

Relationship between audiological and phonological awareness findings in a group of children with cleft lip and / or palate

Relação entre os achados audiológicos e de consciência fonológica em um grupo de crianças com fissura labial e/ou palatina

Relación entre hallazgos audiológicos y conciencia fonológica en un grupo de niños con labio leporino y/o paladar hendido

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Abstract

Introduction: Children with hearing loss may present speech impairment, consequently disadvantages in the development of language skills, such as phonological awareness. Individuals with cleft lip and / or palate are predisposed to hearing loss and difficulties in phonological awareness; however, there are few studies that demonstrate correlation of this interrelation. **Objective:** Analyze the audiological and phonological awareness findings in a group of children with cleft lip and palate. **Methods:** Observational cross-sectional study approved by the Ethical Committee in Research under the number 1.974.629, held with a group of 36 children. Inclusion criteria: children with cleft lip and / or palate aged between 4 and 12 years, of both genders, without a diagnosis of associated syndromes. To investigate the history of otitis media, a reminder was applied to those responsible for the children. The Sequential Evaluation Instrument – CONFIAS was used to analyze phonological awareness. To verify the auditory acuity,

Contribuição dos autores:

AFDR: data collection; analysis of data and results; theoretical reference.

LPR: analysis of data and results; theoretical reference.

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the children performed the pure tone audiometry. **Results:** The sample consisted of 13 children, aged between 4 and 8 years. All of them presented losses in the CONFIAS scores, 92.3% in the syllable level and 84.6% in the phoneme level. It was found that 69.2% had a history of otitis media. In audiometry, 77% presented conductive hearing loss. There was no correlation between phonological awareness and auditory findings. **Conclusion:** Children with cleft lip and palate presented phonological awareness difficulties, even though there was no correlation with the audiological findings, the hypothesis of an interrelationship between the variables.

Keywords: Cleft Lip; Cleft Palate; Child Language; Hearing; Otitis Media.

Resumo

Introdução: Crianças com perdas auditivas podem apresentar comprometimento na fala e, consequentemente, desvantagens no desenvolvimento das competências linguísticas, como a consciência fonológica. Indivíduos com fissura labial e/ou palatina apresentam predisposição para perda auditiva e dificuldades de consciência fonológica, no entanto, há poucos estudos que demonstrem correlatos sobre esta inter-relação. Objetivo: Analisar os achados audiológicos e de consciência fonológica em um grupo de crianças com fissura labial e/ou palatina. **Métodos:** Estudo observacional transversal aprovado pelo Comitê de Ética em Pesquisa (1.974.629), realizado com um grupo de 36 crianças com fissura labial e/ ou palatina. Critérios de inclusão: crianças com fissuras labiais e/ou palatinas com idades entre 4 e 12 anos, de ambos os gêneros, sem diagnóstico de síndromes associadas. Para investigar o histórico de otite média foi aplicado um recordatório junto aos responsáveis pelas criancas. A consciência fonológica foi avaliada com o Instrumento de Avaliação Sequencial - CONFIAS. Para averiguar a acuidade auditiva, as crianças realizaram a Audiometria Tonal Liminar. Resultados: A amostra foi composta por 13 crianças, com idades entre 4 e 8 anos. Todas apresentaram prejuízos nos escores do CONFIAS, sendo 92,3% no nível da sílaba e 84,6% no do fonema. Verificou-se que 69,2% tinham histórico de otite média. Na audiometria, 77% apresentaram perda auditiva condutiva. Não houve correlação entre os resultados de consciência fonológica com os achados audiológicos. Conclusão: As crianças com fissura labial e/ou palatina demonstraram dificuldades de consciência fonológica, mesmo não havendo correlação com os achados audiológicos, não se descartou a hipótese de uma inter-relação entre as variáveis.

Palavras-chave: Fenda Labial; Fissura Palatina; Linguagem infantil; Audição; Otite Média.

Resumen

Introducción: Los niños con pérdida auditiva pueden presentar problemas del habla y, en consecuencia, desventajas en el desarrollo de las habilidades del lenguaje, como la conciencia fonológica. Las personas con labio leporino y/o paladar hendido están predispuestas a pérdida de audición y dificultades en la conciencia fonológica, sin embargo, hay pocos estudios que demuestren correlatos sobre esta interrelación. Objectivo: Analizar los hallazgos audiológica y de la conciencia fonológica en un grupo de niños con hendidura del labio y/o paladar hendido. Métodos: Estudio observacional transversal aprobado por el Comité de ética de la investigación (1.974.629), realizado con un grupo de 36 niños con hendidura del labio y/o paladar hendido. Criterios de inclusión: niños con labio leporino y/o paladar hendido entre 4 y 12 años, de ambos sexos, sin diagnóstico de síndromes asociados. Para investigar el historial de otitis media, se aplicó una recordatória a los responsables de los niños.La conciencia fonológica se evaluó utilizando el Instrumento de evaluación secuencial - CONFIAS. Para comprobar la agudeza auditiva, los niños realizan la medida cautelar de la audiometría tonal. Resultados: La muestra consistió en 13 niños, con edades entre 4 y 8 años. Todos presentaron pérdidas en los puntajes de CONFIAS, 92.3% en nivel de sílabas y 84.6% en nivel de fonemas. Se encontró que 69.2% tenía antecedentes de otitis media. En audiometría, 77% tenía pérdida auditiva conductiva. No hubo correlación entre conciencia fonológica y los resultados de las audiometrias. Conclusión: Los niños con labio leporino y/o paladar hendido demostraron dificultades en conciencia fonológica, incluso con ninguna correlación con los hallazgos audiológicos, no si descartó la posibilidad de una relación entre las variables.

Palabras clave: Hendidura de lábio; Paladar hendido; Lenguaje del niño; Audición; Otitis media.



Introduction

Cleft lip and/or cleft palate (CLP) are orofacial congenital malformations with the highest occurrence rates. In Brazil, it is estimated that its prevalence varies between 1 and 2 cases for each 1,000 live birth of newborns¹.

Craniofacial structural alterations of individuals with CLP can result in disorders in the articulation and resonance of verbal sounds. Alterations of this nature, if not treated, will compromise the speech development of the child with CLP and can influence in the phonetic and phonological systems acquisition².

Another factor that might affect the acquisition of speech sounds is the decrease of auditory acuity, which interferes in the organization and categorization process of acoustic information (central auditory processing)³. Children with CLP present predisposition to conductive hearing loss, especially those who had a palatoplasty carried out late and/or those with a history of diseases in the middle ear, which are developed due to the compromise of the velopharyngeal mechanism, due to lack of soft tissue in the palate^{4, 5}.

Studies have found that mild conductive hearing loss is the most common one amongst individuals with CLP, varying from 60% to 75%^{6,7}. As to the compromise level of hearing loss, a study has shown that 21% of participants have presented moderate conductive hearing loss and 4% have presented severe mixed hearing loss⁷.

The presence of hearing loss can affect the appropriate perception of acoustic information, causing a certain disadvantage for children with CLP when developing linguistic competences, including Phonological Awareness (PA) – the ability to reflect on the sound structure of spoken words⁸.

PA is a metalinguistic ability conceived in different sequential linguistic levels, namely: syllables, intrasyllabic units (rhymes) and phonemes⁸. Since PA is an ability that allows the association of speech sounds to written ones, it becomes one of the requirements for learning how to read and how to write⁸. However, its progress depends on the

linguistic experiences of the child – more precisely, on the child's cognitive development and exposure to the alphabetic system⁹.

Researches have shown that children with CLP have had difficulties with the required competences for learning how to read and write^{10,11}. One of these studies applied the Initial Competence Assessment Battery for Reading and Writing (BACLE) to children between the ages of 5 years and 6 years and 11 months, noticing that most participants had not developed the word, syllabus and phoneme awareness¹⁰. In face of these considerations, this study aims at correlating audiologic and PA findings in a group of children with CLP.

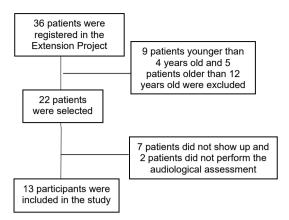
Methods

The research model employed consisted in a quantitative, cross-sectional, observational study, linked to a retrospective recall method, approved by the Research Ethics Committee under number 1.974.629, of the proposing healthcare institution. All procedures followed the recommendations of CNS Resolution 466/2012 regarding ethical aspects for research with human beings.

This study was carried out with a group of 36 patients registered in the extension project of Universidade Federal de Ciências da Saúde de Porto Alegre (Federal University of Health Sciences of Porto Alegre), entitled "Atendimento do paciente portador de fissura labiopalatina" (Health care of patients with cleft lip and palate), who were treated at the Unified Health System (SUS) Specialty Outpatient Clinic of a hospital in the same city.

The inclusion criteria used for this study were: children with chronological age from 4 to 12 years and 1 month, from both genders, without any associated syndrome diagnosis. Those who did not meet the eligibility criteria, those who did not show up for the appointment and those who did not perform an audiological assessment were excluded. Following these criteria, we selected 13 children for investigation, with ages ranging from 4 to 8 years, as shown in the flowchart (Figure 1).





*In accordance with Northern and Downs classification (2002).

Figure 1. Flowchart for sample definition.

Those responsible for the children were invited to take part of the study by the assistant researcher. Those who accepted the invitation answered a questionnaire elaborated by the researchers; then, the children were referred to the audiological assessment and, finally, they had their PA evaluated.

The questionnaires were personally applied in the SUS Specialty Outpatient Clinic, where the assistant researcher helped the responsible ones with their doubts. The questions included identification data (age, date of birth, gender, cleft type), followed by questions about hearing (whether they believed the child listened well or not; if the child had already had otitis and, when the answer was positive, with which frequency did the infection occur). We considered the otitis media as being frequent if it occurred three times in a period of 6 months, or 4 times in a period of 12 months¹².

Participants were referred to the audiological assessment in the Otorhinolaryngology sector of the proposing healthcare institution, where a tonal threshold audiometry was performed, in order to establish their auditory acuity. The assessment was performed from two to three weeks after the referral. 500 Hz, 1 KHz, 2 KHz, 3 KHz and 4 Khz frequencies were tested by air and bone conduction and the degree of hearing loss was established from a three-tone average. The classification used for audiological exams in children considers: regular hearing being less than or equal to 15 dBNA; minimal hearing loss being from 16 to 25 dBNA; mild hearing loss being from 26 to 40 dBNA; moderate hearing loss being from 41 to 65 dBNA; severe hearing loss being from 66 to 95 dBNA,

and profound hearing loss being equal to or more than 96 dBNA¹².

The imitanciometry was not performed, because the researchers of this study, who requested the exams, are graduated in Audiology and Speech Therapy, and only medical doctors are allowed to prescribe requests for this exam, in accordance with Decision No. 12/10, from the Federal Council of Medicine¹³.

After a week from the audiometry referral, we assessed the children writing hypothesis in a room of the SUS Specialty Outpatient Clinic, as well as the PA test application, performed by the assistant researcher and by two students from the Speech-language and Hearing Pathology program, members of the extension project and trained to use the Instrument of Sequential Assessment CON-FIAS test¹⁴. This test was used as PA assessment instrument because it is the one used at the data collection site.

For the writing hypothesis assessment, children were asked to write down their names, some words, and a whole sentence, as follows: "Castelo" (castle), "esqueleto" (skeleton) and "o fantasma abriu a porta" (the ghost opened the door), according to what the author proposed¹⁴. We analyzed and classified the writings in presyllabic, syllabic, syllabic alphabetic and alphabetic¹⁴.

The instrument is composed by 9 tasks concerning syllabus level (total of 40 points) and 7 tasks concerning phoneme level (total of 30 points), both including synthesis, segmentation, identification, production, exclusion and transposition tasks (syllabic and phonemic). The test results allowed a quantitative analysis based on reference values, in accordance with the writing hypothesis level in which the child was.

This instrument's reference values for a child with presyllabic writing hypothesis is of at least 18 right answers and at most 29 for the syllabus level; for the phoneme level, on the other hand, it is at least 6 and at most 10 right answers. For the syllabic writing hypothesis, in the syllabus level, at least 23 and at most 32 right answers, and in the phoneme level, at least 6 and at most 12. In the syllabic alphabetic hypothesis, in the syllabus level, the child must get at least 27 right answers and at most 36, and in the phoneme level, at least 12 and at most 18. Children with the alphabetic writing hypothesis must present, in the syllabus level, at



least 31 and at most 40 right answers, and in the phoneme level, at least 15 and at most 26.

The collected data were typed into Excel spreadsheets, from the Microsoft Office package; then, they were exported to the *Statistical Package for the Social Sciences* (SPSS), version 25, application software. The results were presented through descriptive statistics, in accordance with its nature and variables distribution (frequency, percentage, average and standard deviation). The variables associations were performed through the Student's T test and Fisher's Exact test. Only p values <0.05 were considered statistically significant.

Results

For this study, 22 children were previously selected, 7 of whom were excluded because they did not show up for the scheduled appointment, and 2 of whom did not perform the audiological assessment. Therefore, the sample was composed by 13 children, 3 females and 10 males, of whom 92.3% presented trans-foramen CLP. These data can be seen in Table 1. The children age group was higher between 4 and 6 years old because, at the moment of data collection for this study that was the profile of children assisted in the extension group.

Table 1. Sample Characterization (n=13)

Variables	No.	%
Age group		
4 to 6 years old	11	84.6
7 to 8 years old	2	15.4
Cleft Type		
Trans-foramen CLP	12	92.3
Post-foramen CLP	1	7.7
Otitis History		
No	4	30.8
Yes	9	69.2
Hearing loss		
No	3	23%
Yes	10	77%
Education		
Preschool	10	76.9
1st Year	1	7.7
2nd Year	1	7.7
3rd Year	1	7.7

The questionnaire was answered by those responsible for the children, through which we could verify that 69.2% of them had an otitis media history. As to the audiological assessment, 77% of the participants presented conductive hearing loss, and it was bilateral in 89% of those. The analyzed results were related to the level of hearing loss by ear, due to asymmetry. In Figure 2, the level of mild hearing loss was the most frequent one.

When relating the otitis media history to the time of surgery, the palatoplasty was carried out in 78% of children with otitis media history up to 18 months of age. Therefore, there was no association amongst these variables.

As for the writing development, 84.6% were in the presyllabic level and presented altered results for PA test, both in the syllabus and in the phoneme level (Table 2). The writing hypothesis was linked to the age group by using the *Student's T* test, which showed that children with presyllabic writing were substantially younger than the ones with alphabetic writing (p=0,001).

All children presented lowering in PA test scores, and the results can be seen in Table 2. The total average of right answers that children with presyllabic writing hypothesis got in the CONFIAS test was 8.8 points in the syllabus level tasks and 3.1 in the phoneme ones. In the alphabetic hypothesis, the average was 16.3 points in the syllabus level and 7 in the phoneme one. The most difficult task for them was rhyme production, in the syllabus level. In the phoneme level, the most difficult task was phoneme transposition, which no children could perform.



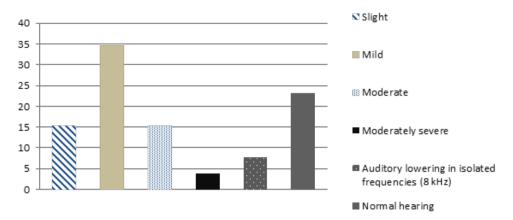


Figure 2. Distribution of the level* of conductive hearing loss and occurrence of hearing lowering in isolated frequencies or hearing threshold inside regular percentage standards.

Table 2. CONFIAS Instrument Results.

Variables	No.	%
Writing Hypothesis		
Presyllabic	11	84.6
Alphabetic	2	15.4
Syllabus Level CONFIAS		
Regular	1	7.7
Altered	12	92.3
Phoneme Level CONFIAS		
Regular	2	15.4
Altered	11	84.6

^{*} Fisher's exact test.

The verification analysis for correlation between the PA test and the audiometry results did not show any significant statistical difference, as seen in Table 3. Another correlation in which there was no statistical difference is the one between otitis media history and CONFIAS test average score (Table 4).

Table 3. Correlation between ear audiometry results and CONFIAS test.

Right ear			Left ear			
Variables	Hearing loss	Regular threshold	p*	Hearing loss	Regular threshold	р*
	n (%)	n (%)	(%)	n (%)	n (%)	•
Syllabus Level CONFIAS			1.000			1.000
Regular	1 (10.0)	0 (0)		1 (10.0)	0 (0)	
Altered	9 (90.0)	3 (100)		9 (90.0)	3 (100)	
Phoneme Level CONFIAS			0.423			0.423
Regular	1 (10.0)	1 (33.3)		1 (10.0)	1 (33.3)	
Altered	9 (90.0)	2 (66.7)		9 (90.0)	2 (66.7)	

^{*} Fisher's exact test.



Table 4. Correlation between otitis media history and CONFIAS test average score.

Variables	Otitis History	Score Average	Standard Deviation	*p value
Syllabus Level CONFIAS	No	8.7500	5.31507	0.545
(Total = 40 points)	Yes	10.7778	5.42627	
Phoneme Level CONFIAS	No	7.7500	4.64579	0.029
(Total = 30 points)	Yes	3.3333	1.93649	

^{*}Student's T Test

Discussion

Rehabilitation of patients with CLP happens in an interdisciplinary way and it takes a long time, starting on the postnatal period and going up to adulthood, depending on the severity of the case^{5,15}. This duration demands financial expenses and family and personal compromises. This study sought the participation of children who had already gone through, or had been going through, clinic speech therapy; therefore, the adherence to the research suffered a loss of approximately 40%, characterized by financial issues that have jeopardized the access to audiological and voice therapy services locations.

The most frequent cleft type in the participants of this study was the unilateral trans-incisive foramen, which affects the lips, the alveolar bone, the hard palate and the soft palate of one side of the hemiface⁽¹⁶⁾. This classification uses the incisive foramen as an anatomic reference, defining them in: pre-incisive foramen cleft (lips and alveolar bone, up to the incisive foramen), bilateral or unilateral, complete or incomplete; post-incisive foramen cleft (hard and soft palate), complete or incomplete; trans-incisive foramen cleft, unilateral or bilateral¹⁶. The cleft type referred as being the most frequent one in the literature is the trans-incisive foramen¹⁷, supporting that findings.

The trans-foramen type CLP compromises anatomic structures in different levels, which might interfere in orofacial and hearing functions¹⁸. Therefore, children with cleft palate might present problems in the middle ear^{4,6}. In this study, it was noticed that the occurrence of otitis media was frequent, and that all participants presented a cleft in the posterior palate. Studies have shown that children with CLP had an occurrence of otitis media significantly higher than the ones without cleft^{6,19}.

Otitis media can cause conductive hearing loss, which happens when there is some type of

obstruction in the sound conduction ¹⁸. The presence of conductive hearing loss, frequent in the children of this study, has been ratified by results reported in researches emphasizing this type of hearing loss as being the most frequent in children with CLP^{4,6}. As to the level of hearing loss, the mild one showed the highest incidence in the ears of the participants of this study, confirming other analysis^{6,7}.

Considering the otitis media history and the time of surgeries to close the palate, it was assumed that most children of this study had their palate reconstruction done during the advised period⁵, up to 18 months of age, and it may not infer an association between the variables. What might be related to the otitis media history in 69.2% of samples, is the compromise of the velopharyngeal mechanism⁴, however, this variable was not investigated by this study.

As to the writing hypothesis, the predominance found amongst the children of this study was the presyllabic one, in which the child considers writing the same as reproducing traces that he/she identifies as being the basic form of writing^{20, 21}. The subsequent writing hypothesis is the syllabic level, in which the child represents each letter with a sound value, writing a letter per syllabus^{21, 22}. The syllabic alphabetic level counts on variations between the syllabic and the alphabetic writing²¹. The progression to the alphabetic stage is given through the understanding that each grapheme represents sound units that are smaller than the syllabus²².

Children in ordinary development of the writing hypothesis, with ages between 4 and 5 years old, are positioned in the presyllabic level²², and that is the same level observed in the children with CLP of this study, in this age group. By the age of 6 years old, children in ordinary development are positioned between the syllabic alphabetic and alphabetic levels²². The findings of this study have shown that children who were 6 years old presented a delay in the writing development, for



they were in the presyllabic level. Researches report that children with CLP present reading and writing difficulties, and this fact is linked to PA alterations, because this ability is one of the learning requirements ^{10, 23}.

It should be highlighted that the children taking part in this study presented PA alterations; that could be verified due to the low score obtained in the CONFIAS instrument. On the other hand, children without CLP, who were analyzed in other studies, have presented higher score averages^{14, 24}. The most difficult test task for the children of this study (transposition of phoneme) was the same as the one considered by children with ordinary development¹⁴.

This study has not illustrated a correlation between hearing loss and PA alterations. However, it does not discard the possibility of existing an association amongst variables, since the integrity of hearing physiological mechanisms is essential to the perception of speech sounds, which will reflect in the development of metalinguistic abilities²⁵.

The PA difficulties presented by children taking part in this study can be inter-related to other non-explored factors, such as social factors, speech problems and disorders in the central auditory processing^{6, 10}. Apart from these, other researches have reported that difficulties in this ability can be correlated to working memory gaps, because it is necessary for the verbal material to be kept in this memory, so that one can perform the requested tasks in the PA tests^{24, 25}.

Conclusion

Children with CLP presented PA difficulties, both in the syllabic and in the phoneme level. There was no correlation between audiological and PA findings. However, it has not discarded the hypothesis of an interrelation between the two variables associated to other factors, since most children presented conductive hearing loss, and all of them presented difficulties with metalinguistic abilities. We suggest the development of more researches related to this theme.

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