ABSTRACT
This study investigated how the difficulties in language in children with Rolandic Epilepsy (RE) could be related to alterations in their development of phonological awareness and/or working memory. We evaluated forty-two children aged 6 to 13 years old. From these, twenty-one children were diagnosed with RE and formed the experimental group; and twenty-one children without RE, paired with the experimental group by sex, age, education and socioeconomic status, formed the control group. The results showed significant differences in the performances of children with RE and healthy children in the tests that evaluated working memory and phonological awareness. Also, positive and high significant correlations were found between working memory and phonological awareness in the RE clinical subgroup. Generally, the results suggest that compromises in both cognitive functions might be associated to loss of language capabilities in children with RE, and also point that the development of working memory and phonological awareness are interconnected.

Keywords
Working memory; Phonological awareness; Language; Rolandic epilepsy

RESUMEN
En este estudio se investigó cómo las dificultades en el lenguaje en niños con epilepsia Rolándica (ER) podrían estar relacionadas con alteraciones en su desarrollo de la consciencia fonológica y/o memoria de trabajo.


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Se evaluaron 42 niños de 6 a 13 años de edad. De éstos, 21 niños fueron diagnosticados con ER y formaron el grupo experimental; y 21 niños sin RE, emparejados con el grupo experimental por sexo, edad, nivel educativo y socioeconómico, que formaron el grupo de control. Los resultados mostraron diferencias significativas en los resultados de los niños con ER y niños sanos en las pruebas que evaluaron la memoria de trabajo y la conciencia fonológica. Además, se encontraron correlaciones positivas y altas entre la memoria de trabajo y la conciencia fonológica en el subgrupo clínico RE. En general, los resultados sugieren que los compromisos en ambas funciones cognitivas pueden estar asociados a la pérdida de capacidades del lenguaje en los niños con RE, y también señalan que el desarrollo de la memoria de trabajo y la conciencia fonológica están interconectados.

Palabras clave
memoria de trabajo; conciencia fonológica; lenguaje; epilepsia rolándica

Introduction

This study aimed to investigate the working memory and phonological awareness in children between 6 and 13 years who were diagnosed with rolandic epilepsy, also known as Benign Childhood Epilepsy with centro-temporal spikes. It is a benign epilepsy syndrome, without apparent anatomical brain lesions, and it is the most common form of focal seizures in childhood, accounting for 25% of all seizures in this period. Usually, RE occurs with infrequent partial seizures which occasionally happen only one time, predominantly in sleep, usually with a satisfactory response to drug treatment and spontaneous remission of seizures in the period of one to three years after beginning. RE typically affects children between 3 and 13 years of age, with a higher incidence of seizures occurring between 6 and 9 years of age; spontaneous remission may occur in adolescence, often before the age of 16 (Berg et al., 2010; Commission on Classification and Terminology of the International League Against Epilepsy, 1989).

Despite the favorable prognosis, studies began to question the “benign” nature of this syndrome when faced with complaints about learning abilities and deficits in neuropsychological tests of children affected by the issue (Beaumanoir, Ballis, Varfis, & Ansari, 1974; Hermann, Jones, Jackson, & Seidenberg, 2012), especially in the areas of language, memory, attention, motor coordination and executive functions (Capelatto et al., 2012; Hwang et al., 2013; Kim, Lee, Chung, Lim & Lee, 2014; Mizira et al., 2012; Neri et al., 2012; Northcott et al., 2005; Overvliet et al., 2013; Smith et al., 2012; Smith, Bajomo, & Pal, 2015; Tovia et al., 2011; Vannest et al., 2013; Verrotti, Filippini, Matricardi, Agostinelli & Gobbi, 2014).

Research on the extension and specificity of the impact of RE on language has been predominant in the literature. This is because this area is considered to be the main weakness in the cognitive functions of this clinical subgroup, which is possibly associated to the harm caused by epilepsy crises on a developing brain (Aquino, Montenegro, Guerreiro & Guerreiro, 2005; Flax et al., 2003; Overvliet et al, 2013; Smith et al., 2012), especially on the areas of the brain related to language: the center of the brain, predominantly in the lower portion of the rolandic and Sylvian areas (Fonseca, Tedrus, Chiodi, Cerqueira & Duran, 2004; Hommet et al., 2001; Staden, Isaaca, Boyd, Brandl &., 1998; Xiao et al., 2016; Zaninotto & Hamad, 2012).

Studies also indicate a correlation between the high incidence of epileptic discharges, the appearance of continuous spikes-waves during sleep and language impairment (Zaninotto & Hamad, 2012). In addition to that, a specific pattern of language dysfunction has been found in these children, showing impairments in reading, writing, spelling, verbal-auditory discrimination and expressive grammar (Staden et al., 1998).

Besides that, deficits in working memory and phonological awareness have also been signaled in children diagnosed with RE (Northcott et al., 2005; Northcott et al., 2007). The literature suggests a strong correlation between these two components and indicates that both are related to language development (Vieira, 2014). Corroborating this perspective, studies demonstrate, for example, a positive connection between the performance in working memory and reading proficiency tasks (Locascio, Mahone, Eason, & Cutting, 2010; Piccolo & Salles,
2013), while delays in phonological awareness are usually related to impacts in writing and reading acquisition (Engel de Abreu et al., 2014; Fricke, Bowyer-Crane, Haley, Hulme, & Snowling, 2013; Krajewski & Schneider, 2009; Salles & Parente, 2008; Savage et al., 2005).

Given the above, this study investigated how the difficulties in language in children diagnosed with RE would be related to alterations in the development of phonological awareness and/or working memory.

**Methods**

**Participants**

The study evaluated forty-two children aged 6 to 13 years old, from both sexes, selected without random assignment. The sample was divided into two groups: one experimental group, and a control one. The experimental group, called G1, was composed of twenty-one children who were diagnosed with RE. The children were recruited at the pediatric neurology service of the Anita Garibaldi Center for Education and Research on Health, at the Children's Neurology Clinic of the Children and Teenagers Ward of the Onofre Lopes University Hospital (HUOL), and at the Specialized Rehabilitation Center (CRE).

The control group, called G2, was structured as a mirror-group of the G1. Thus, it had twenty-one children with typical development recruited from the already existing database from the Laboratory for Research and Extension on Neuropsychology of the UFRN (LAPEN-UFRN). The children in the control group were paired with the ones in the experimental group in relation to sex, age, education and socioeconomic status. It should be noted that this database refers to ruling n. 861.284, approved by the Committee for Ethics in Research of Rio Grande do Norte’s League against Cancer.

As to the characteristics of the groups, the G1 was composed of children who were diagnosed with RE according to the classification of the International League against Epilepsy (ILAE) (Engel, 2001), with average IQ of 60.24 (± 17.50) and enrolled in the regular school system. It should be emphasized that the absence of IQ assessment has been pointed as one of the main weaknesses in studies that associate cognitive difficulties to a diagnosis of RE (Filippini et al., 2016).

The G2 was composed of children with no history of epilepsy, with average IQ of 62.86 (±15.46), and also enrolled in the regular school system. Both groups did not consider children with any other comorbidity or with uncorrected alterations at a visual and/or hearing level, or of motor coordination, or any other impairment that would make executing the proposed tasks impossible.

This study followed the rules of the National Health Council, through Resolution n. 466/2012, which establishes the guidelines and rules for studies involving human beings. Its execution was approved by the Committee for Ethics in Research of the Federal University of Rio Grande do Norte – CEP/UFRN (ruling n. 1178354). All children were authorized to participate in the research by their parents/guardians, who signed a Free and Clarified Consent Term.

**Instruments**

**Raven's Colored Progressive Matrices and Raven's Progressive Matrices – General Scale**

Raven’s Colored Progressive Matrices and Raven’s Progressive Matrices – General Scale were used for IQ evaluation. The goal of these tests is to evaluate general intelligence (g factor), more specifically the eductive ability. In this study, the Colored Progressive Matrices were used to evaluate small children (aged 5 to 11 years old), and Raven's Progressive Matrices – General Scale were used on children aged 12 or higher (Raven, 2003, 2008).
Digit Span Subtest of the Wechsler Intelligence Scale for Children (WISC IV).

The digit span subtest of the WISC is commonly used to evaluate working memory (Abreu & Mattos, 2010; Wechsler, 2013) regarding the phonological loop and the central executive. This study used the forward and backward orders of the subtest.

Corsi block-tapping test

The Corsi block-tapping test was used to assess the visuo-spatial sketchpad and the central executive of the working memory (Abreu & Mattos, 2010).

Phonemic and Semantic Verbal Fluency Test.

The Verbal Fluency Test (Fisk & Sharp, 2004) is considered to be a valid measure for investigating cognitive processes related to both lexical-semantic and executive processes (Elst, Boctel, Breukelen, & Jolles, 2006; Lezak, Howieson, Bigler & Tranel, 2012; Strauss, Sherman, & Spreen, 2006). Thus, this test has been very useful to research the inhibitory component (Capovilla, 2006), the cognitive flexibility (Filippetti, 2011) and working memory (Elst et al., 2006; Troyer, Moscovitch, & Winocur, 1997). In addition to that, the instrument enables the assessment of semantic verbal memory (Elst et al., 2006). This study used limited categories (animals, clothes and fruits) and letters (F, A, M).

Phonological Awareness – Sequential Evaluation Instrument (CONFIAS)

The CONFIAS evaluates phonological awareness in a broad and sequential manner. The Instrument has two parts, evaluation at the level of syllable and phoneme (Moojen et al., 2003).

Procedures

The collection procedures had two stages. The first stage consisted of anamnesis with parents/guardians so as to contextualize the children’s clinical and socio-demographic data. The second stage involved the evaluation of the IQ, working memory and phonological awareness with the application of neuropsychological tests.

The subtests were not applied in a fixed order, so the examiners elaborated a specific work plan for each child, taking into consideration their engagement and motivation in the proposed tasks. The subtests were conducted individually, in a single session (in some cases there was the need for two sessions) of approximately one hour.

Data analysis

The descriptive and inferential statistical data analysis were performed using the statistical software SPSS (Statistical Package for Social Sciences), version 22.0. The study adopted $p<0.05$ as the significance level for all analyses. Non-parametric statistic was used since the sample was small.

Initially, we conducted a descriptive statistical analysis of the performances obtained with the two groups in each of the tests. Later, the performances of the groups were compared using the Mann-Whitney U test. Correlations between variables were tested using Spearman's correlation test. The criterion adopted for interpretation of the correlation coefficients was: 0 to 0.09 (null); 0.10 to 0.39 (low); 0.40 to 0.69 (moderate); 0.70 to 0.99 (high) and 1.00 (very high).

Results

Characteristics of the General Sample

The general sample has forty-two children, twenty-four male (57.1%) and eighteen female (42.9%). The age of the whole sample varied from 6 to 13 years and 11 months, with average
age of 9.14 years (SD = 2.03). In terms of socioeconomic status, 52.4% of the children belonged to classes D and E (family income between R$ 600.00 and R$ 1,350.00) and 47.6% to class C2 (family income between R$ 1,351.00 and R$ 2,250.00) (Table 1).

TABLE 1

<table>
<thead>
<tr>
<th>Characteristics of the sample as a whole</th>
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<tbody>
<tr>
<td>FREQUENCY (N)</td>
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<tr>
<td><strong>SEX</strong></td>
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<tr>
<td>Male</td>
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<td>Female</td>
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<td><strong>AGE</strong></td>
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<td>6</td>
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<td>7</td>
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<tr>
<td>13</td>
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<tr>
<td><strong>INCOME</strong></td>
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<tr>
<td>Classes D and E (from R$ 600.00 to 1,350.00)</td>
</tr>
<tr>
<td>Class C2 (from R$ 1,350.00 to 2,250.00)</td>
</tr>
</tbody>
</table>

Source: own work

Analysis of the performances in the WM and PA tasks between G1 and G2

The comparison of the performances obtained by the groups in the Phonemic Verbal Fluency test (the sum of the words referring to three letters – A, M and F) showed a significant difference between the obtained means (p<0.001), indicating better performance in G2 (mean = 19.52; SD = 4.05) in relation to G1 (mean = 8.48; SD = 4.66). Equally, there was a significant difference between the means of both groups (p<0.001) on the Semantic Verbal Fluency task (sum of the words referring to three categories – Animals, Clothes and Fruits), and the performance of G1 (mean = 21.57; SD = 4.67) was inferior to G2 (mean = 29.10; SD = 7.78).

In terms of general performances obtained the in Digit Span subtest of the WISC IV, there was also significant difference between the means of both groups (p<0.001). Generally, the performance of G2 (mean = 10.38; SD = 2.03) was better than G1 (mean = 5.57; SD = 2.03). Specifically, on the Digit Span Forward, a significant difference was found between the performances of both groups (p<0.001). The performance of G2 (mean = 9.33; SD = 1.46) was significantly better than G1 (mean = 5.86; SD = 1.82); similarly, in the Digit Span Backward task (p<0.001), G2 (mean = 10.76; SD = 1.30) performed better than G1 (mean = 6.19; SD = 1.63).

The analysis of the performances in the Corsi block-tapping task indicates a significant difference between both groups (p<0.001), with G2 performing better (mean = 15.38; SD = 3.02) than G1 (mean = 9.52; SD = 2.06). Specifically, for the Forward Corsi block-tapping task, the performance of G1 (mean = 5.57; SD = 0.98) was inferior to G2 (mean = 7.71; SD = 1.61), evidencing a significant difference (p<0.001). In the Backward Corsi block-tapping task, the results were similar to those obtained in the previous task, with a significant difference (p<0.001) indicating that G2 performed better (mean = 7.76; SD = 1.78) when compared to G1 (mean = 4.05; SD = 1.28).

TABLE 2

<table>
<thead>
<tr>
<th>Mean and standard deviation of the performances between G1 and G2.</th>
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<tbody>
<tr>
<td>G1 (experimental group)</td>
</tr>
<tr>
<td>Phonemic Verbal Fluency</td>
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<tr>
<td>Semantic Verbal Fluency</td>
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<tr>
<td>Digit</td>
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<tr>
<td>Forward Digit span</td>
</tr>
<tr>
<td>Backward Digit span</td>
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<tr>
<td>Confias Blocks</td>
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<tr>
<td>Confias Blocks Forward</td>
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<tr>
<td>Confias Blocks Backward</td>
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<tr>
<td>Confias Backward</td>
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<tr>
<td>Confias Forward</td>
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<tr>
<td>Confias Phonemes</td>
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</tbody>
</table>

Source: own work

The tendency shown above repeated itself in the analysis of the performance of both groups in the Confias syllabic task (p<0.001), indicating inferior performance of G1 (mean = 25.71) in relation to G2 (mean = 34.43). The same was found in the phonemic task of Confias, in which the performance of G1 was worse (mean = 12.71) when compared to G2 (mean = 24). Similarly, the analysis of the performances of both groups on Confias (total score) (p<0.001) found G1 had
an inferior performance (mean = 38.90; SD = 12.46) when compared to G2 (mean = 58.90; SD = 8.20). The summary of the differences in performance between groups are shown on the table below (Table 2).

**Correlational analyses between subtests**

The correlational analyses indicate the presence of positive correlations between the subtests that evaluate working memory and phonological awareness. In this sense, positive and high significant correlations were found between the performances obtained in Confias (total score) and the Phonemic Verbal Fluency task (p < 0.001/R = 0.811); between the performances obtained in Confias (total score) and the Digit Span subtest (p < 0.001/R = 0.748); and between the performances obtained in Confias (total score) and Corsi block-tapping task (p < 0.001/R = 0.814). High, positive correlations were also observed between the performances obtained in Confias (total score) and the Forward Digit Span subtest (p < 0.001/R = 0.741); between the Confias (total score) and the Backward Digit Span subtest (p < 0.001/R = 0.719); between the Confias (total score) and the Forward Corsi block-tapping task (p < 0.001/R = 0.751); and between the Confias (total score) and the Backward Corsi block-tapping task (p < 0.001/R = 0.788). Additionally, the analyses found a moderate positive correlation between the Confias (total score) and the Semantic Verbal Fluency task (p < 0.001/R = 0.694).

It should be noted that high and moderate positive correlations were found between the syllabic task of the Confias and the tasks that evaluated working memory. Similarly, high and moderate positive correlations were observed between the phonemic task and tasks evaluating working memory. The following table shows the correlations identified between the subtests (Table 3).

**TABLE 3**

| Source: own work |

**Correlations between the working memory and phonological awareness subtests**

| Source: own work |

In addition, it is imperative to note that, although not discussed here, this study investigated the performances of G1 based on clinical variables, namely "age of onset of crisis," "number of crises," "age of seizure remission," "exposure time to crises" and "use of AEDs." However, the analysis of variance of one factor (Kruskal-Wallis) revealed the absence of interference of all variables on the performance of children of the G2 in all tests and tasks.

**Discussion**

Despite the good prognosis and remission of seizures during adolescence, a significant number of children with RE has a heterogeneity of cognitive deficits, despite their intelligence levels being classified within the normal range. Commonly, these children have learning difficulties, notably involving language capabilities and executive functions (Filippini et al., 2016; Verrotti et al., 2014).

However, there is no consensus regarding the underlying factors to the presence of cognitive deficits, about which coexist
neurodevelopmental and structural arguments. On the one hand, it is considered that the lesions caused by the manifestations of epileptic seizures are on a developing brain (Overvliet et al., 2013, Smith et al., 2012), suggesting that the reading difficulties presented by children with RE are directly associated with the amount of nocturnal epileptiform activity (Gobbi, Boni, & Filippini, 2006). On the other, there is the occurrence of overlapping cortical areas of language with epileptic activity in the central region, namely the Rolandic and Sylvian regions (Fonseca et al., 2004).

In general, the language deficits in children with RE have been the subject of numerous debates; however, few studies propose to investigate specific abilities connected to the acquisition and development of language skills, which may be associated with this condition. In this scenario, this study presents some considerations about its findings.

Initially, the results show significant differences between the performances of children with RE and healthy children regarding the working memory tasks, suggesting weaknesses in this cognitive domain in the group of children with RE in all investigated components: phonological loop and the visuo-spatial sketchpad.

These findings are consistent with recent studies (Goldberg-Stern et al., 2010; Kárpáti, Donauer, Somogyi & Kónya, 2015; Oliveira, Neri, Capelatto, Guimarães & Guerreiro, 2014) which reported statistically significant differences in working memory tasks, revealing losses in this ability in children diagnosed with RE when compared with healthy children. In addition to that, Goldberg-Stern and colleagues (Goldberg-Stern et al., 2010) found the verbal ability was compromised in the clinical subgroup, through poor performance in tasks considered to be dependent on language and poor performance on the Digit Span task, both forward and backward; they concluded that the working memory is a strong indicator for the acquisition and development of language skills.

It is valid to point out that during the acquisition of language skills the working memory is responsible for children’s ability to analyze the structural properties of language, since this ability is connected to language’s syntactic and phonological components, which gives this executive function a critical part in linguistic processing (Baddeley, 2003). In this direction, working memory deficits compromise the temporary maintenance of linguistic information, causing the formation of simpler, shorter sentences, with less lexical diversity, as well as difficulties in comprehending sentences, acquiring languages and learning new words. Thus, this suggests a bi-directional influence between language and working memory (Montgomery, 2002).

In addition to that, although some studies show that the components of working memory, together, play a fundamental part in language (Alloway et al., 2005), the phonological loop is emphasized regarding the maintenance of phonological information that are necessary for reading. This component is responsible for retaining, for brief periods, the words and sentences while they are being processed, so the longer units of text can be understood (Baddeley, 2012). Significant losses regarding the performance of children with RE in tasks that investigated the phonological loop, a component of the working memory, were also found in this study.

In this context, starting off from the weaknesses in working memory capabilities in children with RE found in this study, as well as considering the fundamental part of the executive component in linguistic processing (Baddeley, 2012; Uehara & Landeira-Fernandez, 2010), our findings suggest that the language compromise in this clinical subgroup seems to be influenced by deficits in working memory.

Regarding phonological awareness, the results point to statistically significant differences between the performances of children with RE and healthy children, both in the syllabic and the phonemic tasks. These findings are consistent with previous studies that suggest that a compromise in this ability in this clinical subgroup impacts the children’s level of reading.
and writing (Amaral et al., 2015; Bedoin et al., 2011; Oliveira, 2011; Oliveira et al., 2014).

It is essential to note that the processes of phonological awareness and reading and writing acquisition are highly complex, reciprocal and mutually strengthened, considering that the initial stages of phonological awareness – that is, awareness of rhymes and syllables – contribute for the level of early reading development and for the development of more complex meta-phonological abilities, like phonemic manipulation and transposition (Grégoire & Piérart, 1997). In this sense, phonological awareness, which begins early (Capovilla & Capovilla, 1998; Gindri, Keske-Soares & Mota, 2007; Krajewski & Schneider, 2009) and gradually (Supple, 1986) is improved and refined by the exposure to words, to the perception of words as being formed by a sequence of identifiable sounds, and to the acquisition of grapheme-to-phoneme conversion, during the formal process of reading and writing acquisition, associated to cognitive development and neural maturation (Bernardino, Freitas, Souza, Maranhe & Bandini, 2006; Freitas, Cardoso & Siquara, 2012).

Thus, just like the working memory, weaknesses in the ability of reflecting about and manipulating the phonological structure of words may be associated to language compromise in children diagnosed with RE, since phonological awareness plays a fundamental part in the development of reading and writing skills, because it helps to establish letter-sound interactions and enables the grasping of the alphabetic principle. Thus, an impairment in this ability is one of the main indicators of difficulties in learning how to write and read (Engel de Abreu et al., 2014; Fostick, Eshcoly, Shtibelman, Nehemia & Levi, 2014; Fricke et al., 2013).

Additionally, in this study, through correlational analyses, positive and high significant correlations were identified between the subtests that evaluate working memory and phonological awareness in the RE clinical subgroup, suggesting that the development of working memory and phonological awareness are interrelated and signaling the components of the working memory as participants in the processes of coding and phoneme storage in the phonological awareness tasks. This finding is consistent with other studies, which indicated a significant connection between working memory and phonological awareness, which are often inextricably linked and dependent on chronological age, maturity and level of education (Cardoso, Silva & Pereira, 2013; Gindri et al., 2007; Northcott et al., 2005; Northcott et al., 2007; Silva & Capellini, 2013).

The lack of consensus among authors is evident, in the scientific field, regarding the interrelation between phonological awareness and working memory. If on the one hand some authors point to the development of working memory and phonological awareness as independent skills, reinforcing the absence of predictive effect of the working memory on phonological awareness (Cárnio, Sá, Jacinto & Soares, 2015), others hold the position in favor of a positive relationship between these skills (Cardoso et al., 2013; Silva & Capellini, 2013), suggesting that the WM and PA skills are developed concurrently and, in the process, influence the literacy process, especially the learning of skills such as reading and writing.

**Conclusions**

In general, the normative results show that the performances of children diagnosed with RE in working memory and phonological awareness tasks are significantly worse when compared to healthy children. These findings corroborate recent studies that question the benign condition of RE, with the goal of identifying specific cognitive deficits in this clinical subgroup, despite normal levels of intelligence.

The fact that significant correlations were found between all the working memory and phonological awareness tests suggests that the development of the skills investigated here are inter-related. Most tasks that evaluate working memory and phonological awareness were found to have high correlations. Therefore, the findings are consistent with other studies that noted the
connection between the two skills and their importance for the acquisition and development of linguistic abilities.

Here, it makes sense to go back to the proposition that the phonological working memory and phonological awareness share phonological processing components. It is assumed that this subsystem of working memory is essential for the execution of tasks that assess the awareness of the sound structure of words. In this sense, understanding the relationship between these skills can help with the early identification of possible deficits in the RE. Certainly, the development of targeted intervention programs for the improvement of phonological awareness and working memory, together with speech therapy, can soften and even prevent alterations in literacy in this clinical group.

Just like any other research, this one also has its limitations and weaknesses, such as the fact that it worked with a wide age range (6-13 years) and a reduced number of participants. However, since Natal is a city with less than 1 million inhabitants, the occurrence of individuals with specific diseases is commonly lower than in other regions of the country, demanding greater flexibility in the selection of the samples and clinical groups.

This study argues that the term benign should be applied with care when talking about RE, since the results show losses in the tasks regarding working memory and phonological awareness, independently from characteristics associated with epileptic discharges and use of medication.

In this sense, despite the spontaneous remission of seizures, RE is an electro-clinical syndrome and, thus, requires longitudinal follow-up, especially regarding the academic performance of these children. Neuropsychological assessments should be done systematically, especially during the phases of reading and writing acquisition. Finally, we hope the results of this study can support different interventions from professionals, allowing for the development of intervention programs turned to minimizing the difficulties this clinical subgroup goes through so as to improve their development and quality of life.

References


**Notes**

* Research Article