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Title: To what extent is bruxism associated with musculoskeletal signs and symptoms? A systematic review

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Article category: Review

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Summary

The aim of the present systematic review was to answer the overall research question: “To what extent is bruxism associated with musculoskeletal signs and symptoms?”. The review was performed in accordance with the PRISMA guidelines. A PubMed search of articles published until November 23, 2017 was conducted. The search string included both MeSH terms and text words. Results were presented in categories according to study design, study population (e.g., adults, children), bruxism sub-type (awake, sleep), assessment methods for bruxism and musculoskeletal symptoms (self-report, validated test), and type of outcome (pain, non-painful musculoskeletal symptoms). It could be concluded that bruxism is to some extent associated with musculoskeletal symptoms, even though the evidence is conflicting and seems to be dependent on many factors, such as age, whether the bruxism occurs during sleep or wakefulness, and also the quality of the diagnostic methodology regarding bruxism and musculoskeletal signs and symptoms. The literature does not support a direct linear causal relationship between bruxism and such symptoms, but points more in the direction of a multifaceted relationship dependent on the presence of other risk factors. Pain is by far the most commonly assessed symptom, whereas non-painful musculoskeletal symptoms have generally not been systematically evaluated. In the light of recent findings indicating that non-painful symptoms may precede TMD pain, it is suggested to increase the scientific focus on non-painful musculoskeletal symptoms in future studies. Also, future studies should use validated methods for case definition and outcome assessments.

Keywords

Bruxism, temporomandibular disorders, musculoskeletal symptoms, pain, unpleasantness, muscle fatigue

1. Background

Until recently, bruxism was defined as “a repetitive jaw-muscle activity characterized by clenching and grinding of the teeth and/or by bracing or thrusting of the mandible”¹. Bruxism activity may occur during wakefulness or sleep.¹ An updated definition was recently published, stating that “sleep and awake bruxism are masticatory muscle activities that occur during sleep (characterized as rhythmic or non-rhythmic) and wakefulness (characterized by repetitive or sustained tooth contact and/or by bracing or thrusting of the mandible), respectively”.² The possible relationship between bruxism and different musculoskeletal symptoms of the masticatory system remains controversial, and the literature is conflicting on this matter.^{3,4} Most often, a possible positive relationship between either awake or sleep bruxism and craniofacial pain has been hypothesized (for a review, please see³) and even sometimes presented as a true and simple cause-effect relationship in non-scientific literature.³ However, it has become increasingly clear that the association between bruxism and craniofacial pain is much more complex, and efforts have been made to understand painful temporomandibular disorders (TMD) along with other chronic pain conditions in a biopsychosocial context,⁵⁻⁸ thereby recognizing the important influence of psychosocial factors.

The study of the possible relationship between bruxism and craniofacial pain has been further challenged by the evolution of definitions and diagnostic criteria for bruxism.² More specifically, the concrete distinction between the two circadian manifestations of bruxism, i.e., awake and sleep, requires that the conditions are separately assessed.¹ Moreover, although the diagnostic criteria for sleep and awake bruxism are still a work in progress, since the 2013 proposal of a bruxism diagnostic grading system it is recognized that commonly used

methods such as self-report and clinical inspection can at best only lead to the suggestion of probable sleep or awake bruxism. Instrumental approaches are required for definitive bruxism assessments.^{1,2} These matters need to be taken into account when interpreting results of related studies.

Advances have also been made in the diagnostic criteria of craniofacial pain conditions, particularly in the field of TMDs. Without any doubt, the introduction of clear and operationalized criteria for TMD pain has been a major achievement in the scientific community with the Research Diagnostic Criteria for TMD⁵, and later the Diagnostic Criteria for TMD.⁹ In contrast to the symptom of pain, other musculoskeletal symptoms that could be speculated to have possible link to bruxism activity, such as jaw muscle fatigue, tenderness, tension, or weakness, are much more rarely evaluated systematically. One reason for that could be that only symptoms and clinical findings of spontaneous craniofacial pain, pain on palpation, changes in jaw movement capacity, and temporomandibular joint sounds may give rise to a definite TMD diagnosis.^{5,9,10} Consequently, non-painful musculoskeletal symptoms are often not taken into account clinically, unless the patient decides to describe them as pain. Hence, the non-painful musculoskeletal complaints fall outside of the TMD umbrella term, and therefore do not qualify for any diagnosis.

Notwithstanding, non-painful symptoms, e.g., muscle fatigue, stiffness, soreness, weakness, etc. may also have a negative impact on the individual. Fields (1999) noted that, in addition to the sensory-discriminative component of noxious input and nociception and the related primary unpleasantness, there could also be a more contextually-dependent degree of unpleasantness – termed secondary unpleasantness.¹¹ In terms of muscle pain, this could mean that stiffness, fatigue, soreness, etc. could represent such non-painful symptoms

resembling secondary unpleasantness. Most of the previous systematic reviews on the relationship between bruxism and pain may have focused on the sensory-discriminative component of pain and primary unpleasantness, leading to the general conclusion that there is little, if any, evidence to support a strong relationship.³ Anecdotally, many clinicians will report that they have experienced an obvious relationship, but the controversy may rely on how pain and non-painful symptoms are reported and recorded. With this in mind, it seems appropriate to re-evaluate the relationship between bruxism and not only pain per se, but also to non-painful musculoskeletal symptoms.

The aim of the present systematic review was to answer the overall research question: “To what extent is bruxism associated with musculoskeletal signs and symptoms?”. To provide the best possible overview, results were reviewed in categories according to study design, study population (adults, children), bruxism sub-type (awake, sleep), assessment methods for bruxism and musculoskeletal symptoms (self-report, self-report and clinical examination, instrumental assessment), and type of outcome (pain, non-painful musculoskeletal symptoms).

2. Methods

The present systematic review was performed in accordance to the PRISMA guidelines.¹² A PubMed search of articles published until November 23, 2017 was conducted. The search string is shown in Table 1. One of the authors (LBH) screened the yielded titles according to the following inclusion criteria: 1. *Bruxism* was present in title and/ or 2. The title was seemingly related to the research question; 3. If any doubt as to criteria 1 and 2: the title was included for abstract review. The abstracts of the chosen titles were then screened according

to two criteria: 1. The article was seemingly related to the research question; 2. If any doubt or abstract was absent: the article as included for full text review. The chosen full text articles were assessed for eligibility by two of the authors independently (LBH and MT) according to the criteria in Table 2. In case of disagreement, authors FL and PS were consulted until consensus was reached. A review of the reference lists of the included articles was then performed, as well as a hand-search for possibly wrongfully omitted articles. Titles obtained were subjected to the same criteria as above. LBH and MT independently assessed the quality of the studies using the Newcastle Ottawa Scale (NOS) for Case Control Studies and Cohort Studies,¹³ the Quality Assessment Tool for Experimental Bruxism Studies (Qu-ATEPS),¹⁴ the Robins-I for assessment of risk of bias on non-randomized intervention studies,¹⁵ and the Cochrane Collaboration's tool for assessing risk of bias in randomized trials.¹⁶ For the NOS and Qu-ATEPS, a cut-of value of 2 stars and 30 points, respectively, was used for including a study for data extraction. If not distinction was made between awake and sleep bruxism, the study was excluded. Data extraction was performed by MT onto a data extraction form (Please see supplementary material. Relevant data from the full text articles included study characteristics (viz., first author, year, journal), participant characteristics (viz., gender, mean age and age range, type of sample), assessed signs and symptoms, methodology used for bruxism assessment, and main results. LBH was consulted in case of any doubts, and consensus was reached through discussion with occasional inclusion of coauthors FL and PS.

A meta-analysis could not be performed due to the heterogeneity of the study designs and outcome parameters assessed of the included studies.

3. Results

A flow chart of the article inclusion is provided in Fig. 1. In total, 51 studies were included in this review. There were 37 studies with a case-control or cross-sectional design, two studies with a prospective cohort design and 14 experimental studies. The studies were published between 1978 and 2017, with almost half (47%) of them having been published between 2008 and 2017 (Fig. 2). Altogether, 20,176 participants were included (8,564 female, 4,244 male, 7,368 no data on gender¹⁷⁻¹⁹) (Table 3). For each included study, a comprehensive description of the definitions of both bruxism cases, as well as outcome measures, i.e., painful and non-painful musculoskeletal signs and symptoms is provided in the supplementary materials.

3.1. Case-control, cross-sectional and prospective cohort studies

The 37 case-control studies were assessed and are presented based on four axes (Table 4): 1) Population age (adults, i.e., participants 18 years or older, and children and/or adolescents, i.e., participants <18 years); 2) Bruxism assessment method (self-report, self-report plus clinical examination, instrumental, i.e., ecological momentary assessment (EMA) and/or single channel electromyography (EMG) for awake bruxism, and single-channel EMG or polysomnography for sleep bruxism); 3) Circadian type of bruxism (awake bruxism, sleep bruxism); and 4) Type of outcome. Regarding the bruxism assessment method, some studies were fit for more than one category. For example, Raphael et al. (2012) provide results for both self-reported, as well as instrumentally assessed sleep bruxism,³¹ and is therefore included in two sections (Adults: Self report; Sleep bruxism, and Adults: Instrumental; Sleep

bruxism, polysomnography). An overview of studies that are included in multiple categories is provided in Table 4.

Variation in the reported outcomes was of a considerable size. As to provide a clear and structured overview of all outcomes, these were grouped into six categories. For acquisition of these categories, all reported outcomes were listed and subsequently grouped based on them referring to a similar theme, a method resembling thematic analysis in qualitative research.²⁰ The categories are mutually exclusive, i.e., each reported outcome could only fit into one category. The six categories were:

- a. Functional signs and symptoms: temporomandibular joint (TMJ) sounds (e.g., clicking, arthrosis /crepitation), disc-related (i.e., when a specific disc-related diagnosis was set by the authors, such as disc displacement), dysfunction (e.g., deviation, restriction, locking, luxation of the mandible)
- b. Muscle pain, or non-painful muscle symptoms (e.g., tenderness, soreness, fatigue, based on self-report and/or clinical examination)
- c. TMJ pain, or non-painful TMJ symptoms (e.g., tenderness, soreness, based on self-report and/or clinical examination)
- d. Function-related pain (e.g., pain only on movement or due to mastication)
- e. Orofacial pain involving musculoskeletal structures, not fit for categories above (e.g., combination of muscle and joint pain, combination of pain with dysfunction) and/or pain characteristics (e.g., intensity)
- f. Structured TMD diagnoses based on a diagnostic system other than RDC/TMD or DC/TMD.^{5,9}

In case the assessment of functional signs and symptoms (category a) and pain (categories b, c & e) is not based on validated clinical criteria, i.e., non-RDC/TMD or DC/TMD clinical

examinations or self-reported symptoms, they are listed using quotation marks. Non-painful muscle and TMJ symptoms (categories b & c) were assessed by unvalidated self-report measures, as standardized clinical criteria do not exist. Function-related pain (category d) was assessed by self-report. The description of outcomes in category e are not provided in detail in this manuscript, and the reader is referred to the original publications.

3.1.1. Adult populations

Adult populations were assessed in 25 studies (Table 4). To be assessed as an adult study, participants should be at least 18 years of age. Some studies reported results on mixed populations of children and adults, (e.g. ²¹). These studies were included in the adult's pool, as long as the reported mean age was ≥ 18 years.

3.1.1.1. Adults: *Self-report*

3.1.1.1.1. Awake bruxism

A self-reported, i.e., report obtained by interview or written questionnaire, awake bruxism diagnosis was obtained in nine studies.

Functional symptoms: TMJ sounds were investigated in two studies.^{22,23} Rossetti et al. ²² found no association between RDC/TMD assessed articular sounds and awake clenching, neither did Duckro et al. ²³ between “popping or clicking jaw sounds” and awake clenching or grinding. However, Michelotti et al. ²⁴ found awake clenching or grinding to be a significant risk factor for RDC/TMD diagnosed disc displacement. Daytime clenching was not found to be associated with mouth opening in myofascial pain patients by Rossetti et al. ²², while Moss et al. (1995) found a negative association between self-reported daily clenching of the teeth

and restricted mandibular movements, or “TMJ dysfunction”,²⁵ and Huhtela et al.²⁶ a positive one between awake bruxism and “TMJ locking”.

Muscle symptoms: The presence of RDC/TMD diagnosed myofascial pain was assessed in the studies of Rossetti et al. (2008) and Michelotti et al. (2010).^{22,24} An association was found with awake clenching, and awake clenching or grinding, respectively. However, the intensity of the pain was not found to be associated with awake clenching.²²

TMJ symptoms: RDC/TMD diagnosed arthralgia, arthritis, or arthrosis were assessed in the study of Michelotti et al. (2010) , which did not show an association with awake clenching or grinding.²⁴

Function-related pain: TMJ pain on jaw movement was found to be related with awake bruxism in the study of Huhtela et al.²⁶

Orofacial pain: Macfarlane et al. 2003¹⁸ assessed the presence of “orofacial pain”, i.e., pain in at least one of the following in the past month: jaw joint(s), area just in front of the ear(s), in or around the eyes, when opening the mouth wide, shooting pains in the face/cheeks, in the jaw joint when chewing, in and around the temples, tenderness of muscles at the side of the face, and prolonged burning sensation in the tongue/mouth, and an association was found with grinding the teeth during waking hours. However, when looking at each pain separately, the authors found an association only for pain in the TMJ.¹⁸ Huhtela et al.²⁶ found a positive association between reported awake bruxism and “TMD pain”, i.e. pain in the temples, TMJ, face, or jaw once a week or more often, while Leketas et al.²⁷ found significant associations between reports of frequent awake bruxism activity and a DC/TMD diagnosed TMD pain.

Non-RDC/TMD diagnoses: Two studies used diagnostic systems other than the RDC/TMD or DC/TMD to diagnose TMD complaints.^{28,29} Macfarlane et al. (2001) found an association

between grinding the teeth during waking hours and pain dysfunction syndrome.²⁸ Likewise, Schiffman et al. (1992) found an association between touching or holding the teeth together, clenching while awake, grinding while awake and/or holding the jaw rigid, and the “level of mandibular dysfunction”.²⁹

3.1.1.1.2. Sleep bruxism

Functional symptoms: RDC/TMD diagnosed locked joints and discopathy (with or without arthropathy) were not found to be associated with grinding or clenching the teeth during sleep in the study of Blanco Aguilera et al. (2014),³⁰ though Huhtela et al.²⁶ did find an association with “TMJ locking”. “TMJ sounds” were not associated with grinding during sleep in the study of Duckro et al.²³

Muscle symptoms: RDC/TMD diagnosed myofascial pain was found to be associated with self-reported tooth grinding during sleep in the study of Raphael et al.³¹ However, the self-report results were in contrast to findings based on PSG-measures.³¹ Please refer to section 3.1.1.3.3.

TMJ symptoms: RDC/TMD diagnosed arthropathy with or without discopathy were assessed by Blanco Aguilera et al.,³⁰ and no association was found with self-reported sleep bruxism.

Function-related pain: “TMJ pain on jaw movement” was found to be related with sleep bruxism in the study of Huhtela et al.²⁶

Orofacial pain: Bivariate analyses in the study of Blanco Aguilera et al.³⁰ showed an association between sleep bruxism and RDC/TMD diagnosed muscle pain, when muscle pain coexisted with discopathy and/or arthropathy, however this association disappeared in subsequent binary analyses. In the same study, pain intensity of RDC/TMD diagnosed pain³⁰ was found to be associated with sleep bruxism. Leketas et al.²⁷ did not find an association

with DC/TMD diagnosed TMD. Macfarlane et al. (2003) found “orofacial pain” (as described above) to be associated with nocturnal grinding of the teeth.¹⁸ In the study of Huhtela et al.²⁶, an association was found for self-reported TMD pain and sleep bruxism.

Non-RDC/TMD diagnoses: As with self-reported awake bruxism, an association was found between “pain dysfunction syndrome” and grinding the teeth during the night,²⁸ and clenching or grinding during sleep and the level of “mandibular dysfunction”.²⁹

3.1.1.2. Adults: Self-report plus clinical examination

3.1.1.2.1. Awake bruxism

There were no studies on this topic.

3.1.1.2.2. Sleep bruxism

Two studies assessed sleep bruxism diagnosed by a combination of self-report and clinical examination. Manfredini et al. (2010) diagnosed sleep bruxism when grinding sounds during sleep were present at least five nights a week during the past six months, according to the participant’s bed partner, and at least one of the following were present: tooth wear or shiny spots on restorations, report of morning masticatory muscle fatigue or pain, and masseteric hypertrophy upon digital palpation.³² Similarly, Fernandes et al. (2012) required patient report or awareness of sounds of grinding during sleep, confirmed by a roommate, and the clinical criteria as described for Manfredini et al. above for their assessment of sleep bruxism.

TMJ symptoms: RDC/TMD diagnosed TMJ pain was not associated with sleep bruxism in the Manfredini et al. (2010) study, unless combined with some occlusal factors (overbite ≥ 4 mm, “asymmetrical molar relationship”, and “laterotrusive interferences”).³²

Orofacial pain: RDC/TMD diagnosed TMD pain was associated with sleep bruxism in the study of Fernandes et al. (2012).³³

There were no studies on the possible association between self-report plus clinical examination diagnosed sleep bruxism and functional symptoms, muscle symptoms, function-related pain, and non-RDC/TMD diagnoses.

3.1.1.3. Adults: Instrumental

In the following three sections, the methods used in each study are briefly presented in a separate paragraph, prior to the report of outcomes.

3.1.1.3.1. Awake bruxism, ecological momentary assessment (EMA) and single channel EMG

There were two studies in which awake bruxism was assessed with the aid of the EMA method.^{34,35} EMA is a method used for tracking experiences and behaviours over time, as they happen in the natural environment of the participant.³⁶ Participants in the study of Glaros et al. (2005) carried pagers for one week and reported on intensity of tooth contact, and jaw, face, or neck tension during wakefulness.³⁴ Chen et al. (2007) used wrist-vibrators for a 10-day period to alert the participants of their study to report on non-functional tooth contact while awake.³⁵ On the other hand, Mude et al.³⁷ performed whole-day EMG recordings of the masseter muscle in a group of participants reporting a history of pain or headache in the area of the masticatory system and a group without such history. Tonic

episodes of muscle activity were measured for both sleep and awake states, and categorized based on duration, i.e., sustained or short, and intensity, i.e., percentages of maximum voluntary contraction (MVC). Alongside, sleep bruxism episodes were score according to established criteria.³⁸

Functional symptoms: Disc displacement was associated with awake bruxism in the form of proportion of time of tooth contact, intensity of tooth contact, and tension in jaw, face, or neck in the study of Glaros et al. (2005).³⁴

Muscle symptoms: RDC/TMD diagnosed myofascial pain was assessed in both of the abovementioned studies.^{34,35} Glaros et al.³⁴ found a positive association between myofascial pain and tension, and myofascial pain and “effort”, i.e., a composite variable, estimating the intensity of tooth contact in combination with the proportion of time in contact. Similarly, Chen et al.³⁵ found an association between myofascial pain and wake-time nonfunctional tooth-contact frequency.

Orofacial pain: The combination of RDC/TMD diagnosed myofascial pain and TMJ arthralgia was investigated by Glaros et al.,³⁴ and an association was found with “tension in jaw, face, and neck”, “proportion of time of tooth contact”, “intensity of tooth contact”, and “effort”. Mude et al.³⁷ found that participants reporting pain showed a significantly higher incidence of sustained tonic EMG episodes (i.e., episodes lasting > 13.65 s), than those without. In the same study, a significantly higher incidence and total duration of sustained, low-intensity tonic EMG episodes were found in the pain history group.

There were no studies investigating the association between an instrumental awake bruxism diagnosis and TMJ symptoms, function-related pain, and non-RDC/TMD diagnoses.

3.1.1.3.2. Sleep bruxism, single channel EMG

In five studies, sleep bruxism was diagnosed with the aid of single channel EMG recordings. The study of Mude et al.³⁷ is described above. In the study of Baba et al. (2005), the masseter EMG activity of participants without, or with minor TMD symptoms, was measured for 5 nights.³⁹ The authors used predefined criteria to score muscle activity, i.e., all periods with EMG activity above a 20% MVC level were considered as potential bruxism events, and calculated the total duration of EMG activity per hour of sleep, averaged across the 5-night study period.³⁹ Shedden Mora et al. (2012) used single-channel EMG devices for recording nocturnal masseter EMG activity during three nights.⁴⁰ The study sample consisted of participants with myofascial pain, pain-free sleep bruxism, and healthy controls. The authors used the Lavigne et al. (1996) criteria³⁸ and a 10 μ V threshold to analyze and score EMG activity, and calculated the mean number and duration of EMG bursts per hour of sleep, the mean number and duration of rhythmic nocturnal masseter muscle activity (NMMA) episodes per hour of sleep, and number of EMG bursts per episode as bruxism outcome measures.⁴⁰ Yachida et al. (2012) performed measurements of the EMG activity of the anterior temporalis muscle for at least 4 nights during a one-week period.⁴¹ EMG activity was analyzed using the Signal Recognition Algorithm described by Jadidi et al. (2008).⁴² An EMG event was scored when the EMG activity exceeded the signal level at rest (i.e., when the participant was asked to relax the jaw muscles), plus 20% of the maximum EMG level as acquired during a 60% MVC clench.⁴² The total number of EMG events, the number of EMG events per hour of sleep and the coefficient of variation (representing night-to-night variability in EMG activity) were used as bruxism outcome measures. Wei et al.⁴³ performed a pilot study with single-channel EMG recordings of the temporalis muscle from a group of women with both TMJ disc displacement and chronic myalgia/arthritis (based on DC/TMD criteria), and a group without those

conditions. The hypothesis that there were diagnostic group differences in frequency, duration and intensity of teeth clenching behavior was tested. The authors developed and validated an automated detection method for scoring sleep clenching behavior.⁴³

Functional symptoms: Clinically evaluated “TMJ sounds” and “active mouth opening” were assessed by Baba et al.,³⁹ and an association with sleep bruxism was found only for TMJ sounds.

Muscle symptoms: Shedden Mora et al. (2012) investigated the presence of RDC/TMD diagnosed myofascial pain,⁴⁰ and no association with sleep bruxism measures was found.

Orofacial pain: The combination of RDC/TMD diagnosed muscle and TMJ tenderness were investigated by Baba et al.,³⁹ and no association with sleep bruxism was found. Shedden Mora et al. reported on pain intensity of RDC/TMD diagnosed myofascial pain, jaw muscle tension in the morning after the EMG recording and TMD related symptoms (e.g., jaw pain, toothache, dizziness),⁴⁰ and an association was found only between TMD related symptoms and the number of EMG bursts per episode (but not for EMG bursts per hour, EMG burst duration per hour, NMMA episodes per hour, NMMA episode duration per hour). Craniofacial pain (i.e., presence of RDC/TMD painful TMD or tension type headache) was assessed in the study of Yachida et al.,⁴¹ and there, no association was found between EMG events per hour and pain, while a positive association was found between the coefficient of variation from multiple night EMG recordings and pain. In the study of Mude et al.,³⁷ significantly longer mean EMG clenching durations and a higher EMG clenching-associated temporalis muscle duty factor (i.e., the sum of EMG clenching episode durations divided by the total recording time) were found in the pain group, compared to the non-pain group. The authors found no differences for the number of EMG clenching episodes per night, EMG episodes per hour and mean clenching bite force between groups. Wei et al. found that clenching duration and

clench-associated temporalis muscle duty factor were significantly higher in the patient group than in the control group.⁴³ However, the authors did not find group differences for the number of clenching episodes per night and per hour, as well as for mean clenching bite force. There were no studies investigating the association between a single-channel EMG diagnosed SB, and TMJ symptoms, function-related pain, and non-RDC/TMD diagnoses.

3.1.1.3.3. Sleep bruxism, polysomnography

Sleep bruxism was diagnosed by laboratory-based polysomnography in eight studies. Rossetti et al. 2008²² performed PSG with audiovisual recordings (PSG-AV) during one night (an adaptation night was carried out, but excluded from analysis). The authors used the Lavigne et al. 1996 criteria³⁸ to score the following sleep bruxism parameters: number of EMG episodes per night, EMG episodes per hour, EMG bursts per hour, EMG bursts per episode, EMG episodes with grinding noise, percentages of EMG episodes in each sleep stage (stage 1-5), and sleep bruxism status, i.e., sleep bruxer and non-sleep bruxer, according to the Lavigne et al. 1996 cut-off values, i.e. ≥ 4 EMG episodes per hour or ≥ 25 EMG bursts per hour of sleep.³⁸ In a smaller pilot study, the same research group used similar outcome measures when analyzing the PSG data from one night of participants with painful TMD and controls.⁴⁴ Abe et al. (2013) performed PSG-AV during one night after an adaptation night was carried out, but excluded from analysis.⁴⁵ The authors used the Lavigne et al. 1996 criteria³⁸ to analyze EMG activity of the masseter and temporalis muscles, in a manner similar to Rossetti et al. 2008,²² with the exception of the percentages of EMG episodes in each sleep stage, which were not addressed. In their retrospective study, Lavigne et al. (1997) investigated sleep bruxism outcomes based on a one-night PSG-AV recording, performed after an adaptation night, of sleep bruxers.⁴⁶ The Lavigne et al. 1996 criteria³⁸ were used to quantify the number

of EMG episodes per hour, EMG bursts per episode, and root-mean-square EMG level per bruxism burst. Smith et al. (2009) performed two nights of PSG and auditory recordings and used the Lavigne et al. 1996 cut-off criteria ³⁸ to diagnose participants as sleep bruxers and non-sleep bruxers. ⁴⁷ A single night PSG-AV was performed in the study of Camparis et al. ²¹ In line with previously mentioned studies, the authors used the Lavigne et al. 1996 criteria ³⁸ to score the number of EMG bruxism episodes and EMG bursts per hour, EMG bursts per episode, total duration of EMG episodes, percentage of EMG episodes in each sleep stage, percentage of bruxism episodes with micro-arousals, and mean bruxism episode amplitude. ²¹ In a sample with RDC/TMD diagnosed myofascial pain and a pain-free control group, Raphael et al. (2012) performed one PSG-AV recording, after one adaptation night. As above, the authors used the Lavigne et al. criteria ³⁸ to score EMG episodes and EMG bursts per hour of sleep, the number of EMG episodes with grinding sounds and the total EMG episode duration. ³¹ The authors assessed the sleep bruxism status, based on the Lavigne et al. 1996 cut-off criteria. ^{31,38} Finally, the study of Muzalev et al. ⁴⁸ was based on the same sample as in Raphael et al. 2012, ³¹ but aimed to investigate whether cases and controls differed in terms of time intervals between sleep bruxism episodes.

Functional symptoms: Rossetti et al. (2008) investigated the presence of TMJ sounds, and restriction of mandibular movement, and no association was found with sleep bruxism measures. ²²

Muscle symptoms: “Transient morning masticatory muscle pain” was assessed in the study of Abe et al. (2013), ⁴⁵ and although the presence of pain in the total sample was low, it was found to be associated with the status of being a sleep bruxer. Within the sleep bruxism group, there were no statistically significant differences between sleep bruxers with and those without pain. ⁴⁵ “Non-myofascial muscle pain”, i.e., presence of pain that did not fulfil

the RDC/TMD criteria for chronic myofascial pain,⁴⁶ and masseter pressure pain sensitivity measured with an algometer⁴⁷ were assessed in the studies of Lavigne et al. (1997) and Smith et al. (2009), respectively, both finding no association with the presence of PSG assessed sleep bruxism. RDC/TMD diagnosed myofascial pain was assessed in two studies.^{22,31} Rossetti et al. found a significant association between a PSG-based sleep bruxism status and myofascial pain,²² however, the study also showed that there was no difference between myofascial pain patients and controls in the percentages of phasic, tonic, and mixed sleep bruxism episodes. Within the myofascial pain group, sleep bruxism was not a significant predictor of TMD intensity index, pain duration, number of tender muscle sites to palpation, sensitivity of tender muscle sites to palpation, and period of worst pain within the day.²² Within the sleep bruxer group, myofascial pain was negatively associated with tonic EMG episodes, i.e., sleep bruxers without pain had more tonic episodes than sleep bruxers with pain.²² Raphael et al.³¹ showed that no sleep bruxism measure was significantly higher in RDC/TMD diagnosed myofascial pain cases than controls, except that controls were more likely to present at least two EMG episodes with grinding per recording than cases. Within the myofascial pain group, pain duration was similar for sleep bruxers and non-sleep bruxers, while characteristic pain intensity was lower for sleep bruxers compared to non-sleep bruxers.³¹ Similarly, in the same group, participants with two or more EMG episodes with grinding noise had significantly lower levels of pain interference with daily activity than those without.³¹ Muzalev et al.⁴⁸ found a similar duration of inter-episode intervals in both myofascial pain patients and controls.

TMJ symptoms: TMJ pain on RDC/TMD palpation was assessed by Rossetti et al. (2008),⁴⁴ who did not find an association with PSG assessed sleep bruxism.

Function-related pain: No studies were available.

Orofacial pain: The combination of muscular and TMJ pain was assessed in two studies.^{21,44} Camparis et al. (2006) found no significant difference in sleep bruxism variables between sleep bruxers with and without painful RDC/TMD diagnosed TMD (muscle pain with or without TMJ pain). However, participants without pain presented 20% more sleep bruxism-related EMG episodes per hour of sleep than those with pain.²¹ Rosseti et al. (2008) studied mixed muscle and/or TMJ types of RDC/TMD assessed TMD pain, and found no association with PSG diagnosed sleep bruxism for neither pain intensity at rest, nor pain on palpation of masticatory muscles and/or TMJs.⁴⁴

There were no PSG-based studies investigating the association between sleep bruxism and function-related pain or non-RDC/TMD diagnoses.

3.1.2. Children and adolescents

Children and adolescents cohorts were evaluated in 10 studies (Table 4).

3.1.2.1. Children and adolescents: Self-report

3.1.2.1.1. Awake bruxism

Functional symptoms: “TMJ sounds” were assessed in the studies of Carra et al. (2011), van Selms et al. (2013), Egermark et al. (2001), Şermet Elbay et al. (2017), and Winocur et al. (2001).^{17,19,49-51} All of these studies found an association between self-reported awake bruxism and “TMJ sounds”, with the exception of Şermet Elbay et al.,⁵¹ who found the association for children living with their parents, but not for children living in child protection institutions. Chun and Koskinen-Moffett (1990) and Şermet Elbay et al. (2017) investigated clinically diagnosed “TMJ sounds”, and found those not to be related with self-reported awake bruxism.^{51,52} RDC/TMD diagnosed anterior disc displacement with reduction was assessed by Kalaykova et al. (2011),⁵³ who did not find an association with awake bruxism.

Self-reported “TMJ locking” and RDC/TMD assessed anterior disc displacement with intermittent locking were found to be associated with self-reported awake bruxism in the studies of Carra et al. (2011) and Kalaykova et al. (2011) respectively,^{17,53} but Winocur et al. (2001)⁵⁰ found no relation between self-reported “locking, catching, or open lock” and self-reported awake bruxism, neither did Şermet Elbay et al.⁵¹ for self-reported “TMJ sticking”. “Range of mouth opening” was not associated with awake bruxism in the studies of Carra et al. (2011) and Winocur et al. (2001),^{17,50} but, on the other hand, Egermark et al. (2001) found an association with “difficulties in mouth opening”.⁴⁹ An association between self-reported awake bruxism and “limited lateral mandibular movements” was shown by Carra et al.¹⁷

Muscle symptoms: “Jaw muscle fatigue upon awakening” was assessed by Carra et al.,¹⁷ who found an association with self-reported awake bruxism. Similarly, Şermet Elbay et al.⁵¹ found an association between awake bruxism and reporting “pain or tiredness in the masticatory muscles”, but only for children living in child protection institutions and not for those living with their parents. In the same study, palpation-induced masticatory “muscle tenderness” was associated with reporting awake bruxism in both groups of children.

TMJ symptoms: Şermet Elbay et al.⁵¹ found an association between reporting awake bruxism and palpation-induced “TMJ tenderness” in a group of children living in child protection institutions, but not for those living with their parents.

Function-related pain: “Tiredness in the jaw while chewing” and “pain in the jaw near the ear while chewing” were examined by Winocur et al.,⁵⁰ and no association was found.

Orofacial pain: RDC/TMD diagnosed TMD pain (myofascial pain with or without limited mouth opening and/or TMJ arthralgia and/or TMJ osteoarthritis) was assessed by Fernandes et al.,⁵⁴ who found an association with awake bruxism. “Orofacial pain”¹⁹ and “pain in jaw or face at rest”⁵⁰ were assessed in the studies of van Selms et al. (2013) and Winocur et al. (2001),

and an association with awake bruxism was found in the former, but not in the latter study.^{19,50} “Jaw fatigue” was assessed in the study of Egermark et al.,⁴⁹ and an association was found with self-reported awake bruxism.

There were no studies on the association between self-reported awake bruxism and TMJ symptoms or non-RDC/TMD diagnoses.

3.1.2.1.2. Sleep bruxism

Functional symptoms: “TMJ sounds” were assessed in three of the previously mentioned studies. Egermark et al. (2001) found an association with self-reported sleep bruxism,⁴⁹ as did van Selms et al. (2013) for “TMJ clicking”, but not for “scraping sounds”,¹⁹ while Carra et al. (2011) did not find any of such associations.¹⁷ The same study did also not find any associations between sleep bruxism and “TMJ locking”, “restricted lateral mandibular movements” and “maximal mouth opening”.¹⁷ On the other hand, Egermark et al.⁴⁹ did find an association with “difficulties in mouth opening”.

Muscle symptoms: Jaw muscle fatigue was assessed by Carra et al.,¹⁷ who found an association with self-reported sleep bruxism.

Orofacial pain: RDC/TMD diagnosed TMD pain (myofascial pain with or without limited mouth opening and/or TMJ arthralgia and/or TMJ osteoarthritis) were assessed by Fernandes et al.,⁵⁴ and an association was found. Van Selms et al. found an association between sleep bruxism and “jaw pain” or “tense feeling in the morning”.¹⁹ Similarly, Egermark et al. (2001) found an association for “jaw fatigue”.⁴⁹

There were no studies investigating the associations between self-reported sleep bruxism and TMJ symptoms, function-related pain, and non-RDC/TMD diagnoses.

3.1.2.2. Children and adolescents: Self-report plus clinical examination

3.1.2.2.1. Awake bruxism

No studies on this topic were found.

3.1.2.2.2. Sleep bruxism

Bruxism diagnosed based on self-report plus clinical examination was assessed in three studies.

Functional symptoms: Emodi-Perlman et al. (2012) and Restrepo et al. (2008) RDC/TMD assessed TMJ sounds in their studies, with the former showing no association,⁵⁵ and the latter finding one.⁵⁶ RDC/TMD diagnosed disc displacement was assessed by Kalaykova et al.,⁵³ who did not find an association with sleep bruxism. Three studies investigated mandibular dysfunction. Emodi-Perlman et al. (2012) did not find an association between sleep bruxism and “TMJ sticking”,⁵⁵ nor did Kalaykova et al. (2011) for RDC/TMD based anterior disc displacement with intermittent locking.⁵³ On the other hand, Restrepo et al. (2008) found an association with (limitation of) the range of mouth opening and with “deviation in mouth opening”.⁵⁶

Muscle symptoms: Reported “pain or tiredness of masticatory muscles” was assessed in the study by Emodi-Perlman et al.,⁵⁵ and no association with sleep bruxism was found. RDC/TMD-based pain or tenderness of masticatory muscles on palpation were assessed in two studies. Emodi-Perlman et al. (2012) found no association with pain on extraoral palpation,⁵⁵ while Restrepo et al. (2008) found no association with intraoral palpation, and a negative association for extraoral palpation.⁵⁶

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TMJ symptoms: Sensitivity of the TMJ on RDC/TMD based palpation was assessed by Emodi-Perlman et al.,⁵⁵ and no association was found.

Function-related pain: Pain when opening or chewing was assessed in the study of Restrepo et al.,⁵⁶ and an association was found with sleep bruxism.

Orofacial pain: The presence of “pain in the temples, face, TMJs, or jaws” was also assessed in the study of Restrepo et al.,⁵⁶ and no association with sleep bruxism was found.

There were no studies using non-RDC/TMD diagnoses.

3.1.2.3. Children and adolescents: Instrumental

3.1.2.3.1. Awake bruxism, EMA and single-channel EMG

There were no studies assessing awake bruxism in children or adolescent populations using EMA methods or single-channel EMG.

3.1.2.3.2. Sleep bruxism, single channel EMG

In one study, sleep bruxism was diagnosed by single-channel EMG. Nagamatsu-Sakaguchi et al. (2008) performed a single night recording of the EMG activity of the masseter muscle in a group of adolescents (mean age 15.4 ± 0.5). The EMG device in this study automatically detected and counted EMG events that exceeded 30% of MVC. Participants were divided into two groups, based on the count of EMG events: a “severe” sleep bruxism group (≥ 125 EMG events) and a “non-severe” sleep bruxism group (< 125 EMG events).⁵⁷

Functional symptoms: RDC/TMD assessed TMJ clicking was found to be associated with a “severe” sleep bruxism diagnosis, while limited mouth opening was not associated with “severe” sleep bruxism in females, while the variable was not observed in males.⁵⁷

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Muscle symptoms: RDC/TMD assessed sensitivity to palpation of masticatory and cervical muscles (digastric, masseter, temporalis, SLM, trapezius) were not associated with “severe” sleep bruxism, except for the digastric muscle in males.⁵⁷

TMJ symptoms: RDC/TMD assessed sensitivity to palpation of the TMJ was, as well, not associated with “severe” sleep bruxism.⁵⁷

There were no studies using single channel EMG for the detection of sleep bruxism that investigated its association with function-related pain, orofacial pain, and non-RDC/TMD diagnoses.

3.1.2.3.3. Sleep bruxism, PSG

There were no studies assessing sleep bruxism in children or adolescent populations using PSG.

3.1.3. Summary of findings from case-control, cross-sectional and prospective cohort studies

In summary, the included studies indicate that in adults, awake bruxism may be positively associated with pain located in the masticatory muscles, and in a more broad sense, with myofascial TMD pain. This association is supported both by studies using a self-report as well as an instrumental diagnosis of awake bruxism. There were too few studies to support a solid conclusion on the relation between awake bruxism and TMJ pain. Studies in children suggest a positive association between musculoskeletal pain and awake bruxism, but since all these studies utilized self-report only diagnoses for both bruxism, as well as musculoskeletal outcomes, these results should be taken with caution.

Studies using EMG or PSG to validate bruxism status are fairly consistent in showing that sleep bruxism status, i.e., receiving a positive assessment of sleep bruxism according to the Lavigne et al. criteria,³⁸ is not positively associated with musculoskeletal pain in adults. Evidence from the reviewed studies suggests that sleep bruxism variables, such as type and length of bruxism episodes, and night-to-night variability in sleep bruxism activity, may be associated with pain symptoms, but did not allow for a solid conclusion regarding the direction of this association. On the other hand, studies using a case definition based on self-reported bruxism, with or without clinical examination, i.e. a less valid case definition, seem to support a positive association with TMD pain diagnoses. However, some studies did not detect this association, especially those focusing on TMJ pain. EMG-based evidence in studies of children was very scarce, and pointed to sleep bruxism not being associated with pain symptoms. Similar results were reported by studies utilizing self-report plus clinical examination. On the contrary, and similarly to adult populations, studies using only self-reported sleep bruxism did show associations with both pain and fatigue symptoms. However, when sleep bruxism was additionally assessed by clinical examination, such associations were no longer found.

There were too few studies assessing non-painful symptoms for allowing solid conclusions to be drawn as for whether these are related to awake and/or sleep bruxism, in both adult and children populations.

As for functional symptoms, results on self-reported TMJ sounds were consistently found to be associated with self-reported awake bruxism in populations of children. However, when clinical examination was used for the detection of TMJ sounds this association was no longer found. For all other types of functional symptoms, as for other awake and sleep bruxism assessment methods, either results of studies were contradicting, or sparse. Consequently,

based on the reviewed literature above, it can be concluded that the association between bruxism and functional symptoms remains unclear.

3.2. Experimental studies

Fourteen experimental studies were included in this review.⁵⁸⁻⁷¹ The experimental conditions involved tooth-clenching tasks (9 studies),^{58,59,61-63,66,69-71} tooth-grinding tasks (2 studies),^{64,65} eccentric contraction of jaw muscles (2 studies),^{58,63} and experimental EMG feedback studies (3 studies).^{60,67,68} The studies evaluated the following range of musculoskeletal symptoms: pain intensity^{58-65,67-71} and pain distribution,^{64,65} soreness/tenderness,^{61,64,65} unpleasantness,^{61,64,65,71} fatigue/ physical tiredness/ exhaustion,^{58,59,62,63,69-71} sensitization to pressure,^{58,59,61-65,69,70} maximum voluntary bite force,⁶¹⁻⁶⁵ muscle endurance time,^{66,70} clenching-related pain and headache pain since the last session,⁶⁷ and maximum pain-free mouth opening.^{58,63}

Overall, the results from the experimental studies above indicate that increasing the masticatory muscle activity experimentally by either tooth-clenching, tooth-grinding, or consciously increasing EMG activity may lead to short-lasting pain and fatigue in healthy individuals.⁵⁸⁻⁷¹ However, none of the tasks of the included studies led to long-lasting pain. Rather, even repeating the task for 5 days resulted in fewer symptoms on day 5 compared with day 1, indicating an adaptation or training effect in the healthy individuals.⁶¹ Using eccentric contractions, though, it was possible to induce delayed onset muscle soreness (DOMS) in the jaw muscles that was resolved after one week.^{58,63}

4. Discussion

The aim of the present systematic review was to answer the overall research question: “To what extent is bruxism associated with musculoskeletal signs and symptoms? It was decided

to evaluate all types of musculoskeletal signs and symptoms, including non-painful symptoms, as they may lead patients to seek evaluation and may possibly precede development of pain. Hence, the aim was not to perform yet another review on the relationship between bruxism and pain. Due to the interest in non-painful symptoms, it was decided not to exclude studies using case definitions based on self-reports of bruxism and musculoskeletal symptoms, but rather to include such studies and report their results in separate paragraphs from studies involving validated diagnostic techniques and assessment methods. Of course, results based on self-reports alone should be interpreted with great caution and it is recommended that future studies always include excellent methodology and valid measures of bruxism and musculoskeletal signs and symptoms.

4.1. Non-painful musculoskeletal symptoms

A few case-control, cross-sectional, or prospective cohort studies included evaluations of musculoskeletal symptoms other than pain, yet none of them as the primary outcome parameter.^{17,19,39,49} The assessed and reported non-painful symptoms were muscle tenderness³⁹ and muscle fatigue.^{17,49,55} However, conclusions are conflicting and vary between studies, although most of them are in favor of an association between bruxism and the studied symptoms. Since the non-painful symptoms were generally not the primary outcome parameter, there is a possibility of publication bias, as it could be speculated that authors might feel more inclined to report significant associations, whereas lack of association between bruxism and a secondary outcome parameter might be left out. Importantly, the risk of Type I error should be taken into account when assessing studies with multiple outcome parameters. Also, the lack of validated assessment methods and potential recall-bias for non-painful musculoskeletal symptoms are important limitations and results

should be interpreted with caution. However, in the experimental studies, non-painful symptoms were much more often taken into account and systematically quantified and reported. Fatigue and sensitization to pressure stimuli were the non-pain symptoms, which were most often reported after a tooth-clenching or grinding task.^{58,59,61-64,70,71}

Assessment of non-painful symptoms could be considered important, since they may be the reason to seek treatment and, recently, a large prospective cohort study showed that reports of three or more non-specific orofacial symptoms, defined as jaw stiffness, cramping, fatigue, pressure, soreness, and ache, can predict subsequent onset of TMD pain.⁷² Hence, a more systematic approach to the evaluation of non-pain musculoskeletal symptoms seems warranted for both clinical and research purposes.

The concept of primary and secondary unpleasantness vs. algosity, suggested by Fields in 1999,¹¹ could also be further explored. Fields suggested that algosity could be seen as the unique sensory-discriminative aspect of pain that separates it from other unpleasant sensations, such as itch or dysesthesia.¹¹ Primary unpleasantness is an aspect of unpleasantness, which is strictly stimulus-bound and therefore should be considered in the context of a sensory-discriminative feature.¹¹ In contrast, secondary unpleasantness is linked to higher-order cognitive processing and largely determined by memories and contextual features.¹¹ It could be speculated that a person experiencing jaw symptoms may be more prone to seek treatment, when secondary unpleasantness dominates the picture, i.e., when the context in which the symptoms occur is negative or when the person has memories of similar symptoms getting worse in the past. In a qualitative study by Rollman et al.,⁷³ it was shown that care seeking for TMD-pain complaints was associated with catastrophizing, i.e., interpreting the pain as alarming and too long lasting. It may be useful to further investigate

the concept of secondary unpleasantness, its possible similarity as a construct with catastrophizing, and the relation with the development of orofacial symptoms and care-seeking behavior. It could be further speculated that some psychosocial factors, such as depression, somatization, and anxiety that are well-known risk factors for chronic TMD pain,⁷⁴ may be closely linked to the experience of secondary unpleasantness. Such psychosocial factors and their relation to non-painful symptoms have been investigated in other chronic pain disorders, such as irritable bowel symptoms.⁷⁵ Taking this information into consideration, it may be prudent to systematically assess non-painful musculoskeletal symptoms and their relationship to awake and sleep bruxism, while taking psychosocial factors into account. Moreover, future longitudinal studies should examine, whether an early intervention in patients with non-painful musculoskeletal symptoms may prevent the development of acute painful TMD, or transition of an acute painful state into a chronic one.

4.2. Pain

Pain as a symptom has much more consistently been evaluated as a possible consequence of awake or sleep bruxism. Overall, studies based solely on self-report of awake or sleep bruxism and pain are more indicative of a positive association than studies supporting bruxism assessment and pain diagnosis with validated clinical examinations and instrumental evaluations of muscle activity. Hence, increasing the validity of the assessment methods seems to weaken the proposed associations, except when it comes to awake bruxism, where studies using EMA demonstrated that TMD patients have more tooth contact than healthy controls.³⁴ As mentioned previously, the validity of self-reported sleep bruxism has been questioned, and in a study using PSG as the gold standard, it was concluded that studies based on self-reports of sleep bruxism should, indeed, be interpreted with caution.⁷⁶ Importantly,

also the use of PSG as a gold standard for sleep bruxism assessment is under current discussion, ² because only frequency measures of EMG episodes or bursts are being considered and not taking into account the amplitude or intensity of such EMG activities. There is a need to reconsider the often-used gold standards of sleep bruxism assessment, with more refined measures of EMG activity over time being suggested valuable for examination of work-related musculoskeletal symptoms. ²

4.3. Functional symptoms

Bruxism is commonly suggested to be a source of microtrauma of the TMJs and masticatory muscles, which, in turn, could lead to functional symptoms of the masticatory system. ⁷⁷ However, the present review could not draw a robust conclusion on this proposed association. A variety of symptoms were evaluated in the reviewed studies, including TMJ clicking and crepitation sounds, disc-related disorders, and deviations in the movement of the mandible in adults ^{22-26,30,34,37,39} and children. ^{17,19,49-53,55,56,57} Although some studies used the RDC/TMD criteria for establishing their diagnoses (e.g., ^{35,53}), there generally was no uniform way of assessing these outcomes across studies, with some solely using self-report measures (e.g., ^{19,23,26}), and others including clinical examination in the assessment of participants (e.g., ^{25,39,52}). This made comparison of the studies difficult. Most importantly though, studies using more valid device-assisted methods for assessing sleep and awake bruxism were extremely sparse ^{22,34,39,57} and did not allow for the drawing of solid conclusions on the topic. Future studies should take advantage of available technologies in order to significantly contribute to the understanding of if and how sleep and awake bruxism are involved in the pathophysiology of functional symptoms of the masticatory system.

4.4. Additional suggestions for future studies

Masticatory muscle activity in terms of bruxism is most likely not enough to cause the onset or persistence of musculoskeletal pain and non-pain symptoms in the absence of other risk factors.^{7,8} Therefore, it is preferable that future high-quality studies assess the multiple interactions between risk factors, such as, but not restricted to, psychological, lifestyle, sleep, and trauma/overloading variables, instead of solely focusing on the assessment of linear relations.^{8,78} Prospective cohort studies using device-assisted bruxism diagnostic methods may prove very valuable in this context. It is of course essential to carefully design and plan such studies by for example, cautiously selecting primary and secondary outcome parameters according to the study aim and plan and perform the statistical analyses using the highest standards to avoid “fishing expeditions”. Also, more focus on effect sizes in addition to statistical significance is advocated.

Bruxism is considered to have two circadian manifestations, namely occurring during sleep and/or awake states.² There is still no evidence to prove that these two manifestations are two distinct conditions, or that they should be considered part of the same entity.² Until this topic is clarified, it is strongly advised that in future studies sleep and awake bruxism are assessed as separate variables. This may additionally contribute to the comparability of the outcomes of different studies. Furthermore, the distinction might make research results more directly applicable to clinical practice, were sleep and awake bruxism are often managed in a different manner.⁷⁹

Furthermore, investigating different expressions of muscle activity may lead to new insights. Alongside with EMG frequency measures during sleep, such as number of episodes, bursts etc., it may be of value to investigate variables like background EMG activity, intensity, and amplitude of activity, variability of activity over time or the timing of activity during sleep,^{31,48}

and their relation to musculoskeletal symptoms. A similar approach can be suggested for awake bruxism studies.^{34,35,37}

Regarding awake bruxism, it is suggested that more instrumental studies are conducted, especially including children and adolescent populations. It is imaginable that the implementation of instrumental awake bruxism studies in these populations is difficult, due to the possible lack of cooperation of young individuals. It may prove useful to design other diagnostic methods for these populations, for example, through direct or indirect (i.e., via video recordings or structured parental-reporting) observation of children in everyday situations, such as attending school, making homework etc.

Future experimental bruxism studies are encouraged to include not only healthy, symptom-free participants, but also individuals with preexisting musculoskeletal symptoms, since it might be speculated that their masticatory system reacts to mechanical loading in a different manner. Similarly to what is mentioned above, it is suggested that experimental studies also take into account other risk factors, such as somatization, depression, fear of movement, beliefs about the effect of bruxism on muscles, etc., as these factors may alter the way that symptoms are experienced and the results of diagnostic test.⁸⁰

As for functional musculoskeletal symptoms, the main suggestion of this review is that future studies use standardized and validated criteria for their assessment, such as instruments included in the DC/TMD.⁹

Importantly, the authors of this review suggest that in future studies non-painful musculoskeletal symptoms are assessed systematically and separately from pain symptoms. For this purpose, standardized criteria need to be developed. This will facilitate research on the importance of such symptoms as risk factors for 'more severe' symptoms, their interplay

with other risk factors in orofacial pain processes, and their role in treatment-seeking behavior.

5. Conclusions

From the present review, it may be concluded that bruxism to some extent is associated with musculoskeletal symptoms. However, the evidence is conflicting and seems to be dependent on many factors such as for example age, whether the bruxism occurs during sleep or wakefulness, as well as diagnostic methodology regarding bruxism and musculoskeletal signs and symptoms. The literature does not sufficiently support a direct linear causal relationship between bruxism and such signs and symptoms, but points more in the direction of a multifaceted relationship dependent on the presence of other risk factors. Pain is by far the most commonly assessed symptom, whereas non-painful musculoskeletal symptoms have generally not been systematically evaluated. In light of recent findings indicating that non-painful symptoms may precede TMD pain, it is suggested to increase the scientific focus on non-painful musculoskeletal symptoms in future studies. Finally, it is strongly recommended that future studies do not rely solely on self-reports for assessment of bruxism status.

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Table 1: Search string
<p>("Bruxism"[Mesh] OR bruxer[text word] OR bruxers[text word] OR bruxing[text word] OR bruxist[text word] OR bruxists[text word] OR bruxofacets[text word] OR bruxomania[text word] OR clenching[text word] OR parafunction[text word])</p> <p>AND</p> <p>("Craniomandibular Disorders"[Mesh] OR "Pain"[Mesh] OR "Muscle Fatigue"[Mesh] OR "Muscle Weakness"[Mesh] OR "muscle hypertonia"[MeSH] OR temporomandibular disorders[text word] OR unpleasantness[text word] OR discomfort[text word] OR tenderness[text word] OR sensitivity[text word] OR hypersensitivity[text word] OR soreness[text word])</p>

Table 2: Inclusion criteria for full text articles

- A. The study is related to the research question
- B. Contains original data and no double data
- C. Study meets minimum quality criteria; see below
- D. Reviews included for reading only and reference list checking
- E. Bruxism is well-defined (e.g., ICHD, Glossary of Prosthodontics, Guidelines AAOP, Lobbezoo et al.)
- F. Well-described bruxism assessment method (e.g., questionnaire, clinical examination, EMG, PSG)
- G. Outcome parameters are well described and quantified (e.g., dichotomized, NRS, VAS)
- H. Includes a control condition or control group
- I. Full text in English is available

Table 3. Demographic characteristics and sample types of included studies.

#	First author, year, journal initials	n	Male	Female	Mean age (range)	Sample type
Case-Control, Cross-sectional						
1	Abe, 2013, JOP	81	34	47	25.8 (18-44)	Participants without TMD
2	Baba, 2005, JOP	93	47	46	24.7 M, 23.7 F (22-32)	Subjects selected at a Medical and Dental University
3	Blanco Aguilera, 2014, JOR	1220	190	1030	? (18-61 or more)	Subjects referred to specialized Orofacial Pain clinic
4	Camparis, 2006, AOB	40	8	32	36.1 (17-54)	Bruxing patients
5	Carra, 2011, EJOS	604	?	?	13 (7-17)	Patients of Orthodontic clinic
6	Chen, 2007, JOP	24	8	16	? (18-67)	Patients referred to Orofacial pain clinic and controls selected among friends and staff
7	Chun, 1990, JCD	96	42	54	12.6 (9-18)	Adolescents attending an Orthodontic and a Pedodontic University department
8	Duckro, 1990, Cranio	500	251	249	? (21-65 or more)	Residents of specific geographic area
9	Emodi-Perlman, 2012, JOR	244	61	183	? (5-12)	Pupils of selected school
10	Fernandes, 2012, JOR	272	34	238	? (21-65 or more)	Residents of specific geographic area
11	Fernandes, 2015, JOR	1307	712	595	? (12-14)	Adolescents attending schools in specific geographic area
12	Glaros, 2005, Cranio	96	26	70	Not provided for total sample, but for groups: [1] 35.4, [2] 39.5, [3] 44.9, [4] 39.2 (?/?)	General population and patients attending a Facial Pain Centre
13	Huhtela, 2016, JOPH	4403	1628	2775	Applied science: 24.4 (male), 23.7 (female), Academic: 25.2 (male), 24.7 (female) (19-35)	Academic and applied science university students

14	Kalaykova, 2011, JOP	260	124	136	14 (12-16)	Adolescents attending a Dental University student's clinic
15	Lavigne, 1997, EJOS	13	8	5	B with pain group: 25.3, B without pain group: 27.3 (?/?)	Sleep bruxers
16	Leketas, 2017, JCS	520	112	408	21.8 (18-64)	Patients visiting a University Clinic of Maxillofacial Surgery, persons recruited by announcements or from acquaintances
17	Macfarlane, 2001, OD	327	56	271	Median, cases: 37, controls: 39	Patients referred to a Temporomandibular Disorder Clinic and patients attending general dental practitioners
18	Macfarlane, 2003, JD	2504	?	?	?(18-65)	Population attending a General Medical Practice
19	Manfredini, 2010, Cranio	276	83	193	32.2 (25-44)	Patients referred to a TMD clinic
20	Michelotti, 2010, JOR	668	182	486	Cases: 33.2, controls: 37.6 (8-79)	Patients referred to the Dental Clinic of a University and their accompanying persons
21	Moss, 1995, JOR	43	3	40	Cases: 42.8, controls: 47.7 (?/?)	General population at University medical centre and community
22	Mude, 2017, JPR	33	11	22	23.9 (?-?)	Students
23	Muzalev, 2017, Sleep	123	0	123	Cases: 37.8, controls: 34.7 (?-?)	Patients seeking treatment at a College of Dentistry
24	Nagamatsu-Sakaguchi, 2007, IJP	127	49	78	15.4 (?/?)	High school subjects of specific geographic area
25	Raphael, 2012, JADA	172	0	172	39.2 (19-78)	Patients attending a University College of Dentistry and their acquaintances
26	Restrepo, 2008, JOR	52	?	?	9.45 (8-11)	Students of school in specific geographical area
27	Rosseti, 2008, Cranio	26	12	14	Cases: 27.14, controls: 27.42 (17-40)	Unspecified population
28	Rosseti, 2008, JOP	60	48	12	Cases: 26.6, controls: 26.0 (19-42)	Unspecified population
29	Schiffman, 1992, JOR	250	0	250	Sophomore: 22, junior: 23, senior: 25 (?/?)	Sophomore, junior and senior nursing students

30	Şermet Elbay, 2017, IDJ	385	191	194	CLCPI group: 13.4 (?/?), CLWP group: 13.5 (?/?)	Children attending public schools and child-protection institutions
31	Shedden Mora, 2012, JPR	106	17	89	TMD cases: 27.4, pain free B cases: 25.7, controls: 24.3 (?/?)	Patients of a Department of Prosthetic Dentistry, an TMD outpatient clinic, university undergraduates and local community
32	Smith, 2009, Sleep	53	10	43	33.6	Patients of a Orofacial Pain Clinic and community
33	van Selms, 2013, CDOE	4208	?	?	14.5 (12-18)	Students attending secondary schools
34	Wei, 2017, JOR	24	0	24	Pain group: 37.4 (21-62), non-pain group: 31.1 (24-56)	Subjects recruited at a Dental University
35	Winocur, 2001, JOR	323	0	323	?(15-16)	Girls attending a high school
36	Yachida, 2012, JDR	115	39	76	Healthy men 25.3, healthy women 27.2, SB men 46.4, SB women 36.0, TTH men 41.1, TTH women 31.0 (?/?)	Students and staff of University, subjects responding to advertisements, patients attending a Headache Centre
Prospective Cohort						
1	Egermark, 2001, AOS	320	153	167	7, 11, 15 (-/-)	Random community sample
Experimental						
1	Arima, 1999, JOP	12	12	0	26.0 (21-42)	Healthy human subjects without history of TMD or bruxism
2	Arima, 2000, JOP	10	10	0	24 (20-30)	Healthy human subjects without history of TMD or bruxism
3	Christensen, 1982, TJPD	6	6	0	35 (?)	Dentist without clinical discerbable diseases and dysfunctions of the manibular locomotor system
4	Dawson, 2013, Pain	30	14	16	Women: mean age 36; Men: mean age 41 (?)	Healthy human subjects
5	Farella, 2010, JDR	10	0	10	23.0 (?)	healthy dental students without report of bruxism, orofacial pain or TMJ clicking in the preceding month

6	Glaros, 2004, JBM	14	8	6	? (21-35)	Healthy human subjects without history of TMD, chronic pain or headaches
7	Glaros, 1998, JOP	5	2	3	? (23-29)	Dental students without TMD or orofacial pain incl. headache
8	Glaros, 2000, Cranio	20	0	20	26.5 (20-51)	Healthy human subjects without history of TMD, chronic pain or headaches
9	Koutris, 2013, CJP	40	20	20	27.7 (?)	healthy subjects without dental pain pathology or self-reported oral parafunctions
10	Takeuchi, 2015, Headache	18	9	9	Women: mean age 22.2; Men: mean age 25.8 (?)	Healthy university grad students without history of TMD
11	Turker, 2010, AOB	6	6	0	35.7 (?)	Healthy subjects without history of TMD
12	Svensson 1996, JOP	10	0	10	24.2 (?)	Healthy women without history of TMD
13	Svensson 2001, AOB	11	11	0	24.2 (23-27)	healthy young men without symptoms of TMD or orofacial pain
14	Suzuki, 2016, JOR	16	7	9	26.3 (21-40)	healthy participants without signs or symptoms of TMD
		n	Male	Female	Unknown gender	
Total (%)		20176	4244 (21)	8564 (42.5)	7368 (36.5)	

Table 4: Overview of included case-control, cross-sectional, and prospective cohort studies.

Dx = Bruxism diagnostic method, B type= Bruxism circadian manifestation, AB= Awake bruxism, SB= Sleep bruxism

A study appears into more than one category when multiple types of bruxism were assessed. Such studies are marked with *.

Dx		Self-report		Self-report plus clinical examination		Instrumental		
B type		AB	SB	AB	SB	AB, EMA / EMG	SB, EMG	SB, PSG
Adults	1	Duckro, 1990*	Blanco Aguilera, 2014	No studies	Fernandes, 2012	Chen, 2007	Baba, 2005	Abe, 2013
	2	Macfarlane, 2001*	Duckro, 1990*		Manfredini, 2010	Glaros, 2005	Shedden Mora, 2012	Camparis, 2006
	3	Macfarlane, 2003*	Raphael, 2012*			Mude, 2017*	Yachida, 2012	Lavigne, 1997
	4	Michelotti, 2010	Macfarlane, 2001*				Mude, 2017	Raphael, 2012*
	5	Moss, 1995	Macfarlane, 2003*				Wei, 2017*	Rosseti, 2008*
	6	Rosseti, 2008*	Schiffman, 1992*					Rosseti, 2008
	7	Schiffman, 1992*	Huhtela, 2016*					Smith, 2009
	8	Huhtela, 2016*	Leketas, 2017*					Muzalev, 2017
	9	Leketas, 2017*						
Children	1	Carra, 2011*	Carra, 2011*	No studies	Emodi-Perlman, 2012	No studies	Nagamatsu-Sakaguchi, 2007	No studies
	2	Chun, 1990	Fernandes, 2015*		Kalaykova, 2011, JOP*			
	3	Fernandes, 2015*	van Selms, 2013*		Restrepo, 2008, JOR			
	4	Kalaykova, 2011*	Winocur, 2001*					
	5	van Selms, 2013*	Egermark, 2001*					

6	Winocur, 2001*		
7	Egermark, 2001*		
8	Sermet Elbay, 2017		

Figure legends

Fig. 1. Flow chart of inclusion of articles in systematic review.

Fig. 2. Overview of study designs and time of publication of included articles.

Fig. 1. Inclusion flow chart

Fig. 2 Type of included study designs over time