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Assessment of obstructive sleep apnea risk in different types of cleft lip and palate

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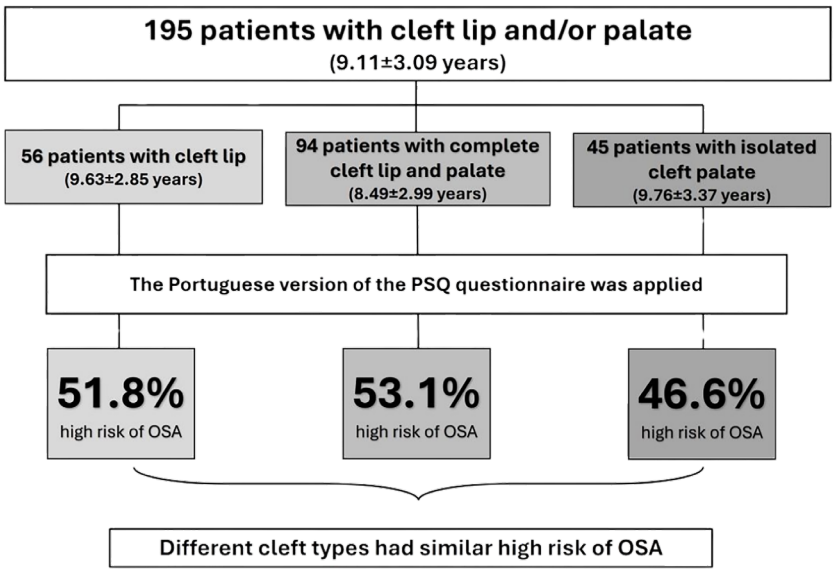
Abstract

Objective: This study aimed to assess the risk of obstructive sleep apnea (OSA) in different types of cleft using the Pediatric Sleep Questionnaire (PSQ). The influence of sex, age, body mass index (BMI), and history of adenotonsillectomy surgery were also evaluated. Methodology: Parents or legal guardians of 195 patients with nonsyndromic cleft lip, lip and palate, and isolated palate aged from 5 to 18 years (9.11±3.09 years) were invited to answer the PSQ. The sample included 94 individuals with unilateral or bilateral complete cleft lip and palate, 56 individuals with cleft lip, and 45 individuals with isolated cleft palate. The frequency of high and low risk was estimated for the complete sample. Multiple logistic regression analysis was used to assess the association between sex, age, cleft type, adenotonsillectomy surgery, and BMI with the occurrence of high-risk of OSA. Significance was considered at 5%. Results: A high risk of OSA was observed in 51.3% of the sample. The high risk of OSA was present in 51.8%, 53.1%, and 46.6% of subjects with cleft lip, cleft lip and palate, and isolated cleft palate, respectively. History of adenotonsillectomy surgery was a predictor of high risk of OSA (OR=6.94). The other variables were not associated with OSA high risk. Conclusion: Different cleft types had similar frequency of OSA high risk. Patients with history of adenotonsillectomy surgery still presented high risk of OSA.

Keywords: Cleft. Airway obstruction. Sleep apnea. Pediatrics.



*The prevalence of high risk of OSA in the total sample was **51.3%**

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Introduction

Sleep is considered essential for the proper development of children and proper health of adults and older adults.[1,2] Studies suggest that quality of life is directly related to an individual's sleep pattern.[1,2] Obstructive sleep apnea (OSA) is a sleep-disordered breathing (SDB) characterized by total (apnea) or partial (hypopnea) upper airway obstruction during sleep.[3] OSA can impact not only adults but also children and adolescents.[1,2] Common clinical signs are episodes of interrupted breathing during sleep, snoring, and nocturnal enuresis.[3] Pediatric OSA causes multiple awakenings and a reduction in oxygen saturation.[1,4] The consequences of untreated OSA in pediatric patients include growth delay, neurocognitive impairment including learning difficulties and failure to thrive, lower quality of life, and increased risk of associated morbidities including cardiopulmonary hypertension, increased blood pressure and risk of diabetes.[1,4,5]

Preliminary studies have shown an increased prevalence of OSA among subjects with cleft lip and palate (CLP) when compared to noncleft individuals.[6,7] CLP are congenital craniofacial anomalies that cause morphological and functional changes that can impact the development of the nasomaxillary complex, reducing the dimensions of the airway in the sagittal, vertical, and transverse directions.[8,9] These changes include nasal septal deviations, bone defects in the nasal floor, maxillary constriction, anteroposterior deficiency of the midface, hyperdivergent facial pattern, and narrowing of the nasopharynx and oropharynx.[8,10-12] Surgical procedures of the soft palate and oropharyngeal muscles further reduce airway dimensions predisposing patients with CLP to airway collapse during sleep.[8,10-13]

The Pediatric Sleep Questionnaire (PSQ), developed by Chervin, et al.[14] (2000), is a screening tool to pre-diagnose OSA in children aged 2 to 18 years. Although polysomnography (PSG) is the gold standard for diagnosing OSA, several barriers prevent its use for routine screening.[15] Previous works using the Pediatric Sleep Questionnaire (PSQ) have shown a prevalence risk of OSA ranging from 12% to 31% in children with syndromic and nonsyndromic cleft lip and/or palate.[6,7,12,16] The presence of associated craniofacial syndromes appears to significantly increase the risk of OSA.[17,18] In a polysomnographic study of pediatric patients with CLP, 72.7% of patients had a confirmed diagnosis of mild apnea and 27.3% had moderate/severe apnea after classified as high risk by the PSQ.[19] The mixed dentition period was reported as the phase of higher prevalence of OSA in patients with CLP.[20]

The increased prevalence of OSA in cleft children is well established.[6,7,12,16] However, there is a lack of studies investigating demographic, anatomic, and surgical factors related to OSA.[15] Data on outcomes about the diagnoses and treatment of OSA in this unique population are lacking.[15]

Therefore, this study aimed to assess the risk of OSA in patients with nonsyndromic cleft lip, cleft lip and palate, and isolated cleft palate and to evaluate the association between OSA high risk, sex, age, bone mass index (BMI), and adenotonsillectomy surgery

Methodology

This observational study was approved by the Ethics and Research Committee of Hospital for Rehabilitation of Craniofacial Anomalies, University of São Paulo (HRAC-USP) (Protocol number 26682619.0.3001.5417). Parents or legal guardians were invited to participate in the study and signed an informed consent form.

From 2020 to 2021, nonsyndromic patients with cleft lip, cleft lip and palate, and isolated cleft palate from the HRAC-USP were interviewed in the Dentistry and Otorhinolaryngology section, before the beginning of orthodontic treatment. Inclusion criteria consisted of patients aged 5 to 18 years, presence of cleft lip and/or palate, as well as absence of craniofacial anomalies, syndromes, and/or Robin's Sequence. Patients in orthodontic treatment were excluded. The patients and their companions were then invited to participate and signed an informed consent form. In the public craniofacial center evaluated, most patients are from the lower income group ($2.16 – $13 per day) based on World Development Indicators.[21] Therefore, a lower schooling level is expected.

The questionnaire was administered by a single orthodontist at the dental clinic. The validated Brazilian Portuguese version of the Pediatric Sleep Questionnaire (PSQ) was applied to parents and legal guardians at the dental section of the Hospital.[22] The questionnaire includes 22 questions related to the frequency and severity of snoring during sleep, apnea and breathing during the night, daytime

sleepiness, and neurocognitive problems. Answers to the questions were scored as yes (1), no (0), and do not know (-). The total score is estimated by dividing the number of "yes" answers by the total number of questions answered (yes + no answers). High risk of OSA was considered when 33% or more of the questions answered by parents or legal guardians were marked as "yes."[14] A percentage less than 33% indicated low risk for OSA.[14] Participants diagnosed as high risk of OSA were referred for evaluation with an otolaryngologist or sleep medicine specialist at the same center.

The final sample comprised 195 patients (109 male, 86 female) with a mean age of 9.1 years (SD=3.09). Data on the cleft type, sex, age (children: 5-11 years/ adolescents: 12-18 years[23]), and surgery history, including pharyngoplasty and adenotonsillectomy, were collected from the patients' records. The final subgroups consisted of 94 patients with unilateral or bilateral complete cleft lip and palate (67 male, 27 female; mean age 8.4 years, SD=2.99), 56 individuals with cleft lip/lip and alveolus (28 male, 28 female; mean age 9.63 years, SD=2.85), and 45 individuals with isolated cleft palate (14 male, 31 female; mean age 9.76 years, SD=3.37). Previous adenotonsillectomy has been performed in 15 out of 195 individuals. The pharyngoplasty factor was excluded considering only one patient from the sample had undergone this type of surgery. Sex, age, and BMI were compared to a control group of 116 no cleft subjects (56 male, 60 female; mean age of 9.79 years, SD=3.38) who had not been submitted to any type of orthodontic treatment. The control group was selected from the files of the Department of Orthodontics of Bauru Dental School, University of São Paulo.

Weight and height were obtained and used to estimate BMI, that were adjusted by age and sex, considering the BMI Calculator for Child and Tenn of Center for Disease Control and Prevention (https://www.cdc.gov/healthyweight/bmi/calculator.html).[24] The sample was classified into four weight-groups: underweight (BMI <5th percentile), normal weight (BMI ≥5th percentile and <85th percentile), overweight (BMI ≥85th percentile and <95th percentile), and obese (≥95th percentile)[24] (Table 1).

Statistical analysis

Statistical data was collected to analyze the frequency of participants with high and low risk of OSA. Intergroup comparisons of sex distribution, age, cleft type, BMI, and Adenotonsillectomy surgery were performed using chi-square test. Sex, age and BMI were also compared to a control group with chi-square test. Multiple logistic regression was conducted to analyze the risk of OSA adjusted associations and interactions between the variables sex, age, cleft type, BMI, and adenotonsillectomy surgery. Variables with significant correlations were included in the final analysis of the multiple logistic regression model using stepwise logistic regression (sex, adenotonsillectomy surgery, and BMI).

Cronbach's alpha test was used to assess the internal consistency of the full scale of the questionnaire, estimated for the total sample of the questionnaire and for the three subscales (snoring, sleepiness, and behavior).

All statistical procedures were performed in the SPSS program version 28 and Jamovi software (version 2.2.5). A 5% significance level was considered.

Results

The risk of OSA (p=0.066), sex (p=0.193), age (p=0.136), and BMI (p=0.133) in subjects with CLP did not show a statistically significant difference compared to the control group (Table 1).

High risk of OSA was observed in 51.3% (100/195) of the total sample (Table 2). Table 2 shows the frequency of patients with high and low risk of OSA according to cleft type, sex, age, BMI, and adenotonsillectomy surgery. The high risk of OSA was present in 51.8%, 53.1%, and 46.6% of subjects with cleft lip, cleft lip and palate, and isolated cleft palate, respectively (Table 2).

The multiple regression final model was significant (p<0.001) and showed that approximately 6.07% of the risk of OSA could be explained by adenotonsillectomy surgery. (Table 3). The variables included in the logistic regression model have not shown collinearity. (Table 3).

Among patients at high risk, the questions that received most "yes" answers were question C3: "Does your child easily distract by extraneous stimuli?" by 67.1% and question A8: "Does your child have a dry mouth on waking up in the morning?" by 64.1% (Table 4). The questions with the least "yes" answers included question B6: "Did your child stop growing at a normal rate at any time since birth?" and question B5: "Does your child wake up with headaches in the morning?",

Table 1- Comparison of sex, age, and BMI with a control group (Chi-square test)

Baseline Characteristics		CLP Group		Control Group		P
		High (n, %) (n=100)	Low (n, %) (n=95)	High (n,%) (n=47)	Low (n, %) (n=69)	0.066
Sex	Male	61 (55.9%)	48 (44.1%)	21 (44.6%)	35 (50.7%)	0.193
	Female	39 (45.3%)	47 (54.7%)	26 (55.4%)	34 (49.3%)	
Age	Children	76 (50.3%)	75 (49.7%)	34 (72.3%)	47 (68.1%)	0.136
	Adolescent	24 (54.5%)	20 (45.5%)	13 (27.7%)	22 (31.9%)	
BMI	Underweight	13 (56.5%)	10 (43.5%)	3 (6.4%)	6 (8.7%)	0.133
	Normal Weight	39 (41.4%)	55 (58.6%)	18 (38.3%)	27 (39.1%)	
	Overweight	15 (57.6%)	11 (42.4%)	7 (14.9%)	15 (21.7%)	
	Obese	33 (63.4%)	19 (36.6%)	19 (40.4%)	21 (30.5%)	

BMI: Bone Mass Index
*Statistically significant at P<0.05.

Table 2- Frequency of patients with high and low risk of OSA according to sex, age, cleft type, BMI, and surgery history (chi-square test).

Baseline Characteristics		Risk of OSA		Age (y)	P
		High (n, %) (n=100)	Low (n, %) (n=95)	Mean (SD)	
Sex	Male (n=109)	61 (55.9%)	48 (44.1%)	8.9 (2.66)	0.141
	Female (n=86)	39 (45.3%)	47 (54.7%)	10.1 (3.57)	
Age	Children (n=151)	76 (50.3%)	75 (49.7%)	8.01 (1.91)	0.623
	Adolescent (n=44)	24 (54.5%)	20 (45.5%)	13.7 (1.83)	
Cleft Type	Lip (n=56)	29 (51.8%)	27 (48.2%)	9.4 (2.57)	0.769
	Lip and Palate (n=94)	50 (53.1%)	44 (46.9%)	8.8 (3.13)	
	Isolated Palate (n=45)	21 (46.6%)	24 (53.4%)	10.7 (3.35)	
BMI	Underweight (n=23)	13 (56.5%)	10 (43.5%)	8.5 (3.38)	0.061
	Normal Weight (n=94)	39 (41.4%)	55 (58.6%)	9.7 (3.50)	
	Overweight (n=26)	15 (57.6%)	11 (42.4%)	9 (2.93)	
	Obese (n=52)	33 (63.4%)	19 (36.6%)	9.4 (2.53)	
Adenotonsillectomy Surgery	Yes (n=15)	13 (86.6%)	2 (14.4%)	10.4 (2.93)	0.004*
	No (n=180)	87 (48.3%)	93 (51.7%)	9.2 (3.10)	

SD: Standard Deviation; OSA: Obstructive Sleep Apnea; BMI: Body Mass Index.
*Statistically significant at *P*<0.05.

Table 3- Results of the multiple logistic regression analysis to determine significant variables affecting risk of OSA.

		Initial Model				Final Model			
Model fit measures		p	R^2N	X^2		p	R^2N	X^2	
		0.003	0.118	18.0		0.003	0.0607	9.09	
Predictor		p	Odds Ratio	95% Confidence Interval		p	Odds Ratio	95% Confidence Interval	
				Lower	Upper			Lower	Upper
Sex		0.258	0.704	0.3838	1.292	-	-	-	-
Adenotonsillectomy Surgery		0.011	0.135	0.0289	0.629	0.012*	6.948	15.240	31.678
BMI	2-1	0.252	1.747	0.673	4.534	-	-	-	-
	3-1	0.651	0.765	0.239	2.444	-	-	-	-
	4-1	0.614	0.765	0.269	2.169	-	-	-	-

*Statistically significant at P less than 0.05; BMI: Body Mass Index

Table 4- Questions that received the most "Yes" answers among high-risk patients.

Question	Yes (n=195)	
	n	%
A1. Snore more than half the time?	57	29.2
A2. Always snore?	45	23.0
A3. Snore loudly?	45	23.0
A4. Have "heavy" or loud breathing?	67	34.3
A5. Have trouble breathing or struggle to breathe?	81	41.5
A6. Have you ever seen your child stop breathing during the night?	25	12.8
A7. Tend to breathe through the mouth during the day?	122	62.5
A8. Have a dry mouth on waking up in the morning?	125	64.1
A9. Occasionally wet the bed?	21	10.7
B1. Wake up feeling unrefreshed in the morning?	43	22.0
B2. Have a problem with sleepiness during the day?	40	20.5
B3. Has a teacher commented that your child appears sleepy during the day?	26	13.3
B4. Is it hard to wake your child up in the morning?	73	37.4
B5. Does your child wake up with headaches in the morning?	19	9.7
B6. Did your child stop growing at a normal rate at any time since birth?	12	6.1
B7. Is your child overweight?	27	13.8
C1. Does not seem to listen when spoken to directly.	86	44.1
C2. Has difficulty organizing task and activities?	88	45.1
C3. Is easily distracted by extraneous stimuli?	131	67.1
C4. Fidgets with hands or feet or squirms in seat?	108	55.3
C5. Is 'on the go' or often acts as if 'driven by a motor'?	124	63.5
C6. Interrupts or intrudes on others (e.g. butts into conversations or games)?	112	57.4

with 6.1% and 9.7%, respectively. (Table 4).

The total internal consistency was 0.73 (substantial). For the subscales of snoring, sleepiness, and behavior, the internal consistency of the questionnaire was 0.68 (substantial), 0.48 (moderate), and 0.64 (substantial), respectively. [25]

Discussion

The Pediatric Sleep Questionnaire provides clinical and research reliability for identifying the risk of OSA.[14,26] Although polysomnography (PSG) is the gold standard for diagnosing apnea, the PSQ offers lower financial cost, greater accessibility, and adequate accuracy in predicting apnea in children with a sensitivity of 85% and a specificity of 87% when compared to PSG.[14,26] The orthodontist plays a crucial role in identifying patients at high risk of OSA, referring those at risk for definitive diagnoses with PSG and contributing to timely multidisciplinary treatment.[27] The value of the internal consistency of the sample was 0.73 (substantial) for the present study.[25] This finding aligns with previous studies using the Brazilian Portuguese version of PSQ, indicating a consistency ranging from substantial to nearly perfect, with values from 0.78 to 0.86.[26,28] The internal consistency of the snoring, sleepiness, and behavior subscales was classified as substantial, moderate, and substantial, respectively.[25] The potential for misdiagnosis between primary snoring and more severe conditions may arise when children do not exhibit pauses or gasps during snoring, and obstructions often occur in the early morning hours when parents are less likely to observe their child's breathing patterns.[28]

In this study, 51.3% of the sample demonstrated high risk of apnea. This result agrees with the higher prevalence of OSA in the CLP population when compared to non-cleft population.[6,7,12,16] The higher risk of OSA in subjects with CLP may be explained by significant and frequent midfacial hypoplasia, airway dysfunction from abnormal muscular attachments, presence of scar tissue and low mobility of the soft palate, irregular morphology of the nasal cavity, airway hypotonia from sensory feedback issues, and central respiratory control alterations.[6,12,29,30] Maclean, et al.[6] (2009) reported a higher risk of OSA (31.4%)

in children. However, only one-quarter of the sample consisted of children aged four to five years.[6] In the present study, three-quarters were younger than four years old, which might explain the increased OSA risk.

Compared to previous studies in oral clefts, a higher prevalence of OSA risk was found in this study. Moreover, no difference was found between the cleft and control groups regarding the risk of OSA. These differences can be explained by the fact that our sample was composed of patients evaluated before the start of the orthodontic/facial orthopedic interventions. Midface hypoplasia is often associated with cleft due to the anatomic structure and primary repair surgeries.[18,31] Maxillary hypoplasia shifts the hard palate backward, pushing the soft palate tissue posteriorly, which reduces the airway and can increase the risk of OSA.[15,18] Another possible explanation for the higher OSA risk in our sample compared to previous studies is the reduced socioeconomic and cultural status of our sample. Most patients attending the hospital have a low to middle socioeconomic status. The full comprehension of the questions in the PSQ might have been hindered by the socioeconomic status. Low schooling level may lead to greater difficulty in understanding the questions, which can contribute to the false-positive diagnosis of high risk of OSA using PSQ. A previous study has also reported an association between lower socioeconomic status and worse quality of life, which influences, consequently, the prevalence of OSA.[32] Therefore, in this study, the PSQ was completed with a professional present to clarify doubts, ensuring accurate answers.

In the present study, the prevalence of high risk of OSA was found similar across various types of cleft. A significant relationship was not found between the high risk of OSA and the cleft type in the multiple regression analysis. These results are in accordance with previous studies[6,7,15] and highlight the importance of routine screening for all children with cleft, including those with isolated cleft lip.[15] Fisher, et al.[15] (2023) found that parental concerns about observed sleep were accurate 67% of the time in identifying OSA, thus the screening questionnaire does not replace the importance of parental observation. This finding emphasized the multifactorial etiology of OSA in CLP and the limitation of relying exclusively on screening questionnaires for OSA diagnosis.

The age was not statistically related to the high risk of OSA in the multiple regression analyses. In a recent study in individuals with cleft lip and palate, no difference was observed in the risk of OSA in individuals younger and older than 10 years of age.[16] Although the peak age of OSA in noncleft individuals corresponds to the peak of adenotonsillar hypertrophy, in individuals with CLP the relative contribution for this factor is unknown.[6] The present study found no difference between females and males for the risk of OSA (p=0.258), agreeing with previous studies conducted in children with CLP.[16,33] Although males have a higher risk compared to females in the general adult population,[34] in children and adolescents with cleft lip and palate the sex seems not be a relevant factor.

Approximately half of the sample with high risk of OSA were overweight or obese (48%), while 35% of patients with low risk of OSA were overweight or obese. However, the regression analysis showed that BMI was not a predictor of higher OSA risk in the sample. This result corroborates a previous study showing no association between an elevated BMI and positive sleep scores.[15] A possible explanation is that OSA in CLP is complex and multifactorial and other factors, such as skeletal deficiencies and irregular morphology of soft and skeletal tissues, play a more important role than only BMI.[6,30]

The adenotonsillectomy surgery was the only factor significantly related to the high risk of OSA. These results showed that most patients who had previous surgery for removal of pharyngeal and palatine tonsils was still demonstrating a high risk of OSA. However, only 7.7% (15/195) of our sample had undergone adenotonsillectomy surgery. A possible explanation is that the adenotonsillectomy surgery may not be enough to solve OSA in all patients with cleft lip and palate.[26,35,36] Tauman, et al. demonstrated that apnea-hypopnea index normalized completely in only 25% of the individuals analyzed before and after adenotonsillectomy surgery.[36] Guilleminault, et al.[35] (2007) have also observed persistent abnormality indices in PSG after adenotonsillectomy surgery. Surgical intervention may not solve others anatomic compromises and residual abnormal respiratory patterns that might be present in children with OSA.[35] In addition, patients with CLP commonly have maxillomandibular retrusion and increased vertical facial height and still show a reduced upper airway volume after the adenotonsillectomy surgery, contributing for the maintenance of high risk of OSA.[10,37] In a study conducted by Muntz, et al.[38] (2008), adenotonsillectomy was the initial

intervention for most children with oral clefts and OSA. While there was a significant overall improvement in sleep, many children continued to experience sleep apnea.[38] Pediatric OSA involves a range of conditions caused by neuromotor dysfunction, lymphoid tissue hypertrophy, and craniofacial skeletal dysmorphology.[38] Adenotonsillectomy is the standard treatment for most healthy children and shows positive outcomes; however, continuous positive airway pressure, medications, orthodontic treatment, and craniofacial distractions should be considered for refractory patients and patients with multilevel obstruction in the setting of craniofacial anomalies.[38,39] These results showed the importance of monitoring patients after adenotonsillectomy. Additional therapeutic approaches might be needed for patients with CLP and persistent OSA symptoms after adenotonsillectomy.

The results of our study should be interpreted with caution. Previous studies have suggested that the sensitivity and specificity of the PSQ are low in children with complex disorders, including craniofacial anomalies, indicating that the PSQ may not be an effective screening tool for OSA in this population.[40,41] A limitation of our study was the lack of PSG data for patients who were referred for further evaluation. A comparative analysis of PSQ and PSG data could be valuable to assess the accuracy and utility of the PSQ as a screening tool in this context. Additionally, these comparative data could aid address issues related to literacy and comprehension of the PSQ across different populations, enhancing the screening process. Future studies with comparative data are recommended.

Conclusion

The prevalence of high risk of OSA among patients with cleft lip and/or palate was 51.3% and different cleft types had similar risk. Patients with cleft lip and/or palate with history of adenotonsillectomy surgery still presented high risk of OSA and need continued monitoring, as well as other possible additional therapeutic approaches. Further research is needed to develop improved screening tools for subjects with oral clefts.

Conflict of interest

The authors declare no conflict of interest.

Data availability

All data generated or analyzed during this study are included in this published article

Authors´ contributions

Jost, Patrícia: Conceptualization (Equal); Data Curation (Equal); Investigation (Equal); Methodology (Equal); Project Administration (Equal); Writing - original draft (Equal). **Utrago, Gabriela:** Writing - original draft (Equal); Writing - review & editing (Equal). **Miranda, Felicia:** Writing - review & editing (Equal). Lauris, **Rita de Cássia Moura Carvalho:** Investigation (Equal); Methodology (Equal); Visualization (Equal). **Poiani, João:** Writing - original draft (Equal); Writing - review & editing (Equal). **Garib, Daniela Gamba:** Conceptualization (Equal); Formal Analysis (Equal), Methodology (Equal); Supervision (Equal); Validation (Equal); Writing - original draft (Equal); Writing - review & editing (Equal).

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