Loci of dysfluency in stuttering

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Similar within-utterance loci of dysfluency in acquired neurogenic and persistent developmental stuttering

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Abstract

Although the underlying neural mechanisms remain unknown for both persistent developmental stuttering (PSD) and acquired neurogenic stuttering (ANS), few studies have examined similarities/differences between these two disorders. We evaluated in both PDS \((n = 35)\) and ANS \((n = 5)\) phonetic, word class, word length, and word position variables that are widely believed to influence at which loci within utterances PDS speakers’ stuttering is most likely to occur. For both groups, (a) word weights based on the combination of variables were greater for stuttered vs. fluent words, and (b) stuttered words were loaded more by individual variables. However, contrary to long-standing views regarding PDS, greater loading for stuttered words was not found for the position variable. Findings suggest similar loci of stuttering in adults with PDS and ANS, and, for both groups, the probability of stuttering on a given word was more influenced by motor production variables than language variables.

Keywords: speech; developmental stuttering; acquired neurogenic stuttering; dysfluency; loci of stuttering
Similar within-utterance loci of dysfluency in acquired neurogenic versus persistent developmental stuttering

Introduction

For both persistent developmental stuttering (PSD) and acquired neurogenic stuttering (ANS), the precise neural mechanisms underlying the overt speech symptoms (i.e., speech dysfluencies consisting of part-word repetitions, audible and inaudible sound prolongations, and monosyllabic word repetitions—Guitar, 2013) remain largely unknown. It is rather surprising that, over more than eight decades of stuttering research, very few studies have directly examined the similarities and differences between these two types of stuttering. Instead, conclusions have been drawn mostly by comparing the highly variable results from ANS case studies with either verified empirical data or clinical insights regarding PDS. This is unfortunate given that direct comparisons of the primary symptoms as well as any associated behavioral and neural characteristics have the potential to elucidate sensorimotor and/or linguistic processes contributing to the observed breakdowns in speech fluency.

In this context, it is interesting that the results from a study in which speech-language pathologists evaluated audio-recordings of individuals with either ANS or PDS suggested that the dysfluencies in the two disorders can be difficult to distinguish (Van Borsel & Taillieu, 2001). On the other hand, results from our own laboratory (Balasubramanian, Max, Van Borsel, Rayca, & Richardson, 2003; Balasubramanian, Cronin, & Max, 2010) as well as other laboratories (Canter, 1971; De Nil, Theys, & Jokel, 2018) have suggested that the situational variability of stuttering across ANS
patients differs from that typically seen across PDS cases: a considerably smaller number of individuals with ANS show improvements in fluency during repeated readings of the same material (adaptation effect) or when hearing delayed or frequency-shifted auditory feedback (DAF, FAF)—two conditions that are known to be fluency-enhancing for individuals with PDS. For example, adaptation across five repeated readings of the same material occurs in as many as 70-85% of individuals with PDS (Gray, 1965; Max & Caruso, 1998; Newman, 1963) but in only ~50% of individuals with ADS (De Nil, Theys, & Jokel, 2018; Theys, van Wieringen, Sunaert, Thijs, & De Nil, 2011).

Remarkably, an even more basic aspect of the symptomatology—namely, the loci of stuttering moments within spoken utterances—has not been directly compared for ANS vs. PDS, despite the availability of extensive data for PDS. Most of the loci data for PDS are based on an early series of studies conducted by Spencer Brown (Brown 1937, 1938a, 1938b, 1938c, 1945; Brown & Moren, 1942; Johnson & Brown, 1935) who found that stuttering moments are not distributed randomly across the words of an utterance. Instead, Brown’s results suggested that, in adults with PDS, stuttering moments are more likely to occur on words with certain phonetic and/or linguistic attributes. Based on the series of studies, Brown (1945) concluded that, although there are large inter-individual differences, four factors affect the probability of stuttering on a given word: (a) its initial phonetic category (more stuttering on words in which the initial sound is a consonant vs. a vowel); (b) its grammatical class (more stuttering on nouns, adjectives, verbs, and adverbs); (c) its length (more stuttering on words that are 5 or more letters long); and (d) its position within the sentence (more stuttering on the first three words of the sentence).
Since Brown’s work in the 1930s and 1940s, studies on PDS have mostly confirmed the increased probability of stuttering on words loaded with these factors. However, it is not easy to estimate precisely how much these factors influence the probability of stuttering because (a) the definitions of each factor have not always been consistent across studies (e.g., for the phonetic factor, there have been discrepancies in terms of whether glides and liquids are considered consonants), (b) the four factors do not have equally strong empirical support, (c) certain effects (i.e., the influence of grammatical class) differ between adults and children who stutter, and (d) there are interactions among these four factors as well as between these and other factors such as syllable stress and frequency of use (Au-Yeung, Howell, & Pilgrim, 1998; Bloodstein & Gantwerk, 1967; Dayalu, Kalinowski, Stuart, Holbert, & Rastatter, 2002; Dayalu, Kalinowski, & Stuart, 2005; Hahn, 1942; Jayaram, 1983; Soderberg, 1962; Taylor, 1966a, Wingate, 1967, 1979, 2002). Interactions of the factors make it difficult to determine how each of them influences the probability of stuttering independently. For example, Taylor (1966a) reported that the grammatical class of words (i.e., content vs. function words) indeed influenced the probability of stuttering, but pointed out that when the data were analyzed separately for words with an initial consonant or with an initial vowel, the difference in the content vs. function words was not observed. Thus, in the analyzed data set, the effect of grammatical class on the loci of stuttering was due entirely to more content words having an initial consonant.

With regard to understanding the neurophysiological mechanisms underlying the occurrence of individual stuttering moments, important new insights may be gained from detailed comparisons of loci analyses in individuals with PDS and ANS. For example,
Canter (1971) claimed that (a) the distribution of stuttering moments across speech sounds differs considerably between ANS and PDS, and that (b) the distribution of stuttering moments is not related to grammatical class in ANS. However, Canter’s (1971) claim about phonetic influences referred to the hierarchy of “difficult” sounds within the group of consonants rather than to the typical distinction between words with initial consonants vs. vowels (as in the studies by Spencer Brown), and Ringo and Dietrich (1995) have pointed out that this within-consonants hierarchy did not hold across different ANS studies. Ringo and Dietrich (1995) further pointed out that neither the phonetic factor (more stuttering on words with initial consonants vs. vowels) nor the grammatical factor (more stuttering on content vs. function words) has been investigated in any of the ANS studies included in their review. A more recent study by Chang and colleagues did investigate the loci of stuttering in four cases with adult onset, but these cases were specifically selected because they showed no evidence of neurological lesions (Chang, Synnestvedt, Ostuni, & Ludlow, 2010).

To address this important gap in understanding even the most fundamental characteristics of ANS and the similarities or differences between ANS and PDS, we compared speakers from both groups in terms of the aforementioned phonetic, word class, word length, and word position variables that are widely-believed to influence where within spoken utterances stuttering moments are most likely to occur. Stuttered words were identified off-line in audio-recordings of oral reading by adults with ANS or PDS. Brown’s (1945) word weights were then determined for each stuttered and each fluent word by assigning weights (minimum 0, maximum 4) to each word that (a) started with a consonant; (b) was a noun, adjective, adverb, or verb; (c) was 5 or more letters
long; and (d) was one of the first three words in a sentence. To avoid potential biases introduced by measuring word length in letters and word position in terms of position in the sentence, complementary analyses were conducted with word length defined in terms of the number of speech sounds and word position defined relative to phrase onset. The PDS and ANS groups were compared with regard to their overall word weight scores for fluent vs. stuttered words as well as with regard to the percentage of fluent vs. stuttered words loaded by each individual factor.

Methods

General participant characteristics

Data were available for five individuals with ANS and 35 individuals with PDS. The PDS group included 8 female and 27 male participants. The ANS group included 1 female and 4 male participants. The mean age for the PDS group was 30 years (ranging from 19 to 49 years), and the mean age for the ANS group was 64 years (57, 67, 58, 60, and 78 years of age for the 5 participants). All participants except one individual with PDS and one individual with ANS were native English speakers. One female participant from the PDS group was a native speaker of Polish, but English was her daily language. Participant ANS5 from the ANS group was a native speaker of German although English had been his primarily language for a long time. All participants from both groups were considered to stutter based on evaluation by at least one of the two senior authors (VB, LM) of this study.

All individuals in the PDS group had been evaluated with the Stuttering Severity Instrument, Fourth Edition (SSI-4; Riley, 2008), a measure of overall stuttering severity
based on the frequency and duration of stuttering moments as well as secondary characteristics (e.g., facial grimacing) during both conversation and oral reading. The average total SSI-4 score across all PDS participants was 22.8 (with a range 11–46). With regard to the individual participants, the stuttering of 12 individuals was rated as very mild, 12 as mild, 5 as moderate, 2 as severe, and 4 as very severe.

Conversational speech samples were not available for all individuals in the ANS group; thus, their overall stuttering severity could not be determined with the SSI-4. Instead, in Figure 1, we show the percentage of stuttered syllables during the passages read for the present study (described below) for each individual subject in the ANS group together with the group mean and standard error of the mean for the PDS group. The ANS group mean percent syllables stuttered was 13.32% \( (SD = 6.76\%) \) versus 6.52% \( (SD = 5.99\%) \) for the PDS group. Due to the large inter-individual variability, this difference between the groups in percent stuttered syllables was not statistically significant \( (Welch t(4.94) = 2.13, p = 0.09) \).

Acquired neurogenic stuttering (ANS) group

Participant ANS1 was male and 57 years old at the time of testing. He had a history of non-insulin dependent diabetes, hypertension, and ischemic disease. Approximately 6 months prior to data collection for the current study, he experienced a cerebro-vascular accident (CVA) that resulted in a new subcortical ischemia involving the right hemisphere and bilateral hypodensity of the pons. These lesions were revealed by a CT scan nine days after the CVA. An evaluation by a speech-language pathologist resulted in the diagnosis of dysphasia and occasional deficits in naming. Non-fluent
speech was also reported since the onset of the CVA. Family members reported that the symptoms resembled those of the childhood stuttering that the patient had experienced, but outgrown, at a young age. More details and CT scans for ANS1 have been published in Balasubramanian et al. (2003).

Participant ANS2 was a 67-year-old male who had been admitted to the hospital with acute speech changes earlier in the same year as the testing for this study. He had a history of poorly controlled diabetes, prostate cancer, and arthritis, and he wore bilateral hearing aids. His medical records indicate a history of cerebrovascular accident and a diagnosis of small vessel ischemic disease. Unfortunately, we were not able to obtain CT or MRI images. ANS2 also had paresthesia along the left side of the face and left upper extremity along with left-handed weakness and short-term memory impairment. Speech and language assessment resulted in the diagnosis of dysarthria and ANS.

Participant ANS3, a 58-year-old male, had an episode of left hemisphere stroke approximately nine months prior to the testing for this study. An MRI scan indicated acute and sub-acute changes in the left mid parietal area. His family also reported that he had a previous stroke at the age of 50, which had resulted in a right posterior parietal lobe infarct. The first stroke affected his speech, but he reportedly recovered and was able to speak relatively unimpaired until the second stroke. At the time of the second stroke, he was also diagnosed with aphasia, moderate-severe apraxia, and deficits in comprehension and reading. A more detailed case history for ANS3 has been reported in Balasubramanian and Hayden (1995).

Participant ANS4 was a 60-year-old woman who suffered left hemisphere ischemic strokes approximately 2.5 years before the testing for this study. The strokes
resulted in infarcts of the white matter underlying left parietal and frontal lobe. She developed aphasia, apraxia of speech, and stuttering immediately after the stroke. A clinical speech-language evaluation conducted two years post-onset confirmed symptoms of apraxia of speech, ANS, and minimal aphasia. More details and MRI images of this participant have been published as “Case 2” in Balasubramanian et al. (2010).

Participant ANS5, a male participant, was 78 years old at the time of testing. He was admitted to a hospital at the age of 76 years, after feeling weakness and having slurred, dysarthric speech. He had experienced two bilateral lacunar infarcts earlier in life. At the time of the first stroke, at the age of 73 years, an MRI scan showed three left lacunar infarcts, and he was diagnosed with dysarthria. At the time of his second stroke, at the age of 75 years, an MRI scan revealed enlargement of the previous three foci and the development of two new foci in the right cerebral white matter. His diagnosis of dysarthria remained. He was also diagnosed with anomia and ANS. The ANS symptoms were reportedly similar to the stuttering that he had experienced in his childhood but from which he had recovered at a young age.

Procedure

PDS data for the present study were derived from participants’ oral readings of the “Washington passage” from the SSI-4 (Riley, 2008). Using audio- and/or video-recordings, stuttered syllables were identified offline by the first author or trained graduate students or post-doctoral fellows in the first author’s laboratory. Expressed as Cohen’s Kappa (Cohen, 1969; Lewis, 1994), each trainee’s token-by-token reliability for identifying stuttering moments (relative to the first author’s judgments) exceeded .80 across a set of five speech samples. ANS data were derived from participants’ oral
readings of passages that differed across participants depending on their reading ability. Stuttered syllables had been identified offline from audio- and/or video-recordings by the first author and trainees in the first author’s laboratory (participants ANS4 and ANS5) or by the last author (ANS1, ANS2, ANS3).

Brown’s (1945) word weights were determined for each word in each of the six reading passages (one common passage from SSI-4 for all participants in the PDS group, and five different passages for the five participants in the ANS group). Weights or points were assigned based on the following four factors identified by Brown (1945): (1) position factor: the word was one of the first three words in a sentence; (2) phonetic factor: the initial sound of the word was a consonant; (3) length factor: the word was five or more letters long; and (4) grammatical factor: the word was a noun, verb, adverb or adjective. Thus, the maximum weight a word could receive was four, and the minimum weight was zero. Note that for the grammatical factor, we used Brown’s (1945) original procedure of assigning a weight for “adjectives, nouns, adverbs, and verbs” and no weight for “pronouns, conjunctions, prepositions and articles” (p. 182) rather than relying on the currently more often used distinction between content words and function words. For example, we assigned a grammatical factor weight to auxiliary verbs given that Brown did not seem to distinguish them from other verbs and did not list them separately in the category of words that were not assigned a weight for this factor. For the phonetic factor, on the other hand, we did use the more common approach of distinguishing between consonants (assigned a weight) and vowels (not assigned a weight) rather than Brown’s (1945) original procedure. In the original procedure, a weight for the phonetic factor was assigned to all words with an initial sound that had “a mean percentage of
stuttering of greater than 9.7 per cent for the entire group of subjects” (Brown, 1945, p. 182). The latter procedure had resulted in no weight being assigned to words with the initial consonants /t, w, ʍ, থ, h/ in addition to those with an initial vowel (Johnson & Brown, 1935). Our decision to use, instead, the global consonants vs. vowels distinction was based on the fact that (a) scientifically, the only appropriate approach is to determine the weights a priori rather than post hoc based on the obtained data (see also Taylor, 1966b, for a critique of the post hoc procedure); (b) Johnson and Brown’s (1935) own analysis did, in fact, lead to the conclusion that “in general more stuttering occurs in relation to consonants than vowels” (p. 493), and (c) the rank-order of stuttered consonants is highly variable across individual subjects (Johnson & Brown, 1935; Wingate 2002; Ringo & Dietrich, 1995).

For each participant, the average weight for the words that contained at least one stuttered syllable (stuttered words) and the average weight for the words with no stuttered syllables (fluent words) were calculated by dividing the total count of weights assigned to all stuttered or fluent words, respectively, by the total number of words identified as stuttered or fluent. Based on all participants in either group, the average weight of all stuttered words was then compared with the average weight of all fluent words. However, for any given participant, the number of fluent words was much greater than the number of stuttered words (e.g., as mentioned above, the mean percent stuttered syllables was only 6.52%). In order to control for any regression-to-the-mean bias in the results due to averaging word weights over such vastly different numbers of tokens, a modified evaluation method was also used. For each participant, a subset of fluent words was randomly selected such that the number of selected fluent words equaled the participant’s
number of stuttered words. The average weight of the stuttered words was then also compared with the average weight of this subset of randomly selected fluent words.

A second set of analyses was carried out to compare the ANS and PDS groups for each of the four factors (phonetic, length, grammar, and position) separately. For these analyses, the number of stuttered words that received a weight for a given factor was divided by the total number of stuttered words and multiplied by 100 to determine the percentage of stuttered words that were “loaded” by that particular factor. For the fluent words, the same analyses were completed with the above described subsets of randomly selected fluent words (i.e., each participant’s number of stuttered tokens matched the number of fluent tokens).

In a final set of analyses, we determined whether results based on traditional word weight measures for the length factor and the position factor (Brown, 1945) might be biased because of their arguably arbitrary definitions. For example, when long words are defined as words that are 5 or more letters, “ghost” is counted as a long word whereas “host” is counted as a short word, even though both words consist of the same number of speech sounds in an identical monosyllabic word structure. For this reason, we examined whether or not the effect of word length is still present if long words are defined as words consisting of 5 or more speech sounds. Similarly, an alternative operational definition was also considered for the effect of word position. For this factor, we evaluated whether or not similar results are obtained if a position weight is assigned to the first three words of each clause rather than each sentence.

Given the extreme difference in sample sizes ($n_{PDS} = 35$ vs. $n_{ANS} = 5$) and given that the results of primary interest relate to within-group comparisons of stuttered versus
fluent words in terms of the effect of Brown’s word weight factors, statistical testing was completed with two-sided paired t-tests for the two groups separately. For each test, Cohen’s $d$ (Cohen, 1988) with an adjustment for paired comparisons (Gibbons, Hedeker, & Davis, 1993) was also calculated to evaluate and compare effect sizes for each group of speakers ($<0.2 =$ negligible, $<0.5 =$ small, $<0.8 =$ medium, $\geq0.8 =$ large; Cohen, 1992). The R software was used for all analyses (R Core Team, 2017).

Results

The PDS and ANS groups’ average Brown’s word weights across the four factors are shown in Figure 2. Each group’s average word weight for the stuttered words is compared with the word weights for the fluent words as obtained with the two different methods of selecting fluent words (using either a participant’s set of all fluent words or only a randomly selected subset of fluent words—see above). Reflecting overall similar patterns, the mean weight of the stuttered words was 2.51 ($SD = 0.37$) for the PDS group and 2.39 ($SD = 0.28$) for the ANS group. For both groups, these weights for stuttered words were higher than those for the fluent words. Using all fluent words, the mean weights were 1.86 ($SD = 0.10$) for PDS and 1.81 ($SD = 0.05$) for ANS. Very similar word weights were obtained when using only the randomly selected fluent words: 1.80 ($SD = 0.41$) for PDS and 1.79 ($SD = 0.28$) for ANS.

For the PDS group, the word weight of stuttered words was statistically significantly greater than that of all fluent words [$t(34)=9.84$, $p<0.001$, $d=1.66$] as well as that of the randomly selected fluent words [$t(34)=7.10$, $p<0.001$, $d=1.20$]. Both effects were associated with large effect sizes. The difference in word weight between the two
methods of selecting fluent words was not statistically significant \([t(34)=0.86, p=0.40]\).

The same pattern was observed for the ANS group: the word weight of stuttered words was statistically significantly greater than that of all fluent words \([t(4)=4.00, p=0.016, d=1.79]\) and that of the randomly selected fluent words \([t(4)=5.01, p=0.007, d=2.24]\), both effects were associated with large effect sizes, and the two methods of selecting fluent words did not yield different results \([t(4)=0.19, p=0.86]\).

Given that there was no significant difference between the results from the two methods of calculating weights for the fluent words, only the data obtained by randomly selecting a subset of the fluent words were used for the remaining analyses. First, we explored the ANS individual participant data, and considered them relative to the PDS averaged group data (Figure 3). The PDS averaged data as well as the data for each individual ANS participant showed a greater word weight for the stuttered words than for the fluent words. Moreover, the ranges of ANS individual subject data for stuttered and fluent word weights encompassed the corresponding average PDS values. Thus, these individual participant data further confirmed the similarity between PDS and ANS in terms of the overall word weight across the four factors.

Next, we compared the two participant groups with regard to the effect of each of the four factors separately. Figure 4 shows, for each factor, the percentages of stuttered and fluent words that were “loaded” by each factor; that is, the percentages of stuttered and fluent words that had received a weight for that particular factor. Neither the ANS group nor the PDS group showed an effect of word position within the sentence: the percentage of stuttered words loaded by the position factor was not statistically significantly different from the percentage of randomly selected fluent words loaded by
the same factor [PDS: \( t(34)=0.20, p=0.84 \); ANS: \( t(4)=0.25, p=0.82 \)]. However, both groups did show statistically greater load percentages for the stuttered words than for the randomly selected fluent words for the length factor [PDS: \( t(34)=8.98, p<.001, d=1.52 \); ANS: \( t(4)=6.18, p=.003, d=2.76 \)], the phonetic factor [PDS: \( t(34)=3.17, p=.003, d=0.54 \); ANS: \( t(4)=2.88, p=.045, d=1.29 \)], and the grammatical factor [PDS: \( t(34)=5.53, p<.001, d=0.93 \); ANS: \( t(4)=4.51, p=.011, d=2.02 \)]. Note that again five of the six statistically significant effects were associated with large effect sizes, the only exception being the phonetic factor in the PDS group which was associated with a medium effect size.

Lastly, we verified whether the above described outcomes for the position and length factors may apply only when these factors are defined in the traditional manner; that is, by considering the first three words of the sentence for the position factor and words of five or more letters for the length factor (Brown, 1945). When data for the position factor were recalculated with a weight assigned to any word that occurred as one of the first three words of a clause, the absolute load percentages were descriptively greater for both groups of speakers and for both the stuttered and randomly selected fluent words because there are more clauses than sentences in a typical reading passage. Most importantly, however, Figure 5 shows that the overall pattern of results remained the same, with again no statistically significant word position effects for either the PDS group [\( t(34)=0.57, p=.57 \)] or the ANS group [\( t(4)=0.59, p=.58 \)]. The pattern of results for the length factor also remained unchanged when defining long words as those with five or more sounds rather than five or more letters: Figure 6 illustrates that both the PDS group and the ANS group still showed a statistically significant (and large effect size)
difference in load percentage between the stuttered words and the randomly selected fluent words [PDS: $t(34)=8.64, p<.001, d=1.46$; ANS: $t(4)=3.32, p=.029, d=1.48$].

Discussion

The primary aim of this work was to apply in both PDS and ANS a similar set of classic analyses that evaluate the influence of phonetic, word class, word length, and word position variables on the distribution of stuttering moments within utterances. Words stuttered during oral reading were identified off-line in audio-recordings of 5 adults with ANS and 35 adults with PDS. Brown’s (1945) word weights were calculated for stuttered vs. fluent words by assigning a point to each word that (a) started with a consonant; (b) was a noun, adjective, adverb, or verb; (c) was 5 or more letters long; and (d) was one of the first three words in a sentence. Analyses based on alternative definitions of word length and word position were also conducted.

Five main findings were obtained. First, analyses based on the combination of all four factors showed that for both the PDS and ANS groups Brown’s word weights were greater for stuttered vs. fluent words. Second, these results—and the associated large effect sizes—were obtained regardless of whether the word weights for fluent words were calculated by averaging across all fluent words (i.e., a number of words that is many times greater than the number of stuttered words) or by averaging across only a subset of fluent words that was selected to match the number of stuttered words. Third, when considering the influence of each factor separately, both groups showed a greater level of loading on stuttered words than fluent words for the length factor, the phonetic factor, and the grammatical factor. Five of these six comparisons were again associated with large effect sizes (one comparison was associated with a medium effect size). Fourth, one
Loci of dysfluency in stuttering

surprising finding that is not in line with a long-standing view regarding the loci of stuttering in PDS is that such greater loading on stuttered vs. fluent words was not found for the position factor. For neither the PDS group nor the ANS group was the percentage of stuttered words that occurred in an early position in the sentence (i.e., among the first three words) greater than the percentage of randomly selected fluent words that occurred in this position. Fifth, the presence of an effect of the length factor and the absence of an effect of the position factor were confirmed to hold when these factors were defined in terms of the number of speech sounds (rather than the number of letters) and early position within a clause (rather than early position within a sentence), respectively.

The novel finding that the same factors influencing the loci of dysfluency in PDS also operate in ANS is of both theoretical and clinical interest. On the one hand, this finding could be considered unsurprising in light of previous work demonstrating that the two disorders share similar phenomenological characteristics. Indeed, even speech-language pathologists can find it difficult to distinguish audio-recordings from individuals with ANS vs. PDS (Van Borsel & Taillieu, 2001). On the other hand, the observed similarity in loci of stuttering in ANS and PDS can be considered unexpected given that potentially different underlying mechanisms are suggested by our own previous research revealing that external situational influences differ between ANS and PDS. For example, many ANS patients tend to not show a decrease in stuttering frequency when repeatedly reading the same passage or when hearing delayed or frequency-shifted auditory feedback whereas these conditions are well known to be fluency-enhancing for most individuals with PDS (Balasubramanian et al., 2003; Balasubramanian et al., 2010; De Nil et al., 2018; Gray, 1965; Max & Caruso, 1998;
Loci of dysfluency in stuttering

Newman, 1963; Theys et al., 2013). Moreover, modern neuropsychology often suggests that even similar but slightly different brain lesions can sometimes result in very selective impairments, for example with differential effects on lexical access of words from different conceptual categories or nouns vs. verbs (Ullmann, 2007). Given the possibility of such lesion-specific effects and the wide variety of brain lesions that may lead to ANS (see the above Method section for information on the current subjects’ neurological findings), one might have predicted not only different loci effects for ANS and PDS but also large between-subject variability within the ANS group. Neither of these potential outcomes were observed in the present study. Instead, each of the five individuals with ANS showed the same word weights effect seen in the averaged PDS data.

Specifically, for both the ANS group and the PDS group, the distribution of stuttering moments showed an effect of the phonetic factor, the length factor, and the grammatical factor. That is, stuttered words—in comparison with fluent words—were more likely words that started with an initial consonant, that were five or more letters (or five or more speech sounds) in length, and that were content words (per the specific operational definition provided in the Method section). The effects of these three factors had been previously observed in PDS with great consistency (for reviews, see St. Louis, 1979; Wingate, 2002), and the present findings add that the effects also apply to ANS.

One possible interpretation is that these particular factors represent the influence of motor variables affecting speech output regardless of the specific mechanisms and neural substrates that underlie the two clinical populations’ disruptions in speech fluency. In contrast with vowels, consonants require oral cavity obstructions and constrictions (partially or completely interrupting airflow) and the precise coordination of oral and
laryngeal muscle activity to distinguish voiced and voiceless consonants. The added motor complexity associated with producing these sounds and transitioning into the subsequent vowel may make breakdowns in fluent speech more likely. Longer words obviously consist of a longer sequence of speech sounds and, thus, are produced as a longer sequence of articulatory gestures. In addition, longer words are more likely than shorter word to start with an initial consonant (Taylor 1966a). Thus, the length factor, too, can be interpreted as resulting from increased motor complexity.

Interpreting the grammatical factor as also representing motor influences might seem more controversial at first, but (a) content words are on average longer than function words (Taylor, 1966a; Wingate, 2002), and (b) content words (as is the case for longer words in general) also start more frequently with an initial consonant whereas proportionally more function words start with a vowel (e.g., Taylor, 1966a, Wingate, 2002). In fact, Taylor (1966a) found that the influence of the grammatical factor on the loci of stuttering was completely accounted for by the influence of the phonetic factor: there was no different effect of content vs. function words when considering only words with an initial consonant or only words with an initial vowel. Moreover, content words form an open class that is much larger than the closed class of function words, and this causes individual content words to be used much less frequently than individual function words (Dayalu et al., 2002; Wingate, 2002). In a study by Dayalu et al. (2002), content words were still more frequently stuttered than function words when both grammatical classes were produced in lists of isolated words (thus as much as possible dissociated from their grammatical role in sentences) and when “matched for initial sound and approximate number of syllables” (p. 871). Dayalu et al. (2002) therefore attributed the
difference in stuttering moments to the difference in frequency of use for these two grammatical categories. It is indeed well known that repeated, more frequent production of words is associated with an adaptation effect (i.e., reduction in stuttering) due to motor learning (Dayalu et al., 2002; Max & Baldwin, 2010; Max & Caruso, 1998).

An unexpected finding of the present study is the absence of a word position effect. That is, our data sets for both the PDS group and the ANS group showed no support for the widely accepted notion that stuttering moments are more likely to occur in the beginning of an utterance, at least not if defined as the first three words of a sentence or the first three words of a phrase. In light of the aforementioned interdependence of some of the word weight factors, this intriguing finding suggests the possibility that prior findings of such a word position effect may have been caused by overlap with the phonetic, grammatical, and length factors. Interestingly, Taylor (1966b) already pointed out many years ago that most studies did not consider “the fact that certain grammatical classes assume particular positions in sentences” (p. 236). Similarly, Wingate (1979, 2002) showed that in much English prose, again including the reading material used in the series of studies by Brown (1945), the first three words of sentences are much more likely to be content words (and, thus, also longer words—see above) rather than function words. In an earlier study, Wingate (1979) had used reading materials with predominantly function words in the first three positions of the sentences. He found that there was not a higher frequency of stuttering (actually, it was descriptively slightly lower) on the first three words of sentences than on the remaining words. Hence, at least some reports of a word position effect on the probability of stuttering may have been confounded by the grammatical factor (and/or the phonetic and length factors—see above
for how the three variables are interrelated). We therefore completed a post-hoc analysis on the “Washington” passage of the SSI-4 (the passage read by all PDS participants in the present study; similar analyses were not completed for the ANS passages because each passage was read by only one single participant). Interestingly, this analysis verified that this particular passage is not confounded in such a manner. Assigning words a point for having an initial consonant, being a content word, or being 5 or more letters long resulted in an average score of 1.48 ($SD$ 0.93) for the first three words of the sentences versus 1.85 ($SD$ 1.05) for the remaining words of the sentences. Although further replication will be necessary, it is remarkable that our relatively large sample of 35 PDS participants showed no word position effect when reading this passage in which the first three words of the sentences were not also more heavily loaded by the other three factors.

In conclusion, an extensive analysis of the within-utterance loci of dysfluency revealed highly similar results for ANS and PDS. Both groups showed greater average word weights for stuttered vs. fluent words when assigning weights based on the word starting with an initial consonant; being five or more letters long; being a verb, noun, adjective, or adverb; or occurring as one of the first three words of a sentence. For both groups, stuttered words were also loaded more by each of three factors individually (namely the phonetic factor, length factor, and grammatical factor). Unexpectedly (based on prior literature), neither group of subjects showed greater loading for stuttered words vs. fluent words when considering only the position factor. Thus, taken together, the overall findings of this study suggest similar loci of stuttering in adults with PDS and ANS. An interpretation that takes into account the substantial interrelation among several
Loci of dysfluency in stuttering

23

of the factors suggests that, for both groups, the probability of stuttering on a given word is influenced more by motor production variables than true linguistic variables.

It should be noted that we do not wish to imply that our findings on a particular behavioral characteristic of ANS and PDS (i.e., in the loci of stuttering moments) suggest overlap in the neural substrates and mechanisms underlying both disorders. The extremely large and highly variable range of both left and right hemisphere cortical (including frontal, parietal, and temporal lobe) and subcortical (including internal capsule, striatum, thalamus, corpus callosum, midbrain, and pons) lesions or degeneration sites that can lead to ANS are very well documented (e.g., Abe, Yokoyoma & Yrifji, 1992; Balasubramanian & Hayden, 1995; Balasubramanian et al, 2003; Balasubramanian et al., 2010; Carluer et al, 2000; Doi et al., 2003; Fleet & Heilman, 1985; Hamano et al., 2005; Jokel, De Nil, & Sharpe, 2007; Lebrun & Leleux, 1985; Ludlow, Rosenberg, Salazr, Grafman, & Smurtok, 1987; Soroker, Bar-Israel, Schechter, & Solzi, 1990; Theys, De Nil, Thijs, van Wieringen, & Sunaert, 2013; Turgut, Utuku & Balci, 2002; Van Borsel, van der Made, & Santens, 2003). PDS, on the other hand, occurs in the absence of actual lesions, although structural abnormalities in white and grey matter and functional hyper- and hypo-activations have also been documented in distributed neural circuits (for reviews, see Craig-McQuaide, Akram, Zrinzo, & Tripoliti, 2014; Etchell, Civier, Ballard, & Sowman, 2018; Neef, Anwander, & Friederici, 2015). Of course, in some cases there will be overlap in the brain regions that are affected by lesions in at least some individuals with ANS and the developmental structural and functional brain characteristics in individuals with PDS (Theys et al., 2013). However, rather than suggesting similar underlying neural and sensorimotor mechanisms for the two disorders,
we propose that finding the described set of identical loci effects *despite* the acquired vs. developmental neural bases can be interpreted as an indication that these effects reflect intrinsic influences of the final stages of speech motor preparation or execution rather than higher-level influences of linguistic processing.

Of course this concluding hypothesis requires future testing. In particular, a number of limitations and caveats should also be considered given that all our data were obtained by analyzing stuttering moments in PDS and ANS speech samples that were originally collected for different purposes (e.g., evaluating stuttering severity). For example, as in most other studies on the loci of stuttering, our reading passages were not constructed in a manner that would allow a fully dissociated analysis of each individual factor. Perhaps even more important for future work is that some loci effects have been claimed to differ in children who are still close to the onset of stuttering vs. adults who stutter. Specifically, children with PDS have been reported to stutter more on function words than on content words, and this finding has been reported for several languages (Au-Yeung, Gomez, & Howell, 2003; Au-Yeung et al., 1998; Dworzynski, Howell, Au-Yeung, & Rommel, 2004; Gkalitsiou, Byrd, Bedore, & Taliancich-Klinger, 2017; Howell, Au-Yeung, & Sackin, 1999; Juste, Sassi, & de Andrade, 2012; Natke, Sandrieser, van Ark, Pietrowsky, & Kalveram, 2004). The exact nature of this phenomenon remains unclear, however, as other work has shown more stuttering on content words even in 7-10-year-old Persian children (Vahab, Zandiyan, Falahi, Howell, 2013). Furthermore, recent studies by Buhr, Jones, Conture, and Kelly (2016) for English and Vahab et al. (2013) for Persian both demonstrated that children who stutter produce part-word repetitions more frequently on content words than on function words. The
same children’s function words were more frequently associated with whole-word repetitions, a type of disfluency that according to some authors should not even be included as a primary symptom of stuttering (Vahab et al., 2013; Wingate, 2001; Wingate, 2002). In fact, neuroimaging work has shown that adult stuttering speakers’ brain activity associated with whole-word repetitions is more similar to that of disfluencies not typical of stuttering than those that are typical of stuttering (Jiang, Lu, Peng, Zhu, & Howell, 2012). This long-standing debate about the inclusion of word repetitions as instances of stuttering cannot be resolved here, but relates to another, final limitation: word repetitions, especially those occurring on function words, may represent attempts to “stall” before an upcoming location where a part-word repetition or sound prolongation is going to occur. Hence, it remains unknown whether the loci of true involuntary fluency breakdowns are different in children vs. adults and whether or not either narrative or reading data from childhood stuttering would lead to different conclusions than those drawn in the present work based on oral reading samples from adults with PDS or ANS. Nevertheless, the main conclusion from the study described here remains that the loci of stuttering in oral readings of adult patients with ANS—who, just like stuttering children, have had fewer years of experience with the behavioral and psychological consequences of stuttering—are highly similar to those of adults with PDS.

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Loci of dysfluency in stuttering

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References


Loci of dysfluency in stuttering


Figure Captions

Figure 1
Percent stuttered syllables during oral reading for the entire group of adults with persistent developmental stuttering (n=35, the error bar indicates standard error of the mean) and for five individual adults with acquired neurogenic stuttering (ANS1 … ANS5).

Figure 2
Brown’s Word Weight group means (and standard errors of the mean) for adults with persistent developmental stuttering and adults with acquired neurogenic stuttering. For each speaker, word weights were calculated for all stuttered words, for all fluent words, and for a number of randomly selected fluent words that equaled the number of stuttered words. Both groups of participants showed higher word weights for stuttered words than for fluent words regardless of whether all fluent words or only the subset of fluent words were used. Asterisks indicate p < .05.

Figure 3
Brown’s Word Weight group data (mean and standard error of the mean) for 35 adults with persistent developmental stuttering and individual subject data for 5 adults with acquired neurogenic stuttering (ANS1 … ANS5). Each of the individuals with ANS showed the same pattern as the developmental stuttering group with higher word weights for stuttered words than for fluent words.
Figure 4
Data for each of the four Brown factors separately. Group data (means and errors of the mean) for developmental stuttering and acquired neurogenic stuttering are shown as the percentage of stuttered versus fluent words loaded by each factor. Results were the same for both groups of subjects: the distribution of stuttering showed no effect of the position factor (the word occurred as one of the first three words of the sentence) but was affected by the length factor (the word was 5 or more letters long), the phonetic factor (the word started with a consonant), and the grammatical factor (the word was a noun, adjective, adverb, or verb). Asterisks indicate p < .05.

Figure 5
Data for the position factor re-analyzed based on clauses rather than sentences (i.e., a word is loaded for the factor if it occurs as one of the first three words of a clause). Group data (means and errors of the mean) for developmental stuttering and acquired neurogenic stuttering are shown as the percentage of stuttered versus fluent words loaded by this factor. Results for the distribution of stuttering moments remained unchanged relative those shown in Figure 4: neither group showed a position effect.

Figure 6
Data for the length factor re-analyzed with long words defined as 5 or more sounds rather than 5 or more letters. Group data (means and standard errors of the mean) for developmental stuttering and acquired neurogenic stuttering are shown as the percentage of stuttered versus fluent words loaded by this factor. Results remained unchanged
relative those shown in Figure 4: both groups of subjects showed a similar effect of word length on the distribution of stuttering moments.
Figure 1

[Bar chart showing percent stuttered syllables for developmental stuttering and ANS1 to ANS5]
Figure 2

![Bar chart showing Brown's Word Weight for Stuttered words, All fluent words, and Random fluent words. Asterisks indicate significant differences.]
Figure 3

The figure shows a bar chart comparing Brown's Word Weight between stuttered words and random fluent words across different conditions. The conditions are labeled as Developmental stuttering, ANS1, ANS2, ANS3, ANS4, and ANS5. The chart indicates a higher Brown's Word Weight for stuttered words compared to random fluent words in most conditions.
Figure 4

Loci of dysfluency in stuttering

- **Position Factor**
  - Stuttered words vs. Random fluent words
  - Developmental stuttering vs. Neurogenic stuttering

- **Length Factor**
  - Developmental stuttering vs. Neurogenic stuttering

- **Phonetic Factor**
  - Developmental stuttering vs. Neurogenic stuttering

- **Grammatical Factor**
  - Developmental stuttering vs. Neurogenic stuttering
Figure 5

Position Factor (clause)

Words with Load (%)

- Stuttered words
- Random fluent words

Developmental stuttering
Neurogenic stuttering
Figure 6

Length Factor (sounds)

- Stuttered words
- Random fluent words

Words with Load (%)

Developmental stuttering

Neurogenic stuttering

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