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Influence of an Intensive Speech Therapy Program on the Speech of Individuals with Cleft Lip and Palate

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Abstract

Introduction Compensatory articulations are speech disorders due to the attempt of the individual with cleft palate/velopharyngeal dysfunction to generate intraoral pressure to produce high-pressure consonants. Speech therapy is the indicated intervention for their correction, and an intensive speech therapy meets the facilitating conditions for the correction of glottal stop articulation, which is the most common compensatory articulation.

Objective To investigate the influence of an intensive speech therapy program (ISTP) to correct glottal stop articulation in the speech of individuals with cleft palate.

Methods Speech recordings of 37 operated cleft palate participants of both genders (mean age = 19 years old) were rated by 3 experienced speech/language pathologists. Their task was to rate the presence and absence of glottal stops in the 6 Brazilian Portuguese occlusive consonants (p, b, t, d, k, g) distributed within several places in 6 sentences.

Results Out of the 325 pretherapy target consonants rated with glottal stop, 197 (61%) remained with this error, and 128 (39%) no longer presented it. The comparison of the pre- and posttherapy results showed: a) a statical significance for the p1, p2, p3, p4, t1, k1, k2 and d6 consonants (McNemar test; $p < 0.05$); b) a statistical significance for the p consonant in relation to the k, b, d, g consonants and for the t consonant in relation to the b, d, and g consonants (chi-squared test; $p < 0.05$) in the comparison of the proportion improvement among the 6 occlusive consonants.

Conclusion The ISTP influenced the correction of glottal stops in the speech of individuals with cleft palate.

Keywords
- cleft palate
- velopharyngeal insufficiency
- speech
- speech therapy

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Introduction

The treatment of velopharyngeal dysfunction (VPD) due to the failure of primary palatal surgery in individuals with cleft palate, when there are speech symptoms, may require physical and/or behavioral treatment, depending on the etiology of the VPD.[1,2] Physical procedures, such as surgery (secondary palatoplasty or pharyngoplasty, for example) or prosthesis (pharyngeal bulb), when the VPD was caused by structural anomalies, that is, when there is a lack of tissue in the soft palate or too much nasopharyngeal space preventing velopharyngeal closure (velopharyngeal insufficiency). Speech therapy is indicated when the cause of VPD is functional, that is, when there is a learning error in the use of velopharyngeal structures.[1] In VPD related to cleft palate, the same individual may present both insufficiency (anatomical or structural defect that prevents adequate velopharyngeal closure) and mislearning (an articulation disorder in which there is a substitution of a pharyngeal or nasal sound for an oral sound), and for this reason, the combination of physical and functional treatment approaches is necessary.[3]

The presence of VPD after surgical correction of cleft palate can lead to the development of speech alterations involving both passive and active errors.[4] Passive speech errors are due to an abnormal velopharyngeal structure, including hypernasality, nasal air emission, and weak intraoral air pressure.[5] Active speech errors involve alteration of articulation placement in response to an abnormal structure, and this compensatory behavior occur in the attempt of the speaker to generate and/or maintain adequate levels of intraoral pressure to produce high-pressure consonants.[5] Although these compensatory speech behaviors may be considered strategies developed in order to achieve the special requirements of a speech regulating system in the presence of VPD; acoustically, these responses tend to undermine rather than improve speech performance.[6]

The incidence of compensatory articulations (Cas) as described in the literature varies between 6 and 63%.[7–10]The effect of erroneously learned neuromotor patterns used during atypical placement production may dominate the phonological development of the child, creating a restricted phonetic repertoire that may persist regardless of the establishment of the potential for velopharyngeal closure. Therefore, it is common for the CA to be incorporated into the speech of the child, to the point of compromising his/her speech intelligibility. Even when the palate is restored with a successful management of velopharyngeal insufficiency, these learned behaviors may persist, always requiring speech therapy to learn an adequate placement and manner of articulation.[11]

Glottal stop (GS) is the most common type of CA found in cleft palate speech.[10,11] When this active speech error becomes habituated and incorporated into the phonological system of the individual, it can be particularly resistant to change even during speech therapy.[12] The difficulty of some clinicians to identify this error may lead them to use inadequate strategies, such as blowing exercises and other activities unrelated to speech to correct the speech error. Besides the selection of an adequate therapeutic approach to correct CA errors, the frequency of speech therapy might also be an important aspect related to the success of the intervention. As described in the literature, the hypothesis of the present study is that a structured intensive speech therapy program (ISTP) meets the facilitating conditions for the correction of CA related to cleft palate and VPD.[12–14]

Objective

The present study investigated the outcome of an ISTP to correct the use of glottal stop productions in the speech of individuals with cleft palate.

Material and Methods

The present retrospective study was approved by the Ethics and Research Committee on Human Subjects of the Hospital of Rehabilitation of Craniofacial Anomalies (1.397.124), where the present study took place. Informed verbal consent was obtained from all participants.

Speakers and Speech Sample

The audio recordings used in the present study were retrieved from the database of the Hospital of Rehabilitation of Craniofacial Anomalies. The samples of interest were obtained from 37 operated cleft lip and/or palate patients presenting with VPD, 16 females (43%) and 21 males (57%), with ages ranging from 6 to 39 years old (mean:19 years old; standard deviation [SD]: 10.8 years old). All of them were Brazilian Portuguese speakers who had participated for the first time in one of the modules of ISTP conducted at the Hospital of Rehabilitation of Craniofacial Anomalies, between 2013 and 2016. The ISTP module involves 45 therapy sessions lasting 50 minutes provided within a period of 3 weeks ($\sim$ 3 daily sessions, from Monday to Saturday), applied by different speech therapists using a combination of phonological and phonetic speech therapy approaches.

Out of the 37 patients, 34 (92%) used a temporary pharyngeal bulb prosthesis to establish potential for velopharyngeal closure during speech therapy, while the remaining 3 (8%) achieved velopharyngeal closure without a prosthesis or pharyngeal flap.

The speech recordings were obtained pre- and post-ISTP in a sound-protected environment with high-quality microphones. The recorded speech samples consisted of 6 sentences, each of them with recurrence of the 6 Brazilian Portuguese stop consonants (total of 24 target consonants), distributed as the following: "p" = **P**a**p**ai olha a **p**i**p**a (*Daddy sees the kite*): 4 target consonants; "b" = A **B**i**b**i **b**a**b**ou (*Bibi drooled*): 4 target consonants; "t" = O **t**a**t**u é **t**eu (*The armadillo is yours*): 3 target consonants; "d" = O **d**e**d**o **d**a **D**u**d**a **d**oeu (*Dudás finger hurt*): 6 target consonants; "k" = O **c**u**c**o **c**aiu a**q**ui (*The cuckoo clock fell here*): 4 target consonants; and "g" = O **G**u**g**u é le**g**al (*Gugu is cool*): 3 target consonants. The 6 sentences are part of the Brazilian cleft articulation screening sentences, which consists of 23 sentences, each with a single target consonant.

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Speech samples were recorded using a Shure PG30 condensed/unidirectional head microphone (Shure, Niles, IL, USA), positioned at $\sim$ 5 cm from the mouth, in an Intel Pentium 4 (256MB HD, 15MB RAM) computer. The files were recorded with wave extension using a Creative Audigy II soundcard (Creative Technology Ltd., Jurong East, Singapore), with the Sony Sound Forge 8.0 software (Sony Corp. Tokyo, Japan), with a sampling rate of 44,100Hz, single channel, 16 bits. To obtain the audio recordings, each patient sat in a comfortable chair in a sound-isolated room in the Phonetic Laboratory. The patients repeated each stimulus sentence after the speech language pathologist (SLP).

Listening Material

The selected samples were edited using the Sound Forge 8.0 software, randomly distributing the recordings (obtained pre- and post-ISTP) into a material presented to three SLPs for a rating task of the 6 phrases analyzed in the present study, 1 for each of the 6 consonants of interest (p, b, t, d, k, and g).

Each listener rated individually the presence and absence of glottal stop articulation in 888 target consonants of the pre- and post-ISTP audio recorded sentences produced by the 37 patients (37 patients $\times$ 24 target consonants = 888 target consonants pre-ISTP and 888 target consonants post-ISTP).

Listeners

Three female Brazilian certified SLPs (listeners), different from those who applied the therapy, with a minimum of 6 years of experience with management of cleft palate speech, rated the samples to identify the presence of glottal stops. The listeners were not aware of the purpose of the present study nor were they familiar with any of the speakers. The SLPs self-reported having normal hearing.

Listening Task

The listeners received an AKG K414P headset earphone (AKG Acoustics, Vienna, Austria) and a USB flash drive containing the speech material. The material included a file with a Microsoft PowerPoint (Microsoft Corp., Redmond, WA, USA) presentation containing instructions for the rating task and a file containing the randomly edited and anonymous recordings to be rated by the SLPs. The listeners reviewed the instructions to become familiar with the rating task. Twenty sentences produced by patients with a history of cleft palate representative of productions with and without glottal stop articulation were presented to the listeners as reference samples. The listeners were also instructed on how to use the form to record their rating. The form was prepared by the first author specifically for this purpose.

The listeners were instructed to rate individually the samples indicating either the presence or the absence of glottal stop articulation. They were also instructed to use their own personal computer with the earphone provided by the investigators to listen to the samples. They could adjust the audio level as needed. The recordings could be listened as many times as the listener deemed necessary until being able to establish the rating of presence or absence of glottal stops.

Inter-rater Agreement

The Kappa index of agreement was used to measure the degree of inter-rater agreement, for each target consonant, pre- and post-ISTP.

Interpretation of Kappa scores[15]: Poor = Kappa < 0.00; Slight = Kappa 0.00–0.20; Fair = Kappa 0.21–0.40; Moderate = Kappa 0.41–0.60; Substantial = Kappa 0.61–0.80; Almost perfect = Kappa 0.81–1.00.

Statistical Analysis

Only the samples rated identically by at least two of the three listeners, pre- and post-ISTP, were used for analysis and comparison. Data analysis was calculated using percentage. The comparison of the occurrence of glottal stop pre- and post-ISTP was calculated using the McNemar test, adopting a significance level of 5% ($p < 0.05$).

The comparison of the proportion improvement among the six stop consonants was calculated using the chi-squared test and proportions. The comparison of the proportion of improvement of each target consonant within the six sentences was calculated using the Cochran test, adopting a significance level of 5% ($p < 0.05$).

Results

The inter-rater agreement and the ratings were compared between the three raters for each target consonant, pre- and post-ISTP (►**Table 1**).

Out of the 888 target consonant possibilities in the pre-ISTP audio recordings, glottal stop articulation was rated to be present in 325 (37%) of the samples. Out of those 325 (100%), 197 (61%) remained with this error, and 128 (39%) no longer presented it, post-ISTP. The comparison of the occurrence of glottal stop among the six stop consonants, pre- and post-ISTP, was statistically significant only for the "p" and "k" consonants (chi-squared test; $p = 0{,}014$). See ►**Fig. 1**.

The comparison of the occurrence of glottal stop and post-ISTP was statistically significant only for the target consonants "p1," "p2," "p3," "p4," "t1," "k1," "k2," and "d6" (McNemar test; $p < 0.05$). See ►**Table 2**.

The comparison of the proportion improvement among the six stop consonants was statistically significant for the *p* consonant in relation to the *k, b, d,* and *g* consonants, and for the *t* consonant in relation to the *b, d,* and *g* consonants (chi-squared test; $p < 0.05$). The comparison of the proportion of improvement of each target consonant within the six sentences was not statistically significant (Cochran test; $p > 0.05$).

Discussion

The aim of the present study was to investigate the influence of an ISTP to correct glottal stop articulation in patients with cleft palate. The results showed a decrease of consonants with glottal stop after therapy, which is in agreement with previous studies that investigated the efficacy of an ISTP for cleft palate speech.[12,13,16,17] Brazilian studies that compared speech outcomes before and after therapy of individuals with

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Table 1 Inter-rater agreement percentage and Kappa values for all 24 consonant targets, pre- and post-intensive speech therapy program

Consonant	Pre-ISTP		Post-ISTP	
	% of agreement	Kappa	% of agreement	Kappa
p1	77	0.54	78	0.29
p2	78	0.58	71	0.21
p3	84	0.69	77	0.27
p4	75	0.51	73	0.25
t1	88	0.74	78	0.39
t2	93	0.86	73	0.30
t3	87	0.74	71	0.35
k1	91	0.82	78	0.50
k2	91	0.82	75	0.46
k3	91	0.82	68	0.37
k4	86	0.71	69	0.38
b1	95	0.83	84	0.33
b2	95	0.83	84	0.33
b3	93	0.73	80	0.12
b4	91	0.68	84	0.18
d1	86	0.64	86	0.40
d2	86	0.64	84	0.36
d3	86	0.64	84	0.37
d4	87	0.68	86	0.42
d5	89	0.73	84	0.29
d6	84	0.56	89	0.42
g1	91	0.74	91	0.69
g2	91	0.74	91	0.69
g3	84	0.51	78	0.31

Abbreviation: ISTP, intensive speech therapy program.

cleft palate also found a significant reduction in the occurrence of CAs.[12,18–21]

Among the 36 patients who presented glottal stop articulation pre-ISTP, 5 (14%) were able to eliminate this error in all target consonants; 4 (11%) did not show any change, which means that they continued to present this error in the same target consonants, and 27 (75%) presented a reduction in the occurrence of this error post-ISTP. It is noteworthy that the patients who remained with glottal stop articulation post-ISTP were referred to participate in the next ISTP module of the Hospital of Rehabilitation of Craniofacial Anomalies or to continue speech therapy elsewhere.

Our results have also shown that, among the six target consonants with glottal stop articulation, the "p" consonant was the easiest to be corrected when compared with the "k" consonant. This can be explained by the fact that the "p" consonant, as an anterior and bilabial consonant, has visual features easier to be learned using the facilitating cues,[18,22] compared with the "k" consonant, which is produced in the back of the mouth, where visual features are difficult to see. Pinto (2016)[20] also found in a retrospective study that the "p" consonant was the easiest consonant to be learned by the cleft palate patients undergoing intensive speech therapy.

Despite the variation in the number of occurrences of glottal stops in all target stop consonants pre-ISTP, it was observed that the greater occurrence of glottal stops was present in the voiceless consonants, when compared with the voiced ones. Voiceless high-pressure consonants require a greater amount of air pressure than the voiced ones, and this may explain the vulnerability of the voiceless consonants to be produced with glottal stop.[5] In general, most target stop consonants (unvoiced and voiced) of the present study have shown reduction in the number of glottal stop occurrences, regardless of their position within the sentence, although the unvoiced ones were those that significantly improved the most. This finding shows the ability of the individual to generalize the correct consonant production by using correct therapeutic strategies[23,24] and approaches.[25]

The findings of our study have also shown that all target consonants presented a reduction of the occurrence of glottal stop articulation regardless of their position within the sentence, with the exception of the consonants "d2" (O dedo da Duda doeu) and "d6 " (O dedo da Duda doeu), which presented a statistically significant difference. This can be explained by the generalizing ability of the patient to use the correct therapeutic strategy to produce the target sound wherever it appears in the word.[26] Some studies show that the generalization process can occur in other positions of the word, in which the patient learns to produce a phoneme in a certain position and proceeds to perform it correctly in other positions.[27,28]

Good speech outcome of patients with cleft lip and palate can be achieved either by intensive speech therapy or conventional therapy, although studies suggest that in ISTPs, the improvement can be achieved in a shorter period of time, at a lower cost.[13,14] Unlike conventional therapy, which is usually based on one or two sessions per week with no fixed time to complete, intensive speech therapy, although based on programmed modules, varies in the number of sessions, on the duration of one module, and on the duration of the therapy session. Intensive speech therapy programs with modules of up to 2 months, with daily sessions ranging from 3 to 7 days a week, with $\geq$ 1times a day, have been reported in the literature.[20,21,29–31]

Studies show that most speech therapists use the phonetic approach to treat individuals with cleft lip and palate, with good results.[32] However, some authors suggest that the phonological approach can also be successfully used for patients presenting with many CAs,[25,33,34] highlighting that future studies should be done to compare the results between patients undergoing both approaches.

In the present study, 72% of the participants used a pharyngeal bulb prosthesis (either temporary or permanent) due to hypodynamic velopharynx. Studies have shown that some individuals can improve the movement of their velopharyngeal structures with the use of the bulb by itself,[12,35–39] and others would only accomplish better

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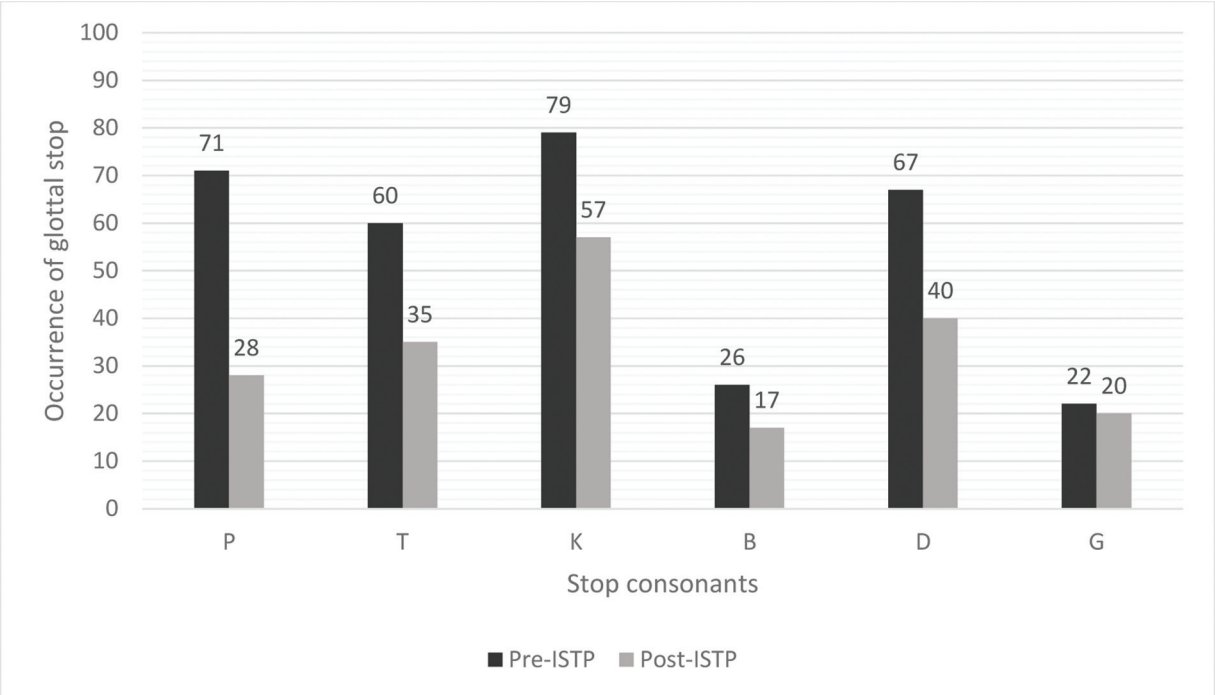

Fig. 1 Distribution of the number of glottal stop occurrences for each of the six stop consonants and post-intensive speech therapy program

Table 2 Distribution of the presence of glottal stops among the 24 target consonants and post-intensive speech therapy program, for the 37 patients

Target consonant	Presence of glottal stop		
	Pre-ISTP	Post-ISTP	Difference
p1 (Papai olha a pipa)	16 / 37	06 / 37	10
p2 (Papai olha a pipa)	17 / 37	07 / 37	10
p3 (Papai olha a pipa)	20 / 37	07 / 37	13
p4 (Papai olha a pipa)	18 / 37	08 / 37	10
t1 (O tatu é teu)	20 / 37	10 / 37	10
t2 (O tatu é teu)	18 / 37	11 / 37	07
t3 (O tatu é teu)	22 / 37	14 / 37	08
k1 (O cuco caiu aqui)	20 / 37	12 / 37	08
k2 (O cuco caiu aqui)	21 / 37	14 / 37	07
k3 (O cuco caiu aqui)	17 / 37	16/ 37	01
k4 (O cuco caiu aqui)	21 / 37	15 / 37	06
b1 (A Bibi babou)	07 / 37	05 / 37	02
b2 (A Bibi babou)	07 / 37	05 / 37	02
b3 (A Bibi babou)	06 / 37	03 / 37	03
b4 (A Bibi babou)	06 / 37	04 / 37	02
d1 (O dedo da Duda doeu)	12 / 37	07 / 37	05
d2 (O dedo da Duda doeu)	11 / 37	07 / 37	04
d3 (O dedo da Duda doeu)	11 / 37	07 / 37	04
d4 (O dedo da Duda doeu)	11 / 37	08 / 37	03
d5 (O dedo da Duda doeu)	10 / 37	07 / 37	03
d6 (O dedo da Duda doeu)	12 / 37	04 / 37	08

(*Continued*)

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Table 2 (*Continued*)

Target consonant	Presence of glottal stop		
	Pre-ISTP	Post-ISTP	Difference
g1 (O Gugu é legal)	08 / 37	06 / 37	02
g2 (O Gugu é legal)	08 / 37	06 / 37	02
g3 (O Gugu é legal)	06 / 37	08 / 37	- 02
Total	325 / 888	197 / 888	128

Abbreviation: ISTP, intensive speech therapy program.

movements with the bulb combined with intensive therapy. Although it is not yet known exactly how the pharyngeal bulb can contribute in increasing the movement of the pharyngeal walls, it is likely to act as a sensorimotor stimulus, facilitating muscular function, especially when its use is associated with speech therapy.[39,40] Dutka and Pegoraro-Krook[3] reported that a structured ISTP using correct therapeutic strategies and facilitating cues with daily monitored practice of the exercises can accelerate the process of assimilation for the replacement of "old speech" by the "new speech." The speech therapist should also train family members to ensure that home exercises are done daily and adequately.[3]

Auditory-perceptual assessment is the routine instrument used to identify and characterize speech disorders, and is therefore essential for speech and velopharyngeal function assessment. Its focus is, obviously, to identify all present speech alterations, mainly the articulatory production during pre-established speech stimuli.[41–44] Although perceptual evaluation is considered the gold standard assessment for speech disorders, it is influenced by several factors, such as the experience/training of the evaluators (multiple or not), the type and quality of the sample, the use of reference samples, among others.[10,42,45–48]

The subjectivity of speech auditory-perceptual assessment is common sense in the literature,[49] and for this reason, many studies point out the importance of using multiple raters to document speech outcomes, as well as the importance of the experience of the evaluators in identifying, characterizing, and classifying speech and voice disorders.[10,47,48] Many authors also advocate the importance of providing training to the evaluators using reference samples.[50,51]

The present study used retrospective data from participants who underwent intensive speech therapy applied by several therapists. We agree with Skidmore[14] that future studies should take into account the variables that could interfere in the results, such as age of the participants, presence of syndromes, and presence of hearing loss, and include a detailed description of the treatment program.

Conclusion

The ISTP corrected glottal stops in the speech of individuals with cleft palate.

Conflict of Interests
The authors have no conflict of interests to declare.

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