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ABSTRACT

Objective

The aim of this article was to systematically review the literature to identify papers dealing with risk factors associated with sleep bruxism (SB) in children.

Design

A systematic search was carried out based on the following databases: PubMed, Embase, Scopus, Cochrane Oral Health Group's Trial Register and Cochrane Register of Controlled Trials, Web of Science, LILACs, SciELO. Studies investigating risk factors related to SB after multiple regression analysis and bruxism symptoms assessed with clinical diagnosis or specific questionnaires were searched. Six out of the 4546 initially identified studies were selected.

This review was conducted according to the guidelines from the Cochrane Handbook for Systematic Reviews of Interventions, with reporting in agreement to the Preferred Reporting Items for Systematic Reviews and Meta-analyses guidelines.

Results

Among the six analyzed articles, one randomized clinical trial (RCT) suggested the increase of SB in heavily exposed patients to second hand smoke (SHS) (OR= 4.5, CI= 2.2-9.4), two cross-sectional studies suggested neuroticism as determinant factor for the development of sleep bruxism (OR= 1.9 C.I.= 1.3-2.6), among children and three case-control studies suggested that children with sleep disturbances were more likely to have SB (OR= 3.3, CI= 1.6-6.6). Parafunctional behaviours (OR= 2.3, CI= 1.2-4-3) had a moderate association.

Conclusions

SHS and sleep disturbances presented the strongest association with SB. The most recurrent source of bias was the lack of blinding procedures. Furthermore the use of reliable SB diagnostic procedures should be recommended to increase the quality of future studies. The evidence emerged from the considered studies was clinically relevant.

Keywords

Sleep bruxism, children, review, risk factors, diagnostic criteria.

INTRODUCTION

Bruxism is a repetitive jaw-muscle activity characterized by clenching or grinding of the teeth and/or by bracing or thrusting of the mandible with circadian manifestations (i.e., sleep or awake bruxism)[1,2]. Sleep bruxism (SB) is the sleep-related motor disorder of primary interest for the health of the craniofacial complex, considering several detrimental consequences on the stomatognathic system, including tooth wear, masticatory muscle tenderness and pain, headache and temporomandibular disorders (TMDs)[3,4]. The prevalence of SB in children varies among different studies. A recent review by Manfredini et al.[5], reported a variability of prevalence between 3.5% and 40.6% with a commonly described decrease with age and no gender differences. The variability was mainly due to methodological reasons avoiding the support of any reliable estimate of the prevalence of SB in children.

According to the World Health Organization (WHO) a risk factor is defined as any attribute, characteristic or exposure of an individual that increases the likelihood of developing a disease or injury[6]. Multiple risk factors have been associated to SB. Nevertheless, there are still many unsolved issues concerning the etiology of bruxism that have consequences on the clinical management strategies.

Based on these premises, the purpose of the current study was to conduct a systematic review of the existing literature to determine the relationship between risk factors and SB symptoms in children from 6 to 11 years of age, in the attempt to find answers to the following two clinical research questions:

1.   Which are the identified risk factors for bruxism in children?
2.   Which is the weight of each risk factor?

MATERIALS AND METHODS

Focused question

The selected articles were evaluated according to the following criteria and the selection procedure was thoroughly described through a detailed flow chart, according to the Preferred Reporting Items for Systematic Reviews and Meta-analyses guidelines (PRISMA) Statement[7] (Figure 1). The PICOS template format was not perfectly fitting to all the included studies because of the different study designs, especially concerning the selection of the target population and the comparison group. Nevertheless, it was the best possible approach to a systematic assessment of the included papers.

Search protocol

This systematic review was conducted with reporting in agreement to the PRISMA statement[7] and according to guidelines from the Cochrane Handbook for Systematic Reviews of Interventions[8]. Searches of the databases MEDLINE, PubMed, Embase, Scopus, Cochrane Oral Health Group's Trial Register and Cochrane Register of Controlled Trials, Web of Science, LILACs, SciELO were completed in March 2015. Articles from 1950 to March 2015 were searched using the following Medical Subject Headings (MeSH) terms and key words and limited to "all children (6 to 11 years old)": (1) MeSH terms: bruxism, sleep bruxism, risk factors (2) key words: (bruxer* or sleep brux* AND ((risk orassoc* or relat*)AND factor*) AND diagnosis.

The a priori inclusion criteria for this study were the following:1)RCTs and observational studies assessing the relation between risk factors and bruxism, 2) RCTs and observational studies assessing the diagnosis of bruxism 3) RCTs and observational studies assessing any therapeutic intervention on bruxers, 4) RCTs and observational studies analyzing patients suffering from SB 5) RCTs and observational studies with a sample of minimum 10 patients.Reference lists from articles were explored for other potential studies. Article authors were contacted by e-mail to clarify any relevant article queries. A web research for ongoing trials using the terms bruxism and risk factors through the metaRegister of Controlled Trials on controlled-trials.com, including the National Institutes of Health ClinicalTrials.gov and the International Standard Randomized Controlled Trial NumberRegisters, was run in March 2015.

The reviewing process included Randomized Clinical Trials (RCTs), Controlled Clinical Trials (CCTs), Cohort Studies, Cross-Sectional and Case-Control studies. After duplicates removal, all articles that appeared to meet the inclusion criteria were reviewed.

All considered participants were bruxers, with tooth grinding and/or clenching (age: 6-11y old). These patients were identified by using specific questionnaires, clinical analysis of tooth wear, diagnostic criteria of the American Academy of Sleep Medicine (AASM)[9].

The following exclusion criteria were applied: lack of standardized measures for bruxism evaluation; lack of effective statistical analysis; case reports; reviews; abstracts and author debates or editorials; studies on patients with systemic diseases, syndromes or neurological or psychiatric disorders.

The primary outcome was represented by risk factors for SB in children.

The research strategy returned 4546 potential articles for inclusion. Non–English language literature and unpublished data were not included. Three authors (T.C., A.D., and G.R.) independently identified appropriate articles through examining relevant abstracts, reviewed trials for eligibility, and assessed the quality of trials. Inconsistencies were solved by consensus.

Quality assessment

According to the CRD (Centre for Reviews and Dissemination, University of York)[10] and to the PRISMA[8] statements, evaluation of methodological quality gives an indication of the strength of evidence provided by the study because flaws in the design or in the conduction of a study can result in biases. However, no single approach for assessing methodological soundness is appropriate to all systematic reviews[8]. The GRADE criteria (Grading of Recommendations Assessment, Development and Evaluation), are widely adopted by several authors and organizations throughout the world to assess the overall quality and the risk of bias level in a systematic review. A shortened version of a GRADE-inspired checklist by Meader et al.[11] was adopted. In order to rate the extent of agreement among data collectors, Kappa statistics as described by McHugh ML[12] were performed. Detailed quality assessment and reliability coefficient are illustrated in Table 1

Statistical analysis

Statistical analysis was performed using the R statistical package (version 3.0.1, R Core Team, Foundation for Statistical Computing, Vienna, Austria). To improve the power of risk factors estimates associated with SB symptoms, papers in which a multiple regeression analysis (adjusted for variables statistically associated with SB symptoms) was performed were selected for the review process. Several data extracted from the selected studies were processed in order to obtain either suitable data for the analysis or for presentation in an evidence table; only statistically significant risk factors were included (p value <0.05).

The primary outcomes were risk factors associated to SB in children, calculated as the standardized Odds Ratio (OR) effect size. This effect size was the result of the OR differences between bruxers and controls. Each OR was then weighted by the inverse of its variance and adjusted for small sample bias.

Non-overlapping 95% CI was considered statistically significant. Based on recommendations of the Cochrane Collaboration [7], one author (G.C.) converted the standardized relative risk into a natural log odds ratio.In order to be considered eligible for the final review process, papers had to include OR analysis for investigated risk factors.

Empirical evidence suggests that relative effect measures are, on average, more consistent than absolute measures[13, 14]. Odds Ratio (OR) is the main way to quantify how strongly the presence or absence of a risk factor is associated with the presence or absence of a disease in a given population.

RESULTS

Study selection

Figure 1 details the articles selection process.

4546 potential articles were identified according to the search strategy. After duplicates removal 3443 papers were analyzed. Then 3393 papers were excluded because not relevant to the subject of the study. Of the remaining 50 papers 37 were excluded because did not meet the inclusion and exclusion criteria and other 7 were excluded because non-

human or in vitro studies. The remaining 6 studies met the inclusion and exclusion criteria. Table 2 summarizes the characteristics of each of the 6 included studies.

Population

The 6 included articles were one RCT [15], two cross-sectional studies[16,17] and three case-control studies[18-20]. All studies were randomized except the study by Castelo et al.[17]

Among the selected articles, one study investigated sleep disturbances[20], one study[18] analyzed functional and parafunctional habits, three studies[16, 17, 19] outlined the role of psycho-social factors and two studies considered vicious habits [15,20].The included articles totaled 1063 children, of whom 360 with clinical diagnosis of SB and 703 without SB. Study sample size ranged from 94 to 652 subjects.

Quality assessment

According to GRADE guidelines[11], among the selected sample, the methodological quality was moderate for all studies, thus representing the overall level of evidence The inter-rater reliability, or the percentage of agreement among the selected papers reviewers accordingly to the simplified GRADE checklist, was high (94%) (Table 1).

Risk Factors

The RCT by Montaldo et al.[15] included 153 children aged 8-11 years suffering by SB, exposed to second-hand smoke (SHS) for a period of six months: one group of children was not exposed to SHS, whereas a second group of children was exposed to SHS. Two cross-sectional studies[16,17] investigated the role of psycho-social factors, the first one[16] in a population aged 7-10 years, and the second one[17] in children aged 6-8 years. Among the three case-control studies[18-20], one[18] observed signs, symptoms and parafunctions inchildren aged 8 years for a period of three days per patient; the second one[19] investigated stress and personality traits in children aged 8 years for a period of three days per patient, and the third one[20]analyzed the role of sleep disturbances in children aged 7-10 years, including multiple risk factors: number of sleep hours, sleep with lights on, noise in room and problems during sleep, all children wre followed for a period of three nights per patient.

Outcome

The primary outcome was represented by risk factors related to SB for all the 6 reviewed articles[15-20].

All studies performed SB diagnosis without PSG analysis, using only questionnaires according to AASM criteria[9] and self-report tools to assess SB in patients, three works[15,17,18] included clinical investigation for oral signs and symptoms of bruxism.

Risk of bias across studies

Randomization procedure was considered adequate for the RCT[15] and for the two case-control studies[18,19], all three studies described adequately the process of randomization sequence generation and the procedures of allocation concealment. No sample randomizations were performed in the other case-control study[20]. Two studies[16,17] made a comparison between groups, while others[18-20] were conducted in a case-control design. Mean age of the evaluated samples ranged from 6 to 10 years.

The most recurrent sources of bias were related to the absence of proper blinding procedures for operators and/or patients in all the six reviewed articles and the lack of randomization in one case-control study[20]. All studies presented outcomes appropriately and avoided selective outcome reporting.

Study Results

Table 3 summarizes the results of each article reviewed, by the tipe of study and risk factor analyzed.

The six articles included in the review[15-20] examined the effects of various risk factors on SB: SHS[15], functional and parafunctional habits[18], psycho-social factors[16,17,19] and sleep disturbances[20]

All the 360 children suffering from SB and the 703 controls were exposed to the same risk factors.

Table 4 shows the results of the review process.

Heavy exposition to SHS presented the strongest association with SB in children (O.R. 4.5) C.I. 95% (from 2.2 to 9.4). Other important risk factors for SB were problems during sleeping (O.R. 3.3) C.I. 95% (from 1.6 to 6.6), together with sleep disturbances such as: noise in room (O.R. 2.7) C.I. 95% (from 1.7 to 4.4), sleeping for ≤8 hours per night (O.R. 2.6) C.I. 95% (from 1.5 to 4.4) and sleeping with light on (O.R. 2.4) C.I. 95% (from 1.5 to 3.9). The association between occlusal factors and SB in children was moderate: primary canine wear and tooth clenching during day presented O.R. 2.3 C.I. 95% (from 1.2 to 4.3), and biting on objects O.R. 2.0 C.I. 95% (from 1.2 to 3.3). Psycho-social factors presented different grades of association with SB: strong for high levels of responsibility (O.R. 2.2) C.I. 95% (from 1.1 to 5) and neuroticism (O.R. 1.9) C.I. 95% (from 1.3 to 2.6), moderate for stress (O.R. 1.8) C.I. 95% (from 1.1 to 2.9) and low for maternal age at birth (O.R. 0.9) C.I. 95% (from 0.8 to 0.9).

DISCUSSION

The systematic review of the existing scientific literature confirmed a probable multifactorial model[21]. Sleep disturbances, functional and parafunctional habits, psycho-social factors and second-hand smoke seem to be risk factors associated to SB.

Sleep disturbances presented the strongest correlations with SB onset, together with neuroticism. Parafunctional behaviours and stress have a moderate association with SB in children. Second hand smoke exposure presented a strong association with SB[15] in children. However the number of considered subjects was low (only 6 patients heavily exposed after an observation period of six months) and thus definite evidence cannot be drawn. Considering that SHS and direct exposure to smoke in adolescents and adult population has been proven to play a great role on SB onset in other works [22,23] it can be postulated that this could be the same for children. However further studies on this topic are recommended.

Teeth clenching and biting on objects are moderate risk factors for SB in children [18,19]. Considering the possibility of a direct causal relationship between parafunctions and SB, lips biting, nail biting, pen biting and the prolonged use of pacifiers in children play a stong role in SB genesis. Thus from a clinical perspective the management of SB in children should consider the avoidance of such behaviours.

The role played by stress on SB in children seems to be moderate[19]. The absence of polysomnographic findings limited the study of the impact of stress and psyco-social factors on SB onset.

Sleep disturbances such as sound and light stimuli and reduced sleep time (≤8 hours) presented strong association with SB[20]. However the lack of randomization and blinding procedures was the source of bias of the studies investigating those risk factors.

As stated by Carra et al. [23], polysomnography represents the gold-standard for the diagnosis of SB. However the cost of PSG limits its use mainly in epidemiological studies[24]. Thus an accurate clinical examination may be a reasonable method to be used in large scale studies. Recently some interesting portable devices were introduced in order to ease data gathering[25-28]. Portable devices adopting a combined EMG and electrocardiographic (ECG) recordings showed an increased accuracy with respect to the EMG-based devices and may represent a promising, simple tool for the diagnosis of SB[25, 26, 28].

This review revealed the need for methodologically well-designed and well conducted studies, with adequate statistical analysis OR, in order to better understand which are the risk factors related to bruxism etiology. Moreover, there is a need for further evidence-based longitudinal studies with standardized and validated diagnostic criteria including clinical assessment associated with an interview with parents or guardians and polysomnography or validated portable devices[25-28], in order to obtain more accurate data regarding the prevalence of SB in children.

CONCLUSIONS

Sleep disturbances presented the strongest association with SB while parafunctional behaviours had a moderate association. SHS needs further investigations on greater population samples. From a clinical point of view the suggestion for good sleep hygiene procedures could be of great help in the management of SB in children. The evidence emerged from the considered studies was clinically relevant.

ACKNOWLEDGEMENTS

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