Contents lists available at ScienceDirect



Journal of Communication Disorders

journal homepage: www.elsevier.com/locate/jcomdis



Communicatio Disorders

Identification of the factors associated with the severity of the speech production problems in children with comorbid speech sound disorder and developmental language disorder

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ARTICLE INFO

Keywords: Phonological awareness Phonological processes Auditory perception Speech sound disorders Working memory Oral motor skills

ABSTRACT

It has been suggested that factors such as auditory perception, oral motor skills, phonological awareness, and working memory are all associated with speech production problems in children with speech sound disorder (SSD) and developmental language disorder (DLD). However, it remains unclear whether the severity of the speech production problems in these children can be explained by an interaction among the aforementioned factors. The aim of this study was to determine which of these four factors best explain the severity of the speech production problems in children with SSD and DLD and whether an interaction between factors occurs. Forty-one children with SSD and DLD between 5 and 5;11 years old were selected. The number of phonological process errors was used as a measure of the severity of the speech production problems. The association between the number of phonological process errors and performance in auditory perception, oral motor skills, phonological awareness, and working memory along with the severity of the DLD was explored using univariate and multivariate regression models (with and without an interaction term). The results showed that the number of phonological process errors was largely explained by working memory and phonological awareness. An interaction between these two factors was also found. This means that working memory and phonological awareness interact to have an effect on the number of phonological process errors that is more than the sum of their parts.

In addition, the severity of the DLD was significantly associated with the number of phonological process errors. These findings suggest that phonological awareness and working memory should be considered when assessing and treating children with comorbid SSD and DLD.

1. Introduction

Speech sound disorder (SSD) is the most common communication disorder observed by speech-language pathologists (SLPs) in the pediatric population (Broomfield & Dodd, 2004). SSD is defined as persistent phoneme deletion and/or distortion errors in speech production as compared with other children of the same chronological age (Shriberg, Austin, Lewis, McSweeny, & Wilson, 1997). The prevalence for this disorder has been estimated to be between 3.4 and 3.8 % (Eadie et al., 2015; Shriberg, Tomblin, & McSweeny,

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https://doi.org/10.1016/j.jcomdis.2020.106054

Received 4 June 2019; Received in revised form 11 August 2020; Accepted 28 August 2020 Available online 30 September 2020 0021-9924/© 2020 Elsevier Inc. All rights reserved. 1999). SSD may occur in children with or without a developmental language disorder (DLD) (Lewis et al., 2015). It has been estimated that between 50 and 75 % of children with SSD also present with DLD (Shriberg & Kwiatkowski, 1994).

SSD is mainly characterized by the presence of speech production problems such as phonological processes (PPs) that continue beyond the age at which children typically produce them (Abou-Elsaad, Afsah, & Rabea, 2019; Bauman-Waengler, 2012; Dodd, 2014). According to the theory of natural phonology (Ingram, 1983), PPs are structured mechanisms used by children to simplify speech. According to Bauman-Waengler (2012), PPs are systematic changes applied to sequences of speech sounds. In typically developing children, PPs begin to rapidly decline very early in life (James, 2001) and disappear by around age 6, when children begin to produce adult-like speech (Lousada, Mendes, Valente, & Hall, 2012). In typically developing Spanish-speaking children, some PPs, such as reduction of vowel groups or addition of phonemes and syllables, tend to disappear completely by the age of 4 (Coloma, Pavez, Maggiolo, & Peñaloza, 2010). However, in children with SSD, PPs persist beyond this age (Lousada et al., 2013). Therefore, the frequency of occurrence of PPs produced by a child can be used as an indicator of the severity of their speech production problems (Edwards, 1992). Note that some clinical tools are available to determine the number of PPs for a closed set of words in both English (Dodd, Zhu, Crosbie, Holm, & Ozanne, 2002) and Spanish (Pavez, Maggiolo, & Coloma, 2008).

Several researchers have been interested in determining the factors associated with the characteristics and severity of speech production problems in children with only SSD and in children with SSD and DLD. Such factors include auditory perception, oral motor skills, phonological awareness, and working memory. Below, we discuss each of these factors and their association with the severity of SSD with and without a concomitant DLD. Then, we discuss the impact of a DLD on speech production in children with SSD. Finally, we describe the research question that has motivated this study along with the aims of this research.

The association between SSD and auditory perception has been investigated by a number of researchers. For example, Tallal, Stark, and Mellits (1985) reported that children with comorbid speech and language disorders performed worse on tasks of auditory discrimination of temporal and spatial cues for nonverbal stimuli than children with typical development. Similarly, Murphy, Pagan-Neves, Wertzner, and Schochat (2014) reported that children with SSD have greater difficulties in tasks of auditory temporal processing for nonverbal stimuli than children of a similar age without SSD. Edwards, Fox, and Rogers (2002) reported that children with SSD performed worse in tasks of auditory discrimination of words in comparison to children with typical development. However, different findings were reported by Preston, Irwin, and Turcios (2015), who found similar results for a phoneme perception task between children with SSD and typically developing children.

With respect to the link between motor oral skills and SSD, Yu et al. (2018) recently showed that an 8-week motor speech therapy significantly improved both oral motor and articulation skills in children with SSD. In addition, in the same sample of participants, magnetoencephalography identified significant post-therapy changes in brain regions associated with oral motor control and speech production (Yu et al., 2018). However, different results were reported by Guisti-Braislin and Cascella (2005), who did not find significant differences in speech production between pre- and post-oral motor training on nonverbal oral movements in children with SSD. Note that systematic reviews on this topic (e.g., Kent, 2015; Ruscello, 2008) have shown that there is not enough evidence supporting the efficacy of training programs based on nonverbal oral movements in reducing speech production problems in children with SSD. Despite the limited evidence in this respect, Lof and Watson (2008) reported that nonspeech oral motor exercises were frequently used by SLPs to treat children with speech production problems.

It has also been suggested that phonological awareness is another aspect that is associated with speech production problems in children with SSD alone or with concomitant DLD (Gillon, 2000; Webster & Plante, 1992). For example, Mann and Foy (2007) found that poor production of consonants was associated with deficient phonological awareness in rhyme tasks in children with comorbid SSD and DLD. The same authors found that children who did not have difficulties in producing consonants presented with better development of phonological awareness than children with consonant production problems. Preston and Edwards (2010) found a significant association between poor phonological awareness and the production of atypical PPs in children with SSD that was not observed in children with typical language development.

Torrington Eaton and Bernstein Ratner (2016) suggested that working memory is the basis for progressing from immature productions to adult-like speech. Therefore, it may be suggested that difficulties with this cognitive system may slow down the reduction of PPs in children's speech. Nijland (2009) found that children with SSD were more likely to obtain poor results on a working memory task (an auditory sequential memory task) than typically developing children. Also, Lewis et al. (2011) found that children with SSD were less accurate in repeating pseudowords (i.e., a working memory task) than typically developing children. Similarly, Lee and Ha (2018) and Peterson, McGrath, Smith, and Pennington (2007) found that children with SSD had more problems with working memory tasks (e.g., repetition of pseudowords, numbers in a backward fashion) than typically developing children.

Finally, note that comorbidity of SSD and DLD should be taken into account when investigating the severity of speech production problems (i.e., PPs) in children with SSD. First, studies investigating children with SSD with and without concomitant DLD have shown that children with comorbid speech and language disorders exhibited more severe speech production problems than children diagnosed with SSD only (Nathan, Stackhouse, Goulandris, & Snowling, 2004; Sices, Taylor, Freebairn, Hansen, & Lewis, 2007). For example, Macrae and Tyler (2014) reported that children diagnosed with concomitant SSD and DLD produced more omissions and fewer substitutions in their utterances than children diagnosed with SSD only. Second, it has been observed that children with DLD exhibit poorer performance in phonological awareness and working memory tasks than typically developing children (Archibald & Gathercole, 2006; Claessen & Leitão, 2012; Dollaghan & Campbell, 1998). Thus, the effect of phonological awareness and working memory on SSD (e.g., number of PPs) may be explained, at least in part, by the potential confound of DLD.

In summary, auditory perception, oral motor skills, phonological awareness, and working memory have been associated with speech production problems in children with SSD. In addition, children with comorbid speech and language disorders seem to have more severe problems in speech production than children diagnosed with SSD only. However, most of the studies discussed above have

investigated each of the four factors alone without evaluating all four factors in the same sample of children. Wren, Harding, Goldbart, and Roulstone (2018) conducted a systematic review about intervention strategies for SSD in preschool children. The authors concluded that interventions that combine different aspects/abilities, including the ones mentioned above, show the strongest evidence. This, therefore, suggests that speech production problems in children with SSD are likely to be explained by more than one factor. A research gap remains regarding whether the severity of the speech production problems in children with SSD and DLD may be explained by interactions between the aforementioned factors.

Considering that a high prevalence of children with SSD also present with DLD (50 %–75 %; Shriberg & Kwiatkowski, 1994) and that the latter seems to influence SSD, in this study we were particularly interested in the population of children diagnosed with comorbid SSD and DLD. The aims of this study were to determine (1) which of the four factors mentioned above best explain the severity of the speech production problems in children with SSD and DLD and (2) whether the severity of the speech production problems in children with SSD and DLD and (2) whether the severity of the speech production problems in children with SSD and DLD may be explained by interactions among these four factors. Considering that DLD has been shown to be associated with both speech production problems in children with SSD (dependent variable in this study) and poor performance in phonological awareness and working memory tasks (independent variables in this study), the severity of the DLD was incorporated as a confounding variable in this study.

2. Methods and procedures

2.1. Participants

A cross-sectional study was performed in 2016 in language schools within a single neighborhood in the Metropolitan Region of Santiago, Chile. Two schools agreed to participate in the study. A non-probabilistic sample of 41 children was obtained. The participants were kindergarten students, with a mean age of 67 months (range 60–71 months), and 19.5 % were female. The parents of all participants signed an informed consent form before the children were enrolled in the study. The study protocol was approved by the Ethics Committee of the Department of Speech Language and Hearing Sciences, Faculty of Medicine, Universidad de Chile.

All children had a previous diagnosis of comorbid SSD and DLD. Such diagnoses were made by SLPs according to the diagnostic criteria recommended by the Chilean Ministry of Education (Ministerio de Educación de Chile, 2010). At the time of the study, all the children were receiving rehab sessions once a week in groups of three children at the language school. Rehab sessions were carried out by a SLP along with a special educator specialized in language disorders, in accordance with the Chilean Ministry of Education's regulations.

Data collection was carried out by a trained SLP not associated with the study.

2.2. Procedures

Initially, the files of all prospective participants were accessed and investigated to determine that the medical reports did not contain a diagnosis for hearing loss or neurological health conditions. Then, prospective participants were scheduled for a 1 h session to confirm the diagnosis of comorbid SSD and DLD. In this session, the Test Exploratorio de Gramática Española de A. Toronto (Screening Test of Spanish Grammar, STSG) (Pavez, 2010) was carried out. This test evaluates grammatical development in Spanish-speaking children between the ages of 3 years and 6 years, 11 months. It contains both a comprehension and an expression subtest with 23 items each. Pairs of sentences are presented to the child with the aim of evaluating the syntactic elements. In the comprehension subtest, four images with similar actions are presented (one image contains the correct action being asked). Then, the examiner says a sentence and the child must point to the correct image. In the expression subtest, two images are presented to the child, and the examiner says two sentences (i.e., one sentence relating to each image without indicating the image they are referring to). Then, the child is asked to produce a sentence for each of the two images that are shown. Both subtests evaluate affirmative and negative sentences, prepositional phrases that only include locative prepositions, and personal pronouns with different functions within the sentence: as a subject, as a direct object and in prepositional syntagma, verb tenses, indefinite pronouns, demonstrative pronouns, adverbs and interrogative pronouns, possessive adjectives, relative pronouns, copulative verbs, and passive sentences. This instrument has validity and reliability criteria and allows a quick detection of syntactic disorders in Spanish-speaking children. The selected children should have presented with a score below -2 standard deviations for the comprehension and/or expression subtest, which determines the presence of DLD. This is because grammatical development has been suggested as a marker for DLD (Rice, 2014). Based on the child's overall score for each subtest (i.e., expression and comprehension) and the normative data according to the age of the child, three categories can be obtained: poor or deficient grammatical performance (below -3 standard deviations), low grammatical performance (between -2 and -3 standard deviations), and normal grammatical development (above -2 standard deviations). Categorical results for each child in each subtest were then combined with the aim to determine the severity of the language impairment. The following grouping, from more to less severe grammatical development, was generated based on the results obtained in the sample of children: poor receptive and expressive grammatical development (PRE), adequate receptive development and poor expressive development (PE), and adequate receptive development and low expressive development (LE). These three categories were then used in the statistical analyses when exploring the variable of severity of DLD (see below for details). Note that all children presented with expressive grammatical development below normal ranges. Therefore, the difference between PE and LE is explained by the severity of the expressive grammatical problems. These problems were more severe in children with PE than in children with LE.

Then, speech production was evaluated with the aim to confirm the diagnosis of SSD. The latter was determined based on a higher than expected number of phonological process (PP) errors for the child's age. PP errors were tested using the Test for the Evaluation of

Phonological Processes (Test de Evaluación de Procesos de Simplificación Fonológicos) (TEPROSIF-R) (Pavez et al., 2008). This test is the only phonological instrument created and normalized for a Chilean population between 3 years and 6 years, 11 months of age. The test consists of 37 images representing words of different length, syllabic stress, and complexity. Examples of these words in Spanish include auto [car], plancha [iron], mariposa [butterfly], tren [train], helicóptero [helicopter], jirafa [giraffe], refrigerador [refrigerator], gorro [hat], and doctor [physician], among others. A flashcard containing a picture is shown to the child, and the evaluator reads aloud a phrase that is related to the picture with the last word missing. The child is asked to complete the sentence with the missing word (i.e., the image of the flash card). Children's responses were written down on the scoring form and further analyzed by two other SLPs. The total number of PP errors produced by the child was calculated, regardless of whether these errors reflected the same phonological process. For example, if a child produced 'tren' as [ten] and 'refrigerador' as [refixerador], these errors were considered as two PPs. Agreement between the two raters was estimated using the intraclass correlation coefficient (ICC = 0.99). All selected children presented with a greater number of PP errors than expected for their age, and thus the presence of speech production problems was confirmed. It has been documented that the most frequently produced PPs by Chilean preschool children are those related to the syllabic structure such as unstressed syllable deletion (e.g., replacement of the word /elikóptero/), cluster reduction (e.g., replacement of the word /trén/ with /tén/), and assimilation (e.g., replacement of the word /ziráfa/ with /firafa/) (Coloma et al., 2010).

With the aim to confirm that the speech production problems were due to an SSD and not due to apraxia of speech, children were asked to repeat isolated syllables containing each of the 17 consonant phonemes of the standard Spanish spoken in Chile (Sadowsky & Salamanca, 2011) combined with each of the five vowels in Spanish. Each stimulus had to be repeated three times in a row (e.g., [pa pa pa], [pe pe pe], [pi pi pi], [po po po], [pu pu pu]). Both error rates and intra-syllable consistency were obtained. On this task, each child performed with a 25 % or less error rate and 75 % or more intra-syllable consistency. In addition, prosody in speech was assessed by means of clinical observation by the examiner (i.e., SLP). Both aspects were used to rule out the presence of apraxia of speech, following the recommendation of the American Speech-Language-Hearing Association (2007).

All the selected children (n = 41) were then scheduled for another 90-minute session (i.e., second session) in which auditory perception, nonverbal oral movements, phonological awareness, and working memory were evaluated. Details for such procedures are given below. The tests were applied in the same order to all research participants. Feedback was not provided during or at the end of the evaluation session.

2.3. Instruments

2.3.1. Auditory perception

To evaluate auditory perception, the auditory perception subtest of the Neuropsychological Assessment of Children battery (*Evaluación Neuropsicológica Infantil*) was used. This battery evaluates 12 neuropsychological domains such as basic cognitive functions, executive functions, and academic ability. There are norms available for Spanish-speaking populations 5–16 years of age (Matute, Rosselli, Ardila, & Ostrosky, 2013).

The auditory perception subtest is part of the basic cognitive functions domain. This subtest evaluates discrimination of musical notes, recognition of environmental sounds, and discrimination of phonemes. The stimuli were played on a portable computer at a comfortable intensity level. First, eight pairs of musical notes were presented, and for each pair, the child was asked to judge whether the two stimuli were the same or different. Next, eight environmental sounds were presented, and the child was asked to identify each one of them, naming the element that produces that sound. Both tasks have a maximum score of 8. Finally, 20 pairs of two-syllable words were presented; 10 pairs contained the same words and the remaining 10 pairs contained different words. Each pair of different words differed in only one consonant phoneme, and this differed in only one distinctive feature. For example, the pair /pesa/-/besa/ might be presented but not /pesa/-/resa/. In Spanish, /p/ and /b/ are unaspirated plosive consonants that only differ in their voice onset times. The child was asked to say whether the two words were the same or different. Each correct response received one point, for a maximum score of 20. According to the test guidelines, the score for each of the auditory perception subtests has to be normalized so the scores can be summed up. Thereby, an overall score for auditory perception was obtained. The instrument overall score sort for the phonemic discrimination task were used.

2.3.2. Nonverbal oral movements

The Guidelines for Evaluating Nonverbal Praxis (*Pauta de Evaluación Observacional de Habilidades Práxicas No Verbales*) (Villanueva, 2014) were used to assess this domain. This instrument has been used in previous research with Spanish-speaking children (Jaque, Jeldes, & Mieres, 2011). These guidelines provide the evaluation criteria for isolated nonverbal movements, including seven lip movements such as bilabial trill, lip protrusion, labial retraction and lateralization, and 15 tongue movements such as lingual click, lingual lateralization, and tongue tip elevation inside and outside the oral cavity, among others. One point is given for each correct oral movement performed, for a maximum of 22 points. The scores were transformed into a percentage of correctly produced nonverbal oral movements.

2.3.3. Phonological awareness

Phonological awareness was evaluated using the Test for Assessing Phonological Metalinguistic Abilities (*Prueba Destinada a Evaluar Habilidades Metalingüísticas de Tipo Fonológicas*) (Yacuba, 1999). Normative scores are available for children aged between 4 years, 9 months and 6 years, 2 months (Yacuba, 1999). The instrument is comprised of six subtests. The first four subtests evaluate

syllable awareness, and the last two subtests evaluate phoneme awareness: (1) final sounds: A reference flashcard is presented to the child, and then, from three flashcards, the child has to choose the one whose image's name (i.e., word) ends with the same phoneme as the reference flashcard; (2) initial sounds: Same as (1) but now the child has to choose the flashcard whose image's name commences with the same phoneme as the reference flashcard; (3) syllable segmentation: In this subtest, a piece of paper that has a drawing and a table containing five cells is presented to the child. Colored pencils are given to the child, and they are asked to color the amount of cells on the table that are equal to the number of syllables that the word represented by the image has; (4) syllable reversal: Initially, a reference flashcard with an image is presented to the child and they are asked to say each of the syllables of the word represented by the image in a backward fashion. By doing so, a new real word is created. Then, given three flashcard in reverse order; (5) letter sound: The examiner says aloud a phoneme, and, given three flashcards each containing a different grapheme, the child has to choose the one that matches with the phoneme produced by the examiner; (6) blending: The examiner says aloud in direct order the phonemes of a word, and, given three flashcards, the child has to choose the one whose image represents the word by blending the phonemes produced by the examiner. Each of the subtests mentioned above is comprised of eight items. For each item, the score is either 1 (correct) or 0 (incorrect). Thus, the overall score is 48 (six subtests with 8 points each).

2.3.4. Working memory

This domain was evaluated using the Pseudoword Repetition Test (*Prueba de Repetición de Pseudopalabras*) (Aguado, 2011). This test has Spanish norms for children aged 5–7 years. The test includes two subtests of 40 pseudowords each. The first subtest contains words created from frequent Spanish syllabic structures (frequent pseudowords), such as [kóte] or [brénodi]. The second subtest contains words created from infrequent Spanish syllabic structures (infrequent pseudowords), such as [geónku] or [blísmu]. Each subtest of 40 pseudowords contains 10 words of two syllables, 10 words of three syllables, 10 words of four syllables, and 10 words of five syllables. The evaluator read each pseudoword aloud, and the child had to repeat it back. For each subtest, one point is given for each correctly repeated pseudoword, for a maximum of 40 points. Points are not deducted for articulation errors. Therefore, following the criterion suggested by Dodd, Reilly, Ttofari, and Morgan (2018), phonetic-based sound distortions such as interdental, dental, and lateral lisps were not counted as errors in this test.

2.4. Statistical analysis

Initially, descriptive statistics were computed for each of the variables of interest. These factors included the continuous variables of number of PP errors as a measure of the severity of the speech production problems (i.e., results of TEPROSIF-R test, dependent variable), the scores for the frequent and infrequent pseudowords subtests of the Pseudoword Repetition Test as two measures of working memory (independent variable), the scores of the Test for Assessing Phonological Metalinguistic Abilities as a measure for phonological awareness (independent variable), the percentage of correctly produced nonverbal oral motor movements obtained using the Guidelines for Evaluating Nonverbal Praxis as a measure for nonverbal oral movements (independent variable), and the scores of the auditory perception subtest of the Neuropsychological Assessment of Children battery as a measure of auditory perception (independent variable). In addition, the severity of the DLD was included in the analyses as a confounding variable. The severity of the DLD was determined from the categories obtained with the STSG (i.e., PRE, PE, and LE).

Then, a Shapiro-Wilk test was used to determine whether the number of PP errors obtained in the TEPROSIF-R test was normally distributed. Because the distribution was not normal (z = 3.36; p < 0.001), a Spearman correlation coefficient was used to analyze the relationships among the continuous variables. With the aim to explore possible associations between the dependent variable (i.e., number of PP errors) and the factors of auditory perception (overall score for the auditory perception subtest and scores for the phoneme discrimination task), nonverbal oral movements, phonological awareness, and working memory (scores for frequent and infrequent pseudowords), along with the covariate of severity of the DLD (categories for the expression and comprehension subtests of the grammatical development test—STSG), five univariate linear regression models were constructed. Considering that the severity of the DLD is a categorical variable, a simple coding system for this variable was used in the linear regression models. The category PE (adequate receptive development and poor expressive development) was used as the reference category because it comprises the same level of receptive grammatical development as the PRE category (poor receptive and expressive grammatical development) and the same level of receptive grammatical development as the LE category (adequate receptive development and low expressive development). Therefore, PE is the only category that has similar characteristics with the other two categories.

Regarding the association between number of PP errors and working memory, four further models were created between the number of PP errors and the scores obtained for the repetition of pseudowords of different length (i.e., disyllables, trisyllables, tetrasyllables and pentasyllables).

With the aim to investigate the effect of DLD on working memory and phonological awareness, two univariate models were constructed using DLD as the independent variable.

Then, two multivariate models including only the variables that were associated (p value <0.1) with the dependent variable in the univariate models mentioned above were constructed. The severity of the DLD (categorical variable) was included in the model as a covariate. For each model, the proportion of variance explained was estimated using the coefficient of determination R^2 . To evaluate for potential interaction effects among the independent variables significantly associated with the number of PP errors, interaction terms between such variables were created. These terms were included in another multivariate linear model with the number of PP errors as the dependent variable. A likelihood-ratio test was computed to compare the multivariate models with and without the interaction terms.

As the distribution of the dependent variable (PP errors) was non-normal, the standard error of linear models was estimated with bootstrapping (10,000 replications). The 95 % confidence intervals (95 % CI) were calculated using the bias-corrected and accelerated method (Poi, 2004). Homoscedasticity was evaluated with the Breusch-Pagan/Cook-Weisberg test. As the assumption of homoscedasticity was not met in most of the models, the Huber Sandwich Estimator was used to estimate the variance (Rogers, 1993). Collinearity was explored among the independent variables included in the models and was evaluated using variance inflation factors (VIFs). STATA version 14 (StataCorp LP, College Station, TX, USA) was used for all analyses.

3. Results

The mean number of PP errors (as obtained by TEPROSIF-R test) per child was $36.6 (\pm 18.8)$, with a range of 15-76 PP errors. The cutoff score (number of PP errors) for the age group investigated is 14. Thus, all children scored above the number of PP errors expected for their age, confirming the presence of SSD (Table 1).

The mean score on the auditory perception subtest was $31.3 (\pm 3.5)$, and the range was 25-40. According to this, 9.8 % of the children obtained a score below the suggested normative value (27 points, which represents the 25th percentile). The mean percentage of correctly performed nonverbal oral movements, evaluated using the Guidelines for Evaluating Nonverbal Praxis, was 70.5 % (\pm 14.8), and the range was 40.9 %-100 %. No normative data for this procedure are available. The mean score on the Test for Assessing Phonological Metalinguistic Abilities (phonological awareness) was $22.4 (\pm 6.4)$, and the range was 13-34. Thirty-one children (75.6 %) obtained scores below the normative values for this test (i.e., 28.05). The mean score on the working memory test was $15.5 (\pm 7.8; range 3-35)$ for pseudowords containing frequent syllabic structures (Table 1). A total of 85.4 % of children obtained scores below the normative values for seudowords with frequent syllabic structures (26 points, which represents the 25th percentile), and 78.1 % obtained scores below the normative values for Spanish Grammar), 7 children were classified as having poor receptive and expressive grammatical development (PRE), 17 children were classified as having adequate receptive development and poor expressive development (LE).

3.1. Correlations among variables

Table 2displays the correlation rank coefficients (Spearman's Rho) among the number of PP errors, auditory perception test results, percentage of correctly executed nonverbal oral motor movements, phonological awareness test results, and working memory test results (scores for frequent and infrequent pseudowords). A significant correlation between the number of PP errors and the score on the phonological awareness test was found. Also, a significant correlation between the number of PP errors and the score on the working memory subtests (frequent and infrequent pseudowords) was found. In addition, phonological awareness was significantly correlated with working memory subtests (frequent and infrequent pseudowords). Finally, regarding working memory scores, the scores for the frequent pseudowords subtest were significantly correlated with scores for the infrequent pseudowords subtest.

3.2. Association between PP errors and the independent variables-univariate models

Univariate regression models were used to explore the associations between the dependent variable (i.e., number of PP errors) and auditory perception, nonverbal oral movements, phonological awareness, and working memory (frequent and infrequent

Table 1

Mean, median, standard deviation and range for the scores of the test procedures used to evaluate the number of phonological process errors, auditory perception, nonverbal oral movements, phonological awareness and working memory in the group of children with comorbid SSD and DLD (n = 41).

Variable	Mean	Median	Standard deviation	Range
PP errors ^a	36.6	29	18.8	15–76
Auditory perception ^b	31.3	31	3.5	25-40
Nonverbal oral movements ^c	70.5	72.7	14.8	41-100
Phonological awareness ^d	22.4	20	6.4	13-34
Working memory ^e				
Frequent pseudowords	15.5	14	7.8	3–35
Infrequent pseudowords	10.8	9	8.3	0–28

^a PP: Phonological process. Scores are in number of phonological process errors obtained using the TEPROSIF test. Higher scores indicate a higher number of phonological process errors and thus worse results.

^b Scores represent the overall performance for the subtest of auditory perception. Higher scores indicate better performance for auditory perception.

^c Scores represent the percentage of correctly produced nonverbal oral movements as specified in the guidelines for evaluating nonverbal praxis. ^d Scores represent the overall performance on the test for assessing phonological metalinguistic abilities. Higher scores indicate better phonological awareness abilities.

^e Scores represent the overall results for the repetition of frequent and infrequent pseudowords. Higher scores indicate better working memory capacities.

Table 2

Correlation coefficients (Spearman's Rho) among the number of phonological process errors, auditory perception test results, percentage of correctly produced nonverbal oral motors, phonological awareness test results, and working memory test results for frequent and infrequent pseudowords (N = 41).

	PP errors	Auditory perception	Nonverbal oral movements	Phonological awareness	Frequent pseudowords	Infrequent pseudowords
PP errors	1.0					
Auditory perception	-0.20	1.0				
Nonverbal oral movements	-0.11	0.12	1.0			
Phonological awareness	-0.44**	0.23	0.23	1.0		
Working memory						
Frequent	-0.79***	0.05	0.05	0.42**	1.0	
pseudowords						
Infrequent	-0.85***	0.08	0.11	0.50***	0.90***	1.0
pseudowords						

PP: Phonological process.

*p < 0.05; **p < 0.01; ***p < 0.001.

pseudowords). Phonological awareness and working memory (frequent and infrequent pseudowords) were significantly associated with the number of PP errors. Phonological awareness explained nearly 20 % of the variance in the number of PP errors, and each subtest of working memory explained around 50 %. Both factors were negatively associated with the number of PP errors. This means that lower (i.e., poorer) results for both the phonological awareness and working memory tests were significantly associated with a higher number of PP errors (see Table 3).

Regarding the effect of the length of the pseudowords to evaluate working memory, the score obtained for each length of pseudoword (i.e., two, three, four, and five syllables) was significantly associated with the number of PP errors. However, the variance explained varied according to the length of the pseudowords, from 20.86 to 61.30 (see Table 4).

The severity of the DLD was significantly associated with the number of PP errors. Children with adequate receptive development and low expressive development (LE) showed fewer PP errors than children with adequate receptive development and poor expressive development (PE). However, no significant differences for the number of PP errors were found between children with poor receptive and expressive grammatical development (PRE) and children with PE. The severity of the DLD explained 16.3 % of the variance in the number of PP errors (see Table 3).

Finally, the number of PP errors was not significantly associated with auditory perception (overall score for the auditory perception subtest and scores for the phoneme discrimination task) and the percentage of correctly produced nonverbal oral motor movements (see Table 3).

3.3. Association between severity of the DLD and working memory and phonological awareness

The severity of the DLD was significantly associated with working memory and phonological awareness, explaining 20.4 % of the variance in working memory and 26.9 % of the variance in phonological awareness. As the DLD was more severe, working memory and

Table 3

Univariate linear regression analyses for the number of phonological process errors (dependent variable) and the independent variables of auditory perception (overall score of the auditory perception subtest and scores for the phoneme perception task), nonverbal oral movements, phonological awareness, working memory, and the severity of developmental language disorder (DLD) (N = 41).^a

Factors	Coefficient (β)	95 % CI	p-value	Variance explained (%)
Auditory perception (overall score)	-1.39	-2.96 to 0.25	0.093	0.67
Phoneme discrimination task	-0.91	-2.57 to 0.86	0.300	0.02
Nonverbal oral movements	-0.08	-0.43 to 0.35	0.680	0.04
Phonological awareness	-1.30	-2.01 to -0.58	<0.001	19.48
Working memory				
Frequent pseudowords	-1.93	-2.34 to -1.46	<0.001	63.67
Infrequent pseudowords	-1.64	-2.12 to -1.24	<0.001	53.46
Severity of DLD ^b				
PRE	-1.78	-18.53 to 15.26	0.836	
PE	Reference	-	-	16.31
LE	-15.65	-26.96 to -4.04	0.008	

PE was considered as the reference category as it is the only category that shares the same level (i.e., adequate, low, poor) of one dimension (i.e., receptive and expressive grammar) with at least another category. Specifically the expressive grammar level is the same for PE and PRE; and the receptive grammar level is the same for PE and LE.

^a Variables significantly associated with phonological process errors in bold.

^b PRE: poor receptive and expressive grammatical development, PE: adequate receptive development and poor expressive development, LE: adequate receptive grammatical development and low expressive development.

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Table 4

Univariate linear regression analyses for the number of phonological process errors (dependent variable) and results for the working memory test according to the number of syllables of the pseudowords (N = 41).^a.

Number of syllables of the pseudowords	Coefficient (β)	95 % CI	p-value	Variance explained (%)
Disyllables	-5.30	-6.32 to -3.89	<0.001	50.28
Trisyllables	-5.87	-7.45 to -4.40	<0.001	54.86
Tetrasyllables	-6.76	-8.48 to -5.04	<0.001	61.30
Pentasyllables	-4.67	-7.65 to -2.29	0.002	20.86

^a Variables significantly associated with phonological process errors in bold.

phonological awareness results were worse.

3.4. Association between PP errors and the independent variables-multivariate model with no interaction term

The multivariate model included the independent variables significantly associated with the number of PP errors in the univariate models. Therefore, phonological awareness and working memory (i.e., frequent pseudowords subtest) were incorporated in the multivariate model as well as the severity of the DLD. The latter was included as a confounding variable, and thus the effect of working memory and phonological awareness on the number of PP errors was controlled for the severity of the DLD. Note that due to collinearity between the frequent and infrequent pseudowords subtests, in the multivariate models only the scores for the frequent pseudowords subtest were included as a measure of working memory.

The results of this multivariate model showed that working memory test scores (i.e., frequent pseudowords subtest) explained a greater proportion of the variance in the number of PP errors. All three variables together (i.e., working memory as measured by the frequent pseudowords subtest, phonological awareness and the severity of the DLD) explained 64.2 % of the variance in the number of PP errors (see Table 5).

3.5. Variance in number of PP errors explained by the multivariate model with the interaction term

Taking into consideration the association between working memory and phonological awareness and that both variables were significantly associated with the number of PP errors in the univariate models, an interaction term between these two variables was constructed. In the multivariate model, this interaction term was significantly associated with the number of PP errors. The model that included the interaction term along with phonological awareness and working memory (i.e., frequent pseudowords subtest) explained 70 % of the variance in the number of PP errors. Note that the severity of the DLD was also included in this multivariate model as a confounding variable, and thus the results were adjusted by the severity of the DLD.

Fig. 1shows that the slope of PP plotted against working memory varies according to phonological awareness, indicating the presence of an interaction effect. When working memory is poor, the number of PP errors varies according to the level of phonological awareness, but when working memory is good, this variability is not observed.

A likelihood-ratio test was computed to compare the multivariate models with and without the interaction term. The model that included the interaction term between phonological awareness and working memory fitted the data significantly better than the model

Table 5

Multivariate regression analyses for the number of phonological process errors and the factors of phonological awareness, working memory and severity of the developmental language disorder (DLD) (n = 41).

Factors	Multivariate model without interaction term ^a	p-value	Multivariate model with interaction term ^b	p-value
Phonological awareness Working memory (Frequent pseudowords) Severity of DLD ^C	-0.51 (-1.09 to 0.16) -1.82 (-2.24 to -1.30)	0.107 < 0.001	-2.43 (-4.41 to -0.76) -4.26 (-6.10 to -2.41)	0.013 < 0.001
PRE PE	-8.85 (-19.36 to 0.90) Reference	0.085	-11.47 (-21.22 to -2.22) Reference	0.018 -
LE Interaction between phonological awareness and working memory (Frequent pseudowords)	-2.14 (-11.92 to 6.64) -	0.653 -	-3.63 (-12.48 to 5.63) 0.11 (0.03 to 0.21)	0.435 0.019
Intercept Variance explained (%)	78.53 (61.30 to 93.02) 64.2	<0.001 -	118.79 70.0	<0.001 -

Coefficients significantly associated with the number of phonological process errors in bold.

PE was considered as the reference category as it is the only category that shares the same level (i.e., adequate, low, poor) of one dimension (i.e., receptive and expressive grammar) with at least another category. Specifically the expressive grammar level is the same for PE and PRE; and the receptive grammar level is the same for PE and LE.

^a Model without including an interaction term between phonological awareness and working memory.

^b Model including an interaction term between phonological awareness and working memory.

^c PRE: poor receptive and expressive grammatical development, PE: adequate receptive development and poor expressive development, LE: adequate receptive grammatical development and low expressive development.



Fig. 1. Number of phonological process errors plotted against working memory performance (frequent pseudowords) for various phonological awareness test scores (5 points represents the lowest performance and 30 points the highest performance). Slopes as predicted by the model with the interaction term.

without the interaction term ($\chi 2$ (1) = 8.43; p < 0.01).

4. Discussion

4.1. Association between predictor variables and the number of PP errors

The primary aim of this study was to determine which of the four factors of interest (auditory perception, nonverbal oral movements, phonological awareness, and working memory) best explained the severity of speech production problems in children with SSD and DLD. The results indicate a significant association between the number of PP errors and phonological awareness and working memory. However, working memory explained the greatest proportion of the variance in the univariate models (63.67 %) as opposed to phonological awareness, which explained 19.48 % in the variance of number of PP errors. In addition, the severity of the DLD explained 16.31 % of the variance in the number of PP errors. The number of PP errors was not significantly associated with auditory perception or nonverbal oral movements.

Regarding working memory in particular, when observing the coefficients obtained in the univariate regression models, for every 1 point of increase on the working memory test (frequent pseudowords subtest), the number of PP errors decreased by 1.93. This is clinically significant. These results are consistent with reports from Torrington Eaton and Bernstein Ratner (2016), who found an association between working memory and SSD in children aged 4–5 years. They found that children with speech production problems showed worse performance in working memory compared to children with typical language development. In addition, performance in working memory impairments were associated with speech sound accuracy and word production. The authors determined that working memory impairments were associated with inaccurate mental representation of words since the input received (i.e., words) decays more rapidly, so the children did not store the correct models, and, therefore, the accuracy of their word production was impaired, giving rise to an increased number of PPs.

In addition, as expected, the length of the pseudowords used to evaluate working memory influenced in the proportion of variance of the number of PP errors. There is a progressive increase in such a variance explained by increasing from two to three syllables and from three to four syllables. However, there is an important decrease in the variance explained when the pseudowords are increased to five syllables. These results are consistent with previous studies reporting that working memory resources depend on the length of the pseudowords used (Baddeley, Thomson, & Buchanan, 1975; Olson, Chun, & Anderson, 2001). The fact that the variance on PP errors explained by the working memory task diminished when five-syllable pseudowords were used may be explained by the difficulty of the task. In other words, it seems that this task was difficult for all children.

As mentioned above, phonological awareness was also significantly associated with the number of PP errors. Mann and Foy (2007) mentioned that the association between the precision of speech production (as evaluated through PP errors in this study) and phonological awareness may be due to the fact that both require internal representations of the phonological structures. However, it is possible that the observed effect of phonological awareness on the number of PP errors in this study is due to the effect working memory has on phonological awareness rather than a primary effect of phonological awareness on the number of PP errors. For example, Gathercole, Willis, Emslie, and Baddeley (1992) observed that working memory and phonological awareness were

associated. Mann and Foy (2007) suggested that working memory may be a moderator of the link between articulatory skills (i.e., number of PP errors in this study) and phonological awareness. The association between working memory and phonological awareness is addressed below in a separate section. Note that the test used in this research to evaluate phonological awareness included a letter sound task. Some authors consider this task a different construct than phonological awareness and suggest that it specifically evaluates alphabetic knowledge (e.g., Anthony et al., 2011; Pavelko, Lieberman, Schwartz, & Hahs-Vaughn, 2018).

The severity of the DLD also explained an important proportion of the variance of the PP errors (16.31 %), although it was lower than the proportion explained by phonological awareness and working memory. Children with adequate receptive development and low expressive development (LE) showed fewer PP errors than children with adequate receptive development and poor expressive development (PE). The difference between both categories relied on the level of expressive grammatical development. Therefore, the results indicate that better expressive grammatical development is associated with fewer PP errors. No significant differences for the number of PP errors were found between children with poor receptive and expressive grammatical development (PRE) and children with adequate receptive development and poor expressive development (PE). This may be explained by the large percentage of children (83 %) who exhibited adequate receptive grammar. In this regard, Fey et al. (1994) argued that there is abundant evidence on the relationship between grammatical performance and speech production in preschool children with language disorders. Further studies should investigate the impact of comorbid speech and language disorders on speech production problems. Note that several studies conducted in children with SSD did not exclude children with expressive language disorders, and thus their results may reflect, at least in part, the effect of DLD on speech production.

Consistent with prior studies, nonverbal oral movements did not predict the number of PP errors. Lof and Watson (2008) concluded that nonverbal oral movements are independent of the act of speech. This finding has implications for intervention decisions, as therapy for this type of impairment is not likely to improve speech production problems in children with SSD. In addition, some studies have shown that nonverbal motor exercises seem to have little effect on phonological development (Lass & Pannbacker, 2008; Ruscello, 2008). However, these results should be interpreted with caution. In a narrow review, Kent (2015) mentioned that the association between speech production tasks and nonverbal motor movements relies on the type of nonverbal oral movements evaluated. In the present study, isolated nonverbal movements, including lip movements and tongue movements, were used. Thus, the results cannot be extrapolated to all nonverbal oral movements. We may argue that if movements in sequences were used, the results may have been different. This is because this type of exercise requires higher motor demands.

As mentioned earlier, in this research, auditory perception was not significantly associated with the number of PP errors. In agreement with our results, Preston et al. (2015) found no significant differences between children with SSD and typically developing children in phoneme perception tasks. However, it is important to consider that the concept of auditory perception is broad and can therefore include different abilities. For example, the studies of Tallal et al. (1985) used tasks of detection, association, temporal resolution, sequencing, rate processing, and auditory serial memory abilities. In this way, under the concept of auditory perception, in the literature, tasks of discrimination of nonverbal sounds, auditory memory, auditory attention, temporal resolution, and discrimination of consonants in the syllabic coda, among others, are found. This is relevant to interpreting our results. It is possible that we did not find a significant association between auditory perception and the number of PP errors because the tasks chosen to measure the auditory perception of the participants were very simple. For example, Rvachew, Nowak, and Cloutier (2004) found that the task to distinguish between a correctly pronounced sound and an incorrectly produced sound by the child is the task that is best associated with speech articulation performance. This type of task was not included in this study. Thus, further research using different measures of auditory perception, including discrimination of correctly and incorrectly produced sounds, should be conducted in children with SDE association.

4.2. Interaction among predictor variables in the number of PP errors

The second aim of this study was to determine whether the severity of the speech production problems in children with SSD and DLD may be explained by interactions among these four factors along with the severity of the DLD. As mentioned above, working memory and phonological awareness were significantly associated with the number of PP errors, and thus we included an interaction term between both factors in a multivariate regression model. This model explained the majority of the variance in the number of PP errors, accounting for more of the variance than the multivariate model with these variables but without the interaction term. Thus, the effect of working memory and phonological awareness on the number of PP errors cannot be attributed solely to covariance between both variables, implying that interaction is present. In other words, the interaction effect between both variables on the number of PP errors is more than the summation of each independent effect. This may suggest that the severity of speech production problems in children with SSD may be explained by the interaction between phonological awareness and working memory rather than by each variable independently.

When there is interaction, the coefficients of the regression are modified; therefore, there is no linearity. The slope for the number of PP errors varies according to the performance of phonological awareness and working memory. On a practical level, this allows us to differentiate among different levels of performance in the variables investigated. As Fig. 1 shows, if the performance on the phonological awareness task is taken as a reference, it is observed that in the group of children with lower scores in this variable, better performance in working memory is associated with a lower number of phonological process errors, on a sharp slope. Therefore, in children with poorer phonological awareness, a better working memory implies the greatest decrease in the number of phonological awareness task, the changes in performance on the working memory task are linked to a more modest decrease in the number of PP errors. As expected, the children with the best performance in working memory and phonological awareness showed the fewest PP errors. Therefore, working

memory is more relevant than phonological awareness to determine the number of PP errors. When working memory shows better performance, there seems to be no added effect of phonological awareness. However, when working memory shows low performance, phonological awareness has an important effect. Therefore, the interaction between working memory and phonological awareness is mainly observed when children have a poor working memory.

We believe that, despite being associated with each other, working memory and phonological awareness represent two distinct constructs, as no significant collinearity was found between them. Passolunghi and Siegel (2001) suggested that working memory and phonological awareness may utilize a common process yet represent distinct cognitive systems. Gorman (2012) studied typically developing children longitudinally and observed that working memory plays an important role in the development of phonological awareness. Therefore, working memory and phonological awareness are different cognitive abilities, but they are closely related. It seems that one requires the other to explain the number of PP errors. In addition, it should be noted that the significant correlation between working memory and phonological awareness may relate to the demand of working memory resources required to perform the phonological awareness task used in this study. Oakhill and Kyle (2000) claimed that demands on working memory resources depend on the type of tasks to evaluate phonological awareness. Gillam and van Kleeck (1996) explained this by pointing out that during a phonological awareness task, children have to convert the acoustic image of a word into a stable phonological representation, a process that requires working memory resources.

In regard to the relationship among working memory, phonological awareness, and PP errors, we believe that there is a cognitive process that is shared among these three variables. We hypothesize that such a cognitive process is the phonological representation of words. Several authors have claimed that weak phonological representations (representations stored in the mental lexicon that contain the phonological representations of words) lead to major speech production problems (Elbro, Borstrøm, & Petersen, 1998; Gillon, 2000; Larrivee & Catts, 1999; Rvachew, 2007; Sutherland & Gillon, 2005). Munson, Edwards, and Beckman (2012) suggested that working memory is crucial to create these phonological representations. Therefore, we suggest that there is a common mechanism among working memory, phonological awareness, and PP errors but that working memory is the most important factor and that is why it explains the largest variance in the number of PP errors in the regression models.

It should be noted that the multivariate model was adjusted for the severity of the DLD as there is a significant association between this variable and the number of PP errors. Also, this adjustment was incorporated because in the current study, the severity of the DLD was associated with the performance on both working memory and phonological awareness tasks. Several studies have shown that children with DLD who exhibit grammatical problems also exhibit poorer working memory and phonological awareness than their peers without DLD (Archibald & Gathercole, 2006; Claessen & Leitão, 2012; Dollaghan & Campbell, 1998). Thus, by controlling for the severity of DLD in the regression models constructed with the data of this research, the effect of working memory and phonological awareness on PP errors cannot be solely explained by the grammatical development of children. Note that these results cannot necessarily be extrapolated to children with SSD because the sample was comprised of children with comorbid SSD and DLD.

Finally, in the present study, phonological difficulties were assessed through the number of PP errors, as suggested by Edwards (1992). However, it should be noted that this is not the only way of evaluating phonological difficulties. Other methods include the percentage of correctly produced consonants (Lousada, Jesus, Hall, & Joffe, 2014; Pascoe, Stackhouse, & Wells, 2006) and the number of atypical PPs (Preston & Edwards, 2010; Rvachew, Chiang, & Evans, 2007).

4.3. Clinical implications

These results show that both working memory and phonological awareness should be assessed as part of the phonological abilities of children with SSD when this disorder coexists with a DLD. Similarly, both aspects (i.e., working memory and phonological awareness) should be incorporated in rehabilitation programs for children with SSD and DLD. Further research should investigate how these aspects can be incorporated in rehabilitation programs and their outcomes in the improvement of speech production in children with SSD and DLD.

Currently, most programs available for treating children with SSD focus each intervention on a single aspect of phonological processing (Joffe & Pring, 2008), most commonly auditory perception (Rvachew, Nowak, & Cloutier, 2004) or phonological awareness (Hesketh, Adams, Nightingale, & Hall, 2000). A survey of SLPs working in the United Kingdom found that interventions for SSD typically involved therapies for auditory perception and phonological awareness (Joffe & Pring, 2008) but not working memory.

In addition, it is important to consider comorbidity of SSD and DLD. From the findings of this study, we suggest that expressive grammar should be taken into consideration when evaluating and/or treating speech production problems in children. Note that Fey et al. (1994) found that a grammar-centered intervention program did not have a positive impact at reducing the speech production disorder and that the latter requires specific intervention. We believe that an intervention program that includes activities for working memory, phonological awareness, and grammatical development all together should have a higher impact at reducing speech production problems than programs where each of these aspects is addressed separately. Rvachew and Brosseau-Lapré (2015) showed that intervention programs that combine more than one aspect/ability produce better results in children with SSD.

4.4. Study limitations

First, the results from this study should be interpreted with caution as all children had comorbid SSD and DLD, and thus the results cannot necessarily be applied to children with SSD only. In addition, from the results of this study, the effect of language development on the PP errors could not be determined accurately. This is because language development and, thus, the severity of DLD were determined from the results of a test that evaluates receptive and expressive grammar (i.e., STSG). No other aspects relating to

language development, such as vocabulary development, were obtained in this study. Also, the grammatical development was considered as a categorical variable and not as a continuous variable. With the aim to better understand the association between language development and PP errors, further studies should consider the inclusion of children with different levels of language development. Also, such studies should incorporate a comprehensive battery of tests that explore different aspects of language development.

Second, it is possible that other variables such as socioeconomic level may affect cognitive aspects, which in turn may impact the association between PP errors and phonological awareness and working memory. However, the study sample was relatively uniform in this regard. Both schools that participated in the research are in an urban area where medium-low-income families live, and they provide therapeutic interventions free of charge. Other variables that were not included in this study are semantic-pragmatic aspects of language, which also may be associated with phonological awareness and working memory. Children with concomitant deficits in other language domains may show different problems of phonological processing compared to children with deficits solely in phonology (Rvachew et al., 2007).

Third, the number of PP errors was not significantly associated with auditory perception or nonverbal oral movements. This may be due to the evaluation procedures used to assess each of these two latter variables, as both instruments were rather simple and did not explore each domain in detail.

Finally, Children's responses and execution of nonverbal oral movements were not video recorded, and thus it was not possible to obtain inter-rater reliability.

5. Conclusion

The present study reveals a significant association between the severity of speech production problems and phonological awareness and working memory in children with SSD and DLD. Working memory accounted for most of the variability in the number of PP errors in the sample of children investigated in this study. Neither auditory perception nor nonverbal oral movements were significantly associated with the number of PP errors. In addition, comorbidity of SSD and DLD influences speech production as the severity of the DLD was significantly associated with the number of PP errors.

In the multivariate models, working memory, phonological awareness, and the interaction term between them explained most of the variability in the number of PP errors in the sample of children with SSD and DLD.

CRediT authorship contribution statement

Felipe Torres: Conceptualization, Investigation, Writing - original draft, Writing - review & editing, Visualization, Supervision, Project administration. Eduardo Fuentes-López: Methodology, Formal analysis, Writing - original draft, Writing - review & editing, Visualization. Adrian Fuente: Writing - original draft, Writing - review & editing. Fabiana Sevilla: Investigation.

Declaration of Competing Interest

The authors have no conflicts of interest to declare.

Acknowledgements

The authors would like to thank Hugo Segura Pujol, Associate Professor at Universidad Autónoma de Chile, Talca, Chile. Carmen Julia Coloma, Full Professor at Departamento de Fonoaudiología, Facultad de Medicina, Universidad de Chile and Camilo Quezada, Assistant Professor at Departamento de Fonoaudiología, Facultad de Medicina, Universidad de Chile, Santiago, Chile.

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