https://doi.org/10.23925/2176-2724.2020v32i3p383-395

Multiple case studies: schoolchildren with cleft lip and palate in context of phoniatric evaluation

Estudos de casos múltiplos: escolares com fissura labiopalatina no contexto da avaliação foniátrica

Múltiplesestudios de caso: escolares conlábio leporino y paladar hendido en el contexto de la evaluación foniátrica

*Mônica Elisabeth Simons Guerra**
*Vanessa Magosso Franchi**
*Beatriz Cavalcanti de Albuquerque Caiuby Novaes**

Abstract

Introduction: Phoniatrics is interested in cleft lip and palate (CLP) due to the complexity in aspects of speech, language and hearing. **Objective:** In context of phoniatric evaluation in schoolchildren with CLP, the objective of multiple cases study was to describe the interference of clinical factors in the performance in tests of auditory perceptual function of language, as well as the association between them. **Method:** Thirty children, between 6 years and 2 months and 9 years and 11 months, were evaluated by Phoniatric doctor, in interdisciplinary treatment service for CLP: 13 (43%) cleft palate and 17 (57%) cleft lip and palate. Clinical factors (including type of cleft, age at evaluation, sex, socioeconomic status, school performance, speech therapy, audiological examination and others) and performance in tests of auditory discrimination, auditory memory and phonological awareness were described, including the association between them. **Results:** There was no significant difference between the cleft groups and the mean age (p = 0.618), sex (p = 0.431), socioeconomic level (p = 0.580) and school performance (p = 0.785). The percentage of children who had attended therapy is higher in cleft lip and palate group (94.1%). Changes in tests: 4 (13.3%) auditory discrimination, 6 (20%) auditory memory and 8 (26.7%)

* Pontifícia Universidade Católica de São Paulo – São Paulo, SP, Brazil.

Authors' contributions:
MESG - Conception and design of the work.
VMF - Critical review of the content.
BCACN - Approval of the final version and guidance of the master.

Correspondence e-mail: Mônica Elisabeth Simons Guerra simons.guerra@gmail.com
Received: 13/03/2020
Accepted: 28/5/2020

phonological awareness. The cases with unsatisfactory school performance, more speech therapy time and worse speech recognition thresholds had a worse performance in auditory memory and phonological awareness. **Conclusion:** The phoniatric evaluation made it possible to identify clinical factors of the cleft, which seems, in some way, to interfere in the language development of these children.

Keywords: Cleft lip; Cleft palate; Child language.

Resumo

Introdução: A foniatria tem interesse em fissura labiopalatina (FLP) devido à complexidade em aspectos de fala, linguagem e audição. **Objetivo:** No contexto da avaliação foniátrica em crianças escolares com fissura labiopalatina, o objetivo do estudo de múltiplos casos foi descrever a interferência de fatores clínicos no desempenho em testes da função perceptiva auditiva de linguagem, assim como a associação entre eles. **Método:** Trinta crianças, entre 6 anos e 2 meses e 9 anos e 11 meses, foram avaliadas pelo médico foniatra, em serviço interdisciplinar de tratamento da FLP: 13 (43%) fissura pós-forame incisivo e 17 (57%) fissura transforame incisivo. Foram descritos os fatores clínicos (incluindo tipo de fissura, idade na avaliação, sexo, condição socioeconômica, desempenho escolar, realização de fototerapia, exame audiológico e outros) e o desempenho em testes de discriminação auditiva, memória auditiva e consciência fonológica, incluindo a associação entre eles. **Resultados:** Não houve diferença significativa entre os grupos de fissura e médias de idade (p=0,618), sexo (p=0,431), nível socioeconômico (p=0,580) e desempenho escolar (p=0,785).A porcentagem de crianças que fez terapia é maior no grupo de fissura transforame (94,1%). Alterações nos testes: 4 crianças (13,3%) discriminação auditiva, 6 (20%) memória auditiva e 8 (26,7%) consciência fonológica. Os casos com desempenho escolar insatisfatório, mais tempo de fonoterapia e piores médias do limiar de reconhecimento de fala tiveram desempenho pior em memória auditiva e consciência fonológica. **Conclusão:** A avaliação foniátrica possibilitou o apontamento de fatores clínicos da fissura, que parecem, de alguma forma, interferir no desenvolvimento da linguagem destas crianças.

Palavras chave: Fenda labial; Fissura palatina; Linguagem infantil.

Resumen

Introducción: Foniátrica está interesado en el labio leporino y paladar hendido debido a la complejidad en los aspectos del habla, el lenguaje y la audición. **Objetivo:** El contexto de la evaluación foniatría en escolares con labio leporino y paladar hendido, el objetivo del estudio de múltiples casos fue describir la interferencia de factores clínicos en el desempeño en las pruebas de la función auditiva perceptiva del lenguaje, así como la asociación entre ellos. **Método:** Treinta niños, entre 6 años y 2 meses y 9 años y 11 meses de edad, fueron evaluados por un médico foniátrico, en un centro de tratamiento interdisciplinario para labio leporino y paladar hendido: 13 (43%) paladar hendido y 17 (57%) labio leporino y paladar hendido. Se describieron y verificaron los factores clínicos (incluido el tipo de hendidura, la edad en la evaluación, el sexo, el estado socioeconómico, el rendimiento escolar, la fototerapia, el examen audiológico y otros) y el rendimiento en las pruebas de discriminación auditiva, memoria auditiva y conciencia fonológica, incluida la asociación entre ellos. **Resultados:** No hubo diferencias significativas entre los grupos de hendidura y la edad media (p = 0.618), sexo (p = 0.431), nivel socioeconómico (p = 0.580) y rendimiento escolar (p = 0.785). El porcentaje de niños que se sometieron a terapia es mayor en el grupo de paladar hendido y labio leporino (94.1%). Cambios en las pruebas: 4 niños (13.3%) discriminación auditiva, 6 (20%) memoria auditiva y 8 (26.7%) conciencia fonológica. Los casos con un rendimiento escolar insatisfactorio, más tiempo de terapia del habla y peores umbrales de reconocimiento del habla tuvieron un peor rendimiento en la memoria auditiva y la conciencia fonológica. **Conclusión:** La evaluación foniátrica permitió identificar factores clínicos de la hendidura, que de alguna manera, parece interferir en el desarrollo del lenguaje de estos niños.

Palabras clave: Labio leporino; Fisura del paladar; Lenguaje infantil.

Introduction

Phoniatrics studies and cares for children and adults with language disorders. It was set as a medical field practice of otolaryngology through the administrative act of Brazilian Federal Medical Board (CFM)[1] in 2006. According to Spinelli (1983), language disorders are manifested in the various modalities of language production and compression, in which impairment grade is variable concerning every single aspect and the combination between them[2].

The first years are important for the development of receptive and expressive language. It depends on several factors such as genetics, environment and emotions in order to stimulate neuroplasticity and learning[2,3]. There is not a single language acquisition rhythm for all children, but there are some milestones of human communication that must be followed[2,3]. Therefore, phoniatrics is interested in patients with cleft lip and palate due to the great complexity and variability in the aspects of speech production, language development and changes in hearing from the first years of life as a result of function failure of muscles involved in velopharyngeal functioning of the auditory tube aeration and changes in craniofacial growth[4,5].

Velopharyngeal closure, which separates the oral and nasal parts of the pharynx, occurs respectively from craniodorsal movement of the soft palate and medialization and anteriorization movements of the lateral and posterior walls of the pharynx[5]. In children with cleft lip and palate, before primary correction of the palate, velopharyngeal dysfunction is always present because of dysplastic changes during embryonic development of the palate levator and tensor veli palatini muscles and it may persist after primary palate surgery. This dysfunction is characterized by several symptoms with an impact on the proper functioning of speech processes, necessary for oral quality of vowels and occlusive, fricative and sibilant consonants, as well as for the processes of swallowing and middle ear aeration[5,6].

As a result, on the one hand, language may be impaired by speech anatomical and functional problems, such as compensatory and articulation disorders, which involve nasal emission and low intraoral pressure. In addition, they are directly related to changes in the palate and lip and may persist even after corrective surgeries of facial defects[4-6].

On the other hand, deprivations of daily-living concerns such as social and affective experiences are also important to build language. They are often true for families whose children suffer from chronic conditions, such as: recurrent hospitalizations and multiple surgeries, difficulties in sucking and swallowing, self-perception of facial appearance and vocal quality and other behavioral factors as well as parents' overprotection[4,5,7-12].

Ultimately, children with cleft lip and palate show high frequency of changes in auditory function because of function impairment of the tensor veli palatini and levator palate muscles. This prevents proper opening of the auditory tube and reduces aeration of the middle ear, which results in high incidence of otitis[5,18-20]. Kuo et al (2014) describe that before one year of age, otitis media with effusion can occur at least once in 90% of children with cleft palate[18].

Changes in hearing can significantly affect the development of auditory function in children with cleft palate Otitis media is the most common cause of temporary hearing loss in this population[13] . And therefore, early in life, otitis media can lead to long-term consequences on various hearing skills at school age, even after clinical recovery[14]. According to the clinical practice guidelines in the management of otitis media with effusion, we know it can occur in any child, ranging from 2 months of life to 12 years of age, with or without disabilities or other conditions that predispose the rise and sequel of it[13]. Therefore, the connection between hearing and language has been often studied in children with mild to moderate fluctuating hearing loss caused by otitis media, which can compromise language skills, especially phonological discrimination and awareness[13-17].

In children with cleft palate, there is consensus on the relationship between otitis media, hearing loss and language development, out of which transient conductive hearing loss is one of the most common causes of language disorders in this population[18]. During the child's first years of life, physicians are advised to identify, monitor and manage otitis media, from the first effusion, enabling early diagnosis. They should particularly inform parents about the natural history of the disease and the importance of follow-up, in addition, they should establish whether the child is at

greater risk for speech, language and/or learning problems[4,13].

In many craniofacial centers, the recommendation for the treatment of otitis media related to tubal dysfunction in cleft lip and palate population is tympanostomy surgery in order to place the ventilation tube, once it can benefit the recovery of hearing, and avoid important sequelae in their speech, language or learning[4,5,18,19]. Other conditions of the middle ear can also occur in this population, more frequently in children with cleft palate, such as: acute otitis media, chronic non-suppurative and suppurative otitis media with cholesteatoma and/or surgical sequelae, for example, tympanic membrane perforation. In all these cases, each condition requires specific treatment, with permanent hearing loss as a potential sequel[4,13].

Thus, in children with cleft lip and palate, there are several causes that can affect full phonological development, impacting auditory discrimination processing, phonological awareness and reading and writing development[4,21]. In recent years, studies have reported changes in aspects involving different organizational levels of language development, such as delayed speech acquisition in the first years of life, inadequate lexical content formation, failure to produce complex sentences, and poor learning performance in reading and writing, which interfere and make it hard for individuals to integrate into society[4,7-11,21]. They have also identified significant variations in cortical functions related to linguistic function, for example, impaired memory capacities, executive and attentional functions[22,23].

In the context of the phoniatric evaluation in schoolchildren with cleft lip and palate, the objective of the present study of multiple cases was to describe the interference of clinical factors in the performance of auditory perceptual language tests, focusing on auditory discrimination, auditory memory and phonological awareness, as well as the relation between them.

Methods

This is a descriptive multiple-case study, derived from partial data from the master's thesis of author Guerra MES[28], and has been approved by the ethics committee as per protocol number 2403672.

The study was carried out at a health service clinic for the diagnosis and treatment of patients with cleft lip and palate, which serves the Brazilian Unified Health System (SUS) and which is philanthropic in nature. The 30 children selected for the study were monitored from birth by a specialized interdisciplinary team and, at data collection phase, they were treated by the phoniatric physician, in a quiet clinic room, in accordance with the phoniatric evaluation published in the Treatise on Phoniatrics[24].

First, clinical factors were identified according to the following procedures: (1) Analysis of medical records to collect variables: gender, age and education at the time of evaluation; type of cleft; primary surgery (cheiloplasty and palatoplasty); otologic surgery procedures for ventilation tube insertion; in addition to duration and work aspects of the speech therapy. (2) Clinical anamnesis performed in the presence of the child's parents or other guardian aiming: to learn about the child's and his/her family's background in relation to language and hearing; to get reports regarding the development of speech and language; to ask questions concerning the child's behavior, school complaints and the child's satisfactory performance within school environment. (3) Socioeconomic analysis was based on the socioeconomic classification methodology proposed by author Graciano MIG (2013)[25] and applied to this health service. (4) Audiological evaluation to obtain the speech recognition threshold (SRT) in two phases – at data collection phase and at around three years of age -, using data from medical records. (5) Otorhinolaryngological examination performed at the time of medical appointment when aspects of the tympanic membrane (otoscopy) were described.

For keeping the study as homogeneous as possible, the children selected were only those with palate malformation, caused by dysfunctions of the muscles of velopharyngeal closure and auditory tube opening. These children were split into two groups: 17 (57%) children with transforamen incisor cleft and 13 (43%) children with post-foramen cleft. Consent for participation in the study along with an explanation on the objectives of the research as well as its possible risks were given to the children in the presence of their guardians. The children excluded were those, according to medical records, with sensorineural hearing loss, intellectual disability, moderate to severe visual impairment and other associated malformations or related syndromes. All individuals were enrolled in a regular school, from the first to the fourth year of

elementary school (*Ensino Fundamental I*); most of them belonged to a lower social class (80%); aged between 6 years and 2 months, and 9 years and 11 months (average of 8 years and 2 months of age) (standard deviation of 2 years and 2 months); there were 14 (47%) male children and 16 (53%) female.

After that, the phoniatric physician applied three tests of perceptual function of language in order to evaluate the auditory perceptual function of language: (1) standardized auditory discrimination for children aged from 5 to 9 years of age and according to male and female genders, consisting of 30 pairs of syllables split into 10 pairs of equal syllables and 20 pairs of different syllables, published by Rodrigues (1981)[24,26]; (2) auditory memory by means of numerical sequence as per age group[24;] (3) phonological awareness by means of oral productio3n for children ranging from 3 to 14 years old, classified in low, medium and high categories, and formed by the skills of syllabic synthesis, phonemic synthesis, rhyme, alliteration, syllabic segmentation, phonemic segmentation, syllabic manipulation, phonemic manipulation, transposition syllabic and phonemic transposition, published by Seabra and Capovilla (2012)[27].

Finally, a statistical analysis of the sample was carried out in two stages: descriptive analysis and statistical association between some variables.

The descriptive analysis was related to clinical factors (including age, gender, socioeconomic status, applied therapy, primary surgeries, ventilation tube insertion surgery, behavioral changes, school performance, type of cleft, otoscope and speech recognition threshold examination in three-year-old children and at data collection phase) and to results in the performance of auditory language perception tests (auditory discrimination, auditory memory and phonological awareness). The statistical association was applied to the type of cleft and to the results in the performance of auditory language perception tests with some variables (clinical factors). Variables with a percentage of less than 0.200 obtained in this analysis were considered as explanatory variables in the adjustment of a multinomial logistic regression model (Hosmer and Lemeshow, 1989). In the adjustment of a binary logistic regression model, the auditory memory is the response variable.

The type of cleft was compared with age averages through a T-Student test. The Chi-square test was applied to evaluate the association between the type of cleft and gender, socioeconomic status and school performance. A likelihood-ratio test was applied to compare the type of cleft with therapies carried out. The result in the performance of the auditory discrimination test was compared with age by means of the T-Student test and compared with SRT, in three-year-old children and at data collection phase, by means of Mann-Whitney test. The result in the performance of the auditory memory test was compared with: the age averages through the t-Student test; gender, socioeconomic status and school performance through a likelihood-ratio test; and with period of speech therapy and SRT in three-year-old children and at data collection phase through the Mann-Whitney test. In the adjustment of a binary logistic regression model, the auditory memory is the response variable.

The data of phonological awareness were published in Guerra and Novaes (2020, in press)[29].

Results

The study was composed of 30 children, who during phonological medical assessment, showed good dialogic attention and interlocution.

All children underwent primary cleft palate and/or lip corrective surgeries, and the majority of them (90%) underwent these procedures at the appropriate age according to the care service protocol – lip correction (cheiloplasty) up to 6 months of age and palate correction (palatoplasty) up to two years of age.

The descriptive data obtained from the phoniatric evaluation are shown in tables 1 and 2. According to medical record data, a speech language pathologist assessed all children throughout their development. Twenty-five of these children (83.3%) had undergone speech therapy, in a period of time that ranged from 6 months to 7 years, due to articulatory and compensatory disorders caused by: intraoral pressure loss as well as omissions and/or replacements of phonemes (53.3%); and unintelligible speech (30%), according to parents' report and medical record data.

Table 1. Descriptive summary of the clinical factors observed in the phoniatric evaluation in children with cleft lip and palate: type of cleft, audiological and otologic data and results of the tests of the auditory perceptual function of language.

Child	Type of cleft *	N. tymp. **	RO ***	LO ***	SRT R 3 years	SRT L 3 years	SRT R collection	SRT L collection	Aud. Disc.	Aud. Mem.	Phon. Awa.
1	T	1	R	R	30	60	25	35	X	X	L
2	P	0	P	P	45	35	50	40	X	X	L
3	P	0	N	R	20	55	10	40	X	X	L
4	P	0	N	N	10	20	5	5	X	X	L
5	T	0	R	R	20	30	10	25		X	L
6	T	0	N	N	25	25	0	0		X	L
7	T	3	E	R	35	55	45	45			L
8	T	2	VT	VT	40	45	15	5			L
9	T	0	R	R	10	15	40	45			M
10	T	1	T	T	15	30	10	20			M
11	T	1	R	R	10	5	5	5			M
12	P	1	T	T	10	10	5	5			M
13	P	1	N	VT	30	25	10	10			M
14	T	1	N	N	20	5	10	10			M
15	P	0	N	N	5	15	5	5			M
16	P	0	N	N	30	55	5	5			M
17	T	1	T	T	25	40	10	10			M
18	T	0	N	R	25	30	20	30			H
19	T	2	P	P	30	40	45	45			H
20	T	3	P	P	15	15	10	10			H
21	P	1	T	T	25	20	15	10			H
22	P	2	R	R	30	30	10	10			H
23	P	2	R	R	15	15	15	5			H
24	T	1	N	N	40	45	0	5			H
25	P	0	N	N	10	10	10	10			H
26	P	1	P	P	40	40	40	10			H
27	T	2	T	T	40	40	10	10			H
28	P	2	N	N	20	5	10	5			H
29	T	1	N	N	10	20	10	10			H
30	T	0	R	R	5	5	25	25			H

Key: *Type of cleft lip and palate: T – transforamen incisor cleft; P – post-foramen cleft. **Number of tympanostomy for ventilation tube insertion: 0 – has not undergone it; 1- once; 2 – twice; 3 – 3 times.***RO- otoscopy in right ear and LO - otoscopy in left ear: N- normal; R – eardrum retraction; T- tympanosclerosis plaque; P – eardrum perforation; VT - ventilation tube placed. SRT - Speech Recognition Threshold; R - right; L - left. Aud. Disc. – Auditory discrimination: X – alteration. Aud. Mem. – Auditory Memory: X – Phon. Awa. alteration.. – Phonological awareness: L – low (light grey lines=8), M – medium (medium grey lines=9); H – high (dark grey lines=13).

Table 2. Descriptive summary of the anamnesis data of the phoniatric evaluation for children with cleft lip and palate.

Child	Surgical			Speech therapy**	Speech delay@(3 years of age)		Parents' report		
	Perinatal	History	Family *	months	Phoneme omission and replacement	Inintelligible speech	Poor@ school performance	School@ com-@ plaints	Behavioral complaints
1							X	X	X
2	X			24	X	X	X	X	X
3	X						X	X	
4	X	X	CLP	48	X	X	X	X	X
5			CLP HI	50	X	X			
6				84	X	X	X	X	X
7	X		HI LANG	48			X	X	X
8	X		LANG	48	X	X	X	X	X
9		X	CLP LANG	24	X	X	X	X	X
10				12					
11				60	X		X	X	
12				48	X		X	X	
13	X	X		12					X
14				72	X	X			
15	X			6				X	
16				6					
17	X			12	X	X			X
18				18					
19				60					
20			LANG	36	X				
21			LANG						X
22	X			72	X	X	X	X	
23				60					X
24				24					
25				48	X				
26			CLP				X		X
27				48	X				X
28									
29		X		6	X				
30		X		24	X		X	X	X
N (%)	9 (30%)	3 (10%)	8 (26,7%)	25 (83,3%)	16 (53,3%)	9 (30%)	13 (43,3%)	13 (43,3%)	15 (50%)

Key: * Family history: CLP – cleft lip and palate, HI - hearing impairment and LANG – language. **Speech therapy. (Colored lines refer to the phonological awareness result: low category in light grey=8, medium category in medium grey =9 and high category in dark grey=13).

Other important data from the phoniatric evaluation for the descriptive analysis of this study were: 9 (30%) children had perinatal history; 5 (16.7%) children underwent other surgical procedures, excluding primary cheiloplasty and palatoplasty; and 8 (26.7%) children had a family history of language, hearing impairment and/or cleft lip and palate. According to parents' report, 13 (43.3%) children showed poor school performance, 13 (43.3%) reported complaints in the school environment and 15 (50%) reported complaints of behavioral changes. Regarding the data on the evaluation of the physical otorhinolaryngological examination, all children (100%) presented a palate corrected by surgical procedures, 21 (70%) children presented changes in otoscopy characterized by tympanic membrane with retraction, perforation, tympanosclerosis and ventilation tube insertion. It was observed that 19 (63.3%) children had undergone tympanotomy for ventilation tube insertion before three years of age. Six (20%) of these children underwent two ventilation tube placement procedures in the middle ear at different surgical times and 2 children (6.7%) to 3 procedures at different surgical times.

It was observed that in three-year-old children, 17 (56.7%) of them presented altered SRT on both ears, 5 (16.7%) presented altered SRT on one ear and 7 (23.3%) did not present any alterations. Besides, at the time of the research, during the data collection phase, it was found that 7 (23.3%) children had altered SRT on both ears and 4 (13.3%)

children had altered SRT on one ear. Therefore, SRT was altered in 23 (76.7%) three-year-old children and in 11 (36.7%) children in the data collection phase.

The sample of 30 children with cleft lip and palate was divided into two types of cleft palate: 13 (43%) children with post-incisive foramen cleft and 17 (57%) children with transforamen incisor cleft.

There was no significant difference regarding the distribution of children between the two groups of type of cleft and average age (p = 0.618), gender (p = 0.431), socioeconomic status (p = 0.580), school performance (p = 0.785) and the number of tympanotomies (p = 0.379).

Most subjects have undergone speech language pathology therapy (Table 3), but the percentage of individuals who have undergone therapy and had transforamen cleft (94.1%) is greater than the percentage of individuals with post-foramen cleft (69.2%). There was no significant difference between the percentages of individuals who have undergone therapy in the two types of cleft, but the difference almost reached a level of significance (p = 0.066). The descriptive summary of the period of therapy for the 25 subjects who have undergone therapy is shown in Table 4.

Table 3. Frequency and percentage distributions of speech therapy by type of cleft lip and palate.

TYPE OF CLEFT	Speech-language pathology therapy		Total
	No	Yes	
POST-FORAMEN	4	9	13
	30.8%	69.2%	100%
TRANSFORAMEN	1	16	17
	5.9	94.1	100
TOTAL	5	25	30
	16.7%	83.3%	100%

There was no significant difference between the percentages of individuals who have undergone therapy in the two types of cleft. (p=0.066).

Table 4. Descriptive summary of the period of speech therapy (months) by type of cleft lip and palate.

TYPE OF CLEFT	N	Average (months)	Standard deviation	Minimum (months)	Median (months)	Maximum (months)
POST-FORAMEN	9	36.0	24.6	6	48	72
TRANSFORAMEN	16	39.1	23.1	6	42	84
TOTAL	25	38.0	23.2	6	48	84

In this study, twenty-five (83.3%) children have undergone therapy and the period ranged from 6 months to 84 months (7 years).

The results in the performance tests of the auditory perceptual function of language are shown in Table 1. It can be observed that only 4 (13.3%) children presented performance below the expected for age and gender in the auditory discrimination test. In the sequential auditory memory test for numbers, 6 children (20%) presented a result below the expected for their age. In addition, in the phonological awareness test, 8 (26.7%) children presented a low category result, 9 children (30%) presented a medium category result and 13 (43.3%) children presented a high category result[29].

The descriptive analysis of auditory discrimination, in which only 4 children (13.3%) presented alterations, demonstrated no significant association between auditory discrimination and: age (p = 0.483); SRT in three-year-old children (p = 0.142); SRT at data collection phase (p = 0.180).

The performance in the auditory memory test was associated with type of cleft, age, gender, socioeconomic status, period of the speech therapy, school performance and SRT in three-year-old children and at data collection phase (Table 5). The distributions of auditory memory percentages between the two types of cleft were similar (Table 5), and there was no significant difference between the probability distributions between the two types of cleft and auditory memory (p = 0.713) (likelihood-ratio test). There was no significant difference between the probability distributions in both genders (p = 0.855) and in socioeconomic status (p = 0.082). The average age observed in the group with adequate auditory memory was higher than in the group with inadequate memory (Table 6). However, the difference was not significant between age averages and auditory memory categories (p = 0.127) (T-Student test). The highest average (34.3 months) and the highest median of speech therapy period (36 months) were observed in children with inadequate performance in the auditory memory test. Nevertheless, there was no significant difference between the distributions of speech therapy period between the two categories of auditory memory (p = 0.917) (Mann-Whitney test) (table 7). The percentage of children with adequate auditory memory was higher in the group with satisfactory school performance (94.1%), compared to the group with poor school performance. The difference was significant (p = 0.024), that is, the probability of adequate auditory memory was significantly higher in the group with satisfactory school performance (Table 8). Averages and medians for SRT in three-year-old children and at data collection phase were worse in children with inadequate auditory memory than in children with adequate auditory memory, but there were no significant differences between auditory memory and distributions between SRT in three-year-old children (p = 0.212) and at data collection phase (p = 0.507) (Mann-Whitney test) (Table 9).

Table 5. Frequency and percentage distributions of performance in the auditory memory evaluation by type of cleft.

TYPE OF CLEFT	Adequate auditory memory	
	NO	YES
POST-FORAMEN (N.13)	3	10
	23.1%	76.9%
TRANSFORAMEN (N.17)	3	14
	17.7%	82.4%
TOTAL (N.30)	6	24
	20%	80%

There was no significant difference between the probability distributions in the two types of cleft and auditory memory (p = 0.713) (likelihood-ratio test).

Table 6. Distribution of children with cleft lip and palate in the auditory memory test according to age at evaluation phase (months).

Tests	Category	N	Average age (months)	Standard deviation	Minimum (months)	Median (months)	Maximum (months)
Adequate Auditory memory	NO	6	90.3	13.1	74	88	113
	YES	24	100.4	11.6	73	101.5	119
	TOTAL	30	98.4	12.4	73	101	119

There was no significant difference between the age average and auditory memory categories (p = 0.127) (T-Student test).

Table 7. Distribution of children with cleft lip and palate in the auditory memory test according to the period of speech therapy (months).

Test / Speech therapy	Category	N	Average period (months)	Standard deviation	Minimum (months)	Median (months)	Maximum (months)
Adequate Auditory memory	NO	6	34.3	32.8	0	36	84
	YES	24	31.0	24.2	0	24	72
	TOTAL	30	31.7	25.6	0	24	84

There was no difference between the distributions of period of therapy in the two categories of auditory memory (p = 0.917) (Mann-Whitney test).

Table 8. Distribution of children with cleft lip and palate in the auditory memory test according to school performance (satisfactory: yes or no).

Satisfactory school performance	Satisfactory auditory memory		
	NO	YES	TOTAL
NO	5	8	13
	38.5%	61.5%	100
YES	1	16	17
	5.9%	94.1%	100

The percentage of children with adequate auditory memory in the group with satisfactory school performance (94.1%) is higher than in the group with poor school performance. The difference was significant (p = 0.024), that is, the probability of adequate auditory memory was higher in the group with satisfactory school performance (likelihood-ratio test).

Table 9. Distribution of children with cleft lip and palate in the auditory memory test according to speech recognition threshold (SRT = dBHL) in both ears at 3 years of age and at data collection phase.

SRT	Adequate Auditory memory	N	Average dBHL	Standard deviation dBHL	Minimum dBHL	Median dBHL	Maximum dBHL
SRT 3 years of age	NO	6	31.3	11.4	15	31.25	45
	YES	24	24.0	13.4	5	22.5	45
	Total	30	25.4	13.1	5	25	45
SRT at data collection phase	NO	6	20.4	16.6	0	21.25	45
	YES	24	15.2	12.7	2.5	10	45
	Total	30	16.3	13.4	0	10	45

There was no difference between auditory memory and distributions between SRT in three-year-old children (p = 0,212) and at data collection phase (p = 0,507) (Mann-Whitney test)

Statistics data about the performance on phonological awareness skills were described in the article under analysis in order to be published by Guerra and Novaes[29]. There was no significant difference between phonological awareness and the probability distributions on two types of cleft, two genders and on socioeconomic status. The highest average age was higher in the high category group and the poor school performance and the longer period of speech therapy were more frequent in the low category group. The worst SRT result observed on three-year-old children and at data collection phase were of the group of children with results in the low category, and there was a significant association only between phonological awareness and SRT in three-year-old children (Guerra and Novaes, in press) [29].

Discussion

The present study evaluated children with cleft lip and palate who had been monitored since birth by a referral health service center equipped with a specialized interdisciplinary team for the treatment of the functional and anatomical conditions caused by the anomaly, and therapy oriented to their speech and language difficulties.

In view of the great variability of the clinical characteristics presented by the studied group and, therefore, the difficulty to establish a control group, the discussion was carried out by applying data from the available literature.

Most of the children (63%) studied underwent tympanotomy for ventilation tube insertion before

the first three years of life. According to Kuo et al. (2014), the occurrence of otitis media with effusion is common in the first years of life of babies with cleft palate, which can lead to conductive hearing loss and, consequently, negatively influence language development[18].

Briscoe, Bishop and Norbury (2001) have reported that the auditory perceptual function is a complex process, and changes that affect it can lead to problems in the phonological structure of language, with consequent adverse effects on the acquisition of vocabulary and literacy[16]. The findings of the present study generally support the statements of Briscoe et al. (2001), especially with regard to the performance of 8 (27%) children in phonological awareness, 6 (20%) children in auditory memory, and 4 (13%) children in auditory discrimination. It has also been observed that there was a direct association between poor performance in phonological awareness and auditory memory tests and unsatisfactory findings regarding poor school performance in 43.3% of the children studied.

Many studies have shown impairment in learning and poor academic performance in the population with cleft lip and palate associated with disorders in the capacities of linguistic functions [7,9,10,11,22,23]. From the point of view of phoniatrics, it has been observed that not only is school performance a reflection of a cascade effect of various situations and conditions in which this population is inserted but that somehow school performance also affects the construction of language[4,5]. Tabith Junior (2002) comments that the factors that interfere with the auditory function as well as the emotional aspects related to facial aesthetics (disfigurement), speech quality and affective relationships can alter the language construction process. Therefore, they are essential for children to use language as a key instrument of interpersonal relationships, which is important for school adaptation in childhood, social adjustment in adolescence and professional and social performance in adult life[4].

It has also been observed that children in the older age group presented a result that was considered high in the phonological awareness test (above 8 years and 11 months of age) and adequate auditory memory (above 8 years and 4 months of age). Firstly, this performance suggests that the fact that these children have been under the care of a referral health service, in which they have benefited from speech therapy according to the demand of each individual, with the purpose of evaluating and giving guidance on speech and language aspects may have contributed to the performance of these children in these age groups. Another important factor to be considered as a potential nurturing element for this better performance is the training of phonological awareness and auditory memory (a requirement, in many schools, for the learning process of reading and writing), which has possibly taken place in the course of literacy[27].

Child 3 – who, at the evaluation phase, was 9 years and 5 months old and was in the 4th year of elementary school – is highly noted for presenting changes in all administered tests. This child in particular presented perinatal changes; had not undergone speech therapy and the child's parents reported school difficulties. This child presented also tympanic membrane retraction on the right side in the otoscope exam and altered SRT result both at the age of three years old and at data collection phase, and was not submitted to tympanotomy for ventilation tube insertion (despite the history of hearing impairment). Some important aspects stand out: personal history may have influenced the neural functioning of cognitive areas and the results of auditory function, thus interfering in auditory processing and, consequently, in the biological maturation of complex areas of language. In addition, the fact that this child had not undergone the recommended speech language pathology therapy may have contributed to the child's worst performance in phonological awareness skills already impaired by the history of otologic and audiological alterations.

In this study, most children (83.3%) underwent therapy and there was no significant difference between the groups according to the type of cleft. The literature highlights that the surgical treatment of the palate aims to close the communication between the oral and nasal cavity and establishes anatomical conditions for adequate functioning of the velopharyngeal mechanism and the auditory tube, mainly targeting on improving the conditions of intraoral pressure and airflow. However, it does not necessarily imply an improvement in speech production and functioning of the auditory tube, requiring speech language pathology therapy as early as possible, which, in addition to preventing or correcting changes in speech sounds, also

lessens the negative impact of the communication disorder in the development of language skills that can reflect on literacy[4,5,9]. Therefore, it can be observed from the descriptive summaries that children with the worst performance in the tests have undergone a longer period of speech therapy, suggesting that children with greater difficulties required specialized therapies owing to various individual demands.

In the context of phonological assessment, child 6 (7 years and 10 months old) is highly noted for presenting changes in the auditory memory tests and phonological awareness despite the child's seven years of speech therapy, the longest period of speech therapy a participant had been submitted to. According to parents' report, the child presented language delay (characterized by omissions and replacements of phonemes and unintelligible speech), difficulties at school and behavioral issues.

As stated on the child's clinical history, at 3 years of age, the child presented SRT alteration in both ears and, at the time of data collection, it was observed that the tympanic membrane and the SRT were within normal aspects in both ears. It is suggested that child 6 presented behavioral and cognitive issues that negatively influenced the development of the tests as both auditory memory and phonological awareness are cortical functions skills that depend on attentional auditory and cognitive factors, as well as others external emotional factors. The auditory discrimination test was performed based on the differentiation of phoneme sounds in a quiet environment and had a better result compared with the other auditory perceptual tests. Auditory discrimination is the ability to distinguish speech sounds and it is a complex process of differentiation[1,24]. In this sense, worse results would be expected to be seen in a population with a clinical history of fluctuating or prolonged hearing loss, as it was observed by the analysis of SRT in two different moments (in three-year-old children and at data collection phase). Thus, one of the limitations of this study is that the analysis of discrimination was performed only with phoneme sounds, excluding complex sounds or words; in a quiet environment with the examiner's presence (which would be a source of sound). This does not necessarily reflect the reality of everyday environments that are mostly noisy. Future studies should consider the present findings and expand the assessment situations.

Child 4 stands out from the four children who had alterations in all tests. This child had a family history of language delay and a case of cleft lip and palate, in addition to a personal history of perinatal disorders and speech delay. The child's SRT at three years of age and at data collection phase, as well as the aspects of the tympanic membrane were normal. In this case, the condition of the audiological and otologic aspects was not relevant in determining the low results of the auditory perception skills. However, other biological elements resulting from situations in the first years of life, such as personal background, may have been the aggravating factors in the acquisition of language and cortical cognitive functions.

In general, most of the children that participated in the present study, despite significant changes in many of the analyzed criteria, performed satisfactorily in the auditory perceptual function tests. These results demonstrated the effectiveness of early intervention and constant monitoring of children's development by specialized professionals. Moreover, the effectiveness of treatment strategies recommended by the referral health service center where the study was carried out suggests that these are the most relevant protective aspects that provided this population with adaptation mechanisms to develop their language skills. Therefore, the target audience of this study represented children who had been treated with the recommended clinical and surgical care within the expected time. In addition, the phoniatric consultation also enabled the identification of aspects not directly related to the anatomical and functional condition of the cleft, but which somehow interfered in the language development of these children and therefore, proving to be a clinical evaluation of great relevance for this population.

Conclusion

In the context of the phoniatric evaluation, the present study has enabled the identification of clinical factors of the cleft, which can interfere in the language development of these children. Auditory memory and phonological awareness were the most affected auditory-perceptual aspects, but they did not always explain school performance. The background and/or behavioral changes in each child as indicators of risk and the absence of therapy were in some cases explanatory factors regarding

school performance, even when perceptual aspects were not altered.

The characteristics and heterogeneity of the studied population represent different clinical conditions of children monitored at a referral health center specialized in the treatment of CLP. The good performance of most of the studied subjects suggests early intervention, constant monitoring by specialized professionals and the treatment strategies recommended by the referral health service center are protective aspects in relation to the development of language skills.

References

1. Tabith Junior A, Favero ML, Pirana S. História da foniatria. In: Pirana S, Favero ML. Tratado de foniatria. 1 Ed. Rio de Janeiro: Thieme Revinter. 2020. p.1-4.

2. Spinelli M. Foniatria: Introdução aos distúrbios da comunicação. Audição / Linguagem. São Paulo. Editora Moraes. 1983.

3. Quirós JB e Gotter R. El lenguaje em el Niño. Buenos Aires. CEDIFA. 1964.

4. Tabith Junior A. Distúrbios da comunicação em portadores de fissuras labiopalatais: aspectos foniátricos. In: Mélega JM. Cirurgia plástica – Fundamentos e Arte. Rio de Janeiro: Editora Guanabara; 2002. p.25-8.

5. Tabith Junior A, Guerra MES. Fissuras labiopalatinas. In: Pirana S, Favero ML. Tratado de foniatria. Rio de Janeiro: Thieme Revinter; 2020. p.161-70.

6. Burg ML, Chai Y, Yao, CA, Magee W3rd, Figueiredo JC Epidemiology, Etiology, and Treatment of Isolated Cleft Palate. Front. Physiol. 2016; 7: 67.

7. Lamônica DAC, Silva-Mori MJF, Ribeiro CC, Maximino LP. Receptive and expressive language performance in children with and without cleft lip and palate. CoDAS. 2016; 28(4): 369-72.

8. Salas-Provance MB, Kuehn, DP, Marsh JL.Phonetic repertoire and syllable characteristics of 15-month-old babies with cleft palate. J. Phonetics. 2003; 31(1): 23-38.

9. Chapman KL. The relationship between early reading skills and speech and language performance in young children with cleft lip and palate. The American Cleft Palate-Craniofacial Association. 2011; 48(3): 301-11.

10. Ha S. Effectiveness of a parent-implemented intervention program for young children with cleft palate. Int J Pediatr Otorhinolaryngol. 2015; 79(5): 707-15.

11. Priester GH, Goorhuis-Brouwer SM. Speech and language development in toddlers with and without cleft palate. Int J Pediatr Otorhinolaryngol. 2008; 72(6): 801-6

12. Fenha M, Santos EC, Figueira L. Avaliação das dimensões cognitivas e sócio-afectivas de crianças com fenda lábio-palatina. Psicologia: Saúde e Doenças. 2000; 1(1): 113-20.

13. Rosenfeld RM, Shin JJ, Schwartz SR, Coggins R, Gagnon L, Hackell JM et al. Clinical Practice guideline: otitis media with effusion (update). Otolaryngology - head and neck surgery. 2016; 154(IS) SI-S41.

14. Zumach A, Gerrits E, Chenault MN, Anteunis LJC. Otitis Media and Speech-in-Noise Recognition in School-Aged Children. Audiol Neurotol 2009; 14: 121-129.

15. Mody M, Schwartz RG, Gravel JS, Ruben RJ. Speech perception and verbal memory in children with and without histories of otitis media. J Speech Lang Hearing Res. 1999 Oct; 42: 1069-79.

16. Briscoe J, Bishop DVM, Norbury CF. Phonological processing, language and literacy: A comparison of children with mild–moderate sensorineural hearing loss and those with specific language impairment. J Child Psychol Psychiatry 2001; 42(3): 329–40.

17. Williams CJ, Jacobs AM. The impact of otitis media on cognitive and educational outcomes. Med J Aust. 2009; 191(9): 69-72.

18. Kuo CL, Tsao YH, Cheng HM, Lien CF, Hsu CH, Huang CY, et al. Grommets for otitis media with effusion in children with clef palate: a systematic review. Pediatrics. 2014 Nov; 134(5): 983-94.

19. Tengroth B, Hederstierna C, Neovius E, Flynn T. Hearing thresholds and ventilation tube treatment in children with unilateral cleft lip and palate. Int J Pediatric Otorhinolaryngol. 2017; 97: 102-8.

20. Amaral MIR, Martins JE, Santos MFCS. Estudo da audição em crianças com fissura labiopalatina não-sindrômica. Braz J otorhinolaryngol. 2010; 76(2): 164-71.

21. Pamplona MDC, Ysunza PA. Total immersion speech camps for patients with cleft palate. Journal of Cleft lip palate and Craniofacial Anomalies. 2017 Nov; 4(3): 132-8.

22. Goldsberry G, O'leary D, Hichwa R, Nopoulos P. Functional abonormalities in the neural circuitry of Reading in men with nonsyndromic clefs of the li por palate. Clef palate craniofac J. 2006 Dec; 43(6): 683-90.

23. Lemos ICC, Feniman MR. Teste de Habilidade de Atenção Auditiva Sustentada (THAAS) em crianças de sete anos com fissura labiopalatina. Braz J Otorhinolaryngol. 2010 Mar-Apr; 76(2): 199-205.

24. Dualibi APF, Favero ML, Pirana S. A consulta Foniátrica. In: Pirana S, Favero ML. Tratado de foniatria. 1 Ed. Rio de Janeiro: Thieme Revinter. 2020. p.119-28.

25. Graciano MIG. Estudo Socioeconômico: Um instrumento técnico-operativo. São Paulo: Veras; 2013.

26. Rodrigues N. Neurolinguística dos distúrbios da fala. São Paulo: Editora Cortez; 1992.

27. Seabra AG, Capovilla FC. Teste de consciência fonológica por produção oral. In: Seabra AG, Dias NM. Avaliação neuropsicológica cognitiva: linguagem oral. Vol 2. São Paulo: Memnon; 2012. p.117-22.

28. Guerra MES. Crianças com fissura labiopalatina: relação entre percepção auditiva e características audiológicas e otológicas no contexto da avaliação foniátrica (dissertação). São Paulo: Pontifícia Universidade de São Paulo; 2019.

29. Guerra MES, Novaes BCAC. Clef lip and Palate: relationship between phonological awareness and audiological characteristics in children. Cleft Palate Craniofac J. no prelo 2020.