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Longitudinal profiles of late phonological development in children with Williams syndrome

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Williams syndrome (WS) is a genetic neurodevelopmental disorder characterized by language skills above what is expected considering non-verbal intelligence. Research on phonological development is scarce, with many studies focusing on grammar in children and adolescents. In one of our previous studies transversally explored the profiles of late phonological development in Spanish-speaking WS children, adolescents, and adults, while our objective is to longitudinally determine these profiles for WS children based on present error indexes in spontaneous speech. Participants were seven WS children (aged 3;7–8;2), engaging in two spontaneous conversations within a 6-month interval. They were compared cross-sectionally with 240 typically developing (TD) children aged 3–6 years, divided into six groups. All speech samples were transcribed and analyzed with the CLAN software package of the CHILDES Project. Phonological profiles were established on the basis of phonological error indexes obtained dividing absolute frequency of errors by the total number of words produced. WS children showed a mean reduction of more than 25% in the absolute frequency of phonological errors after 6 months. As for the comparison with the normative groups, their error index was consistent with the stage of expansion in TD, however, after 6 months, this was consistent with the stage of stabilization. This atypical acceleration in phonological development could be related to lexical growth in the context of relative preservation of phonological memory. Furthermore, the trajectories of late phonological development in WS children might not be linear, as postulated by neuroconstructivist models, suggesting the need for intervention approaches specifically adapted to the phonological profiles of WS children.

KEYWORDS

Williams syndrome, phonological profiles, spontaneous speech, atypical language development, neurodevelopmental disorder

1 Introduction

Williams Syndrome (WS) is a multisystem neurodevelopmental disorder caused by a heterozygous deletion on chromosome 7q11.23 (Pérez-Jurado, 2003), whose prevalence according to the most cited epidemiological study, is estimated at 1 in 7,500 births (Strømme et al., 2002), with no sex difference, racial or ethnic predilection (Morris et al., 2020). The WS physical phenotype includes distinctive and easily recognizable facial features, cardiovascular anomalies, endocrine-metabolic alterations, hoarse voice, and sound sensitivities (hyperacusis, odyacusis, auditory allodynia, and auditory fascinations) (Kozel et al., 2021). Individuals with WS show a specific neurocognitive profile

characterized by a complex pattern of strengths and weaknesses (Karmiloff-Smith et al., 2003b; Vicari et al., 2004; Mervis and John, 2010; Hocking et al., 2015; Wuang and Tsai, 2017; Miezah et al., 2021) and they may show mild-to-moderate intellectual disability (Bellugi et al., 2000; Mervis et al., 2000). In general, deficits in visuospatial construction skills constitute a specific weakness (Mervis et al., 2000; Brown et al., 2003; Farran and Jarrold, 2003; Van der Geest et al., 2005; Porter and Coltheart, 2006), whereas auditory processing and face recognition are strengths in the WS profile (D'Souza et al., 2015; Miezah et al., 2021).

Earlier studies described language as selectively preserved and dissociated from other cognitive functions (Bellugi et al., 1988, 1994, 2000). Further research highlighted that superior verbal skills reported in individuals with WS may be explained in terms of asynchronous trajectories of development, with verbal abilities increasing more rapidly than non-verbal abilities (Jarrold et al., 2001). This asymmetric profile of WS would not be explained as the product of a cluster of damaged or preserved static modules, but as the emergent result of the dynamic processes of development where genes, the brain, cognition, behavior, and the environment interact multidirectionally throughout the life span (Karmiloff-Smith et al., 2003a; Karmiloff-Smith, 2009).

In general, pragmatic abilities have been described as an area of relative weakness in individuals with WS, arising both in narrative and conversational settings (Stojanovik, 2006; Diez-Itza et al., 2018, 2022). In contrast, structural components of language, i.e., phonology, morphosyntax, and vocabulary, have been considered relative strengths. From a preservation perspective, the results of different studies on morphological skills in WS were interpreted in terms of a typically functioning system with some impaired components (Clahsen and Almazan, 1998; Clahsen et al., 2004; Penke and Krause, 2004). However, more recent studies suggest a certain degree of atypical morphological processing (Benítez-Burraco et al., 2017; Diez-Itza et al., 2017, 2019). Several studies have pointed out that grammatical ability is strongly correlated with expressive vocabulary size (Vicari et al., 2002; Volterra et al., 2003).

Regarding lexical acquisition, several studies have emphasized that young children with WS follow an atypical pattern. Unlike typically developing (TD), children with WS produce their first words before understanding or producing protodeclarative gestures (Mervis and Bertrand, 1997) or produce them in smaller quantities even while producing referential language (Laing et al., 2002). It is also known that adolescents and adults have a vocabulary which is extensive and sometimes unusual for their age with an atypical pattern of semantic categorization (Purser et al., 2010). However, the initial stages of development are not clearly indicative of these results.

The idea of good phonological skills in individuals with WS has been mainly consolidated from studies on phonological short-term memory by using word repetition and pseudoword tasks, suggesting that they may depend more on phonology than semantic information (Grant et al., 1997; Majerus et al., 2003), probably because of a semantic-phonological mismatch (Thomas and Karmiloff-Smith, 2003). Nonetheless, very few studies have focused on assessing this level. In this sense, the results of direct studies of production by using articulation test show that these skills are not fully preserved and that difficulties persist

into adolescence and adulthood (Hidalgo, 2019; Hidalgo and Garayzábal, 2019; Huffman, 2019).

A recent cross-sectional study explored phonological development profiles across late stages in Spanish-speaking children, adolescents, and adults with WS based on the analysis of phonological processes in spontaneous speech samples when compared to two TD groups. The results showed atypical and complex trajectories, from the expansion of the system (around 3 years of age) for the children group to its resolution (around 5 years of age) for the adolescent and adult group, which cannot be described as simply delayed or protracted (Pérez et al., 2022). Nevertheless, in late phonological development, individuals with WS reach more advanced stages than other neurodevelopmental disorders, such as Williams duplication syndrome, Smith-Magenis syndrome, Down syndrome, and Fragile X syndrome (Mervis et al., 2015; Huelmo et al., 2017; Hidalgo and Garayzábal, 2019; Diez-Itza et al., 2021).

Regarding phonological development in TD, three late stages have been described using a methodological approach based on the analysis of spontaneous speech corpora in Spanish-speaking children aged 3–5 (Diez-Itza et al., 2001; Diez-Itza and Martínez, 2004; Martínez, 2010). The results showed a reduction of the frequency of errors and changes in their relative distribution as age increased, suggesting a first stage of expansion (age 3), an intermediate stage of stabilization (age 4), and a final stage of resolution (age 5). Within the same theoretical framework, the present study aimed to further advance in a detailed description of longitudinal phonological development in children with WS.

1.1 Objectives

The main goal of this study was to determine the longitudinal profile of phonological development in a group of Spanish-speaking children with WS in order to find out changes across developmental stages and discover whether specific features would be exhibited. The profiles were based on the analysis of five error types (Syllable Structure, Segmental Substitution, Segmental Omission, Assimilation, Segmental Addition) in spontaneous speech. The frequency and percentage distribution of phonological error index were calculated for each of the two assessments. It was hypothesized that children with WS presented a lower frequency of errors from the first to the second assessment times, this reduction affecting differently in quantitative and qualitative terms. A second hypothesis was that phonological development in WS follows the stages of typical development (i.e., expansion, stabilization, and resolution) and that phonological patterns not only show quantitative differences (interpretable as “delayed”) but also, taking into account the error types, atypical characteristics (interpretable as “disordered”).

2 Method

2.1 Participants

The participants were seven individuals with WS, previously diagnosed by the molecular genetic test fluorescence *in situ*

hybridization system and presenting the characteristic physical phenotype. In addition, all participants had associated intellectual disability. The group with WS consisted of three boys and four girls (chronological age: $M = 5.9$; range: 3;07–8;02; verbal age: $M = 3.6$; range: 2;05–5;02). All participants were monolingual Spanish speakers, belonging to urban middle-class families, and attending mainstream schools ($n = 7$), and whose families provided informed consent to participate in the study. Verbal age was obtained from the Peabody Picture Vocabulary Test (Dunn et al., 2010).

The normative group consisted of 240 TD Spanish-speaking preschoolers [part of Martínez, 2010 study] aged 3–6 years divided into six groups based on chronological age and with 40 children in each group (20 girls and 20 boys): 3;0 TD: chronological age: $M = 3.2$; range: 3;00–3;05, 3;6 TD: chronological age: $M = 3.9$; range: 3;06–3;11), 4;0 TD: chronological age: $M = 4.2$; range: 4;00–4;05, 4;6 TD: chronological age: $M = 4.9$; range: 4;06–4;11, 5;0 TD: chronological age: $M = 5.2$; range: 5;00–5;05), 5;6 TD: chronological age: $M = 5.8$; range: 5;06–5;11. These children had no history of language disorder and were enrolled in regular schools distributed in the central area of Asturias (Spain).

2.2 Instruments and procedure

The RETAMHE methodology, -short for Recording, Transcription, and Analysis of Spontaneous Speech Samples (Diez-Itza, 1992; Diez-Itza et al., 1999) was used to obtain the spontaneous speech samples, which were collected via audio-visual recordings of dyadic conversations between each participant and a researcher, with an estimated duration of 45 min in natural settings, and which are part of larger corpora within the Syndroling Project (Diez-Itza et al., 2014). Individuals from the WS group were recorded in two sessions spaced 6 months apart. These conversations were transcribed in CHAT (Codes for the Human Analysis of Transcripts) format and analyzed with the FREQ program, one of the CLAN (Computerized Language Analysis) software programs, both provided by the CHILDES Project (MacWhinney, 2000). Each transcription was completed by a trained researcher and reviewed by two other researchers independently. Difficulties detected were analyzed jointly by the three investigators and discrepancies were resolved by the principal investigator.

The phonological errors were analyzed and classified into one of the following types: Syllable Structure (SYS), Segmental Substitution (SBT), Segmental Omission (OMI), Assimilation (ASM), and Segmental Addition (ADD). The following example illustrates the transcription and coding procedure according to the minCHAT format of the CHILDES Project:

CHI: nombre [] [: name].

%err: **nombre = nombre \$PHO:SYS:CCR;**

2.3 Data analysis

Once the transcriptions were coded, the frequency of lexical variables was obtained using the FREQ program, that is, the total number of words produced (“tokens”) by each participant, as well

as the count of different words (“types”) in each transcription. Next, the frequency of the classes of phonological errors encoded was obtained with the same program. In order to control for variability in the size of the spontaneous speech samples, a Phonological Error Index (PEI) was calculated to indicate the frequency of errors. This index is obtained dividing the absolute frequency of errors by the total number of words produced (tokens) per 100. In addition, the Relative Frequency (RF) was calculated, i.e., the percentage distribution of phonological errors by classes. To calculate the RF, participants in each group who did not present phonological errors in the classes or subclasses analyzed were eliminated.

Intra-group differences in PEI and RF regarding both the total number of errors and error types between the two assessment times were analyzed using the Wilcoxon-signed-rank test for dependent samples.

Additionally, the effect size was calculated by Cohen’s d using G*Power 3.1 statistical software. The d values are typically quantified as small (0.2), medium (0.5), and large (0.8) (Cohen, 1988). In turn, the differences between groups by chronological age groups in total PEI and by error types, and RF were analyzed using the Kruskal-Wallis nonparametric test adjusted with the Bonferroni correction (expressed with the H value) for independent samples, given that the distributions did not always approach normality according to the Shapiro-Wilk test. Spearman correlation was used to analyze the bivariate relationships between chronological age, verbal age, and PEI.

Statistical analysis of the data was performed using SPSS software (Statistical Product and Service Solutions IBM SPSS Statistics 25.0).

3 Results

3.1 Intra-group differences in phonological error index and relative frequency

A strong positive correlation was found between chronological age and verbal age ($r_s = 0.94$; $p = 0.002$) in the WS group. The PEI was negatively correlated with chronological age in the first assessment ($r_s = -0.74$; $p = 0.058$) and in the second assessment ($r_s = -0.72$; $p = 0.068$). Furthermore, PEI correlated negatively with verbal age at the first assessment ($r_s = -0.71$; $p = 0.071$) and at the second assessment ($r_s = -0.64$; $p = 0.012$). However, a strong positive correlation was found between the PEI at both assessments ($r_s = 0.857$; $p = 0.01$).

Table 1 reports the PEI for the WS group in the first and the second assessment, including means for total errors and each class of errors. WS children showed a mean reduction of more than 25% in the absolute frequency of phonological errors after 6 months, although this difference failed to be statistically significant. Wilcoxon comparisons showed statistically significant differences between both assessments only for SYS errors ($p = 0.018$), with a large effect size and with this type of error leading the decrease up to almost 40%. In the OMI and ASM error types there was also a decrease of 28 and 44% respectively, although no statistical differences were observed, with a medium effect size. An increase in segmental SBT and ADD errors was observed in the second

TABLE 1 Phonological error index (total and error types) means and standard deviations for WS group in the first and the second assessment times, Wilcoxon test, and effect size.

	WS1	WS2	Z	p	d
	PEI-M (SD)	PEI-M (SD)			
TOT	22.00 (18.162)	16.19 (18.006)	-1.690	0.091	0.742
SYS	13.25 (9.611)	8.131 (8.732)	-2.366	0.018	1.529
SBT	3.91 (3.886)	4.35 (54.202)	-0.169	0.866	0.130
OMI	3.22 (3.485)	2.37 (2.934)	-1.183	0.237	0.437
ASM	0.95 (1.158)	0.53 (0.543)	-0.734	0.463	0.541
ADD	0.25 (0.207)	0.44 (0.391)	-1.014	0.310	0.487

PEI-M, phonological index mean; TOT, total phonological processes index; SYS, syllable structure; SBT, substitution; OMI, omission; ASM, assimilation; ADD, addition; d, Cohen's effect size.

assessment although these differences were not statistical either, with a small and medium effect size, respectively.

The compared profiles of RF, i.e., the percentage distribution, for error types are shown for the WS group between the first and the second assessment times (Figure 1). At both times, the most frequent error types were those affecting SYS and segmental SBT. Nevertheless, the profile was different, since in the case of SYS a tendency to a reduction in the percentage from the first to the second assessments was observed, whereas in the case of SBT an increase from 16 to 25% was observed, also this difference being statistically significant ($Z = -2.197$; $p = 0.02$; $d = 1.15$). There was also a trend toward a reduction in the percentage of OMI errors and an increase in ASM and ADD errors. However, the Wilcoxon test did not yield statistically significant differences: SYS ($Z = -1.690$; $p = 0.09$; $d = 0.86$); OMI ($Z = -0.845$; $p = 0.39$; $d = 0.22$); ASM ($Z = -0.734$; $p = 0.46$; $d = 0.37$); ADD ($Z = -1.690$; $p = 0.09$; $d = 0.82$).

3.2 Inter-group differences in phonological error index and relative frequency in the first assessment

The Kruskal-Wallis test was applied to analyze whether there were differences between the WS and TD age subgroups in the Total phonological error index and by error types in the first assessment.

Significant differences were observed for all variables: PEI ($H = 80.17$; $p < 0.001$); SYS ($H = 78.80$; $p < 0.01$); SBT ($H = 67.43$; $p < 0.001$); OMI ($H = 36.21$; $p < 0.001$); ASM ($H = 34.08$; $p < 0.001$); ADD ($H = 34.08$; $p = 0.001$). Taking into account age group and after applying the Bonferroni correction, the test specifically showed that there were statistically significant differences between the WS and TD 3;6 years ($H = 59.52$; $p = 0.042$), 4;0 years ($H = 77.05$; $p = 0.008$), 4;6 years ($H = 90.42$; $p = 0.002$), 5; years ($H = 119.15$; $p = 0.001$), and 5;6 years ($H = 142.67$; $p < 0.001$) in the total PEI. In the case of error types, it was observed that in SYS there were also statistically significant differences between WS and TD 3;6 years ($H = 69.09$; $p = 0.018$), 4;0 years ($H = 87.82$; $p = 0.003$), 4;6 years ($H = 97.47$; $p = 0.001$), 5;0 years ($H = 128.29$;

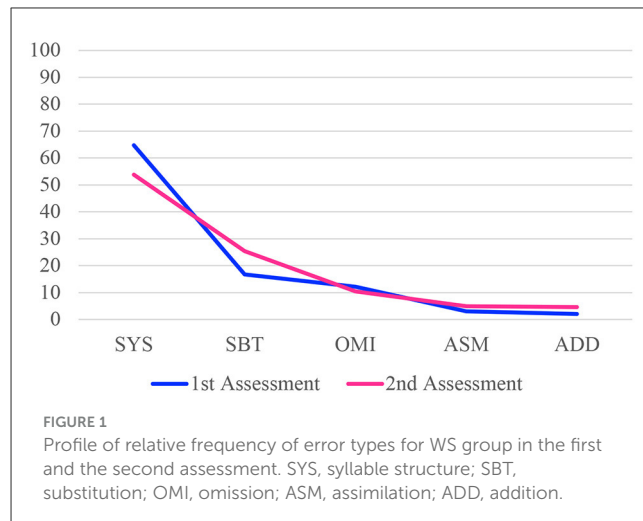


FIGURE 1 Profile of relative frequency of error types for WS group in the first and the second assessment. SYS, syllable structure; SBT, substitution; OMI, omission; ASM, assimilation; ADD, addition.

$p < 0.001$) and 5;6 years ($H = 146.04$; $p < 0.001$). For SBT these differences were observed at 4;6 years ($H = 70.80$; $p = 0.015$), at 5;0 years ($H = 101.55$; $p = 0.001$), and at 5;6 years ($H = 108.07$; $p = 0.001$). Regarding OMI, differences were observed between the WS with the group of 3;6 years ($H = 60.57$; $p = 0.034$), 4;0 years ($H = 88.60$; $p = 0.002$), 4;6 years ($H = 88.72$; $p = 0.002$), 5;0 years ($H = 78.37$; $p = 0.006$), and 5;6 years ($H = 124.42$; $p < 0.001$). Concerning ASM, these differences were found at 5;0 years ($H = 63.08$; $p = 0.028$) and 5;6 years ($H = 67.65$; $p = 0.018$), and for ADD also at 5;0 years ($H = 66.93$; $p = 0.048$) and 5;6 years ($H = 67.93$; $p = 0.019$).

To assess differences in relative frequency of phonological error index by types (Figure 2), the Kruskal-Wallis test was also applied. Statistically significant differences were only observed in terms of relative frequency for segmental OM between the WS and TD 4;0 years ($H = 66.21$; $p = 0.020$), 4;6 years ($H = 63.65$; $p = 0.026$), and 5;6 years ($H = 72.80$; $p = 0.005$).

3.3 Inter-group differences in phonological error index and relative frequency in the second assessment

The Kruskal-Wallis test was applied to analyze whether there were differences between the WS and TD age subgroups for the Total phonological error index and by error types in the second assessment. Significant differences were observed in the variables: PEI ($H = 73.91$; $p < 0.001$); SYS ($H = 71.63$; $p < 0.001$); SBT ($H = 65.07$; $p < 0.001$); OMI ($H = 31.78$; $p < 0.001$); ASM ($H = 35.96$; $p < 0.001$); ADD ($H = 23.57$; $p = 0.001$). Focusing on age group and after applying the Bonferroni correction, the test specifically showed that there were only statistically significant differences between WS and TD 5;0 years ($H = 88.42$; $p = 0.005$) and 5;6 years ($H = 106.21$; $p < 0.001$) in the total PEI. As for error types, statistically significant differences were observed for SYS and TD 5;0 years ($H = 85.86$; $p = 0.003$), and 5;6 years ($H = 103.96$; $p < 0.001$), for SBT and TD 4;6 years ($H = 57.27$; $p = 0.05$), 5;0 years ($H = 88.25$; $p = 0.003$), and 5;6 years ($H = 104.60$; $p < 0.001$), in OMI

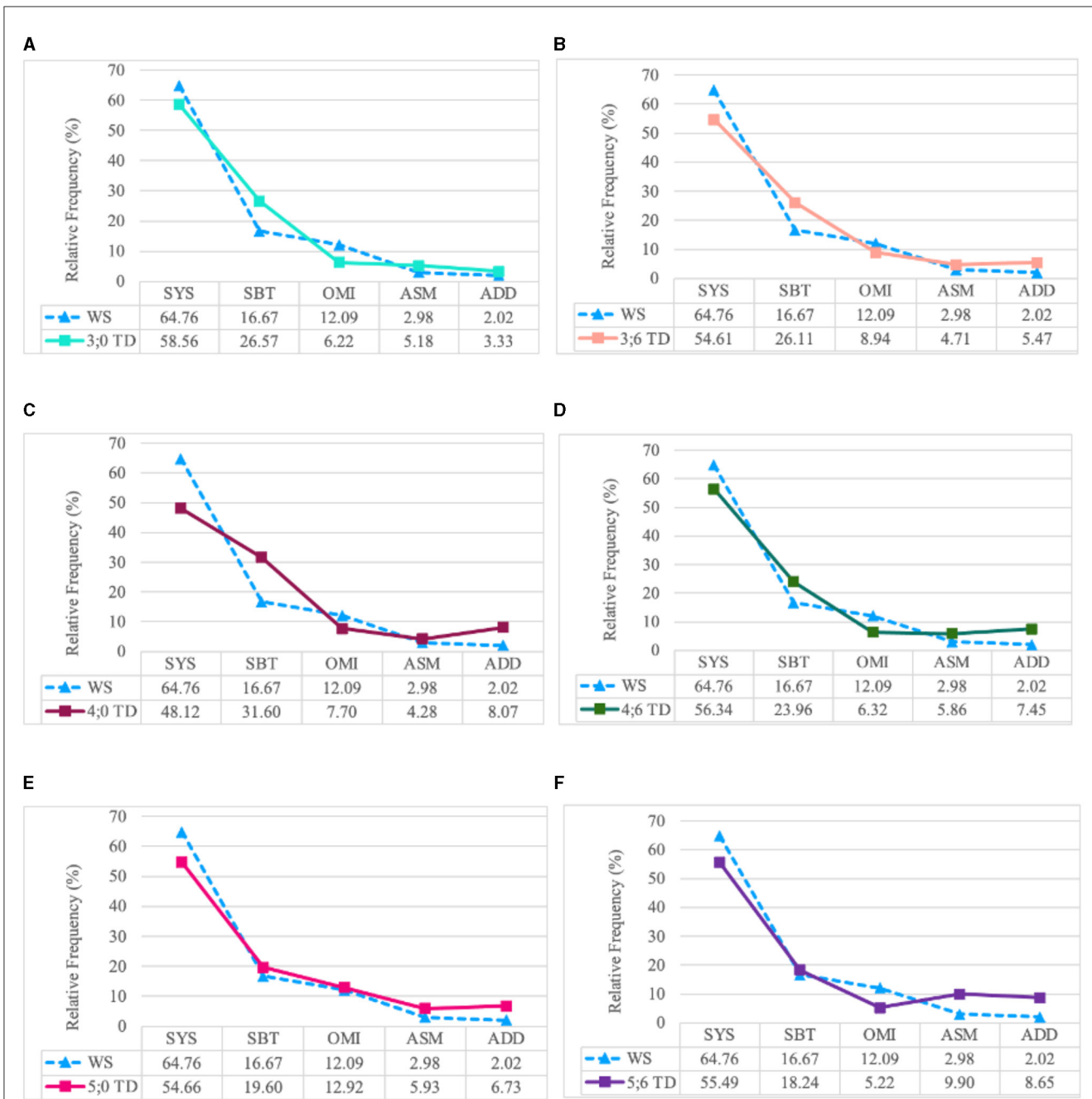


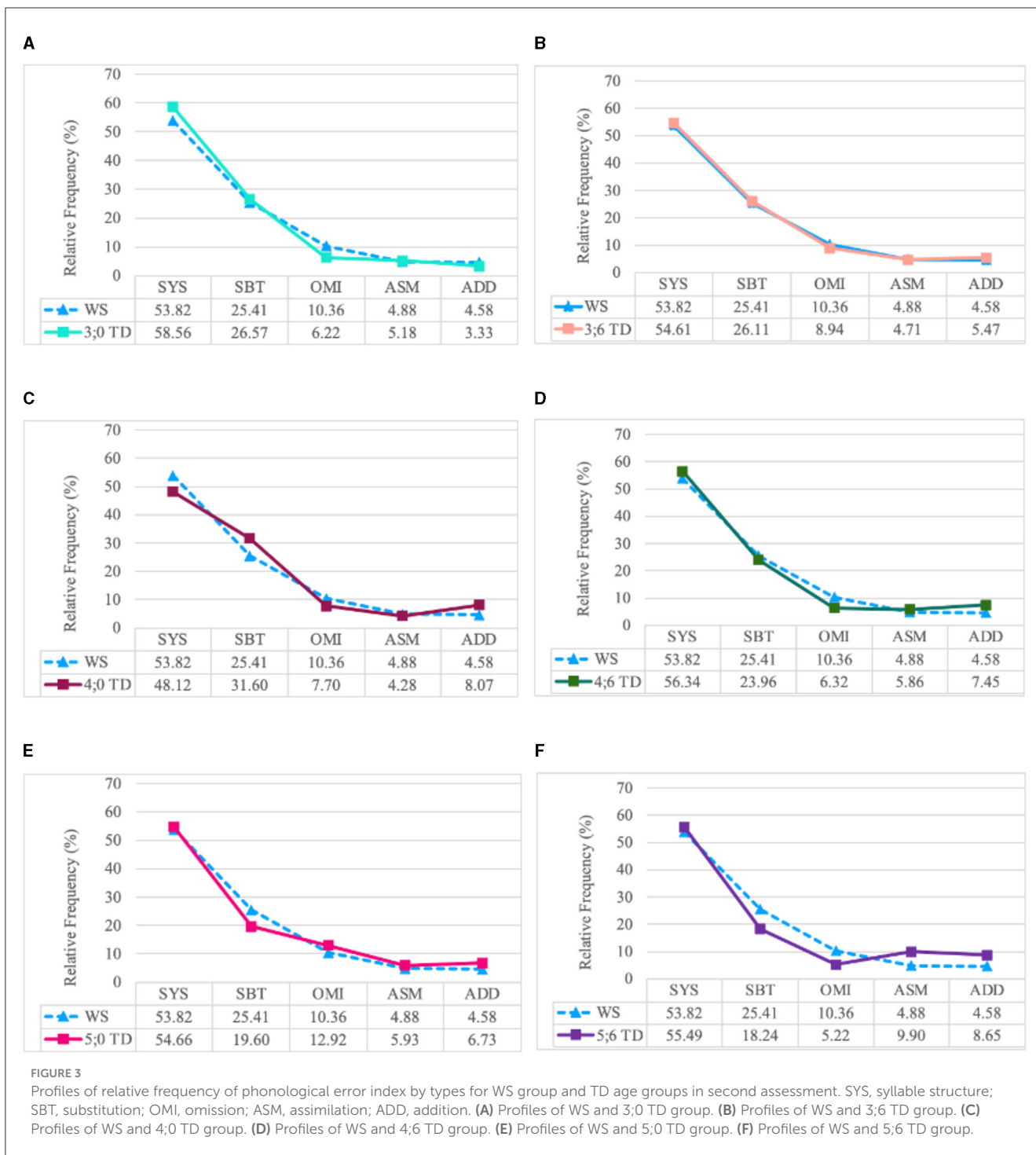
FIGURE 2 Profiles of relative frequency of phonological error index by types for WS group and TD age groups in first assessment. SYS, syllable structure; SBT, substitution; OMI, omission; ASM, assimilation; ADD, addition. (A) Profiles of WS and 3;0 TD group. (B) Profiles of WS and 3;6 TD group. (C) Profiles of WS and 4;0 TD group. (D) Profiles of WS and 4;6 TD group. (E) Profiles of WS and 5;0 TD group. (F) Profiles of WS and 5;6 TD group.

and TD 4;0 years ($H = 65.68; p = 0.021$), 4;6 years ($H = 65.79; p = 0.021$), and 5;6 years ($H = 101.52; p < 0.001$), in ASM and TD 5;0 years ($H = 73.39; p = 0.011$) and 5;6 years ($H = 78.02; p = 0.007$), and in ADD and TD 4;0 years ($H = 56.92; p = 0.049$), 4;6 years ($H = 61.85; p = 0.032$), 5;0 years ($H = 69.92; p = 0.015$), and 5;6 years ($H = 80.77; p = 0.005$).

To assess differences in relative frequency of phonological error index by types (Figure 3), the Kruskal-Wallis test was also applied. Statistically significant differences were only observed in terms of relative frequency in segmental Omissions between the WS group and the 5;6 TD group ($H = 66.45; p = 0.020$).

4 Discussion

The purpose of this study was to determine the longitudinal profile of phonological development in a group of Spanish-speaking WS children in order to find out changes across developmental stages and whether specific features would be exhibited. Profiles were based on five error types (Syllable Structure, Segmental Substitution, Segmental Omission, Assimilation, Segmental Addition) in spontaneous speech, calculating their PEI (frequency of errors/100 tokens) and their RF (percentage distribution) for each of both assessments within



a 6-month interval. To determine if phonological development in WS followed the stages of typical development (i.e., expansion, stabilization, and resolution) and if they presented specific characteristics, not only quantitative differences (interpretable as delayed) but also atypical characteristics (interpretable as disordered), they were also compared with the profiles of TD preschool children of similar verbal age.

Our results showed that, although as chronological and verbal ages of WS children increased, the PEI decreased, but this reduction was not statistically significant for neither assessment. Taking into

account that phonological development in TD children culminates at the age of 7 years (Bosch-Galcerán, 2004), this lack of significance between chronological age and PEI could be explained by age differences, as there were two children aged 3 and 4 years and other two over 7 years of age.

WS children showing a high frequency of phonological errors in terms of PEI in the first assessment were those who continued to present greater PEI in the second one. However, the PEI was reduced by 25% within a 6-month interval, indicating that late phonological development was in progress. The tendency for

phonological errors to markedly decrease over chronological age in WS children suggests that accelerated phonological development occurs, which is consistent with findings previously reported by Martínez et al. (2014) in two WS children. This accelerated rate of phonological development over a 6-month interval would compensate for the delay in language onset, which has been linked to delayed babbling (Masataka, 2001) and auditory-visual integration difficulties observed in young WS children and in other neurodevelopmental syndromes (D'Souza et al., 2015). Despite the PEI reduction in WS children within a 6-month interval, phonological development does not seem to culminate at these ages since WS adolescents and adults, as occurs in other neurodevelopmental disorders such as Down syndrome, Fragile X syndrome, or Smith-Magenis syndrome, continue to manifest phonological difficulties (Huelmo et al., 2017; Hidalgo and Garayzábal, 2019; Diez-Itza et al., 2021; Pérez et al., 2022).

Taking into account the error types, it was observed that WS children showed a higher frequency in SYS followed by SBT in both assessments, which is consistent with previous research in English-speaking WS children and adolescents (Huffman, 2019) and in Spanish-speaking WS children, adolescents, and adults (Hidalgo and Garayzábal, 2019; Pérez et al., 2022). This tendency has also been observed for TD (Bosch-Galcerán, 2004; Martínez, 2010) and in other neurodevelopmental genetic disorders (Barnes et al., 2009; Huelmo et al., 2017; Hidalgo and Garayzábal, 2019; Diez-Itza et al., 2021). Nevertheless, only for errors affecting SYS was a significant reduction observed after 6 months, since SBT segmental errors increased in frequency in the second assessment, a pattern also observed in TD at around 4 years of age (Diez-Itza and Martínez, 2004). It was also observed that the frequency of OMI and ASM segmental errors decreased in the second assessment. However, it was found that OMI errors continued to present a high frequency in WS adolescents and adults (Pérez et al., 2022). In the case of ASM errors, it has been observed that they were still present at ages 6 and 7 in WS (Hidalgo and Garayzábal, 2019), although with a lower incidence. The same occurs in TD (Martínez and Diez-Itza, 2012) where ASM errors have been considered representative of the late phonology of Spanish with a significant percentage at 7 years old (Bosch-Galcerán, 2004).

As for the profile of relative frequency of error types in the first and second assessment, it was observed that, as in absolute terms, the most frequent errors were those of SYS, STB, OMI, ASM, and ADD and this was similar to that observed in TD children of similar verbal age (Martínez, 2010) and DS children and adolescents in spontaneous speech (Diez-Itza et al., 2021). However, the intersections between the relative frequency profiles in WS children might suggest that the trajectories from the first to the second assessment was toward reducing the proportion of SYS and OMI and increasing the proportion of SBT, ASM and ADD errors, although only in the case of SBT this increase was statistically significant. This would suggest that in the second assessment there was a reconversion of the phonological system in relation to SBT segmental errors similar to that observed at the age of 4;6 years for TD (Diez-Itza and Martínez, 2004; Martínez, 2010).

When comparing WS children in the first assessment with the age groups of the normative group, it was observed that WS presented higher PEI than every normative group, except

for the 3;0 TD group. These results would indicate that initially their phonological error index was analogous to that of the expansion stage, corresponding to ages 3;0–3;6 in TD. However, 6 months later, the frequency of the error index was significantly reduced and could be equated with the 4;6 year-old group, thus consistent with the stage of stabilization (ages 4;0–4;6 in TD), therefore showing an accelerated phonological development as previous studies had suggested (Martínez et al., 2014). There appeared to be dynamic development over the 6-month interval as WS children moved from one stage to another. Such atypical acceleration might be related to lexical growth, given the close relationship between lexical and phonological development, and their relatively preserved phonological memory (Majerus et al., 2003; Mervis et al., 2004; Stoel-Gammon, 2011), which would show the interdependence of the processes as well as the dynamic nature of linguistic development (Mareschal et al., 2007).

Concerning error types, the developmental pattern was different in both assessments. Thus, while WS children moved from the expansion stage to stabilization for SYS errors, showing a strongly accelerated growth rate of the phonological system, in the case of SBT, ASM, and ADD errors these children would be in the stabilization stage whereas for OMI segmental errors they would be in the expansion stage, although a reduction in their phonological index was observed in the 6-month interval. This contrast in evolution would suggest a slowdown in the growth rate of the phonological system of WS children, which could be interpreted in terms of delayed phonological acquisition (Pérez et al., 2022).

The study of the relative frequencies of error types showed that in the first assessment the profile was not comparable to that of children aged 3;0 because its frequency is higher for all types. However, in the second assessment the profile overlaps with that of children aged 3;6 years. In relative terms, these WS children would be in the expansion stage at both times although there would be certain progress in their phonological development. On the other hand, the high relative frequency of OMI at both assessment times may be considered atypical and specific to WS since TD children aged 4;0 years no longer produce this type of error with only between 20 and 30% of children showing absence of multiple vibrating/r/ (Bosch-Galcerán, 2004; Diez-Itza et al., 2005). This was confirmed in our previous study where children, adolescents and adults showed a high frequency of vowel omission and liquid consonant omissions compared to the 5-year-old TD normative group therefore suggesting a deviant developmental trajectory (Pérez et al., 2022).

In conclusion, the results of the present study seem to confirm that the frequency of phonological errors in WS children decreases over a 6-month interval, showing an atypical acceleration. Moreover, the trajectories of late phonological development in WS children may not be linear, but dynamic as postulated by neuroconstructivist models since in a short period of time they move from the expansion stage (age 3) to the stabilization stage (age 4), perhaps favored by its interrelation with other components at different levels such as the lexicon (Mervis et al., 2004; Stoel-Gammon, 2011). Although the results are not conclusive on delayed vs. disordered phonological profiles, highly increased frequency of errors at the two time points assessed asynchronous

with verbal age, suggests atypical developmental trajectories of phonological development in the WS children. The description of the detailed longitudinal phonological profile results in a better understanding of the syndrome as well as improved effectiveness of assessments and speech therapy intervention.

The shortcomings of this study stem mainly from the absence of controlled individual differences that could explain significant percentages of the variance observed in WS children. A larger number of participants would have been necessary to minimize these differences and make comparisons by age groups in WS. However, this is a small-scale exploratory study and confidence in the conclusions drawn from the results is reinforced by the large effect size for total errors and for errors affecting SYS. Further study would be necessary to assess the specific features and errors for each of the five types studied.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The study was conducted in accordance with the Declaration of Helsinki and approved by the Ethics Committee of the University of Oviedo for studies involving humans (Approval code 6_RRI_2022 and Approval date 05/10/2022). The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin. Written informed consent was obtained from the minor(s)' legal guardian/next of kin for the publication of any potentially identifiable images or data included in this article.

Author contributions

VM: Conceptualization, Data curation, Formal analysis, Investigation, Supervision, Writing – original draft,

Writing – review & editing, Methodology. VP: Conceptualization, Data curation, Writing – original draft, Writing – review & editing, Investigation. MAA: Investigation, Methodology, Writing – review & editing. MM: Investigation, Methodology, Writing – review & editing. PV: Data curation, Investigation, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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