

Katia Ignacio Meneguetti¹
Laura Davison Mangilli²
Nivaldo Alonso¹
Claudia Regina Furquim de
Andrade¹

Keywords

Cleft Palate
Speech
Surgery
Velopharyngeal Insufficiency
Speech, Language and Hearing
Sciences

Descritores

Fissura palatina
Fala
Cirurgia
Insuficiência velofaríngea
Fonoaudiologia

Correspondence address:

Claudia Regina Furquim de Andrade
Rua Cipotânea, 51, Cidade
Universitária, São Paulo (SP), Brazil,
CEP: 05360-160.
E-mail: clauan@usp.br

Received: September 02, 2016

Accepted: April 30, 2017

Speech profile of patients undergoing primary palatoplasty

Perfil da fala de pacientes submetidos à palatoplastia primária

ABSTRACT

Purpose: To characterize the profile and speech characteristics of patients undergoing primary palatoplasty in a Brazilian university hospital, considering the time of intervention (early, before two years of age; late, after two years of age). **Methods:** Participants were 97 patients of both genders with cleft palate and/or cleft and lip palate, assigned to the Speech-language Pathology Department, who had been submitted to primary palatoplasty and presented no prior history of speech-language therapy. Patients were divided into two groups: early intervention group (EIG) – 43 patients undergoing primary palatoplasty before 2 years of age and late intervention group (LIG) – 54 patients undergoing primary palatoplasty after 2 years of age. All patients underwent speech-language pathology assessment. The following parameters were assessed: resonance classification, presence of nasal turbulence, presence of weak intraoral air pressure, presence of audible nasal air emission, speech understandability, and compensatory articulation disorder (CAD). **Results:** At statistical significance level of 5% ($p \leq 0.05$), no significant difference was observed between the groups in the following parameters: resonance classification ($p=0.067$); level of hypernasality ($p=0.113$), presence of nasal turbulence ($p=0.179$); presence of weak intraoral air pressure ($p=0.152$); presence of nasal air emission ($p=0.369$), and speech understandability ($p=0.113$). The groups differed with respect to presence of compensatory articulation disorders ($p=0.020$), with the LIG presenting higher occurrence of altered phonemes. **Conclusion:** It was possible to assess the general profile and speech characteristics of the study participants. Patients submitted to early primary palatoplasty present better speech profile.

RESUMO

Objetivo: caracterizar o perfil e a fala de pacientes submetidos à palatoplastia primária em um hospital escola de São Paulo, levando-se em consideração a idade do paciente no momento da cirurgia (precoce até os 2 anos de idade e tardio após 2 anos). **Método:** 97 indivíduos, de ambos os gêneros, com diagnóstico de fissura de palato associada ou não à de lábio, divididos em dois grupos: 1) grupo precoce (GP), composto por 43 indivíduos operados até o segundo ano de vida; 2) grupo tardio (GT), composto por 54 indivíduos operados após o segundo ano. Os participantes foram submetidos à avaliação clínica fonoaudiológica. Os parâmetros avaliados e considerados para o estudo foram: classificação da ressonância, presença de ronco nasal audível, ocorrência de fraca pressão intraoral, ocorrência de emissão nasal, classificação da inteligibilidade de fala e presença de distúrbios articulatorios compensatórios (DACs). Uma porcentagem randomicamente selecionada de participantes (30%) foi reavaliada por mais duas fonoaudiólogas e a comparação entre os juízes indicou alta concordância. **Resultados:** (nível de significância de 5%): os grupos não se diferenciaram em relação à classificação da ressonância ($p=0,067$), grau de hipernasalidade ($p=0,113$), presença de ronco nasal ($p=0,179$), ocorrência de fraca pressão intraoral ($p=0,152$), ocorrência de emissão nasal ($p=0,369$) e classificação da inteligibilidade de fala ($p=0,113$). Em relação à presença de DACs, os grupos se diferenciaram ($p=0,020$), com maior ocorrência de fonemas alterados no GT. **Conclusão:** foi possível caracterizar o perfil geral e de fala dos pacientes submetidos à palatoplastia primária do referido hospital escola. Concluiu-se que a realização da cirurgia precocemente traz melhores resultados em relação à fala.

Study carried out at Departamento de Fisioterapia, Fonoaudiologia e Terapia Ocupacional, Faculdade de Medicina, Universidade de São Paulo – USP - São Paulo (SP), Brazil.

¹ Universidade de São Paulo – USP - São Paulo (SP), Brazil.

² Universidade de Brasília – UnB - Brasília (DF), Brazil.

Financial support: nothing to declare.

Conflict of interests: nothing to declare.

INTRODUCTION

Cleft lip and palate is the most common congenital anomaly in the human race. It may occur in isolation or associated with other complex malformations, bringing aesthetic and functional consequences which require lifelong rehabilitation^(1,2). It is the most frequently treated congenital anomaly in craniofacial surgery centers, with incidence of 1:600 live births⁽³⁾. Several anatomical and functional changes are observed as a consequence of cleft lip and palate, causing alterations in midface growth, hearing, and velopharyngeal sphincter function, with the latter interfering directly with the functions of sucking, swallowing, and speaking⁽⁴⁾.

One of the main objectives of cleft palate repair is to establish adequate velopharyngeal function, so that communication development can occur as most appropriately as possible. The chances of adequate speech development decrease significantly when late repair occurs⁽⁵⁻⁷⁾. A previous study⁽⁶⁾ reported statistically significant difference between early and late operated individuals with respect to presence of phonological processes, with these processes appearing less frequently in the group that underwent early surgery.

Failure in velopharyngeal closure, either due to the presence of cleft or velopharyngeal dysfunction, with respect to speech-related aspects, results in loss of part of the airway to the nasal cavity, leading to the appearance of symptoms that may directly or indirectly impair speech understandability⁽⁸⁾. Hypernasality, audible nasal air emission, and weak intraoral air pressure are considered direct or primary consequences of velopharyngeal closure failure^(6,8), whereas compensatory articulation disorders (CAD) - strategies used by individuals with the objective of compensating in other points of the vocal tract the inability to impose pressure to the oral cavity - are defined as indirect or secondary consequences^(6,8).

During adequate functioning of the velopharyngeal mechanism, it is necessary that the soft palate and the posterior and lateral walls of the pharynx perform a wide and synchronized movement, allowing contact between these structures and ensuring total separation between the oral and nasal cavities during the production of oral sounds⁽⁹⁾. Inadequacy of this function causes infants with cleft lip and palate, even in the first attempts of vocalization in the second and third months of life, to produce sounds differently compared with infants without cleft. At 6 months of age, infants begin to produce their first anterior consonants and, in the case of children with cleft, there may be difficulties in performing this task, leading to glottal or pharyngeal articulations⁽¹⁰⁾.

The first year of a child is of fundamental importance for the acquisition of speech and language⁽¹⁰⁾. The specific scientific literature clearly reports that surgical repair of the cleft palate should be performed early, before two years of age^(5,6,11), especially when considering speech^(5,12). The surgical treatment aims to provide adequate velopharyngeal function, which is essential in early childhood⁽⁶⁾. To this end, different palatoplasty techniques have been developed over time aiming to achieve adequate

palate length, in order to allow velopharyngeal closure. Another aspect addressed is the reorientation of the muscle fibers of the palate, creating a muscle strap that will allow better mobility of the soft palate⁽⁵⁾.

Nevertheless, despite the progress made in this area, it is still difficult to predict the efficiency of muscle function for speech after palatoplasty^(13,14). Although there is recognized effort to establish satisfactory velopharyngeal function, a study using magnetic resonance imaging and computerized reconstruction showed that up to 35% of individuals with cleft palate may remain with velopharyngeal insufficiency after surgery⁽¹³⁾. This insufficiency may be due to inadequate dissection of the palatine musculature, insufficient length of the palate, or anatomical changes of the pharynx posterior wall⁽¹³⁾. Another study showed that approximately 30% of patients might remain with changes in articulation and speech resonance⁽¹⁴⁾ after palatoplasty.

In Brazil, as in other developing countries, it is not always possible to follow the stages of cleft lip and palate treatment advocated and conducted by most international and reference national centers, because as previously mentioned, due to socioeconomic and cultural factors, parents seek treatment when the child is already at an older age, past the period of language acquisition, often in adult age^(2,15,16).

Speech alterations associated with velopharyngeal dysfunction would be identified through clinical speech assessment performed by speech-language pathologists and by instrumental evaluation of the velopharyngeal function. The results of these evaluations allow determination of better treatment and prognosis to the case⁽¹⁷⁾.

Perceptual clinical evaluation of speech is considered the main indicator of velopharyngeal dysfunction, and it is an essential part of clinical diagnostics⁽¹⁸⁾. Although subjectively, this assessment allows identification of changes, measurement of severity, and evaluation of effectiveness of the treatments performed^(18,19). Over time, studies⁽¹⁹⁾ have been concerned with improving auditory-perceptual assessment in order to make it less susceptible to errors arising from its subjectivity. Thus, adoption of criteria of scores to represent the speech-language pathologist's judgment is suggested. Among the different scales developed to measure resonance and other characteristics of speech, the most popular is the scale with equal intervals, in which the rater assigns a score to the aspect analyzed in a linear scale, where the smallest number indicates absence of alterations and the highest one denotes the maximum degree of change. Therefore, on a six-point scale in the assessment of hypernasality, a score of one indicates absence of hypernasality and a score of six shows its most severe degree⁽²⁰⁾.

Two domestic studies^(6,21) verified a relation between speech alterations and time of palatoplasty, both considering the best time for surgery until 18 months of age. Palandi and Guedes (2011)⁽⁶⁾ concluded that individuals who underwent late intervention showed statistically significant difference regarding the presence of some phonological processes, and considered that the age at which palatoplasty is performed is an intervening factor for the speech of individuals with cleft palate. The results reported

by Mituuti et al. (2010)⁽²¹⁾ indicate that few individuals present speech changes after cleft repair (ranging from a minimum of 8%, with poor intraoral air pressure, to a maximum of 26%, with CADs).

In view of the presented information, the objective of this study is to characterize the profile and speech characteristics of patients undergoing primary palatoplasty in a university hospital (HCFMUSP) in the city of Sao Paulo, Brazil, considering the time of intervention (early, before two years of age; late, after two years of age). It is a national referral center for cases of cleft lip and palate which receives a large number of new cases annually. Some of these patients start their treatment at the institution, whereas others choose the hospital to continue care. Due to the great demand, the institution identified the necessity to characterize the patients' speech changes, regardless of time of intervention, in order to initiate discussion and propose improvements in the care of these individuals.

METHODS

This is an observational, cross-sectional study whose procedures of selection and assessment of participants only started after the pertinent ethical processes had been completed: approval by the Ethics Committee on Human Research of the aforementioned Institution (CAPPesq HCFMUSP 513.535) and signing of an Informed Consent Form (ICF) by the parents and/or guardians.

Study participants were 97 individuals with diagnosis of non-syndromic cleft palate and/or cleft lip and palate who were referred to speech-language pathology evaluation between May 2011 and July 2015 according to the service's demand. The participants were divided into two groups: Early Intervention Group (EIG) and Late Intervention Group (LIG). The EIG was composed of individuals who had undergone primary palatoplasty up to the second year of life and the LIG comprised individuals who had been submitted intervention after the age of two⁽⁶⁾.

Medical diagnosis and monitoring of the participants were conducted by the Craniomaxillofacial Surgery Team of the HCFMUSP's Department of Plastic Surgery and Burns as a collaboration which evolves the assistance of plastic surgeons and otolaryngologists.

Study inclusion criteria were as follows: individuals 1) of both genders; 2) with minimum age of six years, and no maximum age restriction; 3) diagnosed with non-syndromic cleft palate and/or cleft lip and palate; 4) submitted to palatoplasty at least three months prior to speech-language pathology assessment; 5) who had not undergone previous speech-language pathology therapy and/or treatment with prostheses.

Minimum age of the participants was established based on the studies by Andrade et al. (2004)⁽²²⁾, who advocate that the phonological processes and development of oral language in children are already completed at this age. The use of this criterion aims to exclude the language acquisition variable in this study.

Exclusion criteria comprised: 1) minor surgeries on the palate; 2) presence of neurological disorders; 3) history of face trauma; 4) presence of respiratory diseases; 5) presence

of palate fistulas of any length; 6) presence of speech-language pathology comorbidities (language and auditory complaints and deficits); 7) presence of cognitive impairment or level of consciousness that would prevent evaluators from understanding the verbal information required for speech-language assessment and treatment.

As previously mentioned, some of the study participants were operated at the HCFMUSP and some underwent surgery elsewhere; therefore, it was not possible to control the variables type of surgical procedure and/or number of surgeons involved in these procedures.

All participants were submitted to routine speech-language assessment at the Speech-language Pathology (SLP) Clinic of the HCFMUSP. The evaluations were conducted by a trained speech-language therapist experienced in the area, who belonged to the SLP Team responsible for assistance at the Cleft Lip and Palate Outpatient Clinic.

The data used in this study comprise the range of aspects evaluated during the routine assessment, more specifically the Auditory-perceptual Assessment of Speech. A protocol specifically developed by the Clinic is used in these evaluations. It was prepared by the SLP Clinic after extensive literature review and participation in thematic meetings aimed at standardizing assessment protocols for cleft palate patients. This protocol includes an evaluation of the morphological aspects of the oral motor system, occlusion, and speech. The latter is assessed through analysis of the speech samples, which comprise segments of directed speech (repetition of words and sentences) with recurrence of the target sound, containing all the phonemes of Brazilian Portuguese; automatic speech during counting from one to 10; and spontaneously speech, requesting the patient to report a daily event. To verify the occurrence of nasal air escape during speech, a nasal air emission test is conducted using a Glatzel mirror.

Samples were collected in a SLP room with environment specifically treated for this purpose. The participants remain seated and the voice samples are recorded using a unidirectional microphone positioned at the standard distance of 10 cm away from their mouths at a 45° angle. The stimuli captured by the microphone are digitized using the Praat.wav software with 22.050 Hz sampling frequency and 16-bit resolution.

The following parameters are assessed by the protocol: presence of alterations in resonance, severity of resonance alteration, presence of compensatory articulation disorders (CAD), presence of nasal turbulence, presence of weak intraoral air pressure, presence of audible air emission, and rating of speech understandability - variables considered in the present study.

The samples were analyzed according to the following criteria: 1) presence or absence of hypernasality, with severity rating when present: mild, moderate, or severe; 2) presence or absence of hyponasality, with severity rating when present: mild, moderate, or severe; 3) presence or absence of CADs; 4) determination of the types of CADs present and the number of occurring altered phonemes; 5) presence of nasal turbulence; 6) presence of weak intraoral air pressure; 6) presence or absence of audible nasal

air emission; 7) speech understandability rating: adequate or inadequate, with severity ratings: mild, moderate, or severe.

Hypernasality was considered present when excessive nasal resonance was verified during production of oral sounds⁽⁸⁾ in at least one phoneme of each of the analyzed samples. Hyponasality was identified when decreased or insufficient nasal resonance was verified during production of nasal sounds⁽⁸⁾ in at least one phoneme of each of the analyzed samples.

CADs are called compensatory because they are an attempt to compensate for the difficulty in producing a phoneme at the appropriate place of articulation. They are considered learning disorders related to structural changes, especially with the presence of weak intraoral air pressure for production of plosive, fricative and affricate consonants. In this study, CADs were considered present when there was replacement of orally articulated sounds with sounds articulated at points short of the place of failure, in an attempt to approximate the acoustic result as close as possible to what is considered as the standard sound⁽²³⁾ in at least one speech phoneme of the participant.

The main CADs are glottal stop, pharyngeal plosive, mid-dorsum palatal plosive, pharyngeal fricative, velar fricative, posterior nasal fricative, and nasal fricative⁽²⁴⁾.

Presence of nasal turbulence was defined as the result of air friction between the soft palate and the pharyngeal walls owing to non-closure of the velopharynx⁽²⁴⁾ in at least one phoneme of each of the analyzed samples.

Weak intraoral air pressure was considered present when reduced air pressure was maintained in the oral cavity during production of speech oral sounds⁽¹⁹⁾ in at least one phoneme of each of the analyzed samples.

Nasal air emission was considered present when audible escape of air to the nasal cavity was observed during speech production⁽¹⁹⁾ in at least one phoneme of each of the analyzed samples.

For speech understandability analysis, all speech samples of the participants were considered: directed speech, automatic speech, and mainly spontaneous speech.

Inter-reliability of the speech-language pathologists responsible for the Cleft Lip and Palate Outpatient Clinic

All speech-language pathologists responsible for assistance at the Cleft Lip and Palate Outpatient Clinic attended specific training at the Speech-language Pathology and Audiology Service of the HCFMUSP, and only started their practice after achieving a high degree of agreement with their peers.

However, to ensure reliability to the study, a randomly selected percentage of participants (30%, n=31) were reevaluated by the other speech-language therapists involved in the Cleft Lip and Palate Outpatient Clinic, following the same criteria previously described. Each rater did not have access to the previously conducted evaluations, and the results were used to determine the reliability between them.

Table 1 shows the analysis of the results presented independently by the three speech-language pathologists responsible for the

Table 1. Inter-rater reliability analysis – Kappa coefficient

Variable	K	<i>p</i>	Kappa confidence interval
Hypernasality	1.00	<0.001	0.76-1.0
Severity Rating	0.730	<0.001	0.56-0.90
Hyponasality	1.00	<0.001	0.76-1.0
Severity Rating	0.573	<0.001	0.41-0.74
Speech understandability	1.00	<0.001	0.76-1.0
Rating	0.909	<0.001	0.75-1.0

Caption: K = Kappa coefficient, *p* = statistical significance

consultations, referring to resonance and speech understandability. The inter-rater comparison indicated high agreement for all variables considered, confirming the reliability of the analysis. The inter-rater concordance analysis was calculated by the Kappa coefficient.

Data analysis

Data were submitted to statistical analysis using the SPSS.22 software. Descriptive analysis was performed by the frequency and percentage of distribution for the categorical variables using mean, standard deviation, minimum and maximum values, median, and quartiles for the numeric variables. Inferential analysis was conducted by employing the non-parametric Mann-Whitney test for interrupted comparisons of the numerical variables and the Chi-square or Fisher's exact tests for the categorical variables. A significance level of 5% was adopted for all statistical analyses.

RESULTS

General profile of the participants

Table 2 shows the age of participants in the early (EIG) and late (LIG) intervention groups at the time of palatoplasty and speech-language assessment, where it is possible to observe that a larger number of participants underwent late cleft palate repair. A wide variation in age at the time of palatoplasty can also be observed, including surgeries performed in adulthood.

To confirm the age difference at the time of primary palatoplasty, the median age of the participants in both groups was compared at the times of surgery and of speech-language assessment. In both comparisons, participants in the LIG presented higher median age, with statistically significant difference.

No statistically significant difference was observed between the groups with respect to participants' gender (Table 3). No statistically significant difference was found between the groups regarding the type of cleft, with a larger number of cases of cleft lip and palate in both groups (Table 4).

Speech characterization of participants - general and group analysis

As for the parameter resonance classification in both groups (EIG and LIG), Table 5 shows that most participants presented inadequate results. Language assessment related to speech production did not present statistical difference between the groups for resonance classification, but the *p*-value was marginal (close to 0.05), which can be interpreted as a trend in Biostatistics. Considering this trend, it is possible to state that the LIG would have more participants classified with inadequate resonance.

Regarding the analysis for resonance alteration in both groups, a larger number of participants with hypernasality are

observed (Table 6). As for hyponasality, it was present at mild severity in only four participants of the LIG.

No difference between the groups was found with respect to the presence or absence of hypernasality, but when the frequency distribution for each severity level was verified, the EIG presented a higher percentage of participants with mild level compared with that of the LIG, whereas the LIG showed a higher percentage of participants with severe level than that of the EIG (Table 7).

No differences between the groups were observed for the parameters nasal turbulence, weak intraoral air pressure, and audible nasal air emission (Table 8). With respect to speech understandability, there was no statistically significant difference

Table 2. Age of participants in the early (EIG) and late (LIG) intervention groups at the time of palatoplasty and speech-language assessment

Age	Group	n	Mean	SD	Min	Max	Med	1 st quartile	3 rd quartile	Comparison
Palatoplasty	EIG	43	1.4	0.459	1	2	1.3	1.0	2.0	U=0.0; z=-8.472; p<0.001*
	LIG	54	11.5	10.69	3	50	7.0	4.0	15.8	
Speech-language Assessment	EIG	43	12.3	6.95	6	29	10.0	6.0	18.0	U=724.0; z=-3.183; p=0.001*
	LIG	54	19.8	12.37	6	50	16.0	9.0	29.3	

*statistical significance (*p*<0.05)

Caption: n = number of participants; min = minimum age; max = maximum age; med = median; SD = standard deviation; EIG = early intervention group; LIG = late intervention group

Table 3. Gender comparison between participants in each group

Group	Gender		Total	Statistics
	Female	Male		
EIG	19	24	43	X ² =0.867; df=1; p=0.352*
LIG	29	25	54	

*statistical significance (*p*<0.05) - Chi-square test

Caption: EIG = early intervention group; LIG = late intervention group

Table 4. Comparison of type of cleft between the groups

Type of cleft	Group		Total	Statistics
	EIG	LIG		
Cleft lip and palate	30	31	61	X ² =1.567; df=1 p=0.211*
Cleft palate	13	23	36	

*statistical significance (*p*<0.05) - Chi-square test

Caption: EIG = early intervention group; LIG = late intervention group

Table 5. Resonance classification

Resonance classification	EIG		LIG		Total	Statistics
	Frequency	%	Frequency	%		
Adequate	13	30.2	8	14.8	21	X ² =3.355; df=1 p=0.067*
Inadequate	30	69.8	46	85.2	76	

*statistical significance (*p*<0.05) - Chi-square test

Caption: EIG = early intervention group; LIG = late intervention group

Table 6. Characterization of hypernasality

Resonance alteration	Classification	EIG		LIG		Total	Statistics
		Frequency	%	Frequency	%		
Hypernasality	Absent	12	27.9	8	14.8	20	X ² =2.507; df=1 p=0.113
	Present	31	72.1	46	85.2	77	
Hyponasality	Absent	43	100.00	50	92.6	93	p=0.127
	Present (mild)	0	0.0	4	7.4	4	

Caption: EIG = early intervention group; LIG = late intervention group

between the groups, with most participants presenting inadequate speech (Table 9) regardless of age at the time of palatoplasty. However, high severity ratings are observed in the LIG when analyzing the data according to speech understandability level (Table 10), with the EIG presenting a higher percentage of participants with mild level than that of the LIG and the LIG showing a higher percentage of participants with severe level compared with that of the EIG.

Table 11 presents the general characterization of the participants regarding compensatory articulation disorders. Higher occurrence of participants with inadequate resonance was

observed in the LIG, with glottal stop and pharyngeal fricative as the most common CADs (Table 12). In the LIG, glottal stop also occurred with greater frequency among the other CADs.

Table 13 shows the number of phonemes with identification of CADs according to the groups. As for glottal stop in the LIG, the number of altered phonemes in the participants' speech ranged from one to 12, which indicates variability of the participants' profile. In the LIG, also for glottal stop, the number of altered phonemes varied from one to 13. Statistically significant difference was observed between the groups regarding the number of altered phonemes (Table 14).

Table 7. Hypernasality rating

Severity rating	EIG		LIG	
	Frequency	%	Frequency	%
Mild	15	48.4	15	32.6
Moderate	13	41.9	20	43.5
Severe	3	9.7	11	23.9

Caption: EIG = early intervention group; LIG = late intervention group

Table 8. Presence of nasal turbulence

Variable	Classification	EIG		LIG		Total	Statistics
		Frequency	%	Frequency	%		
Nasal turbulence	Absent	37	86.0	51	94.4	88	$p=0.179$
	Present	6	14.0	3	5.6	9	
Weak intraoral air pressure	Absent	30	69.8	30	55.6	60	$X^2=2.049; df=1; p=0.152$
	Present	13	30.2	24	44.4	37	
Audible nasal air emission	Absent	33	76.7	37	68.5	70	$X^2=0.806; df=1; p=0.369$
	Present	10	23.3	17	31.5	27	

Caption: EIG = early intervention group; LIG = late intervention group

Table 9. Classification of speech understandability

Speech understandability classification	EIG		LIG		Total	Statistics
	Frequency	%	Frequency	%		
Adequate	13	30.2	9	16.7	22	$X^2=2.512; df=1; p=0.113$
Inadequate	30	69.8	45	83.3	75	
Total	43	100.0	54	100.0	97	

Caption: EIG = early intervention group; LIG = late intervention group

Table 10. Speech understandability rating

Severity rating	EIG		LIG	
	Frequency	%	Frequency	%
Mild	17	56.7	11	24.4
Moderate	9	30.0	21	46.7
Severe	4	13.3	13	28.9
Total	30	100.0	45	100.0

Caption: EIG = early intervention group; LIG = late intervention group

Table 11. Classification of the compensatory articulation disorders (CAD)

CADs	EIG		LIG		Total
	Frequency	%	Frequency	%	
Present	24	55.8	41	75.9	65
Absent	19	44.2	13	24.1	32
Total	43	100.0	54	100.0	97

Caption: CADs = compensatory articulation disorders; EIG = early intervention group; LIG = late intervention group.

Table 12. Characterization of the compensatory articulation disorders (CAD)

Group	Types of CADs						
	GS n	MDPP n	PP n	PF N	PNF N	NF N	VF n
EIG	19	6	4	4	0	5	3
LIG	35	5	4	11	2	4	8

Caption: n= number of individuals; CADs = compensatory articulation disorders; GS = glottal stop; MDPP = mid-dorsum palatal plosive; PP = pharyngeal plosive; PF = pharyngeal fricative; PNF = posterior nasal fricative; NF = nasal fricative; VF = velar fricative

Table 13. Number of phonemes with identification of compensatory articulation disorders (CAD) according to the groups (n= 26)

Number of phonemes	GS N		MDPP N		PP n		PF N		PNF N		NF N		VF N	
	EIG	LIG	EIG	LIG	EIG	LIG	EIG	LIG	EIG	LIG	EIG	LIG	EIG	LIG
1	6	2	1	3	2	2	1	3	0	1	3	2	1	3
2	3	10	4	1	1	0	1	3	0	0	1	1	1	2
3	2	4	0	0	0	0	2	0	0	1	1	0	1	2
4	3	5	1	1	1	0	0	3	0	0	0	1	0	1
5	0	1	0	0	0	1	0	1	0	0	0	0	0	0
6	2	5	0	0	0	1	0	1	0	0	0	0	0	0
7	1	3	0	0	0	0	0	0	0	0	0	0	0	0
8	0	1	0	0	0	0	0	0	0	0	0	0	0	0
9	0	1	0	0	0	0	0	0	0	0	0	0	0	0
10	1	0	0	0	0	0	0	0	0	0	0	0	0	0
11	0	0	0	0	0	0	0	0	0	0	0	0	0	0
12	1	1	0	0	0	0	0	0	0	0	0	0	0	0
13	0	1	0	0	0	0	0	0	0	0	0	0	0	0

Caption: n= number of individuals; GS = glottal stop; MDPP = mid-dorsum palatal plosive; PP = pharyngeal plosive; PF = pharyngeal fricative; PNF = posterior nasal fricative; NF = nasal fricative; VF = velar fricative

Table 14. Altered phonemes with occurrence of compensatory articulation disorders (CAD)

Group	N	Mean	SD	Min	Max	Med	1 st quartile	3 rd quartil	Comparison
EIG	43	2.6	3.07	0	12	2.0	0.0	4.0	U=847.5;
LIG	54	4.4	3.92	0	14	4.0	0.8	7.3	z=-2.323; p=0.020*

*statistical significance ($p < 0.05$)

Caption: n = number of participants; min = minimum age; max = maximum age; med = median; SD = standard deviation; EIG = early intervention group; LIG = late intervention group

DISCUSSION

In the Early Intervention Group (EIG), the mean age of participants at the time of palatoplasty was 1.4 years. As reported in previous studies^(5,7,10,11), this age would be compatible with that recommended by the national and international protocols, which affirm that when intervention is performed at this age the chances of individuals presenting speech alterations as a result of palate cleft are reduced.

The specific scientific literature clearly establishes that palate cleft surgical repair should be conducted early, before the age of two^(5,11), especially with regards to speech⁽⁵⁾.

In the Late Intervention Group (LIG), the mean age of participants at the time of primary palatoplasty was 11.5 years. Studies^(16,25) have shown that, in developing countries, lack of information and difficulty in accessing the health system are the main indicators of late primary intervention, and this a characteristic of the population cared at the HCFMUSP, which

often does not have access to this service according to the age recommended by the protocols of the area.

The findings of this study are in agreement with those described in the literature⁽¹²⁾ regarding gender, with predominance of males over females^(12,15), although without statistically significant results.

With respect to type of cleft, only patients with cleft lip and palate and/or cleft palate were included in this study; those with cleft lip only were excluded. Consequently, comparison with the epidemiological data verified in the course of this study is impossible for the population investigated.

Regarding the characterization of speech in general, the study identified a larger number of participants in both groups with alterations in the parameters hypernasality, speech understandability, and presence of CADs. A smaller but considerable number of changes were observed for the parameters nasal turbulence, weak intraoral air pressure, and audible nasal air emission.

According to the results obtained in the present study, it is possible to observe that, even without statistical difference, the data suggest worse performance of the participants who

underwent late cleft repair. Clear difference between the groups was observed regarding the presence of CADs, with a trend for difference in hypernasality as well, as already mentioned in the literature^(6,7,9,21). These differences may be related to the difficulty in velopharyngeal closure, which often occurs in late palatoplasty^(5,6,11).

It is worth noting that this study could not control the type of surgery and the number of surgeons involved in them because of the nature of the patients' admission to the service.

As for resonance classification, a more severe rating of hypernasality was observed in the cases of late cleft repair. This aspect would be justified by the fact that palate closure occurs at a more advanced age, at which the altered speech model would already be established, as well as greater chances of unfavorable anatomical characteristics^(6,13,26). It is also possible to correlate this relationship with an idea widely advocated in the literature^(7,9), that on the importance of conducting early palatoplasty. Concerning hyponasality, it was observed in only four participants in the present study, which confirms hypernasality as the most commonly present resonance alteration in the speech of cleft patients⁽¹⁴⁾.

Other characteristics of speech, which are mentioned in the literature as less common^(14,17), were also identified similarly in this study. Both groups presented low occurrence of nasal turbulence and, clinically, the literature describes it as a seldom-observed parameter⁽⁶⁾. Weak intraoral pressure is also not the most commonly observed change in clinical assessment, and when it occurs, it is generally related to plosive phonemes^(2,6). In this study, its occurrence was relative, but did not reach 50% of the sample in either group. This result can be correlated to the high presence of CADs in the study population. Significant audible nasal air emission was observed in less than 40% of the participants, with no statistically significant difference between the groups. It is also not considered a common disorder in the speech of cleft patients⁽⁶⁾.

With respect to the CADs, higher occurrence was found in the LIG. This result corroborates the findings in the literature^(7,9), and for such changes in speech, speech-language therapy is the most indicated conduct^(7,9,25). According to the literature⁽⁶⁻⁹⁾, when individuals cannot produce a phoneme at the appropriate articulation place, they compensate to emit it as closely as possible to the correct production. According to Perry and Kuehn⁽⁹⁾, the occurrence of CADs justifies speech-language therapy, and the cases without velopharyngeal insufficiency (VFI) justify periodic speech-language pathology monitoring in order to avoid speech and articulation alterations. The CADs, along with hypernasality, are the most common changes in the speech of patients with palate cleft^(14,18).

Speech understandability inadequacy is another commonly present alteration in cleft patients^(5,6,11). Thus, as in the literature, high occurrence of inadequacy was observed in the participants of this study, with greater severity in the LIG. According to previous studies^(14,18), speech understandability inadequacy may also be associated with the presence of CADs, corroborating the findings of this study.

One of the factors associated with the larger number of late palatoplasty in the population cared at the HCFMUSP is the lack of access to information and reference centers for the treatment of cleft patients⁽¹⁷⁾. The literature^(2,15-17) describes this difficulty and mentions that, in developing countries, there is also the difficulty of registering this population. These aspects are associated with a delay in seeking treatment. Many Brazilian centers have the same profile as the HCFMUSP, admitting patients with lack of information on cleft palate and its genetic relationships and on the necessary treatments. International reference centers^(15,16,26) suggest some proposals to minimize this problem, such as population awareness of the risk factors and occurrence of cleft palate and the use of standardized treatment protocols^(15,27). A domestic study⁽²⁸⁾ compared the results of a health service at two different moments, and reached the conclusion that standardization of care for cleft patients by applying protocols showed improvement in the results found in short time.

It is possible to observe a significant difference in access to information, treatment, and adherence in developed countries. According to the literature, in countries such as England⁽¹²⁾ and the USA⁽²⁹⁾, primary palatoplasty occurs up to 1 year of age, minimizing the effects of cleft palate on speech. In the Brazilian service, many patients only have access to it as adults.

Standardized, controlled and randomized studies on the speech aspects of individuals with cleft lip and palate are scarce. Predictive values of prevalence and incidence of speech disorders are not identified in the literature, and the results found are variable. With this study, we expect to obtain material for the formulation of further better-structured studies with greater scientific evidence. It will also allow discussion of clinical protocols, which will directly assist the study population. The determination of evidence-based clinical guidelines and of service indicators would be the high point of speech-language pathology practice after the compilation of these initial data.

It is believed that determination of the profile of the patients cared at the referred hospital will help reflect on speech-language pathology assistance, as well as on the discussion about the best therapeutics, uni- or multi-disciplinary, that could be offered to cleft patients. Determination of the profile of the population cared at the HCFMUSP may assist with the organization of the speech-language pathology care provided, contributing to the organization and planning of a service based on evidence and on the assumptions of the concept of health technology assessment.

CONCLUSION

Based on what has been presented, it was possible to characterize the general profile and speech characteristics of patients undergoing primary palatoplasty in the aforementioned university hospital (HCFMUSP) - a national referral center for treatment of cleft lip and palate located in the city of Sao Paulo.

In general, impairment was observed in the speech of the study participants, evidenced by the statistically significant differences found with respect to presence of CADs between

the early (EIG) and late (LIG) intervention groups. The variable resonance presented marginal statistical results, indicating a trend in differentiating the groups also in relation to this alteration.

In conclusion, patients submitted to early primary palatoplasty present better speech profile.

REFERENCES

- Freitas JAS, Neves LT, Almeida ALPF, Garib DG, Trindade-Suedam IK, Yaedú RYF, et al. Rehabilitative treatment of cleft lip and palate: experience of the Hospital for Rehabilitation of Craniofacial Anomalies/ USP (HRAC/USP) Part 1: overall aspects. *J Appl Oral Sci.* 2012;20(1):9-15. PMID:22437671. <http://dx.doi.org/10.1590/S1678-77572012000100003>.
- Souza J, Raskin S. Estudo clínico e epidemiológico de fissuras orofaciais. *J Pediatr.* 2013;89(2):137-44. PMID:23642423. <http://dx.doi.org/10.1016/j.jpeds.2013.03.010>.
- Cox TC. Taking it to the max: the genetic and developmental mechanisms coordinating midfacial morphogenesis and dysmorphology. *Clin Genet.* 2004;65(3):163-76. PMID:14756664. <http://dx.doi.org/10.1111/j.0009-9163.2004.00225.x>.
- Nahai FR, Williams JK, Burstein FD, Martin J, Thomas J. The management of cleft lip and palate: pathways for treatment and longitudinal assessment. *Semin Plast Surg.* 2005;19(4):275-85. <http://dx.doi.org/10.1055/s-2005-925900>.
- Rohrich RJ, Love EJ, Byrd HS, Johns DF. Optimal timing of cleft palate closure. *Plast Reconstr Surg.* 2000;106(2):413-21, quiz 422, discussion 423-5. PMID:10946942. <http://dx.doi.org/10.1097/00006534-200008000-00026>.
- Palandi BBB, Guedes ZCF. Aspecto de fala de indivíduos com fissura palatina e labial, corrigida em diferentes idades. *Rev CEFAC.* 2011;13(1):8-16. <http://dx.doi.org/10.1590/S1516-18462011005000012>.
- Abdel-Aziz M. Speech outcome after early repair of cleft soft palate using Furlow technique. *Int J Pediatr Otorhinolaryngol.* 2013;77(1):85-8. PMID:23116906. <http://dx.doi.org/10.1016/j.ijporl.2012.09.038>.
- Peterson-Falzone SJ, Trost-Cardamone JE, Karnell MP, Hardin-Jones M. The clinician's guide to treating cleft palate speech. St. Louis: Mosby; 2006. Effects of cleft and non-cleft VPI on speech in older children. p. 17-39.
- Perry JL, Kuehn DP. Magnetic resonance imaging and computer reconstruction of the velopharyngeal mechanism. *J Craniofac Surg.* 2009;20(2, Suppl 2):1739-46. PMID:19816343. <http://dx.doi.org/10.1097/SCS.0b013e3181b5cf46>.
- Hortis-Dzierzbicka M, Radkowska E, Fudalej P. Speech outcomes in 10-year-old children with complete unilateral cleft lip and palate after one-stage lip and palate repair in the first year of life. *J Plast Reconstr Aesthet Surg.* 2012;65(2):175-81. PMID:21978731. <http://dx.doi.org/10.1016/j.bjps.2011.09.015>.
- Chapman KL, Hardin-Jones MA, Goldstein JA, Halter KA, Havlik RJ, Schulte J. Timing of palatal surgery and speech outcome. *Cleft Palate Craniofac J.* 2008;45(3):297-308. PMID:18452355. <http://dx.doi.org/10.1597/06-244>.
- Fitzsimons KJ, Copley LP, Deacon SA, van der Meulen JA. Hospital care of children with a cleft in England. *Arch Dis Child.* 2013;98(12):970-4. PMID:23968774. <http://dx.doi.org/10.1136/archdischild-2013-304271>.
- Perry JL, Kuehn DP. Magnetic resonance imaging and computer reconstruction of the velopharyngeal mechanism. *J Craniofac Surg.* 2009;20(2, Suppl 2):1739-46. PMID:19816343. <http://dx.doi.org/10.1097/SCS.0b013e3181b5cf46>.
- Lima MRF, Leal FB, Araújo SVS, Matos EF, Di Ninno CQMS, Britto ATBO. Atendimento fonoaudiológico intensivo em pacientes operados de fissura labiopalatina: relato de casos. *Rev Soc Bras Fonoaudiol.* 2007;12(3):240-6. <http://dx.doi.org/10.1590/S1516-80342007000300012>.
- Shapira Y, Haklai Z, Blum I, Shpack N, Amitai Y. Prevalence of non-syndromic orofacial clefts among jews and arabs, by type, site, gender and geography: a multi-center study in Israel. *Isr Med Assoc J.* 2014;16(2):759-63. PMID:25630204.
- Ajike SO, Adebola RA, Efunkoya A, Adeoye J, Akitoye O, Veror N. Epidemiology of adult cleft patients in North-western Nigeria: our experience. *Ann Afr Med.* 2013;12(1):11-5. PMID:23480989. <http://dx.doi.org/10.4103/1596-3519.108243>.
- Penido FA, Noronha RMS, Caetano KI, Jesus MSV, Di Ninno CQMS, Britto ATBO. Correlação entre os achados do teste de emissão de ar nasal e da nasofaringoscopia em pacientes com fissura labiopalatina operada. *Rev Soc Bras Fonoaudiol.* 2007;12(2):126-34. <http://dx.doi.org/10.1590/S1516-80342007000200010>.
- Prandini EL, Pegoraro-Krook MI, Dutka JCR, Marino VCC. Occurrence of consonant production errors in liquid phonemes in children with operated cleft lip and palate. *J Appl Oral Sci.* 2011;19(6):579-85. PMID:22230991. <http://dx.doi.org/10.1590/S1678-77572011000600007>.
- Schuster M, Maier A, Bocklet T, Nkenke E, Holst A, Eysholdt U, et al. Automatically evaluated degree of intelligibility of children with different cleft type from preschool and elementary school measured by automatic speech recognition. *Int J Pediatr Otorhinolaryngol.* 2012;76(3):362-9. PMID:22236457. <http://dx.doi.org/10.1016/j.ijporl.2011.12.010>.
- Kuehn D, Moller KT. Speech and language issues in the cleft palate population: the state of the art. *Cleft Palate Craniofac J.* 2000;37(4):348. [http://dx.doi.org/10.1597/1545-1569\(2000\)037<0348:SALIIT>2.3.CO;2](http://dx.doi.org/10.1597/1545-1569(2000)037<0348:SALIIT>2.3.CO;2).
- Mituuti CT, Piazzentin-Penna SHA, Brandão GR, Bento-Gonçalves CGA. Caracterização da fala de indivíduos submetidos à palatoplastia primária. *Rev Soc Bras Fonoaudiol.* 2010;15(3):355-61. <http://dx.doi.org/10.1590/S1516-80342010000300008>.
- Andrade CRF, Befi-Lopes DM, Fernandes FDM, Wertzner HF. ABFW Teste de Linguagem Infantil nas áreas de fonologia, vocabulário, fluência e pragmática. São Paulo: Ed. Pró Fono; 2004.
- Henningsson G, Kuehn DP, Sell D, Sweeney T, Trost-Cardamone JE, Whitehill TL. Universal parameters for reporting speech outcomes in individuals with cleft palate. *Cleft Palate Craniofac J.* 2008;45(1):1-17. PMID:18215095. <http://dx.doi.org/10.1597/06-086.1>.
- Trindade IEK, Genaro KF, Yamashita RP, Miguel HC, Fukushima AP. Proposal for velopharyngeal function rating in a speech perceptual assessment. *Pro Fono.* 2005;17(2):259-62. PMID:16909536. <http://dx.doi.org/10.1590/S0104-56872005000200015>.
- Jajja MR, Gilani A, Cawasji ZF, Imran S, Khan MS, Hashmi SS, et al. Oral clefts: a review of the cases and our experience at a single institution. *J Pak Med Assoc.* 2013;63(9):1098-102. PMID:24601184.

26. Vogel AP, Ibrahim HM, Reilly S, Kilpatrick N. A comparative study of two acoustic measures of hypernasality. *J Speech Lang Hear Res.* 2009;52(6):1640-51. PMID:19951929. [http://dx.doi.org/10.1044/1092-4388\(2009/08-0161\)](http://dx.doi.org/10.1044/1092-4388(2009/08-0161)).
27. Owotade FJ, Ogundipe OK, Ugboko VI, Ojoje VN, Olasoji HO, Makinde ON, et al. Awareness, knowledge and attitude on cleft lip and palate among antenatal clinic attendees of tertiary hospitals in Nigeria. *Niger J Clin Pract.* 2014;17(1):6-9. PMID:24326798. <http://dx.doi.org/10.4103/1119-3077.122822>.
28. Alonso N, Tanikawa DYS, Lima JE Jr, Ferreira MC. Avaliação comparativa e evolutiva dos protocolos de atendimento dos pacientes fissurados. *Rev Bras Cir Plast.* 2010;25(3):434-8. <http://dx.doi.org/10.1590/S1983-51752010000300006>.
29. Basseri B, Kianmahd BD, Roostaeian J, Kohan E, Wasson KL, Basseri RB, et al. Current national incidence, trends, and health care resource utilization of cleft lip-cleft palate. *Plast Reconstr Surg.* 2011;127(3):1255-62. PMID:21364426. <http://dx.doi.org/10.1097/PRS.0b013e3182043af6>.

Author contributions

The study was conceived and designed jointly by all authors; KIM and LDM were in charge of collection, analysis and interpretation of the data; KIM, LDM, and CRFA assisted with the writing or revision of the manuscript; NA and CRFA were responsible for the final approval of the submitted manuscript.