PHONOLOGICAL AWARENESS AND WRITING DEVELOPMENT IN DOWN SYNDROME: A LONGITUDINAL CASE STUDY

Consciência fonológica e desenvolvimento da escrita na síndrome de down: um estudo de caso longitudinal

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ABSTRACT

The aims of this study were to: verify performance advances in phonological awareness and writing skills in a child with Down syndrome speaking Brazilian Portuguese; identify which phonological awareness skills developed; analyze the influence of working memory skills on performance in phonological awareness tasks; verify the participant’s performance in phonological working memory tasks and verbal and executive intelligence. The child was seven years old at the onset of the study and was receiving mainstream schooling. Phonological awareness and writing skills were assessed at three time points (T1, T2, T3) during a period of 4 years and 8 months. The Phonological Awareness: Sequential Evaluation Instrument (CONFIAS) was used to assess phonological awareness. In T1 and T2, writing skills were also evaluated using the same instrument. At T3 we used the writing subtest of the Child Brief Neuropsychological Assessment Battery (NEUPSILIN-INF). At T1 phonological working memory was evaluated using the word span task and at T3 we used the non-words subtest of the NEUPSILIN-INF. To evaluate verbal and performance intelligence quotient (T3) we used the Wechsler Abbreviated Scale of Intelligence (WASI). Progress in writing skills and phonological awareness were identified throughout the study. Some skills in syllabic awareness also improved, but tasks that required handling of the phonemic constituents and rhyming awareness remained difficult for the patient. The participant demonstrated good performance in repeating real words. Word span of real words was superior to pseudowords span. The ability to memorize real words seemed to have improved the patient's performance on phonological awareness tasks. The general intelligence quotient was considered borderline. It is believed that in this case, linguistic and cognitive abilities, such as verbal vocabulary, phonological working memory and intellectual capacity, influenced the performance on phonological awareness tests and enabled the child to learn written language.

KEYWORDS: Down Syndrome; Child Language; Learning; Handwriting

INTRODUCTION

Down syndrome is the most common genetic cause of intellectual disability and learning difficulties in the human population, affecting approximately 1 in every 700 to 800 live births. The overall clinical picture can be caused by three types of chromosomal impairment: simple or non-disjunction trisomy, translocation and mosaicism1. Intellectual disability shows considerable variation. Current data found that most individuals with the syndrome present mild to moderate intellectual disabilities2. Cognitive deficit is not homogeneous, with losses particularly evident in some areas, such as expressive language and working memory, more specifically the phonological component3-5. The term

Conflict of interest: non-existent
working memory can be used to describe a system of processing and storing information in the short term that maintains thinking, learning and communication. Activities that require immediate memory can be used to measure phonological working memory, such as the memorization of sequences of digits (digit-span), repetition of pseudowords, or sequences of real words.

In contrast to the difficulties in oral language and phonological working memory, reading single words is a relatively intact ability in individuals with Down syndrome. Most children affected by the syndrome learn to read, although presenting very different levels. General cognitive ability, expressive and receptive linguistic competences and phonological awareness are among the predictors of reading skills in this population.

Metalinguistic ability related to the awareness of the segmental aspect of oral language in different units (words, syllables and phonemes) is called metaphonology or phonological awareness. This ability can be understood as the capacity to analyze and manipulate speech in its phonological components regardless of the message content.

The role of phonological awareness for learning written language in individuals with Down syndrome began to be discussed since the studies of Cossu et al. These authors demonstrated that individuals with Down syndrome showed significantly lower performance than the controls with the same reading skills in phonological awareness tasks. The conclusion of this study is that individuals with Down syndrome can learn to read in the absence of phonological awareness skills. This statement was strongly criticized and prompted researchers of different nationalities to study phonological awareness and its relationship with reading in children with Down syndrome.

Results of subsequent studies showed that subjects with the syndrome have measurable levels of phonological awareness correlated with reading measures, despite the lower performance in relation to the controls with typical development, matched by the ability to read words. These studies also show that tasks involving phonemic segmentation, detection and production of rhymes are very hard for children with Down syndrome. In a study of Brazilian Portuguese speakers with Down syndrome, significant positive correlations were found between phonological awareness and writing in children with Down syndrome. However, it should be noted that, the understanding of the nature of the relationship between phonological awareness and literacy in children with Down syndrome still raises different opinions among scholars.

Unlike Cossu et al., some authors consider that phonological awareness has a predictive role for literacy in children with Down syndrome. Laws and Gunn point out that phonological awareness in children with Down syndrome appears to develop as a result of literacy. Hulme et al. concluded that phonemic awareness seems to have little influence on the development of reading skills. Other researchers argue in favor of the reciprocal relationship, with some phonological awareness skills giving support to written language learning and others, mainly at the phoneme awareness level, developing together with the literacy process.

Some authors argue that only longitudinal studies can clarify the nature of this relationship. Aspects that may predict written language learning in subjects with Down syndrome, as phonological working memory, are also under discussion among researchers and can only be clarified by longitudinal studies.

It should be noted that no longitudinal studies on aspects related to phonological awareness and written language learning in Brazilian children with Down Syndrome were found in literature. Considering the differences presented and the scarcity of longitudinal research, this study intends to verify whether there were advances in phonological awareness and writing skills in a Brazilian Portuguese speaking child with Down syndrome, over a period of 4 years and 8 months. Identifying which phonological awareness skills developed successfully and which continued being difficult to solve was the second objective of this study. The third was to verify the participant’s abilities on tests of phonological working memory and verbal and performance intelligence.

**CASE PRESENTATION**

This case report resulted from an observational field research of the longitudinal type, outlining itself as a cohort study. The participant was evaluated at three times in terms of phonological awareness and writing skills. The first evaluation took place in March 2008, when the child was 7 years of chronological age and attended the first year of elementary school (Time 1 - T1). The second assessment (Time 2 - T2) occurred two years and eight months later, in November 2010. The participant attended the second year of the elementary school in 2009 and 2010. The third year was attended during the year 2011. In November 2012, when the child was 11 years and 8 months old and was attending the fourth year, the participant was reassessed (Time 3 - T3). This study was approved by the Ethics Committee of PUCRS registered under No. 84124.
Those responsible for the child signed the Informed Consent. A Statement of Consent was signed by the participant in T3.

The evaluated child was born full-term, and is a male carrier of trisomy 21. He started walking at 1 year and 5 months. His first words were pronounced during his second year of life. He underwent early stimulation during the first two years, and at 3, started speech therapy, which was not interrupted until the time of the final evaluation of this study. The child has attended a private school in the city of Novo Hamburgo-RS/Brazil since the age of 2 years and has been monitored by an educational psychologist for 5 years. He attended for 3 years and 9 months the Laboratório de Inclusão e Ergonomia do Centro Universitário (Laboratory of Inclusion and Ergonomics of the University) Feevale-RS. The objective of this laboratory was to stimulate literacy in children with Down syndrome and Infantile Chronic Encephalopathy, using information and communication technologies. In the family environment of the participant, reading and writing practices have always been very stimulated. He began psychological treatment suggested by the educational psychologist of the school in 2013.

He is annually assessed by an otolaryngologist and an ophthalmologist. He underwent surgery to remove the pharyngeal and palatine tonsils in 2005. At the last hearing test conducted, the child was diagnosed with normal auditory thresholds, slight hearing loss at an isolated frequency (8000 Hz), and normal middle ear function (Jerger type A curve, presence of acoustic reflexes and normal static compliance). He presents no visual changes or other clinical abnormalities associated with Down syndrome.

The evaluations carried out in this study occurred in three stages, during the participant’s speech therapy. Phonological awareness and writing skills were assessed at T1, T2 and T3 by the speech therapist of this study. It is noteworthy that the child has not undergone specific training related to phonological awareness during the period of this research. The focus of speech therapy during this period was the treatment of myofunctional disorders and phonological alterations present in the speech. For phonological therapy the Modified Cycles Model was used. The phonological working memory of the participant was assessed at T1 through real words span and pseudowords span in T3. Cognitive abilities related to verbal and performance intelligence were tested by a psychologist only in T3. Speech samples were collected at T1 and T3 through the designation of the figures of the instrument used for the phonological awareness assessment. The criterion for evaluation of progress in phonological awareness tasks was the increase of 2 points or more from T1 to T2 or T3 or T2 to T3. For tasks ranging from 0 to 4 points, improvement was considered when the score increased by 50% or more from one evaluation time to another. The instruments utilized for each item at the three evaluation times are listed below.

### Phonological Awareness

**T1,T2 and T3: CONFIAS – Phonological Awareness:** Sequential Assessment Instrument. This instrument is composed of sixteen phonological awareness tasks divided into syllable level and phoneme level. Both the syllable level and the phoneme level tasks show a variation of 0 to 4 points, with the exception of syllable deletion tasks and phoneme deletion that show variations of 0 to 8 and 0 to 6 points, respectively. The possible variation of scores at different levels appears in the table 1 of results. We used the scoring criteria for individuals with Down syndrome proposed by Lavra-Pinto and Lamprechet.

### Writing

T1 and T2: For data collection, we used the same data collection criteria of children’s writing that were evaluated in the validation of CONFIAS. We used a children’s story book and the participant was asked to write, in addition to his name, three words - gato, castelo and esqueleto - (cat, castle, and skeleton) and a sentence – “O fantasma abriu a porta” - (The ghost opened the door). Data were analyzed according to the conception of the appropriation process of the written language by Ferreiro and Teberosky, which allows the classification of participants in general levels referred to as: presyllabic writing hypothesis, syllabic hypothesis, syllabic-alphabetic hypothesis, and alphabetic hypothesis.

**T3:** writing subtest of the Child Brief Neuropsychological Assessment Battery (NEUPSILIN-INF), composed of 14 real words and 5 pseudowords (total score: 19). Data were analyzed quantitatively (score on the subtest) and qualitatively according to the Theory of Psychogenesis of Writing.

### Phonological Working Memory

**T1:** Real words span test - instrument developed by Lavra-Pinto and Lamprechel to assess the real word span of children with Down syndrome. This instrument is composed of 14 sequences of disyllabic and trisyllabic words not semantically related, containing a total of 46 words. The sequences were presented verbally and the child was asked to repeat the words from memory. The child had two
opportunities to respond and, for scoring purposes, the second response was the one counted. The total score was the number of words repeated correctly during the evaluation. The number of correctly repeated sequences also provided a measure of phonological working memory. The maximum number of disyllabic and trisyllabic words memorized corresponds to the span of the real words with two and three syllables.

**T3:** Pseudowords span of the Child Brief Neuropsychological Assessment Battery (NEUPSILIN-INF). The sequences of pseudowords were presented orally and the participant was asked to repeat the pseudowords in the same order they were presented. In this test the sequences were not repeated. The number of stimuli was gradually increased from 1 to 4 dissyllabic items. The total score of this subtest is 20 points. The greatest correctly repeated sequence corresponds to the span of pseudowords. The authors of NEUPSILIN-INF authorized the use of the writing subtests and pseudowords span for this study.

**Verbal and Performance Intelligence:**

**T3:** WASI- Wechsler Abbreviated Scale of Intelligence in its standardized and validated version for the Brazilian reality to access verbal and performance intelligence. The scale is composed of four subtests, two Verbal (Vocabulary and Similarities) and two Performance (Block Design and Matrix Reasoning). The WASI provides three composit measures: Total Intelligence Quotient, Performance Quotient, and Verbal Quotient. The score of the Total Intelligence Quotient (TIQ) accesses intellectual functioning and is subdivided into Verbal IQ and Performance IQ. The Verbal IQ evaluates the verbal processes and knowledge acquired and is similar to the concept of crystallized intelligence. The Performance IQ measures perceptual organization, ability to manipulate visual stimuli rapidly, and other non-verbal processes, and is closer to the concept of fluid intelligence.

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**RESULTS**

First we will present the quantitative results of the phonological awareness assessment, along with a qualitative analysis of the writing skills and the responses of the participant on tasks of CONFIAS at the three points of evaluation. Then we will describe the other results related to phonological working memory, verbal and performance intelligence.

In Table 1 it is possible to observe the progress achieved by the child in the assessment of phonological awareness. This table shows the raw scores obtained by the participant on the tasks of CONFIAS and the corresponding percentage (total score at the syllable level - LS, scores on each task of the LS, the total score on the phoneme level - PL, scores on different tasks of PL and total score of the assessment) at T1, T2 and T3. The possible range of scores established by the instrument is included in the table 1. Table 2 shows the different writing levels achieved and the chronological age of the child at the three points of evaluation.

The results shown in Tables 1 and 2 allow us to affirm, when responding to the first objective of this study, that the participant showed an increase in phonological awareness and writing skills during the study period. At the first assessment, the participant could not write; however, after 4 years and 8 months, he had reached the level of alphabetic writing. Figure 1 illustrates the performance on tasks of syllabic and phonemic level of CONFIAS at T1, T2, and T3, moments in which the participant had different writing hypothesis. These results showed that the scores on phonological awareness tasks increased as the participant’s level of writing increased.

On the writing subtest of NEUPSILIN-INF, administered only at T3, the total score achieved was 9 points. We observed interference from phonological processes of speech, such as the replacement of voiced and unvoiced sounds. Moreover, speech transcription into words with spelling rules was observed. When he was asked to write the word *texto* (text in Portuguese) he wrote “testo”. The child wrote 2 of 5 pseudowords correctly. It can be inferred that most of the errors occurred because of his speech difficulties. However, despite the
The synthesis and syllable segmentation tasks it was determined that the child had performed well since the first evaluation. On PL, advances were observed in all tasks except that of phoneme transposition, on which the score remained the same compared to previous testings.

It can be said that the task of identifying the final phoneme, phoneme synthesis and segmentation emerged as a result of literacy, since the participant did not obtain a score on T1, when he was found to be at presyllabic writing level. On T3, with the alphabetic writing level, the participant had 100% accuracy on the task of identifying the final phoneme and 50% on the phoneme segmentation and synthesis.

The observation of the raw scores and the percentage of success on each task at different levels of phonological awareness of CONFIAS (Table 1) also allowed us to respond to the second objective of this study. As previously mentioned, it was considered that the participant progressed on the task when an increase of two or more points between T1 to T2 or T3 or T2 to T3 occurred. The tasks of the SL that showed this increase were: identification of medial syllable, syllable deletion, and syllable transposition. It can be argued that these tasks developed with the process of appropriation of written language by the participant. On the synthesis and syllable segmentation tasks it was determined that the child had performed well since the first evaluation. On PL, advances were observed in all tasks except that of phoneme transposition, on which the score remained the same compared to previous testings.

It can be said that the task of identifying the final phoneme, phoneme synthesis and segmentation emerged as a result of literacy, since the participant did not obtain a score on T1, when he was found to be at presyllabic writing level. On T3, with the alphabetic writing level, the participant had 100% accuracy on the task of identifying the final phoneme and 50% on the phoneme segmentation and synthesis.

Table 1 - Participant’s performance on the phonological awareness assessment

<table>
<thead>
<tr>
<th>Scores</th>
<th>Possible variation</th>
<th>T1 RS (%)</th>
<th>T2 RS (%)</th>
<th>T3 RS (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>SL - Total</td>
<td>0-40</td>
<td>16</td>
<td>27</td>
<td>32</td>
</tr>
<tr>
<td>S1 - Synthesis</td>
<td>0-4</td>
<td>4 (100)</td>
<td>3 (75)</td>
<td>4 (100)</td>
</tr>
<tr>
<td>S2 - Segmentation</td>
<td>0-4</td>
<td>3 (75)</td>
<td>4 (100)</td>
<td>4 (100)</td>
</tr>
<tr>
<td>S3 - Identification of the initial syllable</td>
<td>0-4</td>
<td>2 (50)</td>
<td>2 (50)</td>
<td>3 (75)</td>
</tr>
<tr>
<td>S4 – Identification of rhyme</td>
<td>0-4</td>
<td>1 (25)</td>
<td>2 (50)</td>
<td>2 (50)</td>
</tr>
<tr>
<td>S5 - Production of a word with the given syllable</td>
<td>0-4</td>
<td>2 (50)</td>
<td>3 (75)</td>
<td>3 (75)</td>
</tr>
<tr>
<td>S6 - Identification of the medial syllable</td>
<td>0-4</td>
<td>0 (0)</td>
<td>3 (75)</td>
<td>4 (100)</td>
</tr>
<tr>
<td>S7 – Production of rhyme</td>
<td>0-4</td>
<td>0 (0)</td>
<td>1 (25)</td>
<td>1 (25)</td>
</tr>
<tr>
<td>S8 - Deletion</td>
<td>0-8</td>
<td>3 (37,5)</td>
<td>6 (75)</td>
<td>8 (100)</td>
</tr>
<tr>
<td>S9 - Transposition</td>
<td>0-4</td>
<td>1 (25)</td>
<td>3 (75)</td>
<td>3 (75)</td>
</tr>
<tr>
<td>PL- Total</td>
<td>0-30</td>
<td>3</td>
<td>14</td>
<td>19</td>
</tr>
<tr>
<td>F1 – Production of a word that starts with a given sound</td>
<td>0-4</td>
<td>1 (25)</td>
<td>4 (100)</td>
<td>4 (100)</td>
</tr>
<tr>
<td>F2 - Identification of the initial phoneme</td>
<td>0-4</td>
<td>1 (25)</td>
<td>3 (75)</td>
<td>3 (75)</td>
</tr>
<tr>
<td>F3 – Identification of the final phoneme</td>
<td>0-4</td>
<td>0 (0)</td>
<td>4 (100)</td>
<td>4 (100)</td>
</tr>
<tr>
<td>F4- Deletion</td>
<td>0-6</td>
<td>1 (16,6)</td>
<td>2 (33,3)</td>
<td>4 (100)</td>
</tr>
<tr>
<td>F5- Synthesis</td>
<td>0-4</td>
<td>0 (0)</td>
<td>1 (25)</td>
<td>2 (50)</td>
</tr>
<tr>
<td>F6- Segmentation</td>
<td>0-4</td>
<td>0 (0)</td>
<td>0 (0)</td>
<td>2 (50)</td>
</tr>
<tr>
<td>F7- Transposition</td>
<td>0-4</td>
<td>0 (0)</td>
<td>0 (0)</td>
<td>0 (0)</td>
</tr>
<tr>
<td>Total Score</td>
<td>0-70</td>
<td>19</td>
<td>41</td>
<td>51</td>
</tr>
</tbody>
</table>

SL: Syllable level, PL: Phoneme level, T1: time 1, T2: time 2, T3: time 3
RS: Raw score (% - percentage of correct answers on the task)

Table 2 – Writing Assessment

<table>
<thead>
<tr>
<th>Assessment Period</th>
<th>Writing Hypothesis</th>
<th>Chronological Age (years,months)</th>
</tr>
</thead>
<tbody>
<tr>
<td>T1</td>
<td>Presyllabic</td>
<td>7,0</td>
</tr>
<tr>
<td>T2</td>
<td>Syllabic-Alphabetic</td>
<td>9,8</td>
</tr>
<tr>
<td>T3</td>
<td>Alphabetic</td>
<td>11,8</td>
</tr>
</tbody>
</table>

T1: time 1, T2: time 2, T3: time 3
Among the tasks of the SL, the child did not progress, according to the criteria established in this study, on rhyme identification and production tasks. On the task of phoneme transposition, the participant earned no score on any of the three assessment moments. These findings suggest that awareness of rhyme or the ability to perform phoneme transposition may not be necessary for the appropriation of the alphabetic system in children with Down syndrome, a hypothesis which deserves to be more deeply and specifically investigated.

During the evaluation of phonological awareness it was observed that phonological processes in the child’s speech interfered with the responses provided at the three assessment moments. At T1, the main phonological processes found were: replacement or deletion of liquids - /l/ /λ/ /r/ /ɾ/ - and replacement of voiced and unvoiced phonemes. As an example of this interference, we can cite the response given on the task of producing a word with the target sound [ʃ]. His answer was the word “girafa” (giraffe in Portuguese), pronounced [ʃilafa], observed in T1. The difficulty with voiced and unvoiced sounds, mainly involving fricative phonemes, remained at the last evaluation. Within the liquids, the participant continued to present the replacement of the non-lateral liquid /ɾ/ by the lateral /l/. On T3, interferences related to the knowledge of writing were observed, involving, more specifically, the name of the letters. For example, when the child was asked to say what the initial syllable of target words were, he said the name of the first two letters. On the task of rhyme production, he responded with words that started with the same phoneme of the target word or with another semantically related word, instead of with a word that rhymed with the target. As an example, for the target word balão (balloon in Portuguese), he said “bola” (ball in Portuguese), for the target word café (coffee in Portuguese), his response was “maçã” (apple in Portuguese).

The phonological working memory was assessed at T1, through repetition of real words. The participant’s performance on the real words span task is shown in table 3. The ability to memorize up to three real words may have positively influenced the performance on phonological awareness tasks measured by CONFIAS. For solving the tasks of this instrument it is necessary to compare one target word with a group of up to 3 words, which must be memorized. The total score obtained on the pseudowords subtest of NEUPSILIN-Inf, applied at T3 was 8 points, which is equivalent to 40% accuracy. It was observed that the child can repeat a maximum of two disyllabic pseudowords (pseudowords span). In a few moments, he presented lexicalization of terms: the pseudowords jolha and prina were repeated as “Júlia” and “prima” (words in Portuguese).

Through the application of the Wechsler Abbreviated Scale of Intelligence (WASI) it was found that the IQ of the participant can be classified as borderline level (IQ between 70 and 79) and he performed better on Vocabulary and Similarities subtests. The Vocabulary Subtest assesses the ability to define objects and concepts verbally and
the development of expressive language. The Similarities Subtest examines the ability to identify similarities between objects or concepts, and is an indicator of the level of verbal comprehension. Thus, it is possible that the language skills and intellectual capacity, as defined by WASI, have influenced the progress reported in phonological awareness and writing skills. The numerical results of the WASI subtests - Verbal IQ, Performance IQ and total IQ - appear in Table 4.

**DISCUSSION**

Cross-sectional studies on the learning of reading or writing are limited in the ability to elucidate the development of literacy. Only longitudinal studies allow the investigation of the development of written language learning and have greater potential for the exploration of causal relations.

In the present study, the child showed measurable levels of phonological awareness at the three assessment times, confirming previous research results. In a longitudinal study by Hulme et al., as well as on this case report, progress in terms of written language learning and phonological awareness in children with Down syndrome was observed. The authors found among the participants with the syndrome a significant positive relationship between phonemic awareness in the second evaluation and the abilities to read words at the end of the study.

In most of the studies conducted, although there is significant positive correlation between phonological awareness and reading and writing measures, the performance of children with the syndrome is lower than that of participants with typical development at the same reading level. In this study it was observed that at T1, the participant’s performance on tasks of CONFIAS was lower than what is expected for children with typical development and same writing level. However, in T2 and T3, his performance on phonological awareness tasks was quite close to what is expected for typically developing children at the same writing level. The expected scores for typically developing children according to the writing level are provided by CONFIAS.

<table>
<thead>
<tr>
<th>Scores</th>
<th>Possible Variation</th>
<th>Score</th>
</tr>
</thead>
<tbody>
<tr>
<td>PWM (total score)</td>
<td>0-46</td>
<td>35</td>
</tr>
<tr>
<td>Number of correctly repeated sequences</td>
<td>0-14</td>
<td>7</td>
</tr>
<tr>
<td>Dissyllabic words span</td>
<td>0-5</td>
<td>3</td>
</tr>
<tr>
<td>Trisyllabic words span</td>
<td>0-3</td>
<td>3</td>
</tr>
</tbody>
</table>

PWM: Phonological Working Memory

<table>
<thead>
<tr>
<th>Subtest</th>
<th>IQ</th>
</tr>
</thead>
<tbody>
<tr>
<td>Vocabulary</td>
<td>20</td>
</tr>
<tr>
<td>Similarities</td>
<td>16</td>
</tr>
<tr>
<td>Cubes</td>
<td>6</td>
</tr>
<tr>
<td>Matrix Reasoning</td>
<td>15</td>
</tr>
<tr>
<td>Verbal IQ</td>
<td>76</td>
</tr>
<tr>
<td>Performance IQ</td>
<td>74</td>
</tr>
<tr>
<td>Total IQ</td>
<td>72</td>
</tr>
</tbody>
</table>

IQ: Intelligence Quotient

In relation to the predictors of literacy, Kay-Raining Bird et al. postulated that phonemic awareness in this initial phase of the study predicts the development of reading skills in children with Down syndrome, which were evaluated at three moments over 4.5 years. Cuples and Iacono evaluated individuals with Down syndrome at two
times during 8,9 months and found poor performance on the task of phonemic segmentation, however, they concluded that this ability is a strong predictor of reading development in children with Down syndrome. It should be noted that, in the initial evaluation of both studies, participants were already able to read words despite varied levels, making it difficult to conclude whether the phonemic awareness skills are predictors or consequence of contact with formal written language instruction.

Hulme et al. \(^{21}\) assessed the development of metaphonological and reading skills in 49 children with Down syndrome and 61 with typical development, paired by their level of reading words. The participants were evaluated at three times over 2 years and possible predictors of reading capacity were investigated. The authors concluded that although there was correlation between phonemic awareness and reading skills in children with Down syndrome, measures of vocabulary, instead of phonemic awareness, are more robust predictors of reading capability. Among children with typical development, the predicted relationship between phonological awareness and reading was stronger. These findings were corroborated by another recent study \(^{27}\). Roch and Jarrold \(^{28}\), during their longitudinal investigation of the abilities to read words and pseudowords in children with Down syndrome, also found no predictive relationship between decoding skills and awareness at the phoneme level.

Lavra-Pinto and Lamprecht \(^{12}\) found that the performance of literate children with Down syndrome in phonological awareness tasks is significantly higher than that of illiterate individuals with the syndrome. The authors suggested that phonemic awareness in children with Down syndrome is a result of written language learning. The participant’s performance in this case report is consistent with this assumption. Most phonemic awareness skills in children in this study came with learning the alphabetic principle. At T1, the participant obtained a total score of 3 points on phoneme level tasks, which is equivalent to 3% accuracy. At T3, when he had reached the alphabetic writing hypothesis, the percentage of correct answers was 63.3% (19 points). On the phonemic segmentation task the participant only registered score at T3, which seems to indicate that this ability is a consequence of literacy.

In relation to syllabic phonological awareness, the development of metaphonological skills at the syllable level was observed during the process of appropriation of writing by the participant. The tasks of identifying the initial syllable, syllable deletion and transposition showed progress in accordance with the criteria established in this study. However, the ability to identify medial syllabic segments seems to have emerged only with the learning of written language. No improvements were observed on the task of identification and production of rhymes over time. It is also noteworthy that the participant showed great performance on the tasks of synthesis and syllable segmentation at the three assessment times. This data is consistent with results of previous studies which assessed phonological awareness at the syllable level \(^{8,9}\). In the study by Lavra-Pinto and Lamprecht \(^{12}\) no significant difference among individuals with Down syndrome was found with alphabetic and presyllabic writing hypothesis with regard to syllabic synthesis and segmentation skills. Other authors concluded that some skills involved in syllabic awareness tasks can be well developed in phases prior to literacy \(^{9}\).

In most published studies on phonological awareness in children with Down syndrome, as in this case report, difficulties with tasks involving identification and production of rhymes have been observed. In the conducted study \(^{12}\), no significant differences were found in the production of rhymes among literate or illiterate children with Down syndrome. Individuals with the syndrome have shown significantly lower performance on tasks that involve rhyme awareness \(^{19,20}\) when compared to controls with typical development and at the same reading level. It has been suggested that, when compared to children with typical development, the phonological awareness development of individuals with Down syndrome follows a different path \(^{19}\).

Results of this study show that, as pointed out by previous studies, some syllabic awareness skills improve with written language learning, while most phonemic awareness skills arise as a result of literacy in individuals with Down syndrome \(^{7,9,12,17,20}\). It was observed that the tasks of phonemic synthesis, segmentation and transposition were very hard for the child evaluated in this study at the three assessment times. It can be said that the skills involved in these tasks, as well as in the production of rhymes, do not seem to play an important role for individuals with Down syndrome in terms of learning the written language \(^{12,21,27}\).

The impairment in the phonological component of working memory found in children with the syndrome \(^{29}\) may be related to poor performance on tasks involving phonemic synthesis, segmentation, and manipulation. The deficit in phonological working memory may also explain the low performance in the span test of pseudowords used in the last assessment. Speech changes may also have hampered the repetition of pseudowords.

Laws and Gunn \(^{10}\) longitudinally analyzed factors that could differentiate children with Down syndrome who became readers from those who failed to learn...
how to read. The participants were analyzed at two times over a period of 5 years. The performance on the test of repeated pseudowords did not increase significantly throughout the study and was not considered a predictor of reading abilities achieved at the end of the research. The principal predictors were: non-verbal skills, knowledge of the name of letters, and performance on tasks of receptive vocabulary and grammatical understanding. The results of Hulme et al. confirm the findings regarding the predictive relationship between measures of vocabulary, non-verbal intelligence, and knowledge of the names of letters.

Despite the poor performance on the span of pseudowords test, the participant of this investigation has achieved good results in the repetition of real words test administered at T1. He displayed the ability to memorize up to 3 disyllabic or trisyllabic words, which may explain the good performance on CONFIAS tasks. This instrument requires, on identification tasks, the memorization of sequences of three disyllabic or trisyllabic words. These results are consistent with findings from a previous study. In studies evaluating the digit-span, the mean score of children with Down syndrome is 3 or 4 digits.

The total score of the participant on the real words span task at T1 (35 points), when he was not literate yet, is higher than that found among illiterate individuals in the study of Lavra-Pinto and Lamprecht. Many studies show significant positive correlation between measures of reading and phonological working memory in children with Down syndrome. However, the understanding of phonological working memory as a predictor of literacy in individuals with Down syndrome requires further investigation.

It is believed that the good working memory capacity, as measured by the real words span task in the first assessment, verbal language skills and intellectual capacity, evaluated as borderline by the intelligence scale used, enabled the evaluated child to learn the written language. The child in this study, despite speech interferences, demonstrated the capacity to write pseudowords, which shows the ability to make connections between phonemes and graphemes. A similar case was reported by Groen et al. Snowling et al. reported that the verbal mental age seems to be a strong predictor of individual differences related to the decoding capability.

It is also noteworthy that extrinsic factors, such as supportive therapies to which the child was submitted and the influence of family and school environments may have functioned as critical factors for literacy and the advances obtained in phonological awareness.

CONCLUSIONS

Based on the results presented, it is possible to conclude that the child evaluated showed improvement in writing and phonological awareness skills throughout the three times of this study. Some syllabic awareness skills improved with literacy, but most of the skills used on phonemic awareness tasks emerged as a result of the appropriation of the alphabetic system. Some skills that require handling phoneme constituents and rhyme awareness remained difficult to resolve even after literacy. The participant of this study performed well on tests of phonological working memory and verbal intelligence. The capacity for memorization of up to three real words may have helped in solving phonological awareness tasks of the instrument used. It is believed that in this case, cognitive and linguistic skills, such as working memory and large verbal vocabulary played an important role in the learning of written language. However, further longitudinal studies with a larger number of participants, including comparisons with control groups, should be performed in order to investigate the nature of the relationships between the variables studied and the predictors of reading and writing skills. In particular, we suggest the investigation of the predictive role of general cognitive skills, such as phonological working memory, as well as verbal and performance intelligence, on literacy in individuals with Down syndrome.
RESUMO

Os objetivos deste estudo foram: verificar a existência de avanços em habilidades de consciência fonológica e escrita em uma criança com síndrome de Down, falante do português brasileiro, em um intervalo de tempo de quatro anos e oito meses; identificar quais habilidades de consciência fonológica desenvolveram-se e quais continuaram sendo de difícil resolução; verificar o desempenho do participante em testes de memória de trabalho fonológica e de inteligência verbal e de execução. A criança frequenta escola regular e tinha 7 anos de idade cronológica no início do estudo. A consciência fonológica e a escrita foram avaliadas em três momentos (T1, T2, T3) em um período de 4 anos e 8 meses. Para a avaliação da consciência fonológica foi utilizado o teste Consciência fonológica: instrumento de avaliação sequencial (CONFIAS). No T1 e T2, a escrita foi avaliada de acordo com critérios do mesmo instrumento; no T3, foi utilizado o subteste de escrita do Instrumento de Avaliação Neuropsicológica Breve Infantil (NEUPSILIN-INF). No T1, a memória de trabalho fonológica foi avaliada por meio do span de palavras e, no T3, foi utilizado o subteste de pseudopalavras do NEUPSILIN-INF. Para avaliação da inteligência verbal e de execução (T3), utilizou-se a Escala de Inteligência Wechsler Abreviada (WASI). Foram verificados progressos em habilidades de escrita e consciência fonológica ao longo do estudo. Algumas habilidades de consciência silábica aprimoraram-se, já tarefas que exigem manipulação de constituintes fonêmicos e consciência de rima continuaram de difícil acesso. O participante apresentou um bom desempenho no teste de repetição de palavras reais. O span de palavras reais foi superior ao span de pseudopalavras. O quociente de inteligência geral foi considerado limítrofe. Acredita-se que, neste caso, habilidades linguísticas e cognitivas, como o vocabulário verbal, a memória de trabalho fonológica e a capacidade intelectual, influenciaram o desempenho no teste de consciência fonológica e capacitaram a criança para o aprendizado da língua escrita.

DESCRITORES: Síndrome de Down; Linguagem Infantil; Aprendizagem; Escrita Manual

■ REFERENCES