

Speech-language-hearing therapists' perception of orofacial myofunctional changes in subjects with congenital Zika syndrome

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ABSTRACT

Purpose: to investigate the profile of speech-language-hearing therapists from Northeastern Brazil assisting children presented with congenital Zika syndrome and their perception of these children's orofacial myofunctional aspects and speech-language-hearing intervention procedures.

Methods: a cross-sectional observational study with quantitative/qualitative analysis. The data collection instrument was an online questionnaire with 34 items on the speech-language-hearing therapists' sociodemographic information and perception of the study topic. The sample comprised 23 speech-language-hearing therapists from the nine Northeastern states.

Results: altogether, 96% of the sample were females. Most professionals had more than 3 years of experience with congenital Zika syndrome. Concerning stomatognathic system characterization, the professionals described a greater degree of change in aspects of orofacial muscle posture and mobility. Regarding oral functions, they perceived greater changes in speech and swallowing liquids and solid foods. There was a greater degree of change in masticating solids than in swallowing liquids.

Conclusion: most speech-language-hearing therapists in the research had a specialization degree, especially in dysphagia and oral-motor control. Most of them had more than 3 years of experience following up with children with congenital Zika syndrome, which indicates a satisfactory time working with this population to help identify orofacial myofunctional changes in them. The professionals stated that orofacial muscle posture and mobility, speech, and swallowing liquids and solid foods were the most changed aspects.

Keywords: Child; Zika Virus; Microcephaly; Speech, Language and Hearing Sciences

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INTRODUCTION

In the second half of 2015, especially in Northeastern Brazil, there was an increase in the number of children born with microcephaly¹. This is a neurological condition in which the brain is underdeveloped, and the fontanels close prematurely, resulting in a smaller head than those of other children of the same age and sex. Since then, various studies have established a relationship between microcephaly and congenital Zika virus (ZIKV) infection²⁻⁴.

Most recorded characteristics of these cases of microcephaly were severe, with an important craniofacial disproportion. Moreover, microcephaly was not the only consequence, as neurological abnormalities were also observed, such as severe global hyper-tonia with hyporeflexia, irritability, excessive crying, swallowing disorder, and impaired auditory and visual responses²⁻⁵.

Neurological damage and abnormal neuropsychomotor development were also detected even with normal head circumferences. Hence, since microcephaly is not present in all cases, the name congenital Zika syndrome (CZS) is suggested for the new condition caused by such virus infection during pregnancy⁶⁻⁸.

Health professionals had to aid children with this new syndrome using the knowledge they had while recognizing the importance of making efforts to better learn its progress and their therapeutic needs. Thus, research focused on prevention (such as fighting the virus vector mosquito), assessment, and therapeutic intervention in this population.

Speech-language-hearing (SLH) therapists are among the professionals indicated by the Ministry of Health to follow up with these children, especially regarding their orofacial structures, swallowing and its disorders, language, and hearing – which are important aspects to their survival, growth, and development⁹.

According to the National Plan to Address Microcephaly, set by the Ministry of Health⁹, orofacial structures should be assessed by SLH therapists, considering their structural and functional dimensions, and indispensably assessing their breathing and swallowing skills and functions⁹.

The importance of orofacial structures to these functions makes it relevant to find the most affected aspects from the perspective of professionals who follow up on children with CZS in their clinical practice. They can provide parameters to be compared with those of other studies, describing aspects, and guiding professionals in the therapeutic process. Therefore,

this research aimed to investigate the profile of SLH therapists from Northeastern Brazil who treat children with CZS and the perception of these professionals on orofacial myofunctional aspects and SLH intervention procedures.

METHODS

This research is part of a larger project named “SLH Assessment and Intervention in Children with CZS”, approved by the Research Ethics Committee of the Department of Health Sciences at *Universidade Federal da Paraíba* (UFPB), Brazil, under evaluation report number 4.101.795 and CAAE number 32451820.0.0000.5188.

This is a cross-sectional observational study with quantitative/qualitative analysis, whose sample comprised 23 SLH therapists. The data collection instrument, developed by the researchers, was an online questionnaire on the profile of the SLH therapists and the orofacial characteristics and functions of children with CZS. The Northeast Region was chosen because of its greater incidence of CZS cases. The questionnaire could be filled out between August 3 and October 9, 2020, on Google forms, made available through a link sent via social media (e.g., Facebook, Instagram, and WhatsApp) or e-mail to SLH therapists who were interested in participating, and worked directly with the population in question. It had 34 subjective and objective (multiple choice) questions and was divided into two parts.

The first one aimed to characterize SLH professionals, analyzing the following data:

- Sex: the options were male and female.
- Age: recorded by the date of birth.
- State of residence: the options were the states in Northeastern Brazil.
- Data on professional training: the highest educational attainment, the year when they obtained their bachelor's degree, main specialization (all those recognized by the Federal SLH Council), and qualification and training to follow up children with CZS.
- Professional experience: time working with children with CZS (up to 1 year, from 1 to 3 years, or more than 3 years), therapeutic resources (kinesiology tape, laser, electrostimulation, and others), and their frequency of use.

The second part of the questionnaire focused on the professionals' perception of stomatognathic system characteristics, represented in degrees on a scale

from 1 to 10 – in which 1 was the lowest and 10, the highest degree of change – regarding the frequency of pulmonary complications and changes in their teeth, posture, mobility, tonus, intraoral and extraoral sensitivity, breathing, swallowing (solid, semisolid, and pureed foods, and liquids), mastication, and speech.

The inclusion criteria were as follows: being a Brazilian SLH therapist, living in the Northeast Region, and currently working in the rehabilitation of children with CZS. Evidently duplicated questionnaires (which had been previously answered by the same subject) and those with incomplete answers were excluded from the sample.

All participants read the informed consent form and agreed to participate voluntarily in the research.

The results were presented in tables and figures. Categorical variables were presented as absolute and

relative frequencies, and the quantitative ones, as measures of position (mean) and dispersion (standard deviation). The degree of oral-motor and function changes was also presented in figures (boxplots) with information such as the median and minimum, maximum, and discrepant values.

RESULTS

After analyzing the inclusion and exclusion criteria, 15 questionnaires were excluded – five due to evident duplication, nine for being filled out by professionals who were not working with children with CZS at the time, and one for being incomplete.

The results presented below are related to the distribution of SLH professionals who work with children with CZS in the nine Northeastern states (Table 1).

Table 1. Distribution of speech-language-hearing therapists per state where they worked

State	N	%
Maranhão	1	4.3
Piauí	2	8.7
Ceará	2	8.7
Rio Grande do Norte	2	8.7
Pernambuco	4	17.4
Paraíba	7	30.4
Sergipe	1	4.3
Alagoas	1	4.3
Bahia	3	13.0

Source: The authors, 2021

Captions: N – number; % - percentage.

Altogether, 96% of the sample of SLH therapists were women, and their mean age was 35.56 years (± 7.99). The mean time since their graduation was 10.60 years (± 6.30).

Table 2 shows the characterization of SLH professionals in the research.

Table 2. Characterization of the speech-language-hearing therapists in the research

Categorical variables	N	%
Educational attainment		
Bachelor's degree	4	17
Specialization degree	13	57
Master's degree	6	26
Doctor's degree	0	0
Specialization degree		
None	6	26
Oral-motor control	5	22
Voice	1	4
Public health	3	13
Dysphagia	6	26
Neuropsychology	1	4
Neurofunction	1	4
Experience with CZS		
Up to 1 year	4	17
1 – 3 years	8	35
More than 3 years	11	48
Qualification for practice		
None	8	35
Course	10	43
Conference	1	4
Bobath concept	1	4
Residence	1	4
Not reported	2	9

Source: The authors, 2021

Captions: CZS – congenital Zika syndrome; N – number; % - percentage.

Table 3 presents the degree of change the SLH therapists ascribed to each orofacial aspect and oral function. They were asked to fill out a scale from 1 to

10, in which 1 corresponded to the lowest and 10, to the highest degree of change.

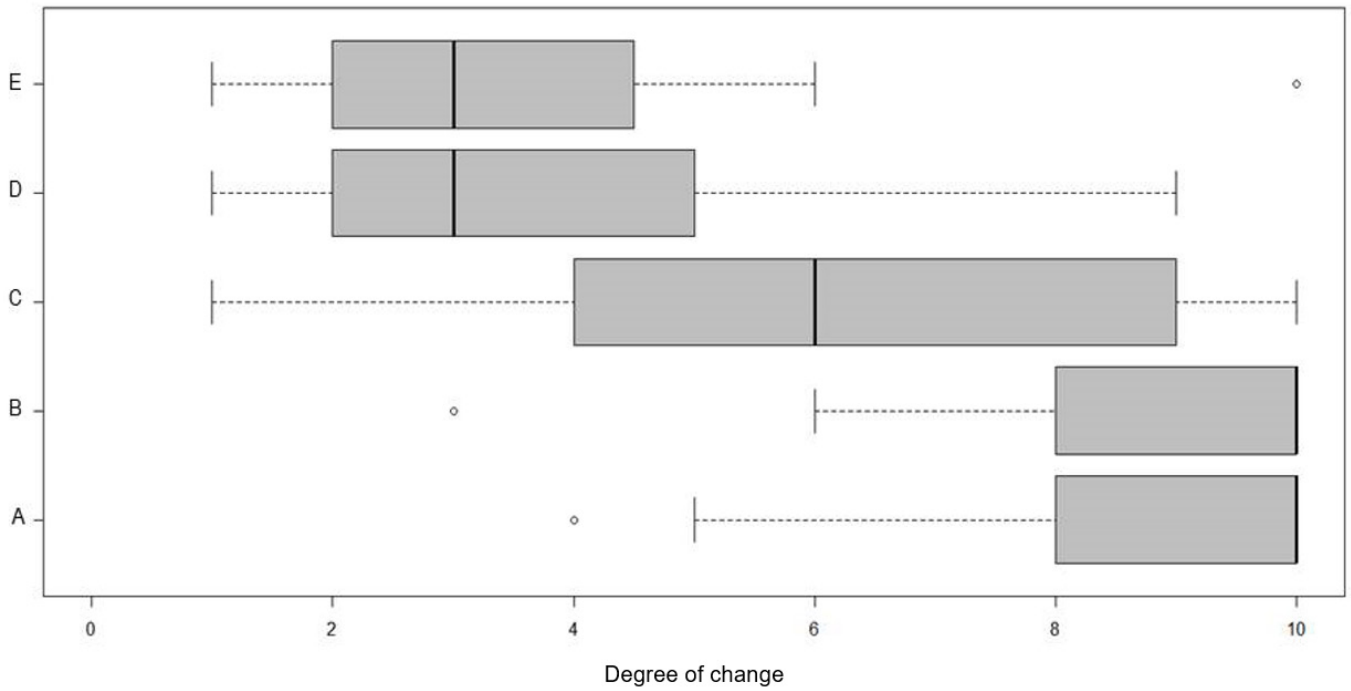
Table 3. Reported degrees of change in structures and oral functions

Structures	Degree of change
Dental changes	7.26 (\pm 2.22)
Posture	8.82 (\pm 1.77)
Mobility	8.78 (\pm 1.80)
Muscle tonus	5.82 (\pm 3.02)
Intraoral sensitivity	3.52 (\pm 2.31)
Extraoral sensitivity	3.60 (\pm 2.48)
Functions	
Breathing	7.39 (\pm 2.16)
Sucking/swallowing	8.21 (\pm 1.41)
Mastication	8.78 (\pm 1.59)
Swallowing solid foods	9.00 (\pm 1.85)
Swallowing semisolid foods	7.95 (\pm 1.84)
Swallowing pureed foods	6.34 (\pm 1.69)
Swallowing liquids	8.26 (\pm 2.19)
Speech	9.47 (\pm 1.87)

Source: The authors, 2021

Figures 1 and 2 present information (median and minimum, maximum, and discrepant values) on the

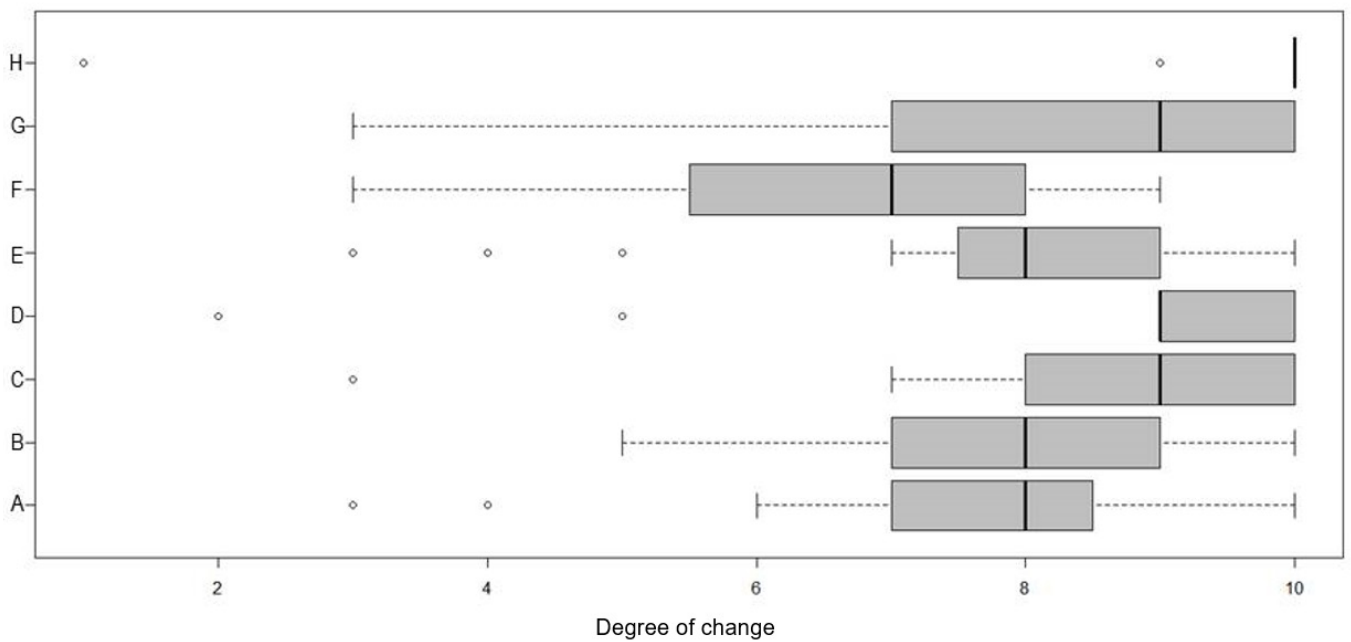
degree of change respectively of stomatognathic system structures and oral functions.



Source: The authors, 2021.

Captions: A) Posture; B) Mobility; C) Tonus; D) Intraoral sensitivity; E) Extraoral sensitivity.

Figure 1. Degree of change in aspects of posture, mobility, tonus, and intraoral and extraoral sensitivity



Source: The authors, 2021.

Captions: A) Breathing; B) Sucking/swallowing; C) Mastication; D) Swallowing solid foods; E) Swallowing semisolid foods; F) Swallowing pureed foods; G) Swallowing liquids; H) Speech.

Figure 2. Degree of change in oral functions

The professionals were also asked about the frequency of pulmonary complications in the previous year, to which the mean answer was 5.08 (± 2.77).

The most recurrent answer about the frequency of visits was once a week, while the mean was 1.78 (± 1.20).

Regarding SLH intervention, 4% reported using only myofunctional therapy, 17% most of the time used only

myofunctional therapy, and 78% used technological resources along with myofunctional therapy.

Kinesiology tape was used by 87%, whereas only 9% used laser and electrostimulation. Table 4 shows the absolute and relative frequency of the use of these strategies/resources in therapy. As for other strategies, 24% reported using tactile thermal oral stimulation.

Table 4. Therapeutic procedures used by the speech-language-hearing therapists

Therapeutic procedures	N	%
Kinesiology tape		
4 - 8 times/week	12	52
9 - 15 times/week	4	17
16 - 20 times/week	4	17
Not used	3	13
Laser		
4 - 8 times/week	0	0
9 - 15 times/week	1	4
16 - 20 times/week	1	4
Not used	21	91
Electrostimulation		
4 - 8 times/week	1	4
9 - 15 times/week	1	4
16 - 20 times/week	0	0
Not used	21	91

Source: The authors, 2021

Captions: N – number; % - percentage.

DISCUSSION

Most participating SLH therapists were from the states of Paraíba, Pernambuco, and Bahia, in this order. This can be explained by the greater occurrence of CZS in these Northeastern states¹⁰, likely requiring more SLH therapists to follow up on these children.

The sudden appearance of a health condition with peculiar characteristics led to new demands for these children's rehabilitation in a rather short time, which were beyond the professional skills used until then¹¹. This study observed the importance of training, as these professionals are responsible for planning procedures and following up with children with CZS. Nonetheless, the results showed that 35% of them had not received any training to address the new syndrome, which may directly interfere with procedures and decision-making in such children's rehabilitation, given the unfamiliarity with its characteristics. As for those who received training, 43% reported having taken

some type of course, mostly focusing on early stimulation – i.e., to follow with 0-to-3-year-old children with CZS. It is greatly important to continuously follow up on these children's orofacial development and assess the impact of such changes in their stomatognathic system as they grow up¹².

The results of this research show that most SLH therapists had a specialization degree, while 26% had a master's degree. Most specializations were in the areas of dysphagia and oral-motor function – both related to orofacial myofunctional changes, which are prevalent in CZS. Children with this syndrome have eating difficulties – 51.28% choke when they eat¹³, and dysphagia occurs simultaneously with orofacial myofunctional disorder¹⁴. Hence, they need qualified assistance from the SLH therapists who provide care to this group.

Most SLH therapists reported more than 3 years of experience following up on children with CZS. Hence, it can be stated that they have had not only significant

contact with these children but also knowledge of their current changes.

Regarding stomatognathic system characterization, this study found the greatest change in orofacial muscle mobility and posture. Another study confirmed the occurrence of changes in the posture of children with microcephaly – concerning the mouth posture at rest (lips and tongue), they found that 46% of patients had closed lips, 52% had parted lips, and 2% had open lips; also, 54% had a flat tongue, and 41% had a raised tongue¹⁴.

The high frequency of dental changes was another relevant aspect of the present research. Other studies likewise found such high frequencies and emphasized the need to draw the attention of dental surgeons and other health team members to treat the most frequent mouth problems in these patients^{15,16}. Children with CZS are more prone to the delayed eruption of teeth, inadequate tongue posture, and short lip and tongue frenula¹⁷.

The professionals in this research reported that speech was the most impaired function. According to the American Speech-Language-Hearing Association (ASHA), about one in every 200 people is unable to communicate orally due to cognitive, physical, or neurological factors¹⁸. The relationship between ZIKV and neurological changes is evident^{19,20}, identifying both calcifications and other neurological changes that damage the neurons, which may delay their neurological development². Thus, regarding development, children with CZS may have severe delays, being likely to have limited skills and require continuous care²¹.

The central nervous system regulates functions necessary to eating, such as mastication, swallowing, and breathing; hence, once it is affected, impairments to these functions manifest in the development of children with ZIKV. Therefore, adequate prospective multidisciplinary follow-up of these patients aims to understand the natural history of this new agent and ensure better development and quality of life for the children and their families²².

Due to neurological changes, swallowing disorders are quite frequent²³. According to instrumental assessment, eight out of nine babies in a research sample did not have sensitivity in the mouth or upper airway, delaying the beginning of the pharyngeal phase of swallowing. Consequently, a rather big risk of food aspiration (especially liquids) was identified in combination with severe oral dysfunction²⁴. Babies with microcephaly have considerably affected swallowing

and breathing and impaired muscle tonus and dental eruption²⁵. The present research observed, from the professionals' perspective, significant changes in the swallowing of liquids and solid foods.

Currently, different areas of rehabilitation can count on various resources to aid the therapeutic process, ensuring more effective and lasting results. The therapeutic resource most used in this study was the kinesiology tape, followed by tactile thermal oral stimulation.

Rehabilitation services and professionals who follow up on children with disabilities must get organized to use new interventions for children with CZS and their families. Even though these strategies are not often used, the findings point to advancements in therapies for children with microcephaly⁴.

Given the findings in this research, further studies are suggested to assess the orofacial structures and stomatognathic functions of children with CZS – especially comparing them with healthy children and revealing the effectiveness of SLH treatment for these children. Concerning the limitations of conducting this study, attention is called to the need for a register of professionals who follow up on these children and a register of children assisted by SLH therapists at the Unified Health System (SUS).

CONCLUSION

Most professionals who participated in this research were specialists, particularly in dysphagia and oral-motor control, and had more than 3 years of experience in assisting children presented with CZS. In the perception of these SLH therapists, orofacial muscle posture and mobility are the most impaired aspects, as well as speech and swallowing liquids and solid foods.

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MLV: participated in the data curation, investigation, methodology, writing original draft, writing review, and editing;

LAP and RLCM: participated in the formal analysis, methodology, writing, review and editing;

GASA: participated in the conceptualization, data curation, formal analysis, funding acquisition, investigation, methodology, project administration, supervision, visualization, writing original draft, review, and editing.