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ORIGINAL ARTICLE

Prevalence and Risk Factors for Bruxism in Children with Congenital Zika Virus Syndrome: A Case-Control Study

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ABSTRACT

Objective: To determine the prevalence of possible sleep and awake bruxism and its related risk factors in children with Congenital Zika Virus Syndrome. **Material and Methods:** The case group consists of 20 children with congenital Zika, age range from 5 to 6 years old, and the control group consists of 120 regular children of the same age group. A questionnaire was used to assess bruxism and associate possible risk factors such as medications, systemic disorders, and sleep relationships, and a validated Oral Behaviors Checklist (OBC) was applied. Absolute and percentage frequencies of each outcome variable and mean and standard deviation of each item of the OBC questionnaire were calculated, respectively, compared between case and control groups using Fisher's exact or Pearson's chi-square and Mann-Whitney tests. **Results:** Only 25% of the children with microcephaly do not present swallowing difficulty, significantly lower compared to the 95.8% of the children in the control group who do not ($p<0.001$). The occurrence of bruxism was significantly higher in the case group (47.4%) compared to the control group (10.2%). In addition, 89.8% of children did not grind their teeth in the control group, compared to only 52.6% in the case group. There was a statistically significant difference between the case and control group ($p<0.001$). **Conclusion:** Dysphagia, feeding routes, and reflux in children with cerebral palsy are risk factors for possible awake bruxism, and children with congenital Zika virus syndrome showed a prevalence for possible bruxism compared to regular children.

Keywords: Bruxism; Microcephaly; Zika virus.

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Introduction

In late April 2015 and early 2016, a ZIKV epidemic occurred in Brazil. It is estimated that more than 1 million Brazilians had ZIKV infections in 2015. In Brazil, six months after the start of the ZIKV outbreak, there was an unusual increase in newborns with microcephaly [1,2]. From 2016 to 2020, 1,638 cases of Zika were confirmed in the Municipality of Fortaleza. In the first weeks of 2022, only 66 notifications of Zika were registered in the information system: 51 already duly investigated (1 confirmed and 50 discarded) and 15 still under investigation [3].

The most severe neurological sequela of phenotypically Zika virus infection is certainly microcephaly, which is not a disease but a sign of destruction or deficit of brain growth [4]. Thus, boys with a measurement equal to or less than 31.9 centimeters (cm) and girls with a value equal to or less than 31.5 cm born at 37 weeks of pregnancy, or more are considered neonates with microcephaly.

In addition, it is suggested that neurological alterations lead to hypotonia of orofacial musculature, followed by inadequate lingual posture, thus resulting in poor swallowing reflex and frequent mouth breathing. When the mouth is left ajar for long periods, adequate transverse maxillary growth is limited, leading to a tendency towards maxillary atresia and an ogival palate [5-7]. Moreover, childhood Bruxism (Bx) has been reported by some mothers of patients with CZVS. However, no other works on this topic were found in the databases searched, to date.

In 2018, an international consensus was reached on a simple and pragmatic definition of Bx as a repetitive masticatory muscle activity that is characterized by clenching or grinding of the teeth and/or by bracing or thrusting of the mandible, without the necessary presence of tooth contact, and that is specified as either sleep bruxism (SB) or awake bruxism (AB), depending on its circadian phenotype. Moreover, it cannot be consider either a sleep disorder or a movement disorder in otherwise healthy individuals [8].

Along with the new definition, Lobbezoo et al. [8] proposed a system for grading assessments of bruxism. Accordingly, possible sleep/awake bruxism is based on self-report only; probable sleep/awake bruxism on self-report plus clinical inspection; definite sleep bruxism on self-report, clinical inspection plus polysomnography (preferably combined with audio/ video recordings); and definite awake bruxism on self-report, clinical inspection plus electromyography (preferably combined with ecological momentary assessment/experience sampling methodology [EMA/ESM]).

The literature points out that the risk factors for bruxism are the consumption of tobacco, alcohol, coffee, obstructive sleep apnea syndrome (OSAS), and anxiety disorders, usually present in adults. Emotional factors, behavioral disorders, genetic abnormalities, and sleep syndromes or pathologies have been pointed out in children and adolescents [9]. Conditions such as reflux esophagitis, depression, respiratory diseases, or nocturnal frontal lobe epilepsy, as well as emotional instability, tension, and attenuated psychotic personality, also appear as features associated with bruxism [9].

The relevance of the present study is due to the need to investigate the prevalence and risk factors associated with sleep and awake bruxism in children born with CZVS so that this condition can be better managed, bringing greater well-being to the child. Thus, this study aimed to determine the prevalence of possible sleep and awake bruxism and its related risk factors in a sample of Brazilian children with Congenital Zika Virus Syndrome, compared with regular children of the same age group, by a parent-report questionnaire.

Material and Methods

Study Design and Ethical Clearance

This case-control study was approved by the Research Ethics Committee of the Christus University Center with opinion number: 1,881,086. The research participants signed the Informed Consent Form to participate in the research, where the objectives, methodology, risks, and benefits related to the research were clearly explained, as required by the Guidelines and Regulatory Standards of the National Health Council (Resolution No. 466/2012).

Sample Calculation

The study sample consists of children in the age group of 5 and 6 years, created in 2016 to care for children with Congenital Zika Virus Syndrome (CZVS). As CZVS is a rare condition, to make the study more reliable at the population comparison level, a control group was carried out with regular children of the same age group - between 5 and 6 years old. In this way, in addition to the risk factors for childhood possible awake and sleep bruxism, it will be possible to observe its prevalence in the local sample.

The inclusion criteria for the case group were children born with Congenital Zika Virus Syndrome (CZVS), aged 5 and 6 years, and who are part of the Center for the Study and Care of Microcephaly – NEAM. The exclusion criteria were children affected by diseases other than CZVS or children whose parents did not want to participate in the study. For the control group, the inclusion criteria were children in the same age group of 5 and 6 years, with the absence of congenital Zika virus or any other relevant systemic manifestation, and the exclusion criteria were parents who did not agree to participate in the research and/or who were not available and children who presented any systemic or neurological alteration.

A study observed that the frequency of seizures is higher in patients with microcephaly due to the Zika virus than in patients without this condition (30% vs 0%). Since this parameter is directly related to TMD [13], it was estimated necessary to evaluate 116 patients to obtain a sample that represents with 90% power and 95% confidence the alternative hypothesis of this work and, because of the possibility of sample loss, 20% was added to this totaling 140 patients. As microcephaly due to Zika is an uncommon disease, a ratio of 6 controls for each case was established [14], thus totaling 20 cases and 120 controls.

Data Collection

Two different questionnaires were applied: one with 26 objective questions that were elaborated by the researchers, to be answered by the parents or guardians of the child with Congenital Zika Virus Syndrome; the second questionnaire was the Oral Behaviors Checklist (OBC), which is a validated self-report questionnaire of 21 questions, two about nighttime habits and 19 about daytime. For each statement, there is an option to mark among five options related to the frequency of each event, ranging from "never" = score 0 to "4 to 7 nights a week" = score 4, which quantifies the frequency of Oral Behaviors (OBs) [10]. Its validity was successfully verified concerning EMG [11]. OBC was included in the Diagnostic Criteria for Temporomandibular Disorders (DC/TMD) as a screening tool due to the known contribution of OCs to TMD [12].

The questions, both the Appendix and the OBC, are objective, multiple-choice, being possible to mark only one alternative per question. The question banks are intended to assess the manifestation of possible sleep and awake bruxism in children, evaluate their pattern, and relate possible risk factors, such as medications in use, other systemic disorders, such as those of the respiratory system, and those related to sleep. Since those children could not answer the questions, the results of the present study are based on parents' reports for both questionnaires.

Through the application and subsequent analysis of the forms, we also seek, with the publication of the results, to estimate the epidemiology of possible awake and sleep bruxism in children with CZVS, to evaluate the prevalence of this condition, to establish the risk factors, and to seek the control of this condition, so that it does not negatively affect the quality of life of children.

Data Analysis

The data were tabulated in Microsoft Excel and exported to the Statistical Package for Social Science (SPSS) software version 20.0 for Windows (IBM Corp., Armonk, NY, USA), in which the analyzes were performed adopting a 95% confidence. Absolute and percentage frequencies of each outcome variable and mean and standard deviation of each item of the OBC questionnaire were calculated, respectively, compared between case and control groups using Fisher's exact or Pearson's chi-square and Mann-Whitney tests. The strategies for dichotomizing the variables were based on the median.

Results

About 30.8% of deliveries in the control group were average, while 69.2% were cesarean sections. In the case group of patients with microcephaly, 25% births were natural deliveries and 75% of cesarean delivery (p=0.598). 74.2% of the control group was born of premature birth, and 90% of the case group was also born (p=0.122).

Regarding heredity, about 72.5% of the parents of the control group did not have bruxism, while 80% of the case group did not. When only the mother was considered, the control group presented 13.3%, and the case group presented 10%. When considering only the father, 12.5% of the control group presented bruxism compared to 10% of the case group — considering both the father and the mother, 1.7% of the control group presented, while 0.0% of the case group (p=0.873) (Table 1).

Table 1. Distribution of children according to age, type of delivery, and spouse has bruxism.

Variables	Group		p-value
	Control N (%)	Case N (%)	
Age			
Up to 6 years	52 (43.3)	7 (35.0)	0.485
>6 years	68 (56.7)	13 (65.0)	
Cesarean delivery			
No	37 (30.8)	5 (25.0)	0.598
Yes	83 (69.2)	15 (75.0)	
Premature birth			
No	31 (25.8)	2 (10.0)	0.122
Yes	89 (74.2)	18 (90.0)	
Spouse has bruxism			
No	87 (72.5)	16 (80.0)	0.873
Yes, the mother	16 (13.3)	2 (10.0)	
Yes, the father	15 (12.5)	2 (10.0)	
Yes, mom and dad	2 (1.7)	0 (0.0)	

The number of children in the control group who didn't breastfeed was 7.5% and in the study group, 20% didn't breastfeed. By three months, 13.3% of the children in the control group suckled, and 35% in the case group. By six months, 13.3% of the control group and 10% of the case group breastfed. For over 12 months, the control group had 65.8%, and the case group had 35% of the children (p=0.014) (Table 2).

Regarding swallowing difficulty in the case group, 75% of the children have difficulty swallowing, while 4.2% of the control group has difficulty swallowing. Only 25% of the children with microcephaly do not present swallowing difficulty, significantly lower compared to the 95.8% of the children in the control group who do not. There was a statistically significant difference between the means presented for the case group and control ($p<0.001$) (Table 2).

When considering the feeding route, the control group had 99.2% of feeding by mouth of both solid and liquid foods. In the case group, the same data is 30%, while feeding by mouth only pasty and liquid foods is 45%, and by tube (enteral feeding) is 25%. There was a statistically significant difference between the means presented for the case and control group ($p<0.001$) (Table 2).

When pacifier sucking habit was assessed, 50% of the case groups did not use it, and 55% of the control group did. With 30.8% of the control group children having the habit for more than 12 months compared to 45% of the case group ($p=0.684$) (Table 2).

When bruxism was analyzed, the result was significantly higher in the case group, with 47.4% of children grinding their teeth while sleeping and awake compared to 10.2% in the control group. In addition, 89.8% of children did not grind their teeth in the control group, compared to only 52.6% in the case group. There was a statistically significant difference between the means presented for the case and control group ($p<0.001$) (Table 2).

When asked if any professional dental surgeon had already examined and clarified the possible causes of bruxism, 47.4% of the mothers in the case group answered yes, and 52.6% answered no. In the control group, 10.2% answered yes, while 89.8% answered no. In the control group, 10.2% answered yes, while 89.8% answered no. There was a statistically significant difference between the means presented for the case and control groups ($p<0.001$) (Table 2). Regarding snoring, 50% of the case group snores, and 50% do not. Of the control group, 52.5% did not snore, and 47.5% did. Regarding snoring, 55% of children with microcephaly never snore, 15% snore 1-2 times a week, 10% snore 3-4 times a week, and 20% daily. In the control group, 60% never snore, 7.5% snore 1-2 times a month, 15% 1-2 times a week, 5.8% snore 3-4 times a week, and 11.7% snore daily (Table 2).

Table 2. Distribution of children according to breastfeeding, dysphagia, feeding routes, pacifier, bruxism and snoring.

Variables	Group		p-value	OR	CI95%
	Control N (%)	Case N (%)			
Child Suckled					
No	9 (7.5)	4 (20.0)*	0.014	RC	-
Yes, around 3 months	16 (13.3)	7 (35.0)*		0.9844	0.2251 to 4.305
Yes, around 6 months	16 (13.3)	2 (10.0)		0.2813	0.04274 to 1.851
Yes, for more than 12 months	79 (65.8)*	7 (35.0)		0.1994	0.04872 to 0.8159
Dysphagia					
No	115 (95.8)	5 (25.0)	<0.001	RC	-
Yes	5 (4.2)	15 (75.0)*		69.00	17.86 to 266.6
Child feeds like					
Mouth (Pasty/liquid foods only)	1 (0.8)	9 (45.0)*	<0.001	RC	-
Mouth (solid and liquid foods)	119 (99.2)*	6 (30.0)		0.00560	0.00060 to 0.0517
Probe (Enteral feeding)	0 (0.0)	5 (25.0)*		1.737	0.05977 to 50.47
Pacifier Habit					
No	66 (55.0)	10 (50.0)	0.684	RC	-
Yes, around 3 months	5 (4.2)	1 (5.0)		1.320	0.1394 to 12.50
Yes, around 6 months	6 (5.0)	0 (0.0)		0.4872	0.02550 to 9.307
Yes, for more than 12 months	43 (35.8)	9 (45.0)		1.381	0.5188 to 3.678

If the Child has Bruxism, CD Clarified?					
No	88 (89.8)*	10 (52.6)	<0.001	RC	-
Yes	10 (10.2)	9 (47.4)*		7.920	2.602 to 24.11
Snores					
No	63 (52.5)	10 (50.0)	0.836	RC	-
Yes	57 (47.5)	10 (50.0)		1.105	0.4287 to 2.850
Snoring Frequencies					
Never	72 (60.0)	11 (55.0)	0.564	RC	-
1-2 times a month	9 (7.5)	0 (0.0)		0.3318	0.01804 to 6.102
1-2 times per week	18 (15.0)	3 (15.0)		1.091	0.2751 to 4.325
3-4 times a week	7 (5.8)	2 (10.0)		1.870	0.3433 to 10.19
Practically every day	14 (11.7)	4 (20.0)		1.870	0.5200 to 6.726

RC = Reference Group; *p< 0.05, chi-square test.

When the presence of sleep disorders was assessed, 5.0% of the case group answered yes, while 95.0% answered no. The control group, on the other hand, 100%, did not present data regarding sleep disorders (p=0.014) (Table 3). It was observed that 50% of the children with microcephaly had reflux, while 50% did not. In the control group, only 13.3% of the sample declared having reflux, and 86.7% answered that they did not. There was a statistically significant difference between the means presented for the case and control group (p<0.001) (Table 3).

When sleep and sleep quality were assessed, the following data were obtained: 85% of the Zika group sample reported having a good night's sleep, and 15% reported not. In the control group, the percentage was higher, with 91.7% affirmative response and 8.3% the percentage of children who do not sleep well (p=0.342). Sleep duration varied considerably between the groups. In the control group, 45% of children slept 9-11 hours, 35% 8-9 hours, 10.8% 7-8 hours, 7.5% 6-7 hours, and 1.5% less than 5 hours. In the study group, the average number of hours of sleep was lower, with 35% sleeping 9 to 11 hours, 30% 8 to 9 hours, 15% 7 to 8 hours, 10% 6 to 7 hours, and 10% less than 5 hours (p=0.279).

Regarding time to sleep onset, in the study group, 25% took about 15 minutes to fall asleep, 40% took 15 to 30 minutes, 15% took 30 to 45 minutes, and 20% took more than 60 minutes. In the control group, 47.5% took up to 15 minutes, 29.2% from 15 to 30 minutes, 8.3% from 30 to 45 minutes, 5.8% from 45 to 60 minutes, and 9.2% more than 60 minutes (p=0.166) (Table 3).

Table 3. Distribution of children according to sleep sickness, reflux and sleep quality and duration.

Variables	Group		p-value	OR	CI95%
	Control N (%)	Case N (%)			
Diagnosis of sleep-related illness					
No	120 (100.0)	19 (95.0)	0.014	RC	-
Yes	0 (0.0)	1 (5.0)		18.54	0.7282 to 471.9
Reflux					
No	104 (86.7)*	10 (50.0)	<0.001	RC	-
Yes	16 (13.3)	10 (50.0)*		6.500	2.338 to 18.07
Child sleeps well at night					
No	10 (8.3)	3 (15.0)	0.342	RC	-
Yes	110 (91.7)	17 (85.0)		0.5152	0.1286 to 2.064
Child sleeping hours					
Less than 5h	2 (1.7)	2 (10.0)	0.279	RC	-
6 to 7h	9 (7.5)	2 (10.0)		0.2222	0.01845 to 2.676
7 to 8h	13 (10.8)	3 (15.0)		0.2308	0.02250 to 2.367
8 to 9 am	42 (35.0)	6 (30.0)		0.1429	0.01682 to 1.213
9 to 11 am	54 (45.0)	7 (35.0)		0.1296	0.01567 to 1.072
Child time to fall asleep					

<15 min	57 (47.5)	5 (25.0)	0.166	RC	-
15 to 30 min	35 (29.2)	8 (40.0)		2.606	0.7893 to 8.602
30 to 45 min	10 (8.3)	3 (15.0)		3.420	0.7033 to 16.63
45min to 60min	7 (5.8)	0 (0.0)		0.6970	0.03490 to 13.92
>60min	11 (9.2)	4 (20.0)		4.145	0.9579 to 17.94

RC = Reference Group; *p< 0.05, chi-square test.

Table 4 presents the values referring to the Oral Behaviors Checklist (OBC) questionnaire. The results were:

- Q1. Clenches or grinds teeth when asleep, based on any information you may have: The control group's mean was 1.03, and the case group was 1.40 with no significance (p>0.05).
- Q2. Sleeps in a position that puts pressure on the jaw: The control group presented a mean of 2.25 and the case group of 0.35, being possible to observe a higher frequency of sleeping in a position pressing the mandible in this group than in the case group of children with microcephaly. There was a statistically significant difference between the means presented for the case and the control group (p<0.001).
- Q3. Grinding teeth when awake: It was observed in the control group a mean of 0.13 and in the case group a mean of 1.15, thus concluding that the case group of children with microcephaly had a higher frequency of grinding teeth awake than the control group. There was a statistically significant difference between the means presented for the case and the control group (p<0.001).
- Q4. Clenches teeth when awake: The control group's mean was 0.18, and that of the case group was 1.10. Thus, children with microcephaly had a higher frequency of clenching their teeth than the control group. There was a statistically significant difference between the means presented for the case and the control group (p<0.001).
- Q5. Presses touch or holds teeth in contact other than when eating (i.e., makes contact between upper and lower teeth): The control group presented a mean of 0.37 and the case group of 1.05, thus showing that children with microcephaly press, touch, or keep their teeth in contact beyond when eating more frequently (p=0.001).
- Q6. Holds, stiffens, or tenses muscles without clenching or pulling teeth together: The control group had a mean of 0.20 and the case group of 0.80, also showing that children with microcephaly are more likely to hold, tense, or stiffen their muscles without touching their teeth than the control group. There was a statistically significant difference between the means presented for the case and control group (p<0.001).
- Q7. Holds or protrudes the jaw forward or to the side: The control group had a mean of 0.34 and the case group 0.65 without significance (p=0.068).
- Q8. Presses tongue hard against teeth: The case group presented a mean of 0.89 compared to the control group, with a mean of 0.21 being more significant (p=0.001).
- Q9. Put tongue between teeth: The mean of the children with microcephaly was higher than that of the control group, being 1.25 case group and 0.20 control group. Thus, it was statistically significant that the case group more often put the tongue between the teeth. There was a statistically significant difference between the means presented for the case and the control group (p<0.001).
- Q10. Bites, chews, or plays with tongue, cheeks, or lips: The control group presented 0.38 in its meanwhile, the case group presented a higher mean of 1.00 (p=0.009).
- Q11. Holds the jaw in a rigid or tense position, such as to hold or protect the jaw: The case group of children with microcephaly presented a mean of 0.75, higher than the control group's mean of 0.22. There was a statistically significant difference between the means presented for the case and the control group (p<0.001).

- Q12. Holds between teeth or bite objects such as hair, pipe, pencils, pens, fingers, nails, etc.: The control group had a mean of 1.15, and the case group had a mean of 0.95 (p=0.332).
- Q13. Chews gum: The control group had a mean of 0.66 for chewing gum use, higher than the case group with a mean of 0.00, with significance concerning the case group that does not use it. There was a statistically significant difference between the means presented for the case and the control group (p<0.001).
- Q14. Plays a musical instrument that involves the use of the mouth or jaw (e.g., wind, brass, or string instruments): The control group mean was 0.18, and the case group mean was 0.05 (p=0.253).
- Q15. Leans with hand on the jaw, as if to place or rest the chin on hand: A control group with a mean of 0.56 and the case group with a mean of 0.30 (p=0.054).
- Q16. Chews food only on one side: A control group, with a mean of 0.65, and the case group, with a mean of 0.40, not showing statistical significance (p=0.082).
- Q17. Eats between meals: The mean of the case group showed that children with microcephaly eat less between meals, with a value of 0.45 being lower than the mean of the control group of 1.71. There was a statistically significant difference between the means presented for the case and control group (p<0.001).
- Q18. Prolonged speech: The study showed that the case group had a lower speech frequency, with a mean of 0.35, compared to the control group, with a mean of 1.71; children with microcephaly hardly speak. There was a statistically significant difference between the means presented for the case and the control group (p<0.001).
- Q19. Sings: The study showed that the case group had a lower singing frequency, with a mean of 0.15, compared to the control group, with a mean of 1.68; children with microcephaly hardly sing. There was a statistically significant difference between the means presented for the case and the control group (p<0.001).
- Q20. Yawns: The study showed that the case group had a higher yawning frequency r with a mean of 1.75 compared to the control group with a mean of 0.72; children with microcephaly yawn more than children without microcephaly. There was a statistically significant difference between the means presented for the case and the control group (p<0.001).
- Q21. Holds the phone between head and shoulders: The groups did not differ much in the means, with the control group means being 0.11 and the case group 0.10 (p=0.925).

Table 4. Mean and standard deviation values and p-value of the Oral Habits Questionnaire (OBC) regarding the case and control groups' sleep bruxism and waking bruxism.

OBC Questions	Group		p-value	Δ	CI 95%
	Control	Case			
Q1	1.03±1.58	1.40±1.50	0.179	0.37	-0.38 to 1.12
Q2	2.25±1.76	0.35±0.93	<0.001	-1.90	-2.43 to -1.37
Q3	0.13±0.46	1.15±0.99	<0.001	1.03	0.56 to 1.49
Q4	0.18±0.51	1.10±1.02	<0.001	0.93	0.44 to 1.41
Q5	0.37±0.85	1.05±1.15	0.001	0.68	0.13 to 1.24
Q6	0.20±0.54	0.80±0.95	<0.001	0.60	0.15 to 1.05
Q7	0.34±0.68	0.65±0.99	0.068	0.31	-0.16 to 0.79
Q8	0.21±0.53	0.89±1.15	0.001	0.69	0.13 to 1.25
Q9	0.20±0.51	1.25±1.12	<0.001	1.05	0.52 to 1.58
Q10	0.38±0.61	1.00±1.12	0.009	0.63	0.09 to 1.16
Q11	0.22±0.47	0.75±0.79	<0.001	0.53	0.16 to 0.91
Q12	1.15±1.04	0.95±1.10	0.332	-0.20	-0.74 to 0.34
Q13	0.66±0.76	0.00±0.00	<0.001	-0.66	-0.80 to -0.52
Q14	0.18±0.48	0.05±0.22	0.253	-0.13	-0.26 to 0.01
Q15	0.56±0.70	0.30±0.66	0.054	-0.26	-0.59 to 0.07

Q16	0.65±0.97	0.40±0.99	0,082	-0.25	-0.74 to 0.24
Q17	1.71±0.91	0.45±1.00	<0.001	-1.26	-1.75 to -0.77
Q18	1.71±1.43	0.35±0.75	<0.001	-1.36	-1.78 to -0.93
Q19	1.68±1.05	0.15±0.67	<0.001	-1.53	-1.89 to -1.17
Q20	0.72±0.72	1.75±0.79	<0.001	1.03	0.65 to 1.42
Q21	0.11±0.36	0.10±0.31	0.925	-0.01	-0.16 to 0.15

Discussion

The prevalence of awake and sleep bruxism found in patients with Congenital Zika Virus Syndrome was higher compared to the control group, which means that patients in the study group must have a greater predisposition to possible awake and sleep bruxism compared to the control group. According to Lavigne et al. [15], the prevalence of bruxism in children with neurological disorders is significantly higher than the rate found in normal children.

The study by Simões-Zenari and Bitar [17] investigated the relationship between bruxism and factors associated with oral habits, orofacial motricity, and chewing, breathing, and swallowing functions in 141 children aged 4 to 6 years. Through questionnaires applied to parents for bruxism diagnosis and clinical evaluation of orofacial motricity, results were obtained that children with sialorrhea during sleep, who had the habit of using a pacifier, biting lips, biting nails or with altered cheek tone and bite type, have an increased risk of triggering bruxism.

According to the data analysis performed, it was observed that there is statistical relevance regarding the number of children with Zika virus who have dysphagia, reaching 75% of the percentage of them who have difficulty swallowing solid, pasty, or liquid foods. According to Matsuo et al. [18], dysphagia can result from a wide variety of diseases or disorders, as is the case with Zika Virus Congenital Syndrome, causing deficiencies in function or structure in the oral cavity, pharynx, larynx, or esophageal sphincters.

In the study by Oliveira et al. [19], the authors compared dysphagia in children with microcephaly associated with SCZV and normal children, born in Pernambuco, Brazil, a region close to and with similar cultural characteristics to where the present research was carried out. The study was carried out through assessments based on three instruments: a questionnaire applied to caregivers, a clinical assessment of the stomatognathic system and clinical assessment of swallowing. It was seen that the first group had a considerably higher incidence of the mentioned condition, being 79.3% in relation to the 20.7% of the control group. In addition, it was observed that the oral and pharyngeal phases of swallowing were the most affected, having as characteristics in the oral phase the loss of food through the mouth, abnormal cervical auscultation, and oral food stasis, ranging between 56.9% and 75.9%. In the pharyngeal phase of swallowing, it can be seen that coughing, choking, and reduced laryngeal elevation were frequent, being present between 21% and 31% of children with CZVS. The study points out that this condition has a multifactorial cause. However, it is estimated that there is a cortical lesion, which can affect neuromotor function. Hypotonia in the lips and cheeks in 70% of the study group may also have played an essential role in swallowing difficulty.

In this study, the degree of dysphagia was also measured; Amorim et al. [20] believe that the deficiency in suction and swallowing, dysphagia, mouth breathing, delayed eruption of deciduous teeth, alteration in the sequence of dental eruption and enamel hypoplasia present in children with microcephaly is a consequence of altered muscle tone, whether hypotonia or hypertonia. No study reports a direct relationship between dysphagia and a greater predisposition to bruxism, so we cannot list dysphagia as a possible risk factor.

According to the data collected by this research, the majority of the study group, 75%, can feed by mouth, except that some children have solid and liquid feeding while others only liquid and pasty. On the other hand, 25% of the evaluated group uses a nasogastric tube, which is justified by the presence of a tracheostomy to assist in breathing. In the study by Oliveira et al. [19], about 20.7% of children with microcephaly associated with the Zika virus required an alternative feeding route, which in 75% of cases corresponded to a nasogastric tube. In the literature, no study reports a direct relationship between the feeding route and bruxism, so we cannot conclude a possible risk factor related to this.

In the present study, about 50% of the participants in the case group had gastroesophageal reflux, and several studies have shown a strong association between gastroesophageal reflux disorder (GERD) and sleep disturbances such as shorter sleep duration, difficulty falling asleep, and awakenings during sleep. Gastroesophageal reflux also appears to be a risk factor for sleep bruxism, and it has been hypothesized that this association has a role in stimulating the protective function of salivary flow [22]. Miyawaki et al. [23] found that subjects with SB assessed by sleep laboratory sessions had more episodes of GERD and that 60% of sleep bruxism (SB) episodes occurred during reflux, and that administration of proton pump inhibitor medication led to a reduction in SB episodes compared to placebo and controls [24].

Gastroesophageal reflux is frequent among children with neurological impairment [25,26]. Cerebral palsy and anticonvulsant medication are predisposing factors to gastroesophageal reflux [27]. Sakaguchi et al. [28] investigated the relationship between behavioral problems, sleep disorders such as bruxism and gastroesophageal reflux. Children with a mean age of 13.3 years (n=1840) were divided into two groups: normal behavior and behavioral problems. The Gastroesophageal Reflux Symptom Frequency Scale (GSSF) was used to assess reflux. For the analysis of nocturnal bruxism and other sleep-related disorders, parents were questioned about the presence of episodes. In this study, they concluded that there is a correlation between these variables influencing lifestyle and eating habits. The study showed similarity with the research, presenting reflux as a risk factor associated with bruxism in children with congenital Zika virus syndrome.

Simões-Zenari and Bitar [17] observed that children with oral habits such as sialorrhea during sleep, pacifier use, lip biting, and nail-biting are at increased risk of developing bruxism. Similar to children with congenital Zika virus syndrome. In addition to the psychological factor, an association has been found between this parafunction and systemic alterations such as gastroesophageal reflux, as well as in the study by Sakaguchi et al. [28] with reflux being a risk factor for nocturnal bruxism. This correlation is anchored in the increased rhythmic activity of the chewing muscles induced by esophageal acidification from gastroesophageal reflux [8]. This study reaffirms that reflux is a risk factor associated with bruxism in children with ZKV.

Neurological disorders, especially when associated with cognitive disorders like Down Syndrome, are considered risk factors for stereotyped movement disorders such as secondary bruxism [29]. In the study by Ortega et al. [16], parafunction was significantly higher in the first group when comparing the presence of dental clenching in children with cerebral palsy and those without this disorder [16]. The same research showed that the study group of children with ZKV had significantly higher bruxism than the regular children.

The main limitation of this study is the sample size, as a convenience sample was used based on the number of children affected by the Zika virus in the Fortaleza region. This may limit the generalization of the results to other regions or populations. Moreover, the awake and sleep bruxism diagnosis were made only by parent-report questionnaire, witch lids to the diagnosis of a possible awake and sleep bruxism, not definitely.

Conclusion

Children with congenital Zika virus syndrome had possible awake and sleep bruxism. Other results such as dysphagia, feeding routes, can feed by mouth and makes use of nasogastric tube and reflux were statistically significant as risk factors for bruxism. We also observed a prevalence of possible awake bruxism, with means in clenching (1.10), grinding (1.15), tensing the muscles (0.80), projecting the tongue between the teeth (1.25), keeping the jaw in a rigid position (0.75) and yawning (1.75) among children with the syndrome higher than regular children. The characteristics of congenital Zika virus syndrome may also influence these risk factors, especially the shape of the ogival palate, maxillary atresia, and microcephaly. All these factors contribute to the survival of these children, and multidisciplinary care and routine dental follow-up are of great value.

Authors' Contributions

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All authors declare that they contributed to critical review of intellectual content and approval of the final version to be published.

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Conflict of Interest

The authors declare no conflicts of interest.

Data Availability

The data used to support the findings of this study can be made available upon request to the corresponding author.

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