NONWORD REPETITION TASK PERFORMANCE IN PRESCHOOLERS WHO STUTTER

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ABSTRACT

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Research reveals that children who stutter (CWS) complete nonword repetition tasks (NRTs) with poorer accuracy compared to children who do not stutter (CWNS). The primary aim of this study was to examine the contribution of phonological abilities to NRT performance accuracy in CWS. The second aim of this study was to assess error characteristics, specifically the location of errors on the longest nonword, to determine if error profiles differentiated children in each of the groups. We examined NRT scores from 54 preschoolers (35 CWS and 19 CWNS). CWS were then placed into different groups based on phonological abilities: CWS with speech sound disorders (CWS+SSD) and CWS with typical speech sounds (CWS+TSS). On average, CWS+SSD had significantly higher error rates than CWS+TSS and CWNS on each of the four nonwords. Children in all groups made the highest number of errors on the 4-syllable nonwords. Errors committed on all four syllables of NW 4 predicted membership into CWS+SSD group, while error rates on the first syllable also predicted membership into CWS+TSS group. Comparing performance among all groups revealed that having a concomitant SSD played a significant role in poorer NRT performance in the subgroup of CWS. Errors committed on the 4-syllable nonwords also revealed subtle differences in phonological working memory strategies between CWS and CWNS. Taken together, these findings help shed light on poorer NRT performance in stuttering and enhance generalization of findings to populations of CWS with a range of phonological abilities.

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Introduction

Stuttering, a speech disorder affecting approximately 1% of the population, is characterized by overt and covert characteristics (Bloodstein & Bernstein-Ratner, 2008). Overt symptoms of stuttering include speech disfluencies or involuntary disruptions in the flow of speech. These may be classified as part-word repetitions ("I see a b-b-baby!"), or whole single-syllable word repetitions ("You...you...you...go next"), sound distortions and prolongations ("I ate a ssssssssandwhich for dinner"), and/or blockages of air flow ("Do you see the c----at?") (Yairi & Ambrose, 2005). Observable symptoms may also include physiological concomitants, such as rapid blinking, facial grimacing, and extremity movements (Bloodstein & Bernstein-Ratner, 2008).

Stuttering also consists of covert symptoms, which could be related to anxiety, depression, avoidance, frustration, and shame or guilt regarding their speech (Douglass et al., 2018). Stated differently, stuttering affects more than just a person's speech output. For example, adults who stutter may be more likely to suffer from anxiety disorders (Craig & Tran, 2014; Davis et al., 2007; Iverach et al., 2009). Stuttering impacts psychosocial development in some children resulting in long-lasting repercussions (Beilby, 2014).

As many as 5-11% of the preschool population go through a period of stuttering; however, most will spontaneously recover with or without fluency intervention (Yairi & Ambrose, 2005; Reilly et al., 2013). After approximately one year of stuttering, however, the chance of recovery decreases (Yairi & Ambrose, 2005). Stuttering emerges in early childhood, generally between the ages of two and five years, when speech and language skills are being learned and expanded (Kloth et al., 1999). Because stuttering emerges during significant speech and language development, there have been a number of studies examining potential

relationships between the onset of stuttering and language development (Bauman et al., 2012; Bloodstein & Bernstein-Ratner, 2008; Ntourou et al., 2011; Yairi & Ambrose, 2005). In particular, links between disordered phonology and stuttering have been suggested (Arndt & Healey, 2001; Blood et al., 2003; Louko et al., 1990; Paden et al., 1999; Spencer & Weber-Fox, 2014; Wolk et al., 1990; Yaruss et al., 1998; cf. Nippold, 2002). It is unclear, however, whether disordered phonology is associated with stuttering or occurs comorbidly.

Nonword repetition tasks (NRTs) have been used to assess certain aspects of phonological abilities in children who stutter (CWS), more specifically, phonological encoding, storage/phonological working memory, and phonological execution processes (Coady & Evans, 2008). Overall, results show less accurate performance achieved by CWS compared to children who do not stutter (CWNS), particularly during the production of longer nonwords (NWs) (Anderson et al., 2006; Anderson & Wagovich, 2010; Hakim & Bernstein-Ratner, 2004; Pelczarski & Yaruss, 2016). Given the phonological skills inherent in the NRT task, it is important to consider the comorbidity of stuttering and phonological disorders to understand whether poorer nonword production accuracy in CWS is related to the presence of an underlying speech sound disorder (SSD) in some of the population. Furthermore, to our knowledge, there has been no research looking at where error breakdowns occur during performance by CWS on NRTs to clarify their poorer performance. Therefore, the purpose of this study is to shed light on potential underlying factors leading to poorer nonword production performance. Our first aim is to assess how differences in phonological abilities affect NRT performance in CWS with and without SSD. Our second aim is to determine if NRT error characteristics, specifically the location of errors on NW 4 can be used to differentiate performance among participant groups.

Literature Review

Stuttering Etiology

Stuttering is classified by the DSM-V (American Psychiatric Association, 2013) as a neurodevelopmental disorder; it arises from atypical development in the central nervous system circuitry (Chang et al., 2008). For example, CWS exhibited reduced fractional anisotropy, or water diffusivity as an index of white matter organization, in tracts in the brain compared to their typically developing peers (Chang et al., 2015). Another study examined the gray matter development of frontal regions, including Broca's area, and found differences in maturational patterns between groups of children and adults who do and do not stutter (Beal et al., 2015). There is also evidence to suggest a genetic component. Although it remains unclear how stuttering is inherited, a child who stutters is more likely to have a relative who stutters than a child who does not stutter (Yairi & Ambrose, 2005).

Theoretical Underpinnings of Stuttering

Although genetic and neurophysiological factors likely contribute to stuttering, there are diverse causal theories of stuttering. For example, some theories fall under a physiological category, while others fall under psychological/learned theories. Physiological theories focus on the physiology of speech-related brain regions. Nearly 90 years ago, Travis (1931) proposed the Cerebral Hemispheric Dominance theory that posits an imbalance of control between the two cerebral hemispheres of the brain (Travis, 1931). A more modern version of this theory examined the idea that rather than an imbalance of control between the two hemispheres, the right hemisphere is over activating compared to the left. More specifically, there is more activation in the right frontal lobe in persons who stutter compared to people who do not stutter

to compensate for deficient speech networks located in the left hemisphere (Watkins et al., 2007).

On the other hand, learning theories, or psycho-behavioral/emotional theories, propose that learned behaviors may cause speech disruptions in CWS. For example, in the Conflict theory (Sheehan, 1953) the underlying cause of stuttering is fear/anxiety stemming from learned fear of certain words or speaking situations that creates a conflict for CWS who want to speak but wish to avoid stuttering. This is thought to result in a disruption of speech (Sheehan, 1953).

Relevant to the current study, psycholinguistic accounts of stuttering maintain that stuttering is a downstream result of weakened "upstream" encoding processes of the syntactic, lexical, phonological, or suprasegmental goals of speech production. The Covert Repair Hypothesis explains that stuttering is the result of excessive numbers of errors in the speaker's phonetic plan (Postma & Kolk, 1993; Kolk & Postma, 1997). The EXPLAN model combines theories regarding the base of planning difficulty (phonetic or lexical complexity) with an atypical synchrony in the planning and execution of speech and language (Howell, 2004). Empirical evidence shows that CWS do have a higher probability of developing a language and/or phonological disorder (Arndt & Healey, 2001; Paden et al., 1999; Bloodstein & Bernstein-Ratner, 2008; Louko et al., 1990; Pelczarski & Yaruss, 2014; cf. Nippold, 2002).

Many researchers and clinicians, however, view stuttering as a multifactorial disorder, meaning that there is not a single cause underlying this complex disorder. (Smith & Kelly, 1997; Smith & Weber, 2017; Adams, 1990; Starkweather and Gottwald, 1990). Rather, stuttering results from the dynamic interactions among multiple factors. For example, the Multifactorial Dynamic Pathways theory offers a way to explain the different variables that may contribute to stuttering. Even slight differences in a given factor for a child may have significant

"downstream" effects that ultimately cause that child to stutter (Smith & Kelly, 1997; Smith & Weber, 2017). Related to the current study, the onset of stuttering coincides with the rapid development of speech/language skills and phonological abilities. For example, in some CWS, deficient phonological processing may interact with speech motor networks that are developmentally delayed resulting in breakdowns in speech fluency (Smith & Weber, 2017).

Phonological Abilities of Children Who Stutter

There are different pieces of evidence implicating atypical phonological abilities in children who stutter. For example, Louko et al. (1990) found that in a group of 30 CWS, 40% of the children exhibited 18 different phonological processes, compared to their typically developing peers who only exhibited 11 processes, a significant difference. Some of these atypical phonological processes included glottal replacement, backing, and lateralization (Louko, et al., 1990).

In a longitudinal study of early childhood stuttering, Paden, et al. (1999) compared phonological abilities with the Assessment of Phonological Processes – Revised (Hodson, 1986) between groups of children whose stuttering had persisted (n = 22) with those who would eventually recover from stuttering (n = 62). These phonological scores were recorded soon after stuttering onset for both groups. After a period of time when it could be determined which children would persist and which would recover, they found that the children who persisted in stuttering performed more poorly on this assessment than the group who would later recover (Paden et al., 1999).

There is also evidence suggesting that some CWS have less accurate phonological encoding abilities, defined as the ability to process verbal language, and convert it to

words/sentences, than their typically developing peers. Pelczarski and Yaruss (2014) compared phonological awareness abilities of 10 CWS and 10 age matched CWNS. Results showed that the CWS performed significantly less accurate on elision (the omission of a sound or syllable) and sound blending tasks (blending two or more sounds to create a word) compared to the CWNS (Pelczarski & Yaruss, 2014).

Nonword Production Performance by Children who Stutter.

Finally, there is empirical evidence suggesting that CWS generally perform more poorly on phonological NRTs compared to typically developing peers (Anderson et al., 2006; Hakim & Bernstein-Ratner, 2004; cf. Bakhtiar et al., 2007). NRTs were originally designed to combat the biases of normative-referenced tests when assessing children with developmental language disorders. Often, norm-referenced language assessments depend on the test-taker's familiarity with the syntax and vocabulary of the language being assessed. NRTs tap into phonological working memory and phonological encoding/execution abilities. Phonological codes are held in working memory and NRTs are thought to measure how well someone can maintain and retrieve the codes to produce the nonwords (Archibald & Gathercole, 2007, Dollaghan & Campbell, 1998). An advantage of nonword repetition tasks is that they help pinpoint potential phonological deficiencies.

A meta-analysis conducted by Ofoe, et al. (2018) compared scores on NRTs between CWS and CWNS and found that CWS scored more than half a standard deviation below the average scores of the control group. They noted that, "the average participant in the CWNS group scored higher on verbal short-term memory (VSTM) measures than 73% of the CWS group" (Ofoe et al., 2018, pp. 1633). Performance decreased with increased nonword length

(Ofoe et al., 2018). Several studies from this meta-analysis are highlighted in the following paragraphs.

Anderson et al. (2006) assessed the nonword production skills of 12 3- to 5-year-old CWS and 12 age-matched CWNS using the Children's Test of Nonword Repetition (Gathercole et al., 1994). Results showed that the CWS performed with reduced accuracy on two and three-word syllable NWs compared to their typically fluent peers. Both groups of children performed with reduced accuracy on the longer, four- and five-syllable nonwords. Overall, the CWS performed, on average, with lower accuracy, than their typically fluent peers. Anderson and Wagovich (2010) conducted a follow-up study of phonological working memory and attention in preschool CWS using the same assessment, the Children's Test of Nonword Repetition (Gathercole et al., 1994). They replicated their initial study documenting significantly poorer performance on this NRT in 9 CWS and 14 CWNS (Anderson & Wagovich, 2010). Note that in the follow-up study, the group utilized a portion of the children from the first study combined with children from another study assessing repetition priming (Anderson & Wagovich, 2010).

Another study conducted by Hakim and Bernstein-Ratner (2004) assessed the language abilities of 8 CWS and 8 CWNS between the ages of 4-8 years including their nonword repetition abilities with the Children's Test of Nonword Repetition (Gathercole et al., 1994).

Hakim & Bernstein-Ratner (2004) altered the lexical stress of the nonwords to resemble a non-English stress pattern—as emphasis was consistently placed on the last syllable of the stimuli.

The role that syllabic stress plays in stuttering is unclear, although evidence shows that children stutter more commonly on stressed syllables compared to unstressed (Natke et al., 2004). They found that all children performed more poorly when presented with the altered stimuli compared to the unaltered stimuli. However, the CWS still performed less accurately than the control group

overall. Contrary to their expectations, Hakim and Bernstein-Ratner (2004) did not find that the altered stimuli resulted in more disfluencies (stuttering) in the group of CWS compared with the stimuli that conformed to English stress patterns.

Pelczarski and Yaruss (2016) examined the phonological memory skills of 11 5- to 6-year-old CWS and 11 CWNS matched for age, sex, general language skills, and socio-economic status. Pelczarski and Yaruss compared performance on NRTs, digit span task (which measures the capacity of a person's phonological working memory), expressive and receptive vocabulary measures (Expressive Vocabulary Test; EVT; Williams, 1997; Peabody Picture Vocabulary Test - III; PPVT-III; Dunn & Dunn, 1997), and a standardized articulation assessment, the Goldman-Fristoe Test of Articulation (GFTA-2; Goldman & Fristoe, 2000). The two groups performed similarly on the assessments of vocabulary and articulation. No significant differences were found between groups on the digit span tasks as well, suggesting memory capacity for this task was similar between groups of CWS and CWNS. However, the CWS performed more poorly on the NRTs compared to the control group (Pelczarski & Yaruss, 2016).

Taken together, empirical evidence reveals that CWS tend to perform more poorly on NRTs compared to typically developing peers, with some evidence showing that phonological abilities may aid in predicting children at higher risk for persistence of stuttering. In a longitudinal prospective study, Spencer and Weber-Fox (2014) administered two measures of phonological abilities, the Bankson-Bernthal Test of Phonology (BBTOP), a standardized measure that looks at phonological and articulatory ability (Bankson & Bernthal, 1990), and nonword repetition performance (Dollaghan & Campbell, 1998) to preschool children diagnosed with early childhood stuttering. They found that children who would eventually persist

performed with significantly reduced accuracy on the NRT and the BBTOP compared to children who would eventually recover from stuttering (Spencer & Weber-Fox, 2014).

In summary, there is ample evidence suggesting that CWS generally perform more poorly on NRTs and that accuracy decreases with increases in nonword length. There has been no research, however, examining performance on NRTs in groups of CWS with and without SSD nor on the characteristics of the errors produced during these tasks. Examining whether having a concomitant SSD impacts NRT performance and about the nature of errors committed during these tasks may help shed light on the underlying factors leading some CWS to perform more poorly on NRTs. Toward this aim, we examined where errors occurred on the longest, 4-syllable nonword (i.e., on the first, second, third, or fourth syllable).

In many studies, groups of CWS and CWNS were matched for speech sound production abilities using standardized assessments that focus exclusively on articulation abilities such as the GFTA-2 or articulation and phonological abilities using the BBTOP. Given that a significant number of CWS present with concomitant phonological and/or articulation disorders (Blood et al., 2003; Wolk et al., 1990), we elected to specifically examine NWR performance in groups of CWS with and without phonological and articulation disorders, henceforth, speech sound disorders (SSD) to enhance the generalization of findings to the population of CWS at large. Children with SSD also perform more poorly on NRTs (Munsen et al., 2005; Pigdon et al., 2020; Martikainen et al., 2020; Archibald & Gathercole, 2007; Vuolo & Goffman, 2020). Thus, it is important to carefully account for children's phonological abilities as these abilities may affect performance on nonword production tasks. Anderson et al. (2006) found significant correlations between GFTA-2 scores and nonword repetition accuracy in their cohort of preschool CWS (without SSDs) but not in CWNS. In the one study that grouped participants based on their

phonological abilities, findings revealed that CWS with a concomitant phonological disorder produced significantly more errors on these NRTs than both the CWNS and CWS without a concomitant phonological disorder (Smith et al., 2012). However, the sample size of the group with concomitant phonological disorder was small at only 6 participants. This is a limitation of many of the NRT studies in CWS with modest sample sizes ranging from 8-12 participants in each group. Given the considerable heterogeneity in pediatric populations, larger sample sizes will allow us to detect differences among our groups of children should these differences exist.

Current Study

The overarching goal of this study is to conduct a comprehensive analysis of nonword production performance in a large cohort of preschoolers who stutter to shed light on poorer nonword production performance in CWS. We will fulfill this goal through two aims: The first aim is to clarify the potential role that phonological abilities play in poorer NRT performance in CWS. We will achieve this aim by comparing the frequency of errors during nonword production between three groups: children who stutter with typical speech sound production abilities (CWS+TSS), children who stutter with speech sound disorder (CWS+SSD), and children who do not stutter or have SSD (CWNS). Our second aim is to determine whether error characteristics, specifically the location of errors differentiate children in the three participant groups. We examined this for the longest and most complex nonword (NW 4). This is the nonword on which CWS and CWNS made the highest number of errors in our earlier studies (Spencer & Weber-Fox, 2014; Smith et al., 2012).

Given findings from previous studies (e.g., Anderson et al., 2006; Hakim & Bernstein-Ratner, 2004), we hypothesize that overall, CWS in both groups will produce a significantly higher number of errors overall on the NRT compared to CWNS. We further hypothesize that the

presence of an SSD will affect performance such that CWS+SSD will make significantly more errors compared to CWS+TSS and CWNS (Smith et al., 2012). Finally, we hypothesize that differences will emerge between groups CWS+TSS and CWNS when the difficulty of the task increases. Thus, CWS+TSS will perform more poorly on the longest, most challenging NW, NW 4, compared to CWNS (Anderson et al., 2006; Hakim & Bernstein-Ratner, 2004).

Methods

Participants

Fifty-four preschoolers, 35 CWS (26 males, 9 females) and 19 CWNS, (14 males, 5 females) participated in this study. The participants were between the ages of 3;9 and 6;0 (CWS M = 4;5, CWNS M = 4;6). All participants spoke North American English as their native language. These children were part of a larger longitudinal study by the Purdue Stuttering Project examining the development of stuttering in young children. The research protocol was approved by the Institutional Review Board at Purdue University.

All preschoolers passed a standard hearing screening at 20dB at 500, 1000, 2000, 4000 and 6000 Hz. Each participant had normal or corrected-to-normal vision, no history of neurological problems, and were not taking medication expected to affect the central nervous system per parent report. There was no indication of impaired reciprocal interaction and restriction of activities in these children (autism spectrum disorder), as assessed by the Childhood Autism Rating Scale, 2nd edition (Schopler et al., 2010). All participants scored within normal limits on the Primary Test of Nonverbal Intelligence (Ehrler & McGhee, 2008). In addition, all participants passed a screening of the oral-motor mechanism to ensure they did not have a motor-speech disorder (Robbins & Klee, 1987)

Stuttering Diagnosis

Children were diagnosed as stuttering using the following criteria established by Paden et al. (1999): (1) the child was regarded as stuttering by the parent/caregiver (2) the child was regarded as stuttering by the project speech-language pathologist (SLP) experienced in childhood stuttering, (3) the child's stuttering severity rating was 2 or higher on an 8 point severity scale by

either a parent/caregiver or the project SLP (with 0-1 indicating typical fluency, 2-3 indicating mild stuttering, 4-5 indicating moderate stuttering, and 6 or higher indicating severe stuttering), (4) the child exhibited at least three stuttering-like disfluencies per 100 syllables of spontaneous speech. Spontaneous speech samples were collected from each child during two play-based sessions: One with the parent and one with the SLP. Speech samples were analyzed and transcribed by the trained graduate clinician then confirmed by the SLP. Any discrepancies between the transcripts were resolved by consensus (i.e. consensus reached while the pair listened to the speech sample together).

Phonological and Language Assessments

As a part of a comprehensive speech and language assessment battery, each child was given the Structured Photographic Expressive Language Test 3rd Edition, a standardized measure assessing morphology and syntax abilities (SPELT-3; Dawson et al., 2003), as well as the Clinical Evaluation of Language Fundamentals 2nd Edition (CELF-2; Semel et al., 2004), a standardized measure of expressive and receptive language abilities. Each child was also given the BBTOP (Bankson & Bernthal, 1990). The BBTOP is as assessment of children's articulation and phonology administered to children aged 3-9 years. The phonology portion comprises analyses of the 10 the most frequently occurring phonological processes. These include assimilation, fronting, final consonant deletion, weak syllable deletion, stopping, gliding, cluster simplification, depalatalization, deaffrication, and vocalization. BBTOP produces three diagnostic inventories: the word inventory (the number of words produced without any consonant misarticulation), the consonant inventory (the number of consonant phonemes/clusters in error), and the phonological process inventory (the number of errors reflecting one of the 10 processes assessed by the BBTOP). A standard score of less than 85 on any of the three sections

is considered a "no pass." All CWNS achieved scores within normative limits on all speech and language standardized testing, indicating that no CWNS had a concomitant speech or language disorder. CWS were divided into groups based on their speech production abilities measured by the BBTOP. There were 17 CWS who did not pass the BBTOP, and therefore were placed into a group with CWS+SSD. Children who stutter without an SSD, who passed the BBTOP, are referred to as CWS+TSS (see Table 1).

Table 1: Average scores for participants in all three groups on the BBTOP subtests, the SPELT-3, and the CELF-2.

Group	N	Age in Months M (SD)	BBTOP- CI M (SD)	BBTOP- PPI M (SD)	SPELT-3 M (SD)	CELF-2 M (SD)
A – CWS+TSS	18	54.8 (7.0)	105.2 (9.3)	107.7 (9.7)	101.8 (7.9)	102.1 (10.3)
B - CWS+SSD	17	57.7 (7.0)	78.7 (9.8)	76.3 (8.4)	86.3 (16.7)	90.4 (16.7)
C - CWNS	19	54.6 (5.9)	110.3 (8.6)	110.1 (9.1)	109.6 (9.1)	110.5 (9.8)

Note: 1 child in group A did not pass the SPELT and CELF; 5 children in group B did not pass the SPELT and CELF; 2 additional children in group B did not pass the SPELT; 1 additional child in group B did not pass the CELF.

Experimental Stimuli and Procedures

All children completed the NRT (Dollaghan & Campbell, 1998) to assess his/her nonword repetition skills. The nonword stimuli were recorded by a native English-speaking adult female and were presented through two speakers, one on each side of the child. Following instructions from Dollaghan & Campbell (1998), all of the one-syllable NWs were presented first, followed by the two-syllable, three-syllable, then finally four-syllable NW. This entire task took less than ten minutes to administer. The directions, adapted for preschoolers, were as follows, "You are going to hear some "alien" words. These are words you have never heard before because we made them up! You will hear each word one time. We want you to be a

copycat and repeat each word exactly how you hear it. Some of the words get long, but just listen and do the best job you can being a copycat. Are you ready for the first word?" The experimenter then played each nonword one time and the SLP, who was sitting next to the child, indicated whether a phoneme was produced incorrectly using a form with the phonetic transcription of each nonword (Appendix A). The nonword task was audio recorded for later, offline analysis.

The stimuli consisted of sixteen nonwords ranging from one to four syllables in length (see Table 1 below). Every nonword began and ended with a consonant and there were no consonant clusters. There were also no unstressed syllables in any of the stimuli. The nonwords used met the following requirements:

1. To ensure that nonword repetition would not be affected by a subject's vocabulary knowledge, nonwords were constructed such that none of their individual syllables (CV or CVC) corresponded to an English word. 2. To minimize the articulatory difficulty of the repetition task, enabling the inference that errors resulted from a lack of recall of target phonemes, rather than from an inability to produce them, the nonwords were constructed to exclude the consonants described by Shriberg and Kwiatkowski (1994) as the "late eight" (i.e., /s, z, l, r, \int , \int , ∂ , ∂ /), as well as consonant clusters. 3. The nonwords were constructed to contain only tense vowels, for two reasons. First, being longer in duration than lax vowels, tense vowels are inherently less susceptible to being reduced to schwa. Thus, by contrast with lax vowels, errors on tense vowels cannot easily be attributed to the vowel reduction associated with a casual speech style. Second, the increased perceptibility of tense vowels increases confidence in interpreting errors as problems with recall, rather than perception, of vowel targets.

As a result of including only tense vowels, the stimuli contained no weak syllables, by contrast with the typical English metrical stress pattern in which strong and weak syllables alternate. However, the lack of conformity to real words may be seen as an additional control for familiarity effects, further reducing the possibility that the correct vowel in any syllable could be guessed.

- 4. To reduce the predictability of consonant phonemes in the two possible syllable positions within the nonwords (onset or coda), consonants were assigned to occupy only those syllable positions in which they occurred $\leq 25\%$ of the time, according to data on the percentage of occurrence of each consonant in word-initial and word-final position (Shriberg & Kent, 1982, p. 429). Word-medial consonants were treated as syllable onsets and thus also had to occur $\leq 25\%$ of the time in word-initial position.
- 5. To ensure that accurate repetition of a nonword required that each of its phonemes be recalled independently, no consonants or vowels occurred more than once within a given nonword. (Dollaghan & Cambell, 1998, p. 1138-1139)

Table 2: Dollaghan Nonword Stimuli (Dollaghan & Campbell, 1998).

Two syllables	Three syllables	Four syllables
/teɪ - vak/	/tsi - noı - taub/	/veɪ - ta - t∫aɪ - dɔɪp/
/tsov - væg/	/naɪ - tʃoʊ - veɪb/	/dæ - vou - noi - tsig/
/væ- tʃaɪp/	/dɔɪ - taʊ - væb/	/naı - tʃəɪ - taʊ - vub/
/noʊ - tʃaʊf/	/tei - voi- tʃaig/	/tæ – va - tʃi - naɪg/
	/tei - vak/ /tʃoʊ - væg/ /væ- tʃaip/	/tei - vak/ /tʃi - nɔi - taub/ /tʃoʊ - væg/ /nai - tʃoʊ - veib/ /væ- tʃaip/ /dɔi - taʊ - væb/

Analysis of Nonword Repetition Task Performance

A trained research assistant listened to each recorded nonword production as many times as needed and phonetically transcribed it using a blank form (Appendix A). Any errors in question were resolved by consensus agreement with the project SLP. Substitutions and omissions of phonemes were considered errors, while phoneme additions and disfluencies were noted, but not counted as errors (See Figure 1). For nonwords that were not attempted, the phonemes were noted as "not attempted" and were entered in the total error count. This happened infrequently, on one of the 4-syllable nonwords for two participants. All errors for each participant were entered into a database, with each column in the database corresponding with each syllable of each nonword. Every row in the database represents data for a particular participant. The total number of phonemic errors for each nonword length: nonword 1, 2, 3 and 4, henceforth, (NW 1), (NW 2), (NW 3), and (NW 4) was then tabulated for all participants. Phoneme additions were also noted but were not included in the final error count, so the final tabulated error count for CWS was not skewed (Smith et al., 2012). Finally, I noted the location of errors on each syllable (first, second, third, or fourth) of NW 4 for each participant. Interrater agreement among the first author and two committee members for all nonword coding reached 97%.

Statistical Analysis

NRT performance variables may not be normally distributed as they often comprise percentages or counts within a narrow range and may include zero values. Visual inspection of the distributions and residuals and Shapiro-Wilk tests of normality confirmed that many variables of interest were not normally distributed (i.e., right-skewed). Log and arcsine transformations were applied; however, the data still violated the assumption of normality.

Therefore, we calculated nonparametric Mann-Whitney U tests to examine whether there were differences in the distributions of nonword error rates between CWS and CWNS, and Kruskal-Wallis (KW) tests to assess potential differences in the distributions of error rates among CWS+TSS, CWNS, and CWS+SSD. If KW tests reached significance, post hoc Mann-Whitney tests using a Bonferroni adjusted alpha level of .017 were used to compare error rates among the 3 groups.

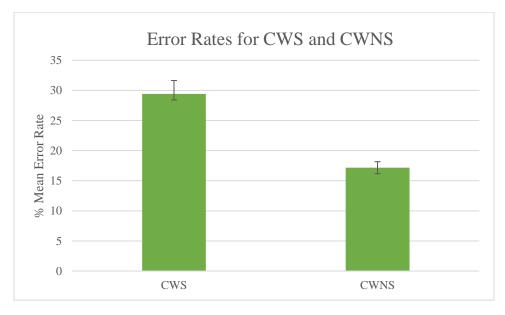
To address our second aim of whether characteristics of nonword errors in the two stuttering groups (CWS+TSS and CWS+SSD) predicted membership into those respective groups, multinomial regressions were performed with CWNS serving as the baseline comparison group for syllable location of errors (first, second, third, or fourth syllable) on NW 4.

Results

Nonword Error Rates Between All Children Who Stutter and Children Who Do Not Stutter

In order to compare findings from the current study with those from previous studies of NRT accuracy between CWS and CWNS, we assessed average error rates (i.e. collapsed across the four nonword lengths) from CWS (with and without SSD) and CWNS. As Figure 1 reveals, CWS with and without SSD had an average error rate that was 42% higher than the CWNS. A Mann-Whitney U test indicated that the distribution of the average error rate (U = 557.5, p < .001) for CWS (Mdn = 26.04) was significantly different (on average, higher) than the average error rate for CWNS (Mdn = 17.71).

Figure 1: Overall mean error rate collapsed across nonwords with standard error bars for all CWS and CWNS.



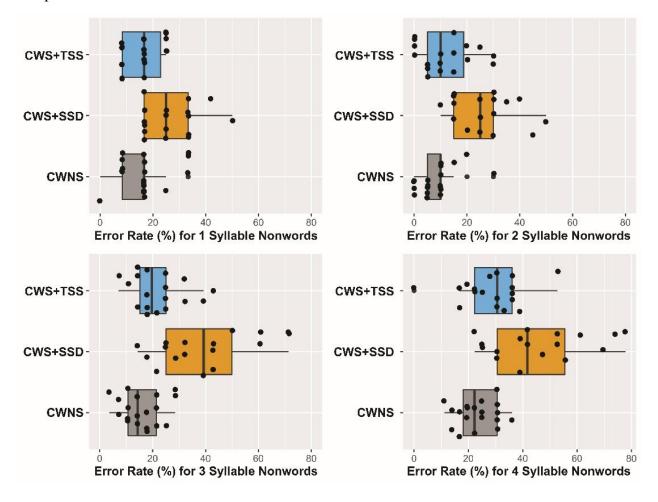
Nonword Error Rates Among Children Who Stutter with and Without a Speech Sound Disorder and Children Who Do Not Stutter

We then compared NRT error rates for the four NWs for the CWS with and without an SSD. The three groups of children for the subsequent analyses are: CWS+SSD, CWS+TSS, and CWNS. A series of four Kruskal-Wallis tests revealed a significant difference in NRT error rates for NW 1, H(2) = 12.43, p = .002, NW 2, H(2) = 21.54, p < .001, NW 3, H(2) = 20.92, p < .001, and NW 4, H(2) = 20.02, p < .001. Post-hoc Mann-Whitney tests using a Bonferroni adjusted alpha level of .017 were used to compare performance among the three groups. Overall, CWS+SSD had significantly higher error rates on all four NW lengths than the CWS+TSS and CWNS groups. No comparison between the groups of CWS+TSS and CWNS was significant (all ps > .26) (see Figure 2). As seen in Figure 2, on average, children in all groups had the highest error rates on the longest nonword, NW 4.

Table 3: Median, standard error, U test statistic, and adjusted p-value for the comparisons between the three groups of participants: children who stutter with typical speech sounds (CWS+TSS), children who stutter with SSD (CWS+SSD) and children who do not stutter (CWNS).

Comparison	Median Group 1	Median Group 2	Std. Error	U Test Statistic	Adjusted <i>p</i> -value
			NW 1		
CWS+SSD/CWS+TSS	25.00	16.67	5.09	15.70	.006
CWS+SSD/CWNS	25.00	16.67	5.02	15.38	.007
CWS+TSS /CWNS	16.67	16.67	4.95	0.32	1.00
	NW 2				
CWS+SSD/CWS+TSS	25.00	10.0	5.27	17.95	.002
CWS+SSD/CWNS	25.00	10.0	5.20	23.20	.001
CWS+TSS /CWNS	10.0	10.0	5.12	5.25	.92
	NW 3				
CWS+SSD/CWS+TSS	39.29	19.64	5.29	15.02	.014
CWS+SSD/CWNS	39.29	14.29	5.23	23.72	.001
CWS+TSS /CWNS	19.64	14.29	5.15	8.70	.27
	NW 4				
CWS+SSD/CWS+TSS	41.67	30.56	5.29	14.78	.016
CWS+SSD/CWNS	41.67	22.22	5.23	23.19	.001
CWS+TSS /CWNS	30.56	22.22	5.15	8.41	.31

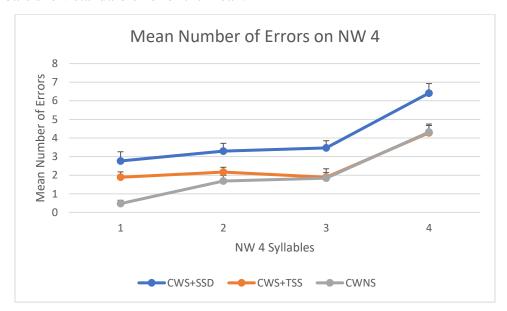
Figure 2: Error rates percentages for the three groups of participants for each nonword length. Box edges represent the 25th (lower quartile) and 75th (upper quartile). Whiskers cover the 95th percentile with the center line representing the mean and black circles representing individual data points.



Location of Errors on Nonword Four

As Figure 3 reveals, all three groups of children made, on average, the most errors on the last (fourth) syllable of NW 4 compared to the first, second, or third syllables.

Figure 3: Mean number of errors for the three groups of participants as a function of syllable order for nonword 4 for children who stutter with speech sound disorder (CWS+SSD), children who stutter with typical speech sounds (CWS+TSS) and children who do not stutter (CWNS). Error bars show standard error of the mean.



We used multinomial regression analyses with CWNS serving as the baseline comparison to investigate whether the location of nonword errors on each syllable of the 4-syllable nonword predicted group membership for the groups of children who stutter with and without SSD (Table 4). Results showed that errors made on the first syllable of NW 4 predicted membership into both groups of children who stutter, CWS+TSS and CWS+SSD (top left graph of Figure 4). For each one unit increase in number of errors made on the first syllable of the NW 4, the odds of being a CWS+TSS increase by 3.54. For one unit increase in number of errors made on the first syllable of NW 4, the odds of being a CWS+SSD increase by 4.95. After, on average, 2-3 errors made on the first syllable, the odds of being a CWS+TSS begin to decline, while the odds of

being a CWS+SSD continue to increase. Additional errors on the second, third, or fourth syllable did not significantly predict membership into the CWS+TSS group (Table 4; Figure 4). However, for every additional error made on the second syllable of NW 4, the odds of being a CWS+SSD increased by 2.30, for the third syllable by 1.82, and for the fourth syllable, by 1.91 (top right and two bottom graphs of Figure 4).

Figure 4: Probability of being a child who stutters with a speech sound disorder (CWS+SSD) or child with typical speech sounds (CWS pass BBTOP) as a function of the location of errors on all four syllables of NW 4 (data from children who do not stutter CWNS served as a baseline for each syllable).

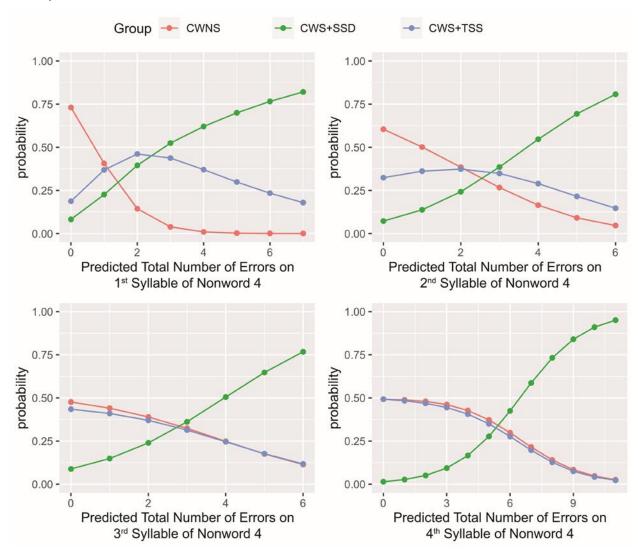


Table 4: Table 4: Multinomial regression results with children who do not stutter (CWNS) serving as baseline. Syllable locations of NW 4 (predictors), odds ratios with confidence intervals (CI) and *p*-values for children who stutter with typical speech sounds (CWS+TSS) and children who stutter with speech sound disorders (CWS+SSD).

Predictor	Group	Odds Ratio	CI lower	CI upper	<i>p</i> -value
Syll1_NW 4	CWS+SSD	4.95	2.10	11.67	<.001
Syll1_NW 4	CWS+TSS	3.54	1.57	7.99	<.001
Syll2_NW 4	CWS+SSD	2.30	1.30	4.07	<.001
Syll2_NW 4	CWS+TSS	1.35	0.80	2.27	.26
Syll3_NW 4	CWS+SSD	1.82	1.16	2.86	.01
Syll3_NW 4	CWS+TSS	1.02	0.67	1.56	.93
Syll4_NW 4	CWS+SSD	1.91	1.19	3.07	.01
Syll4_NW 4	CWS+TSS	0.99	0.68	1.44	.95

Discussion

The overarching goal of this study was to take an in-depth look into NRT performance in preschoolers who stutter to shed light on poorer nonword production performance in these children. As a first step, we compared performance between groups of preschool-aged children who do and do not stutter. We found that the total error rate (i.e., collapsed across the four nonwords) for CWS, regardless of speech sound production abilities, was higher than CWNS. This result replicates findings from many previous studies noting higher total error rates in CWS on an NRT (Anderson et al., 2006; Anderson & Wagovich, 2010, Pelczarski & Yaruss, 2016; Hakim & Bernstein-Ratner, 2004; cf. Bakhtiar et al., 2007). We then divided participants who stutter into two groups based on children's speech sound production abilities. The CWS+SSD group had significantly higher error rates on all four NWs compared to the CWS group, while the error rates between CWS+TSS and CWNS were not significantly different for any of the four NWs.

Children in all groups made the most errors on the longest nonword, NW 4. We examined where errors occurred within this nonword to determine whether error characteristics on this nonword might help distinguish NRT performance across the groups of children. The number of errors committed on all four syllables of NW 4 predicted membership into the CWS+SSD group, while errors committed on the first syllable of NW 4 also predicted membership into the CWS+TSS group.

Comparison Between Children Who Do and Do Not Stutter

Previous studies indicate that CWS generally perform more poorly on NRTs compared to CWNS (Anderson et al., 2006; Anderson & Wagovich, 2010; Hakim & Bernstein-Ratner, 2004;

Pelczarski & Yaruss, 2016; cf. Bahktir, Ali, & Sadegh, 2007). The current study replicated this finding. We found that the total error rate (i.e. averaged across all four nonwords) for CWS was significantly higher, on average, compared to CWNS. When CWS were combined into one group that included a range of speech sound abilities, they had an average error rate that was 42% higher than the CWNS group (see Figure 1).

Given the number of processes involved in hearing and repeating unfamiliar nonwords including perception of phoneme strings, phonological encoding and phonological storage/working memory, and motor planning and execution (Burke & Coady, 2015; Dollaghan & Campbell, 2004), differences in performance between CWS and CWNS have been interpreted to result from deficient encoding, storing, and/or retrieval mechanisms (Anderson et al., 2006; 2010; Hakim & Bernstein-Ratner, 2004, Pelzcarski & Yaruss 2016). For example, Anderson et al. (2006) cited potential deficits in phonological working memory in their group of preschool CWS. On the other hand, Pelzcarski & Yaruss (2016) showed that working memory abilities were comparable between groups of children who do and do not stutter (as measured by performance on a digit span task) and discussed the possibility of weaker phonological representations in CWS and/or more subtle differences in phonological working memory that their control task did not identify.

Performance Among Groups of Children Who Stutter With and Without a Speech Sound Disorder and Children Who Do Not Stutter

The first aim of the study was to clarify the potential role that speech sound production abilities play in poorer NRT performance in CWS. Children with SSD also perform more poorly on these tasks (Munson et al., 2005; Pigdon et al., 2020; Martikainen et al., 2020; Archibald & Gathercole, 2007; Vuolo & Goffman, 2020). Therefore, we examined error rates on all four

nonwords while considering the speech sound production abilities of CWS. Specifically, we compared performance of CWS+TSS, CWS+SSD and CWNS. Both the current study and Smith et al., (2012) accounted for the presence of concomitant disorders in the participants by grouping children based on phonological abilities in two separate cohorts of preschool CWS. The current study replicated findings from Smith et al. (2012) with larger group sizes. We found that the CWS+SSD group made significantly more errors on all four nonwords than both the CWS+TSS and CWNS groups. NRT performance for the CWS+TSS was not statistically different from the CWNS group. Similarly, once speech sound production abilities were taken into consideration, Smith et al. (2012) also found significant differences in NRT performance between CWNS and CWS with phonological and language deficits, whereas CWS with typical phonological abilities had error rates similar to the CWNS group. Taken together, these results suggest that having a concomitant SSD was likely a driving factor behind poorer performance on the NRT in this subgroup of children who stutter.

Growing evidence suggests that children with SSD may have deficits in speech sound perception (Hearnshaw et al., 2019; Rvachew & Brosseau-Lapre, 2018). This deficit in speech perception may lead to difficulties forming more well-specified phonological representations (Sosa & Stoel-Gammon, 2012; Macrae et al., 2014; Hearnshaw et al 2019; Rvachew & Brosseau-Lapre 2018). As a child's vocabulary grows, more detailed representations are needed to differentiate words that sound similar, such as "cup" versus "cut" (Metsala & Walley, 1998). This has been directly related to NRTs, as children with larger vocabularies achieve higher nonword repetition accuracy compared to children who possess smaller vocabularies (e.g., Gathercole & Baddeley, 1989). Children with higher vocabularies are thought to have more detailed phonological representations which can be more flexibly arranged into novel phoneme

sequences necessary for nonword repetition (Edwards et al., 2004). Therefore, children with SSD may show poorer NRT performance due to difficulties forming flexible, well-specified phonological representations.

Location of Errors on Nonword Four

The second aim of this study was to determine whether error characteristics differentiated children in the three participant groups. We examined the specific location of errors committed on the longest and most complex nonword, NW 4. Gupta (2005) conducted a study to determine whether there are serial-position effects in NRTs and how these effects could affect NRT accuracy. More specifically, they examined if primacy effects (the tendency to recall items presented at the beginning of a task) and recency effects (the tendency to recall items presented at the end of a task) occurred in NRTs as they do in serial word list recall tasks (Murdock, 1962). Twenty adults participated and recalled two nonword lists (consisting of 30 four-syllable nonwords each) in Experiment 1 and 2, and a list of longer, 7-syllable nonwords in Experiment 3, all created specifically for this study. They found that both primacy and recency effects were present during the nonword repetition tasks. Overall, adults were better able to recall syllables near the beginning and end of all nonwords, supporting the idea of utilization of primacy and recency effects when recalling nonwords (Gupta, 2005).

In the current study, it appears that many preschoolers, regardless of speech status, were not taking advantage of the recency effect, whereas adults in the Gupta (2005) study clearly took advantage of this effect to support more accurate attempts at recalling nonwords. Children in all groups made, on average, the highest number of errors on the last syllable of NW 4. It is possible that they were able to take advantage of the primacy effect to some degree but perhaps the recency effect applied to NRT performance may emerge over development. In support of this

hypothesis, Walsh (in preparation) found that school-aged (7- to 12-year-olds) CWS with typical speech and language development also produced significantly more errors compared to CWNS on the last syllable of NW 4, whereas CWNS were able to take advantage of the recency effect and committed the fewest errors on the last syllable. Therefore, it seems that by the time preschool children with typical speech and language development reach school age they were able to take advantage of the recency effect whereas school-aged CWS did not on this NRT. Accordingly, it is plausible that the recency effect arises as preschool children develop.

Using the CWNS as a baseline, the analysis of NW 4 also showed that group membership could be predicted by error rates for both groups of CWS, with and without SSDs. The number of errors made on all four syllables significantly predicted membership into the CWS+SSD group. Alternatively, only errors made on the first syllable predicted CWS+TSS membership. Errors made on the first syllable of NW 4, increased the chances of being a member of the CWS+TSS increased by 3.54. However, after three or more errors made on the first syllable of NW 4, a child who stutters would be more likely to be categorized as a CWS+SSD than a CWS+TSS. Compared to both groups of CWS, CWNS made significantly less errors on syllable 1. Overall, this result suggests that CWNS may be able to take advantage of the primacy effect to a greater extent than CWS, meaning they were able to recall the first syllable more accurately, while CWS were less likely than their peers who do not stutter to do so.

It has been shown that the primacy effect emerges over development, as seen in a longitudinal study of 54 children around 8 years of age. They were reassessed intermittently (every six months) for the following four years; the final assessment occurred at 12 years of age. This study looked at the development of recall processes during three free recall tasks, each list consisting of 12 words each. As participants aged, recall accuracy increased as well as utilization

of the primacy effect. It was shown that children around 8 years of age showed virtually no primacy effect during the recall tasks. As they aged, however, utilization of the primacy effect was more prominent (Lehmann, 2015). In the current study, both CWS+TSS and CWS+SSD showed higher error rates than CWNS on the first syllable of NW 4 indicating they may not be exploiting the primacy effect to the same extent as their peers with typical speech and language. Taken together, our results reveal differences in the strategies CWS used to perceive, encode, store, and produce strings of syllables.

Limitations and Future Directions of the Current Study

While the current study extended previous research by investigating the role of speech sound production, it was not within the scope of the current study to also investigate the role of language impairment. It has been shown that children with language impairments also perform more poorly on NRTs (Archibald & Gathercole, 2007; Dollaghan & Campbell, 1998; Estes et al., 2007; Vuolo & Goffman, 2020; Pigdon et al., 2020). There were simply not enough children in each of the groups (CWS with and without language impairment) to properly assess this in the current study. It should be noted that all participants were assessed on their expressive and receptive language abilities, and one child in the CWS+TSS group and several children in the CWS+SSD group did not pass these assessments. Co-occurring language impairments could also have affected overall NRT performance, especially in the group of CWS+SSD (Dollaghan & Campbell, 1998; Macrae & Tyler, 2014; Pigdon et al., 2020).

In addition, results of the current study may have been enhanced by including a measure of vocabulary. Vocabulary size has been associated with nonword repetition abilities and controlling for vocabulary size has allowed for investigation of additional factors, for example

encoding abilities, influencing nonword repetition performance in children with speech and language disorders (Edwards et al., 2004; Roepke et al., 2020).

Finally, we could have unknowingly counted phonemes as errors that were not in a child's inventory. We had access to BBTOP scores, but not the BBTOP test forms for each participant, so were not able to assess whether phonemes not in a child's inventory inflated the error count. However, it should be noted that the NRT used in the current study contained nonwords that did not include the later developing eight phonemes (Dollaghan & Campbell, 1998). Thus, the phonemes in the NRT were more likely to be in participants' phonemic inventories.

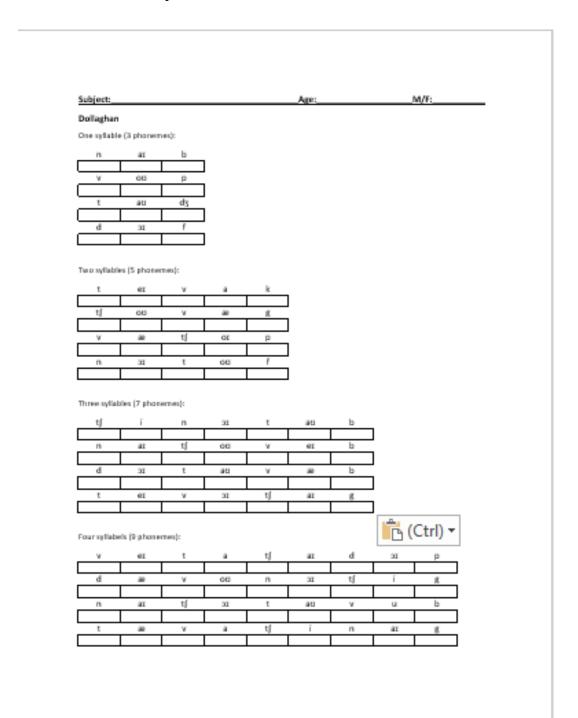
Conclusion

NRTs involve multiple processes; therefore, it is unsurprising that children with a range of speech and language disorders have difficulty with these tasks (Anderson et al., 2006; Coady & Evans, 2008; Munson et al., 2005; Vuolo & Goffman, 2020). In the current study, the CWS+SSD group had significantly higher error rates across all NW lengths compared to other participant groups. However, error rates for CWS+TSS were not significantly different from CWNS supporting the assertion that having a concomitant SSD played a significant role in poorer NRT performance in this subgroup of CWS. We also noted that both groups of CWS—with and without SSD—may have relied upon different phonological working memory strategies compared to CWNS. We found that errors committed on syllable 1 of NW 4 predicted membership into both groups of CWS. Thus, CWNS may be taking advantage of the primacy effect compared to CWS. Taken together, these findings help shed light on poorer NRT performance in stuttering and enhance generalization to populations of CWS with and without concomitant SSD.

APPENDIX

APPENDIX: Blank Nonword Repetition Form

Figure 5: Blank Nonword Repetition Form



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