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ARTICLE

Oral dyspraxia in self-limited epilepsy with centrotemporal spikes: a comparative study with a control group

Dispraxia oral em epilepsia autolimitada com espículas centrotemporais: um estudo comparativo com grupo controle

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ABSTRACT

Background: self-limited epilepsy with centrotemporal spikes, previously considered benign focal childhood epilepsy with centrotemporal spikes show clinical signs of involvement of Rolandic areas, mainly lower area, which may affect the planning and execution of motor sequences. **Objective:** This study aimed to evaluated oral praxis in children with self-limited epilepsy with centrotemporal spikes and compare to the age-matched control group. **Methods:** This was a descriptive study with 74 children with self-limited epilepsy with centrotemporal spikes, with the classical forms according to International League Against Epilepsy, and between 4 and 15 years of age, selected from the child neurology outpatient clinic of the Hospital das Clínicas, Faculdade de Medicina da Universidade de São Paulo, São Paulo, Brazil, and 239 age-matched and educational level-matched (convenience sampling) control children. All children were submitted to the battery of oral volitional movements, which consisted of 44 tests for oral movement (tongue, lip, cheek, jaw, and palate) and 34 phonemes and consonant cluster tasks, with simple and sequenced oral movements. **Results:** The mean age and standard deviation (SD) of children with epilepsy was 9.08 years (SD 2.55) and of controls 9.61 years (SD 3.12). The results showed significant differences between the groups with a poorer performance of children with epilepsy compared to children without epilepsy in simple and particularly in sequenced movements. **Conclusion:** These findings can be attributed to the genetically determined immaturity of cortical structures related to motor planning in children with self-limited epilepsy with centrotemporal spikes.

Keywords: Epilepsy; Epilepsy, Rolandic; Apraxias.

RESUMO

Antecedentes: Epilepsia autolimitada com descarga centrotemporal, previamente designada por epilepsia benigna focal infantil com espículas centrotemporais, mostra sinais clínicos de envolvimento de áreas rolândicas, principalmente área inferior, que podem afetar o planejamento e a execução de sequências motoras. **Objetivo:** Este estudo visou avaliar a práxis oral em crianças com epilepsia autolimitada com espículas centrotemporais e comparar com o grupo de controle de mesma idade e grau de escolaridade. **Métodos:** Tratou-se de um estudo descritivo, com 74 crianças com epilepsia autolimitada com espículas centrotemporais selecionadas no ambulatório de neurologia infantil do Hospital das Clínicas da Faculdade de Medicina da Universidade de São Paulo, São Paulo, Brasil, e 239 crianças do grupo controle da mesma faixa etária e grau de escolaridade. Todas as crianças foram submetidas à bateria de tarefas de movimento oral volitivo, que inclui movimentos orais simples e sequenciados. **Resultados:** A idade média das crianças com epilepsia era de 9,08 anos (desvio padrão — DP 2,55) e dos controles 9,61 anos (DP 3,12). Os resultados mostraram diferenças significativas entre os grupos, com desempenho mais fraco das crianças com epilepsia em comparação ao das crianças saudáveis, em movimentos simples e particularmente em movimentos sequenciados. **Conclusão:** Esses resultados podem ser atribuídos à imaturidade geneticamente determinada das estruturas corticais relacionadas com o planejamento motor em crianças com epilepsia autolimitada com espículas centrotemporais.

Palavras-chave: Epilepsia; Epilepsia Rolândica; Apraxias.

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INTRODUCTION

Self-limited epilepsy with centrotemporal spikes (SLCT) or Rolandic epilepsy, previously considered benign focal childhood epilepsy with centrotemporal spikes, is the most common form of self-limited or drug-responsive focal epilepsy[1,2,3]. It represents almost 15% of all childhood epilepsy cases and 13 to 25% of new-onset epilepsy in children, beginning between 2 and 12 years of age (peak incidence around 6 to 7 years)[2,4,5], and predominantly in males[6,7]; it is not associated with an underlying structural lesion[8]. The abnormalities observed in the electroencephalogram (EEG) are probably associated with an autosomal dominant inheritance with age-related penetrance[9] and consist of centrotemporal spikes (CTS), focal, high-amplitude central or mid-temporal surface negative spike, or sharp waves followed by slow waves. Spontaneous recovery occurs before adolescence[5].

Seizures are infrequent, often single and brief, and awareness is usually preserved. Oro-pharynx-laryngeal and unilateral facial manifestations (motor and/or sensitive) suggest that the focal pathology is related to the inferior Rolandic cortex, where the face and oropharynx are represented[10].

Therefore, although normal neurological and intellectual development are accepted as criteria, some studies have shown that children with SLCT may develop other deficits involving attention, perception, short- and long-term declarative memory with both verbal and non-verbal material, visuomotor coordination in reading, spelling, and speaking, and these problems may be associated with patterns of EEG abnormalities in SLCT[11,12]. The pathophysiological mechanisms by which SLCT induces neuropsychological impairment remain unclear; however, the relation between neuronal network disorganization promoted by epileptic discharge and neuropsychological dysfunction[13] might be involved.

Lundberg et al. reported that children with SLCT showed abnormalities on the performance of tongue movements and on the emission of nonsense words, compared to a control group[14]. Previously, Deona et al. reported patients with interictal oral dyspraxia[15], and Scheffer et al. described an Australian family with oral dyspraxia and cognitive impairment, associated with epileptiform discharges over the centrotemporal region[16]. This family presented clinical anticipation of these symptoms because they had been genetically determined. SLCT with language and speech disorders is an autosomal dominant disease and shows anticipation[17]. This phenomenon may be due a specific gene for SLCT, not identified yet, or due to a mutation in Xq22, of gene SRPX2, which was identified by Roll et al. as the responsible for Rolandic seizures associated with cognitive impairment and oral dyspraxia[18].

The exact definition of dyspraxia in children remains controversial, although there is broad agreement that it involves a disorder of planning, organization and coordination movements. Two suggestions were offered to define dyspraxia in the inaugural United Kingdom (UK) interdisciplinary forum in 1994: "In the absence of any known neurological condition or intellectual impairment, dyspraxia is the inability to plan, organize and coordinate movements. It results in fine and gross motor problems and/or speech difficulties". "Dyspraxia children are those who, in the absence of physical and/or neurological disorder, have difficulties in control and coordination of voluntary motor activity. The condition is developmental, rather than acquired"[19].

Thus, oromotor dyspraxia is a specific form of developmental dyspraxia that occurs in children without neurological abnormalities, which appear to prevent or make difficult the movement of isolated or sequential laryngeal or supralaryngeal muscles. The difficulty in making and coordinating the movements of the oral muscles is not related to speech production. Concerning the difference between oral dyspraxia and verbal dyspraxia, Rimmer and Hartley affirm that in oral dyspraxia there is difficulty making and coordinating movements of the oral and buccal musculature unrelated to speech production[20].

During the first years of life, children acquire many motor abilities, and this learning is not strictly related to motor maturation, but it also requires interaction with the environment. The non-speech oral movements depend on the mouth area of the premotor cortex. The frontal lobes are the main brain structures responsible for planning, organizing and executing movements. No studies have addressed oral movements in children without structural brain damage. This investigation may be difficult in children, because many oral skills are still developing[21]. Also, the cortico-cerebellar activation is evident in sequence learning[22].

A study by Ciumas et al. showed that children with SLCT have alterations in the microstructure of the white matter, predominating over the regions displaying chronic interictal epileptiform discharges[23]. The association between diffusion tensor imaging changes, duration of epilepsy and cognitive performance appears compatible with the hypothesis that interictal epileptic activity alters brain maturation, leading to cognitive dysfunction[23].

Considering the above information, the evaluation of oral movements should be part of the neurological examination of children with SLCT. The need to evaluate children with epilepsy compared to age-matched controls is justified by the different stages of cortical maturation, thus avoiding misunderstandings in the interpretation of the oral volitional movements. This study aimed to assess oral praxis in children with self-limited epilepsy with centrotemporal spikes and compare to the age-matched control group. In the present study, there was no address of speech dyspraxia, only of oral-motor dyspraxia.

METHODS

This descriptive study was conducted at Hospital das Clínicas, Faculdade de Medicina da Universidade de São

Paulo, São Paulo, Brazil. The patient group was recruited from the child neurology outpatient clinic of the Hospital das Clínicas, and the control group was selected in public and private schools in the same district of residence of patients, preferably.

The participants were divided into two groups: Group 1 — the experimental group consisted of 74 children with typical SLCT, according to ILAE[7], and group 2 — the control group consisted of 239 children without epilepsy. Groups were similar in age, sex, and schooling, verified by the Kolmogorov-Smirnov test ($p>0.10$ for all variables) and by the Kruskal-Wallis test ($p=0.17$). The control children were selected by convenience sampling, but priority was given for children from the same school or neighbors of the patients.

Inclusion criteria

Group 1: signing the informed consent term by the mother or legal guardian of the child; both sexes; between 4 and 15 years of age; clinical and EEG diagnosis of SLCT; last seizure more than 30 days before the test; no abnormalities in neuroimaging; no history of gestational, delivery or neonatal problems; no developmental delay; no chronic disease; no severe psychiatric illness; no learning disorder according to school report and family information; no mental disabilities (all children in group 1 underwent neuropsychological assessment — Wechsler Intelligence Scale for Children [WISC 3]); and no diagnosed genetic diseases.

Group 2: signing the informed consent term by the mother or legal guardian of the child; both sexes; between 4 and 15 years of age; no personal or family history (up to third degree) of epilepsy; no previous history of gestational, delivery or neonatal problems; no developmental delay; no chronic disease; no severe psychiatric illness; no learning disorder according to school report and family information; no genetic diseases diagnosed; and no medications, except antiepileptic medication.

Exclusion criteria (both groups): three or more failed attempts in any of the 34 tasks of Portuguese sounds. All children were submitted the oral volitional movement tasks, which include simple and sequenced oral movements. This was an adaptation and expansion of the battery proposed by Crary and Anderson (1990), which included 44 tests of oral movement (Table 1) and 34 phonemes and consonant clusters tasks. The stimulus modality was by imitation[24].

The tasks were divided into five parts: tongue movements (T1 to T19), lip movements (T20 to T35), cheek movements (T36 to T39), jaw movements (T40 to T43), and palate movements (T44).

Position of the examiner and demonstration of the exam: the examiner sat in front of the child, without any object between them, and explained what the test would look like and made a demonstration. In sequenced tests, in which movements were counted, the test time was five seconds, because in the pilot tests the children under seven years of age had great difficulty in sustaining attention in the tests for more than five seconds. To maintain the standard for all ages researched, this time was established.

In simple tasks, a single movement was reproduced, while in sequential tasks, movements were repeated for five seconds. The examiner presented the movement to the child and the child was oriented to proceed with the imitation. When the child was unable to perform the movement, the examiner repeated the orientation and performed the proposed movement again; after the third failure, the task was considered incorrect. The simple tests were: T1, T2, T3, T4, T5, T6, T7, T8, T9, T13, T14, T16, T18, T19, T20, T21, T22, T23, 24, T25, T26, T28, T29, T30, T31, T32, T33, T34, T35, T36, T37, T38, T39, T40, T41, T42, T43, T44 (Table 1).

In the sequenced tests, the child was asked to start the movements and stop at the examiner's command. The examiner would demonstrate the test for 5 seconds. Each completed movement was counted as one point.

The other battery was composed of 34 tasks to evaluate the production of Portuguese language sounds, using phonemes and consonant clusters (Table 2). The examiner emitted the sound and asked the child to repeat it; only after three failed attempts, was the task considered incorrect.

The study protocol, registrations, and patient consents were approved by the Ethics Committee of the Comitê de Ética da Faculdade de Medicina da USP. This study followed the ethical criteria determined by the Resolution of the National Health Council no. 466 of 2012, which is based on the Declaration of Helsinki.

Statistical analysis was done using the Kolmogorov-Smirnov non-parametric test to compare the performances (number of movements performed in five seconds) of the experimental and control groups on the sequenced movement tasks (T10, T11, T12, T15, T17 and T27).

For qualitative variables, contingency tables were built, and the chi-square tests was used to compare the performances (number of correct vs. number of incorrect answers) of the experimental and control groups on the simple tasks (Table 1). If any category had an $n<5$, Fisher's exact test was used (Hollander and Wolfe, 1973). The level of significance was set at 5%. The software Statistica 12.0 was used for all the analyses.

RESULTS

Three hundred ninety-seven children were evaluated (104 with epilepsy and 293 controls) and 313 (74 children with epilepsy and 239 controls) were selected. The main cause of exclusion (65 children, 87.8%) was the failure to complete the test battery: 30 were from the epilepsy group and 54 from the control group.

Table 1. Tasks.

Tongue movements tests	
T1. Protrude the tongue	T11. Lateralize the tongue tip to the left angle of the lip and to the right angle quickly (repeat for 5")
T2. Put the tongue tip on the right lip angle	T12. Move the tongue tip up and down touching the lips quickly (repeat 5")
T3. Put the tongue tip on the left lip angle	T13. Elevate and keep the tongue tip on the papillae (repeat for 5")
T4. Put the tongue tip, internally, on the right cheek	T14. Click the tongue tip against the papillae
T5. Put the tongue tip, internally, on the left cheek	T15. Click the tongue tip against the papillae quickly (repeat for 5")
T6. Put the tongue tip on the papillae with the open mouth	T16. Suck the tongue against the palate
T7. Turn down the tongue internally with the open mouth	T17. Suck the tongue against the palate and click quickly (repeat for 5")
T8. Put the tongue tip on the upper lip	T18. Vibrate the tongue tip
T9. Put the tongue tip on the lower lip	T19. Elevate the dorsal face of the tongue several times emitting the sound "KA"
T10. Move the tongue in and out quickly (Repeat for 5")	
Lip movements tests	
T20. Protrude the lips	T28. Vibrate the lips
T21. Protrude the lips forming a closed "beak"	T29. Click the lips as kissing
T22. Retract the lips as a closed smile	T30. Click the retracted lips as kissing
T23. Retract the lips as an open smile	T31. Bite the lower lip
T24. Retract the lips inside the oral cavity	T32. Bite the upper lip
T25. Make a closed "beak" and divert to right without move the jaw	T33. Blow out
T26. Make a closed "beak" and divert to left without move the jaw	T34. Whistle
T27. Alternate the closed "beak" to left and right quickly (repeat for 5")	T35. Suck the own finger
Cheek movements tests	
T36. Fill the cheek up with air	T38. Fill the left cheek up with air
T37. Fill the right cheek up with air	T39. Pass the air from right to left
Jaw movements tests	
T40. Open and close the mouth	T42. Move the jaw left
T41. Move the jaw right	T43. Move the jaw forward
Palate movements test	
T44. Provoked move of the palate (a, ã)	

Table 2. List of phonemes and consonant clusters.

45. Pê	53. Nhê	62. Rê	70. Frê
46. Tê	54. Fê	63. Rrê	71. Vrê
47. Kê	55. Sê	64. Pré	72. Plê
48. Bê	56. Chê	65. Tre	73. Tlê
49.Dê	57. Vê	66. Krê	74. Klê
50. Guê	58. Zê	67. Brê	75.Blê
51. Mê	59. Ge	68. Drê	76. Glê
52. Nê	60. Lê	69. Grê	77. Flê
	61. Lhe		78. Vlê

The mean age of children with epilepsy was 9.08 years (SD 2.55) and of controls 9.61 years (SD 3.12), the age difference in the groups was not significant (p=0.17) (Table 3). The epilepsy group was composed of 30 girls and 44 boys, and the control group was composed of 95 girls and 144 boys.

The test battery that assesses oral praxis was composed of 19 movements of the tongue, 16 of the lip, 4 of the cheeks and one of the palate, and none of the oral gestures involved speech.

The sequenced oral tasks (T10, T11, T12, T15, T17 and T27) differed between the experimental and control groups. In all six tasks, the experimental group showed lower medians than the control group (Table 4). Task 13 (T13 - elevate

Table 3. Descriptive measures and test for evaluation of age distribution for case and control groups.

Group	n	Median	SD	n	Min	Max	Median	p-value
Control	239	9.61	3.12	239	4.00	15.00	10.00	0.17*
Experimental	74	9.08	2.55	74	4.00	15.00	9.00	

*Kruskal-Wallis test.

Table 4. Descriptive measures and test for assessing the distribution of praxis assessments measured quantitatively for the experimental and control groups.

Group	Variables	Mean	SD	n	Min	Max	Median	p-value
Control	T10	16.08	5.75	239	0.00	32.00	15.00	<0.0001*
Experimental	T10	11.28	5.39	74	0.00	24.00	12.00	
Control	T11	16.80	5.57	239	0.00	32.00	16.00	<0.0001*
Experimental	T11	12.54	5.98	74	0.00	24.00	12.00	
Control	T12	9.54	4.79	239	0.00	24.00	10.00	<0.0001*
Experimental	T12	6.68	4.30	74	0.00	16.00	7.50	
Control	T15	19.20	4.41	239	0.00	29.00	20.00	<0.0001**
Experimental	T15	14.96	5.64	74	0.00	24.00	16.00	
Control	T17	13.97	4.45	239	0.00	30.00	13.00	<0.0001*
Experimental	T17	9.14	4.92	74	0.00	19.00	10.00	
Control	T27	8.92	6.43	239	0.00	22.00	10.00	<.0001**
Experimental	T27	4.77	5.36	74	0.00	15.00	0.00	

*ANOVA; **Kruskal-Wallis test. T10 - Move the tongue in and out quickly; T11: Lateralize the tongue tip to the left angle of the lip and to the right angle quickly; T12: Move the tongue tip up and down touching the lips quickly; T15: Click the tongue tip against the papillae quickly; T17: Suck the tongue against the palate and click quickly; T27: Alternate the closed "beak" to left and right quickly.

and keep the tongue tip on the papillae (repeat for 5") was not analyzed, as the data was lost.

Significant differences were found between the groups for correct and incorrect answers, with a significantly higher number of errors in the experimental group for the tasks T5, T6, T7, T16, T18, T33 and T43 (Table 5).

DISCUSSION

The present study evaluated simple and sequenced oral gestures (oral praxis) in children with SLCT and in the control group without abnormalities in speech production. The imitation stimulus was applied in this research, and correct and incorrect responses were analyzed using one qualifier. The results show that the performance of the experimental group was poorer than that of control group in simple and sequenced production of gestures, especially related to the tongue and lips.

Oral motor actions required in the production of speech sounds were not evaluated, as the objective was to evaluate the planning and execution skills of single or sequential voluntary movements (praxis) of the orofacial systems in children with Rolandic epilepsy.

The children in the epilepsy group failed in the six sequenced movement (tongue and lips) tasks, differently than the control. Among the qualitative tasks, there were differences between the groups in five tongue movements, four lip movements, and one mandibular movement.

Praxis skills progress according to brain maturity and, for this reason, research involving praxis assessment in children must consider the neurological evolution in different age groups before considering the failure of responses as an abnormality. One strategy to avoid this problem is to conduct studies with an age-matched control group.

The evaluation of oral praxis in children is particularly challenging because the cortical maturation is still under way. Motor learning is based on the content and construct of a motor program. In this way, orofacial praxis "is the ability to plan and execute movements or sequences of voluntary movements, meaningful or not, using the muscles of the pharyngo-buccofacial system or the orofacial region", according Bearzotti et al.[25], reinforcing the importance of evaluating oral praxis in children considering their age.

Table 5. Qualitative tasks with significant difference between groups (experimental and control).

Tasks	Production	Control	Experimental	Total	p-value
T5	Correct	238	71	309	0.04
	Incorrect	01	03	04	
T6	Correct	239	72	311	0.05
	Incorrect	0	02	02	
T7	Correct	231	66	297	0.02
	Incorrect	08	08	16	
T16	Correct	237	68	305	0.00
	Incorrect	02	06	8	
T18	Correct	191	51	242	0.05
	Incorrect	48	23	71	
T25	Correct	195	58	253	0.01
	Incorrect	44	16	60	
T26	Correct	194	53	247	0.00
	Incorrect	45	20	65	
T28	Correct	218	65	283	0.01
	Incorrect	45	20	65	
T33	Correct	238	66	304	<0.00
	Incorrect	1	07	08	
T43	Correct	229	66	295	0.04
	Incorrect	10	08	18	

Chi-squared. T5: Put the tongue tip, internally, on the left cheek; T6: Put the tongue tip on the papillae with the open mouth; T7: Turn down the tongue internally with the open mouth; T16: Suck the tongue against the palate; T18: Vibrate the tongue tip; T25: make a closed "beak" and divert to right without move the jaw; T26: make a closed "beak" and divert to left without move the jaw; T28: Vibrate the lips; T33: Blow out; T43: Move the jaw forward.

Few studies have investigated the performance of oral praxis tasks, and little data are available for both adults and children. Similarly, very little is known about how to develop oral praxis in children according to age groups[26].

In the quantitative oral praxis tasks, the epilepsy group was significantly worse than the control. Volitional sequenced movements involve many complex conditions or factors that depend on the synchronous operation of several cortical and subcortical areas[27], and in the motor cortex the movement is planned and implemented[28]. Children may not be able to perform some movements due to incomplete motor neuronal maturation[29], and for this reason, it is important to consider the children's age during the praxis evaluation.

Studies about non-speech oral tasks that evaluated the motor control of the orofacial and laryngeal systems show that young children depend on guiding movements, during the onset of movement. This occurs probably due to the instability in the central nervous system[30]. The importance of the perisylvian region for language functions is undeniable, but this area is also involved in motor execution, especially in sequenced movements[31].

Epileptic discharges involving the perisylvian neuronal network region might be responsible for oral dysfunctions observed in children with SLCT, such as oral dyspraxia. Halász et al. have described the occurrence of epileptic activity in the surrounding areas of the Sylvian fissure[32].

Many studies correlate the performance of verbal tasks to the activation of the Broca's area (Brodmann's area B44 and B45) during speech generation and of perception[33,34]. In addition, Broca's area is involved in motor processing and control[35], and by proximity to the premotor cortex, this area is related to oral and facial control, especially the lips and tongue[36]. It is important to emphasize the great cerebral representation of the oral cavity, "reflecting its sensitivity and dexterity"[37].

Besseling et al. showed that there is reduced structural connectivity in children with SLCT in the several connections involving the Rolandic regions, from which the epileptiform activity originates. Most of these aberrant tracts involve the left hemisphere (which mediates language skills), notably the pars opercularis of the inferior frontal gyrus (Broca's area) and the supramarginal gyrus (Wernicke's area). The authors described that microstructural white matter alterations were correlated with language impairment in children with SLCT[38].

Watanabe et al. analyzed left and right movements of the tongue and noted the involvement of the bilateral dorsal premotor area, superior parietal lobule and the inferior parietal lobule. The bilateral parietal lobule was involved in the processing of the human tongue movement[37].

A study by Ayaz et al. evaluated fine motor skills in children with SLCT, and the results showed that the children with self-limited epilepsy did not perform as well as the controls. Epileptic focus, treatment status, type of antiepileptic treatment, age at the time of the first seizure, time since the last seizure, and total number of seizures did not affect motor skills. SLCT negatively affected fine motor skills, regardless of the children's level of intelligence[39].

The presence of oral dyspraxia in children with SLCT may indicate a dysfunction of brain regions involved in the planning and/or execution of complex volitional movements, e.g. the inferior motor Rolandic area[14]. According to Staden et al., the combination of oral dyspraxia and seizures with oral deficiencies (drooling and speaking problems) suggests that the areas responsible for ideation and execution of complex movements of speech overlap[11].

We believe that SLCT is an excellent model for the evaluation of cognitive functions in children with epilepsy, especially those focusing on praxis, attention and memory capacities, since children with SLCT do not present structural abnormalities or severe symptoms. In most cases, antiepileptic medication is not used, which also avoids deleterious bias effects.

In conclusion, children with SLCT showed poorer performance than children without epilepsy on tasks involving

simple and, more obviously, sequenced oral movements. Many children with SLCT showed oral dyspraxia in the oral volitional movements' battery. The major importance of this study was the realization of praxis assessment in children with and without epilepsy (matched) because it eliminated the possibility that the oral dyspraxia observed in children with SLCT is due to developmental dyspraxia, reinforcing the probability that the neurological abnormality is due to the influence of epileptic discharges in the Rolandic area.

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In memory of Letícia Lessa Mansur.

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