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Myofunctional orofacial findings in children with microcephaly

Achados miofuncionais orofaciais em crianças com microcefalia

Hallazgos Miofuncionales Orofaciales en Niños con Microcefalia

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Abstract

Objective: To identify the main orofacial myofunctional changes in patients who have microcephaly and describe the predominant speech-language syndromic hypotheses. **Methods:** This is a retrospective, cross-sectional and descriptive study. The sample consisted of 46 medical records of children with microcephaly in the age group from zero to seven months attended at the Specialized Center for Rehabilitation of Maceió in 2016, disregarding the records that did not have a defined etiological diagnosis of microcephaly or did not present evaluative data. The data were treated statistically using the Chi-square test, with a significance level less than or equal to 5%. **Results:** Considering the correlation between orofacial structures and oral reflexes; orofacial structures and syndromic speech-language diagnostic hypotheses; oral reflexes and syndromic speech-language diagnostic hypotheses and orofacial structures and type of food, there was significance for the first three correlations, which can be inferred that structural changes have repercussions on the development and functionality of the stomatoglossognathic system. **Conclusion:** It is noticed that the orofacial myofunctional changes in children with microcephaly include structural changes (lips parted at rest and anterior insertion of the lingual frenulum) with a predominance of Orofacial Myofunctional Disorder and changes in oral reflexes; in addition, dysphagia and Orofacial Myofunctional Disorder occurred simultaneously. Exclusive breastfeeding occurred only in children without changes in orofacial structures.

Keywords: Microcephaly; Diagnosis; Child; Speech, Language and Hearing Sciences.

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JBCF - Study design; data collect; outline of the article.
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Resumo

Objetivo: Identificar as principais alterações miofuncionais orofaciais dos pacientes que possuem microcefalia e descrever as hipóteses sindrômicas fonoaudiológicas predominantes. **Métodos:** Trata-se de um estudo retrospectivo, de corte transversal e de caráter descritivo. A amostra foi de 46 prontuários de crianças com microcefalia na faixa etária de zero a sete meses atendidas no Centro Especializado em Reabilitação de Maceió no ano de 2016, desconsiderando-se os prontuários que não possuíam diagnóstico etiológico definido de microcefalia ou não apresentaram dados avaliativos. Os dados foram tratados estatisticamente por meio do teste Qui-quadrado, com nível de significância menor ou igual a 5%. **Resultados:** Considerando-se a correlação entre estruturas orofaciais e reflexos orais; estruturas orofaciais e hipóteses diagnósticas fonoaudiológicas sindrômicas; reflexos orais e hipóteses diagnósticas fonoaudiológicas sindrômicas e estruturas orofaciais e tipo de alimentação, observou-se significância para as três primeiras correlações, o que se pode inferir que as alterações estruturais repercutem no desenvolvimento e funcionalidade do sistema estomatoglossognático. **Conclusão:** Percebe-se que as alterações miofuncionais orofaciais em crianças com microcefalia englobam alterações estruturais com predomínio do Distúrbio Miofuncional Orofacial e alterações dos reflexos orais; além destes, houve ocorrência simultânea de disfagia e Distúrbio Miofuncional Orofacial. O aleitamento materno exclusivo ocorreu apenas em crianças sem alterações das estruturas orofaciais.

Palavras chave: Microcefalia; Diagnóstico; Criança; Fonoaudiologia.

Resumen

Objetivo: identificar las principales alteraciones miofuncionales orofaciales de los pacientes que poseen microcefalia y describir las hipótesis sindrómicas fonoaudiológicas predominantes. **Métodos:** consiste en un estudio retrospectivo, de corte transversal y carácter descriptivo. La muestra consistió en 46 prontuarios de niños de cero a siete años atendidos con microcefalia en el Centro Especializado en Rehabilitación de Maceió, en el año 2016, desconsiderándose los prontuarios que no especificaran diagnóstico etiológico definido de microcefalia o que no presentaran datos evaluativos. Los datos sí trataron estadísticamente mediante la prueba de chi-cuadrado, con un nivel de significancia menor o igual al 5%.**Resultados:** considerándose la correlación entre estructuras orofaciales y reflejos orales; estructuras orofaciales e hipótesis diagnósticas fonoaudiológicas sindrómicas, reflejos orales e hipótesis diagnósticas fonoaudiológicas sindrómicas e estructuras orofaciales y medio de alimentación, se observó significancia para las tres primeras correlaciones, por lo que se puede afirmar que las alteraciones estructurales están relacionadas con el desarrollo y funcionalidad del sistema estomatoglosognático. **Conclusión:** se observa que los cambios miofuncionales orofaciales en niños con microcefalia incluyen cambios estructurales con predominio del trastorno miofuncional orofacial y cambios en los reflejos orales, donde hubo una ocurrencia simultánea de disfagia y trastorno miofuncional orofacial. La lactancia materna exclusiva se produjo solo en niños sin cambios en las estructuras orofaciales.

Palabras clave: Microcefalia; Diagnóstico; Niño; Fonoaudiología.

Introduction

Microcephaly is a pathology of neurological origin, in which there is a reduction in head circumference as a consequence of a decrease in the thickness of the cerebral cortex associated with malformations or other possible significant neurological changes. The diameter considered standard would be 42 cm, according to WHO (World Health Organization), so the child has microcephaly when the head circumference is less than two or more standard deviations than the parameter for age or gestational time and sex.[1-4]

It can be classified as congenital microcephaly, which is present at birth and postnatally, and is related to changes in the normal growth of the head circumference after birth, that is, the brain is normal at birth.[5]

The neurological condition can be seen during routine prenatal examinations, among which stands

out the pregnant woman's obstetric ultrasound and computed tomography, at birth, through physical examination, after delivery, through the Apgar test - which consists of assessing the newborn's vitality condition and involves: heart rate, reflex response, skin color, muscle tone; in addition, congenital infections, such as meningitis and meningoencephalitis, can also be an etiology of microcephaly.[5,6]

The parameters related to height, baby weight and head circumference are also detected in the exams. The child diagnosed with microcephaly may have delayed neuropsychomotor development, some degree of mental retardation, as well as visual and auditory changes.[6]

Some changes present in microcephaly are related to Orofacial Motricity (MO). The MO consists of a field of Speech Therapy that studies the interventions of the structural and functional aspects of the orofacial and cervical regions, as well as the anatomy and physiology of these structures, enabling an effective performance through understanding the proper development of stomatoglossognathic functions and facial mimicry.[7]

Children who have the affected neurological system are predisposed to changes related to orofacial motricity, so it is necessary to evaluate structures of the stomatoglossognathic system and their functions, highlighting, in this study, mobility, posture and tonus of the tongue and the classic functions of breathing, sucking and swallowing, encompassing the child's feeding process through breastfeeding[2,8] and / or complementation, for better perception and knowledge of the changes. This assessment is very relevant, because if these aspects are developing improperly, the baby is at risk of complications such as aspiration pneumonia, weight loss, malnutrition, dehydration, characterizing dysphagia, due to changes in the phases of swallowing.[9,10]

It was found that from the early stimulation of babies born with microcephaly it is possible to promote the harmony of development between the various functional organic systems, motor, sensory, perceptual, proprioceptive, linguistic, cognitive, emotional and social areas, dependent or not on maturation of the Central Nervous System (CNS), that is, this stimulation will provide a balance in the evolution of this individual and this may or may not be related to the CNS maturation.[10]

Microcephaly is a very compromising alteration for the individual, even so there is a lack in literature regarding this theme in relation to speech therapy aspects. The way professionals can act in the face of the patient's difficulties needs to be further discussed in order to favor an effective treatment and a better quality of life for these individuals.

Through this study, we intend to find changes regarding the phonoarticulatory organs and their functions. The analysis aims to identify the main orofacial myofunctional changes in patients who have microcephaly and describe the predominant speech-language syndromic hypotheses.

Methods

This article was approved by the Research Ethics Committee (CEP) of the Alagoas State University of Health Sciences (UNCISAL) under CAAE: 58444516.0.0000.5011 and number: 2,103,441.

This is a retrospective, cross-sectional and descriptive study. It was carried out by analyzing the medical records of subjects with microcephaly, aged between zero and seven months, attended at a Specialized Rehabilitation Center in Maceió city, during 2016. Sampling was established for convenience considering the presence of diagnosed Microcephaly by a neurologist through image exams such as Transfontanela ultrasonography, skull tomography and head circumference measurement.

As inclusion criteria, we considered the medical records of patients, whose parents accepted to participate in the research, which had microcephaly and were seen in the area of Orofacial Motricity, in 2016, at the Assistance Unit of the Specialized Center for Rehabilitation III of the University. Regarding the exclusion criteria, the medical records of patients who did not have a defined medical etiological diagnosis of microcephaly or died before the beginning of the research were disregarded.

The speech-language syndromic diagnostic hypothesis was referred to as the primary variable and the occurrence of orofacial structural and myofunctional changes in microcephalic patients as a secondary variable.

Data were collected from medical records using the structured extraction form from an instrument developed by the researchers, with which data were obtained regarding sex, age group, state of behavioral organization (state of consciousness,

posture and global tone), posture oral (lips and tongue), oral reflexes (looking, sucking, biting and vomiting) and type of feeding (exclusive breastfeeding, artificial or complementary feeding). To observe such items, data related to the structures and functionality of the face and oral cavity were extracted from the medical records.

To cover knowledge about myofunctional changes in children with microcephaly, items essential to the description of the structures of the stomatoglossognathic system and their functions were related, considering the anatomofunctional aspects, among them: orofacial structures and oral reflexes (Table 1); orofacial structures and syndromic speech-language diagnostic hypotheses (Table 2); oral reflexes and syndromic speech-language diagnostic hypotheses (Table 3) and orofacial changes and type of child feeding (Table 4).

After collection, the information was tabulated in the Excel 2016 program and analyzed using the Chi-square test in the Bioestat 5.3 program, considering a significance level of p value <0.05. This test analyzes nominal and ordinal variables from independent samples, in order to verify whether there is significance or not.[11,12]

Results

It was not possible to analyze all medical records due to the lack of information in the evaluated protocols of the service, justified by factors of referral of the patient to other services, difficulty in completing the assessment related to crying and irritation of the child or death of the individual. Thus, the final sample was established through 46 medical records.

In view of the medical records analyzed, through the evaluation data described, a higher prevalence of males was perceived, with 52.2% (N = 24) and 47.8% (N = 22) of females, aged between six and seven months. Of these, 22% (N = 10) were on exclusive breastfeeding and 78% (N = 36) on supplemented / mixed food with formula milk and / or other foods of a more solid consistency.

In the clinical aspect, considering oral posture at rest (lips and tongue), it was found that 46% (N = 21) of the patients had sealed lips, 52% (N = 24) lips parted and 2% (N = 1) of the patients demonstrated open lips posture. Regarding the tongue posture, 54% (N = 25) of the medical records contained a flat tongue, that is, a higher prevalence of normality, 41% (N = 19) with elevated posture, 2% (N = 1) with a retracted tongue and protruded.

Considering other structures of the stomatoglossognathic system, normal insertion of the frenulum was observed in 44% (N = 20), anterior insertion 35% (N = 16), short frenulum 6% (N = 3) and in 15% (N = 7) the medical records did not contain enough information to identify this aspect. Regarding the mandible, there was 59% (N = 27) of age-appropriate anatomical growth and 24% (N = 11) was inadequate, 17% (N = 8) of the medical records did not present this data in the evaluations. The maxilla showed 61% (N = 28) of age-appropriate anatomical growth, 22% (N = 10) inadequate and 17% (N = 8) of the patients did not have such information described in their medical records. Regarding the hard palate, it showed 43% (N = 20) without alteration, 28% (N = 13) ogival, 4% (N = 2) deep and 25% (N = 11) of the patients did not have enough evaluative information about this structure.

Relating the orofacial structures and the oral reflexes, 72% (N = 33) of altered reflexes were determined in children who had structural changes, while 28% (N = 13) demonstrated presence of oral reflexes, despite the altered structures. Constant choking and neurogenic dysphagia were noted in 4% (N = 2) of patients who had structural impairment of the stomatoglossognathic system.

Table 1. Description of orofacial structures and oral reflexes in microcephaly

Oral reflexes**	Orofacial Structures*		p-value***
	Normal	Altered	
	n (%)	n (%)	
Present	33 (72%)	13 (28%)	*<0,0001*
Absent	13 (28%)	33 (72%)	

Subtitle: n – number;
* Tongue, lingual frenulum, lips, palate, maxilla and mandible.
** Demand reflexes, sucking, swallowing, GAG, bite;
Test: ***Qui-scare, p-value ≤0,05

In the correlation between orofacial structures and speech-language diagnostic hypotheses, a statistically significant relationship was identified with regard to the association of the presence of orofacial structural changes and Orofacial Myofunctional Disorder (OMD) (p-value 0.0001).

Table 2. Description of orofacial structures and speech-language diagnostic hypotheses in microcephaly

HDF	Orofacial structures *		p-value**
	Normal	Altered	
	n (%)	n (%)	
Dysphagia	44(96%)	2 (4%)	
OMD	14 (30%)	32 (70%)	*0,0001*
Dysphagia and OMD	20 (43%)	26 (57%)	

Subtitle: N – number; HDF – Speech Therapy Diagnostic Hypotheses; OMD – Orofacial Myofunctional Disorder.
* Tongue, lingual frenulum, lips, palate, maxilla and mandible.
Test: ***Qui-scare, p-value ≤0,05

The association between oral reflexes and syndromic speech therapy diagnostic hypotheses demonstrated statistical significance (p-value 0.0001). It was found that 63% (N = 29) of the analyzed children had altered oral reflexes with syndromic characterization of the simultaneous occurrence of dysphagia and OMD.

Table 3. Description of oral reflexes and diagnostic speech-language hypotheses in microcephaly

HDF	Oral reflexes*		p-value**
	Normal	Altered	
	n (%)	n (%)	
Dysphagia	44(96%)	2(4%)	
OMD	33(72%)	13(28%)	*0,0001*
Dysphagia and OMD	17(37%)	29(63%)	

Subtitle: N – number; HDF – Speech Therapy Diagnostic Hypotheses; OMD – Orofacial Myofunctional Disorder.
* Tongue, lingual frenulum, lips, palate, maxilla and mandible.
Test: ***Qui-scare, p-value ≤0,05

Regarding the orofacial structures and the type of feeding, there was a predominance of artificial feeding in 43% (N = 20) of the medical records; of these 80% (N = 16) presented structural and functional basis changes (anterior frenulum insertion, tone and mobility of lips and tongue reduced). The lowest index referred to exclusive breastfeeding, presenting 22% (N = 10) of the total sample collected.

Table 4. Description of orofacial structures and type of feeding in microcephaly

Type of feeding	Oral reflexes *		p-value**
	Normal	Altered	
	n (%)	n (%)	
Exclusive breastfeeding	10(22%)	36(78%)	
Artificial feeding	20(43%)	26(57%)	*0.0839*
Complementary feeding	16(35%)	30(65%)	

Subtitle: N – number; *Demand reflexes, sucking, swallowing, GAG, bite;
Test: ***Qui-scare, p-value ≤0,05

In view of the relationship established in Table 4, mentioned above, it was noted that there was no statistical significance between the orofacial structures and the type of food.

Discussion

The incidence of the Zika virus represented a strong impact in Brazil, affecting a large part of the population, including pregnant women who abruptly discovered that the pathology had irreversible consequences for babies, involving neurological syndromes, such as microcephaly[13,14]. Since then, several rehabilitation services have endeavored to cover the needs for therapeutic stimulation, necessary for the more effective development of these children[3,6,15-17], with microcephaly due to Zika virus or other gestational infections.

In this study, it was possible to identify the presence of changes in the stomatoglossognathic system and to verify the most recurrent speech-language diagnostic hypotheses in children with microcephaly. The data allowed to reveal a predominance of the male gender, corroborating with another study that stated that there is a higher prevalence of some type of disability in the male gender in relation to the female.[13,18]

The relationship between structural changes and oral reflexes is highly significant, patients who have changes in the stomatoglossognathic system present concomitantly changes in oral reflexes, with a greater incidence of the absence of these reflexes in individuals who had altered structures. This finding confirms data from another study that described that the complications presented during the neonatal period, and the physiological and neurological maturation deficits, are other factors that can influence weakness or absence of oral reflexes[18].

Another study reported a high prevalence of changes in the development of orofacial structures, mainly of the palate, subsequently impairing the development of oral reflexes in individuals who have microcephaly. However, they associate this fact as a possible consequence to the exposure of the Zika virus, due to the verification of malformations of this structure in the embryonic period, corresponding to the period of infection of the mother[19,20].

Correlating the orofacial structures and the syndromic diagnostic hypotheses of speech therapy, a high incidence of OMD is perceived. A similar factor was found in a study that evaluated orofacial myofunctional changes in children seen at the pediatric clinic of a university hospital in Aracaju/SE, in which the authors observed structural changes such as the absence of a lip seal at rest, and changes in mandibular movements as risk factors for the occurrence of OMD. Thus, it is evident that these changes cause impairment in the functions of the stomatoglossognathic system leading to a myofunctional imbalance.[20,21]

Regarding oral reflexes and diagnostic hypotheses, a higher rate of reflex alterations was noticed when the individual had been affected by OMD concomitant with dysphagia, corroborating the findings of a study that found a preponderance of swallowing changes and inadequacy in swallowing, mobility and posture of the phonoarticulatory organs, in addition to the perception of alteration of the oral reflexes in the face of a neurological pathology[18]. This can reinforce the fact that these reflexes have a primordial influence for the good performance of the functions of the stomatoglossognathic system, including the functional bases of this system, tonicity, mobility and sensitivity, therefore, it can be inferred that these changes determine the presence of syndromic speech-language hypotheses..

Despite the low incidence of oropharyngeal dysphagia, it is important to note that it is quite recurrent in individuals with neurological damage, featuring, among other signs, eating difficulties, which were described in the present study. A study carried out with microcephaly children in Ceará, whose main objective was to analyze the dietary pattern of these children, showed that the majority of the mothers of children with microcephaly reported persistent feeding difficulties, with difficulty in positioning the child in the breast during breastfeeding and the presence of gagging[22]. Consequently, this increases the risk of aspiration pneumonia, due to changes in oral motor control, tonus and oral reflexes, factors that are relevant to the effectiveness of swallowing.[23,24]

According to the data identified in the relationship of orofacial changes and the type of food, it was noticed that artificial food had a higher prevalence of changes. These data match the information found in a study that described the use of the bottle as responsible for developing other muscles that were not prepared for such an act, which can cause disturbances in growth and / or oral breathing, causing changes in the development of the head, face, poor dental positioning, chewing difficulties, behavioral changes and body posture.[25]

A study carried out aiming to identify the anthropometric profile and food consumption of children with microcephaly, pointed out that more than half of the analyzed population had some difficulty in feeding. The individuals had structural alterations, functional bases and functions of the stomatoglossognathic system, including: increase or decrease in muscle tone, facial asymmetry, alterations or absence of oral reflexes, malformation of the orofacial structures and dysphagia. These factors were identified as influencing the determination of the type of food, with preference being given to artificial food with formula and / or other complementary foods[26]. It can be inferred that the anatomical changes of the stomatoglossognathic system have repercussions on the functionality of swallowing and chewing, triggering dysphagia and OMD consequently.

It is worth mentioning that one of the medical records characterized a patient with a complete cleft lip, a lowered general clinical condition, due to the constant use of medications, and submitted to gastrostomy, due to respiratory fatigue during feeding. In this case, it can be suggested that microcephaly is related to syndromic aspects.

There is an evident scarcity of studies with a well-defined methodological design in the literature on microcephaly and speech-language aspects. Therefore, it is suggested to implement new studies in order to expand the scientific knowledge of health professionals, and, especially speech therapists, considering that the demand for patients affected by microcephaly increased significantly after the Zika virus outbreak.

Conclusion

Through this study, it was found that microcephaly causes myofunctional changes in the stomatoglossognathic system, modification of the pattern of oral reflexes and differentiated characterization of speech-language syndromic diagnostic hypotheses, considering structural changes and oral reflexes. The structural changes show a higher incidence of orofacial myofunctional disorder and the change in oral reflexes presented simultaneous occurrence of dysphagia and OMD.

It should be noted that the alterations vary according to the degree of severity of the brain impairment, and it is not prudent to define a pattern of dysfunctions in the stomatoglossognathic system. Thus, the need for an individualized therapeutic plan and adapted to the manifestations presented during the child's development period is reinforced.

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