Original article

# Multiple diagnoses, increased kinesiophobia? - Patients with high kinesiophobia levels showed a greater number of temporomandibular disorder diagnoses

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ABSTRACT

Objectives: The aim of this study was to empirically derive subgroups according to pain-related fear of movement beliefs using cluster analysis within a sample of TMD patients and asymptomatic volunteers.

Methods: 129 volunteers participated in this cross-sectional study (34.78, standard deviation [SD]: 12.49 years; 92 TMD patients and 37 symptom-free volunteers). Mechanical pain sensitivity through pressure pain threshold (PPT) on orofacial and remote sites, kinesiophobia, pain catastrophizing, anxiety and depression were assessed. A cluster analysis was used to derive subgroups according to kinesiophobia scores (TSK/TMD).

Results: Three subgroups were derived: cluster 1 (high kinesiophobia [n = 53], TSK score: 33, SD[standard deviation] = 2.9), cluster 2 (moderate kinesiophobia [n = 50], TSK score: 26.2, SD = 2.14) and cluster 3 (no/ low kinesiophobia [n = 26], TSK score 12.12, SD = 2.08) which included patients with higher overall PPT and lower scores on psychosocial variables. The group with high kinesiophobia showed high levels of pain catastrophizing, anxiety, and orofacial pain-related disability compared to the other subgroups and mechanical pain hyperalgesia in remote site compared to the low-kinesiophobia group. Also, we found a greater prevalence of triple diagnosis for the high-kinesiophobia subgroup compared to the moderate kinesiophobia group - odds ratio: 12.6 (95% confidence interval [CI]: 3.31–43.52, p < 0.01).

Conclusion: These results suggested that patients with TMD and higher levels of kinesiophobia beliefs may show a more complex clinical feature, with high psychosocial distress, widespread mechanical pain sensitivity, and a more complex TMD disorder. In this way, we suggest a relationship between the number of TMD diagnoses and kinesiophobia severity.

## 1. Introduction

Temporomandibular disorders (TMD) are complex and multifactorial conditions which affect masticatory muscles and/or the temporomandibular joint (TMJ), and may result in pain and disability (Mujakperuo et al., 2010). The prevalence of TMD ranges between 5 and 12% in the general population (Schiffman et al., 2014). Diagnostic criteria for TMD with simple, clear, reliable, and valid operational definitions are needed for accurate physical diagnosis in clinical and research settings (Schiffman et al., 2014). The Research Diagnostic

Criteria for Temporomandibular Disorders (RDC/TMD) has been the most widely employed diagnostic protocol since its publication in 1992 (Dworkin and LeResche, 1992). However, there was a need to investigate the accuracy of the diagnostic algorithms of the RDC/TMD in terms of reliability and criterion validity (Schiffman et al., 2014). Therefore, the RDC/TMD was revised and a new Diagnostic Criteria for TMD (DC/TMD) was proposed (Schiffman et al., 2014). Before this new diagnostic criteria can be used to evaluate individuals from other countries and different cultures, it must be translated into the intended language and culturally adapted to the country in which it will be used

(Guillemin et al., 1993; Beaton et al., 2000). However, it is still not available in the Brazilian Portuguese language.

TMDs are caused by altered pain mechanisms in the peripheral or central nervous system, and may be characterized by generalized hypersensitivity of the somatosensory system to both noxious and non-noxious stimuli (Nijs et al., 2017). In addition, the Orofacial Pain - Prospective Evaluation and Risk Assessment emphasized the importance of psychosocial factors as preditors of TMD incidence and highlighted the need to identify subgroups of TMD patients according to biological and psychosocial features (Slade et al., 2016).

Higher psychological and affective distress, elevated levels of depression, greater perceived stress and catastrophizing, and increased somatic awareness are commonly found in patients with TMD (Fillingim et al., 2011; Kothari et al., 2017). Several studies have focused on psychosocial factors in TMD (Kothari et al., 2017; Velly et al., 2002; Suvinen et al., 2005a; Bair et al., 2016). However, in contrast to other musculoskeletal disorders, kinesiophobia is an undervalued factor in TMD, and no study has used kinesiophobia to identify subgroups of TMD patients.

Kinesiophobia or pain-related fear of movement, is a psychological construct within the fear-avoidance model of pain (Leeuw et al., 2007; Vlaeyen et al., 2016). This model was developed in an attempt to identify the factors which act to maintain the vicious cycle of persistent pain. People with predisposing psychological profiles (negative affect), may develop pain catastrophizing, which may exarcebate fear and activity avoidance (Leeuw et al., 2007; Linton et al., 2011). This results in disability and deconditioning, contributing to pain chronification and preventing recovery (Leeuw et al., 2007; Linton et al., 2011). Psychosocial factors may be key to the development of persistent pain. The Tampa Scale of Kinesiophobia (TSK) was adopted to assess fear of movement in TMD patients. It was reported that kinesiophobia is associated with clinical features such as pain, joint sounds, and jaw locking (Visscher et al., 2010). The use of cut-off values to distinguish levels of fear of movement beliefs may help clinicians to better understand phenotypes and manage TMDs. To the best of our knowledge, no study has identified subgroups of TMD patients based on pain-related fear of movement and investigated the resulting profiles according to pain sensitivity, clinical features, and other psychosocial aspects.

We therefore aimed to empirically identify subgroups according to pain-related fear of movement beliefs using cluster analysis in a group of TMD patients and a group of asymptomatic volunteers, and to describe the characteristics of the resulting clusters. Additionally, we aimed to classify clusters using the remaining psychosocial factors, mechanical pain sensitivity, subtypes of TMD, anthropometry, and clinical variables. We hypothesized that high kinesiophobia scores would be associated with high global psychosocial distress and increased mechanical pain sensitivity (low-pressure pain threshold [PPT]). Ultimately, we hypothesized to find more mixed TMD diagnosis in the subgroup with high kinesiophobia.

## 2. Materials and methods

### 2.1. Study participants

This is a cross-sectional study and data was collected between August 2015 and November 2016 in a local Orofacial Pain clinic. Female volunteers aged from 18 to 60 years were recruited. Only female volunteers were recruited due to the low number of males who seek treatment in the clinic. A convenience sample of 129 volunteers was recruited with mean age: (34.78, standard deviation [SD]: 12.49 years). This sample comprised of 92 TMD patients and 37 symptom-free volunteers, who were recruited from the university staff. For the TMD group, only patients who were diagnosed with painful TMD according to the Research Diagnostic Criteria for Temporomandibular Disorders (Dworkin and LeResche, 1992), by a professional dentist experienced in using the criteria, and additionally experienced pain within the last

three months were recruited. Participants with only one diagnosis of disc displacement (non-painful diagnosis according to RDC/TMD) were not considered in the study. However, these participants were recruited if the non-painful diagnosis was in association with a painful one. In addition, volunteers with a clinical history of tumors in the craniofacial region, volunteers with acute musculoskeletal disorders, volunteers who recently underwent dental surgery, volunteers with infections, volunteers with whiplash-associated disorders, and volunteers with systemic chronic degenerative inflammatory or neurologic disorders were excluded from this study. The symptom-free group comprised of women who were free of any orofacial pain, with neither a current nor a past history of painful TMD or other forms of chronic orofacial pain. Individuals aged between 18 and 60 years were enrolled. For the symptom-free group, (1) individuals with abnormal cognitive function and communication skills; (2) individuals who used pain medication daily; and (3) individuals who experience headaches or have fibromyalgia or other chronic painful conditions were excluded. Symptom-free participants were enrolled in the present study as controls for comparison with the TMD group.

The study was conducted in accordance with the Declaration of Helsinki and was approved by a local Ethics Committee, process number: 201/2015. Informed consent was obtained from each study participant.

### 2.2. Instruments

#### 2.2.1. Clinical instruments
*2.2.1.1. Temporomandibular Disorder assessment – Research Diagnostic Criteria for Temporomandibular Disorders (RDC/TMD).* The TMD was evaluated using axis I of the RDC/TMD (Dworkin and LeResche, 1992). The RDC/TMD is a noninvasive clinical evaluation instrument that has been widely used in the literature; thus, levels of reliability and validity of most items and diagnoses have been previously assessed (Look et al., 2010). The same experienced examiner conducted the TMD assessment for all patients.

*2.2.1.2. Pain intensity.* To assess pain intensity, a numerical pain rating scale (NPRS) was used. The NPRS ranged from 0 to 10 (0 = "no pain" and 10 = "worst pain imaginable"). Evidence supports the reliability and validity of the NPRS. Furthermore, the NPRS may be slightly more sensitive than the other pain measures (Visual Analogue Scale, Verbal Rating Scale, and the Faces Pain Scale-Revised) (Ferreira-Valente et al., 2011).

#### 2.2.2. Psychosocial instruments
Kinesiophobia beliefs also called fear of movement beliefs, commonly observed in patients with chronic pain, are related to several other constructs including anxiety, depression, and pain catastrophizing as described in the fear-avoidance model (Leeuw et al., 2007; Vlaeyen et al., 2016). Moreover, previous findings showed that disability in TMD is associated with depression and catastrophizing (Velly et al., 2011), and anxiety and depression are prevalent in TMD (Bertoli and de Leeuw, 2016). There are questionnaires available in the literature that reliably evaluate these constructs. Thus, these questionnaires were applied and completed by self-report.

*2.2.2.1. Tampa scale for kinesiophobia for Temporomandibular Disorders (TSK/TMD).* The TSK/TMD is a self-report questionnaire that assesses beliefs regarding fear of movement (kinesiophobia), activity avoidance, general health fears and fear of (re)injury (Visscher et al., 2010). In this study, the TSK/TMD comprising of 12 items validated in the Brazilian Portuguese language was used (Aguiar et al., 2017). Each item is scored on a four-point ordinal scale, ranging from 'strongly disagree' (score = 1) to 'strongly agree' (score = 4). Ratings are summed to yield a total score, where higher scores reflect greater fear of movement (12–48 points). The TSK/TMD Brazilian Portuguese version (TSK/TMD

– Br) has two domains: the activity avoidance domain, comprising of seven questions (scores ranging from 7 to 28) and the somatic focus domain, which comprises five questions (scores ranging from 5 to 20).

*2.2.2.2. Anxiety and depression – hospital anxiety and depression scale (HADS).* The HADS is used to identify symptoms of anxiety and depression among patients in non-psychiatric hospital settings. It was translated into Brazilian Portuguese and validated (Pais-Ribeiro et al., 2007). The HADS contains 14 items; seven relate to anxiety (0–21 points) and the remaining seven relate to depression (0–21 points). A cut-off of $\geq 8$ was described, with good sensitivity and specificity values (0.70–0.90) for anxiety and depression symptoms (Bjelland et al., 2002).

*2.2.2.3. Pain catastrophizing scale (PCS).* The Brazilian Portuguese PCS (Sehn et al., 2012) is a self-administered questionnaire that consists of 13 items used to assess pain catastrophizing. It is divided into three domains: helplessness, magnification, and rumination. In the original PCS, items are rated on a five-point ordinal scale: (0) not at all, (1) to a slight degree, (2) to a moderate degree, (3) to a great degree, and (4) all the time. The Brazilian Portuguese PCS (B-PCS) total score ranges from 0 to 52 points; higher values indicate greater pain catastrophizing. Acceptable values for validity, internal consistency, and test-retest reliability were described for the Brazilian PCS (Sehn et al., 2012).

### 2.2.3. Mechanical pain sensitivity

*2.2.3.1. Pressure pain sensitivity – pressure pain threshold (PPT).* Evaluation of participants' widespread mechanical hyperalgesia was performed using pressure algometry, a technique that quantitatively assesses PPT in individuals with or without musculoskeletal pain symptoms (Farella et al., 2000). Pressure algometry enables the control of speed and direction of the pressure applied.

The PPT evaluation was performed by a previously trained researcher to ensure the application of constant pressure of approximately $0.5 \, kg/cm^2/s$ and the correct positioning of the metallic tip of the equipment. PPTs were measured using a 1-cm (Schiffman et al., 2014) flat-tipped algometer device (Kratos, model DDK-20 kgf, Cotia, Brazil, 2013) perpendicular to the anatomical areas evaluated. All the procedures were performed as previously described (Chaves et al., 2007).

The areas evaluated by algometry were: 1) the midpoint of the thenar region of the nondominant hand; 2) the anterior fibers of the temporalis muscle, 3) the middle fibers of the temporalis muscle, 4) the posterior fibers of the temporalis muscle, 5) the masseter muscle origin (zygomatic arch), 6) the masseter muscle belly, 7) the masseter muscle insertion (masseteric tuberosity), 8) the TMJ, 9) the midpoint on the upper trapezius muscle, 10) the origin of the sternocleidomastoid muscle (mastoid process), and 11) the suboccipital muscles (spinous process of vertebra C2). The PPT evaluation of the masseter and temporalis muscles followed Álvarez-Méndez et al. protocol (Álvarez-Méndez et al., 2017), in which the muscles were divided and numbered into 15 anatomical sites (3 vertical x 5 horizontal for the masseter muscle and 5 vertical x 3 horizontal for the temporalis muscle). In our assessment, we applied the mechanical stimulus on the sites 2, 8 and 14 for temporalis muscle (posterior, middle and anterior fibers, respectively) sites 2, 8 and 14 for masseter muscle belly according to the anatomical map of the face described by Álvarez-Méndez (Álvarez-Méndez et al., 2017).

*2.2.3.2. Statistical analysis.* Missing data values were handled by imputation. Only 5% of the values were missing, particularly values for the self-reported variables.

A cluster analysis was performed to categorize the participants (TMD patients and asymptomatic volunteers) according to their kinesiophobia scores since, despite the existence of several cut-offs

described in the literature for fear-avoidance scores (Wertli et al., 2014a), there are no clearly defined and valid cut-offs for grading kinesiophobia (Wertli et al., 2014a; Neblett et al., 2016). Because of the aforementioned reasons, we adopted an unsupervised statistical method to identify severity subgroups. We used an exploratory hierarchical agglomerative cluster analysis involving the Ward's clustering method and squared Euclidean distances to identify unique subgroups based on the total kinesiophobia score on the TSK/TMD. Whereas we assumed that it would be clinical meaningful to classify participants in three subgroups of kinesiophobia (high, moderate and low kinesiophobia), we pre-defined a 3-cluster solution aiming to obtain the three subgroups.

A discriminant function analysis with a stepwise method was used to determine the accuracy (cross-validation) of the cluster subgroups. It is a multivariate statistical procedure used to determine whether a set of variables can predict group membership (Brown and Wicker, 2000).

To validate the cluster subgroups clinically, differences in anthropometry, social and clinical variables, psychosocial factors, and mechanical pain sensitivity were examined using the one-way analysis of variance (ANOVA; Bonferroni post-hoc analysis). The chi-square test was used to analyze percentage variables ($p < 0.05$).

The strength of the association between the levels of kinesiophobia and the presence or absence of single, dual or triple TMD diagnoses was expressed with odds ratios (ORs) and 95% confidence intervals (CI). ORs with CIs above 1.0 suggest a significantly greater likelihood of occurrence and those below 1.0 suggest a significantly lower likelihood of occurrence.

In addition, to investigate the correlation between variables, the Spearman's rank correlation was used ($p < 0.05$). The strength of the correlations was determined as follows: $R < 0.39$ = weak; $0.4 < R < 0.69$ = moderate; $R > 0.7$ = strong (Dancey and Reidy, 2004). A point-biserial correlation was used to correlate continuous and dichotomous variables. It is a special case of the Pearson's product-moment correlation.

The software program SPSS was used for statistical analysis (SPSS Statistics for Windows, Version 22.0, Armonk, NY: IBM Corp, 2013).

## 3. Results

### 3.1. Empirically derived subgroups according to kinesiophobia

The cluster analysis identified three subgroups: Cluster 1 (high kinesiophobia [n = 53]), which comprised of TMD patients and asymptomatic volunteers with high scores for kinesiophobia beliefs (TSK/TMD scores ranged from 30 to 43); Cluster 2 (moderate kinesiophobia [n = 50]), which comprised of TMD patients and asymptomatic volunteers with moderate scores for kinesiophobia beliefs (TSK/TMD scores ranged from 22 to 29); and Cluster 3 (no/low kinesiophobia [n = 26]), which comprised of only asymptomatic volunteers with no kinesiophobia (a score of 12 on the TSK/TMD means that the participant answered disagree to all questions) or low scores for kinesiophobia beliefs (TSK/TMD scores ranged from 12 to 18). The 3-cluster solution identified is depicted in Fig. 1.

### 3.2. Discriminant analysis

Validation of the cluster solution was performed using discriminant function analysis with TSK/TMD scores simultaneously included in the analysis: 1) activity avoidance score (Wilk $\lambda = 0.14$, $p < 0.001$) and 2) focus somatic score (Wilk $\lambda = 0.11$, $p < 0.001$). Two discriminant functions were extracted, with significant results only for function 1 ($\chi$ (Schiffman et al., 2014)$_4$ = 305.342, $p < 0.001$, Wilk $\lambda = 0.088$). These findings showed that the two-domains of the kinesiophobia scale were able to discriminate the subgroups into high-, moderate-, and low/no-kinesiophobia subgroups. Function 1 accounted for 99.9% (canonical correlation = 0.95) of the total relationship between variables
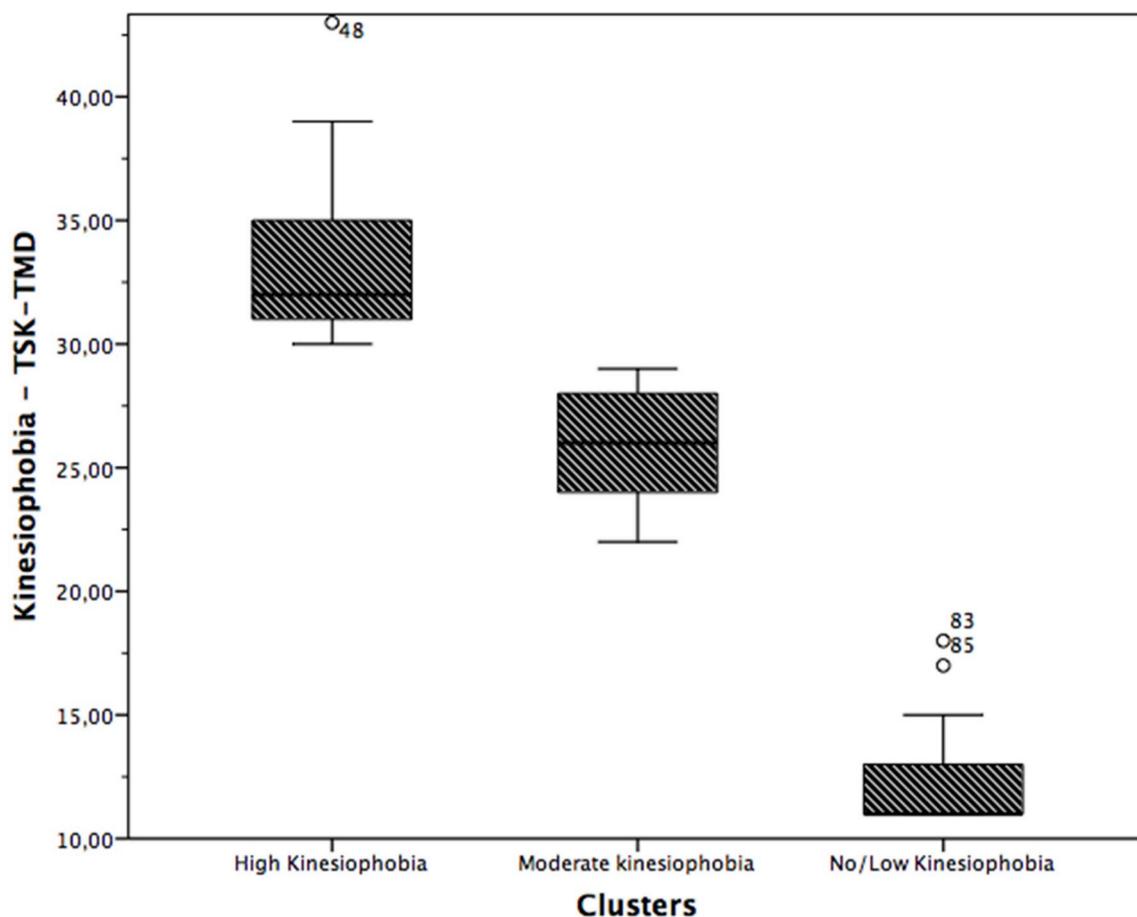
**Fig. 1.** Cluster subgroups according to high (n = 53), moderate (n = 50) and no/low kinesiophobia (n = 26) (total score TSK/TMD). The circles represent the outliers (cases: 48, 83 and 84).

and cluster groups with an eigenvalue of 10.28. The discriminant function canonical standardized coefficients for the activity avoidance domain was 0.53 and that for the focus somatic domain was 0.69. Cross-validation classification showed that the function was able to correctly classify 99.2% of the participants included in the analysis.

### 3.3. Differences between cluster subgroups

Anthropometry and social and clinical features were analyzed in the cluster subgroups. The no/low-kinesiophobia subgroup had spent more years in school. Higher levels of orofacial pain-related disability were reported by the high-kinesiophobia subgroup compared to the other subgroups. However, for pain-free jaw opening, lower values were obtained for the high-kinesiophobia subgroup compared to the no/low-kinesiophobia subgroup only. In addition, higher scores for pain intensity, years living with pain, headaches which interfered with jaw activities, and low range of motion for protrusion and lateral deviation were observed for both high and moderate kinesiphobia subgroups compared to the no/low-kinesiophobia subgroup (Table 1).

We found significant differences in catastrophizing, anxiety, depression, and mechanical pain sensitivity among the three clusters (Table 2). Pairwise comparisons revealed that the high-kinesiophobia subgroup had higher pain catastrophizing and anxiety scores compared to the moderate- and no/low-kinesiophobia subgroups. Specifically, for the thenar region, a significantly lower mean PPT was observed for the high-kinesiophobia subgroup compared to the no/low-kinesiophobia subgroup. We found significantly lower values of PPT on orofacial sites and greater depression scores for both high- and moderate-kinesiophobia subgroups compared to the no/low-kinesiophobia subgroup (Table 2).

### 3.4. Differences between cluster subgroups based on TMD diagnosis

A high prevalence of mixed diagnosis (concomitant muscle and joint diagnoses) was observed for the moderate- and high-kinesiophobia subgroups. However, when the mixed diagnoses were separated into dual and triple diagnosis, we found a higher prevalence of dual diagnosis of Myofascial pain + Disc displacement with reduction in the moderate-kinesiophobia subgroup compared to the high-kinesiophobia subgroup. Additionally, we found a greater prevalence of triple diagnosis of Myofascial pain (with/without limited opening) + Disc displacement (with/without disc reduction) + arthralgia in the high-kinesiophobia subgroup compared to the moderate-kinesiophobia subgroup (Table 3). The presence or absence of a single TMD diagnosis was equal in the high or moderate kinesiophobia subgroups (Table 3). On the other hand, the chance for a dual diagnosis was lower in the high kinesiophobia subgroup compared with the moderate kinesiophobia subgroup (OR = 0.33, an odds of 1 to 3). Also, the chance of the presence of a triple diagnosis was 12.6 times higher for the high-kinesiophobia subgroup than for the moderate-kinesiophobia subgroup (Table 3).

### 3.5. Correlations between clinical findings and kinesiophobia scores in TMD patients

A weak/moderate positive correlation was observed between orofacial pain disability and kinesiophobia in the TMD patients. Weak negative correlations were also observed between protrusive excursion and mechanical pain sensitivity in the thenar region, and activity avoidance domain score and TSK/TMD total scores. Additionally, weak

**Table 1**
Mean values, standard deviations (SDs) and percentage of clinical and anthropometric data between Temporomandibular Disorders subgroups.

| Characteristics | Cluster 1 – High kinesiophobia (n = 53) | Cluster 2– Moderate kinesiophobia (n = 50) | Cluster 3 – No/Low kinesiophobia (n = 26) | $F_{(2,126)}$ and $X^2$, p-value |
|---|---|---|---|---|
| Age (years) | 37.15 (12.26) | 34.52 (11.01) | 30.42 (14.71) | 2.61, p = 0.08 |
| Education level[b] | 4.06[c] (1.75) | 3.94[c] (1.68) | 5.38 (1.65) | 6.92, p < 0.01 |
| Weight (kg) | 66.35 (11.16) | 67.27 (12.71) | 62.24 (10.17) | 1.68, p = 0.19 |
| Height (m) | 1.63 (0.06) | 1.62 (0.05) | 1.62 (0.05) | 0.15, p = 0.86 |
| Pain intensity (mean last 30 days) | 5.77[c] (1.35) | 4.96[c] (2.44) | 0 | 100.49, p < 0.01 |
| Years living with orofacial pain | 6.28[c] (5.71) | 5.72[c] (7.80) | 0 | 10.19, p < 0.01 |
| Orofacial pain disability[a] | 3.43[d] (1.05) | 2.68[c] (1.50) | 0.04 (0.20) | 76.54, p < 0.01 |
| Headache report (last 30 days) | 81%[c] (n = 43) | 60%[c] (n = 30) | 18% (n = 5) | 18.7, p < 0.001 |
| Headache interfering with jaw activities[a] | 2.11[c] (1.80) | 1.44[c] (1.64) | 0 | 16.27, p < 0.01 |
| Years living with headache | 8.93[c] (8.64) | 6.08[c] (8.73) | 1.69 (4.58) | 7.13, p < 0.01 |
| Pain free mouth opening (mm) | 32.13[c] (7.51) | 33.62 (8.39) | 36.77 (7.36) | 3.06, p = 0.05 |
| Maximum mouth opening (mm) | 38.30 (7.27) | 39.94 (7.60) | 40.81 (7.28) | 1.18, p = 0.31 |
| Protrusion (mm) | 4.72[c] (2.51) | 5.20[c] (2.40) | 7.96 (3.29) | 13.90, p < 0.01 |
| Lateral deviation (mean right and left) (mm) | 6.81[c] (2.51) | 7.12[c] (3.27) | 8.92 (1.85) | 5.54, p = 0.01 |

[a] Four jaw functional activities: chewing, mouth opening, clenching/grinding and jaw activities (talk, kiss) (score range 0–4, 0-no pain, 1 = pain report for each question).
[b] Education level: 1 = primary level incomplete, 2 = primary level complete, 3 = secondary level incomplete, 4 = secondary level complete, 5 = technical education, 6 = tertiary education incomplete, 7 = tertiary education complete and 8 = post-graduation.
[c] Difference in relation to cluster 3 (Bonferroni correction, p ≤ 0.05).
[d] Difference in relation to the other groups (Bonferroni correction, p ≤ 0.05).

negative correlations between orofacial and neck mechanical pain sensitivity and the somatic focus domain score and weak/moderate positive correlations between orofacial pain disability and TSK/TMD total score were observed (Table 4) in the TMD patients.

## 4. Discussion

The aim of this study was to empirically identify subgroups based on kinesiophobia using cluster analysis in a sample of TMD and asymptomatic volunteers. We also aimed to describe the profiles of subgroups according to clinical features, other psychosocial factors, and pain sensitivity. Three subgroups were identified: cluster 1, comprised of participants with high kinesiophobia scores (mean TSK/TMD score 68%); cluster 2, comprised of participants with moderate kinesiophobia scores (mean TSK/TMD score = 54%) and cluster 3, comprised only of asymptomatic volunteers with low kinesiophobia scores (mean TSK/TMD score = 25%). In summary, the high-kinesiophobia subgroup comprised of patients with high scores for psychosocial variables and

**Table 2**
Comparison of psychosocial and pain sensitivity variables between subgroups clustered according to kinesiophobia (n = 129). TSK: Tampa Scale of Kinesiophobia for Temporomandibular Disorders (TMD), PCS: Pain Catastrophizing Scale, HADS: Hospital Anxiety Depression Scale.

| | Cluster 1 – High kinesiophobia (n = 53) | Cluster 2 – Moderate kinesiophobia (n = 50) | Cluster 3 – No/Low kinesiophobia (n = 26) | $F_{(2,126)}$ and p-values |
|---|---|---|---|---|
| **Variables included in the cluster analysis** | | | | |
| **Kinesiophobia** (TSK/TMD: 12–48 points) | 33.00[b] (2.90) | 26.20[a] (2.14) | 12.12 (2.08) | 621.96/p < 0.001 |
| Activity Avoidance | 18.19[b] (1.85) | 14.32[a] (1.68) | 6.85 (1.54) | 375.72/p < 0.001 |
| Somatic Focus | 14.81[b] (1.57) | 11.88[a] (1.08) | 5.27 (0.87) | 489.66/p < 0.001 |
| **Psychosocial and Clinical variables – characterization** | | | | |
| **Catastrophizing** (PCS total: 0–52) | 33.92[b] (9.32) | 24.36[a] (11.51) | 3.27 (7.22) | 83.95/p < 0.001 |
| Magnification | 8.02[b] (2.66) | 5.84[a] (3.52) | 0.69 (1.38) | 57.64/p < 0.001 |
| Rumination | 11.30[b] (3.09) | 8.94[a] (3.85) | 1.12 (2.67) | 82.75/p < 0.001 |
| Hopelessness | 14.60[b] (4.65) | 9.58[a] (5.30) | 1.46 (3.23) | 69.08/p < 0.001 |
| **Anxiety** (HADS: 0–21 points) | 10.83[b] (4.62) | 8.02[a] (3.53) | 4.69 (3.28) | 21.41/p < 0.001 |
| **Depression** (HADS: 0–21 points) | 7.94[a] (4.87) | 5.98[a] (3.90) | 2.35 (2.98) | 15.64/p < 0.001 |
| **PPT – thenar region** (kpa) | 310.22[a] (88.80) | 331.63 (94.30) | 378.27 (96.69) | 4.71/p = 0.01 |
| **PPT – neck sites (overall)** (kpa) | 217.46 (87.80) | 214.20 (78.29) | 236.89 (72.39) | 0.70/p = 0.48 |
| **PPT – orofacial sites (overall)** (kpa) | 177.65[a] (80.82) | 176.55[a] (73.89) | 229.11 (49.75) | 5.28/p = 0.01 |
| Anterior Temporalis | 212.85 (103.86) | 198.75[a] (89.71) | 260.81 (62.96) | 4.03/p = 0.02 |
| Middle Temporalis | 207.10[a] (104.89) | 209.07[a] (92.94) | 271.43 (62.62) | 4.81/p = 0.01 |
| Posterior Temporalis | 209.35[a] (95.71) | 216.42[a] (104.76) | 274.08 (62.29) | 4.50/p = 0.01 |
| Masseter – origin | 174.40 (96.24) | 174.62 (84.35) | 224.60 (74.21) | 3.39/p = 0.04[c] |
| Masseter – belly | 149.45[a] (79.48) | 146.50[a] (64.37) | 191.73 (54.62) | 4.15/p = 0.02 |
| Masseter – insertion | 159.03 (96.23) | 157.27 (86.00) | 197.01 (71.98) | 2.03/p = 0.14 |
| Temporomandibular joint (TMJ) | 131.39[a] (68.80) | 133.23[a] (62.30) | 184.15 (38.87) | 7.43/p < 0.01 |
| Upper trapezius | 243.32 (122.96) | 229.95 (94.66) | 263.18 (87.50) | 0.85/p = 0.43 |
| Sternocleidomastoid | 203.68 (84.98) | 194.51 (73.06) | 226.28 (77.06) | 1.39/p = 0.25 |
| Suboccipitalis | 205.38 (90.04) | 218.15 (93.52) | 221.20 (73.71) | 0.39/p = 0.68 |

[a] Difference in relation to cluster 3 (Bonferroni Correction, p ≤ 0.05).
[b] Difference in relation to cluster 2 and 3 (Bonferroni Correction, p ≤ 0.05).
[c] Anova showed a significant main effect, but the post hoc correction did not support the results from ANOVA.

**Table 3**
Comparisons (percentage) of diagnostic subtypes between Temporomandibular Disorders (TMD) subgroups according to kinesiophobia scores (n = 103).

| TMD Diagnosis | Cluster 1 – High kinesiophobia (n = 53) | Cluster 2 – Moderate kinesiophobia (n = 50) | $X^2_{(2, 103)}$/p-value |
|---|---|---|---|
| Mixed TMD | 82% (n = 43) | 82% (n = 41) | 0.02, p = 0.86 |
| Myogenous | 17% (n = 9) | 15% (n = 7) | 0.12, p = 0.72 |
| Arthrogenous | 2% (n = 1) | 2% (n = 1) | 0, p = 1 |
| Asymptomatic | 6% (n = 3) | 16% (n = 8) | 1.15, p = 0.27 |
| **One diagnosis** | | | |
| Myofascial pain | 17% (n = 9) | 15% (n = 7) | 0.12, p = 0.72 |
| Arthralgia/Osteoarthritis | 2% (n = 1) | 4% (n = 2) | 0, p = 1 |
| One diagnosis (total prevalence) | 18% (n = 10) | 18% (n = 9) | 0.02, p = 0.88 |
| Odds Ratio one diagnosis (95%CI) | *1.09 (0.38-2.86), p = 0.91* | | |
| **Two diagnoses** | | | |
| Myofascial pain + arthralgia/Osteoarthritis | 17% (n = 9) | 10% (n = 5) | 0.58, p = 0.44 |
| Myofascial pain + DDwR | 2% (n = 1) | 28%* (n = 14) | *6.81, p < 0.01* |
| Arthralgia + DDwR, with/without limited opening | 11% (n = 6) | 20% (n = 10) | 0.51, p = 0.47 |
| DDwR + Osteoarthritis | 2% (n = 1) | 0 | NA |
| Two diagnoses (total prevalence) | 32% (n = 17) | 58%* (n = 29) | 5.98, p = 0.01 |
| Odds Ratio** two diagnoses (95%CI) | *0.33 (0.15-0.76), p < 0.01* | | |
| **Three diagnoses** | | | |
| Myofascial pain (with/without limited opening) + DDwR/DDwoutR + arthralgia | 38%* (n = 20) | 4% (n = 2) | *6.99/p < 0.01* |
| Myofascial pain + DDwR + Osteoarthritis | 6% (n = 3) | 2% (n = 1) | 0.53, p = 0.46 |
| Three diagnosis (total prevalence) | 43%* (n = 23) | 6% (n = 3) | 17.13, p < 0.001 |
| Odds Ratio** three diagnoses (95%CI) | *12.6 (3.31-43.52), p < 0.01* | | |

* Chi-square (p < 0.05).

NA: Not Applicable, DDWR: Disk Displacement with Reduction; DDwoutR: Disk Displacement without Reduction.

**Odds Ratio $= \dfrac{[\text{cases with two/three diagnoses}_{\text{high kinesiophobia}}] - [\text{cases without two/three diagnoses}_{\text{high kinesiophobia}}]}{[\text{cases with two/three diagnoses}_{\text{moderate kinesiophobia}}] - [\text{cases without two/three diagnoses}_{\text{moderate kinesiophobia}}]}$

orofacial and non-orofacial mechanical hyperalgesia, and a high prevalence of triple diagnosis; the moderate-kinesiophobia subgroup comprised of patients with moderate scores for psychosocial variables and orofacial mechanical hyperalgesia, and a high prevalence of dual diagnosis; and finally, the no/low-kinesiophobia subgroup comprised solely of asymptomatic volunteers.

Our findings support our hypothesis which states that the high-kinesiophobia subgroup has higher likelihood to global psychosocial distress and mechanical hyperalgesia at orofacial and remote sites. One previous study (Suvinen et al., 2005b) reported three subgroups of TMD patients according to psychosocial variables; the first subgroup of patients experienced high levels of psychosocial distress, the second

subgroup experienced moderate levels of psychosocial distress, and the third subgroup experienced low levels of psychosocial distress. Interestingly, the study did not show differences among the TMD subgroups according to their clinical symptoms. Bair et al. (2016) clustered patients with TMD and asymptomatic participants using multiple variables (pain sensitivity and psychosocial variables). However, contrary to our results, they identified a TMD subgroup with high pain sensitivity and mild psychosocial distress scores, a subgroup (global symptoms) with both high pain sensitivity and psychosocial distress scores, and a subgroup called the adaptive subgroup, with low pain sensitivity and psychosocial distress scores. The difference in results between the aforementioned studies and the present study may be due to the

**Table 4**
Correlation between jaw function and pain sensitivity outcomes and kinesiophobia scores in patients with Temporomandibular Disorders (n = 103). AA: Activity Avoidance, FS: Somatic Focus, TSK: Tampa Scale of Kinesiophobia for Temporomandibular Disorders (TMD), PPT: Pressure Pain Threshold.

| Jaw function and pain sensitivity | kinesiophobia | | |
|---|---|---|---|
| | Activity avoidance | Somatic Focus | Total Score TSK |
| Locking jaw – not opening[b] | 0.01, p = 0.93 | −0.03, p = 0.77 | −0.01, p = 0.92 |
| Locking jaw interfering with eating[b] | 0.05, p = 0.66 | 0.01, p = 0.90 | 0.04, p = 0.71 |
| Locking jaw – last 30 days[b] | −0.01, p = 0.95 | 0.00, p = 0.99 | −0.01, p = 0.96 |
| Jaw locked – not closing[b] | 0.13, p = 0.24 | 0.09, p = 0.39 | 0.14, p = 0.20 |
| Any maneuver to close the jaw | −0.05, p = 0.62 | 0.02, p = 0.85 | −0.03, p = 0.81 |
| TMJ sounds | 0.13, p = 0.21 | 0.17, p = 0.12 | 0.16, p = 0.13 |
| Orofacial pain disability[c] | 0.37[a], p < 0.01 | 0.30[a], p = 0.03 | 0.42[a], p < 0.01 |
| Headache interfering with jaw activities[c] | −0.04, p = 0.74 | 0.03, p = 0.78 | −0.01, p = 0.94 |
| Lateral deviation to right (mm) | −0.19, p = 0.07 | −0.10, p = 0.36 | −0.19, p = 0.08 |
| Lateral deviation to left (mm) | −0.12, p = 0.26 | −0.06, p = 0.55 | −0.12, p = 0.27 |
| Protrusion (mm) | −0.28, p = 0.01[a] | −0.13, p = 0.24 | −0.16, p = 0.02[a] |
| Pain free jaw opening (mm) | 0.09, p = 0.42 | 0.01, p = 0.93 | 0.07, p = 0.54 |
| Maximum jaw opening (mm) | 0.16, p = 0.13 | 0.00, p = 0.99 | 0.11, p = 0.29 |
| PPT – neck sites (overall) (kpa) | −0.07, p = 0.49 | −0.23[a], p = 0.03 | −0.17, p = 0.10 |
| PPT – orofacial sites (overall) (kpa) | −0.13, p = 0.23 | −0.29[a], p = 0.01 | −0.24[a], p = 0.02 |
| PPT – thenar region (kpa) | −0.26[a], p = 0.01 | −0.13, p = 0.24 | −0.25[a], p = 0.02 |

[a] Spearman Correlation, $p \leq 0.05$.

[b] Pearson Product Correlation (Point biserial).

[c] Four jaw functional activities: chewing, mouth opening, clenching/grinding and jaw activities (talk, kiss) (score range 0–4, 0-no pain, 1 = pain report for each question).

variables used to identify the subgroups. Also, the aforementioned studies did not include fear-avoidance beliefs as a variable in the cluster analysis for identifying subgroups.

The high- and moderate-kinesiophobia subgroups showed increased mechanical pain sensitivity at orofacial sites, although TMD patients without nociplastic pain features may experience pain sensitivity at orofacial sites (Ballenberger et al., 2018). In addition, a previous systematic review (La Touche et al., 2018) reported that patients with TMD show decreased PPTs in trigeminal and remote areas. Remarkably, only the subgroup with high kinesiophobia demonstrated mechanical hyperalgesia in the non-orofacial region (thenar region of the hand). One can argue that nociplastic pain features may be predominant among patients with TMD and high levels of kinesiophobia **beliefs**. This suggests that a subgroup of TMD patients may exhibit hyperalgesia at remote sites which may be associated with mechanism(s) of central sensitization (Fernández-de las Peñas et al., 2009). As reported above, a previous report showed that higher psychosocial distress is associated with higher pain sensitivity only in a subgroup of TMD patients (Bair et al., 2016).

The fear-avoidance model (FAM) demonstrates that the development of fear-avoidance behavior increases the risk for disability and depression, which may favor the maintenance of the deleterious cycle of persistent pain (Leeuw et al., 2007; Vlaeyen et al., 2016; Vlaeyen and Linton, 2000), with increasing disability and pain. One can suggest that the resulting chronic process may favor changes in plasticity in nociceptive pathways during chronic pain (Woolf, 2001), resulting in increasing pain and the spread of the pain to remote areas. Two previous systematic reviews reported that fear-avoidance and pain intensity are positively correlated in patients with chronic musculoskeletal pain (Kroska, 2016; Luque-Suarez et al., 2018).

When the TMD subtypes were considered, no differences were observed among the kinesiophobia subgroups. Consistent with our study, Gil-Martínez et al. (2016) did not find differences in kinesiophobia scores among TMD patients with joint pain, muscle pain, and mixed diagnosis. A previous study reported no significant differences among TMD patients subgrouped using psychosocial factors such as physical/symptom features (face pain, difficulty or pain during mouth opening, and TMJ clicking or grating) (Suvinen et al., 2005b).

An interesting clinically relevant finding of the current study is the high prevalence of multiple TMD diagnoses according to the severity of kinesiophobia. High-kinesiophobia subgroup showed a lower likelihood of dual TMD diagnosis and a higher likelihood of triple TMD diagnosis compared with the moderate-kinesiophobia subgroup. To the best of our knowledge, no study has investigated the number of TMD diagnoses among subgroups according to psychosocial variables such as kinesiophobia. Additionally, our results suggest that fear of movement beliefs must be considered when managing patients with TMD and that high levels of fear of movement are associated with more complex TMD clinical features. Despite the fact that the cross-sectional nature of this study prevents the inference of a cause-effect relationship, one can speculate that the severity of the clinical frame (more diagnoses), may increase the risk for the development of greater fear of movement. The hypervigilance associated with the presence of more symptoms at orofacial sites may increase fear and avoidance behaviors, resulting in an increase in orofacial pain related-disability.

In the present study we found a statistically significant decrease in lateral deviation and protrusive range of motion in the TMD subgroups with moderate and high kinesiophobia compared to the control group. The mean differences in protrusion between the moderate- and high-kinesiophobia subgroups and the control group were greater than the smallest detectable change (SDC; 2–3 mm) reported by Julsvoll et al. (2017). However, for lateral deviation, only the TMD subgroup with high kinesiophobia showed a clinical meaningful reduction compared to the control group. For maximum mouth opening, no difference between groups was observed, although the subgroup with high-kinesiophobia showed a lower pain-free mouth opening compared to the

control group (Julsvoll et al., 2017). In addition, the restriction in the range of motion in TMD patients was slightly affected by the level of kinesiophobia particularly for protrusive movement (Table 4). Therefore, the range of motion restriction may be due to less exposure to movement, which can trigger a vicious cycle in which the patient is physically affected, leading to muscle deconditioning. FAM postulates that the final part of the fear-avoidance cycle represents the development of disuse and increased perceived disability (Vlaeyen and Linton, 2000). On the other hand, one can assume that the greater the complexity of the clinical picture, the greater the risk for range of motion limitation.

Surprisingly, no correlation between TMJ sounds and kinesiophobia was observed among TMD patients, contrary to the findings of Visscher et al. (2010). However, we observed a positive correlation between orofacial pain disability and kinesiophobia in TMD patients, suggesting that the higher the orofacial perceived pain disability, the higher the kinesiophobia beliefs. This finding was highlighted by a previous systematic review (Luque-Suarez et al., 2019) on chronic musculoskeletal pain and by a previous study in which kinesiophobia and neck pain disability explained 34% of orofacial pain disability (Beltran-Alacreu et al., 1992). Additionally, we found that high kinesiophobia scores, particularly related to activity avoidance, may be associated with mandibular range of motion limitation (or vice-versa) and heightened mechanical pain sensitivity in TMD patients. In accordance with our results, a previous study in chronic neck pain patients showed a negative, low correlation between mechanical pain sensitivity at a remote site and neck range of motion and kinesiophobia (Walton et al., 2014). Brown et al. (2016) showed that the kinesiophobia score may be a valuable tool for identifying patients at risk for decreased range of motion after Total Knee Arthroplasty. This suggests that maladaptive beliefs regarding movement may cause range of motion limitations, and not the inverse, i.e., range of motion restriction may result in increased kinesiophobia. Moreover, why did the results show a negative correlation only between kinesiophobia and protrusive movement? Considering that more than 50% of our sample were diagnosed with disc displacement with or without reduction, one can argue that during protrusive movement, the displaced disc works as an anatomical barrier, which inhibits the forward movement of both condyles. This interferes more specifically with protrusion due to the lack of rotation during such a movement. We can assume that the anatomical barrier imposed by the dislocated disc, which impairs movement, may contribute to the exacerbation of the belief that "*the movement would aggravate the TMJ injury.*" Additionally, it is possible that maladaptive beliefs may be more related to a perception of mechanical dysfunction in the TMJ, and not poorly related to hearing of TMJ weird sounds. However, this assumption is speculative and should be investigated in future studies.

Cut-off values to clinically classify fear avoidant beliefs were proposed in the literature using TSK-17: low risk (< 17 points), moderate risk (17–37 points), and high risk (> 37points) (Wertli et al., 2014b). A cut-off score of 54% on the TSK (TSK-17: 37/68 and TSK/TMD-12: 26/48) indicates relevant fear avoidance in chronic low back pain. By using such cut-off values, both subgroups in the present study with high and moderate kinesiophobia could be considered as relevant fear-avoiders Our study, using unsupervised cluster analysis, showed that among TMD patients, those with scores higher than 68% on the TSK-12 had worst clinical and psychological features than those with scores lower than 54%. This suggests that the cut-off values must be condition-specific.

Four psychosocial variables were analyzed in the present study. Two cognitive (catastrophizing and kinesiophobia) and two affective (anxiety and depression) factors (Pincus and McCracken, 2013). Catastrophizing, anxiety, and depression were higher for both high- and moderate-kinesiophobia subgroups compared to the asymptomatic group. However, the high-kinesiophobia subgroup showed worst scores for catastrophizing and anxiety, but not for depression. Conversely, a

previous study reported that the subgroup with high psychosocial distress scores had significantly higher scores for catastrophizing and depression, and not anxiety (Suvinen et al., 2005b). As we used kinesiophobia to identify subgroups among TMD patients, this may explain the differences between results. Another previous study in TMD patients reported that catastrophizing and depression contributed to the progression to chronic TMD and pain-related disability (Velly et al., 2011).

Considering the differences in anxiety and depression scores in the present study, one can assume that in patients with high levels of kinesiophobia, anxiety and depression states might be triggered by distinct mechanisms. The tripartite model of emotion helps explain common and distinct aspects of depression and anxiety, using three factors: positive affect, negative affect, and physiological hyperarousal (Clark and Watson, 1991). For instance, physiological hyperarousal is defined as increased activity in the sympathetic nervous system, in response to threat (Gencoz, 2000) and is unique to anxiety disorders (Clark and Watson, 1991). Considering pain as a threat and fear-avoidance as a possible behavior in response to pain, we can suggest that the greater the kinesiophobia, the greater the hyperarousal, which may influence anxiety regardless of depression. A previous study suggested alternative pathways for the fear-avoidance model in which depression is not necessarily a resulting factor from pain-related fear of movement and catastrophizing (Pincus et al., 2010).

Several limitations should be noted during the interpretation of our results. First, we included only female volunteers because of the limited availability of male TMD patients in the orofacial clinic. There is evidence in the literature regarding differences in the experience of pain between men and women (Bale and Epperson, 2015). Second, this study is a cross-sectional study. Therefore, it is impossible to assign a causal relationship between TMD severity (multiple diagnoses) and kinesiophobia levels. Future studies may investigate such a relationship using a longitudinal study design. Third, although there is a new Diagnostic Criteria for TMD (DC/TMD) in the literature, we could not use it because it is still not available in Brazilian Portuguese. Therefore, we cannot guarantee that future studies using DC/TMD to diagnose TMD patients will find similar results.

## 5. Conclusion

Our results showed that the high-kinesiophobia subgroup comprised of patients with high scores for psychosocial variables and orofacial and non-orofacial mechanical hyperalgesia, and greater prevalence of triple diagnosis. While the moderate-kinesiophobia subgroup comprised of patients with moderate scores for psychosocial variables and orofacial mechanical hyperalgesia, and greater prevalence of dual diagnosis, the no/low-kinesiophobia subgroup comprised solely of asymptomatic volunteers. These results suggest that patients with TMD and high levels of kinesiophobia beliefs may show more complex clinical features, including high psychosocial distress, widespread mechanical pain sensitivity, and a more complex TMD disorder. We conclude that there is a relationship between the number of TMD diagnoses and kinesiophobia severity.

## Funding sources

## Conflicts of interest

None declared.

## Ethical approval

This study was approved by ethics committee from Ribeirão Preto Medical School, from the University of São Paulo, process number 40470114.3.3001.5419.

## Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.msksp.2019.102054.

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