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Original article

Relationship between neck motion and self-reported pain in patients with whiplash associated disorders during the acute phase

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ABSTRACT

Background: Biomechanical measures quantify motor control and functional deficits in Whiplash Associated Disorders (WAD), but few studies relate those measures to the clinical scales that are routinely used to assess patients. Most studies are limited to chronic neck pain, and report poor to moderate correlations.

Objective: To define a statistical model that relates measures of neck kinematics with clinical scales of neck pain, in WAD patients during the rehabilitation process in the acute phase (less than 3 months since the accident).

Methods: 96 WAD patients self-assessed their pain using VAS and NPQ, and passed neck motion tests as part of their rehabilitation program. Four regression models were fitted to analyze the effects of the measured kinematic parameters and subject-specific characteristics on VAS and NPQ. Model errors were compared to minimal clinically significant differences.

Results: Multiple correlation coefficients of the models were between 0.74 and 0.90. More than 66% of that correlation was accounted for by subject-specific factors, and most of the other half by the measured kinematic parameters. Range of motion of flexion-extension and axial rotation, and harmonicity of flexion-extension, where the variables most consistently related to the decrease of pain. The error of the models was within the MCSD in more than 50% of the observations.

Conclusions: Part of the individual progression of pain and pain-related disability in acute WAD patients, as rated by NPQ and VAS, can be mapped to objective kinematic parameters of neck mobility tests, like ranges of motion, velocities, repeatability and harmonicity of movements.

1. Introduction

Neck pain is a health problem with an important social and economical impact, reported as the fourth leading cause of years lost to disability (Cohen, 2015). One of the leading causes of neck pain is Whiplash Associated Disorder (WAD), typically associated with traffic accidents, which is specially problematic due to its high incidence (about 300 per 10,000 inhabitants in western countries), the difficulty of its clinical diagnostic, and the controversial relationship with insurance compensations, litigation and malingering (Holm et al., 2008).

Methods for assessing WAD may be broadly classified into patient-reported outcome measures (PROMs) and objective measures of physical ability (Misailidou et al., 2010; Sterling and Kenardy, 2008). PROMs assess physiological and psychosocial factors, which strongly influence the course of pathology and pain coping attitudes and behaviors (Vargas-Prada and Coggon, 2015). There are several scales for such measures whose validity and prognostic value have been demonstrated in multiple studies, like the Neck Disability Index (NDI, Pietrobon et al., 2002), the Visual Analogue Scale (VAS, Dimitriadis et al., 2014), and the Northwick Park Neck Pain Questionnaire (NPQ, González et al., 2001; Leak et al., 1994), among others.

Objective physical measures are also valuable assessment instruments, specially in medico-legal frameworks that require objective evidence of injury or significant consequences for the claimant's life (Bošnjak-Pašić et al., 2007; Oliphant, 2016; Pujol Robinat, 2017). They can be useful to objectify persistent pathology (Elliott et al., 2013), or as complementary measures to quantify specific motor control and function deficits (Misailidou et al., 2010). They normally involve the analysis of neck motion, which forensic medical examiners and rehabilitation physicians hold as a useful instrument in the assessment of WAD sequelae and recovery control (Vivas Broseta et al., 2017). However, the evidence of their validity and prognostic value is still limited in comparison with PROMs (Nordin et al., 2008).

In the last decade many studies have pursued the validation of

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motion analysis for WAD assessment, chiefly in terms of the reliability of the kinematic measures (Hanney et al., 2014; Michiels et al., 2014; Popovich Jr. et al., 2015; Sarig Bahat et al., 2016; Vorro et al., 2013; Williams et al., 2011) and their capacity to discriminate between patients and controls (Baydal-Bertomeu et al., 2011; Sarig Bahat et al., 2015; Stenneberg et al., 2017; Tsang et al., 2014). The most frequently studied kinematic parameter is neck range of motion (ROM), although some of those studies also focus on other motion measures like velocity, smoothness or harmonicity of repeated movements.

But to demonstrate the utility of motion analysis to support WAD assessment, it is also important to determine whether they can explain the course of pain, which is normally assessed by PROMs (Ailliet et al., 2018; Dunn et al., 2017). However, only a few papers have studied the relationship between neck kinematics and clinical scales, reporting poor to moderate correlations. Most of them focus on chronic neck pain, comparing clinical scales with cervical ROM, either measured in conventional tests (Howell et al., 2012; Ylinen et al., 2004), or with virtual reality games that allowed velocities and other kinematic parameters to be measured (Sarig Bahat et al., 2014; Treleaven et al., 2016).

Two longitudinal studies analyzed neck pain and motion during periods between 2 and 6 months (Chiu et al., 2005; Meisingset et al., 2016). In another study, patients were classified as “acute” (< 2 weeks), “subacute” (< 6 months) and “chronic” (> 6 months), according to the time passed between onset of pain and the assessments (Hermann and Reese, 2001). Only one study focused specifically on WAD patients, who were reassessed at different times over a 6-month period (Kasch et al., 2001).

The objective of this study is to define statistical models that relate neck motion parameters in WAD patients with the course of PROMs that represent the perception of pain and disability, during the rehabilitation process in the acute phase (less than 3 months after the accident). The relationship between neck motion and PROMs was analyzed controlling for factors that other studies have considered separately, as time, patient demographic characteristics, and baseline values of reported pain. The models did not only consider the relationships between inputs and outputs, but also their variations between sessions. Such models will help us understand what part of the changes observed in the course of PROMs are predictable from objective measures.

2. Methods

2.1. Participants

A statistical analysis was conducted on measurements from patients who reported neck pain after a traffic accident graded as WAD II/III (Spitzer et al., 1995), and followed a physiotherapy program administered at three rehabilitation clinics in Spain, which included biomechanical assessments after every 5 rehabilitation sessions.

The study sample included 96 patients who underwent two assessments, and a subgroup of 87 who had a third assessment. They were evenly distributed by gender and age, with similar time elapsed between assessments (Table 1).

Table 1
Description of the subjects' sample.

	N2	N3	Age	T1	T1-2	T2-3
Female	51	47	39.67 (13.48)	19.34 (9.81)	10.00 (2.55)	9.74 (3.91)
Male	45	40	38.89 (12.56)	21.08 (11.79)	9.32 (3.26)	8.63 (3.23)

N2, N3: Sample size at the 2nd and 3rd session, respectively.
Age: Age in the first assessment, measured in years (mean and std. deviation).
T1: Days elapsed from the accident to the first measurement (mean and std. deviation).
T1-2, T2-3: Days elapsed between successive measurements (mean and std. deviation).

2.2. Statement of ethical approval

All participants gave informed consent to participate in the study and process the data recorded in the tests. This study was approved by the Ethics Committee of the Universitat Politècnica de València (ref. P7_11_01_18).

2.3. Measurement of neck pain and kinematic parameters

At the beginning of each session, the subjects rated their level of pain and pain-related disability on a 10-cm VAS scale and with the NPQ. Then neck motion was analyzed using the procedure described by Baydal-Bertomeu et al. (2011): subjects sat on an adjustable chair, with their trunk secured to the backrest. A photogrammetry system (Kinescan-IBV) recorded a 4-marker set mounted on a helmet (Fig. 1). At the start of each measurement the subjects looked straight at a mirror fixed at eye level, and then made cyclic neck motions around a given axis, with the maximum achievable range at their preferred speed during 30 s. Each subject made two trials around each axis, in the following order: flexion-extension (FE), lateral flexion (LF), and axial rotation (AR). The time series of the main rotation angles were extracted and differentiated to obtain angular velocities and accelerations.


Fig. 1. Instrumented subject.

Those signals were used to calculate the following kinematic parameters, which were averaged across the two trials of each axis (Baydal-Bertomeu et al., 2011), see Fig. 2 for details:

- Range of motion (ROM).
- Maximum absolute angular velocity (MV), calculated as the 95th percentile of absolute angular velocity.
- Phase area ratio (PAR), as indicator of the motion variability across cycles. The PAR is the ratio between the areas traced by the angle-angular velocity diagram, as $S_B \div S_M$, where S_M is the area inside the trace of the mean cycle, and S_B is the area enclosed in the band around the mean cycle ± 1 standard deviation.
- Harmonicity (H), which represents the similarity of the cyclic motion and a sine wave. It is calculated as the absolute value of Pearson's correlation coefficient between the main angle and

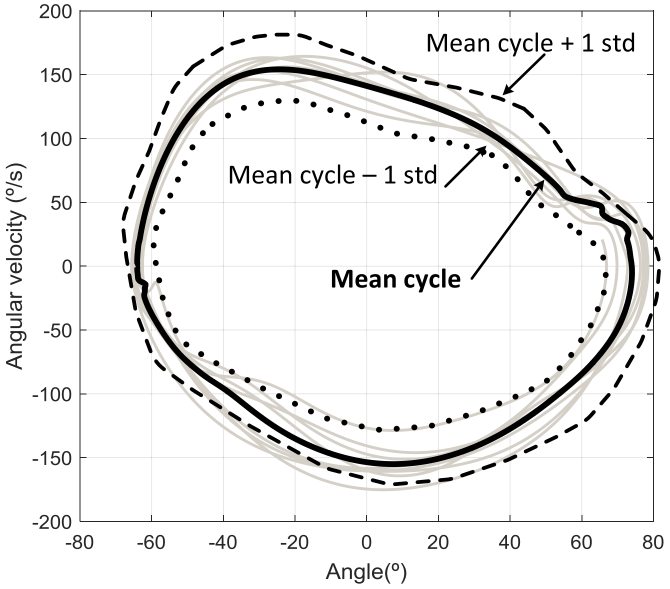


Fig. 2. Angle-angular velocity diagram used in the calculation of ROM, MV and PAR.

angular acceleration.

Since MV values had a heavily long-tailed distribution, it was log-transformed in order to avoid excessive influence of extreme values on the statistical models; likewise, PAR was logit-transformed since it was bounded in the [0–1] interval, highly concentrated at the lower end of the scale (Fox and Weisberg, 2011). To improve numerical stability and facilitate the comparison of model coefficients, all were rescaled to variables with zero mean and unit variance.

2.4. Statistical analysis

For each output (NPQ, VAS) two models were fitted: one that related the absolute values of PROMs and kinematic parameters, and another that related their variations, taking the first session as baseline. This was done to test whether analyzing variations instead of absolute values might compensate individual differences between patients.

2.4.1. Model fitting

Each model was calculated following the steps presented in Fig. 3. The mathematical details are described in a separate appendix. All analyses were performed using the R package for statistical computing (R Core Team, 2017).

In all cases, a basic model was first fitted using the PROMs or their variations as output, and the session number plus subject characteristics (age, gender) as possible inputs, together with a "random" effect associated to the patient's code. In the models for variations, the "baseline" scores reported in the first session (NPQ_0, VAS_0) were also considered as inputs. In order to ensure parsimonious models, the relevant inputs were selected by the stepwise AIC method (Yamashita et al., 2007), in order to obtain the "variables of the basic model".

The process was then repeated adding the kinematic parameters or their variations to those basic variables. In order to increase the robustness of the models, they were refitted after removing influential outliers — assessed by Cook distances associated with both fixed and random effects (Van der Meer et al., 2010).

Model fitting was done with linear mixed models for NPQ scores (Bates et al., 2015). For VAS scores we used a cumulative link mixed model, since they were not continuous, but discrete values in a scale from 0 to 10, (Christensen, 2015). To avoid sparse observations at the ends of the scale, VAS scores smaller than 2 or greater than 9 were merged, and the variations of VAS with respect to the baseline value were reduced to a factor with values $\Delta VAS \in \{\leq 3, -2, -1, 0, 1\}$. The resulting frequencies of VAS scores and variations kept a "bell shape", steadily decreasing towards the ends of the scale.

2.4.2. Analysis of the models

The resulting models contained three sources of information: (1) "basic" measurable characteristics of the subject and the session (gender, age, initial scores, session number), (2) measures of neck kinematics, and (3) subject-specific unknown or "random" effects. The model coefficients associated to kinematic parameters were examined in order to evaluate the influence of neck kinematics. The relative importance of each source of information in the estimation of PROMs was assessed through the multiple correlation coefficients (MCC) associated to different "sub-models": the full models, "fixed effects models" ruling out the random effects, and "basic fixed effects models" that only took into account the variables selected for the basic models (ruling out the kinematic parameters).

The MCC of the NPQ sub-models were calculated as the square root of marginal and conditional coefficients of determination R^2 (Nakagawa and Schielzeth, 2013). For VAS sub-models, the calculation and decomposition of R^2 in cumulative linked mixed models does not have a straightforward interpretation, so the MCC were calculated as the Pearson's correlation coefficient between the observed PROMs and the outputs of the sub-models.

The errors of the "fixed effects models" were compared to minimal clinically significant differences (MCSD) for change in scores on the pain scales, for which 30% of the full scale is advocated (Dworkin et al., 2009; Ostelo et al., 2008).

3. Results

3.1. Description of the variables

In the first measurement session after the accident, the mean NPQ score was 22.9 (std. deviation 9.2) and the mean VAS was 5.8 (standard deviation 1.6). In subsequent sessions the scores tended to decrease, although there was a large variance between subjects (Fig. 3).

3.2. Statistical models

3.2.1. Effects of kinematic parameters and other factors

In the "basic" models, neither gender or age were selected as influential variables. Only the number of the session was selected in all models, as well as the initial value of NPQ in the model that assessed the NPQ changes from baseline.

Table 2 shows the influence of those factors and the kinematic parameters in the models. The selected kinematic parameters were different for each model, with the exception of flexion-extension ROM and harmonicity, which appeared in all models.

The absolute values of NPQ scores in each session were related to ROM (FE, AR), MV (FE, LF), and H (FE). According to the coefficients of the session number, NPQ scores decreased between 2 and 4 points between sessions on average, and the effect of the kinematic parameters was comparable to that independent contribution of time. NPQ variations from baseline were related to the same parameters except ROM in AR, with similar coefficients as the model of absolute values.

The absolute values of VAS scores in each session were related to ROM (FE, AR), MV(AR), PAR (LF, AR) and H (FE). VAS variations were related to the variations of the same kinematic parameters except MV and PAR in AR, which were replaced by MV in FE.

3.2.2. Goodness of fit

All models had a good fit, with MCCs ranging between 0.74 and 0.90 for the "full models", i.e. including both fixed and random effects (Fig. 4). The correlations of the sub-models that ruled out subject-

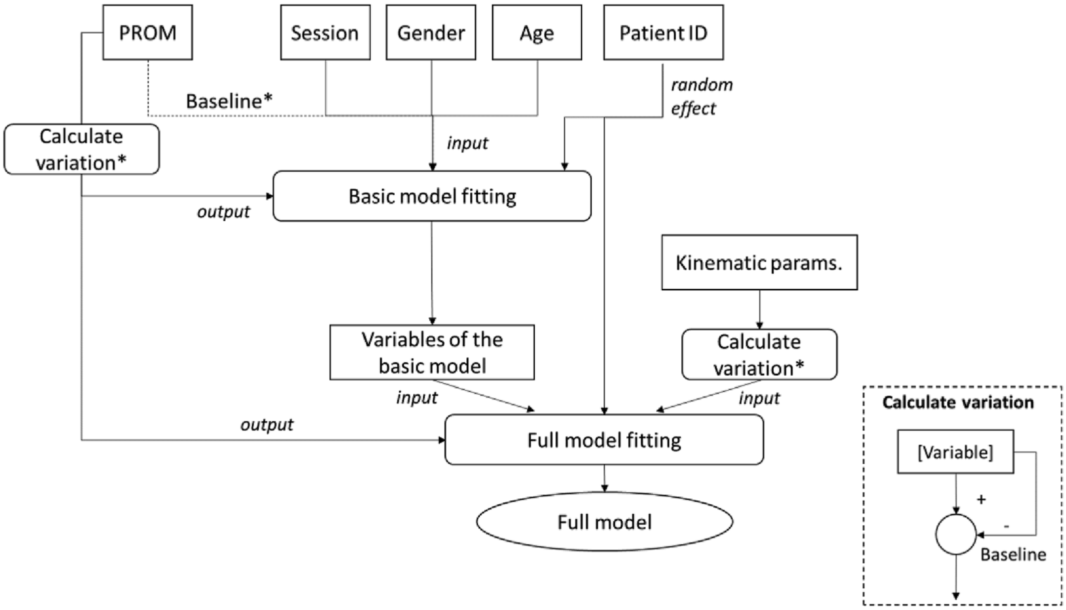


Fig. 3. Model fitting procedure. The variables and operations marked with an asterisk (*) were only used to fit the models for variations.

Table 2
Estimates of the fixed effects coefficients (standard error in parentheses) for the four fitted models.

	NPQ (absolute value)	NPQ (variation)	VAS (absolute value)	VAS (variation)
Other factors				
Base value	–	−0.17 (0.06)	–	–
Session 2	−2.16 (0.75)	–	−1.12 (0.33)	–
Session 3	−3.86 (0.83)	−1.82 (0.79)	−1.62 (0.39)	−0.43 (0.35)
Kinematic par.				
ROM (FE)	−3.50 (1.08)	−3.59 (1.25)	−0.59 (0.38)	−1.47 (0.61)
ROM (AR)	−2.09 (0.93)	–	−0.94 (0.46)	−1.23 (0.57)
MV (FE)	3.48 (1.54)	2.97 (1.80)	–	0.24 (0.74)
MV (LF)	−2.58 (1.19)	−3.08 (1.37)	–	–
MV (AR)	–	–	−0.60 (0.44)	–
PAR (LF)	–	–	−0.54 (0.27)	−0.54 (0.29)
PAR (AR)	–	–	0.48 (0.32)	–
H (FE)	−2.04 (0.89)	−1.91 (1.06)	−0.70 (0.30)	−1.02 (0.50)

Abbreviations: ROM: Range of Movement, PAR: Phase-Area Ratio, MV: Max. Velocity, H: Harmonicity; FE: Flexion-Extension, LF: Lateral Flexion, AR: Axial Rotation.

specific random factors were from 66% to 72% of the full model correlations. Those proportions decreased to values between 17% and 34% in the "basic" models that did not take into account kinematic parameters (see Fig. 5).

The correlations were higher for the models that related the absolute values of NPQ and VAS with the measures at each session (around 0.90 in both cases). However, the size of errors was smaller for the models that related variations of NPQ and VAS with variations of the measures. In all cases, the errors were smaller than the MCSD more than 50% of cases for NQP, and more than 85% of cases for VAS.

4. Discussion

4.1. Relationship between neck motion and NPQ and VAS scores

Pain is a subjective experience that is normally evaluated by PROMs, but it has physical manifestations that can be quantified objectively. The statistical models presented in this paper quantify the relationship between neck motion and NPQ and VAS scores in WAD patients in the acute phase, where ROM and H in FE, and ROM in AR, were the kinematic parameters with the clearest influence. That relationship is consistent with the findings of previous studies mainly done with chronic patients, although it is conditioned by a large variability between subjects, which has been controlled by the statistical approach.

The proposed models had three types of inputs: (1) basic information that did not require any type of specific measurement, like the subject's gender or age and the time since the accident (indirectly coded as the number of assessment sessions), plus baseline pain perception measures when the variations from baseline were studies; (2) kinematic parameters in neck mobility tests; and (3) subject-specific "random" aspects that were not characterized in the study.

Considering the MCCs of the different sub-models, roughly one third part of the variability in the scales predicted by the models was accounted for by such subject-specific factors, whereas neck kinematics accounted for the greatest part of the remaining variability. Gender or age did not have any significant influence on the outcomes, and the estimated changes over time independent of variations in kinematic parameters made little difference with a null model that only considered average patterns.

4.2. Interpretation of the models

The examination of the models' coefficients determines the relationship between performance in neck motion and perceived pain or disability. In most cases better performance was related to less pain, as expected, but not always. That relationship was always observed for ROM and H in FE, which were the common parameters to all models, as well as for ROM in AR, which appeared in three out of the four models. On the other hand, the contribution of MV in FE (for three models) and PAR in AR (for one) was the opposite. Since the correlation between kinematic parameters was always positive, that contrast may be understood as a "moderating effect": i.e. the reported pain tended to increase for subjects who, while being able or confident enough to perform relatively fast flexion-extension movements or regular left-right rotation, had limited or irregular mobility as measured by other parameters.

Contrary to the initial hypothesis, the fit was poorer when we studied the relationship between variations of kinematic parameters and PROMs from baseline, possibly due to the accumulation of unexplained residual variance in the calculation of differences, which would

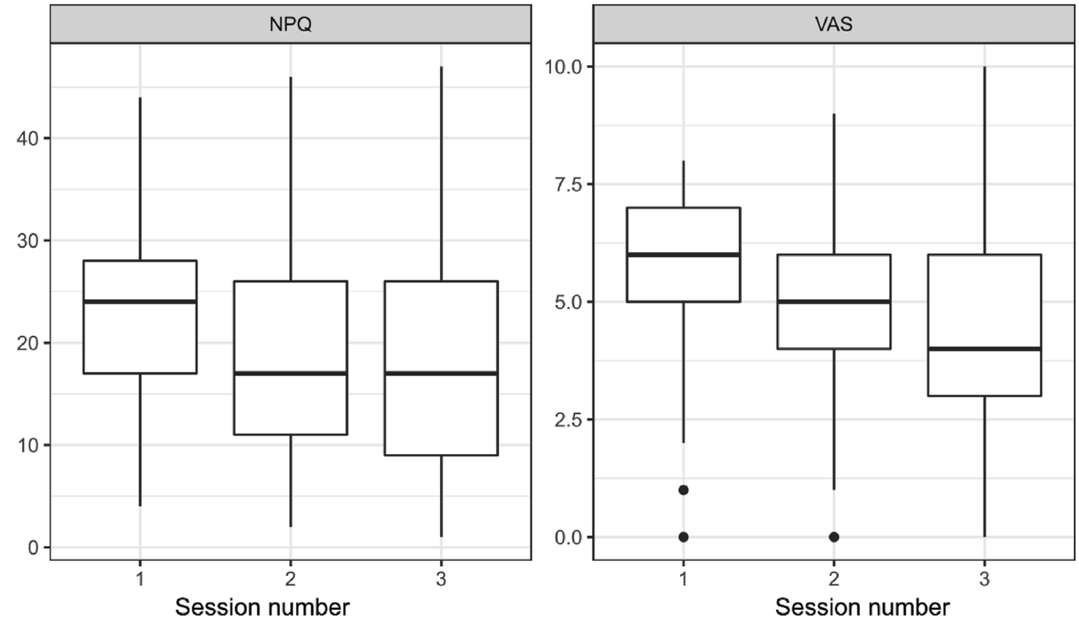


Fig. 4. Distribution of NPQ and VAS scores across sessions.

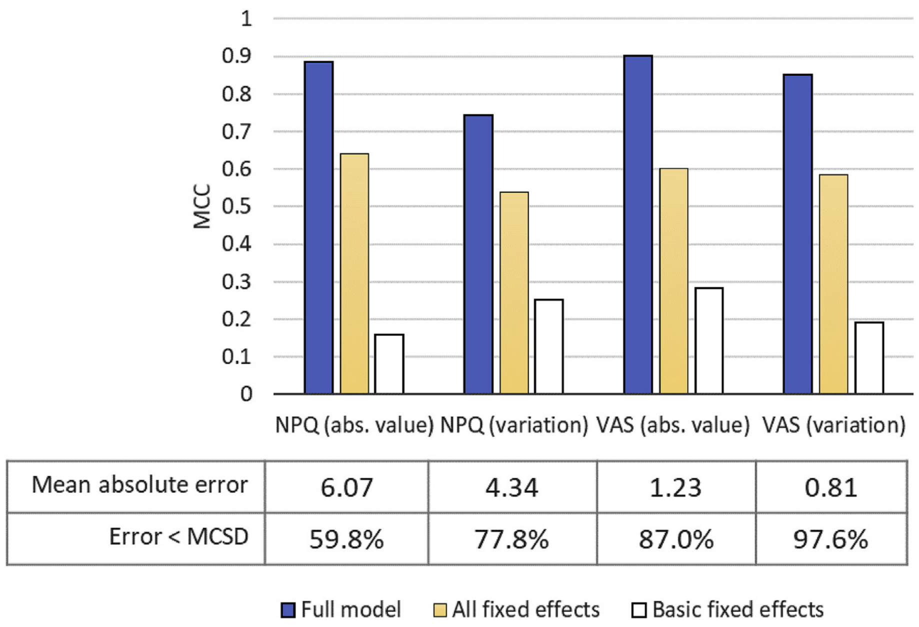
overinflate the denominators used in the calculation of the MCCs. On the other hand, the size of the errors in the analysis of variations from baseline was around 2:3 smaller than the errors of absolute scores.

4.3. Comparison with previous studies

This is the first study of this kind applied to WAD patients in the acute phase, but the results are consistent with other studies that showed that neck kinematics are generally related to pain and pain-related disability (Childs et al., 2008; Hermann and Reese, 2001; Vogt et al., 2007). Some differences can be explained by the statistical approach. Most previous papers have focused on the analysis of correlations and linear regressions between pairs of variables. Treleaven et al. (2016) recently published a paper with a multiple regression for the opposite relationship to the one presented here, in which the inputs were six different PROMs (including VAS and NDI as pain-related measures) and the output were the kinematic variables. Meisingset et al. (2016) modeled other pain scales (Numerical Rating Scale and NDI) by regression with numerous mobility and motor control variables for different parts of the body.

But all those models left out individual-specific variability, which were included in this study as “random” effects. The discrete nature of the VAS scores was also considered, using appropriate statistical methods for ordinal outcomes. Yet another difference with respect to the previous literature is that this study did not use mobility measures based on single head movements, but continuous cyclical motions that facilitated the analysis of movement repeatability and dynamic features like harmonicity, which turned out to be a significant factor (Baydal-Bertomeu et al., 2011).

The fit of these models can be compared with the ones reported in previous studies. Pearson's correlation coefficients between −0.16 and −0.58 have been reported for ROM or peak velocity against VAS (Kasch et al., 2001; Sarig Bahat et al., 2014) or NPQ (Chiu et al., 2005; Howell et al., 2012), which is less than the correlation obtained with



	NPQ (abs. value)	NPQ (variation)	VAS (abs. value)	VAS (variation)
Mean absolute error	6.07	4.34	1.23	0.81
Error < MCSD	59.8%	77.8%	87.0%	97.6%

Fig. 5. Multiple correlation coefficients and mean absolute error of the models. Correlation coefficients are shown for the “Full model” containing all fixed and random effects, “All fixed effects” (i.e. excluding subject-specific random effects), and “Basic fixed effects” (excluding kinematic parameters). Mean absolute errors and the proportion of errors smaller than the MCSD are given for the “All fixed effects” models.

the scores estimated from those and other kinematic variables. Meisingset et al. (2016) obtained models with R^2 between 0.19 and 0.25, which would account for MCCs between 0.44 and 0.50, comparable to the contribution of neck motion parameters alone in this study. By adding other controlled factors, like time or baseline pain scores, better fits were obtained.

4.4. Limitations

The sample of participants and the analyzed variables were a realistic representation of the patients that follow a rehabilitation program and the measurements that are taken during its administration in a clinic. The downside of that study methodology is that the characteristics of the patients and their treatments were not randomized, and their behavior during measuring sessions might be conditioned by fear of pain, and possible litigation of their case with potential secondary gain.

There are more scales that can be used for the assessment of neck pain, but this study focused on the analysis of NPQ and VAS, which were incorporated to the routine of the clinic that provided the data. Likewise, there are other physical measures that have been reported to relate to pain scores, like ROM, velocities and jerk of single neck movements, upper limb mobility and strength, and postural sway, while this study focused on the kinematics of cyclic neck movements, which Baydal-Bertomeu et al. (2011) reported to have a good discriminative power to differentiate between people with no history of neck pain, WAD patients with chronic pain, and recovered WAD patients trying to reproduce learnt patterns associated to neck pain.

Although the models include the effects of successive rehabilitation sessions, only three longitudinal measures were taken, and the time passed between the accident and each of the three sessions was not the same for all patients. All measures were taken within the first 3 months after the accident, before chronic pain could be diagnosed, but the second and third sessions were normally after the fourth week, so they could be considered as measures in the “sub-acute” period according to some definitions of pain time scales (Nyirö et al., 2017). The rehabilitation program consisted of physiotherapy interventions, which were adapted to the needs of each patient.

All these issues were sources of uncontrolled subject-specific variability, which was accounted for in the “random” factors of the models, but may have distorted some results of the study. On the other hand, the results obtained in such real-life circumstances, which are comparable to those reported in other studies with different profiles of patients suffering from neck pain, support the suitability of the methodological and statistical approach of our study.

4.5. Conclusion

We may draw some conclusions from this study: it is possible to find consistent relationships between PROMs or their variations and time, baseline values and kinematic parameters; and more than 66% part of such relationships can be attributed to objective kinematic parameters in neck motion tests. All in all, neck motion analysis is a useful objective tool to estimate part of the course of pain-related disability in WAD patients during the first months of rehabilitation.

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Appendix A. Supplementary data

Supplementary data related to this article can be found at https://doi.org/10.1016/j.msksp.2018.09.004.

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