Atypical Cortical Tracking of the Speech Envelope in Children Who Stutter: A Potential Contributor Towards Phonological Processing Differences

by

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A growing body of evidence suggests that individuals with developmental stuttering exhibit phonological processing differences when compared to fluent peers. However, it has yet to be unveiled which factors may contribute towards this atypical processing. It has been argued that the speech mechanisms which process these phonological units are monitored within a hierarchical system, whose foundation is controlled by low-frequency neural oscillating networks (Giraud & Poeppel, 2015). Thus, phonological processing differences may arise due to impairments in fundamental mechanisms associated with low-frequency neural oscillating networks, such as temporal speech encoding. For this reason, this study sought to investigate cortical temporal response functions in 14 children who stutter (3-7 years of age) compared to 13 normally fluent peers. EEG data were recorded as participants encoded natural speech during a dichotic listening task. When comparing between groups, the results provide evidence that children who stutter experience significantly weaker cortical tracking for unattended speech and more efficient cortical tracking for attended speech, suggesting that phonological processing is atypical at the level of speech envelope encoding. Considering these findings, we propose that children who stutter may be increasing cognitive effort during speech and language processing, in order to compensate for an atypical phonological processing mechanism.

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Preface

This thesis would not have been possible without the guidance and support of Dr. Bharath Chandrasekaran and Dr. Amanda Hampton. Thank you both for dedicating your time and expertise to assist me with this endeavor amongst your numerous other obligations. Your collaboration has allowed me to connect my interests in stuttering and neuroscience into one amazing and exciting project. Additionally, I would like to thank Nike Gnanateja Gurindapalli for showing me the steps to preprocess EEG data, along with teaching me how to analyze my data and understand the results. Your guidance has significantly helped me accomplish this project. I would also like to thank Dr. Lauren Calandruccio for traveling from Cleveland to act as my external examiner and Dr. Jason Bohland for acting as a committee member. Lastly, I would like to thank my Mother, Father, and friends for being very good listeners, even though you all had no idea what I was talking about. I am incredibly grateful for everyone's help and support.

1.0 Introduction

Stuttering is a fluency disorder characterized by involuntary disruptions in speech known as repetitions, prolongations, or blocks. The individual knows exactly what they wish to say, but has trouble expressing their words overtly. Stuttering occurs in nearly 1 in every 100 individuals (Guitar & Conture, 2006), and can often have detrimental effects on one's confidence to communicate. These emotional effects may persist throughout the individual's lifespan, impacting both their personal and professional experiences. Developmental stuttering has been shown to typically arise around 3-5 years of age, when there are rapid demands for language, motor, emotional, and cognitive development (Guitar & Peters, 2014). This suggests that there may be a relationship between the high cognitive demands required by linguistic processes and the onset of this disorder. Although most children recover, approximately 25% continue to stutter into adulthood (NIDCD, 2017). Despite extensive knowledge within the field, the exact causes of stuttering remain elusive. However, research has linked gender, age of onset, and genetics as major predictors for persistence or recovery (Guitar & Peters, 2014).

While the covert repair hypothesis (Postma & Kolk, 1993) emphasizes differences in phonology and phonological processing mechanisms as the root of stuttering, it is becoming increasingly clear that other factors interacting within the speech motor system partake in the development of this disorder. More specifically, the multifactorial dynamic pathways theory posits that developmental stuttering is primarily a disorder disrupting the sensorimotor control of speech that is influenced by supplementary cognitive, linguistic, and emotional factors (Smith & Weber, 2017). Based on this model, phonology may act as one contributor in the phenotypic expression and variable overt speech characteristics of this disorder.

1.1 Phonological Disorders in Individuals Who Stutter

Several studies have provided evidence for a link between stuttering and phonological disorders. In past research, it was discovered that 30-40% of children who stutter (CWS) may experience additional phonological disorders, compared to an incidence of 2-6% in the general population (Beitchman et al., 1986; Bernstein Ratner, 1995; Louko, 1995; Wolk, 1998; Melnick & Conture, 2000). However, it is important to note that when comparing the results between studies, there were not consistent methods for classifying phonological disorders. Regardless, this information still holds significance. Research has also proposed that phonological disorders may influence the overt speech characteristics in individuals who stutter. Wolk, Conture, & Edwards (1993) found that individuals with stuttering and co-existing phonological disorders experienced significantly more sound prolongations and fewer iterations of whole-word repetitions than children who only experienced stuttering.

In addition, it is suggested that performance on phonological tasks may predict stuttering persistence or recovery. In a longitudinal study conducted by Spencer and Weber-Fox (2014), they examined phonological skills using the Bankson-Bernthal Test of Phonology, Consonant Inventory subtest (BBTOP-CI; Bankson & Bernthal, 1990) and the Dollaghan and Campbell nonword repetition test (NWR; Dollaghan & Campbell, 1998). Participants were re-assessed each year, ranging from 1-4 years after the initial study visit, to determine which children recovered or continued to stutter. Results indicated that poorer performance on these two tests correlated with stuttering persistence. It is also worth noting that 13 CWS displayed delayed phonological abilities (represented by a score greater than one SD below the normative mean on the BBTOP-CI) and of these 13 children, nine persisted and 4 recovered. These reports suggest that stuttering and phonology may be associated with one another.

1.2 Behavioral Performance During Phonological Tasks

Even without the consideration of co-occurring phonological disorders, research indicates that individuals with chronic stuttering may experience further difficulties with phonological tasks. Hakim and Ratner (2004) sought to determine if CWS exhibited more errors during the Children's Test of Nonword Repetition (CNRep; Gathercole, Willis, Baddeley, & Emslie, 1994). Their results showed a higher percent of phonemic errors in children who stutter across all syllable lengths, with significant group differences at the 3-syllable level. However, the researchers acknowledged that this may be due to ceiling and floor effects for the longer and shorter syllable lists. A larger sample size was also needed. To address these concerns, Anderson, Wagovich, and Hall (2006), replicated the experimental design using a larger sample size of young children with a narrower age range. Their findings supported Hakim and Ratner's work; CWS produced significantly less 2- and 3-syllable nonwords correctly and CWS had significantly more phoneme errors at the 3-syllable level. Taken together, these findings suggest that individuals who stutter may exhibit poorer performance on nonword repetition tasks.

Mahesh, Geetha, Amulya, and Ravel (2018) also found that CWS had significantly slower speeds in monitoring phonemes in word initial and medial positions, which provides evidence that individuals who stutter may experience difficulties encoding phonological units. Other studies examining phoneme monitoring have noted significant differences in phonological performance, especially as cognitive demands increase. For instance, Sasisekaran and Byrd (2013) found that CWS took longer to monitor consonant clusters than singletons, particularly those in the syllable offset position.

1.3 ERP Evidence

To date, only three studies have been completed examining the neurophysiological mechanisms of phonology in individuals who stutter. However, these few studies have found significant results. For instance, Weber-Fox et al. (2004), found atypical neural processing in adults who stutter (AWS) during a rhyme judgement task when cognitive demands were highest. Otherwise, accuracy and reaction times between AWS and adults who do not stutter (AWNS) were similar. Event-related potentials (ERPs) showed that AWS engaged greater right hemisphere activation for late cognitive processes mediating rhyme decisions. As a follow-up to this study, Weber-Fox et al. (2009) assessed the behavioral performance and ERPs in CWS completing the same rhyming task. Results showed that CWS had significantly lower accuracy of rhyming judgements when compared to the fluent group. In addition, the peak latency of the N400 was earlier in the right hemisphere in CWS, whereas this pattern occurred over the left hemisphere in children who do not stutter (CWNS). These results provide ERP evidence for phonological processing differences in CWS, even in the absence of engaging the speech-motor system.

Neural activity for rhyme judgements may also act as a predictor for stuttering persistence or recovery. Mohan and Weber (2015) compared the neural activity of three groups: children who stutter, children who recovered from stuttering, and children who were fluent. Upon analyzing the behavioral results, all groups were highly accurate in their rhyme judgement performance. When evaluating event-related potentials, peak latency and mean amplitude of the N400s elicited by the stimuli indicated a typical ERP central-parietal rhyme effect. However, over anterior electrode sites, this effect was absent in children with persistent stuttering, occurred bilaterally in children who do not stutter, and was greater over the right hemisphere in children who recovered. The results suggest that even when there are not significant performance differences, CWS may still exhibit atypical neural activity for phonological processes.

1.4 Cortical Tracking of the Speech Envelope

It is evident that individuals who stutter may exhibit differences in phonological processing at many levels. In addition to this, it has been argued that the speech mechanisms which process phonological units are controlled within a hierarchical system, whose foundation lies in low-frequency neural oscillating networks (Giraud & Poeppel, 2015). Therefore, phonological processing differences may arise due to impairments in these fundamental low-frequency oscillating mechanisms, such as temporal speech encoding.

The temporal properties of speech sounds have recently been of great interest in auditory neuroscience. While the auditory system primarily acts as a frequency analyzer, it is now widely acknowledged that place-frequency mechanisms cannot solely account for speech perception. Physiological evidence suggests that temporal information is integral for both the perception of melodic pitch and the auditory depiction of spectral shape (Sachs & Miller 1985; Sachs et al. 1983). Rosen (1992) developed a framework for describing the temporal fluctuations of speech, which included three features: envelope, fine-structure, and periodicity. Each of these features conveys different areas of phonological information, such as segmental cues, voicing, and manner of articulation. In more recent literature, researchers have supported Rosen's notions with similar findings from low-frequency oscillations (Doelling et al., 2014; Ding et al., 2017; Ghitza, 2017). Therefore, inefficient encoding of temporal speech envelope may account for the phonological performance and processing differences that have been observed in children who stutter.

One recent machine learning approach, which provides insight into the human ability to encode the temporal properties of speech, involves cortical tracking of the speech envelope. This involves analyzing the synchronization between speech-evoked neurophysiological responses and the acoustic structure of speech. A filter is applied, known as the Multivariate Temporal Response Function (mTRF), which describes the linear mapping between features of the auditory stimuli and the neural response. Low-frequency cortical oscillations phase-lock to acoustic characteristics of the speech envelope, depending on how robustly the brain encodes this information (Peelle et al., 2013). Temporal Response Functions (TRFs) therefore describe how efficiently the brain encodes temporal features in speech.

2.0 Current Study

2.1 Aims and Hypothesis

The current study aims to analyze cortical tracking of the temporal speech envelope in CWS, which may act as a contributor toward phonological processing differences. A thorough understanding of phonological processing abilities in individuals who stutter would lead to more robust theories and clinical treatments, therefore improving fluency outcomes. Temporal response functions (TRFs) were analyzed in CWS, which describe how efficiently the brain encodes temporal speech information. Since it is suggested that these temporal properties act as a foundational framework for phonological processing, a deficit at this foundational level would likely have implications for all phonological processing abilities. Therefore, we hypothesized that CWS may have atypical cortical tracking to the temporal speech envelope, given phonological processing differences that have been noted in literature (Beitchman et al., 1986; Ratner, 1995; Louko, 1995; Wolk, 1998; Melnick & Conture, 2000; Wolk, et al., 1993; Spencer & Weber-Fox, 2014; Hakim & Ratner, 2004; Anderson, et al., 2006; Mahesh et al., 2018; Mohan & Weber, 2015). This task requires auditory attention and the attention deficits that have been seen in CWS (Felsenfeld et al., 2010; Karrass et al., 2006; (Bosshardt, 1999, 2002, 2006; Bosshardt et al., 2002; Vasic & Wijnen, 2005), may influence both speech envelope processing and phonological encoding.

2.2 Materials and Method

2.2.1 Participants and Screening

Participants were recruited from the mid-Michigan community via flyers and word-of-mouth from speech-language pathologists and pediatricians. Twenty-seven children, ranging from 3-7 years of age, were included in the present study. Of these children, 14 were children who stutter (M = 5.40 years, SD = 1.15, 8M) and 13 were perceptually fluent peers (M = 5.11, SD = 1.28, 8M) with no presence or history of stuttering. Data collection was completed at Michigan State University and all procedures were approved by Michigan State University's Institutional Review Board.

All participants were native, monolingual speakers of English with no history of neurological disease or injury, and no language, reading, visual, or hearing impairments, other than stuttering for the experimental group. Each child was required to pass a hearing screening at 20 dB HL at 500, 1000, 2000, 4000, 6000, and 8000 Hz bilaterally. In addition, all participants performed within or above one SD of the norm-based mean on a nonverbal intelligence quotient (IQ) task, the Primary Test of Nonverbal Intelligence (PTONI; Ehrler & McGhee, 2008). The socioeconomic status (SES) of each child was coded by trained research assistants, in accordance with the Hollingshead Four Factor Index of Social Status (Hollingshead, 1975). For CWS, the mean SES was 44.32 (SD = 12.99). For CWNS, the mean SES was 48.23 (SD = 13.64). An independent samples t-test indicated no significant differences between groups ($p > .05$). In addition, all children exhibited normal language skills for their age determined by either the Clinical Evaluation of Language Fundamentals – Preschool – Second Edition (CELF-P2; Wiig, Secord, & Semel, 2004) or the Clinical Evaluation of Language Fundamentals–Fifth Edition

(CELF-5; Wiig, Semel, & Secord, 2013). For the CWNS, one child was ambidextrous, and all others were right-handed. For the CWS, four children were ambidextrous, one child was left-handed, and all others were right-handed. This was determined by the Edinburgh Inventory for Assessment of Handedness (Oldfield, 1971).

A child was diagnosed with stuttering if: 1) stuttering severity was rated as a 2 or greater on an eight-point (0–7) scale by a speech-language pathologist (0 was equivalent to no stuttering and 7 was equivalent to the greatest severity of stuttering); 2) stuttering severity was rated as a 2 or greater on the same eight-point (0–7) scale by the parent; and 3) the child displayed at least three stuttering-like disfluencies (SLDs) per 100 syllables during a language sample acquired in the lab. SLDs included part-word repetitions, sound prolongations, and/or silent blocks.

Table 1. Participant Information

ID	Group	Sex	Age	SSI Severity	ID	Group	Sex	Age
1	CWS	M	4.17	Mild	15	CWNS	F	3.75
2	CWS	M	3.33	Moderate	16	CWNS	M	4.75
3	CWS	F	7.75	Moderate	17	CWNS	M	5.17
4	CWS	M	5.58	Moderate	18	CWNS	F	3.75
5	CWS	F	5.42	Mild	19	CWNS	F	5.00
6	CWS	F	4.50	Very Mild	20	CWNS	F	6.25
7	CWS	M	6.33	Moderate	21	CWNS	M	4.75
8	CWS	M	6.50	Moderate	22	CWNS	M	5.25
9	CWS	M	4.58	Moderate	23	CWNS	M	4.08
10	CWS	F	6.00	Moderate	24	CWNS	M	7.92
11	CWS	F	6.25	Moderate	25	CWNS	M	4.67
12	CWS	M	4.67	Mild	26	CWNS	M	7.08
13	CWS	F	5.92	Mild	27	CWNS	F	4.00
14	CWS	M	4.67	Mild				

Note. SSI = Stuttering Severity Instrument

In addition, the Dollaghan and Campbell nonword repetition test (NWR; Dollaghan & Campbell, 1998) was administered to each child. The NWR test consisted of a set of 16 nonsense words ranging from one to four syllables in length. The child was required to accurately repeat the stimulus item after hearing each recorded token once. Scoring followed the guidelines described by Dollaghan and Campbell (1998) and was calculated as the number of correct phonemes produced in each nonsense word. Since the total number of syllables varied across syllable lengths, percent accuracy was calculated for each syllable length. The children's performances on the NWR task at all syllable lengths were analyzed using a repeated measures ANOVA. From the CWNS group, one child refused to complete the task (RTD) and one child did not understand the task (DNU). Therefore, the data from these two children were excluded from analysis. The CWS and CWNS performed similarly across all syllable lengths on the NWR task, [$F(1, 23) = 0.29$, $p = .59$].

Table 2. Individual Scores from Nonword Repetition Task

ID	Group	1 Syllable Score	2 Syllable Score	3 Syllable Score	4 Syllable Score
1	CWS	10	19	20	21
2	CWS	11	17	24	1
3	CWS	11	17	24	27
4	CWS	11	17	25	18
5	CWS	10	14	18	11
6	CWS	11	15	18	23
7	CWS	11	19	23	29
8	CWS	11	18	28	34
9	CWS	10	18	12	9
10	CWS	11	19	26	32
11	CWS	10	16	24	23
12	CWS	9	16	15	10
13	CWS	11	13	24	26
14	CWS	12	20	18	11
15	CWNS	RTD	RTD	RTD	RTD
16	CWNS	10	18	18	27
17	CWNS	11	17	18	9
18	CWNS	9	17	14	17
19	CWNS	8	19	21	18
20	CWNS	11	20	27	27
21	CWNS	8	18	23	16
22	CWNS	11	20	21	7
23	CWNS	10	16	26	23
24	CWNS	12	20	28	32
25	CWNS	12	19	25	14
26	CWNS	10	20	27	34
27	CWNS	DNU	DNU	DNU	DNU

Note. RTD = refused to do; DNU = did not understand. The highest possible score for each condition was as follows: 1 syllable (12), 2 syllable (20), 3 syllable (28), and 4 syllable (36).

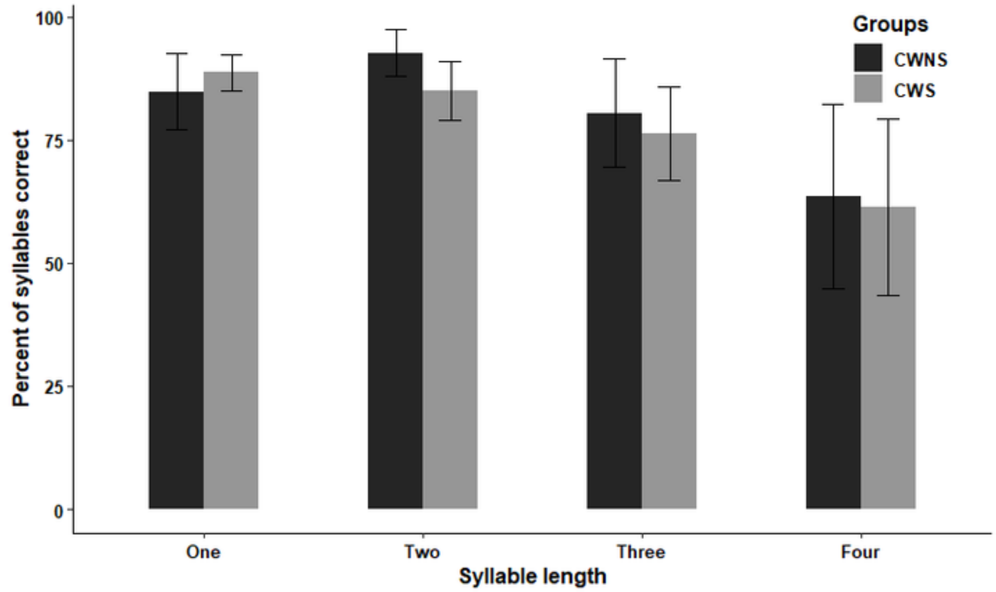

Figure 1. Percent of Syllables Correct on NWR Task for CWS and CWNS

Note. Error bars indicate one standard error from the mean

Given that our study tests inhibition and switching processes in young children, we included the Shape School task (Espy, 1997) as part of our test battery. This task involves individually administering a storybook designed to examine inhibition, cognitive flexibility, and attention in young children. There were five conditions: color naming, color inhibit, shape naming, color-shape switch, and color-shape inhibit. The last condition, color-shape inhibit, was only completed by some of the children due to the difficulty level of the task.

The results from the Shape School tasks were analyzed using a repeated measures ANOVA. One child in the CWS group RTD the Color-Shape-Switch condition and one child in this group DNU this condition. In addition, in the CWNS group one child RTD all conditions of the Shape School task and one child from this group DNU the Shape Naming condition. Data for incomplete or misunderstood tasks were not included in analysis. Also, the inhibition-switch condition was excluded from analysis, due to a limited number of children being able to complete the task. The CWS and CWNS performed similarly across all conditions, [$F(1, 22) = .08$, $p = .78$].

Table 3. Individual Scores from Shape School Task

ID	Group	Color Naming	Color Naming Inhibition	Shape Naming	Color-Shape Switch
1	CWS	10	17	10	12
2	CWS	8	14	9	RTD
3	CWS	10	18	11	16
4	CWS	12	16	8	DNU
5	CWS	9	14	9	6
6	CWS	12	18	10	15
7	CWS	9	17	10	11
8	CWS	9	17	11	15
9	CWS	11	17	12	13
10	CWS	10	17	12	12
11	CWS	11	16	9	13
12	CWS	9	4	11	8
13	CWS	10	18	10	16
14	CWS	12	18	11	16
15	CWNS	RTD	RTD	RTD	RTD
16	CWNS	10	16	12	14
17	CWNS	11	17	11	15
18	CWNS	9	15	10	7
19	CWNS	12	5	9	16
20	CWNS	12	17	10	14
21	CWNS	10	16	7	10
22	CWNS	9	14	DNU	1
23	CWNS	11	18	11	8
24	CWNS	12	18	12	13
25	CWNS	11	18	10	9
26	CWNS	12	17	11	12
27	CWNS	12	18	11	12

Note: RTD = refused to do; DNU = did not understand. The highest accuracy scores that could be achieved in each condition were: Color naming (12), Color-naming inhibition (18), Shape naming (12), and Color-Shape Switch (16).

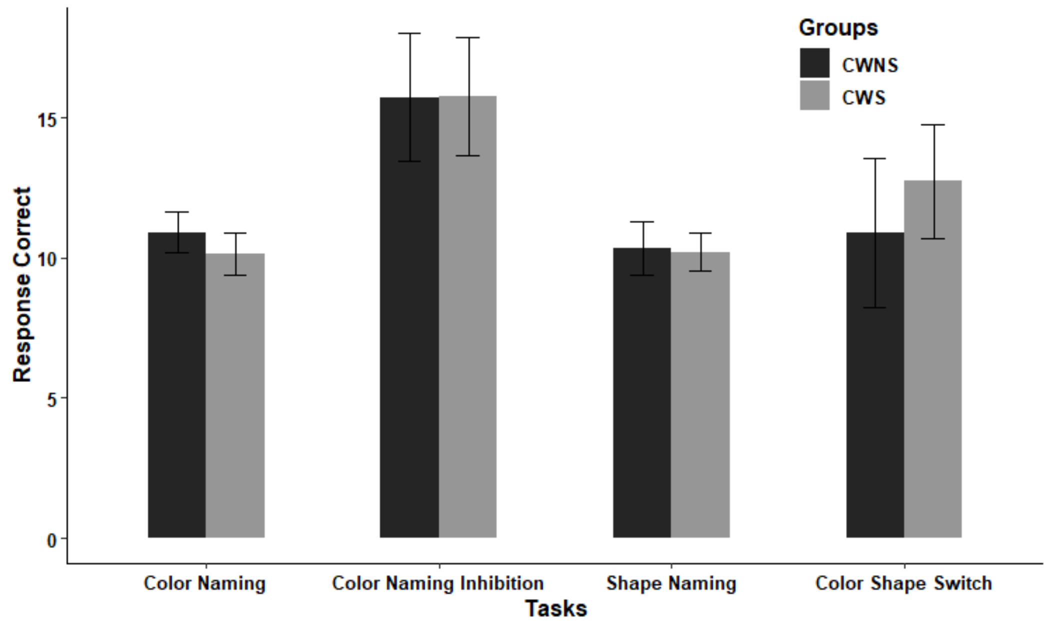

Figure 2. Responses Correct on Shape School Tasks

Note. Error bars indicate standard error from the mean

2.2.2 Stimuli

The current study applied a selective auditory attention paradigm used in past research (Hampton Wray et al., 2017; Isbell et al., 2016; Karns et al., 2015; Neville et al., 2013; Coch et al., 2005). Four narrative stories were played from *Blue Kangaroo* series (Clark, 1999, 2001a, 2001b, 2002), four from the *Harry the Dog* series (Zion & Graham, 1956, 1958, 1960, 1965), four from *Max and Ruby* series (Wells, 1991, 1997, 2000, 2002) and four from the *Classic Munch* series (Munsch & Martchenko, 1988, 1989, 1992; Munsch & Petricic, 2004). The stories were between

150 seconds – 210 seconds in length and were digitally recorded (16 bit, 22 kHz) by means of an Electro Voice 1750 microphone connected to a Macintosh computer running a sound-editing program (SOUNDEDIT 16, Version 2). Within each audio file created, there were two stories. One story was played in the right audio channel and a second story (which consisted of a separate story read by a narrator of the opposite sex) was played in the left audio channel. Children were seated midway between the right and left speakers and 150 cm away from a screen monitor. Children listened to two stories presented simultaneously that varied in location (left/right), voice (male/female), and content, presented at an average of 60 dB SPL. Children were tasked with attending to one of the two stories. In front of them, the screen monitor displayed illustrations corresponding to the story that they must attend to. Images subtended a visual angle of 5° or less and changed every 5–15 seconds to indicate the start of a new audio file. By the end of the experiment, children attended to two stories presented on the right side and two stories presented on the left. All stories were counterbalanced between participants. Figure 3 provides an illustration of the paradigm.

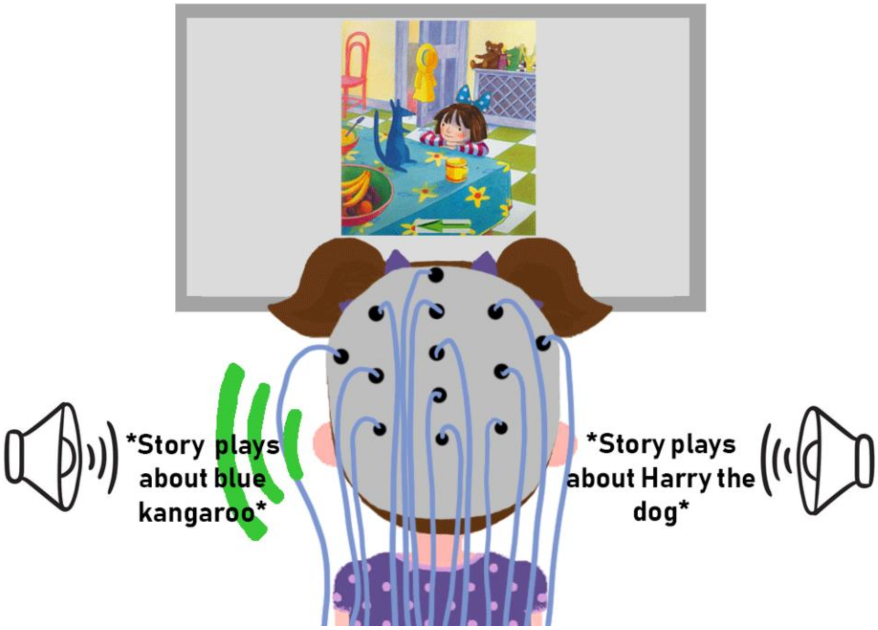

Figure 3. Illustration of the Experimental Paradigm

In addition, a researcher remained in the booth next to the child during the experiment. Upon completion of each audio file, the researcher asked the child three basic comprehension questions about the story they were meant to attend to (for a total of twelve questions throughout the experiment). Questions had two alternatives. If the child responded with "I don't know," the response was marked as incorrect.

Furthermore, linguistic and nonlinguistic probes were embedded in the audio files for a separate study that analyzed ERPs. The linguistic probe /ba/ was recorded by a female speaker, who was different from the female narrators. This probe was digitized and edited to 100 ms. The nonlinguistic probe was created by scrambling 4-6 ms segments of the linguistic /ba/ probe, which resulted in a 100 ms broad spectrum 'buzz' sound. All probes were presented at 70 dB SPL. Probes were embedded over one of the two auditory channels within each session and an equal number of probes (approximately 400 probes per attend/unattend condition) were randomly presented every 200, 500, or 1000 ms. Throughout all sessions, the identical probe stimuli were used.

2.2.3 Procedure

Upon arrival, children were given time to adjust to the laboratory. Following this, parents/caregivers signed a consent form prior to children providing verbal consent. The experimental task and procedures were thoroughly explained to the parent and child prior to beginning the study and questions were encouraged. Children completed behavioral testing with a speech-language pathologist on a separate day than the EEG data collection. After providing consent, behavioral testing with the child was administered by a certified speech-language pathologist. Parents were able to monitor all testing through cameras in a room adjacent to the testing room.

A 32-eclectrode cap was placed over the child's scalp. The child was then seated in a comfortable chair inside a sound-isolated booth. Children were advised to limit their movement during the experiment, which was reinforced by the research assistant if necessary. Prior to recording, children completed a practice session to familiarize themselves with the task. The child was advised that they would either hear a male or female speaker read the story. A small green arrow at the bottom of the screen would point to the speaker that they should listen to and the story they were meant to listen to would also relate to the pictures on the screen. In addition, they were instructed that unrelated sounds ('bas' and 'buzzes') would play, but that these sounds should be ignored.

By the end of the experiment, children attended to four narratives selected from the four story sets. Two of the stories were attended from the left side and the other two stories were attended on the right side (order either RLLR or LRRL). Two stories were always presented by a male and two stories presented by a female. A video camera and intercom monitored the child inside the booth so that other researchers and the caregiver(s) could observe from outside the booth. A trained researcher remained in the booth next to the child to ensure that they remained seated between the two speakers and that they were completing the task.

2.2.4 Electrophysiological Acquisition

An elastic electrode cap (Biosemi Active 2, Amsterdam, Netherlands) with 32 electrodes was used to record electrical activity from the scalp. The scalp was measured, and the appropriate cap size was placed snugly over the head. Thirty-two electrodes were positioned in homologous locations across the left and right hemispheres according to the criteria of the International 10-20 system (Jasper, 1958). The electrode channels included lateral sites F7/F8, FT7/FT8, T7/T8, P7/P8, medial sites FP1/FP2, F3/F4, C3/C4, FC5/FC6, C5/C6, CP1/CP2, CP5/CP6, P3/P4, PO3/PO4, O1/O2, and midline sites FZ, CZ, PZ, and OZ.

Recordings were referenced offline to the average of data recorded from the left and right mastoids. Horizontal eye movement was monitored through electrodes placed on the left and right outer canthi. Electrodes were also placed on the superior and inferior orbital ridge to monitor vertical eye movement. Eye channels were used to determine EEG artifact and were not included in analyses. Left and right horizontal eye channels were re-referenced to one another offline. All electrical impedances were adjusted to 20 kΩ or less. The EEG signals were recorded with a digitized sampling rate of 512 Hz.

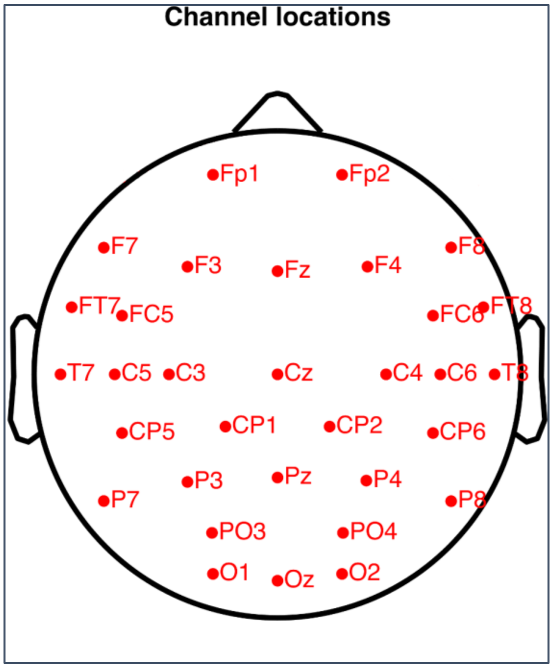

Figure 4. Electrode Locations for Event-related Brain Potential Recording

2.2.5 EEG Data Preprocessing

EEG data were preprocessed using EEGLAB 14.1.2 (Delorme and Makeig, 2004) in MATLAB® (The MathWorks Inc.). The raw EEG data were down-sampled to 128 Hz to improve computer competence. Minimum-phase causal windowed sinc FIR filters were then applied to down-sampled data using a band pass filter between 1 and 15 Hz. Filtered data were then re-referenced to the average of the two mastoid channels. Re-referenced channels were rejected if they had electrical activity that varied more than 3 standard deviations from the mean of the surrounding channels. Spherical spline interpolation was implemented for rejected data, which was established by the activity in surrounding channels. Artifacts in the EEG data were reduced with artifact subspace reconstruction (ASR) (Mullen et al., 2015). Clean sections (~60 seconds) were visually identified within the data and were entered as the calibration data for ASR. The ASR cleaned data were then separated into epochs, resulting in 4 epochs for each condition. Independent component analysis (ICA) was implemented using the infomax algorithm in EEGLAB on the epoched data. Independent components were then manually rejected by visually identifying components that included horizontal eye movements, eye blinks, and voltage drifts. Clean EEG data were built based on the remaining components.

2.2.6 Cortical Tracking to Speech Analysis

Cortical tracking of the speech envelope was estimated using the Multivariate Temporal Response Function (mTRF) Toolbox in MATLAB®. The multiband speech envelope was obtained using Hilbert decomposition of the output of 16 frequency-bands logarithmically spaced gamma tone filters between 250 Hz and 8000 Hz (Slaney, 1998). The amplitudes of these envelopes were raised to a power of 0.6 to replicate inner ear compression (Decruy et al., 2019; Vanthornhout et al., 2018). The envelopes were then down-sampled to 128 Hz, to align with the EEG sampling rate. Multivariate linear regression was applied to obtain the linear function between the speech envelope and the EEG data (Crosse et al., 2016) in every channel and at different time lags:

$$\beta = \quad [EEG(t, chan) = \sum_{\tau=-100}^{450} TRF(t, chan) \times env(t-\tau) + \varepsilon(t, chan)]$$

The EEG epochs were trimmed in alignment with the duration of the stimulus and the time lags used for TRF estimation were from -100 ms – 450 ms. In order to enforce a smoothness constraint on the TRFs and reduce overfitting to high-frequency noise, a regularization parameter was applied during model estimation. This parameter was optimized from $2^{0,1,2\ldots,20}$ using cross validation. TRFs were estimated using a 15-fold cross validation. Three trials were used for the TRF estimation in every iteration, then these trials were averaged and used to predict the neural response of the excluded (fourth) trial. Pearson's Correlation coefficient (r-values) were used to estimate the model fit of the TRF. After estimating the ridge parameters, the TRFs of all four trials were averaged to obtain the final averaged TRF. A schematic is displayed in Figure 5.

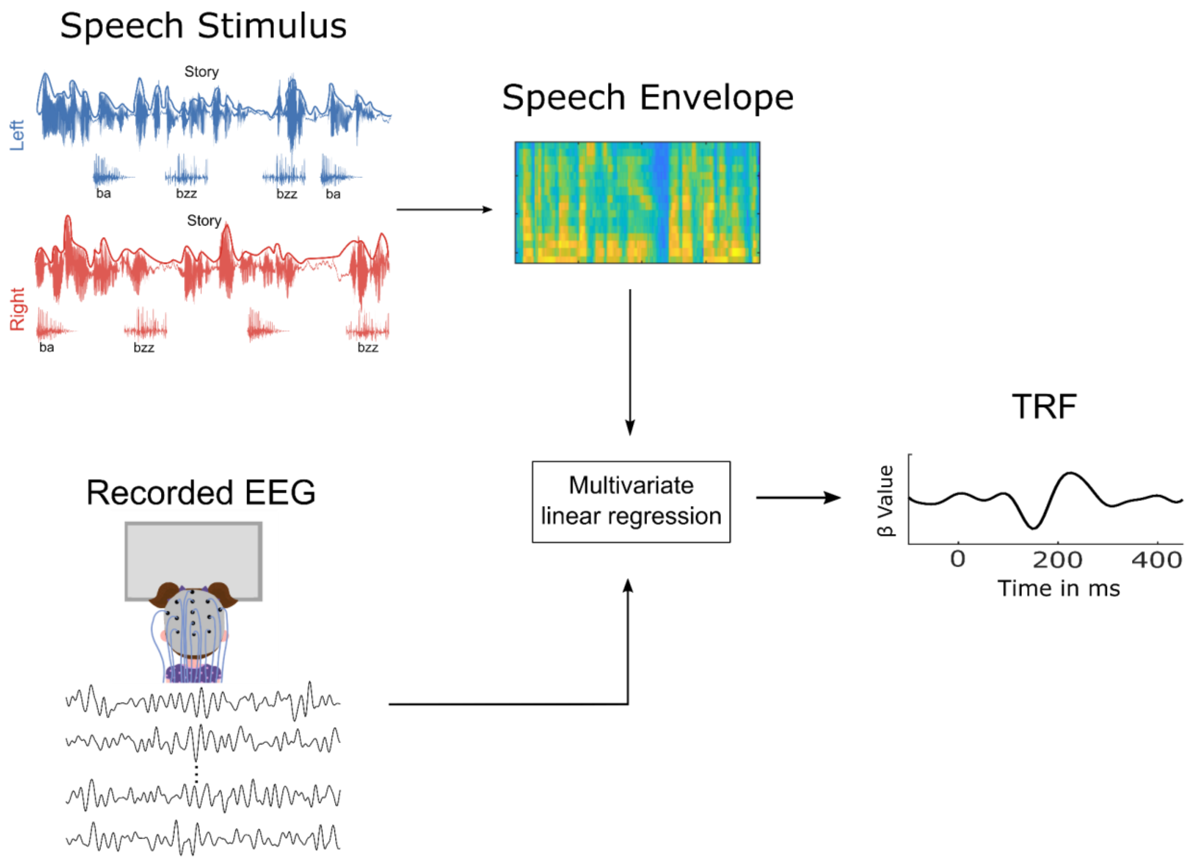

Figure 5. Cortical Tracking Schematic

Note. Schematic representation of the multivariate linear regression procedure used to obtain cortical tracking metrics. The low frequency temporal speech envelope was extracted across sixteen gammatone filters. Multivariate linear regression was used to estimate the (delayed) covariance of the low frequency temporal speech envelope and EEG data, which resulted in a TRF.

3.0 Results

Results from the TRFs, along with performance on the comprehension questions administered about the attended stories, were assessed. Figures 6, 7, 8, and 9 (below) depict TRFs across all electrode locations. Periods of significant differences ($p < .05$) are indicated in grey shaded areas, which were established by a pointwise t-test with cluster-based permutation analysis to correct for multiple comparisons (Maris & Oostenveld, 2007). In addition, each of these figures includes an isolated waveform in the bottom right which depicts the average across significant electrode locations. In these waveforms, all insignificant differences have been masked. Figure 10 is a display of all four of these isolated waveforms together.

Higher beta values (irrespective of polarity) suggest more efficient cortical tracking. The results indicate that both groups exhibit more efficient cortical tracking to the attended speech and less efficient cortical tracking to the unattended speech. When comparing differences between groups, CWS seem to have less efficient cortical tracking than CWNS for the unattended condition and greater cortical tracking than CWNS for the attended condition.

An independent samples t-test was completed on the individual scores from the comprehension questions, indicating no significant differences between groups; $t(25) = -0.70$, $p = .49$.

3.1 TRF Results

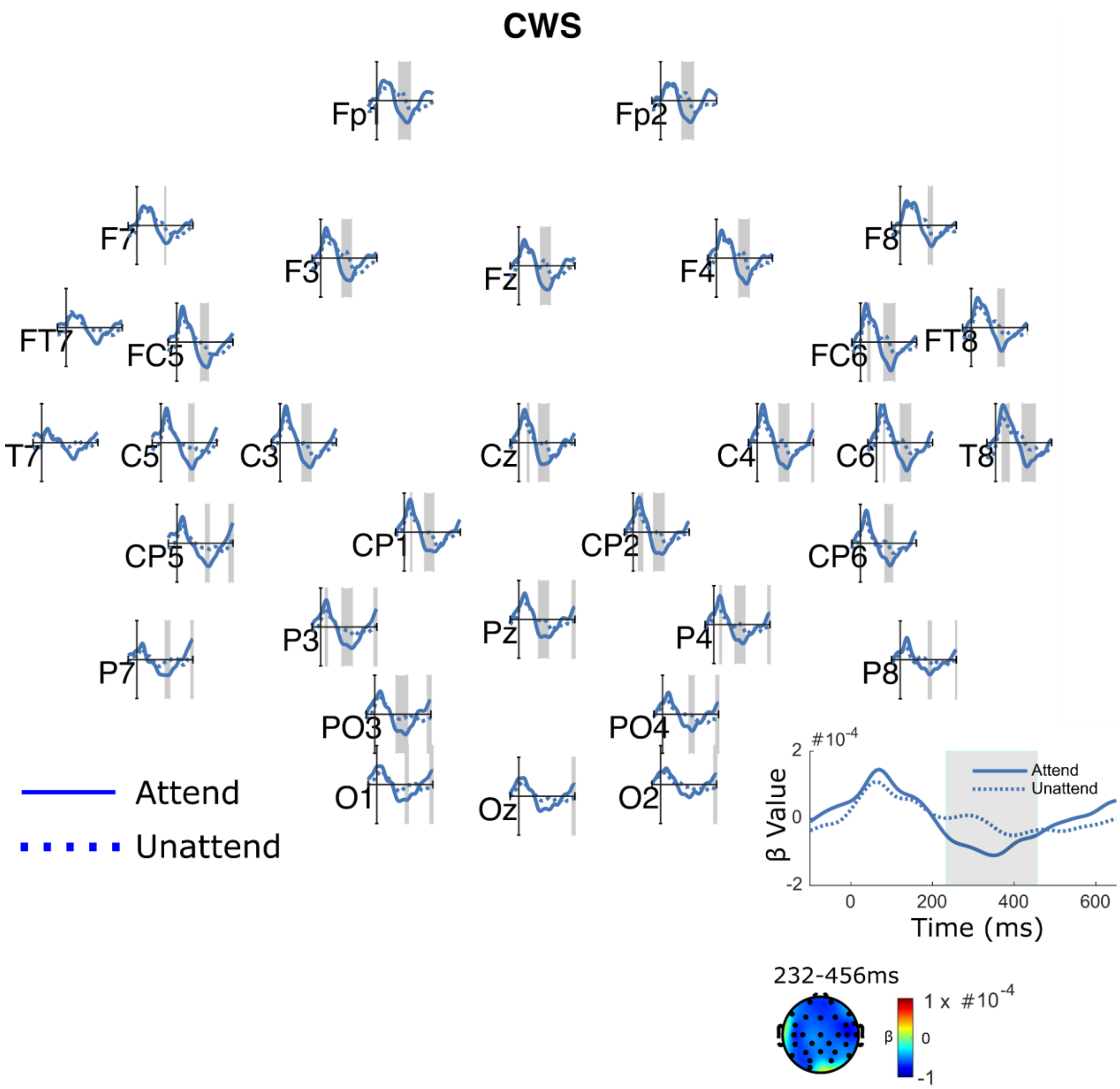

Figure 6. Attention Effects in CWS

Note. An average of the waveforms across significant electrode locations is in the bottom right.

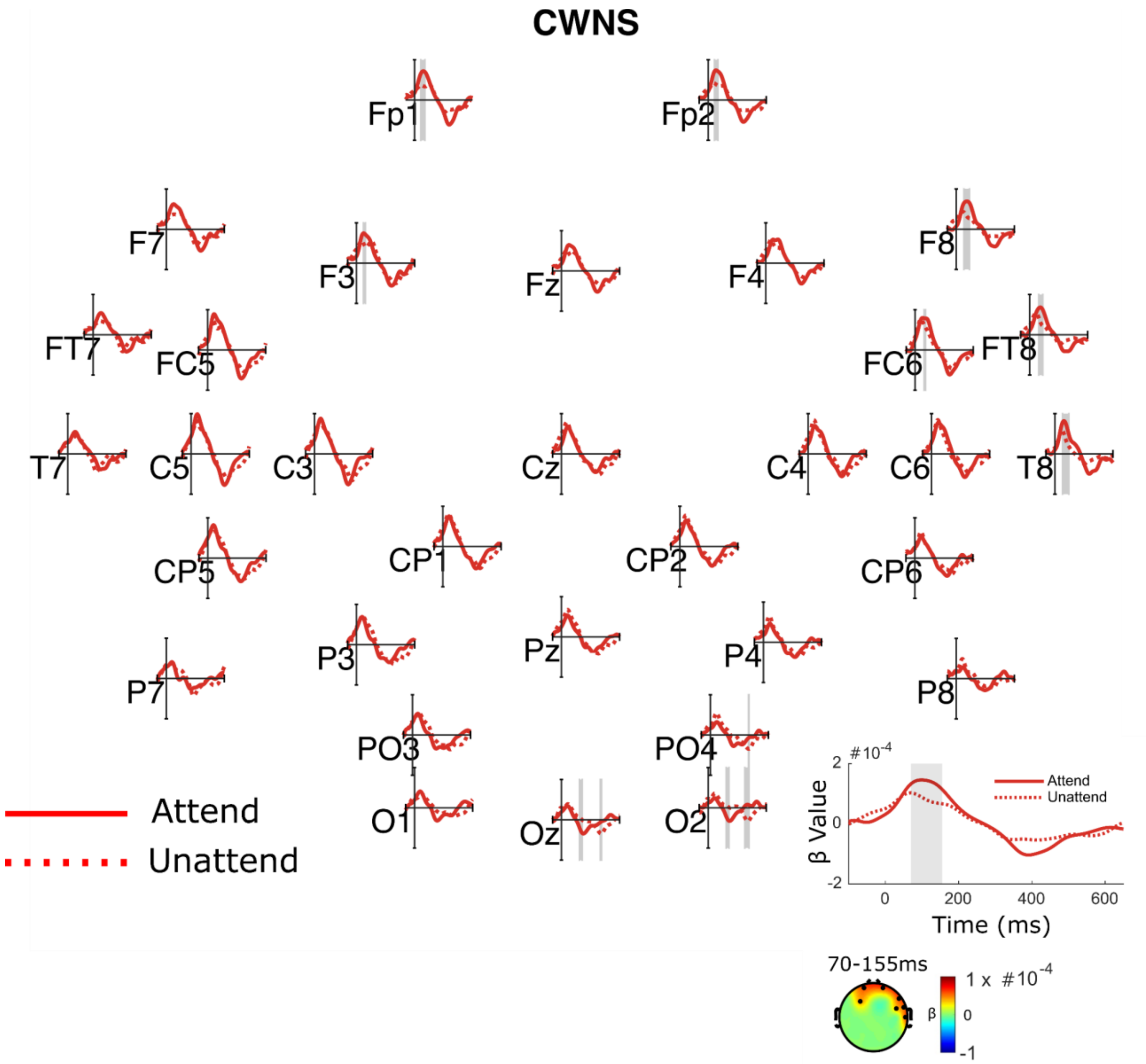

Figure 7. Attention Effects in CWNS

Note. An average of the waveforms across significant electrode locations is in the bottom right.

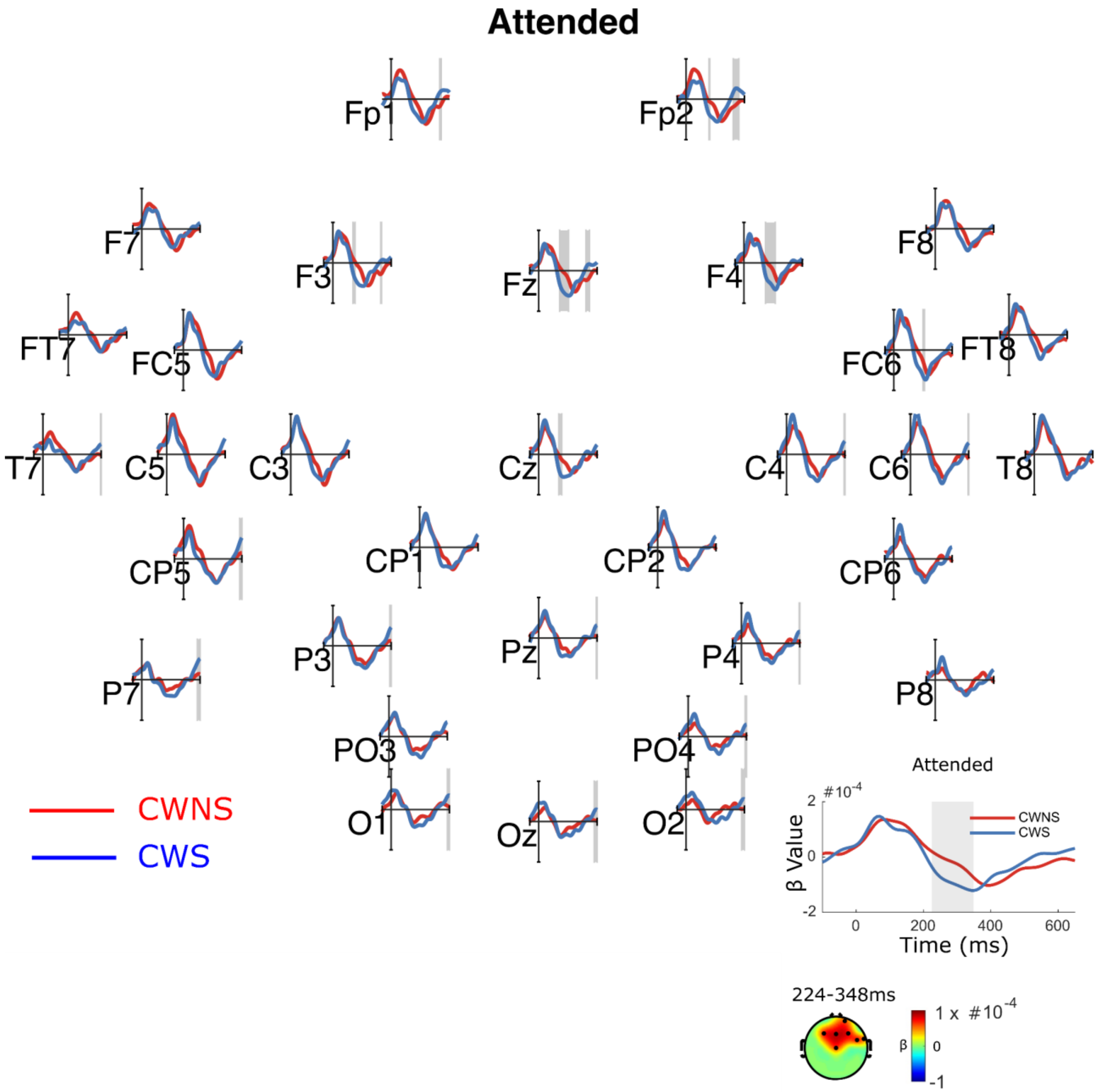

Figure 8. Group Effects in the Attended Conditions

Note. An average of the waveforms across significant electrode locations is in the bottom right.

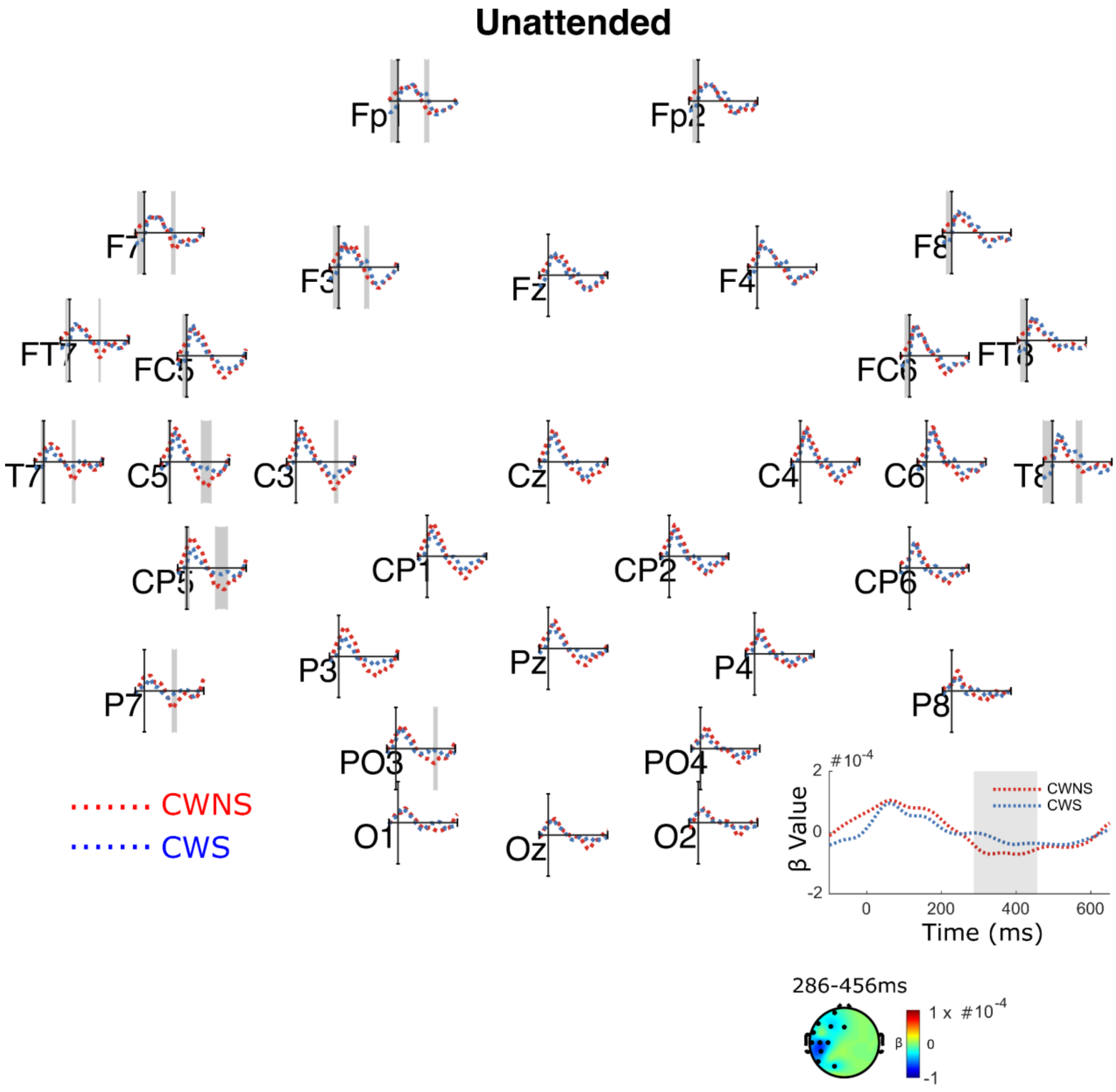

Figure 9. Group Effects in the Unattended Conditions

Note. An average of the waveforms across significant electrode locations is in the bottom right.

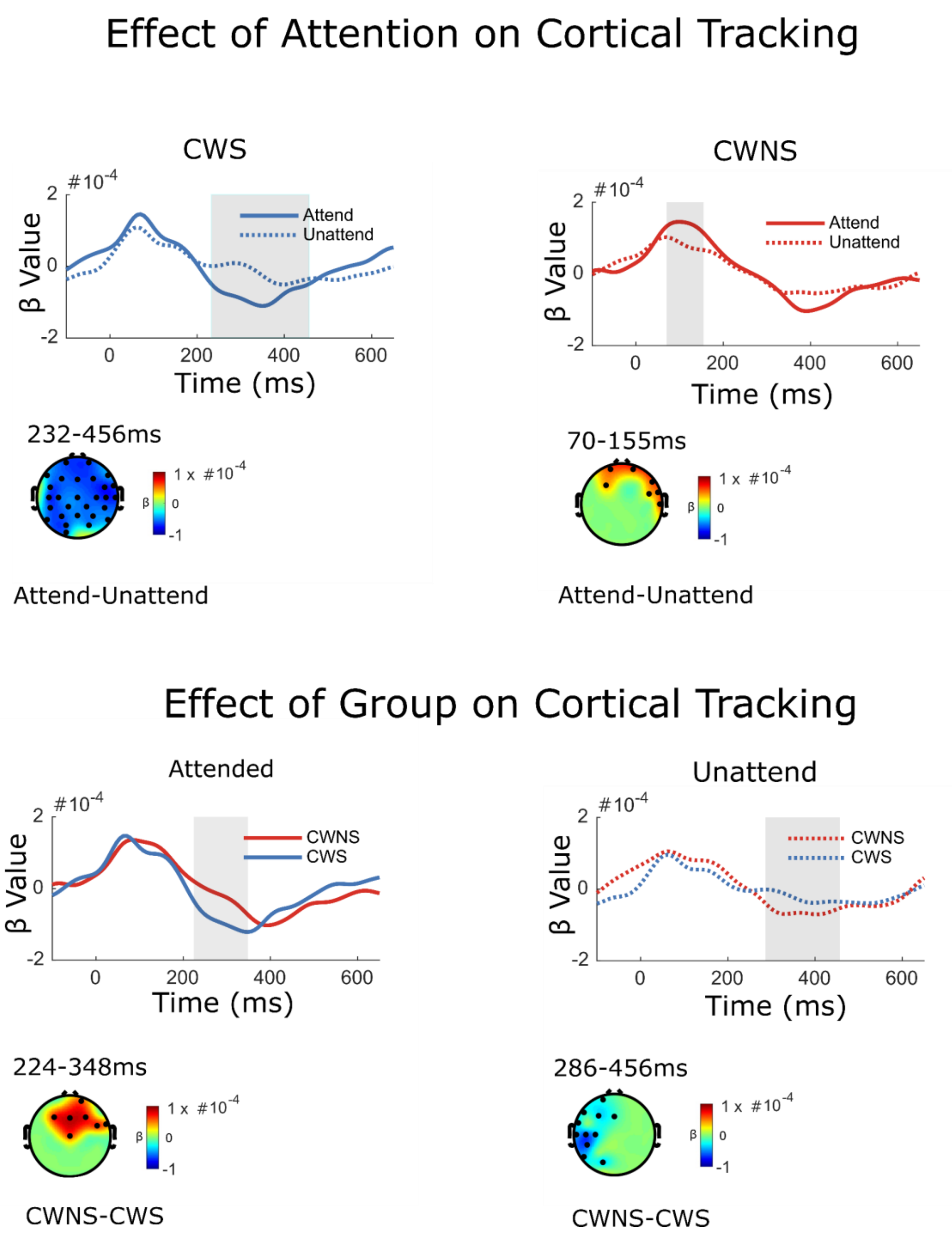

Figure 10. Waveforms Averaged Across Significant Electrode Locations

Note. Electrodes displayed indicate areas of significant differences. All insignificant differences have been masked.

3.2 Behavioral Results

Individual scores from the auditory attention questions are displayed in Table 4. The best score possible was 12 (12 questions were asked). Mean scores were compared between CWS and CWNS using an independent samples t-test with a 95% confidence interval. There were no significant differences between the auditory attention (AA) scores for CWS (M = 9.29, SD = 1.77) and the scores for CWNS (M = 8.77, SD = 2.05); t(25) = -0.70, p = .49.

Table 4. Individual Auditory Attention Scores

ID	CWS AA score	ID	CWNS AA Score
1	9	15	10
2	8	16	10
3	9	17	9
4	8	18	6
5	8	19	7
6	11	20	8
7	11	21	7
8	10	22	7
9	7	23	10
10	11	24	12
11	12	25	6
12	6	26	11
13	11	27	11
14	9		
	M = 9.29		M = 8.77

Note. M = mean.

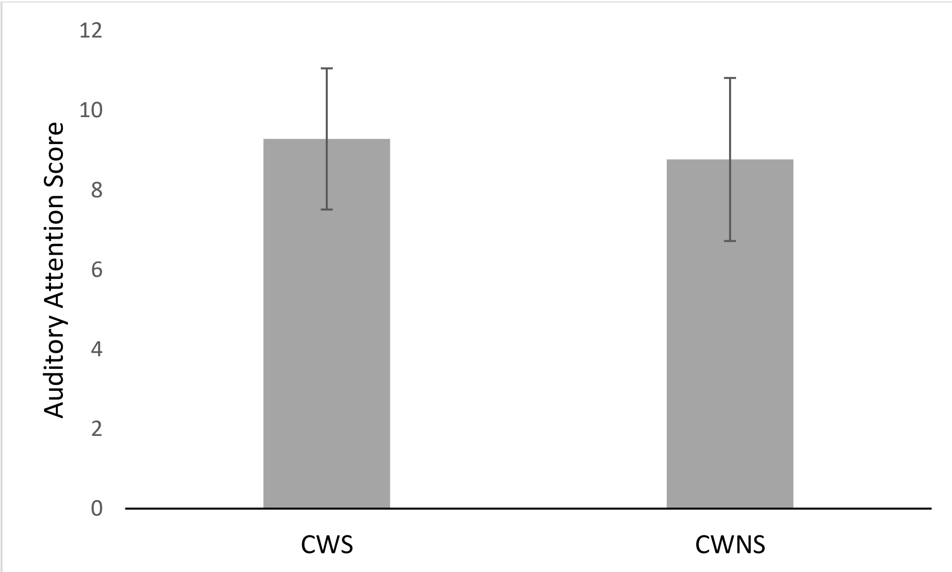

Figure 11. Mean Auditory Attention Scores Between Groups

Notes. Error bars indicate standard deviation of the mean. Best possible score = 12. CWS: (M = 9.29, SD = 1.77). CWNS: (M = 8.77, SD = 2.05).

4.0 Discussion

This study investigated the cortical underpinnings of developmental stuttering, a fluency disorder whose specific etiology is highly debated. The multifactorial pathways theory suggests that multiple factors interacting within the speech-motor system may result in this disorder (Smith & Weber, 2017). Of such factors, a growing body of evidence supports an interaction between stuttering and atypical phonological processing (Beitchman et al., 1986; Ratner, 1995; Louko, 1995; Wolk, 1998; Melnick & Conture, 2000; Wolk, et al., 1993; Spencer & Weber-Fox, 2014; Hakim & Ratner, 2004; Anderson, et al., 2006; Mahesh et al., 2018; Mohan & Weber, 2015). It has been proposed that phonological processing is a part of a hierarchical system, whose foundation lies in low frequency neural oscillations (Giraud & Poeppel, 2015). For this reason, measures of cortical tracking to the low-frequency temporal speech envelope were investigated. We hypothesized that CWS atypically encode the temporal speech envelope, which may contribute towards phonological processing difficulty. To test this hypothesis, cortical tracking was derived from continuous EEG recorded while participants performed a dichotic listening task to natural speech (Karns et al., 2015). This task requires auditory attention and attention deficits that have been seen in individuals who stutter (Felsenfeld et al., 2010; Karrass et al., 2006; (Bosshardt, 1999, 2002, 2006; Bosshardt et al., 2002; Vasic & Wijnen, 2005) may influence both speech envelope processing and phonological encoding. The comprehension of the target stimuli was determined by asking participants questions relating to the narrative played to the attended ear.

The results indicated that while both groups performed similarly on the behavioral task, the underlying neural mechanisms differed between CWS and CWNS. Consistent with our results, Mohan and Weber (2015) have also found that CWS may exhibit atypical neural activity, despite similar behavioral performance to CWNS.

There were attention effects; both groups exhibited more efficient cortical tracking for the attended speech and weaker cortical tracking to the unattended speech. This is as expected, since the auditory cortical system phase-locks to the temporal envelope of attended, but not ignored, speech (Kerlin et al., 2010; Power et al., 2012; Horton et al., 2013; O'Sullivan et al., 2014). However, these differences were seen at different latencies across groups. In CWNS, attention effects were seen at latencies of 70 ms - 155 ms and were restricted to frontal and fronto-temporal electrodes. However, in CWS attention effects occurred at latencies of 232 ms – 456 ms, which were distributed across all scalp electrodes.

When looking at group effects on cortical tracking, CWS showed higher cortical tracking for the attended speech and weaker cortical tracking for the unattended speech, when compared to CWNS. These group effects were seen in the latencies from 224 ms - 348 ms for the attended speech (in the frontal electrodes) and from 286ms-456ms for the unattended (throughout all the electrodes in the left hemisphere). These cortical tracking differences may indicate that CWS exude more attentional effort to overcompensate for an inadequate encoding mechanism. When they are not required to actively attend to the stimuli, there is an inherit deficit in encoding (significantly less efficient cortical tracking). However, an increase in attention is exerted to make up for this processing difference (significantly more efficient cortical tracking for the attended, even greater than cortical tracking for fluent peers). This atypical neural activity may only affect behavioral performance when cognitive demands are high, resulting in a breakdown of the system. This would explain why some studies have only found significant behavioral performance differences during higher cognitive demands (Byrd et al., 2015; Jones et al., 2012; Bosshardt, 2009; Weber-Fox et al., 2004; Sasisekaran & Byrd, 2013).

It is also worth noting that our results from the NWR task were not consistent with previous literature. We did not find performance differences between groups, while previous research has found significant differences at the 2- and/or 3-syllable level (Hakim & Ratner, 2004; Anderson et al., 2006). However, these studies included grouping variables which differed from our own. While Hakim & Ratner's study also had a wide age range (their study included children who were 4-8 years old, while our study included children who were 3-7 years old), all their CWS were classified as having moderate stuttering. In contrast, many of the CWS in our group only had very mild or mild stuttering. A higher level of stuttering severity may contribute towards behavioral differences, despite a wide age range.

In the study completed by Anderson, Wagovich, and Hall (2006), the experiment included a much larger sample size (24 CWS) and a narrower age range (children who were 3-5 years old). Given a wide range of factors that may interact with stuttering, a larger sample size would be more likely to account for differences within CWS. In addition, a younger group of children may have found the NWR test more difficult, thus increasing cognitive load and leading to performance differences. Furthermore, most of the children in their study were classified as having at least moderate stuttering. Once again, this suggests that stuttering severity may influence results.

In addition, Spencer and Weber-Fox (2014) noted that performance on the Dollaghan and Campbell nonword repetition test may be linked to stuttering persistence or recovery. However, their study included 40 CWS, which is a significantly larger sample size than our own. The age range was also restricted to children who were 3-5 years old, like the Anderson, Wagovich, and Hall (2006) study. Our group likely includes CWS who will go on to recover as well as persist, so we may not see group differences in NWR because some kids may be performing comparably to CWNS, as seen by Spencer and Weber Fox (2014). Overall, these findings suggest that sample size, age range, and SSI score may contribute towards performance differences.

We also did not find performance differences between groups for the Shape School task. Previous literature has incorporated dual-task paradigms to investigate inhibitory control, with significant findings that individuals who stutter perform less efficiently on these inhibitory tasks (Eggers & Jansson-Verkasalo, 2017; Eggers et al., 2013; Ofoe et al., 2018; Anderson & Wagovich, 2017; Piispala et al., 2018; Piispala et al., 2016). However, many of these studies incorporated auditory attention and examined response times. None of these studies involved the Shape School Task. In fact, to the best of our knowledge, our study is the first to examine Shape School performance in CWS. Therefore, while CWS may have performed similarly to CWNS, it is unknown whether they take longer to respond. This may reflect a delayed time in encoding (and processing) information.

It is now widely acknowledged that a plethora of factors interact with stuttering. Therefore, the heterogeneity within its population is likely a large contributor to the varied performances throughout much of stuttering research. This likely contributed to why our behavioral results were inconsistent with previous findings. In addition, our study included a wide range of participant ages and a small sample size. For some of the older children, the NWR task may not have been difficult enough to observe differences between groups. A larger sample would better account for overall differences within the dynamic stuttering population. In addition, this was the first study ever completed to examine cortical tracking of speech in individuals who stutter. No children in the current study had severe stuttering and 6 out of the 14 children (42%) only had very mild or mild stuttering. A sample size that includes children who have a higher SSI is likely to show poorer behavioral performance and larger differences in neural oscillating networks.

Our study provides new insights on the neural underpinnings of cortical tracking to continuous speech in individuals who stutter. The results support our hypothesis that CWS exhibit atypical cortical tracking to continuous speech. Atypical processing at the neural level may therefore act as a contributor towards phonological processing differences in CWS. This study provides a foundation for further investigation on the neural oscillating networks in individuals who stutter, as this is the first study to examine cortical tracking of continuous speech in this population.

The temporal properties of speech are only one of many features that can be analyzed using cortical tracking. Recent data show that stimulus-induced modulations to the delta (1–3 Hz), theta (4–8Hz), and low gamma frequency bands (25–35 Hz) reflect processing related to different speech units (Giraud & Poeppel, 2015; Ghitza, 2011; Poeppel, 2003). Cortical tracking of the temporal speech envelope (1-15Hz) has been cited as the most studied speech feature in examining continuous speech (Ding & Simon, 2014), and was analyzed in our study. However, it is also possible to study additional phonemic, phonetic, and semantic features of the speech stimulus (Di Liberto et al., 2015; Di Liberto & Lalor, 2017; Brodbeck, 2018). These different areas reflect various hierarchical levels of speech processing.

While temporal speech encoding may provide insight into syllabic parsing and prosidic information, examining cortical tracking to phonemic units would provide a more detailed analysis of higher level phonological processing. This is one area we hope to investigate within our next project. Since CWS have exhibited atypical cortical tracking at the broader level of temporal speech encoding, we hypothesize that CWS may also exhibit cortical tracking differences to the phonemic properties of speech. Atypical neural oscillations at this higher level may also account for the performance and processing differences that individuals who stutter have shown during various phonological tasks (Beitchman et al., 1986; Ratner, 1995; Louko, 1995; Wolk, 1998; Melnick & Conture, 2000; Wolk, et al., 1993; Spencer & Weber-Fox, 2014; Hakim & Ratner, 2004; Anderson, et al., 2006; Mahesh et al., 2018; Mohan & Weber, 2015).

There are also different machine learning-based approaches to analyze cortical tracking abilities. In the present study, we incorporated a forward modeling approach. Forward modeling uses acoustic speech features to predict EEG and offers insight on how the auditory system maps to different frequency bands. However, modeling can also be mapped in the reverse direction (backwards modeling). This method offers a complementary way to investigate speech encoding at the neural level. Backward modeling has many advantages to its counterpart. For instance, because reconstruction projects back into the stimulus domain, it does not require a pre-selection of neural response channels. This provides low weighting to irrelevant channels and allows for all channels to be included (certain channels are typically be excluded in feature selection approaches, like in forward modeling) (Pasley et al., 2012). In addition, backward modeling offers increased sensitivity to signal differences between response channels that have a high correlation with each other. This is possible because the data from all response channels are mapped simultaneously (Mesgarani et al., 2009). In contrast, each analysis in forward modeling is univariate and thus ignorant to the other data in the EEG channels. Despite its advantages, backwards modeling was not used in the present study due to time constraints associated with its implementation. We intend to implement this model within our next research project.

The present study has many implications for a future shift in research. Humans have the unique ability to generate and comprehend complex language, yet the most common methods for examining speech and language processes incorporate brief, sound-isolated stimuli. This is because estimating event-related potentials requires time-locking to discrete sensory events (ERPs; Handy, 2005; Luck 2014). The impulse of the response function is approximated by convolving the system with discrete probes and averaging over hundreds of response trials. While recent studies have attempted a more naturalistic approach by incorporating multiple repetitions of the same speech segment (Zion-Golumbic et al., 2013), instead of brief phonemes or sounds, this is far from real-life scenarios. It is an improved approach, but still does not align with how the human brain commonly processes speech and language. For this reason, there is a strong need for studies to incorporate natural, continuous speech stimuli. Consistent with this notion, Bonte, Parviainen, Hytönen, and Salmelin (2006) found that neural responses to syllables embedded in continuous speech are different from identical syllables that are presented in isolation. Natural speech incorporates linguistic information, co-articulation, and syntactic structure. Therefore, this underexplored method may provide better insight on speech and language in ethological settings. We encourage a shift within EEG methodology to incorporate machine learning-based approaches. This approach adapts an efficient and realistic examination of speech and language processing in both typical and clinical populations and could profoundly add to our knowledge on human processing abilities.

Our methods offer a novel way to investigate stuttering using objective measures of cortical tracking derived from natural speech and EEG. Using this study as a preliminary investigation, next we intend to examine cortical tracking to phonemic properties, which will provide greater insight on neural oscillations relating to phonological processing in CWS. We also intend to incorporate a much larger sample size to account for overall differences within the stuttering population. We expect that our next study will provide evidence for atypical cortical tracking of phonemic properties in CWS, as an additional contributor towards phonological processing differences.

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