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Oral diadochokinesia and lisp in speech production in children with surgically repaired cleft lip and palate

Diadococinesia oral em crianças com fissura labiopalatina operadas e presença de ceceio na produção da fala

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ABSTRACT

Purpose: To correlate the performance of the oral motor tasks of diadochokinesis in children with repaired cleft lip and palate to the type of cleft and to the presence of lisp. **Methods:** We studied 30 children, aged 9 to 12 years, with complete unilateral cleft lip and palate and 30 with isolated cleft palate. Two speech language pathologists analyzed repetition phrases with the phones [s] and [z] to identify the presence of lisp. The analysis of diadochokinesis was performed by registering the repetition of the syllables "ta" and "ka" and the sequence "pataka". The Kappa test was used to verify agreement between the evaluators, the Chi-square test was used to compare the lisp frequency between the groups, and the t-test was used to compare the diadochokinesis values between the same groups as well as between individuals with and without lisp. **Results:** The mean number of emissions per second and the mean time between emissions were not significantly different between groups. Good agreement was observed between the examiners for the presence of lisp in [s] and [z], with the most evident agreement in the complete unilateral cleft lip and palate group. The comparison between the diadochokinesis values and the presence of lisp revealed no significant difference. **Conclusion:** The performance of diadochokinesis tasks did not change according to the type of cleft. Further, the presence of lisp speech was higher in patients with complete unilateral cleft lip and palate, but this did not affect performance of diadochokinesis tasks.

Keywords: Cleft lip; Cleft palate; Speech disorders; Child; Evaluation

RESUMO

Objetivo: Correlacionar o desempenho motor oral nas tarefas de diadococinesia, de acordo com o tipo de fissura e com a presença de ceceio na fala, em crianças com fissura labiopalatina, operadas. **Métodos:** Foram estudadas 30 crianças com fissura completa unilateral de lábio e palato e 30 com fissura isolada de palato, operadas, com idade entre 9 e 12 anos. Dois fonoaudiólogos analisaram a repetição de frases com os fones [s] e [z] para identificar a presença de ceceio e a análise da diadococinesia ocorreu a partir do registro da repetição das sílabas "ta" e "ca" e da sequência "pataca". Na análise, o teste Kappa verificou a concordância entre os avaliadores, o teste Qui-quadrado a frequência de ceceio entre os grupos e o teste t os valores da diadococinesia, segundo o tipo de fissura e a presença de ceceio. **Resultados:** O número de emissões por segundo e os tempos médios entre as emissões não demonstraram diferença significativa entre os grupos. Houve boa concordância entre os avaliadores quanto à presença de ceceio, com maior frequência nos casos com fissura completa unilateral de lábio e palato. A comparação entre os valores da diadococinesia e a presença de ceceio não evidenciou diferença significativa. **Conclusão:** O desempenho nas tarefas da diadococinesia não sofreu alteração segundo o tipo de fissura labiopalatina. A presença de ceceio na fala foi maior no grupo com fissura completa unilateral de lábio e palato, sem modificação, porém, no desempenho das tarefas da diadococinesia.

Descritores: Fenda labial; Fissura palatina; Distúrbios da fala; Criança; Avaliação

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INTRODUCTION

In general, a cleft lip and palate compromises speech because this physiological function requires anatomical and functional integrity of the associated organs, adequate articulation, and proper functioning of the velopharyngeal valve[1,2]. In these cases, speech alteration occurs due to compromised velopharyngeal function[3,4], such as obligatory disturbances and compensatory articulations, as well as the dento-occlusal balance condition[5-8], which aids functional adaptation of the tongue and lips. In individuals with a cleft lip and palate, regardless of age and the surgical technique used for repair, the dento-occlusal balance condition affects lingual-alveolar phoneme production, i.e., [s] and [z], the lingual-dental phonemes [t], [d], and [n], and the alveolar phoneme [l], with interdentalization of the tongue and lisp, which account for the observed alterations[5,7-9].

The condition of the muscle or oral motor ability is of great importance during the speech production process, and these characteristics can be evaluated by using the diadochokinesis (DDK) test. This test is used to evaluate neuromotor maturation and integration, thereby providing an acoustic index of movement speed and articulatory positioning through quick repetitions of opposite muscular contraction patterns[10,11]. In order to judge the obtained values, the Motor Speech Profile Advanced (MSP) software, KayPENTAX™, automatically provides parameters that are used for 2 types of analyses: quantitative and qualitative. The quantitative analysis refers to the amount and duration of the emissions. The amount, which corresponds to the number of emissions per second, is represented by the average DDK rate, and the duration is represented by the mean time between such emissions. The qualitative analyses takes into account the parameters of the emissions, such as regularity, rhythm, and/or stability, and is represented by the standard deviation of the DDK period, coefficient of variation of the DDK period, perturbations of the DDK period, and coefficient of variation of the DDK peak intensity[11].

Several previous studies, evaluated children without cleft lip and palate who had a speech disorder[10-16]. These studies demonstrated that children with a speech disorder produced most of the emissions at a slower rate[15,16], with a significant positive correlation between age, percentage of correct consonants, and speed of speech[17]. Further, an increase in the speed of syllable emission was observed with increased age[11].

A few studies analyzed the correlation between DDK and tongue protrusion strength in children with an anterior lisp and in those without a speech disorder. The results showed a higher tongue protrusion strength and greater number of emissions in children without speech alterations, while children with a lisp presented with lower tongue protrusion strength and were slower in accomplishing the tasks evaluated by the DDK test[12,14]. According to the authors of one of these studies, this finding indicates that a correlation exists between anterior lisp, tongue weakness, and reduced DDK speed with low speech intelligibility[14].

Although several studies analyzed DDK in children without a cleft lip and palate who had different speech disorders, only a few used this test to evaluate individuals with a cleft lip and palate[18-20]. One study evaluated 3 teenagers with a cleft lip and palate by using electropalatography and the DDK test. The teenagers had difficulties elevating the tongue tip and showed decreased speed of tongue movements when a cleft was associated with the Robin Sequence[18]. Another study evaluated 10 children with a complete cleft lip and palate, 10 with a cleft palate, 7 with velocardiofacial syndrome, and 47 without a cleft. This study reported similar results among the groups, thereby verifying that children with a complete cleft lip and palate did not show apraxic characteristics, unlike those with a cleft palate and those with velocardiofacial syndrome[19]. Another study evaluated children and young adults with and without a cleft lip and palate, and the results indicated lower speed in the group with a cleft, thereby explaining the alterations in intraoral pressure[20].

Consequently, one can conclude that, even though these few studies included individuals with a cleft lip and palate, they used limited and heterogeneous samples, despite the use of distinct DDK parameters. Therefore, it is unclear whether the DDK parameters were altered in these populations. Clarifying these questions would facilitate adequate therapeutic planning with regard to the inclusion of activities for motor abilities. In individuals without a cleft lip and palate, the DDK is altered when speech disorders are present. A structurally compromised oral cavity is a long-term condition present in people with a cleft lip and palate even after surgery, thereby resulting in functional speech adaptations. This study aimed to correlate oral motor performance in DDK tasks based on the type of cleft and the presence of a lisp in the speech of children who had undergone surgery for a cleft lip and palate.

METHODS

This retrospective, non-randomized study was approved by the Human Research Ethics Committee of the Hospital for Rehabilitation of Craniofacial Anomalies at the *Universidade de São Paulo* (USP) under the number 406,375. Further, this study was part of a wider prospective research project that was approved by this committee under the number 288/2009. Administration of the Free and Informed Consent Form was not required because previously collected data were evaluated in this study. The documents from the complete sample population in the larger study were analyzed. The study comprised 60 children, aged between 9 and 12 years, who were randomly selected and distributed into 2 groups: 1 group comprised 30 children (14 girls and 16 boys) with a unilateral cleft lip and palate (UCLP) and the other group comprised 30 children (18 girls and 12 boys) with an isolated cleft palate (CP).

The children underwent primary surgeries for lip repair until they were 6 months of age and palate repair surgery between 12 and 24 months of age in the same institution where the study was designed. Children were excluded from the study if the following criteria applied: secondary lip repair surgeries; intraoral braces that interfered with execution of the required tasks; oronasal fistulas; moderate to severe dysphonia; lingual frenulum with short or fixed extension in close proximity to the tongue tip; neurologic conditions or syndromes; compensatory articulations while pronouncing plosive phonemes [t] and [k], and speech disfluency.

Diadochokinesis test

The number of emissions produced per second (s) as well as the mean time between the emissions in milliseconds (ms) were analyzed by using data obtained from the DDK test. The DDK test was conducted in an acoustically appropriate room, where the individuals remained seated with a microphone (AKG, C420) laterally positioned at 60°, 10 cm from the labial commissure. The microphone was adapted to a sound table (Behringer® 8 channels, XENYX/X1204USB) connected to an Intel® Pentium® 4 computer (CPU 1.80 GHz and 256 MB RAM; sound module Audigy II, Creative).

Sample speech recordings of repetitions of the monosyllables "ta" and "ka" and the sequence "pataka" were acquired for 6 seconds each[11]. The "pa" syllable was not tested because it is a bilabial articulatory point and the objective was to study the lisp, which is a disorder that involves the motor ability of the tongue rather than the lip. All children had been previously instructed and trained to produce emissions as fast as possible during the evaluation with clear and precise articulation[11]. After training, the emissions were recorded 2–3 times, and the samples were stored by using the SoundForge 8.0 software (Sony®) at a sampling rate of 44.100 Hz, mono-channel in 16-bit.

One person performed and edited the recordings for subsequent analyses of the emissions of both the monosyllables "ta" and "ka" and the "pataka" sequence. The first 2 seconds of the emissions of "ta" and "ka" were excluded from the analyses by using the MSP software Model 5141, version 2.5.2 (KayPENTAX™), and the number of emissions was considered in 3-second intervals[11]. The same software, which automatically provides the number of emissions per second and the mean time between the emissions, was used to perform the analyses. For the "pataka" sequence, both editing and quantitative analysis were performed by using the SoundForge 8.0 (Sony®) editing software, excluding the first 2 seconds and retaining the next 3 seconds. The number of emissions per second was manually counted by using visual and auditory tracks because the MSP software considers the trisyllabic sequence to be 3 separate monosyllables rather than a single emission[11].

The emission for the "pataka" sequence was not registered when the child could not perform the emissions correctly even after training.

Analysis of the presence of lisp speech

We analyzed speech samples that were recorded with an audio-visual system by using a digital camera recorder (Sony® DCR-DVD810) supported by a tripod placed 1 meter from the seated patient. Such samples involved the repetition of sentences that comprised words belonging to a script of target sounds, each of which contained 10 occurrences: 4 in the initial syllable, 3 in the middle syllable, and 3 in the final syllable[8]. The alveolar phonemes [s] and [z] in a total of 20 emissions were taken into account during the analysis. The speech samples were stored on portable devices in ascending order according to the case number of the study, and were given to each evaluator.

Two experienced speech evaluators received verbal and written instructions to analyze the samples and register the presence of a lisp when an audible phonetic distortion was detected during production of phonemes [s] and [z][21]. To this end, an individual and separate analysis was conducted in a quiet environment with the use of headphones. The results were subsequently analyzed. In cases with contradictory results, a third experienced person who was informed that he/she served as a third experienced evaluator, was required to analyze the speech samples in order to resolve the impasse. Therefore, considering the agreement between the 2 evaluators, the presence of a lisp was registered in at least 5 out of 10 emissions of each phoneme[22]. It is important to note that sample randomization was not performed for data analysis.

The agreement of the evaluators was verified with a Kappa test, which revealed good agreement, i.e., 85% (Kappa=0.85) and 90% (Kappa=0.90) for the phonemes [s] and [z], respectively. Because the agreement between the evaluators was satisfactory, the intra-evaluator reliability was not verified. The analysis of speech disorder frequency according to the cleft type was verified by using the Chi-square test. The DDK results between the 2 cleft groups, as well as between groups with and without a lisp, were compared by using the t-test for independent samples. In all analyses, p values <0.05 were considered significant.

RESULTS

The DDK results between the UCLP and CP groups with regard to the number of emissions per second and the mean time between emissions in milliseconds were compared by using the t-test for independent samples. No significant differences were observed between the groups in either comparison (Table 1).

Analysis of the frequency of a lisp based on the type of cleft revealed a higher frequency (60%) in the UCLP group (p=0.00882) (Table 2).

No significant differences were observed while comparing the values of the DDK parameters between groups with and without a lisp when the group was considered as a whole, regardless of the type of cleft (Table 3), or when each group was considered individually (Tables 4 and 5).

Table 1. Comparison between the means and standard deviations of the number of emissions per second and mean time per second between the groups

Groups	Emissions				
	"ta"		"ka"		"pataka"
	Emissions per second	Mean time between the emissions (ms)	Emissions per second	Mean time between the emissions (ms)	Emissions per second
CP	4.66 ± 0.61	217.66 ± 26.25	4.06 ± 0.46	249.74 ± 31.98	1.64 ± 0.16
UCLP	4.65 ± 0.48	217.49 ± 23.60	3.86 ± 0.58	264.27 ± 38.46	1.67 ± 0.16
p-value	0.922	0.985	0.149	0.117	0.614

T-test for independent samples (p<0.05)
Note: CP = isolated cleft palate; UCLP = unilateral cleft lip and palate

Table 2. Lisp speech frequency according to the type of cleft

Lisp	Group		p-value	Total
	CP	UCLP		
Present	23.33% (n=7)	60.00% (n=18)	p=0.00882*	41.67% (n=25)
Absent	76.67% (n=23)	40.00% (n=12)		58.33% (n=35)

Chi-square test (p<0.05)
*Higher frequency in the UCLP group
Note: CP = isolated cleft palate; UCLP = unilateral cleft lip and palate

Table 3. Comparison of the diadochokinesis values according to the presence of lisp in speech while considering the whole group

Emissions		Lisp		p-value
		Present (n=25)	Absent (n=35)	
"ta"	Emissions per second	4.56 ± 0.45	4.72 ± 0.60	0.252
	Mean time between the emissions (ms)	221.40 ± 21.76	214.81 ± 26.65	0.313
"ka"	Emissions per second	3.92 ± 0.57	3.99 ± 0.50	0.590
	Mean time between the emissions (ms)	260.30 ± 37.47	254.59 ± 34.95	0.541
"pataka"	Emissions per second	1.67 ± 0.18	1.64 ± 0.15	0.528

T-test for independent samples (p<0.05)

DISCUSSION

Evaluating the oral motor function of patients with a cleft lip and palate is critical since these individuals have a structurally compromised oral cavity from birth, and this condition can persist for a long time during rehabilitation. Structural alterations of the oral cavity favor functional adaptations, especially of the tongue, in order to perform orofacial functions[2,6-8]. In addition, the presence of velopharyngeal dysfunction and dento-occlusal alterations cause specific speech disorders[1,2,5,9,23].

Therefore, evaluating oral motor performance in individuals with a cleft lip and palate could help clarify whether the functional adaptations developed by the individual to compensate for the structural deformity would compromise this ability. DDK analysis enables evaluation of oral motor ability by using tasks that involve quick repetitions of syllables and syllabic sequences[10,11].

Table 4. Comparison of the diadochokinesis values according to the presence of lisp in speech in the UCLP group

Emissions		Lisp		p-value
		Present (n=18)	Absent (n=12)	
"ta"	Emissions per second	4.61 ± 0.48	4.70 ± 0.49	0.605
	Mean time between the emissions (ms)	219.21 ± 23.45	214.91 ± 24.63	0.633
"ka"	Emissions per second	3.81 ± 0.58	3.95 ± 0.58	0.507
	Mean time between the emissions (ms)	263.33 ± 38.97	258.19 ± 38.53	0.489
"pataka"	Emissions per second	1.70 ± 0.16	1.59 ± 0.17	0.106

T-test for independent samples (p<0.05)
Note: UCLP = unilateral cleft lip and palate

Table 5. Comparison of the diadochokinesis values according to the presence or absence of lisp in speech in the CP group

Emissions		Lisp		p-value
		Present (n=7)	Absent (n=23)	
"ta"	Emissions per second	4.43 ± 0.35	4.73 ± 0.66	0.253
	Mean time between the emissions (ms)	227.02 ± 16.86	214.75 ± 28.18	0.287
"ka"	Emissions per second	4.21 ± 0.45	4.02 ± 0.46	0.340
	Mean time between the emissions (ms)	239.96 ± 25.23	252.71 ± 33.68	0.364
"pataka"	Emissions per second	1.59 ± 0.20	1.65 ± 0.15	0.399

T-test for independent samples (p<0.05)
Note: CP = isolated cleft palate

In order to obtain homogeneous groups, children who had undergone surgery in the same institution at the ages recommended in the literature were selected for this study. This study group included children who had mastered the analyzed phonemes and had precise articulation; further, the selected children had mixed dentition with the central and lateral incisive teeth already present[(21)] because the absence of these teeth could cause false results.

The analysis of the obtained results revealed a similarity between the UCLP and CP groups with regard to the parameters of the mean time between emissions (ms) and number of emissions per second for all tested repetitions. Low variation was observed in the values for both the groups, as indicated by the standard deviation. This result indicates that, despite the more extensive structural alterations in the UCLP group, these alterations did not affect motor ability with regard to production of the syllables "ta" and "ka" and the "pataka" sequence.

Even though a control group was not included, the results of the present study were consistent with the previously reported values for individuals (aged 5–93 years) without a cleft when the number of emissions per second was compared[(10,11)]. The values obtained for the syllable "ta" were 4.66 ± 0.61 in the CP group and 4.65 ± 0.48 in the UCLP group, with a possible variation between 3.33 and 7.1. The values for the syllable "ka" were 4.06 ± 0.46 and 3.86 ± 0.58 for the CP and UCLP group, respectively, with a possible variation between 3.18 and 6.6.

For emission of the trisyllabic sequence "pataka," the values of 1.64 ± 0.16 and 1.67 ± 0.16 obtained for the CP and UCLP group, respectively, are consistent with those reported in previous studies[(10,11)].

Lisp frequencies of 60% and 23.33% were observed during speech production in the UCLP and CP groups, respectively, with a significantly higher alteration frequency in the group with a cleft lip and palate. This result can be explained by the incorrect positioning of the tongue, which affects articulatory points and is occasionally found as a result of the dento-occlusal balance condition, structural alteration, and reduced intraoral cavity, even after surgical repair[(1,5-8,10-19, 23-25)].

On the other hand, the presence of a lisp was not expected in the CP group because this type of cleft does not require extensive occlusal alterations. However, while evaluating 40 children aged 4–10 years without a cleft, a single study reported that the phonemes most frequently distorted were [s] and [z][(21)].

Analysis of oral motor performance during DDK tasks while considering lisp interference in both of the studied groups did not reveal any differences between the values for individuals with and without a lisp. This result indicates that, despite a more anterior tongue position to compensate for structural alteration and to enable the production of alveolar phonemes, motor ability was not affected according to the DDK evaluation. This may be due to the functional adaptation acquired by these individuals during their lifetime. This finding is inconsistent with those of previous studies in which DDK was compared between individuals with different types of articulatory disorders and individuals without speech problems[(12,13,16)].

This study had several limitations. First, we were not able to study a large sample because of the specific inclusion criteria; this was especially true for the criterion regarding the presence of compensatory articulations during the production of phonemes that are the most affected in this population, i.e., [t], [k], [s], and [z][(1,3,9)]. Furthermore, because only a few published studies used DDK to evaluate individuals with a cleft lip and palate, our ability to compare our results with those of previous studies was limited.

CONCLUSION

DDK task performance was not altered by the type of cleft palate. The presence of a lisp was more frequent in children with a complete UCLP. However, the presence of a lisp during speech production did not affect DDK task performance.

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