Risk Factors Associated with Probable Sleep Bruxism of Children and Teenagers with Cerebral Palsy

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Aim: *To assess the association between probable sleep bruxism (PSB) and other occlusal characteristics in children and teenagers with Cerebral Palsy (CP).* ***Study design:*** *A cross-sectional study was carried out with 148 participants (74 with CP and 74 without special needs) aged between 2 and 14 years old. Participants underwent an oral clinical examination to evaluate the occlusal characteristics. Parents/caregivers filled out a questionnaire with information related to the typical sounds of PSB, sociodemographic factors and the presence of harmful oral habits in the study participants. Data analysis was carried out, using Chi-square or Fisher's exact test and Odds Ratio ($p<0.05$).* ***Results:*** *PSB and malocclusion (68.9% and 95.9%, respectively) were more prevalent in participants with CP than in participants without CP. The association between PSB and the presence of a wear facet was statistically significant ($p < 0.001$) in participants with CP. In this group, 64.8% of participants with PSB presented tooth wear.* ***Conclusion:*** *Presence of dental wear facets was significantly associated with PSB in individuals with cerebral palsy.*

Keywords: *Cerebral palsy, bruxism, malocclusion.*

INTRODUCTION

Individuals with Cerebral Palsy (CP) are more susceptible to altered masticatory muscle function which predisposes them to orofacial disorders and changes in dental occlusion[1-3]. However, due to the limitations in obtaining an adequate sample and the difficulties of the clinical examination of patients, few studies have reported the occlusal characteristics of individuals with CP[1].

Sleep bruxism is a rhythmic or non-rhythmic masticatory muscle activity that manifests during sleep. It is not a movement disorder or a sleep disorder in otherwise healthy individuals[4]. The prevalence of probable sleep bruxism (PSB) in individuals with CP varies between 25% and 69.4%; the former figure is similar to that found in individuals without CP[5], the latter is higher than that of the general population[6]. Likewise, the prevalence of malocclusions in CP individuals is controversial; a similar prevalence to that of people without special needs has been found[7,8] or, as more often reported, an association between cerebral palsy and malocclusion has been observed[1,9-11].

The constant activity of masticatory muscles during sleep or wakefulness may have clinically detectable consequences. The frequent presence of this activity from childhood to adulthood can lead to conditions that affect the temporomandibular joint, the facial muscles, tooth wear, periodontal diseases and even tooth loss due to trauma[12,13] and impair general health.

Recent studies have sought to determine the prevalence of PSB, its associated factors and clinical manifestations in various populations[14-17]. In individuals with CP, there are reports on the prevalence of malocclusion[9,11,13], but its association with possible protective or

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risk factors, such as bruxism, is still unclear. Thus, the present study aimed to assess the association between probable sleep bruxism and other occlusal characteristics in Brazilian children and adolescents with Cerebral Palsy.

MATERIALS AND METHOD

This study was conducted according to the norms of Resolutions 466/2012 and 510/2016 of the National Health Council. The research project was approved by the Research Ethics Committee of the Federal University of Piauí (CAAE 19201913.7.0000.5214).

Study Design and Participants

A study with cross-sectional design was conducted. At the moment when data was collected there were 92 individuals with CP at the Integrated Center for Special Education (CIES).

This study was carried out at the Integrated Center for Special Education (CIES), a public referral center for the health care of children and adolescents (0-14 years old) with special needs, in the state of Piauí, northeastern Brazil[18,19]. There are no data available on the prevalence of CP in the state or records that show the location of individuals with this disorder outside the institution.

To calculate the sample size, a finite population formula (http://www.openepi.com) was used considering a confidence level of 5%, test power was set at 80% and the prevalence at 50% for the study group. Seventy four individuals were necessary for the study.

Individuals were considered eligible for the study if they had a medical diagnosis of Cerebral Palsy, were aged between 2 and 14 years and presented first deciduous or permanent molars erupted at the time of the data collection.

Individuals who were undergoing or had previously had orthodontic treatment and individuals who had cerebral palsy associated with any other disabilities (such as autism or hydrocephalus) were not included. Thus, 74 individuals (39 males and 35 females) were included in the SG.

The control group (CG) consisted of the same number of individuals (74) with no special needs. Groups were matched for age, gender, and socioeconomic status, and undergoing treatment at the Pediatric Dentistry clinic at the Federal University of Piauí.

Calibration and pilot study

To assess the time necessary for the clinical examination and filling in the questionnaire and also the feasibility of the study, a pilot study was performed. The Kappa-values[20] (0.87 and 0.82 for intra and inter agreement, respectively) were obtained after calibration with clinical examination of 10 children with CP who were not included in the sample.

Data collection

After signing the informed consent form, parents/guardians of all participants filled a multiple choice questionnaire on socioeconomic variables including gender, age, family income in minimum wages, and the use of medication due to the presence of CP.

Information about the gross motor function of participants with CP on the basis of their self-initiated movement with particular emphasis on sitting, walking, and wheeled mobility, the need for assistive technology, including hand-held mobility devices (walkers, crutches, or canes) or wheeled mobility were collected. The gross motor function classification system (GMFCS) was used to classify the participants[21,22].

Clinical examination

The participants' oral hygiene was performed before the exam with toothbrush and fluoride toothpaste. One examiner only carried out the clinical examination of the oral cavities under an artificial lighting reflector in a dental setting in the institution where the study took place. Mouth openers, a dental mirror, an explorer recommended by the WHO, silicone cursor and an endodontic millimeter ruler were used.

Data on the dental occlusion of the participants included the following variables: 1- presence or absence of a posterior crossbite (posterior teeth of the upper arch placed in a palatal position to those of the lower teeth, unilaterally or bilaterally), 2- overbite (normal, increased or decreased), which included cases of anterior open bite or edge-to-edge overjet, 3- overjet (normal, increased or decreased in cases of an anterior cross bite) and, 4- the first molar Angle's classification (Class I, Class II or Class III)[1]. This relative ranking is based on the ratio of the mesio-buccal cusp of the first upper molar placed in the buccal groove of the opposing first molar[10]. An overbite was considered normal when the incisal edge of the maxillary central incisors covered the lower central incisor by 1-2 mm[23]. A normal overjet was when the values of the maximum distance horizontally between the upper and lower central incisors was between 2 to 3 mm[23,24].

Reports of parents and/or caregivers of participants about the participants' sleep were obtained. Grinding the teeth, keeping the teeth together or tightening of the mandible whilst sleeping were considered as a positive report of bruxism. The presence of masticatory muscle hypertrophy, indentations on the tongue or lip and/or a line alba on the inner cheek, damage to the dental hard tissues, repetitive failures of restorations or mechanical wear of the teeth were considered as a positive clinical inspection and indicative of bruxism. Furthermore, PSB was considered as present when a positive clinical inspection was observed, regardless of the parents'/caregivers' report[4,25].

Statistical Analysis

Data analysis was conducted using the statistical software SPSS® version 21.0 for Windows. The descriptive analysis showed the frequencies and percentages in the two groups. The Shapiro Wilk test was applied to analyze the data distribution (p=0.078) and therefore, parametric tests were applied. Pearson's chi-square and Fisher's exact tests were applied to analyze the association between PSB and occlusal characteristics in both groups. In cases where a significant difference was observed between the groups, an odds ratio was applied. The significance level of 5% was considered for all analyses of this study.

RESULTS

Most study participants were male (52.7% for both groups) and had a family income of less than 2 Brazilian minimum wages. Table 1 shows the frequency of ages for the control (CG) and study groups (SG) (GC: 7.38 ± 3.625; SG: 7.36 ± 3.606).

Table 1: Age frequency of the study participants

Age	N	%
2 – 6 years	56	37.8
7 – 14 years	92	62.2
Total	148	100.0

The descriptive characteristics of SG regarding the impairment of cerebral palsy are presented in table 2. A higher proportion of individuals with most severe CP impairment (quadriplegia and level V) were observed.

Table 2: Descriptive characteristics of the individuals with cerebral palsy

		N	%
Clinical Standard			
	Monoplegia	2	2.7
	Diplegia	4	5.4
	Hemiplegia	25	33.8
	Paraplegia	5	6.8
	Quadriplegia	38	51.4
GMFCS Level			
	I	8	10.8
	II	13	17.6
	III	2	2.7
	IV	12	16.2
	V	39	52.7

A higher prevalence of PSB was observed in SG (68.9%; $p < 0.001$). Participants with CP had 6 times higher chance of presenting PSB than participants without special needs (Table 3).

The presence of malocclusion, overjet or overbite was significantly different between groups (Table 3). Only 4.1% of the individuals with CP had a normal occlusion (normal overjet and overbite, right and left molar Angle class I relationship and absence of a posterior crossbite). The chance of malocclusion of participants with CP was 6.535 times greater than that of an individual without special needs.

Most of the participants (72.97%) in SG presented a higher that 3mm overjet; the same condition in CG was present in 52.7% of the participants (Table 3). Normal overbite was more frequent in CG. In SG, the three types of overbite (normal, increased, reduced) had a similar prevalence.

Table 3: Association between PSB and occlusal characteristics in the individuals with (SG) and without Cerebral Palsy (CG)

n		CG		SG		p-value
		%	n	%		
Bruxism†						
	Present	20	27.0	51	68.9	<0.001[a]
	Absent	54	73.0	23	31.1	
	Total	74	100.0	74	100.0	
Malocclusion‡						
	Present	58	78.4	71	95.9	0.002[b]
	Absent	16	21.6	3	4.1	
	Total	74	100.0	74	100.0	
Overjet						
	Normal	30	40.54	15	20.27	
	Larger	39	52.70	54	72.97	0.024[a]
	Reduced	5	6.76	5	6.76	
	Total	74	100.0	74	100.0	
Overbite						
	Normal	42	56.75	22	29.73	
	Larger	20	27.00	27	36.49	0.003[a]
	Reduced	12	16.25	25	33.78	
	Total	74	100.0	74	100.0	

[a]Pearson's chi-square test; [b]Fisher's Exact Test; †Odds Ratio 5.987 (CI 95% 2.940-12.190; p-value < 0.001); ‡Odds Ratio 6.535 (CI 95% 1.814-23.255; p-value < 0.001)

PSB was more frequent in participants with who were quadriplegic (73.6%) or hemiplegic (64%). As for the medication used, among the bruxists: 31% used anticonvulsants, 66.7% used anticonvulsants along with other medications such as a muscle relaxant, and 2.4% used another type of medication such as antipsychotics. No statistically significant association was found between PSB and the type of dentition or PSB and the presence of posterior crossbite.

Table 4 shows a higher prevalence of harmonic occlusion (Angle's class I on both sides) in CG (75.7% CG, 54.1% SG, $p = 0.006$). A statistically significant association was observed between the presence of PSB and the presence of wear facets ($p < 0.001$); 64.8% of the participants with bruxism presented incisor and/or first molars wear facets (Table 5).

Table 4: Distribution of participants with (SG) and without cerebral palsy (CG) according with their Angle Classification

		Left Class			
		Class I	Class II	Class III	Total
SG					
Right Class	Class I	56 (75.7%)	2 (2.7%)	1 (1.4%)	59 (79.7%)
Class II	2 (2.7%)	10 (13.5%)	0 (0%)	12 (16.2%)	
Class III	0 (0%)	1 (1.4%)	2 (2.7%)	3 (4.1%)	
Total	58 (78.4%)	13 (17.6%)	3 (4.1%)	74 (100%)	
CG					
Right Class	Class I	40 (54.1%)	3 (4.1%)	4 (5.4%)	47 (63.5%)
Class II	10 (13.5%)	9 (12.2%)	0 (0%)	19 (25.7%)	
Class III	2 (2.7%)	0 (0%)	6 (8.1%)	8 (10.8%)	
Total	52 (70.3%)	12 (16.2%)	10 (13.5%)	74 (100%)	

Table 5: Association between probable sleep bruxism (PSB) and presence of wear facets in individuals with Cerebral Palsy

	Wear facets				Total		p-value
	Present		Absent				
	n	%	N	%	n	%	
PSB							
Present	46	64.8	25	35.2	71	48.0	<0.001[a]
Absent	16	20.8	61	79.2	77	52.0	
Total	62	41.9	86	58.1	148	100.0	

[a]Pearson's chi-square test.

DISCUSSION

Individuals with CP are predisposed to have occlusal alterations and oral disorders due to their orofacial dysfunctions[1,3,26]. In the present study, a high prevalence of PSB (68.9%) and occlusal alterations (96.9%) in individuals with this condition was observed. These results corroborate most studies in the literature, which indicate a higher prevalence of PSB[6,7,13,27] and malocclusion[1,9-11,26,28] in individuals with CP when compared with individuals with no special needs.

The prevalence of PSB observed in our study is similar to the prevalence reported in individuals with CP in other studies, which varies from 25% to 69.4%[2,6,7,13]. However, these studies include possible, probable or definitive sleep bruxism. This variability is due to the different diagnosis methods used (questionnaire, self or caregivers' reports, clinical examination, polysomnographic or electromyographic)[4], which makes it difficult to compare and discuss the results. Therefore, a standardization of the diagnostic method for this condition is encouraged, so that reliable statistic data on the prevalence of possible, probable and definitive sleep bruxism in individuals with CP are available.

Studies that diagnosed PSB in the same way that it was diagnosed in this study, report a lower prevalence[7,13]. Rosenbaum *et al* [7] found a 44% prevalence of PSB among individuals with cerebral palsy. The diagnosis was made through the observation of facets of clinically detectable attrition and questioning of the parents/caregivers. Another study presented a prevalence of 36.9%, with a diagnosis made through a questionnaire applied to caregivers[13]. In our study, the clinical features were considered for the diagnosis of PSB, regardless of the positive report of parents or caregivers. In addition, our study only included participants with the absence of any other medical or psychological disorder that could be associated with abnormal movements of the mouth and mandible during sleep and who had no other sleep disorders, in order to reduce detection bias.

The prevalence measured by Peres *et al* [6] in individuals with CP (69.4%) are similar to that measured here. The diagnosis was based on the caregivers' reports and clinical examination. In the present study, a positive diagnosis for PSB was considered when the presence of the wear facets in the first molars and/or incisors was observed, regardless of the report of parents/caregivers.

Malocclusions may predispose one to the occurrence of carious lesions due to the difficulty in biofilm control[29]. For individuals with CP, who additionally have difficulty maintaining their oral hygiene, this is a combination of risk factors for poor oral health[30]. Malocclusions in individuals with CP[1,9-11,26,28] and other conditions[31-33]. such as those found in this study, are well documented.

No association was found between the type, topographic classification and GMFCS of CP and the occurrence of bruxism and malocclusion. The low number of individuals in some groups (monoplegia and level III, for example), could explain this finding. A larger sample could detect different results. The scarcity of studies with this type of analysis makes it difficult to compare these aspects. However, cerebral palsy seems to be significantly associated with the occurrence of bruxism and the occlusal characteristics compatible with wear[11,13,25].

The reports in the literature on the association between bruxism and malocclusion are conflicting[6,34]. Studies have shown no association between a para-functional habit and the presence of malocclusion, nor with the type of dentition[6,34]. However, an epidemiological study showed that bruxism was associated with occlusal changes such as posterior crossbite, tooth loss and incisor relationship[35]. It is believed that during chewing movements, the change in dental contact due to malocclusion may create a greater tendency for deviation of the mandible, predisposing one to the occurrence of bruxism[36].

Studies have shown the association between bruxism and the use of drugs such as anticonvulsants and antipsychotics[37-39]. There was a reduction in bruxism with the use of clonazepan[37] and lamotrigine[38], drugs commonly used by individuals with CP. In the present study, the report of caregivers on the use of antipsychotics and anticonvulsants by individuals with CP may be one of the factors that justify the high occurrence of PSB.

An association between the presence of wear facets and PSB in individuals with CP was observed in this study. The clinical condition most frequently associated with bruxism is an abnormal wear of

the dental structure[39]. Consistent with other reports in the literature, it has been stated that sleep bruxism increased the risk of the presence of facets and therefore, is directly associated with the clinical manifestation of bruxism[5,40].

The presence of wear facets is common in individuals with bruxism[41]. However, their presence is not a method to be adopted for the diagnosis of this condition[4]. The level of wear may not directly reflect the degree of bruxism present. The individual may have marked wear from past chewing muscle activity, no longer present at the time of evaluation, leading to an overestimated prevalence. On the other hand, individuals who have recently developed the habit may still not show signs of tooth wear, underestimating the occurrence of bruxism[5]. For these reasons, in the present study, the presence of wear facets was considered as a clinical consequence of bruxism. The occurrence of PSB is a risk factor for wear facets in the participants of this study.

This investigation has shortcomings, such as the sample size and the absence of a a definite sleep bruxism diagnosis (i.e., by means of polysomnographic recordings). However, a recent study indicated that in PSB cases there was a 83% concordance between the parent's report and the polysomnographic diagnosis[42].

Therefore, the high occurrence of bruxism and malocclusion observed among individuals with CP in this study supports the importance of preventive oral health care, in addition to orientations to parents and caregivers.

CONCLUSION

The results of our study showed that the prevalence of PSB is high among children and teenagers with CP. The novel finding of the present study was that the occurrence of PSB is a risk factor for dental wear facets in children and teenagers with CP. Future studies are encouraged for a better understanding of the multifactorial mechanism involved in the onset of PSB and the associated factors in individuals with special needs.

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