

Could speech disorders in children with cleft lip and palate be justified by impaired central auditory skills?

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ABSTRACT

Purpose: to verify the association between central auditory skills and speech disorders related to velopharyngeal dysfunction.

Methods: forty-five children, with repaired non-syndromic cleft lip and palate or cleft lip only, aged 7-11 years old, were divided into three groups: G1 (n=15), children with hypernasality, nasal air emission, and compensatory articulations; G2 (n=15), children with hypernasality and nasal air emission, but without compensatory articulations; and G3 (n=15), children without hypernasality, nasal air emission, and compensatory articulations. The medical records of all participants were analyzed to verify the eligibility criteria and obtain speech assessments, and then, they were submitted to an assessment of central auditory skills. Statistical analysis comprised descriptive and chi-square test with a significance level of 5%.

Results: G1 presented a higher occurrence of impairment in central auditory skills differing from the other groups, particularly in the temporal ordering and binaural integration skills. A significant difference was observed among groups in temporal ordering ability. No significant association was found between the use of specific types of compensatory articulations and impaired auditory skills.

Conclusion: there was an association between changes in temporal ordering auditory skills and binaural integration in children with velopharyngeal dysfunction, regardless of the presence or type of compensatory articulation found.

Keywords: Cleft Lip; Cleft Palate; Hearing; Speech; Velopharyngeal Insufficiency

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INTRODUCTION

Among craniofacial anomalies, cleft lip and palate (CLP) is the most prevalent malformation affecting the lip, alveolar ridge and palate. It stands out for the number of complex alterations affecting aesthetics, speech, hearing, social and psychosocial interactions if left untreated¹. Surgical correction of the lip and/or palate, however, does not guarantee normal function of the velopharyngeal mechanism and auditory tube, which is necessary for proper speech and good hearing, respectively. A considerable number of patients with history of CLP, therefore, may present with speech disorders² and middle ear complications³. Studies have shown that middle ear problems and the most frequent speech disorders in this population can be related to velopharyngeal dysfunction (VPD)^{4,5}.

Hearing disorders in children with CLP are associated with an immature development of the auditory tube, because in children, in general, the more horizontal positioning of this structure, the anatomy and physiology is not the same as that found in adults; Abnormalities in the muscles of the auditory tube, as a result of the structural changes caused by CLP, there is inadequate functioning of the velopharyngeal musculature, which can result in edema and inflammation of the auditory tube itself, as well as adenoid hypertrophy, causing negative pressure in the middle ear, leading to episodes of otitis media with effusion (OME); and finally, craniofacial bone abnormalities in children with CLP, mainly related to alterations in the structure of the auditory tube. All these factors contribute to episodes of peripheral auditory alterations, mainly of the conductive type^{6,7}. Regarding changes in central auditory skills, it was observed that regardless of the history of the presence or absence of OME, children with CLP have changes in central auditory processing, with worse results being verified in those children with a history of OME⁸.

Hypernasality, nasal air emission and use of atypical place of articulation (compensatory articulations) are speech disorders related to impaired velopharyngeal function for speech, a condition known as velopharyngeal dysfunction⁹. Auditory tube dysfunction (ETD) in children with CLP can lead to OME and consequently impaired hearing function⁶; also, OME and hearing loss can have an important impact on speech development and intelligibility¹⁰. That is, secretion in the middle ear causes difficulties in sound transmission¹¹, producing sensory deprivation¹² and restraining adequate

reception of speech sounds and causing changes in different auditory skills^{13,14}.

The history of sensory deprivation of individuals with OME is well known, as is the fact that hearing loss has a profound effect on some of the abilities involved in speech perception and speech production¹¹. While many studies have focused on hearing skills for the population with CLP^{13,15,16}, fewer studies investigated the relationship between auditory skills and speech disorders in this same population. A previous study¹⁷ reported a statistically significant association between hearing loss, VPD and use of compensatory articulations (CA).

The hypothesis of the present study is that central auditory skills in children with CLP who present speech disorders related to VPD (hypernasality, nasal air emission and compensatory articulations) are different from the skills of children with CLP without speech disorders related to VPD. In other words, it is assumed that there is an association between speech disorders related to VPD and central auditory skills.

This study is justified because some children presented with cleft lip and palate and compensatory articulations may have difficulties in modifying these patterns in speech, even during the speech therapy process. The persistence in the articulatory patterns may be related to difficulties in auditory perception of patterns involved in speech production because of altered auditory skills. In this way, the present study aims to verify the association between central auditory skills and speech disorders related to velopharyngeal dysfunction.

METHODS

This cross-sectional and prospective study was conducted after approval by the Research Ethics Committee of the Hospital for Rehabilitation of Craniofacial Anomalies, University of São Paulo (HRAC-USP), Brazil, (CAAE 235661513.0.0000.5441, under protocol number: 501.124) and included 45 children with history of cleft lip only (CL) or cleft lip and palate (CLP). The participants were selected randomly among a group of children treated at a hospital specialized in the management of craniofacial malformations. Only those participants whose guardians signed the free and informed consent form were included in the sample.

The inclusion criteria for participation in the study included: a) children with ages ranging from 7 to 12 years with operated cleft lip and/or palate, not associated with syndromes, and b) children with

prior assessments of speech and peripheral hearing available in the patient's records. Participants who presented complaint and/or hearing loss at the time of the central auditory skills evaluation performed during this study were excluded. The presence of hypernasality and nasal air emission, with or without compensatory articulation, was defined as the indicator of VPD. The sample was obtained by convenience, in which 45 children (18 females, 27 males) scheduled for routine care at the institution where the study was carried out, with repaired non-syndromic cleft lip and palate or cleft palate only, fulfilled the inclusion criteria and were recruited into the study. According to the protocol of the institution where the study was carried out, palate surgery is performed at around 12 months of life for babies.

Speech (regarding velopharyngeal dysfunction and the use of compensatory articulations) and audiological (to include only children with hearing thresholds lower than 15 dB HL¹⁸, obtained from the last audiological assessment in the medical record) data were recorded

in patient's records before this study and were used to distribute participants into groups. The audiological assessments for analysis regarding inclusion in the study and speech assessments for division between the study groups were collected retrospectively from the medical records, within a maximum period of one year.

The participants were divided into three groups (Table 1): G1 was composed of 15 patients with hypernasality, nasal air emission and compensatory articulations; G2 composed of 15 patients with hypernasality and nasal air emission, but without compensatory articulations; G3 (control group) composed of 15 patients without hypernasality, nasal air emission or compensatory articulation. Participants in G1 and G2 were grouped according to presence of velopharyngeal dysfunction and the presence (G1) or absence (G2) of compensatory articulation, and G3, by the absence of all grouping variables (velopharyngeal dysfunction and compensatory articulation).

Table 1. Distribution of participants by type of cleft, sex and mean age into the three groups (G1, G2, G3)

Type of cleft	G1 (n=15)	G2 (n=15)	G3 (n=15)
Bilateral cleft lip and palate	8 (53%)	5 (33%)	3 (20%)
Unilateral cleft lip and palate	3 (20%)	5 (33%)	8 (53%)
Cleft palate only	4 (27%)	5 (34%)	4 (27%)
Sex	G1 (n=15)	G2 (n=15)	G3 (n=15)
Females	5 (33%)	7 (47%)	6 (40%)
Males	10 (67%)	8 (53%)	9 (60%)
Age (years)	G1 (n=15)	G2 (n=15)	G3 (n=15)
Mean (\pm SD)	9.53 \pm 1.99	9.73 \pm 1.57	9.53 \pm 1.84

G1 = participants with hypernasality, nasal air emission and compensatory articulations

G2 = participants with hypernasality and nasal air emission, but without compensatory articulations

G3 = participants without hypernasality, nasal air emission or compensatory articulations

n = number; SD = standard deviation

All participants had their data checked from their medical records. Speech disorders related to VPD, such as hypernasality, nasal air emission and compensatory articulation, were identified and registered by a speech-language pathologist (SLP) during a routine face-to-face evaluation at the research site. The outcome of the Nasal Air Emission (NAET) and the Hypernasality Cul-de-sac (HCST) Tests¹⁹ were retrieved from prior assessment. The consistent presence of nasal air emission and hypernasality in all 10 productions of the tests (scores 10 and 10 on NAET and

HCST, respectively) was considered as the indicator of presence of VPD. The consistent absence of nasal air emission and hypernasality (tests scores of 0 and 0 on NAET and HCST, respectively) was considered as the indicator of absence of VPD. The participants' use of compensatory articulation was also identified in patient's records. In assessments conducted prior to this study, an SLP identified the use of compensatory articulation during participant's repetition of syllables, words and phrases. The types of compensatory articulation identified in the records included: glottal stops

(GS), pharyngeal fricatives (PF), mid-dorsum palatal stops (MDPS), velar fricatives (VF), posterior nasal fricatives (PNF), and pharyngeal stops (PS). Of interest for this study, therefore, was the history of signs of VPD and history of use of compensatory articulation. After analyzing the medical records, the participants were submitted to an assessment of peripheral hearing and central auditory skills.

During this study, all participants selected were initially submitted to a peripheral audiological evaluation in order to verify the need for exclusion. Pure tone audiometry was performed with TDH-39 headphones and a Midimate 622 Madsen audiometer (Copenhagen, DK). The standard of normality for children aged 7 to 12 years was determined according to norms¹⁸. The average of frequencies 500, 1000, 2000 and 4000 Hz for each evaluated ear ranged from 0 to 15 dB for all children. Tympanometry was performed using a GSI 33 v.2 Middle Ear Analyzer (Grason-Stadler), with results demonstrating type A curve¹⁸, and the absence of middle ear pathology, verified by otoscopy, performed by the otorhinolaryngologist.

Since hearing loss was not identified among the selected participants, all 45 patients were submitted to a series of tests for central auditory skills evaluation, including: the Sound Localization Test (SLT)²⁰, the Sequential Memory for Verbal (SMVS) and Non-Verbal (SMNV) Sounds Test²⁰, the Random Gap Detection Test (RGDT)²¹⁻²³, and the Dichotic Digit Test (only binaural integration) (DDT)²⁰. All tests were applied and

interpreted according to the standardized procedures described in the literature.

The data collected were presented descriptively, and the chi-square test were used to verify the postulated hypothesis that central auditory skills would be different among the tree groups of participants. Chi-square test or Fisher's Exact Test were used to verify the postulated hypothesis that central auditory skills would be different according to the different compensatory articulations. The significance level of 5% was adopted for interpretation of statistical tests outcome.

RESULTS

Table 2 shows the distribution of compensatory articulations by each participant in G1. With regards to the type of compensatory articulations: 7 participants (47%) presented with glottal stop, in which only 2 participants presented this compensatory articulation isolated and, the others being combined with other compensatory articulations; 6 (40%) with pharyngeal fricative, 2 isolated and 3 associated with glottal stop; 4 (27%) with mid-dorsum palatal stop, isolated in 1 participant and associated with other compensatory articulations in the other participants; 3 (20%) with pharyngeal stop and 3 (20%) with posterior nasal fricative, in which only 1 participant presented each of the compensatory articulations isolated; and, finally 2 (13%) with velar fricative, associated with other compensatory articulations (Table 2).

Table 2. Distribution of the types of Compensatory Articulation (CA) and number of affected sounds per participant of Group 1 (G1)

G1 Participants	Types of CA (Affected Sounds)	Number of Sounds Affected
1	GS (k,t); PF (tʃ,ʃ,ʒ)	5
2	GS (p,t)	2
3	MDPS (d,g); VF (z)	3
4	PNF (s,z,ʃ,ʒ)	4
5	MDPS (t,d)	2
6	PF (ʃ,ʒ)	2
7	MDPS (t,d,k); PNF (s,z)	5
8	GS (p,t,k)	3
9	GS (t,d,p,k); PNF (z); MDPS (g)	6
10	PF (ʃ,ʒ)	2
11	PS (k,t,g,p,b)	5
12	GS (p,b,t,g); PS (k)	5
13	GS (p); PF (dʒ,ʃ,ʒ)	4
14	VF (ʒ,z); PF (ʃ)	3
15	GS (p); PF (tʃ,ʃ); PS (k)	4

Types of CA: GS: glottal stop; PF: pharyngeal fricative; MDPS: mid-dorsum palatal stop; VF: velar fricative; PNF: posterior nasal fricative; PS: pharyngeal stop

Tables 3 and 4 present altered or normal auditory abilities, MSSV and MSNV tests were grouped, as they evaluate the same ability, which is temporal ordering. Thus, when one of the two tests showed an altered result, the temporal ordering auditory ability was considered altered.

When the overall performance on the central auditory skills evaluation was considered, the largest number of children with impaired hearing skills was found in G1. Impairments were found in the following auditory skills in decreasing order of frequency: temporal resolution, binaural integration, temporal ordering and sound localization. The ability to localize sounds was the least impaired among the children; a significant difference between groups was not found. On the other hand, with values ranging from 23.75 ms to 60.0 ms, impairment in temporal resolution was found in all participants (n=45) who composed the three groups (Table 3).

Only 3 participants had problems with sound localization. None of the participants of the control

group (G3=no speech disorders and without velopharyngeal dysfunction) presented impairment in sound localization, 1 (7%) presented impairment in temporal ordering, while 6 (40%) had impairment in binaural integration. The group with hypernasality and nasal air emission but without compensatory articulation (G2) had 1 (7%) individual with impairment in sound localization, 3 (20%) with impairment in temporal ordering and 11 (73%) with impairment in binaural integration. The group with hypernasality, nasal air emission and compensatory articulation (G1) had 2 (13%) individuals with impairment in sound localization, 9 (60%) with impairment in temporal ordering and 14 (93%) with impairment in binaural integration. Results show that individuals with speech disorders related to VPD and history of using compensatory articulation presented with significantly higher occurrence of alterations in temporal ordering ($p=0.003$) and binaural integration (0.006) skills.

Table 3. Average performance of G1, G2, G3 according to outcome in the central auditory skills evaluation

TESTS	SKILLS	G1 (n=15)		G2 (n=15)		G3 (n=15)		p value
		Normal	Altered	Normal	Altered	Normal	Altered	
SLT	Sound Localization	13(87%)	2(13%)	14(93%)	1(7%)	15(100%)	0	0.342
SMVS SMNV	Temporal Ordering	6(40%)	9(60%)	12(80%)	3(20%)	14(93%)	1(7%)	0.003*
RGDT	Temporal Resolution	0	15(100%)	0	15(100%)	0	15(100%)	N/A
DDT	Binaural integration	1(7%)	14(93%)	4(27%)	11(73%)	9(60%)	6(40%)	0.006*

G1: participants with hypernasality, nasal air emission and compensatory articulations; G2: participants with hypernasality and nasal air emission, but without compensatory articulations; G3: participants without hypernasality, nasal air emission or compensatory articulations; n: number
 SLT: Sound Localization Test; SMVS: Sequential Memory for Verbal Sounds Test; SMNV: Sequential Memory for Non-Verbal Sounds Test; DDT: Dichotic Digit Test; RGDT: Random Gap Detection Test; * $p < 0.05$
 Statistical test: Chi-square test. Statistical significance: $p < 0.05$

The performance in the different central auditory tests for the 15 participants of G1 was further analyzed taking into consideration the types of compensatory articulations used (Table 4). Statistical testing revealed no significant ($p > 0.05$) association between central auditory skills and compensatory articulation. The distribution of participants into the 6 different types of CAs (GS, PF, MDPS, VF, PNF, PS) varied from a minimum of 2 to a maximum of 6 participants with presence of CA, limiting the interpretation of the statistical findings

due to the small sample size. When sound localization skills were considered, 3 (20%) participants in G1 failed this ability (which presented compensatory articulations related to GS, MDPS and VF), differing from the temporal resolution skills in which all 15 participants in G1 (100%) failed this ability. Impaired temporal ordering skills were worse for participants who used MDPS, PNF and PS while impaired binaural integration skill were worse for participants who used GS, MDPS, VF, PNF and PS.

Table 4. Distribution of participants from G1 with altered central auditory skills tests according to presence or absence of the different types of compensatory articulation

Skills Tested Impaired Skill (n=15)	Sound Localization		Temporal Ordering		Binaural Integration		Temporal Resolution	
	YES	NO	YES	NO	YES	NO	YES	NO
GS Present (n=7) ^F	1 (14%)	6 (86%)	5 (71%)	2 (29%)	7 (100%)	0	7 (100%)	0
GS Absent (n=8) ^F	1 (13%)	7 (87%)	5 (63%)	3 (37%)	7 (88%)	1 (12%)	8 (100%)	0
p value	1,000		1,000		1,000		N/A	
PF Present (n=6)	0	6 (100%)	3 (50%)	3 (50%)	5 (33%)	1 (7%)	6 (100%)	0
PF Absent (n=9)	2 (22%)	7 (78%)	6 (67%)	3 (33%)	9 (100%)	0	9 (100%)	0
p value	0.215		0.447		0.268		N/A	
MDPS Present (n=4)	1 (25%)	3 (75%)	3 (75%)	1 (25%)	4 (100%)	0	4 (100%)	0
MDPS Absent (n=11)	1 (9%)	10 (91%)	5 (45%)	6 (55%)	10 (91%)	1 (9%)	11 (100%)	0
p value	0.423		0.310		0.533		N/A	
VF Present (n=2)	1 (50%)	1 (50%)	1 (50%)	1 (50%)	2 (100%)	0	2 (100%)	0
VF Absent (n=13)	1 (8%)	12 (92%)	7 (54%)	6 (46%)	12 (92%)	1 (8%)	13 (100%)	0
p value	0.101		0.919		0.685		N/A	
PNF Present (n=3)	0	3 (100%)	2 (67%)	1 (33%)	3 (100%)	0	3 (100%)	0
PNF Absent (n=12)	2 (17%)	10 (83%)	6 (50%)	6 (50%)	11 (92%)	1 (8%)	12 (100%)	0
p value	0.448		0.605		0.605		N/A	
PS Present (n=3)	0	3 (100%)	2 (67%)	1 (33%)	3 (100%)	0	3 (100%)	0
PS Absent (n=12)	2 (17%)	10 (83%)	7 (58%)	5 (42%)	11 (92%)	1 (8%)	12 (100%)	0
p value	0.448		0.792		0.605		N/A	

n: number; GS: glottal stop; PF: pharyngeal fricative; MDPS: mid-dorsum palatal stop; VF: velar fricative; PNF: posterior nasal fricative; PS: pharyngeal stop
^F: Fisher's Exact Test

Statistical test: Chi-square test and Fisher's Exact Test. Statistical significance: $p < 0.05$

DISCUSSION

A systematic review pointed out that individuals with CLP have more difficulties in auditory skills when compared to their peers without CLP, considering global information processing deficits and slow speed processing and memory problems, which can possibly affect language, speech and learning²⁴, suggesting that the findings of the present study related deficits in the oral language of children who have compensatory articulations may be associated with difficulty in auditory feedback, as a result of alterations in auditory skills, mainly related to temporal resolution and temporal ordering skills.

In the present study, G1, the children with compensatory articulations, hypernasality and nasal air emission, presented with higher occurrence of impairment in central auditory skills differing from the other groups particularly in the temporal ordering and binaural integration skills. The distribution of G1 participants across the 6 different types of CAs, however, yielded subgroups too small for further interpretation of the findings.

Temporal resolution was the most impaired auditory ability in this study. This ability is responsible for detection of time intervals, the rapid and sudden changes in sound stimuli necessary for an individual to perceive differences among sound signals²⁵. The fact that the three groups studied have presented impairment in this ability corroborates prior findings^{8,15,26} that suggest a relationship between history of otitis and auditory tube dysfunction and impairment in the temporal resolution ability. While there was no difference between G1, G2 and G3 regarding temporal resolution ability (all 45 participants presented impairment in temporal resolution), a significantly different behavior was identified for temporal ordering skills among the groups (Table 3).

Defined as the ability to correctly discriminate the order of occurrence of an acoustic signal within a specific time interval^{27,28}, it is known that impairment in temporal ordering creates difficulties in perceiving, rapidly, changing stimuli^{29,30}, which can affect sound discrimination. The majority of children in G1 (60%) presented with impaired temporal ordering skills, behavior found to be significantly different from G2 and

G3 groups (20% and 7% impairment, respectively). Prior research²⁷ reported a statistically significant relationship between severity of the phonological disorder and temporal ordering ability suggesting, along with other studies³¹⁻³³, that compromised auditory skills during the phonological development interferes with speech comprehension and consequently can affect speech production.

In the current study the binaural integration ability also showed significant signs of impairment in the group of children with CLP and speech disorders involving CAs (G1) compared to the other groups. Some studies pointed out that children with CLP may present poor binaural integration skills compared to children without CLP^{14,15,34}. Impaired binaural integration or separation skills can hinder analysis and synthesis of speech sounds^{35,36} and may be one of the factors that explain why some children with CLP choose using atypical place of articulation.

The findings regarding the association between impaired hearing abilities and the use of the specific compensatory articulations studied was not statistically different among the groups and this may be explained by the limitation in samples in each CA category. Further research involving larger samples controlled for type of CAs are needed for a better understating of the relationship between central auditory abilities (i.e., impaired binaural integration and temporal ordering skills) and use of CA by children with CLP. The current findings, therefore, must be interpreted carefully.

Information about school performance, otological history, time of speech therapy, and other changes such as attention deficit hyperactivity disorder were not included in the analysis of results, being listed as limitations of the study. The clinical importance of this study is suggested considering the prognosis of children with speech disorders due to compensatory articulations, as these may be persistent due to the difficulty of auditory perception because of alterations in auditory skills that may occur, regardless of the history of recurrent otitis media in children with cleft lip and palate. Thus, it is considered important in clinical practice that, in addition to peripheral audiological assessment and auditory training, the need to assess and treat central auditory processing in children with cleft lip and palate, especially those with compensatory articulation, is considered.

CONCLUSION

There was an association between changes in temporal ordering auditory skills and binaural integration in children presented with velopharyngeal dysfunction, regardless of the presence or type of compensatory articulation found.

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JLC and MRF participated in the conception of the study project, data acquisition, analysis and interpretation of data, article preparation, critical review for relevant intellectual content and final approval of the version to be submitted for publication;

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