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Original Article

Artigo Original

Verification of the therapeutic process in cleft patients

Verificação do processo terapêutico em pacientes fissurados

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Keywords

Cleft Palate
Cleft Lip
Articulation Disorders

ABSTRACT

Purpose: This study was conducted to verify the origin of a longer or even failed therapeutic process in patients with cleft lip and palate as to its difficulty. **Methods:** Eighteen children undergoing therapeutic process were observed for at least 6 months and divided into two groups: presenting isolated cleft lip and palate (group I) and having been diagnosed by a Speech-Language Pathologist with reading and writing disorders, with manifestation of phonological awareness deficit (group II). Two tests were applied for the evaluation of speech and language: ABFW Language Test for Young Children (phonology) and Phonological Awareness: Instrument of Sequential Assessment (CONFIAS). **Results:** Group I presented higher percentages in ABFW test than group II, except in the "simplification of consonant cluster" and "plosive devoicing" variables. It was also observed that, in the process of omission, group I hardly omits the vibrant consonant, as observed in group II. At the syllable level of CONFIAS, the percentages observed in group I tended to be higher than in group II, with the exception of the following tasks: "medial syllable," "production of rhyme," and "exclusion." At the phoneme level, the percentages observed in group II tended to be higher than in group I, with the exception of the following tasks: "starts with given sound," "exclusion," "synthesis," and "segmentation." No significant difference was observed between percentage distributions in groups I and II ($p>0.118$). **Conclusions:** The differences found between groups I and II, although not statistically significant, may suggest that the presence of malformation hinders speech and language acquisition and development and prolongs the therapeutic process if directive interventions are not carried out, including phonological awareness therapy.

Descritores

Fissura Palatina
Fenda Labial
Transtornos da Articulação

RESUMO

Objetivo: Verificar a origem de um processo terapêutico mais longo ou até com insucesso de pacientes com fissura labiopalatina quanto à sua dificuldade de fala. **Métodos:** Foram observadas 18 crianças em processo terapêutico há pelo menos seis meses, divididas em dois grupos: com fissura labiopalatina isolada (grupo I), e com diagnóstico fonoaudiológico de transtorno de leitura e escrita com manifestação de déficit da consciência fonológica (grupo II). Aplicaram-se dois testes para avaliação de fala e linguagem: ABFW – Teste de Linguagem Infantil (área de Fonologia) e Consciência Fonológica: Instrumento de Avaliação Sequencial (CONFIAS). **Resultados:** No ABFW, o grupo I apresentou porcentagens maiores do que o grupo II, menos nas variáveis "simplificação de encontro consonantal" e "ensurdecimento de plosiva". Também observou-se que, para o processo de omissão, grupo I dificilmente omite as vibrantes como ocorreu no grupo II. No CONFIAS nível sílaba as porcentagens do grupo I tendem a ser maiores que as do grupo II, com exceção das tarefas "sílaba medial", "produção de rima" e "exclusão". No nível do fonema, as porcentagens do grupo II tendem a ser maiores do que no grupo I, com exceção das tarefas: "inicia com som dado", "exclusão", "síntese" e "segmentação", mesmo sem diferença significativa entre as distribuições da porcentagem nos grupos I e II ($p>0,118$). **Conclusões:** As diferenças encontradas entre os grupos I e II, apesar de não significativas estatisticamente, podem sugerir que a presença da malformação dificulte a aquisição e o desenvolvimento da fala e linguagem, e prolongue o processo terapêutico se não realizadas intervenções diretivas, como incluir terapia de consciência fonológica.

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INTRODUCTION

All children start their speech and language acquisition process early, and any environmental, anatomical, and/or physiological factors can affect it. The first years of life and experiences with the world of sound are paramount to the development and acquisition of speech and language. Therefore, intrauterine congenital disorders have a deleterious effect on the learning of symbolic language and the development of a speech pattern[1].

This process is based on specific anatomical structures and on the ability of oral motor control of speech; hence, its development is affected in children presenting with anatomical and physiological impairments[2].

According to recent studies, about 50% of the children born with cleft palate have speech difficulties at around 3 years of age, even after palatoplasty[3-5]. Several studies in other languages, mainly in English, report that children with cleft palate present deficits in both phonological processes and phonetic and resonance changes[6-9].

As the whole experience of early language influences the development of skills of perception and acquisition of phonological processes, children with cleft palate can present phonetic alterations because of changes in anatomical structure, which influences the learning and acquisition of speech and language, and are at risk for deficits in phonological awareness.

Other factors that can impair language learning and acquisition of speech and language are otologic problems, such as otitis media and hearing loss, very common in these children[10-14]. There are studies that indicate that otologic problems hinder the accomplishment of tasks involving verbal memory and sentence comprehension[15]. Furthermore, the mode and time of start of the intervention can also change this scenario. Recent studies highlight that, either in schools or by specialists, the later the intervention occurs, the greater the risk of difficulties in communication and cognition; the sooner and rigorous the intervention strategies are, the better the outcome in children with cleft lip or palate[14].

The reference to the therapeutic process in children with cleft lip and palate tending to be long term is very common. Having knowledge of the occurrence of compensation and possible changes that may be present in children with this malformation and observing reports of speech and language alterations in international articles, this study aims to verify the origin of a longer or even failed therapeutic process and how to modify or redirect the therapeutic approach, in addition to evaluating and comparing patients with cleft lip and palate as to their speech difficulties.

METHODS

All participants in this study signed the informed assent. Their legal representatives also agreed by signing the Free and Informed Consent Form, so that their children could be subjected to any kind of evaluation.The study was analyzed and approved by the Institutional Ethics Committee under number 0170/11, being mainly a descriptive and observational cross-comparative study.

Children from fourth to seventh year of primary school who received outpatient care in Speech-Language Pathology and Audiology in a public hospital, with a minimum of 6 months of therapeutic intervention and an average age of 9 years, were enrolled, observed, and evaluated.

The participants were divided into the following groups:

- Group I: 11 children, eight boys and three girls, enrolled in primary education in state and municipal schools, with congenital cleft lip and palate or isolated cleft palate and with an average age of 8 years, who received outpatient care in the Speech-Language Pathology and Audiology Craniofacial Malformation and Associated Syndromes department of a public hospital;
- Group II or control: seven children, three boys and four girls, enrolled in primary education in state and municipal schools without craniofacial malformation or genetic syndrome, with a mean age of 10 years, who received outpatient care at the Speech-Language Therapy Department at the same institution.

The variables of education, age, and socioeconomic conditions for the two groups were not controlled. Data regarding audiologic and central auditory processing evaluation in children of both groups were not considered in the overall results.

Inclusion criteria were as follows: the presence of congenital fissure and a therapeutic process of more than 6 months for group I and a therapeutic process of more than 6 months and the absence of craniofacial malformation for group II.

Exclusion criteria for both groups were motor abnormalities, moderate to severe degree of hearing loss, serious neurological alterations, and consecutive absences during testing. Specific exclusion criterion for group I was any child who had sought care before 4 years of age and, for group II, any child who presented alterations in the central auditory processing. There was a sample loss of 10 children in total from both the groups.

Data collection was conducted in the outpatient clinics of the Speech-Language Pathology and Audiology for Cleft Palate and Associated Syndromes and Speech-Language Therapy Departments of a public hospital. Two tests were applied for evaluation of speech and language: ABFW Language Test for Young Children; and Phonological Awareness: Instrument of Sequential Assessment (CONFIAS), and their performance was analyzed; no written test was used. The tests were administered by the researcher by recording the voice of the children during procedures and not assisted by filming[16,17].

The ABFW test of child language in the field of phonology consists of two tests: naming and imitation. For this study, only the naming phase was used, allowing the quantitative and qualitative evaluation of the phonetic inventory and 14 phonological processes: syllable reduction, consonant harmony, plosivation of fricatives, velar posteriorization, palatal posteriorization,

velar anteriorization, palatal anteriorization, simplification of liquid, simplification of consonant cluster, simplification of final consonant, plosive voicing, fricative voicing, plosive devoicing, and fricative devoicing.

The purpose of the evaluation of the phonological system is to check the child's phonetic inventory and analyze the phonological processes that have been developed, in addition to the presence of phonological simplifications and compensatory disorders, such as glottal stop, pharyngeal fricative, and nasal lisp. The glottal stop can be interpreted as the replacement of the articulation point of plosive phonemes in the glottis, whereas the pharyngeal fricative is produced by friction in the pharyngeal region, and the nasal lisp occurs by directing air into the nasal cavity during the emission of fricative phonemes.

The application of the ABFW test was conducted in a room of the outpatient clinic of a public hospital during their clinical routine. An excerpt from the spontaneous speech of children and the application of the naming task were recorded in an MP3 player, and later, the phonetic and phonological processes analyses were performed. In the naming task, the child was asked to name the 34 figures shown in the form of boards, measuring 20 cm by 23 cm. If the child was not able to name, the examiner named that figure and, after five subsequent figures, asked the child to name it again, according to the instructions specified by the authors. All responses were recorded in two forms: the phonetic transcription form and the nomination form.

Then, the CONFIAS test was applied, a tool for sequential evaluation divided into two levels: the syllable and the phoneme. In the first level, nine aspects are analyzed: synthesis, segmentation, identification of initial syllable, identification of rhyme, production of word with given syllable, identification of medial syllable, rhyme production, deletion, and transposition. In the phoneme level, aspects verified were production of word starting with the given sound, identification of initial phoneme, identification of final phoneme, exclusion, synthesis, segmentation, and transposition. The child should respond to 16 tasks presented orally by the examiner, so that the phonological aspects of the child at the syllable and phoneme levels could be assessed. All responses were registered in a form of rights and wrongs, through which the analysis of the percentage of correct answers of each aspect and the calculation of the percentage of success in relation to the possible total of the test were performed according to the given instructions. No test was applied for collecting data on reading and/or writing.

In this study, the significance level was set at 5% ($p<0.05$). To calculate this index, the PASW Statistics software was used. The p-value was calculated according to the n sample collected (n=15).

The χ^2 test was applied to the values of the distributions of the phonetic inventory, the presence or absence of phonological processes, and the percentage of absence of omission, glottal stop, pharyngeal fricative, and nasal lisp observed in the application of the ABFW test in both the groups.

To compare the distributions of responses in the aspects of the CONFIAS test in the two groups, the Kruskal-Wallis test was applied. The same procedure was adopted to compare the distributions of responses in ABFW in both the groups. When necessary, the Bonferroni procedure was considered to find differences between them[18].

In the figures of this study, a color was set for each patient, thereby facilitating the analysis of the difficulties individually and in groups.

RESULTS

ABFW test

In Table 1, the means of the variables of the phonetic inventory of words are very close. We can conclude that the groups had similar errors and there are no specific errors to each of them.

Figure 1 shows the individual values of the ABFW test observed at a similar concentration of percentage of phonetic right answers and of absence of the phonological processes.

Table 1. Amount of correct answers of the phonetic inventory performed by the children of the two groups

Variable	n	Mean±SD	Min–Max	Amplitude	p-value*
Phonetic inventory					
I	11	45.8±31.1	6–94	88	
II	7	58.9±19.0	38–94	56	
Total	18	52.4±	6–94	88	0.199

*p<0.05; SD: standard deviation; Min: minimum; Max: maximum.

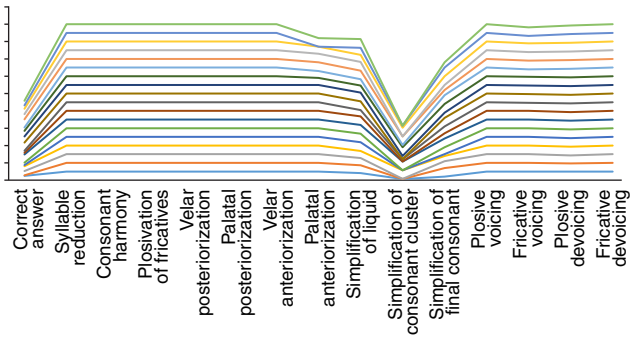

Figure 1. Phonological processes in the ABFW test

Further analysis of the ABFW test indicated that, for the phonological process of palatal anteriorization, a p-value with a significance level of 5% (p=0.013) was observed (Table 2). In other aspects, group I presented higher percentages than group II, except for the variables "simplification of consonant" and "plosive devoicing."

Table 2. Demonstration of the percentage of correct answers in the phonological processes of ABFW in each group

Variable	n	Mean±SD	Min–Max	Mediana	Amplitude	p-value
Palatal anteriorization						
I	11	100.0±0.0	100–100	100	0	
II	7	77.1±35.5	0–100	80	100	
Total	25	92.8±20.7	0–100	100	100	0.013*
Simplification of liquid						
I	11	91.8±10.2	73–100	100	27	
II	7	88.4±11.3	73–100	82	27	
Total	25	91.0±12.2	55–100	100	45	0.565
Simplification of consonant cluster						
I	11	25.5±38.9	0–100	2	100	
II	7	50.0±47.9	0–100	25	100	
Total	25	37.7±41.5	0–100	25	100	0.364
Simplification of final consonant						
I	11	70.9±25.9	20–100	80	80	
II	7	82.9±21.4	60–100	100	40	
Total	25	79.2±23.4	20–100	80	80	0.269
Fricative voicing						
I	11	99.4±2.1	93–100	100	7	
II	7	96.0±6.8	86–100	100	14	
Total	25	98.6±4.0	86–100	100	14	0.219
Plosive devoicing						
I	11	98.7±2.8	93–100	100	7	
II	7	100.0±0.0	100–100	100	0	
Total	25	98.9±3.3	86–100	100	14	0.526
Fricative devoicing						
I	11	100.0±0.0	100–100	100	0	
II	7	100.0±0.0	100–100	100	0	
Total	25	99.4±2.8	86–100	100	14	>0.999

*p<0.05; SD: standard deviation; Min: minimum; Max: maximum.

Figure 2 shows the values for the percentage of nonoccurrence of omission of phonemes and the presence of compensatory articulation disorders (glottal stop, pharyngeal fricative, and nasal lisp) in both the groups. A p-value with a significance level of 5% (p<0.05) was verified for the variables "omission" and "pharyngeal fricative." Group I presented lower percentages for all variables because of their underlying disease, except for the variable omission. Conversely, group II did not show the occurrence of compensatory articulation disorders, meeting the expectations, because they did not show cleft lip or palate, which was not expected to occur.

Table 3 shows that, for the process of omission, the child with cleft palate hardly omits vibrant consonants, as occurred in group II. Children with cleft lip or palate made more changes than omissions, preserving the "space" of the phoneme.

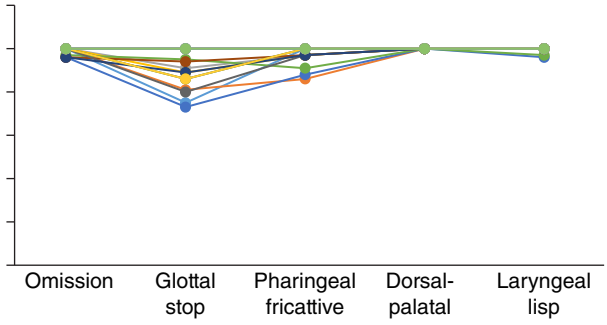

Figure 2. Distribution of compensatory speech articulation disorders

Table 3. Description of the nonoccurrence of omission and presence of compensatory mechanisms (glottal stop, pharyngeal fricative, and nasal lisp) in both groups

Variable	n	Mean±SD	Min–Max	Amplitude	p-value
Omission					
I	11	100±0	100–100	0	
II	7	98.6±1.9	96–100	4	
Total	18	99.3±	96–100	4	<0.055*
Glottal stop					
I	11	87.9±9.4	73–100	27	
II	7	100±0	100–100	0	
Total	18	93.9±	73–100	27	0.124
Pharyngeal fricative					
I	11	95.5±4.9	86–100	14	
II	7	100±0	100–100	0	
Total	18	97.75±	86–100	14	0.016*
Nasal lisp					
I	11	99.4±1.4	96–100	4	
II	7	100±0	100–100	0	
Total	18	99.7±	96–100	4	0.266

*p<0.05; SD: standard deviation; Min: minimum; Max: maximum.

CONFIAS test

The analysis of the CONFIAS test at the syllable level (Table 4) aimed to identify the differences between groups. The percentage distributions of the groups I and II (p>0.088) did not show a significance level.

The percentages of group I (Figure 3) tend to be higher than those of group II, with the exception for the "medial syllable," "rhyme production," and "exclusion" tasks.

In the analysis of the CONFIAS test, the phoneme level indicates no significant difference between the distributions of the percentages of correct answers in both the groups (Table 5). The difficulties in performing tasks are again similar.

Further analysis (Figure 4) found that the values of the percentage of correct phonemes in group II tend to be higher than in the group I. However, in the specific task of starting with the given sound, exclusion, synthesis, and segmentation, group I shows a higher percentage of correct answers than group II. However, there is no significant difference between the percentage distributions of groups I and II (p>0.118).

Table 4. Description of the percentages of correct answers in the phonological aspects of Phonological Awareness: Instrument of Sequential Assessment at the syllable level in each group

	n	Mean±SD	Min–Max	Amplitude	p-value
Syllable					
I	11	85.9±17.1	37–97	60	
II	7	82.3±9.8	70–95	25	
Total	18	84.1±	37–97	60	0.094
Synthesis					
I	11	95.5±15.1	50–100	50	
II	7	89.3±19.7	50–100	50	
Total	18	92.4±	50–100	50	0.609
Segmentation					
I	11	97.7±7.5	75–100	25	
II	7	89.3±19.7	50–100	50	
Total	18	93.5±	50–100	50	0.558
Identification of initial syllable					
I	11	95.5±15.1	50–100	50	
II	7	89.3±19.7	50–100	50	
Total	18	92.4±	50–100	50	0.609
Identification of rhyme					
I	11	90.9±23.1	25–100	75	
II	7	85.7±13.4	75–100	25	
Total	18	88.3±	25–100	75	0.347
Syllable given					
I	11	97.7±7.5	75–100	25	
II	7	85.7±13.4	75–100	25	
Total	18	91.7±	75–100	25	0.088
Medial syllable					
I	11	86.4±30.3	25–100	75	
II	7	89.3±13.4	75–100	25	
Total	18	87.8±	25–100	75	0.255
Production of rhyme					
I	11	65.9±25.7	25–100	75	
II	7	71.4±22.5	50–100	50	
Total	18	68.7±	25–100	75	0.747
Exclusion					
I	11	72.5±27.9	12–100	88	
II	7	82.0±17.7	62–100	38	
Total	18	77.3±	12–100	88	0.435
Transposition					
I	11	86.4±30.3	0–100	100	
II	7	82.1±18.9	50–100	50	
Total	18	84.3±	0–100	50	0.242

SD: standard deviation; Min: minimum; Max: maximum.

DISCUSSION

Because the groups have similar errors in the phonetic aspect and no significant differences were observed, one can conclude that there is no standard or specific error for each group, confirming the results of previous studies[15-17].

All children underwent therapeutic intervention for at least 6 months, and one of the aspects addressed in the therapy for phonetic production in children with cleft lip and palate was the airflow associated with sensory stimuli such as tactile, visual, and auditory sensations. Thus, it is believed that, for the production of a palatal fricative consonant, the child with cleft lip or

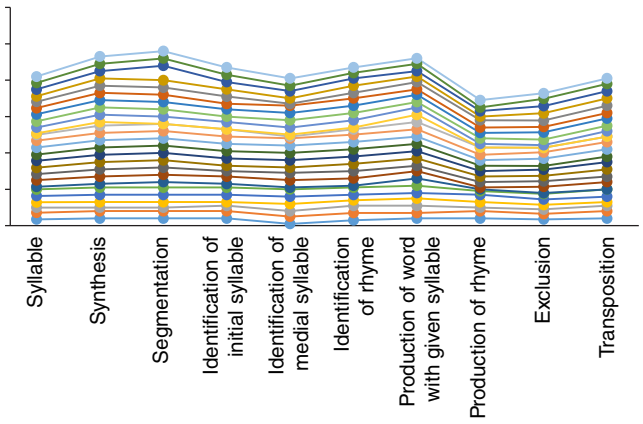

Figure 3. Distribution of Phonological Awareness: Instrument of Sequential Assessment percentages at the syllable level

Table 5. Description of the percentages of correct answers in the aspects of Phonological Awareness: Instrument of Sequential Assessment at the phoneme level in each group

	n	Mean±SD	Min–Max	Amplitude	p-value
Phoneme					
I	11	77.2±8.5	63–96	33	
II	7	77.6±12.8	60–94	34	
Total	18	77.4±	60–96	34	0.747
Starts with given sound					
I	11	100.0±0.0	100–100	0	
II	7	96.4±9.4	75–100	25	
Total	18	98.2±	75–100	25	0.609
Identification of the initial phoneme					
I	11	100.0±0.0	100–100	0	
II	7	100.0±0.0	100–100	0	
Total	18	100.0±0.0	100–100	0	>0.999
Identification of the final phoneme					
I	11	84.1±20.2	50–100	50	
II	7	85.7±13.4	75–100	25	
Total	18	84.9±	50–100	50	0.375
Exclusion					
I	11	74.0±20.3	50–100	50	
II	7	68.7±22.5	33–100	67	
Total	18	71.4±	33–100	67	0.134
Synthesis					
I	11	72.7±13.5	50–100	50	
II	7	71.4±17.3	50–100	50	
Total	18	72.1±	50–100	50	0.845
Segmentation					
I	11	75.0±25.0	25–100	75	
II	7	60.7±24.4	25–100	75	
Total	18	67.9±	25–100	75	0.431
Transposition					
I	11	38.6±20.5	0–100	75	
II	7	64.3±28.3	25–100	75	
Total	18	51.5±	0–100	75	0.118

SD: standard deviation; Min: minimum; Max: maximum.

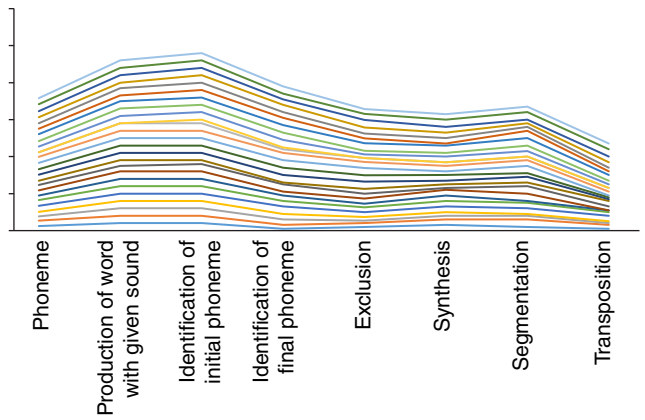

Figure 4. Distribution of Phonological Awareness: Instrument of Sequential Assessment percentages at the phoneme level

palate benefits more from the introduction and acquisition of the phoneme than a child with no malformation from group II, which generally uses the technique only for demonstration of articulation point, favoring the performance of alveolar fricatives instead of palatal fricatives.

Previous studies have reported that children with cleft lip or palate perform more exchanges than omissions, which was also observed in this study, leading to the conclusion that children with cleft lip or palate hear the phoneme but has difficulties in discriminating and producing it[19-21].

Since the 1960s, speculations made through evidence already pointed that children born with malformation could be affected in the performance of verbal and nonverbal tasks, which would modify the expressive modalities. In these studies, there are also reports that the children presented delays in aspects of verbal behavior before school age[22,23]. Back then, these differences could be considered as difficulties in auditory discrimination, thus affecting the verbal tasks and verbal behavior.

Knowing that auditory discrimination is a distinct perception ability than just receiving auditory stimuli and the ability to distinguish one sound from another and distinguish small differences in them, it is thought that the child learns to associate a sound with the source that produced it and that a child with alterations in discrimination confuses and changes phonemes at the time of its enunciation[11,12].

The processes of attention and listening work together with the development of children's capacity to handle the sounds received (input). Children with this alteration may vary the development of auditory and speech skills, presenting developmental delay, which can lead to the emergence of a phonological disorder[7,10,12].

The initial hypothesis of this study was that group I would present more difficulties in the acquisition of speech and language, such as perceiving sound without identifying it or being able to build words that start with this sound. The alteration that occurs in these children is apparently an imbalance of phonological processing.

A widely believed assumption says that children born with malformation correctly identify the phoneme (correct input), but due to anatomical alterations, such as history of otitis media, lower ability to control the oral motor speech, and alteration in the acquisition and development of speech and language, produces the output in the incorrect place with compensation, and the feedback itself reinforces this production as the child imagines to be producing the phoneme correctly[6,7]. When a task of constructing words from a phoneme or syllable is required, they have no record of this sound, hence sounds like a new sound[13].

The results of the CONFIAS test indicate that there is no significant difference between the distributions of the percentage of correct answers in both the groups. However, group I shows, in the aspects related to the phoneme, difficulties in phonological tasks. Because the patient with cleft lip or palate respects the space of the phoneme but does not identify it, in tasks where discrimination is required, they have more difficulties.

In the aspects that were not the object of therapy, the group with malformation presented difficulties, confirming the suspicion that, in the aspects they presented better percentages, therapy may have helped already, as their percentage of concentration is very similar to group II.

Studies conducted between 2005 and 2010 reported that early language influences the development of the perception skills and phonological processes, that children with a history of otitis media differ on memory and sentence comprehension tasks, and that children born with cleft lip or palate are significantly worse in reading, phonological memory, and reading fluency[13,14].

As physical conditions or inabilities and incompetence interfere with functional deficits, it was found that children with malformation showed no greater phonological awareness deficits than the control group.

This finding explains the facts that occur in the therapeutic experience, in which the child with cleft lip or palate, regardless of age, presents, in the stage of automation, a longer and more tiring period if phonological aspects are not addressed. However, in the stages of phoneme awareness, production, and correction, during which sensory therapeutic techniques are used to facilitate perception, children do not demonstrate great difficulties.

From the reports of several authors, it is known that the spoken language and the recognition of the code are precursors of reading, that orthographic knowledge influences the phonological awareness tasks, that speech disorders are correlated with phonological awareness, and that this affects word recognition and speech[15-17]. Probably, group I will present greater risks for the development of reading and writing disorders.

Therefore, it is believed that it would be interesting to take the phonological aspect always into consideration in speech therapy in children with cleft lip or palate. Because these children do not have, in their therapeutic intervention plan, the concern with the analysis and treatment of phonological aspects, it is believed that this is one of the reasons for a more lengthy process.

In this study, reading and writing disorders were not assessed in groups I and II. Future studies on these changes can define how the manifestations of deficits in phonological awareness will interfere with these functions.

CONCLUSIONS

With the revelations of deficits in phonological awareness from the analysis of the results obtained in this study, especially at the phoneme level, we conclude that isolated children born with cleft lip or palate (group I) will have a risk to acquire the ability to analyze words in relation to phonemes and syllables, explaining the reason for the difficulty in speaking in these patients.

As deficits were not known previously, the therapeutic approach was incomplete. Therefore, the therapeutic process eventually took longer than necessary.

The differences found between groups I and II, although not statistically significant, may suggest that the presence of the malformation impairs the acquisition and development of speech and language and, as said earlier, extend the therapeutic process if directive interventions, such as the inclusion of phonological awareness therapy in speech therapy, are not carried out.

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