized as a tendency to have overly negative thoughts in response to pain or pain-related cues [24,25]. Both constructs have been included in the Fear-Avoidance model of pain [21,26,27] which has been highly supported [28–33]. The application of the fear-avoidance model of pain to WAD [15,16] proposes that people with a trait tendency to have fearful or catastrophic thoughts in response to pain, are more likely to develop chronic pain after a whiplash injury. According to this model, individuals with WAD with high levels of kinesiophobia and/or pain catastrophizing, not only tend to overreact in response to actual or potential threats, but also tend to avoid activities which could be associated with pain. This then causes them to be more attentive to pain, resulting in greater perceived levels of pain intensity, pain-related disability and depressive symptoms. A great deal of evidence suggests that kinesiophobia and pain catastrophizing are associated with, and lead to, negative outcomes in the development [28,29] and maintenance [30,32,33] of chronic pain.

There is preliminary evidence showing that kinesiophobia [34–37] and pain catastrophizing [34,37,38] may be associated with the course of neck symptoms following a whiplash injury facilitating the transition from acute to chronic WAD. However, findings of these preliminary studies

remain inconsistent [34–38] regarding both the strength and even the direction of the associations. Several systematic reviews [14,39–42] have analysed whether pain catastrophizing and kinesiophobia are prognostic factors of WAD chronicity. Carroll et al. [14] and Walton et al. [39,42] concluded that kinesiophobia [14] and pain catastrophizing [14,39,42] were associated with poorer or less recovery, while Williamson et al. [41] and Campbell et al. [40] found inconclusive evidence. Based on this heterogeneity of findings, an updated and more rigorous systematic review was deemed necessary [43]. Furthermore, assessing the inconsistency, reporting bias, imprecision and indirectness is crucial to determine the level of the evidence for the studied outcomes [44]. The Grading of Recommendations Assessment, Development and Evaluation (GRADE) criteria can assess this and was employed in the current rigorous systematic review whereas it was not in past reviews. The aim of our study was to systematically review the literature including longitudinal cohort studies to determine whether kinesiophobia and/or pain catastrophizing associated with the progression to, and maintenance of, chronic pain and disability following a whiplash injury.

## **METHODS**

This review was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement [45]. The review protocol was registered at the International Prospective Register of Systematic Reviews (PROSPERO: CRD42016053864) prior to the identification of articles and data extraction.



### *Data Sources and Search Strategy*

Two reviewers (JMC and ALS) independently searched the following electronic databases from inception to September 2017 using optimized search strategies: PubMed, AMED, CINAHL, PsycINFO, and PubPsych. Manual searches of relevant eligible studies were also carried out through cross-references identified in journals associated with the topic of this review, and reference lists within both original and review articles, selecting studies which may have been missed during the electronic search. A sensitive search strategy using relevant search terms from Medical Subject Headings (MeSH) was developed, and keywords generated from the subject headings were used: "whiplash injuries" [MeSH Terms], "prognosis" [MeSH Terms], "catastrophization" [MeSH Terms], "fear" [MeSH Terms], whiplash associated disorder, whiplash syndrome, cervical spine disorder, cervical spine injury, psychological factors, kinesiophobia, fear of movement. The complete search strategy report is shown in **Appendix A**. The grey literature was also searched in the following databases in order to identify any relevant unpublished work: the British National Bibliography for Report Literature, New York Academy of Medicine Grey Literature Report, the System for Information on Grey Literature (Open Grey), and Google Scholar [46]. References were exported, and duplicates were removed using citation management software (Mendeley desktop v1.17.4).

### *Eligibility Criteria*

Based on the Cochrane PICO framework (P=population; I=intervention; C=comparator; O=outcome) [47], the PECO (P=population; E=exposure; C=comparator; O=outcome) framework was followed to determine which studies satisfied our inclusion criteria and were included in the present review:

- (i) Longitudinal cohort studies examining whether kinesiophobia and/or pain catastrophizing are associated with the progression to, and maintenance of chronic pain intensity and disability.
- (ii) Studies where participants were adults diagnosed with acute or subacute WAD.
- (iii) Studies where the follow-up was at least three months (time necessary to observe the possible transition to chronic pain).

Exclusion criteria were:

- (i) Studies examining kinesiophobia and/or pain catastrophizing in the context of a behavioural task or treatment.
- (ii) Studies examining the influence of pain catastrophizing and/or kinesiophobia on outcomes (pain intensity and disability) after applying any treatment.
- (iii) Cross-sectional studies, case-control studies, reviews, clinical studies, case reports, editorial, and abstracts.
- (iv) Studies exploring psychometric properties of kinesiophobia and/or pain catastrophizing measuring instruments.

Finally, no restriction was applied in terms of gender, ethnicity, language and settings for recruiting participants (general population, primary, secondary, or tertiary care).

### *Study Selection*

All studies identified by the search strategy were screened using the eligibility criteria listed above by two independent reviewers (JMC and ALS). The first stage of the assessment involved screening of titles and abstracts. The same reviewers undertook the second stage by screening the full text. In the case of disagreement, a decision was made by consensus or, when necessary, a third reviewer (DF) was consulted. A short checklist was adapted to the present review and was used to guide the selection of relevant studies (see **Appendix B**) [48].

### *Data Extraction*

Risk of bias assessment, quality of the evidence and data extraction were performed by cohort rather than by individual studies, in order to avoid giving greater weight to any factors that were investigated in the same cohort but in multiple publications. Two reviewers (JMC and ALS) independently extracted the following relevant data from each study: study details (first author, year of publication), study participant characteristics (mean age and pain duration (duration since WAD at baseline)), sample size, self-reported tools assessing pain catastrophizing, kinesiophobia, pain intensity and/or disability, as well as the duration of follow-up. When consensus could not be reached, we intended to ask a third reviewer (DF) to resolve the discrepancy, although this was not required. An email would have been sent to the original authors to obtain further information regarding their findings, although this step was not necessary.

### *Risk of Bias Assessment*

Two reviewers (JMC and ALS) independently assessed the risk of bias of the included studies using the Quality in Prognostic Studies (QUIPS) tool [49], which grades six separate study domains (selection of participants, study attrition, prognostic factor measurement, outcome measurement, study confounding and statistical analyses) according to their risk of bias (low, medium, or high risk of bias).

### *Statistical Analysis*

The same pair of reviewers independently evaluated the overall quality and the strength of the evidence per outcome using the GRADE criteria [50]. The GRADE criteria classified the type of evidence as either (1) high (further research is unlikely to change our confidence in the estimate of effect and there are no known or suspected reporting biases); (2) moderate (further research is likely to have an important effect on our confidence in the estimate of effect and might change the estimate); (3) low (further research is likely to have an important effect on our confidence in the estimate of effect and is likely to change the estimate) or; (4) very low (we are uncertain about the estimate) [51].

The quality of the evidence can decrease or increase based on the presence, or not, of the following factors: serious (-1) or very serious (-2) limitation to study quality (risk of bias); some (-1) or important (-2) inconsistency of results; some (-1) or major (-2) uncertainty about directness; some (-1) or important imprecision or sparse data and; some (-1) or high (-2) probability of reporting bias [50]. Specifically, regarding the risk of bias, when 50% or more of the included studies presented moderate or high risk, the GRADE system recorded “very serious” risk of bias. Inconsistency was graded “very serious” when 50% or more of the studies showed heterogeneity in terms of statistical significance and/or the

direction of the relationship. Imprecision was graded “very serious” when optimal information size (OIS) criterion was not met and the sample size was small; OIS criterion was met but the 95% CI around an effect did not exclude 1.0 (wide confidence intervals); 95% CI was not reported. For the primary analysis, studies were grouped per outcome: pain intensity and disability. A meta-analysis could not be carried out due to large heterogeneity in terms of: (i) participant’s age; (ii) different periods of follow-up across studies; (iii) variable management of the main outcome measures, e.g., pain intensity was measured as a dichotomous variable (minimal pain vs considerable pain) (55), by frequency of presentation (“never”, “sometimes”, “often” or “always”) (61), and as a continuous variable (35). Consequently, a descriptive quantitative analysis (the most relevant summary measure with a precision estimate) for each study was provided. For those studies that reported results with several degrees of adjustment for confounders in different models, we extracted the estimate from the model which showed the best goodness of fit. Review Manager (RevMan) version 5.3 (Copenhagen: The Nordic Cochrane Centre, The Cochrane Collaboration, 2014) software was used to process the data.

## **RESULTS**

### *Study characteristics*

A total of 1,827 citations were identified through electronic databases, with 17 additional studies identified following screening of references. After removing the duplicates, 945 titles and abstracts were screened with 397 full-text articles then evaluated. Of these, 14 longitudinal studies

describing 12 independent cohorts with a total of 2,733 participants with WAD satisfied our inclusion criteria and were included in this review [34,36,38,52–62]. The number of studies retrieved from each database and the number of studies excluded in each screening phase are shown in

**Figure 1.** The characteristics and the statistical results of the included studies are reported in **Appendix C.**

*Risk of bias assessment*

The degree to which the studies met the quality criteria varied considerably, ranging from moderate (study participation, prognostic factor measurement, outcome measurement as well as statistical analysis and reporting) to high (study attrition and study confounding) risk of bias. The risk of bias assessment for all of the included studies is presented in **Table 1.**

**Table 1.** Methodological quality for cohort studies (The QUIPS tool).

First Author	Study participation	Study attrition	Prognostic factor measurement	Outcome measurement	Study Confounding	Statistical analysis and reporting
Andersen et al. [38]	L	H	M	L	H	L
Asenlof et al. [34]	L	H	L	L	H	M
Bostick et al. [59]	L	H	M	L	M	M

Buitenhuis et al. [60]	H	H	L	M	H	H
Carstensen et al. [58]	L	M	M	M	M	M
Casey et al. [61,62]	M	M	M	M	H	M
Kivioja et al. [52]	M	M	M	H	H	M
Nederhand et al. [56]	M	L	M	M	H	M
Nieto et al. [36]	M	L	L	M	M	L
Pedler et al. [53]	M	L	M	M	H	M
Sterling et al. [54,55]	H	M	M	H	H	H
Williamson et al. [57]	M	H	H	H	H	M

Note: L = low risk of bias; H = high risk of bias; M = moderate risk of bias.

*Kinesiophobia and pain catastrophizing in the transition to chronic WAD: quality of the evidence based on the GRADE criteria*

According to the GRADE criteria, the overall quality of the evidence was very low regarding how pain catastrophizing and/or kinesiophobia are associated with both pain intensity and disability (**Table 2**).

**Table 2.** Summary of findings and Quality of evidence assessment based of the GRADE criteria.

Summary of findings			Quality of evidence assessment (GRADE)						
Outcome	No of studies	No. of participants	Risk of bias <sup>1</sup>	Inconsistency <sup>2</sup>	Indirectness <sup>3</sup>	Imprecision <sup>4</sup>	Other considerations <sup>5</sup>	Level of evidence	Importance
Kinesiophobia									
Pain intensity	3	227	Very serious	No	No	Very serious	Reporting bias detected	Very low	Critical
Disability	6	513	Very serious	Very serious	No	Very serious	Reporting bias detected	Very low	Critical
Pain catastrophizing									
Pain intensity	5	1,382	Very serious	Serious	No	Serious	Undetected	Very low	Critical
Disability	8	1,392	Very serious	Very serious	No	Serious	Reporting bias detected	Very low	Critical

Note: 1. Observational studies (failure to develop and apply appropriate eligibility criteria; flawed measurement of both exposure and outcome; failure to adequate control confounding; incomplete follow-up; non-presence of an unexposed cohort) 2. Point estimates vary widely across studies; confidence intervals show minimal or no overlap. 3. Differences in population, differences in intervention, differences in outcome, indirect comparison. 4. Optimal information size (OIS) criterion is not met and the sample size is small; OIS criterion is met but the 95% CI around an effect does not exclude 1.0 (wide confidence intervals); 95% CI is not reported. 5. Outcome data not included in the predictive model.

*Very low evidence regarding how kinesiophobia is associated with pain intensity*



A total of three studies [36,54,55] examined how kinesiophobia is associated with pain intensity in the transition to chronic WAD. Pain intensity over time did not differ among patients with different levels of kinesiophobia at baseline (see **Appendix D**). Nieto et al. [36] reported a small regression coefficient  $\beta = 0.26$   $p = 0.053$ . However, Sterling et al. [54,55] did not report this information in their logistic regression models.

*Conflicting results with very low evidence regarding how kinesiophobia is associated with disability*

A total of six studies [34,36,53–56] were included in this analysis. There were conflicting results across the included studies. Three studies revealed that higher levels of kinesiophobia at baseline was associated with the progression and maintenance of disability at six months [36,53,56]. The values of the regression coefficients were small  $\beta = 0.29$   $p = 0.01$  [36];  $B = 0.341$   $p = 0.002$  [53] and  $AUC[95\%CI] = \text{probability of } 83.3\% [70.3–91.3]$   $p < 0.05$  [56].

On the other hand, three studies showed non-significant results [34,54,55] (see **Appendix E**). Åsenlöf et al. [34] reported a small values of regression coefficients  $\beta[95\%CI] = -0.06[-0.34 \text{ to } 0.147]$   $p = 0.43$ . However, Sterling et al. [54,55] did not report this information in their logistic regression models.

Thus, overall the findings were inconsistent and imprecise (Table 2).

*Conflicting results with very low evidence regarding how pain catastrophizing is associated with pain intensity*

Five studies examined this issue [36,38,52,58,59]. Three studies showed that higher levels of pain catastrophizing at baseline was associated with the progression and maintenance of pain intensity at three [59], six [38,59] and twelve months [58], with a small value of the regression coefficient of  $\beta = 0.08$   $p = 0.01$  [38]; OR [95%CI] = 1.14[1.10 to 1.18]  $p < 0.000$  [58];  $\beta$ [95%CI] = 0.09[0.03 to 0.16]  $p < 0.05$  (3 months) [59];  $\beta$ [95%CI] = 0.10[0.0 to 0.18]  $p < 0.05$  (6 months) [59].

However, non-significant results were found in two studies [36,52] (see **Appendix F**) with a small value of regression coefficient  $\beta = 0.02$   $p = 0.89$  [36] and OR[95%CI] = 1.0[0.9 to 1.08]  $p = 0.913$  [52]. Thus, overall the findings were inconsistent and imprecise (Table 2).

*Conflicting results with very low evidence regarding how pain catastrophizing is associated with disability*

A total of eight studies [34,36,56,57,59–62] evaluated this topic. Higher levels of pain catastrophizing at baseline was associated with the progression and maintenance of disability at three [59], six [56], twelve [61] and twenty-four months [62], while one study was associated with the opposite at six months [60]. One study reported a moderate value of the regression coefficient ( $\beta = 2.16$   $p < 0.001$ ) [61], based on the analysis of the helplessness dimension of the PCS. However, most of the coefficients were small ( $B = 0.12$   $p \leq 0.01$  [62];  $B$ [95%CI] = 0.08[0.01 to 0.16]  $p < 0.05$  [59]; OR[95%CI] = 0.885[0.814 to 0.962]  $p = 0.004$  [60]; AUC[95%CI] = 0.78 [0.67-0.88]  $p < 0.05$  [56]).

On the other hand, non-significant results were observed in five studies [34,36,57,59,60] (see **Appendix G**), with a small value of the regression coefficient  $\beta = 0.09$   $p = 0.35$  [36];  $\beta$ [95%CI] = 0.08[-0.11 to 0.35]  $p = 0.30$  [34];  $B$  [95%CI] = 0.06[-0.01 to 0.14]  $p > 0.05$  [59]; OR[95%CI] =

0.942[0.878 to 1.011] p= 0.097 [60]. Williamson et al. [57] did not report any figures regarding the regression logistic model. Thus, overall the findings were inconsistent and imprecise (Table 2).

## **DISCUSSION**

The purpose of this study was to systematically review and critically appraise the literature to determine whether kinesiophobia and pain catastrophizing are associated with greater likelihood of patients developing chronic pain and disability following a whiplash injury, based on the analysis of 14 longitudinal studies in 12 independent cohorts. Of these, six studies were included in the current review which were not previously examined in prior systematic reviews [14,39–42]. The synthesis of data from the included studies revealed the following findings: (i) higher levels of kinesiophobia at baseline were not associated with greater pain intensity over time; (ii) conflicting results were found on how kinesiophobia at baseline is associated with disability; (iii) there were also conflicting results on how pain catastrophizing at baseline is associated with both pain intensity and disability. The level of evidence of all these findings was very low in terms of risk of bias, inconsistency and imprecision of the results. Our findings do not support kinesiophobia and pain catastrophizing in being able to be associated with pain or disability in the prognosis of WAD, as previous reviews have shown [40,41]. However, our confidence in these results have improved due to the increased rigour of the current study and the inclusion of six additional studies which were not previously evaluated.

Previous systematic reviews have been conducted in order to detect potential prognostic factors for the perpetuation and maintenance of chronic pain and disability following a whiplash injury [14,39–42]. Carroll et al. [14] explored the course of neck pain and associated symptoms in WAD, based on the analysis of 70 articles. They concluded that both higher levels of kinesiophobia and catastrophizing at baseline were associated with more pain intensity and disability over time. However, these conclusions about the influence of kinesiophobia and catastrophizing were only based on one study. Williamson et al. [41] evaluated the role played by some psychological factors in the development of late whiplash syndrome. Twenty-five articles were included with a total of five studies specifically analysing kinesiophobia and catastrophizing. Their findings were inconclusive. Walton et al. [39,42] assessed potential risk factors for persistent problems following whiplash after a motor vehicle accident, based on a total of fifteen cohorts. Of these, only three longitudinal cohort studies explored the role of kinesiophobia and catastrophizing in the progression and maintenance of WAD. They concluded that catastrophizing was associated with the course of symptoms in WAD, while kinesiophobia did not. Finally, Campbell et al. [40] explored potential psychological factors in the development of chronic whiplash symptoms, based on the inclusion of thirty-one studies. Seven longitudinal cohort studies out of thirty-one analysed the potential role of kinesiophobia and catastrophizing in WAD. Their findings showed inconclusive evidence about the potential role that kinesiophobia and catastrophizing play in the development of chronic WAD.

The inconsistency of the results observed in previous systematic reviews is supported by our findings since the current systematic review also revealed conflicting results on how kinesiophobia and pain catastrophizing are associated with disability and pain intensity. The current findings

were based on the application of a robust review tool, the GRADE criteria, which showed that the quality of the evidence was very low in terms of inconsistency, imprecision, reporting bias, indirectness and risk of bias of the reported findings. However, previous systematic reviews showed inconclusive [40,41] or good [14,39,42] evidence based exclusively on the quality of their included articles in terms of risk of bias, and based on a scarce number of studies.

The fear-avoidance model of pain [21,27] is probably the framework which has received more empirical attention to explain the transition to chronic pain. Specifically, in WAD, individuals who show a certain level of kinesiophobia and/or catastrophizing about pain often try to avoid specific movements and activities. These activities are thought to be potentially dangerous, which is problematic since active exercise is usually effective in WAD [63]. These maladaptive behaviours give rise to sedentary behaviour in people with musculoskeletal pain [64], leading to physical impairment. Such changes affect the mood state, causing more pain and emotional distress [65]. This negative vicious cycle can affect rehabilitation efforts, interfere directly on patient recovery, and can maintain a negative pain experience [27]. Nevertheless, the results of the current review do not support this model, thus, more rigorous studies are needed in order to draw firm conclusions.

An integrated biopsychosocial model was recently proposed by Walton and Elliot [66]. Contrary to the fear-avoidance model of pain, this proposed new framework considers not only psychological factors (e.g., mostly fear and pain catastrophizing) as the key drivers to develop chronic pain, but also a range of factors such as biological, social, and contextual factors, which interact with each other contributing to the perpetuation of symptoms in WAD. This may explain why, despite the existence of clinical prediction rules in people with WAD [9,10], this

knowledge has still not resulted in better outcomes [67,68]. Furthermore, multiple pathways associated with the development and perpetuation of pain intensity and disability should be considered. The number and duration of episodes, fluctuation of symptoms, biopsychosocial profile of every individual and their health care use, can vary considerably between individuals with WAD. Therefore, it seems unlikely that kinesiphobia and pain catastrophizing can be associated with pain intensity and disability in the simplistic pathway proposed in the fear-avoidance model of pain [69].

#### *Methodological considerations*

There are some limitations in the evaluated studies that should be mentioned. Mediation models for longitudinal data offer improved statistical inference between variables [70]. When determining the impact of a determined prognostic factor, variables which mediate the relationship between the exposure and the outcome are important. However, mediation analyses were only carried out in one study [71]. Furthermore, the presence of confounding variables should be considered when regression models are developed. However, statistical analyses adjusted for potential confounders were not always reported.

There are some limitations of the current review. First, case-control studies were not considered during the search strategy. Therefore, potential articles for inclusion may have been missed. However, a complete search strategy was applied, including five databases, the grey literature, and a

manual search. Second, some studies did not report data regarding association. Third, there were some modifications to the original protocol registered in PROSPERO (CRD42016053864): (i) the included psychosocial factors were reduced to two specific psychological factors: kinesiophobia and pain catastrophizing; (ii) the Modified Cochrane Back and Neck Pain Group Criteria and the adapted version of NOS were replaced by the GRADE criteria and the QUIPS tool; (iii) the focus of the review was the transition to chronic WAD and not the prognosis in chronic WAD. The reasons for these modifications were, as follows: (i) kinesiophobia and pain catastrophizing are two constructs that have received more attention in the prognosis of chronic musculoskeletal pain; (ii) we came to the conclusion at the beginning of this review that the most appropriate tools to analyse the evidence in systematic reviews are those proposed by the Cochrane group, (iii) there is inconclusive evidence to explain why some patients with acute WAD are more susceptible to develop chronicity than others.

### *Future Research*

The risk of bias identified in most of the included articles needs to be addressed in future studies. This includes consideration of confounding variables when regression models are carried out, adequate sample size, and the sufficient power to detect a meaningful difference in the outcomes of interest.

## **CONCLUSION**

This systematic review analysed how kinesiophobia and pain catastrophizing are associated with with greater likelihood of patients developing chronic pain and disability following a whiplash injury. The findings suggest that kinesiophobia at baseline is not associated with pain intensity over time. There were conflicting results regarding how pain catastrophizing at baseline is associated with pain intensity and disability, and how kinesiophobia at baseline is associated with disability. Additionally, the overall quality of the evidence was very low.

## **COMPETING INTERESTS**

The authors declare to have no competing interests

## **CONTRIBUTORSHIP**

All authors have made a substantial scientific contribution to the study and they are thoroughly familiar with the primary data. All authors have read the complete manuscript and take responsibility for the content and completeness of the manuscript

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### **ETHICAL APPROVAL INFORMATION**

Not applicable

### **DATA SHARING STATEMENT**

Not applicable

### **KEY POINTS**

**Findings:** Kinesiophobia at baseline is not associated with pain intensity over time. There were conflicting results regarding pain catastrophizing at baseline and pain intensity and disability, and the role of kinesiophobia at baseline on disability.

**Implications:** Given the low level of evidence present, further research in the field is needed

**Caution:** The results of this review provide low-level support that both factors may be associated with outcome in WAD.

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## **LEGENDS**

Figure 1: Flow diagram of review process