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Editor-in-Chief
Burak BULDUR

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Contemporary Pediatric Dentistry

Editorial
Contemp Pediatr Dent 2021:2(3):134-134
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Hi Dear Friends and Colleagues,

I am pleased to inform you some of good news regarding the development and process of the Contemporary Pediatric Dentistry (CPD). Since its establishment, CPD has received 80 submissions from all over world within sixteen months, and published 25 articles including review, original research and case reports. CPD has also moved to be indexed in reputed databases.

Another achievement was establishment of 1st International Congress of Contemporary Pediatric Dentistry (ICCPD), organized by the CPD, which was taken place on 1- 3 November 2021 as a fully virtual meeting to celebrate the first anniversary of the launch of CPD. The ICCPD hosted 257 registered colleagues from 47 countries. The website of the ICCPD had been viewed more than 5000 visitors from 102 countries in only 2 months.

As an exciting action, CPD is now getting ready to initiate the 2nd International Congress of Contemporary Pediatric Dentistry (www.iccpd.org). I feel confident that the ICCPD 2022 will serve as a place of learning where we can all share the latest knowledge and learn the contemporary view, approaches, techniques and treatment options from highly eminent clinicians and/or academicians from different countries. The theme of the ICCPD 2022 is "*Contemporary Clinical Techniques in Pediatric Dentistry*". The 2nd ICCPD will also host oral and poster presentation. All abstracts will be published in the Supplementary Issue of Contemporary Pediatric Dentistry.

It is my honor to invite all colleagues, who have sincere enthusiasm to help children to let them have better oral health, quality of life, smiling and development, to attend 2nd International Congress of Contemporary Pediatric Dentistry to be held on 2-4 September 2022 as a fully virtual meeting.

Please follow website and social media accounts of CPD to get latest information regarding Congress and the Journal.

Thanks and Best regards,

Burak Buldur
Editor-in-Chief, CPD
Congress President, ICCPD

Contemporary Pediatric Dentistry

Review
Contemp Pediatr Dent 2021:2(3):135-141
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Does toxic stress impact paediatric dental procedures? An integrative review

Victor Cavallaro Bottesini[1] ✉, Gabriel Florio Cairo[2], Gabriel Marcelino[3], Danilo Antonio Duarte[3]

Highlights

- Invasive paediatric dental procedures can produce a stressful situation in young children.
- The topic of toxic stress is relevant and should be included in paediatric dentistry research and clinical trials.
- Paediatric dentistry needs to recognize and diagnose toxic stress as an aggravating factor in child health.

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Abstract

Toxic stress is defined as strong, repetitive stress with prolonged activation, without the child having the support of an adult caregiver. It is plausible that invasive and complex paediatric interventions produce or contribute to the development of toxic stress. This article aims to evaluate, discuss and synthesize the association between toxic stress and paediatric dental clinical practice through an integrative review of studies published in several databases. The results of this study allow us to state that this subject is little explored in the dental literature and, therefore, that it is essential to encourage clinical studies and specific research. The expansion of these studies will provide health professionals with scientific evidence regarding the implementation of proper clinical practice, thus reducing or even eliminating the development of toxic stress.

Keywords: Adverse Childhood Experiences; Anxiety; Child; Pediatric Dentistry

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INTRODUCTION

Stress is a relevant condition in the overall development of the human body and influences the tolerability of future adverse situations, the acquisition of habits and behaviours, and the learning and development of brain architecture and neural systems.[1,2]

Stress is classified into three levels based on the biological responses of the body and is dependent on the duration and intensity that the individual is exposed to the stressor factor. Thus, stress can be positive, tolerable, or toxic.[3,4]

Positive stress occurs when an experience is brief and mild, and the affected individual can return to normal standards in an acceptable time. Stress is characterized as tolerable when an individual experiences stressors for a sustainable period of time that are capable of increasing the risk of impaired brain architecture. Toxic stress is defined as intense and continuous stress that can cause irreversible damage to the neuropsychomotor development of an individual, increasing the risk of organic diseases throughout the life cycle.[5,6]

Given that brain development occurs in early childhood, with the consequent construction of habits and development of learning to address adverse or unfavourable situations, strong scientific evidence has shown that persistent, toxic stress in early childhood produces changes in the body biology and behaviour of children.[1,2,6] All these changes affect the overall health of children, causing immune system impairments, increases in inflammatory markers, and impacts on oral health.[7–11] In addition, previous studies[12–14] reported that family factors, such as maternal or paternal stress, can lead to the development of parafunctional oral habits such as finger sucking and bruxism, caries lesions and other oral disorders. Scientific evidence[5,7,11,12,13,14,15] has indicated that children with previous potentially stressful experiences in paediatric dental procedures, manifested uncooperative behavior in relation to those who didn't suffer negative experiences. Furthermore, is reasonable to infer a strong association between a child's young age, negative dental experiences, toxic stress, and uncooperative behavior.[16-19] Thus, it is necessary to identify and discuss the impact that toxic stress has on the oral health of and dental procedures in children.

METHODS

Study design

This study is an integrative review, which consists of locating, interpreting and synthesizing the volume of scientific evidence produced on a subject, which in this case is the relationship between toxic stress and paediatric dentistry procedures. For this, the following steps were undertaken: identification of the theme and design of the guiding question: "Which is the toxic stress influence in paediatric dentistry?", establishment of inclusion and exclusion criteria for the articles, gathering of data from the articles, analysis and interpretation of the results and synthesis of the content. This study was based on steps proposed by Hermont et al.[21]

Search strategies

The literature review was performed by three reviewers, based on active searches for information contained in the following databases: PubMed, SciELO, ProQuest and Lilacs (Bireme's databases). The selection of articles was based on health descriptors (Decs.), combined with the Boolean operators "and" and "or", based on the relevance of the articles. The descriptors "Childhood stress", Toxic stress", Resilience", "Developmental origins", "Family paediatrics" and "Psychological adaptation" combined with "Pediatric dentistry" and with the Boolean operators "and" and "or" were used for the searches in the databases used.

Search filters were selected to explore more relevant and recent studies (01/2011 to 01/2021) and exclude bibliographic productions represented by books, monographs and theses. Therefore, the remaining studies were clinical trials, meta-analysis, randomized controlled trials, and systematic reviews.

Eligibility criteria of the articles

The articles selected for the study were evaluated based on their relevance to the guiding question

of this study. Articles were selected by reading the titles, abstracts and eventually the full text of articles that met the inclusion and exclusion criteria. The inclusion criteria were the study correlation between toxic stress and paediatric dentistry. While the exclusion criteria were the opposite of this, when the study approached only

one of them or when it didn't do a correlation between them. This process was performed by the researchers in a critical and independent manner. Disagreements between the researchers were resolved by consensus and ultimately by the research supervisor, with the goal of reducing bias.

Selection of publications and data extraction

The remaining articles were organized in a spreadsheet on Google Sheets, the titles were analysed, and duplicates were excluded. Next, articles whose titles did not fit the study were excluded; and finally, those that did not fit the study objective were excluded. After this filtering process, the articles were read and judged based on their information presented and topics covered.

RESULTS

The active search in the various databases retrieved 128 articles. As the articles came from different databases, filtering and exclusion of duplicates was performed – 11 studies – resulting in 117 studies. After this, the articles relevant to the study were selected based on the titles, among which 30 articles were selected. Subsequently, the abstracts of those studies were analysed individually, and among the 30 studies, 9 were designated for full reading. All the articles elected for the research addressed the early childhood population. Among the articles cited for full reading, 2 studies fully covered the focus of the research, which is toxic stress and child dental care (Flowchart 1). All articles found were published in English (Table 1).

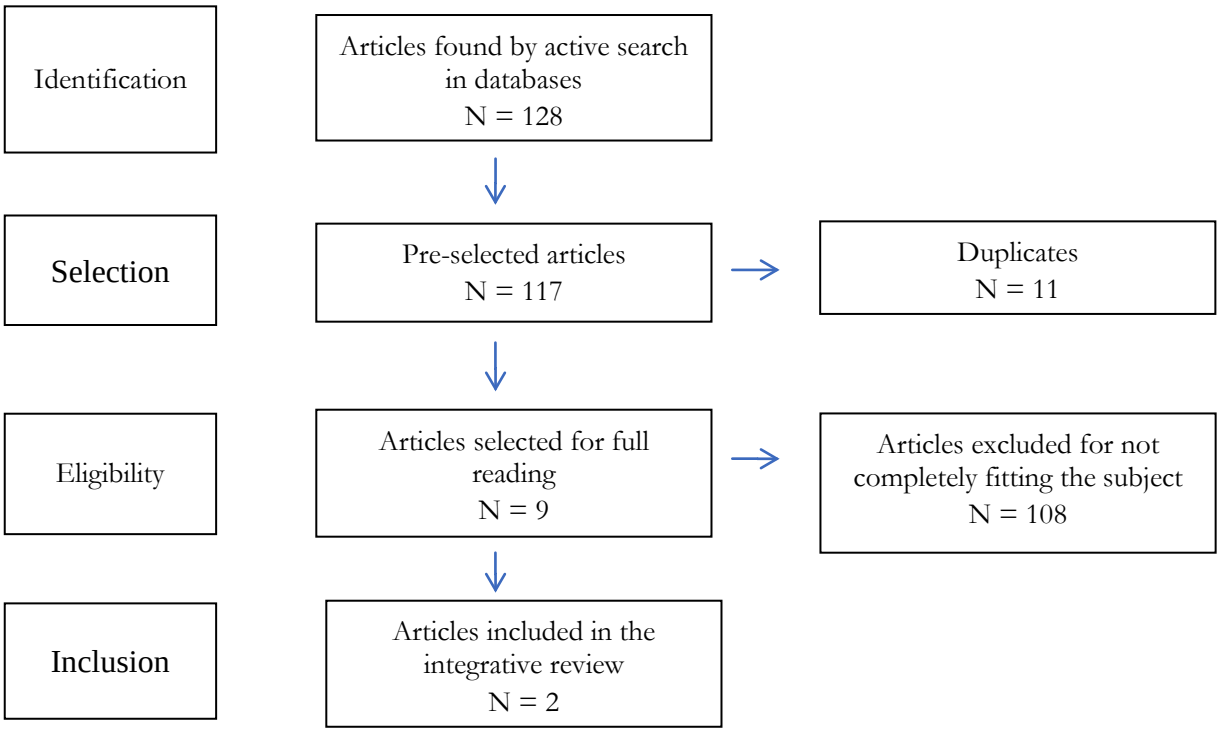

Figure 1. Flowchart of selection and filtering of scientific articles

Table 1. Selected articles

Author	Title	Main subject studied	Journal	Language	Month/year
Bright et al.	Adverse childhood experiences and dental health in children and adolescents	Association between social stressors and oral health	Community Dentistry and Oral Epidemiology	English	June/2015
Long	Stress and economic hardship: the impact on children and parents	How economic difficulties, parent depression and other stressors influence general and oral health throughout life and the implications for paediatric dentistry	Paediatric dentistry	English	April/2014

Considering the number of articles initially identified and especially those submitted to all selection steps, it is reasonable to conclude that there is a small number of publications and production of knowledge on this subject. Thus, there is a need for research on this subject to expand the literature and share knowledge about toxic stress and its impact on children undergoing paediatric dentistry procedures.

DISCUSSION

Contemporary paediatric care is not restricted to the contexts of the diagnosis and treatment of oral pathologies, and the need to know all the environments in which children live and develop, such as family, school and society, is necessary.[5,21]

The interpretation of a child's growth and development is extremely important in understanding the expectation of a healthy life, from early childhood to adulthood. Scientific evidence[3,17] has shown that the infant brain begins developing during the foetal period and that the various brain zones expand and modulate with extreme ease until the end of early childhood, hence the importance of experiences during this period, including dental procedures, as this resilience makes the brain sensitive to chemical changes. Thus, neural damage caused in this life cycle, although not physically visible, leads to permanent changes in brain structure and function.[3,15]

These hypertrophic changes generate hyperactivity in these areas, consequently stimulating the degree of anxiety in the child. The cerebral amygdala is an important physiological factor involved in the stress response; due to the cascade effect of stress, in the amygdala, there is an increase in the concentration of cortisol, which is a biomarker of responses to stressful experiences.[22,23]

In paediatric dentistry, it is recognized that invasive and complex clinical interventions may cause situations of stress. In addition, invasive procedures can cause pain, whose presence increases cortisol levels. We emphasize that in the first years of a child's life, levels of anxiety are naturally higher, given that cognitive and emotional development is immature at these ages in relation to that in older children.[1,2]

For all these reasons, it is essential for paediatric dentists to master and manage the care of children regarding pain control, differentiating the types of crying, behavioural control techniques and time needed for procedures.[13,15]

Pain can be controlled with appropriate anaesthetic techniques. Crying can be the result of pain or fear, and behavioural assessment methods can be used to make such differentiations.[16] Another important parameter is the time children spend undergoing procedures; importantly, the time required for the cortisol level to peak is twenty to thirty minutes after a stress stimulus.

Thus, the evolution of patient behaviour combined with a good clinical sense will indicate the continuity or suspension (when possible) of a clinical intervention.

Evidently, children subjected to toxic stress become more resistant to paediatric dental procedures, given the high degree of anxiety and fear. To facilitate the management of these patients, it is advisable to measure the degree of anxiety using a behavioural assessment method.[16-18] One of the established techniques for controlling anxiety is "talking, showing and doing", which consists of introducing children to elements of the dental office, verbally explaining the procedures to be performed and demonstrating the procedures through visual, auditory and tactile ways.[13] The patient modelling technique is another alternative and consists of an introductory consultation to become familiar with the office, followed by a consultation for a preventive procedure and ending with the most complex procedures. Studies indicate that this pattern of care tends to reduce stress and anxiety.[17,18,24]

We emphasize the importance of paediatric dentists understanding the characteristics of the family nucleus as part of the care of children, given that families can have a supportive function or can aggravate toxic stress conditions. In addition, studies indicate that children who belong to minority groups or ethnic groups and who suffer any type of racism, lack educational opportunities and experience residential segregation experience an exacerbation of this stress process.[15,25] Thus, paediatric dentists must recognize, protect and even neutralize the negative impacts resulting from oral diseases on children, especially those of a young age, who require continued treatment that poses a potential to cause toxic stress.[3,15]

Knowledge of these theories and clinical criteria will allow paediatric dentists to work at positive and tolerable stress levels by eliminating the toxic stress induced by clinical procedures, reducing the emotional cost, and neutralizing the negative impacts on the development and health of children.

CONCLUSIONS

Toxic stress, which is widely described and discussed in paediatric medicine, has not been given the same prominence in the dental literature. However, the literature on dental specialties, notably paediatric dentistry, indicate high degrees of anxiety and fear that impact and limit clinical procedures.

It is reasonable to infer that such facts are consequences of underdiagnosed toxic stress in child patients, possibly due to dental professionals misunderstanding the subject. Thus, it is important to produce research and clinical studies on toxic stress and its impact on paediatric dentistry to guide dentists in identifying psychosocial stressors, such as low family income, low education, family structure, racism, neglect and maltreatment, among others, which should be considered in the health of children.

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Contemporary Pediatric Dentistry

Original Research
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Reliability and validity of the Turkish version of the Abeer Children Dental Anxiety Scale (ACDAS)

Aylin Aslan[1] ✉, Tamer Tüzüner[2], Özgül Baygın[3], Nagehan Yılmaz[4], Serpil Sagdıc[5]

Highlights

- Dental anxiety is a common problem that can lead to adverse conditions in dentists and patients.
- Addressing childhood dental anxiety is a critical step in improving children's oral health and dental experience. It is important to identify the anxious child as early as possible.
- The Turkish version of the Abeer Children Dental Anxiety Scale (ACDAS) could be considered as a valid and reliable dental anxiety scale that helps us to identify and treat anxious children.

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Abstract

Aim: This study aimed to assess the validity and reliability of the Turkish version of the Abeer Children Dental Anxiety Scale (ACDAS) as a tool used for the identification and treatment of anxious children. **Methods:** The study sample included 122 children aged ≥7 years, and the intervention consisted of implementation of the ACDAS and the Children's Fear Survey Schedule-Dental Subscale (CFSS-DS) by the lead researcher during the first dental visit for each child. ACDAS application was also repeated by another pediatric dentist in a smaller sub-sample of 20 children during the same visit. The lead researcher repeated the process, (i.e. application of ACDAS) in the whole study sample after two weeks. **Results:** The mean age of the participants was 9.84 ± 1.696 years (range: 7–12 years), and anxiety (ACDAS ≥ 26) was detected in 36.1% of the children. The Turkish version of the ACDAS exhibited high correlation with the CFSS-DS ($r = 0.760$; $p < 0.001$), and the Cronbach Alpha value ($\alpha = 0.934$) showed good internal consistency. No statistically significant differences were observed in the dentist's responses to questions in the Dental and Cognitive sections when comparing the first and second applications of the scale ($p > 0.05$), although significant differences were observed in the Child Evaluation section ($p < 0.01$). The Area Under the Curve (AUC), obtained by Receiver Operative Characteristic Curve (ROC) Analysis was 0.849 indicating good diagnostic performance. **Conclusions:** Based on these findings, the Turkish version of the ACDAS can be considered as a valid and reliable dental anxiety scale for the identification of children concerned about dental procedures.

Keywords: Dental Anxiety; Dental Care; Pediatric Dentistry

INTRODUCTION

Although dental anxiety, a very common condition that may cause various problems for dentists and patients,[1,2] can appear at any age, it is typically observed in childhood or adolescence[2] as a result of social factors such as the individual's personality traits, socioeconomic status, family history of dental anxiety, parental expectations regarding their child's behavior in a dental settings, and various factors associated with the dental environment itself.[3] Childhood dental anxiety may also be carried into adulthood, resulting in the individual avoiding any form of dental treatment. Therefore, identification of an anxious child and management of their dental anxiety at an early stage[4] plays a critical role in the improvement of an individual's oral health and dental experience.[5] In order to achieve this, evaluation of dental anxiety must be child-oriented, reliable, valid, and practical,[6] and anxiety measurement techniques include scoring the child's behavior during dental visits, projective techniques, physiological measurements, and psychometric scales[7-11] such as the Children's Fear Survey Schedule-Dental Subscale (CFSS-DS) which assigns scores based on the level of fear exhibited by the child in response to dental-related conditions or treatments (e.g., "dentists," "injections," and "examining someone's mouth").[12,13] Although the CFSS-DS has been shown to have high reliability and validity in previous studies,[12,13] it does not evaluate the physical signs, thoughts, and behaviors of the child which may contribute to a better understanding of their dental anxiety.[2] The Abeer Children Dental Anxiety Scale (ACDAS) includes cognitive questions and is a valid scale for the measurement of dental anxiety in children over 6 years of age.[14] Therefore, the aim of this study was to investigate the validity and reliability of the Turkish adaptation of the ACDAS.

METHODS

Ethical approval

Ethical approval for this study was obtained from the Directorate of Scientific Research Ethics Committee of Karadeniz Technical University, Faculty of Medicine (No. 2019/18; 11/02/2019).

Sample size

The study was conducted at the Children's Dentistry Clinic of Karadeniz Technical University, Faculty of Medicine. The power calculation, based on a previous study done by Temel G and Erdogan S[15], yielded a sample size of 116. Based on this, this study aimed to include 140 children in order to allow up to 20% loss to follow-up. The inclusion criteria were as follows: age ≥ 7 years; absence of any systemic, mental and/or physical disorders; no history of medications for any psychiatric reasons; absence of any learning and understanding disabilities; and fluency in reading, speaking and understanding Turkish.

Translation of the scale into Turkish was carried out in three stages, and the language validity was tested using the translation/retranslation method. Two pediatric dentists who were proficient in English translated the scale into Turkish independently, following which the original scales and translations were evaluated by two different pediatric dentists to allow selection of the most appropriate one. At the final stage, a dentist who was blinded to the original scale translated the Turkish text into English, compared it with the original text, and translated the nonconforming items back into Turkish to obtain the final scale (Figure 1).

The ACDAS was applied to 20 participants by the lead researcher as well as another pediatric dentist in the same visit to allow measurement of interobserver reliability. ACDAS was further

administered by the lead researcher in all participants (excluding 20 children) after an interval of two weeks to measure other reliability parameters (internal consistency and test-retest). Additionally, the Children's Fear Schedule Scale-Dental Subscale (CFSS-DS) was also applied to the same children to assess the validity of ACDAS in the first visit.

Data collection tools

Abeer Children Dental Anxiety Scale

The ACDAS is a cognitive scale consisting of 19 questions and three sections, as follows: dental, cognitive, and child evaluation. In the dental section, consisting of 13 questions, the children were asked to show how they felt in response to the question by selecting the appropriate option out of three facial expressions provided. The responses were scored on a scale of 1–3, with the total scores ranging between 13 and 39 and a score of 26 and above indicating anxiety. The Cognitive and Child Evaluation sections of the scale were completed by the child's legal guardian and the dentist.[14]

Child Fear Survey Schedule-Dental Subscale (CFSS-DS)

The Turkish version[16] of the CFSS-DS consisted of 15 questions addressing the various stages of clinical dental treatments.

The children were asked to score their response to the question using a scale ranging from 1 to 5 (1: I am not scared at all; 2: I am scared very little; 3: I am a little scared; 4: I am very scared; 5: I am extremely scared). The total score ranged between 15 and 75, and a score of 36 and above was considered indicative of anxiety.

Statistical analysis

All statistical analyses were carried out using the SPSS 17.0 Statistical Package Program (Chicago: SPSS Inc.). Cohen's Kappa test was used to determine interobserver agreement, and Spearman's Correlation and ROC analyses were used to determine the validity of the ACDAS and CFSS-DS scales. The Cronbach Alpha test was used to assess internal consistency of the entire scale, and the Wilcoxon test was used to test the reliability of each section. The McNemar and McNemar–Bowker tests were used for detailed analysis of the survey questions, and an ACDAS-Turkish cut-off points ≥ 26[14] and CFSS-DS ≥ 36[17] were considered to be indicative of anxiety.

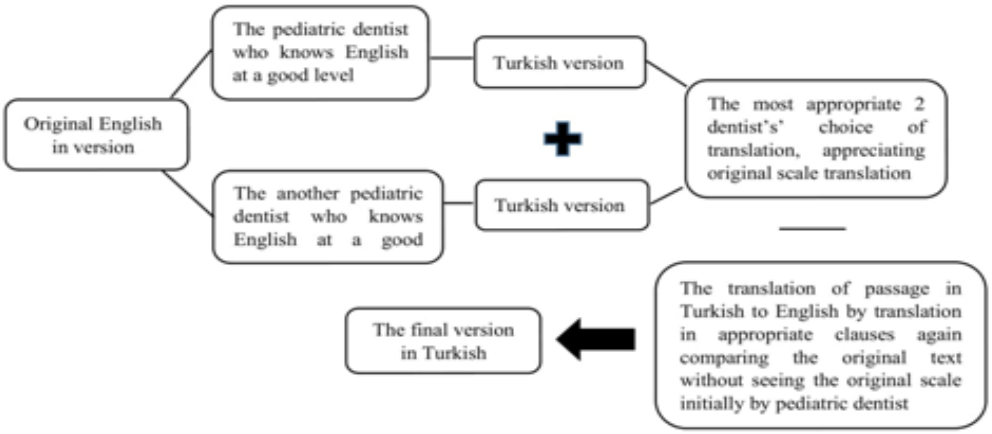

Figure 1. Flowchart of language validity

RESULTS

The study sample included 122 children, of which 65 were girls (53.3%) and 57 were boys (46.7%), and the mean age was 9.84 ± 1.696 years (range: 7–12 years).

Findings in relation to the cut-off score

The first application of the scale (ACDAS1) by the lead researcher found that 44 (36.1%) children were anxious, while 78 (63.9%) participants exhibited no anxiety based on their total scores being less than the cut-off values described previously. In contrast, 30 (24.6%) children were found to be anxious and 92 (75.4%) children exhibited no anxiety when using the CFSS-DS scale in the same visit (Table 1).

Reliability analysis

The internal consistency Cronbach Alpha coefficient ($\alpha = 0.934$) and the Cohen's Kappa test ($\varkappa = 1$, $p < 0.001$) showed high reliability of the ACDAS scale and good interobserver agreement between the two researchers, respectively (Table 2).

No statistically significant differences were observed in the dentist's responses to the questions in the Dental and Cognitive sections of the ACDAS scale when comparing the first application by the lead researcher to the second application two weeks later (ACDAS2) ($p > 0.05$). The frequency and distribution of the total scores for the questions in the Cognitive and Child Evaluation sections have been shown in Table 3. Sstatistically significant differences in the legal guardian's responses to the questions in the Child Evaluation section ($p = 0.004$; $p < 0.01$) (Table 3) were observed. Approximately 29.5% of the respondents reported feeling ashamed when they went to the dentist; 40.1% said that they were ashamed of the appearance of their teeth; and 36.9% said that they were worried about losing control when visiting the dentist.

Table 1. Distrubution of groups to anxious and non anxious

	N	%
ACDAS1		
< 26	78	63.9
≥ 26	44	36.1
ACDAS2		
< 26	77	63.1
≥ 26	45	36.9
CFSS-DS		
< 36	92	75.4
≥ 36	30	24.6
Total	122	100.0

Table 2. Interobserver agreement

		ACDAS1		Total	$\varkappa$ (Cohen's Kappa)	p
		<26	≥26			
ACDAS (Researcher 2)	<26	10	0	10	1.000	p<0.001
	≥26	0	10	10		
	N	10	10	20		

**Values of p<0.05 were considered statistically significant* (N=20, κ=1, p<0.001)

Table 3. Cognitive part and information related to child assesment

	ACDAS 1		ACDAS 2		p
The Cognitive Part	N	%	N	%	
3	12	9.8	8	6.6	
4	28	23.0	30	24.6	p=0.398
5	37	30.3	37	30.3	
6	45	36.9	47	38.5	
TCA-For Legal Guardian					
2	55	45.1	69	56.6	
3	54	44.3	45	36.9	*p=0.004
4	8	6.6	8	6.6	
5	5	4.1	-	-	
TCA-For Operator					
1	50	41.0	64	52.5	
2	61	50.0	43	35.2	p=0.150
3	11	9.0	15	12.3	
Total	122	100	122	100	

**Values of p<0.05 were considered statistically significant*

Statistically significant differences in the legal guardian's responses to the question "has your child had dental treatment before? (Yes/No)" in the Child Evaluation section was observed ($p < 0.001$) (Table 4). No statistically significant differences were observed in the answers to the question "how do you expect your child's behavior to be like today? (Happy/Well/Afraid)" between the first and second applications of the scale ($p > 0.05$) (Table 4).

Table 4. Child assessment- Time-related changes in legal guardian responses

		ACDAS1		ACDAS2		p
		N	%	N	%	
Has your child had previous dental treatment?	Yes	109	89.3	122	100	*p<0.001
	No	13	10.7	-	-	
How do you expect your child's behaviour today?	Happy	58	47.5	69	56.6	p<0.199
	Ok	55	45.1	45	36.9	
	Scared	9	7.4	8	6.6	
	Total	122	100	122	100	

**Values of p<0.05 were considered statistically significant*

Validity analysis

The criterion validity of the scale was evaluated using CFSS-DS, and a highly statistically significant correlation was observed between the ACDAS and CFSS-DS (Spearman's Correlation Analysis $r = 0.760$; $p < 0.001$).

The AUC obtained from the ROC analysis was 0.849 (Figure 2) indicating good diagnostic performance.

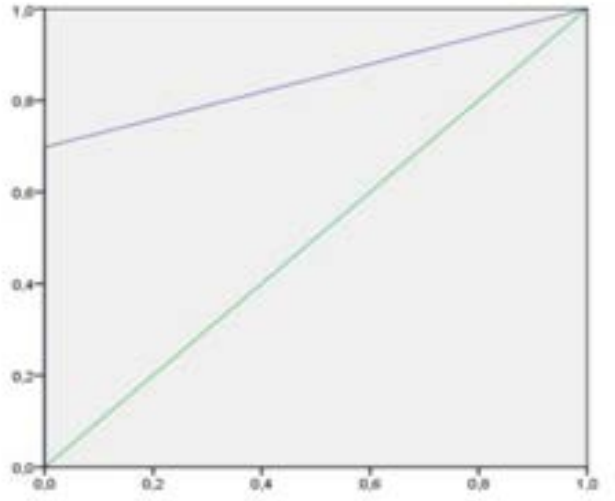

Figure 2. Roc curve

DISCUSSION

The main aim of this study was to evaluate the psychometric characteristics of ACDAS after translation to Turkish by analyzing its validity and reliability. The ACDAS may have advantages over the Turkish measures, such as the inclusion of a separate cognitive section with only 3 faces in response. In addition, the scale can uniquely measure dental anxiety by asking questions to the child, mother/legal guardian, and dentist simultaneously. This feature can provide 360^0 ratings of children's dental anxiety. Therefore, this scale can be considered as a much more beneficial and valid Turkish anxiety scale compared to the previously reported scales.[14,16,18]

This study design had several limitations. Firstly, recruitment of participants was restricted to a single clinic and, for this reason, the results may not be generalizable to all children of this age in Turkey. Secondly, this study only evaluated the validity and reliability of the scale, and the dental anxiety observed in the sample and the factors associated with it were not explored.

Cohen's Kappa test ($\kappa = 1$, $p < 0.001$) showed excellent interobserver agreement in this study, and this was similar to a previous study conducted by Al-Namankany et al.[18] who observed slightly lower inter observer agreement ($\kappa = 0.89$; 95% CI: 0.82–0.96) and an internal consistency Cronbach Alpha coefficient ($\alpha = 0.90$) similar to findings of this study ($\alpha = 0.934$), indicating homogeneity in all items of the scale. Moreover, the internal consistency of the ACDAS was found to be comparable to that of the CFSS-DS (α = 0.8–0.9).[2] Based on these findings, the Turkish version of the ACDAS was found to be a reliable dental anxiety scale suitable for use in children and adolescents. Mafla et al.[19] previously evaluated the validity and reliability of the Spanish version of ACDAS and reported a high internal consistency coefficient ($\alpha = 0.88$). The Cronbach Alpha reliability coefficient observed in this study was similar to that reported by a systematic review examining anxiety scales.[2]

Although no statistically significant differences were detected in the repeatability of the dentist's answers in dental, cognitive and child evaluation section in the evaluation of the repeatability of ACDAS ($p>0.05$), a statistically significant difference was observed in the legal guardian's response to the question "has your child had dental treatment before? (Yes/No)" between the first and second applications of the scale ($p < 0.001$). A total of 109 participants answered "Yes" and 13 participants responded "No" in the first application of the scale and, as dental treatment of the participants was carried out in the same visit,

all participants were able to respond "Yes" to this question in the second appointment. This significant difference between the two applications of the ACDAS over a two week interval could be attributed to the fact that patients who had never had dental treatment before at the time of the first appointment went on to receive treatment and, therefore, this status changed at the time of the second appointment.

The high correlation ($r = 0.760$; $p < 0.001$) between the ACDAS and the CFSS-DS supports the validity of ACDAS in the field of dentistry, indicating that it was capable of accurately measuring the characteristic of interest without confusing it with any other characteristics. Although the ACDAS evaluates the child's experience of dentistry, similar to the CFSS-DS, it also evaluates other important factors such as the perception of losing control, feeling shame, and loss of self-confidence in the child, all of which are related to the cognitive nature of the child and may play a role in anxiety. The correlation values obtained in this study were similar to those observed in a previous study comparing the validity of MCDAS with CFSS-DS ($r = 0.82$; $p < 0.001$)[20], and higher than that observed in another study also comparing the validity of ACDAS with CFSS-DS but in a different population ($r = 0.46$, $p = 0.007$; $p < 0.001$)[21].

The area under the ROC curve, used to assess the ability of the ACDAS scale to identify an anxious individual accurately, was high (EAA = 0.849) in this study, and these findings were similar to that reported by Al-Namankany et al.[14] (EAA = 0.80).

This study showed that acceptable results were obtained in terms of both numerical and categorical data. The study sample included children in the age range of 7–12 years, similar to previous studies[19,21], as younger children would not have the cognitive complexity necessary to accurately report their reactions to dental treatments.[22] Moreover, as the questions on the scale were answered by the participants themselves, only those who had the ability to read Turkish (7 years and older) would be eligible.

Evidence suggests that the prevalence of dental anxiety varies globally, with estimates ranging between 3% and 43%,[23-25] and Folayan et al.[26] argued that these differences could be attributed to the method of study as well as environmental factors themselves. This study found that 36.1% of children aged 7–12 years exhibited anxiety (≥26 points) and, although higher than the estimates provided by previous studies[5,27,28] evaluating anxiety in a similar age group, this proportion was within the global range.

CONCLUSIONS

Based on these findings, the Turkish version of the ACDAS may be considered as a valid and reliable scale for the identification of children with dental anxiety, thus enabling recognition of the causes of their concerns and facilitating treatment and prevention of further anxiety.

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Assessment of oral health literacy of principal caregivers and its association with dental caries in Mexican disabled pediatric patients

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Highlights

- Disabled persons have a higher incidence of caries and periodontal disease than the general population.
- Oral health literacy level of principal caregivers was significantly associated with disabled pediatric patient´s high result in the DMFT index.
- Low oral health literacy level of principal caregivers is a risk factor to have a high DMFT in disabled pediatric patients.

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Abstract

Aim: To assess oral health literacy (OHL) of principal caregivers (PC) and its association with dental caries in Mexican disabled pediatric patients (DPP). **Methods:** 40 disabled patients and their PC were recruited from the pediatric dentistry Clinic (UAS) and the Center of Rehabilitation and Special Education of the Institute for Integral Development of the Family in Sinaloa, Mexico. Spanish Oral Health Literacy Scale (SOHLS) was used for the OHL assessment of PC. DPP were subject to clinical examinations to observe the mean number of decayed, missing, or filled teeth through the DMFT index. Disabilities were classified in neuromotor, auditive and language, visual, intellectual, or multiple. To estimate OHL differences, the variable was divided into 3 categories according to the obtained score: low (0-16), medium (17-22), or high (23-29). For the logistic regression model, the variable was categorized on 2 according to ANOVA results: lower (0-22) and high (23-29) to estimate the association between PC low OHL and DMFT percentual increase. **Results:** Regarding DPP, there were no differences in age expressed in years (12.47(±8.16) (p=0.673)), DMTF (0.34(±0.26) (p=0.673)) or the number of teeth (23.32(±5.29) (p=0.653). Besides, no differences were found in PC age expressed in years (41.22(±8.91) (p=0.795)), PC scholarship expressed in years (3.95(±8.91) (p=0.128)) or PC OHL (22.27(±3.99) (p=0.205)). The logistic regression model categorizing OHL on high (>22) and low (≤22) found 0.97% more risk to be a PC with low OHL per percentual unit of DMFT increment (p=0.041). **Conclusions:** The OHL level of PC is significantly associated with DPP´s high result in the DMFT.

Keywords: Caregivers; Dental Caries; Dentistry; Literacy

INTRODUCTION

According to World Health Organization (WHO), about 15% of the world population lives with some form of disability, and 2-4% of them can experience significant difficulties in self-care functions given by developmental or sensitive disorder[1,2], which result in communicative or intellectual impairments[3] that conducts to a long-term dependence.[4] In this regard, critical aspects such as oral health care depend on the knowledge, attitude, and actions of the PC.[5,6]

This is relevant since oral health is a major constituent of general health.[7] It is widely reported that disabled persons (DP) have a higher incidence of caries and periodontal disease than the general population.[8-11] Oral health problems like caries and periodontitis can lead to pain[12] and eating difficulties, poor nutrition [13,14], digestive and respiratory problem associated to recurrent infections[15,16], sleep disturbance [17], and decreased self-esteem[5] (Figure 1). Furthermore, dental caries and gingivitis in DP (2-19 years old) have been strongly associated with the level of the mother´s education.[18]

Oral health literacy (OHL) plays a central role in the relationship between the perception about signs and symptoms, perceived value of good oral health, and the ability to understand the health care system.[19] This relationship has been associated with several oral health actions and outcomes. There is evidence that OHL of PC is a strong predictor of oral health in the DP under their care.[20] Health literacy includes the ability to understand instructions on prescriptions or basic health information, medical education brochures, and health personnel directions.[21] PC are in charge of preventive oral care, diet, oral hygiene, and attendance at dental consultations.[22] Besides, there is a strong relation between OHL of PC and decayed, missing, and filled teeth (measured by DMFT index) in the persons under their care.[23] This is relevant since dental caries is the most incidental disease of the oral cavity in Mexico and worldwide.[24] Therefore, the purpose of this study was to associate the OHL of PC with dental caries in Mexican DPP.

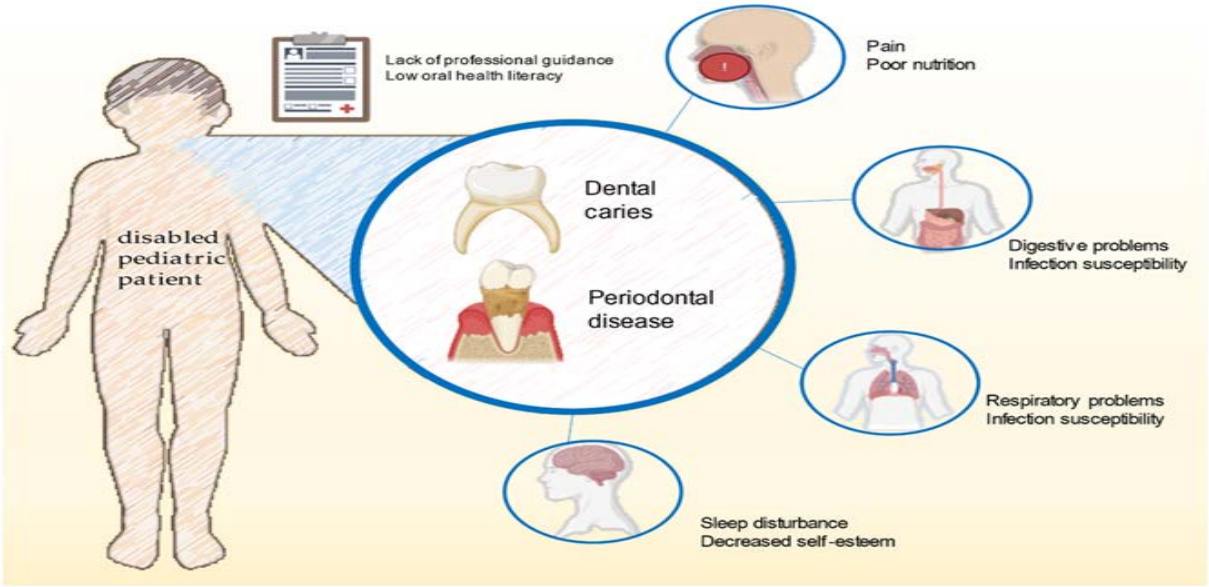

Figure 1. Oral health problems in DPP like caries and periodontitis can lead to other affections such as pain and eating difficulties, poor nutrition digestive and respiratory problem associated to recurrent infections, sleep disturbance, and decreased self-esteem. Lack of professional guidance or low OHL of PC could be key for the development of illness

METHODS

Study sample

A sample size of 8 per disability class was calculated for detecting differences between 2 proportions on DMFT index, estimated by a X^2 test, using epidemiologic data of Down´s syndrome patients[25] that reports 37% on DMFT index vs 77% of cerebral palsy patients [26], using a $\alpha = 0.05$ and $\beta = 0.8$ for a 2 tiled test. In this cross-sectional study, 40 disabled patients and their PC were recruited from the pediatric dentistry Clinic (Universidad Autónoma de Sinaloa UAS) and the Center of Rehabilitation and Special Education of the Institute for Integral Development of the Family in Sinaloa, Mexico**.** Informed consent was obtained from all subjects involved in the study. This study was previously approved for ethical committee of Universidad Autónoma de Sinaloa. DPP included had more than 20 teeth and their PC accepted to participate and filled the instrument. DPP with genetic syndromes, systemic health problems, illiterate PC, or incomplete questionnaires were excluded.

Measurements

Spanish Oral Health Literacy Scale SOHLS (reliable and validated instrument)[27] was used for the OHL assessment of PC. This instrument is a self-answered questionnaire for Mexican Spanish speakers that evaluates how a person understands, processes, and applies specific information. It contains 29 items that assess five literacy skills: location, integration, generation, calculation, and return (9 correspond to cycling skills, 4 to location, 2 to formulation, 3 to generation, and 11 to integration skills). Each item has a value of one if answered correctly. The complete test has a summative score from 0 (lowest literacy) to 29 (highest literacy).

DMFT

DPP were subject to clinical examinations through DMFT Index. This index is the most important used in epidemiological studies of the health status of the community.[28] The DMFT score of the samples was determined based on the results of clinical examination and calculation of the number of decayed (D), filled (F), and missed (M) teeth due to caries. The data were collected through observation and direct examination using a mirror number #4. During the inspection, the subjects under examination and the researcher sat close to the window to examine under the maximum natural light.

Disability classification

Disability classification was analyzed as a confusing variable and decoded in agreement to the Social Integration Law of Sinaloa for disabled people (neuromotor, auditive and language, visual, and intellectual or multiple).

Standardization

SOHLS standardization was performed with a pilot probe of 10 PC. Complications on lecture, understanding, or tiredness were evaluated during questionnaire filling without finding any problem. DMFT index standardization was performed using intraoral pictures of 10 patients. DMFT results of every patient were assessed by 2 evaluators, and exercises were repeated 3 times in 1-week intervals using the Intraclass concordance coefficient. A 0.998% concordance was obtained between evaluators.

Statistical analysis

Significance and differences were assessed across disability classes using different statistical tests. ANOVA was used for parametric variables. To estimate OHL differences, the variable was divided into 3 categories according to the obtained score: low (0-16), medium (17-22), or

high (23-29). For the logistic regression model, the variable was categorized on 2 according to ANOVA results: lower (0-22) and high (23-29) to estimate the association between PC low OHL and DMFT percentual increase.

RESULTS

Descriptive statistic was developed to find differences between disability classes (Table 1). Regarding DPP, there were no differences in age expressed in years (12.47(±8.16) (p=0.673)), DMTF (0.34(±0.26) (p=0.673)) or the number of teeth (23.32 (±5.29) (p=0.653) showing homogeneity of the main confounding variables of the DPP.

Besides, no differences were found in PC age expressed in years (41.22(±8.91) (p=0.795)), and PC scholarship expressed in years (3.95(±8.91) (p=0.128)) or PC OHL (22.27(±3.99) (p=0.205)).

DMFT analyses associated with PC OHL raised lower values of DMTF related to high OHL group (0.26 (±0.21)) when compared to medium OHL (0.42 (±0.29)) or low OHL ((±0.32) (p=0.041)) groups. Logistic regression model categorizing OHL on High (>22) and Low (≤22) found 0.97% more risk to be a PC with low OHL per percentual unit of DMFT increment (p=0.041) (Table 2). Considering the uniformity of the confounding variables, we assume that the differences detected were not influenced by the main confounding variables.

Table 1. Descriptive analysis for disability class

Disability class	DPP Age	DMFT	Teeth	PC age	PC scholarship	OHL
Neuromotor	11.40(±6.71)	0.32(±0.3)	23.50(±6.34)	39.90(±7.82)	4.50(±7.82)	23.2(±3.11)
Auditivea nd language	11.80(±6.80)	0.39(±0.22)	24.30(±3.40)	40.80(±6.17)	4.10(±6.17)	20.9(±5.62)
Visual	15.30(±12.72)	0.37(±0.33)	21.50(±7.33)	43.70(±11.46)	4.60(±11.46)	24.0(±3.09)
Intellectual or multiple	11.40(±4.83)	0.29(±0.19)	24.00(±3.23)	40.50(±10.18)	2.60(±10.18)	21.0(±3.16)
Total	12.47(±8.16)	0.34(±0.26)	23.32(±5.29)	41.22(±8.91)	3.95(±8.91)	22.27(±3.99)

DPP: Disabled pediatric patients; DMTF: Index to measure Decay, Missing or filled teeth; PC: Principal Caregivers; OHL:Oral Health Literacy

Table 2. Logistic regression analysis found more risk to be a PC with low OHL per percentual unit of DMTF increment

OHL	N	DMFT		CI (95%)	Significance	
Low (10-16)	4	0.54 (±0.32)		0.29 - 1.05		
Medium (17-22)	15	0.42 (±0.29)		0.26 - 0.57	p=0.041	
High (23-28)	21	0.26 (±0.21)		0.17 - 0.36		
Total	40	0.35 (±0.26)		0.26- 0.43		
Logistic regression	**N**	**DMFT %**	**X^2**	**CI (95%)**	**Exp (B)**	**Significance**
High	19	44 (±29)	p=0.019	30-58	0.970	p=0.041

OHL: Oral Health Literacy; DMTF: Index to measure Decay, Missing or filled teeth; CI: confidence interval; X^2: squared Chi; Exp (B): exponentiation of the B coefficient.

DISCUSSION

The results demonstrate that the OHL level of PC is significantly associated with DPP´s high result in the DMFT index. Besides, improving the OHL of PC is the best option to help strengthen their abilities to promote oral health and DPPs general health. These findings are opposite to those obtained by Divaris and Cols in 2012[29], who found poor correlations between the assessment of PC clinical needs of patients. This difference could be explained given the age of children included in the study who were less than two years old. Besides, as mentioned by Baskaradoss & Cols in 2018[20], parents of very young children may overestimate the oral health status of their child.

This study has some limitations regarding the use of the DMFT index for dental caries detection, which fails to detect interproximal and incipient lesions. Besides, the DMF score is a count that does not indicate the number of teeth that are at risk and does not discriminate between the mix of decayed, missing, and filled teeth or surfaces, and whether teeth are lost for reasons other than caries; therefore, the validity of the DMF can be considered as reduced.[30] Also, a larger or multi-centered study can improve the validity of results. In this regard, more work is required to further explore the different aspects of OHL. On the other hand, the strength of this work is the sampling method employed since aimed to provide a comprehensive picture of the DPP in this country, as patients of the study come from different attention centers of Sinaloa and have different socioeconomic and cultural statuses.

In 2019, 1.2 million of DPP were reported in Mexico. The results of this study suggest that the age of DPP, teeth number, as well as descriptive data of PC, have no differences regardless of disability class. These similarities are given by the presence of physical or communicative barriers at the time of oral health care. This could be due to a lack of experience of professionals in the management of information and the lack of adequate guidance to their PC.[31]

Findings showed that Low PC OHL is a risk factor to have a high DMFT in DPP which agrees with data suggesting that patients with lower values of health literacy had a poor general and oral health status.[20] Besides, there is evidence that shows that PC with high OHL are more attentive to prevent oral health of the patients they take care.[32,33] As has been previously reported[34], knowledge and oral health literacy of PC are important predictors that must be taken into account to reduce social inequalities in health through actions undertaken at the local level in patients with disabilities.

Dental caries in Mexico is a public health problem. Therefore, the promotion of primary prevention and understanding of oral health care in DPP should be an integral part of dentistry. These individuals are a risk group[35] since from a young age they require specialized dental care. Besides, it is important to ensure PCs adequate literacy given by trained professionals that allows understanding preventive care, the importance of diet, frequency of visit to the dentist, and other instructions to guarantee oral health of DPP.

CONCLUSIONS

The OHL level of PC is significantly associated with DPP´s high result in the DMFT index. Improving the OHL of PC might, therefore, help strengthen their capacities to promote oral health, thus helping to improve DPP general health.

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Surgical management of central giant cell granuloma of mandible and prosthetic rehabilitation in a nine year girl: A case report

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Highlights

- Central giant cell granuloma is an idiopathic, osteolytic lesion of jaws.
- A rare case of large destructive CGCG involving anterior region of mandible in a nine-year girl is presented in this case report.
- The case was treated successfully by enucleation and curettage with satisfactory preservation of the continuity of mandible.

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Abstract

Central giant cell granuloma (CGCG) is an uncommon, benign, idiopathic, osteolytic lesion of jaws, histologically characterized by multinucleated giant cells distributed in fibrovascular connective tissue stroma. Accurate diagnosis of the lesion is essential for the successful management and the prognosis of this locally destructive lesion. In this paper, a rare case of large destructive CGCG involving anterior region of mandible, causing expansion of labial cortical plate and mobility of teeth in a nine-year girl is presented. It was treated successfully by enucleation and curettage with satisfactory preservation of the continuity of mandible. Nine months post operatively, the child was rehabilitated with a temporary partial denture to improve esthetics, phonetics and function. One year clinical and radiographic follow up showed new bone formation and no evidence of recurrence.

Keywords: Curettage; Giant cell; Granuloma; Rehabilitation

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INTRODUCTION

Central giant cell granuloma (CGCG) is an uncommon, idiopathic, osteolytic lesion of multinucleated giant cells. It is histologically benign but locally aggressive lesion of bone occurring most commonly in craniofacial skeleton.[1] The lesion was initially described by Jaffe in 1953 as a reparative reaction, later this term was discontinued because it is more destructive than reparative. The World Health Organization (WHO) has defined it as an intra-osseous lesion consisting of cellular fibrous tissue that contains multiple foci of hemorrhage, aggregations of multinucleated giant cells and occasionally trabeculae of woven bone.[2] It occurs most commonly during the first 30 years of life, with nearly 50% occurring in patients younger than 16 years of age. CGCG occurs more often in females than in males, the mandible is involved twice as often as the maxilla.[3]

CGCG is usually asymptomatic and it may present with aggressive growth and expansion of jaws. Radiographically, CGCG may vary from a unilocular radiolucency to a multilocular bone-destructive lesion with displacement of teeth, root resorption and cortical perforation. Sensory disturbance has been rare.[3,4] The lesion should be differentiated with other benign jaw lesions such as Brown's tumor, Ameloblastoma, aneurysmal bone cyst, odontogenic cysts, myxomas, fibrous dysplasia and ossifying fibroma.[5,6,7]

Histologically, the lesion is characterized by loosely arranged fibrous stroma with presence of multinucleated giant cells. These giant cells resemble osteoclasts with 20 or more nuclei.[5] Treatment for CGCG ranges from local enucleation and curettage to enbloc resection which can lead to large defects in the jaws. Recently for management of non-aggressive variants, intralesional steroids, systemic calcitonin, α-interferon and denosumab have been considered.[5,8,9,10] Oral rehabilitation after surgical intervention proves challenging and affected patients generally require prosthetic rehabilitation to facilitate function.[8] The potential for recurrence further complicates this process as recurrence rate[9] ranges from 11-49%. The purpose of reporting this case is to discuss the management of central giant cell granuloma involving mandible in a nine-year old female child followed by prosthetic rehabilitation.

CASE REPORT

A nine-year old girl reported with a chief complaint of swelling in the front region of lower jaw from one month. The swelling increased in size gradually. It was not associated with pain, difficulty in mouth opening and paresthesia. Extra oral examination revealed a solitary, diffuse swelling in symphysis area extending between the corners of the mouth (Figure 1). No abnormalities were reported with mouth opening. Skin over the swelling was normal. Swelling was hard in consistency and non-tender on palpation and it was not associated with lymphadenopathy. Intra-orally diffuse swelling in labial sulcus extending from teeth #31 to #84 region with obliteration of labial vestibule was observed. The mucosa over the swelling was stretched, no sinus opening or any discharge was present (Figure 1). The associated teeth 31,41, 42, 83, 84 showed grade-2 mobility. The swelling was bony hard in consistency and non-tender on palpation.

Panoramic radiograph showed a diffuse radiolucent area in the anterior region of mandible with displacement of incisors (Figure 2). Since the borders and extent of lesion was not clear, Cone beam computed tomography (CBCT) of anterior region of mandible was made. Various sections of CBCT showed unilocular radiolucency with expansion of labial cortical plate and resorption of roots of 31,41 (Figure 2). However, the lingual cortical plate was intact. The lesion measured 3x2cms in maximum dimensions on the orthopantomogram. nose, webbed neck, low posterior hair line, and with competent lips (Figure 2).

Figure 1. Clinical appearance of the lesion. 1.a: Extra-oral appearance; 1.b: Intra-oral appearance of the lesion showing obliteration of labial vestibule

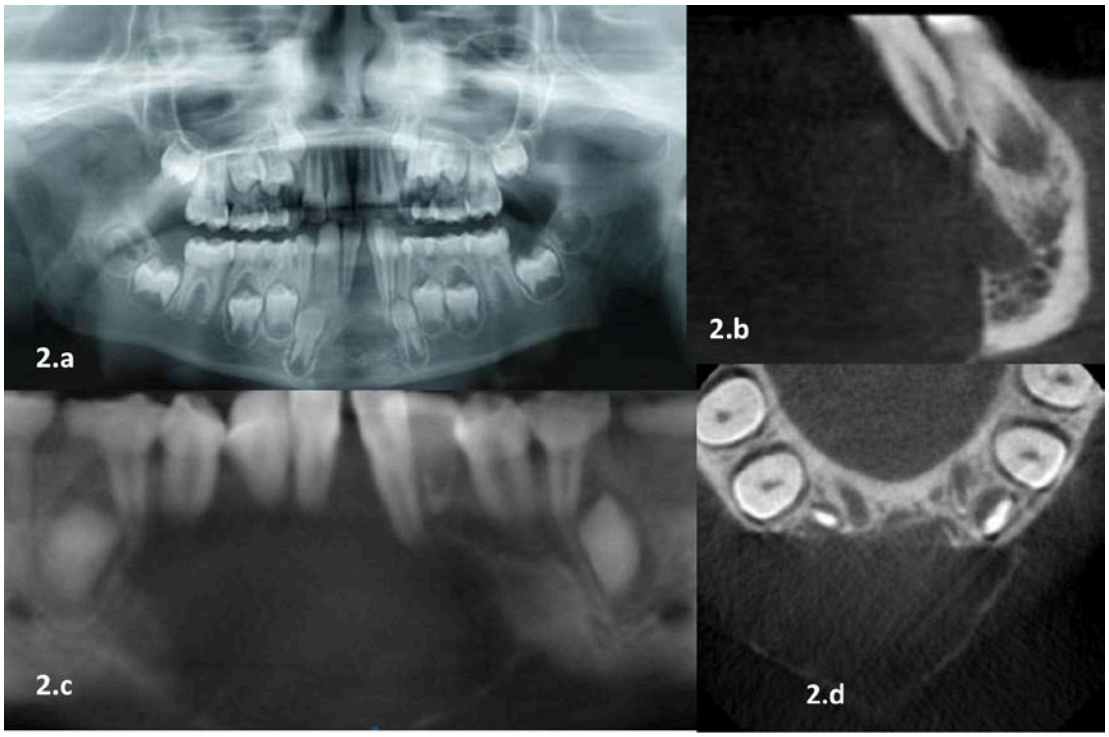

Figure 2. Radiographic appearance of the lesion 2.a: OPG showing diffuse radiolucency in the anterior region of mandible; 2.b, 2.c and 2.d: Lesion in various sections of CBCT

Procedure was explained to the parents and written consent was obtained for the treatment and publication of the case report. A provisional diagnosis of Central giant cell granuloma was made. The differential diagnosis was Odontogenic Cyst, Ameloblastoma, Brown's tumor and Aneurismal bone cyst. Biochemical investigations showed normal serum calcium, phosphorous, alkaline phosphatase and parathormone levels, ruling out the Brown's tumor. Incisional biopsy was performed which showed multinucleated giant cells surrounded by a loose fibrous stroma with areas of hemorrhage and inflammatory cell infiltrates (Figure 3).

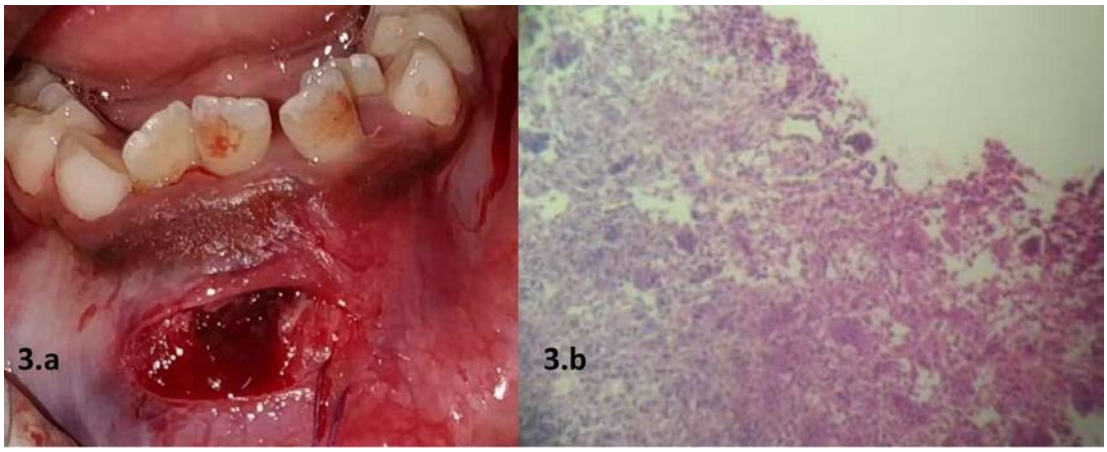

Figure 3. 3.a: Lesion during incisional biopsy; 3.b: Histopathological slide showing multinuclear giant cells in fibrous stroma

This confirmed the diagnosis of central giant cell granuloma. The child was referred to endocrinologist and with his opinion and also by considering the age of the patient, intralesional steroid was the initial chosen treatment modality.[10,11,12] Treatment was started with triamcinolone acetonide 10mg/ml and lignocaine 2% with adrenaline 1:200,000 (50:50), at the dose of 1ml solution for every 1cm of radiolucency, as determined on an orthopantomogram (OPG). The total dose of triamcinolone administered was 60mg over six weeks in increments of 6 injections of 10mg each. Clinical and radiological evaluation one week after the last dose showed no regression in radiolucency (Figure 4). Hence, with the parent's consent surgical enucleation and curettage[13] was planned.

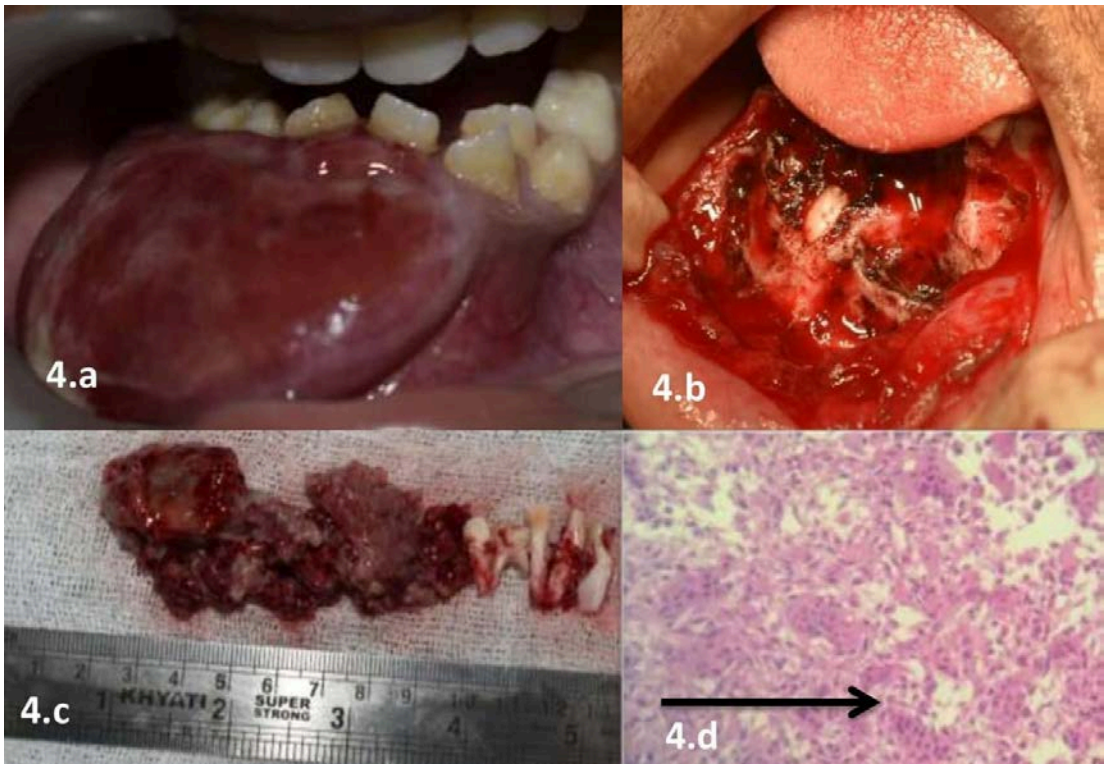

Figure 4. 4.a: Increase in size of the lesion after intralesional steroid therapy; 4.b:Surgical enucleation of lesion; 4.c: Gross appearance of the excised tissue and extracted teeth 31,41,42, 83, 84; 4.d: Histopathological slide showing multinuclear giant cells in fibrous stroma confirming the diagnosis

After taking anesthesiologist's opinion, routine blood investigations were done and the child was scheduled for surgery under general anesthesia. The surgical procedure was performed in association with oral and maxillofacial surgeon. Incisions were made 1cm away from the margins of the bony defect and flap was raised. Since, the patient was a child in the age of growth and development, a radical surgery in the form of bone resection was not considered as the first step. However, complete surgical enucleation[13] of the lesion along with extraction of the mobile teeth 31, 41, 42, 83 and 84 (Figure 4) followed by aggressive curettage was done. This avoids formation of large defects that would compromise esthetics in the young child.[13] Neurovascular bundle was not involved. Tooth buds of 43, 44 were left intact. The bony defect was then irrigated and resorbable sutures were placed. Histopathology of the excised lesion showed multiple giant cells in fibrous stroma (Figure 4). The patient was instructed to use frequent warm saline rinses starting from the day after surgery to keep the wound free of food debris. Child was regularly reviewed until healing was complete. Child was kept under observation and six months post operatively complete healing with new bone formation was seen in the defect area on the panoramic radiograph (Figure 5). At nine months follow up a temporary partial denture (Figure 6) was fabricated with C clasp on 85, Adam's clasp on 75 and it was inserted in the oral cavity.

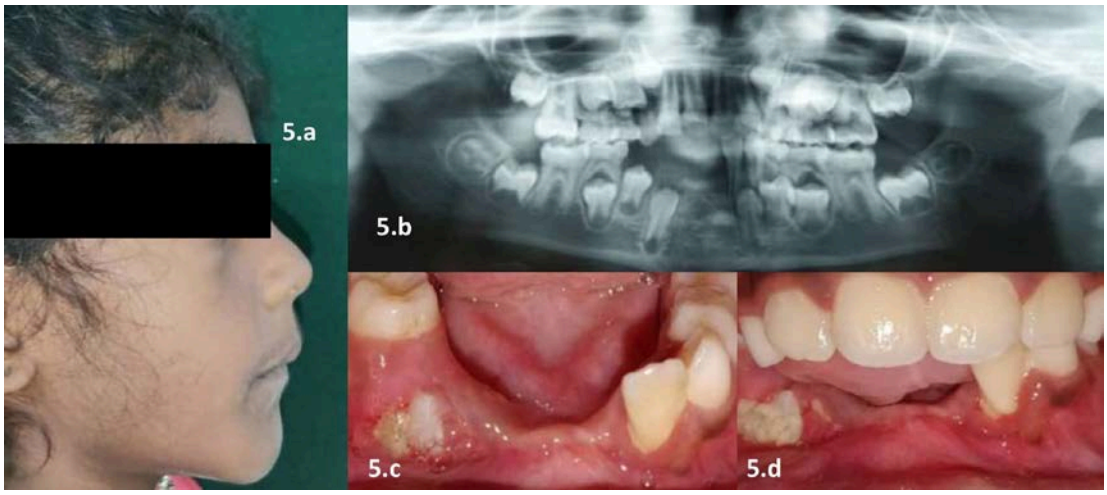

Figure 5. 5.a, 5.c & 5.d: Extra and intra oral image showing 6-months post operative healing, 5.b: 6-month follow up OPG showing satisfactory healing and new bone deposition

Figure 6. 6.a shows 9-months post-operative healing; 6.b, 6.c & 6.d: Fabrication and insertion of treatment partial denture to improve the esthetics, phonetics and function

Thus, it improved esthetics, phonetics and function in the child patient. One year clinical and radiographic (Figure 7) follow up showed new bone formation and no evidence of recurrence. The child will be followed up for next three years.

DISCUSSION

Central giant cell granuloma is a benign tumor of unknown etiology, it belongs to a group of giant cell tumors.[2] Etiology and pathogenesis of CGCG of jaw bones have not been explicitly defined or recognized clearly. It is believed that it can occur as a response to past injury or as a reactive response to changes in blood supply to the bone.[8] Based on the clinical and radiographic features, it can be classified into two types, non-aggressive and aggressive type. Non-aggressive lesion is slowly expanding and do not show cortical erosion, where as aggressive type is more destructive, grows rapidly with cortical perforation, displacement of teeth and resorption of roots.[5] Correct identification and diagnosis can be quite challenging since clinical and radiographic appearance is not pathognomonic and it is similar to aneurysmal bone cyst, Ameloblastoma, ossifying fibroma and brown's tumor.[4] Final diagnosis can be made by correlating clinical, radiographical findings with histological examination.

In this case, age and gender of the child, location of the lesion and expansile nature of the lesion were indicative of central giant cell granuloma. To rule out Brown's tumor and hyperparathyroidism, biochemical investigations were done which showed normal serum calcium, phosphorous levels, normal alkaline phosphatase levels, normal parathormone levels. Incisional biopsy confirmed the final diagnosis of central giant cell granuloma.

The treatment advocated for CGCG has predominantly been varying extents of surgery, ranging from simple curettage to radical resection.[6] Depending on the size and location of the lesion, surgical intervention will be associated with varying degrees of morbidity. Although radical surgical intervention can be effective, an inevitable loss of teeth and tooth germs will result.[6] Conservative medical regimens have been extensively reported, including systemic calcitonin, α-interferon, denosumab and intralesional corticosteroids.[4] Intralesional corticosteroid use for the treatment of CGCG was first described by Jaco-way et al in 1988. The rationale for its use was that steroids might act by suppressing any angiogenic components of the lesion and inhibit the osteoclast activity. In present case triamcinolone acetonide diluted in an anesthetic solution was infiltrated similar to the protocol recommended by Terry and Jacoway.[10]

Figure 7. One-year follow up orthopantomogram (OPG) shows new bone formation and no evidence of recurrence.

However at the end of six weeks the lesion did not reduce in size, the reason for this can be attributed to a population of altered osteoclasts that do not have cell membrane receptors to corticosteroids.[4] Since, the patient was a child in the age of growth and development, a radical surgery in the form of bone resection was not considered as the first step. However, complete surgical enucleation of the lesion along with extraction of the mobile teeth 31, 41, 42, 83 and 84 (Figure 4) followed by aggressive curettage was done. This avoids formation of large defects that would compromise esthetics in the young child.[13] The tooth buds of 43, 44 were left intact. Literature search revealed other innovative methods to restore the lost tissues and hasten bone fill in the defect. Use of PRF, native extracellular matrix (ECM) and autologous bone grafts in large defects have been reported. In our case since the lingual cortical plate was intact, adequate healing and bone fill after six months was seen on panoramic radiograph. Hence, at nine months follow up the child patient was rehabilitated with a temporary partial denture to replace the missing teeth.[8] One year clinical and radiographic follow up showed new bone formation and no evidence of recurrence. The child will be followed up periodically.

CONCLUSIONS

Although benign jaw lesions constitute only a small number of pathologic conditions seen by a pediatric dentist, they are of great significance since they have the potential ability to jeopardize the health and longevity of the patient. Hence, early diagnosis and management are very essential.

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Multidisciplinary approach in the anterior open bite using a McNamara expander with palatal crib during mixed dentition stage: A case report

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Highlights

- Pediatric dentists need to be aware of the multidisciplinary work involved in the treatment of anterior open bite as a result of its multifactorial etiology.
- This case report presents an orthodontic treatment option based on the elimination of confounding factors during diagnosis of the malocclusion.
- This article seeks to delegate competencies regarding the treatment of anterior open bite since the dentist is not able to fully intervene in some circumstances.

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Abstract

Open bite can be defined as an absence of occlusion, most frequently located in the anterior region of dental arches and its etiology is multifactorial. We present a clinical case of an 8 years and 10 months child presenting an anterior open bite (AOB) with transverse maxillary deficiency caused by tongue thrust during mixed dentition. The malocclusion was corrected by means of a McNamara expander with a palatal crib jointly with the association of speech therapy for tongue repositioning, and otolaryngology to treat adenoid hypertrophy due to its correlation with AOB. The multidisciplinary approach was effective in correcting the malocclusion with stable results after 2 years post-treatment.

Keywords: Interceptive Orthodontics; Malocclusion; Open Bite

INTRODUCTION

Open bite can be defined as the absence of occlusion primarily in the anterior region of the arch[1] in the presence of negative overbite, while the posterior teeth are in contact.[2]

The etiology of anterior open bite (AOB) is considered multifactorial because a multitude of variables can work in the bone-dental configuration as well as in the magnitude of dysplasia associated with it.[3,4] Therefore, several etiological factors are associated with the emergence of the AOB, such as genetic factors and environmental, standing out in addition to harmful oral habits of non-nutritive sucking, tongue thrust, mouth breathing, and interposition of the tongue between dental arches. [5]

Genetic factors expressing skeletal AOB are due to craniofacial dysplasia and are characterized by a rotation of the palatine process in the counterclockwise direction, associated with an increased anteroinferior facial height, obtuse gonial angle, and the short mandibular ramus.[6,7] On the other hand, deleterious oral habits are more frequently associated with AOB of dental origin, which results from the interruption of the normal vertical development of the anterior teeth, while skeletal open bite results from an alteration in the vertical growth of the face.[7]

The AOB is considered challenging to develop a treatment plan that results in long-term stability, given that the solidity of the results is directly related to the diagnosis, etiology, and the time of intervention.[6] Early intervention of AOB can prevent the installation of its permanent form[8], as well as time consuming treatments whose long-term stability cannot be ensured, including orthodontic extrusion of anterior teeth, orthognathic surgery with impaction of the posterior region of maxilla, or by the management of posterior teeth eruption in growing patients.[9,10]

Within an early and multidisciplinary perspective approach, the development of a rational treatment plan that takes into consideration the different variants that may be associated with the presence of deleterious oral habits will allow that the timely interception can yield satisfactory and stable results in the functional and aesthetic perspective, in addition to the psychosocial gains that early intervention can offer to the child with this type of malocclusion.

Our objective is to report a clinical case of a dental AOB, corrected employing interceptive orthodontics associated with speech therapy and otolaryngology, during the mixed dentition stage.

CASE REPORT

An 8 years 10 months white female patient attended the Preventive and Interceptive Orthodontic Clinic of FORP/USP with the chief complaint of her anterior teeth were crowded. On extraoral examination was evidenced facial symmetry, good lip sealing, obtuse nasolabial angle, defined cervicomandibular angle, convex facial profile, and slightly increased facial lower third (Figure 1).

Figure 1. Initial extraoral photographs. A: Front; B: Front smiling; C: Right side

In the functional examination, the patient was asked to retain some amount of water in her mouth for a few moments, to check whether there would be a need to use her mouth to breathe, resulting in the patient swallowing the liquid. At the moment of swallowing, the patient's lower lip was lowered to check for anterior projection of the tongue. It was observed mouth breathing and lingual thrust during deglutition characterizing the presence of atypical swallowing.

In the intraoral examination, the patient presented a molar Class III relationship subdivision left side, positive overjet (2 mm), and negative overbite (-3 mm) characterizing an AOB. It was observed the premature loss of teeth 84 and extensive crown destruction of teeth 74, which has already been endodontically accessed, as well as the presence of the first molars, lateral and central permanent incisors erupted. Her maxillary dental arch was skeletally constricted, the palate was ogival and V-shaped and the tooth size-arch length discrepancy was -3 mm while in the lower arch was +1.0 mm (Figure 2).

In panoramic radiography was verified the presence of all germs of permanent teeth, except for the 3rd molars, normality in the stage of rhizogenesis of the permanent teeth, and absence of peripicopathies.

Due to the premature loss of teeth 84, were observed the early eruption of tooth 44 and advanced rhizolysis of the roots of teeth 74. The E-space was shown to be favorable to the permanent teeth eruption (Figure 3A).

In the lateral cephalometric radiograph (Figure 3B) and the cephalometric analysis (Table 1), showed the patient was skeletal Class I malocclusion (SNA = 81º; SNB = 79º; ANB = 2º), therefore, the bone bases were well-positioned to each other and with the anterior base of the skull.

Table 1. Initial and final cephalometric measurements

Magnitude	Normal	Initial	Final
SNA	82º ± 2	81º	83º
SNB	80º ± 2	79º	81º
ANB	0º a 4º	2º	2º
SN.GoGn	32º	33º	33º
Sn.Gn	68 º	68º	69º
Facial Axis	90 º	90º	91º
1.NA	22 º	26º	24º
1-NA	4 mm	5 mm	4 mm
1.NB	25 º	25º	23º
1-NB	4 mm	5 mm	4 mm
1.1	131 º	134º	130º
S-Ls	0 mm	1 mm	2 mm
S-Li	0 mm	0 mm	-1 mm
NA.Pog	-8º a 10º	3º	4º

Figure 2. Initial intraoral photographs. A: Front; B: Left side; C: Right side; D: Upper occlusal; E: Inferior occlusal

Figure 3. A: Initial panoramic radiograph; B: Initial lateral cephalometric radiograph suggesting the presence of hypertrophied adenoid tissue

Vertically, the mesofacial skeletal pattern was verified (SN.GoGn= 33º; SNGn= 68º; Facial Axis= 90º). In relation to the dental pattern, the maxillary incisors were protruded (1.NA = 26º; 1-NA= 5mm) and mandibular presented with suitable axial inclination, however, they were protruded in relation to the apical bone base (1.NB = 25º; NB-1= 5mm). The soft-tissue profile was slightly convex (S-Ls = 1mm / S-Li = 0 mm) and the hard-tissue profile was straight (NA.Pog= 3º). There was also possible to be observed an enlarged radiopaque image in the posterior area of the nasopharynx suggestive of hypertrophy of the adenoids, which could be leading to the mouth breathing since the superior airways could be obstructed (Figure 3B).

Based on clinical and cephalometric findings, the AOB presented by the patient was diagnosed as a dental one, originating from the habit of tongue thrust during atypical swallowing. At the time, the prognosis was favorable, since the magnitude of the malocclusion was not severe and other possible confounding factors for the diagnosis were ruled out, making the correct diagnosis led to adequate treatment.

Treatment plan

At first, the treatment plan consisted of Rapid Maxillary Expansion (RME) through the installation of a modified McNamara expander associated with an anterior palatal crib (Figure 4A) and a lower lingual holding arch in the mandibular arch (Figure 4B) since the premature loss of tooth 84 has already occurred and, due to the great loss of tooth structure and advanced root resorption of teeth 74, was chosen for its extraction. The patient was also referred to an otolaryngologist (ENT) evaluation. The McNamara-type appliance is a bonded expander which is belived to present a better vertical control of posterior teeth due to its acrylic splint[11]. Therefore, in addition to expanding the maxilla, it would prevent the worsening of the anterior open bite, since for every 1 mm of posterior tooth extrusion, the bite opens in the anterior region twice this value.[12]

In the second phase, after the activation and retention period of the expander, was installed a removable appliance with a palatal crib and indicated speech therapy for the correct lingual position. This referral had to be done later since the fixed appliance on the palate would not allow the development of speech and articulation exercises. For the third planed phase, it was carried out monitoring the exfoliation of the remaining primary teeth and eruption of the permanent ones, for later referral to corrective orthodontics.

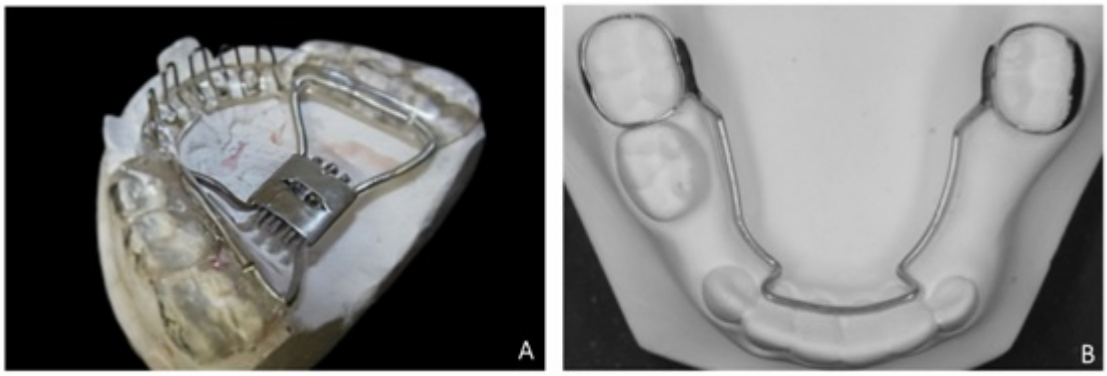

Figure 4. A: McNamara expander with vertical palatal crib; B: Illustrative photo of the lingual holding arch

Treatment goals

The first phase of the treatment aimed at gaining space in the maxilla for permanent teeth, correct the deep palate for better accommodation of the tongue, improvement in breathing and lip sealing. The choice of this type of appliance was due to its cost and ease of installation by the professional. The modified expander with palatal crib was carried out to provide the blocking of the interposition of the tongue during swallowing and combined with speech therapy and ENT approaches lead to correction of the dental anterior open bite. The lower lingual holding arch aimed at maintaining space, due to the premature loss of primary teeth 74 and 84.

Treatment alternatives

The McNamara-type appliance is tooth-borne, but there is also a tooth-tissue-borne alternative, the Haas expander. We did not consider miniscrew-assisted rapid palatal expander (MARPE), which is a bone-borne device, since our patient was only 8 years old and still presented the midpalatal suture opening.

However, taking into account the need to intercept the atypical habit of protruding the tongue during swallowing, interceptive approaches to the habit would be necessary. The use of lingual spurs could also be employed; however, as we see this therapeutic option may cause pain in the child, we chose not to use it.

Progress of treatment

Although the maxillary transverse deficiency was not as severe, the patient presented mouth breathing and a negative maxillary dentoalveolar discrepancy. The modified McNamara expander was made with a Hyrax screw (Morelli, Brazil, 11mm) positioned centrally to the midpalatal suture in the region of the primary second molars. The vertical palatal crib was made of stainless steel Ø0,70mm (.028") (Morrelli, Brazil). The central region of the crib was sectioned to allow the opening of the screw. The acrylic resin was added at the interface between the crib and the palatal mucosa to avoid damage to it. The acrylic occlusal covering of this type of appliance works as a "bite block" in the posterior segment of the arch, which prevents the extrusion of the maxillary posterior teeth, helping to close the bite. The activation protocol of the Hyrax screw was 2/4 turns per day[13], which corresponds to 0.5mm of screw opening per day, with 1/4 turn in the morning and a 1/4 turn at night, until the palatal cusps of the upper molars touch the buccal cusps of the lower molars. The expander screw was opened up to a width of 7mm and the opening of the midpalatal suture was followed by occlusal radiography of the maxilla. After the activation period, there were 6 months of retention with the same device, with assisted control for ossification of the midpalatal suture. In addition to activating the expander, the patient was instructed about oral hygiene and care with the appliance.

Treatment result

The patient was followed up monthly for one year, and her final orthodontic records were obtained two years after the correction of the AOB, to monitor the stability of the case.

A satisfactory long-term result was obtained with the fixed vertical palatine crib coupled to the McNamara expander, speech therapy, and ENT, both in the facial (Figure 5) and occlusal aspects (Figure 6). The expander also provided space for the alignment of permanent maxillary anterior teeth, while the use of the vertical crib associated with speech therapy has contributed to the interception of tongue thrust and correction of atypical swallowing.

The lower lingual holding arch showed a benefic effect on the maintenance of the perimeter of the lower arch, which was a concern due to loss of primary teeth, however, it enabled the eruption of all permanent teeth (Figure 6), and the obtaining of positive overjet of about 2mm, and overbite of 3mm.

Figure 5. Final extraoral photographs. A: Front; B: Front smiling; C: Right side

The final panoramic radiograph did not show any periacopathy or root resorption. It was also possible to observe the germs of the lower third molars in an advanced stage of rhizogenesis (Figure 7A).

In the cephalometric analysis (Table 1), the bone bases kept in a good relation with each other (ANB=2º), and the facial pattern remained mesofacial (SN.GoGn =33 º ; SNGn = 69 º ; Facial Axis = 91 º). The maxillary incisors (1.NA= 24º; 1-NA= 4 mm) and mandibular incisors (1.NB = 23º; NB-1 = 4 mm) were well positionated in relation to their apical base with a good interincisal relationship (1.1= 130º). The integumentary profile was considered convex (S-Ls = 2mm/ S-Li = -1 mm), and the bone profile straight (NA.Pog = 4º). It was also possible to verify that adenoid lymphoid tissue involuted, resulting in a wider space in the upper airway.

After the final orthodontic evaluation, it was possible to observe that the clinical case had a satisfactory and stable result after 2 years of intervention, requiring corrective orthodontics only for refinement of the case.

The prognosis due to the patient's age and the facial pattern was considered favorable. Furthermore, early correction provides stimuli that provide adequate craniofacial growth.

Figure 6. Final intraoral photos. A: Front; B: Left side; C: Right side; D: Upper occlusal; E: Inferior occlusal

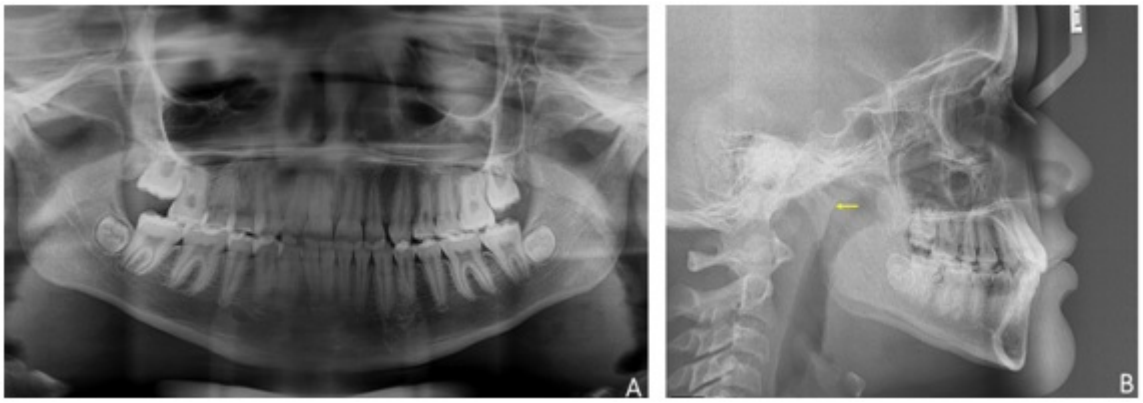

Figure 7. A: Final panoramic radiograph; B: Final lateral cephalometric radiograph showing adenoid tissue regression

DISCUSSION

Although the prevalence of AOB tends to decrease in patients in the mixed dentition[8] as a result of predisposition to cease adopting deleterious oral habits, it is reported that the later the malocclusion is intervened, the chances of success and long-term stability decrease.[14]

The morphological configuration of the AOB presented by the reported patient showed a diffuse and narrow shape, not consistent with the interposition of a finger or a pacifier. Furthermore, anamnesis helped us to rule out the hypothesis of a prior non-nutritive sucking habit. As the cephalometric findings indicated that the patient had a mesofacial skeletal pattern (SN.GoGn=33º ; SNGn=69º ; Facial Axis=91 º), there was no skeletal predisposition to the onset of malocclusion, since there was no tendency of counterclockwise rotation of the palatal plane.[15,16]

The functional examination was essential to identify the tongue as the etiological agent of this AOB, since when swallowing, the patient protruded the tongue anteriorly to prevent the escape of liquid and, thus, allow the swallowing to occur.

The study by Gutiérrez et al.[17] showed that patients with AOB are more likely to present constriction of the anterior portion of the maxilla, as well as a smaller intercanine distance and deep palate. The authors attributed these conditions to the lowered positioning of the tongue during rest, which would cause an imbalance of forces

between the tongue and the cheek muscles, leading to a lack of transverse stimulation of the maxilla.[17] This lowered position of the tongue has also been correlated with an increase in the perimeter of the mandibular arch, as the teeth undergo its expansive pressure.[17]

Although our patient suffered a premature loss of tooth 84 and the E-space[18,19] was still preserved, some space gain in the arch could also have occurred as a result of the lingual pressure, since it is clinically perceived that there was a slight loss of space by the mesialization of tooth 85, which was rotated (Figure 2E), but teeth 44 erupted without hindrance.

Although it is still controversial in the literature if the unusual position of the tongue comes as a result of a pre-existing condition or as the primary factor[20], our patient presented a suggestive radiographic finding of hypertrophy of the adenoids (Fig. 3B), which could be leading to the mouth breathing. However, as the x-ray does not provide a three-dimensional visualization of the tissue volume, the patient was referred to the ENT, which carried out the nasal endoscopy for diagnosis.[21,22] Although the adenoid suffers physiological hypertrophy up to approximately 7 years of age[22,23], treatment with topical corticosteroids was used for regression of the lymphoid tissue[24] (Figure 7B), as there were already evident craniofacial alterations, such as maxillary constriction.

In the maxillary arch, considering the narrow palate and the bone-tooth negative discrepancy presented by the patient, it was decided to proceed with RME. This procedure would provide, in addition to a gain in the arch perimeter[25], space for the tongue to be correctly accommodated on the palate, providing a character of normality in its positioning.

The removable cribs can be less effective than fixed by their dependence on the collaboration of the patient[8], for this reason, and considering that the patient had reasonable oral health, despite the premature loss of two elements in the mandibular arch, the use of fixed appliance was the choice performed in the treatment plan.

The use of the McNamara expander type was given the advantage it provides, such as the ease of cleaning, not occurring irritation tissue due to the interposition of food between the palate and acrylic, as can occur more often with the Haas expander, and by the fact that the acrylic resin in the posterior region works as one bite block, preventing the extrusion the posterior maxillary teeth[26], which would lead to an increase in AOB.

The AOB originated from deleterious oral habits only cease after the removal of these factors[27] , for this reason, the expander was modified with the addition of a vertical palatal crib, given they are devices very effective in trapping of AOB and that can restore the overbite up to 3mm [8].

It is worth noting that the momentary remission of the habit in virtue of the use of the appliance does not ensure the stability of correction, since it is necessary relearning the proper positioning of the tongue during swallowing and rest.[26] Therefore, the speech therapy monitoring of the patient is essential for the success of the treatment.

CONCLUSIONS

It is concluded that the correct diagnosis based on the elimination of confounding etiological factors provides the necessary basis for carrying out the treatment plan for AOB based on tongue thrust origin. However, only the multidisciplinary treatment between speech therapy, otolaryngology, and the dentist will guarantee the success of the treatment. The rapid maxillary expansion through the McNamara expander associated with the fixed palatal crib is an adequate option for both the correction of anterior open bite and narrow palate, which ultimately favors the treatment and stability of the correction.

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