

Title: Speech Planning and Execution in Children Who Stutter: Preliminary findings from a fNIRS investigation

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Abstract

Few studies have investigated the neural mechanisms underlying speech production in children who stutter (CWS), despite the critical importance of understanding these mechanisms closer to the time of stuttering onset. The relative contributions of speech planning and execution in CWS therefore are also unknown. Using functional near-infrared spectroscopy, the current study investigated neural mechanisms of planning and execution in a small sample of 9-12 year-old CWS and controls (N=12) by implementing two tasks that manipulated speech planning and execution loads. Planning was associated with atypical activation in bilateral inferior frontal gyrus and right supramarginal gyrus. Execution was associated with atypical activation in bilateral precentral gyrus and inferior frontal gyrus, as well as right supramarginal gyrus and superior temporal gyrus. The CWS exhibited some activation patterns that were similar to the adults who stutter (AWS) as reported in our previous study: atypical planning in frontal areas including left inferior frontal gyrus and atypical execution in fronto-temporo-parietal regions including left precentral gyrus, and right inferior frontal, superior temporal, and supramarginal gyri. However, differences also emerged. Whereas CWS and AWS both appear to exhibit atypical activation in right inferior and supramarginal gyri during execution, only CWS appear to exhibit this same pattern during planning. In addition, the CWS appear to exhibit atypical activation in left inferior frontal and right precentral gyri related to execution, whereas AWS do not. These preliminary results are discussed in the context of possible impairments in sensorimotor integration and inhibitory control for CWS.

Keywords: stuttering, fluency, fNIRS, planning, execution

Abbreviations

CON: controls

CWS: children who stutter

DSN: different syllable non-words

fNIRS: functional near-infrared spectroscopy

HbO: oxy-hemoglobin

HbR: deoxy-hemoglobin

IFG: inferior frontal gyrus,

IPL: inferior parietal lobule

MFG: marginal frontal gyrus

MNI: Montreal Neurological Institute

OcL: paracentral lobule

preCG/postCG: precentral and postcentral gyrus,

preSMA/SMA: pre-/supplementary motor area

RO: Rolandic operculum

SMG: supramarginal gyrus

SPL: superior parietal lobule

SSN: same syllable non-words

STG: superior temporal gyrus

1. Introduction

Stuttering is a neurodevelopmental communication disorder that impacts speech production in five to eight percent of young children and one percent of older children and adults (Yairi & Ambrose, 2012, 2005). The mechanisms underlying stuttering persistence are poorly understood. We previously found that for adults who stutter (AWS), speech planning was associated with left hemisphere speech network differences and speech execution was associated with right hemisphere speech network differences (Jackson, Wijekumar, et al., 2019). The current study aimed to collect preliminary data towards elucidating the neural correlates of speech planning and execution in children with persistent developmental stuttering closer to the age of onset and to relate these findings to the broader literature. To achieve our aim, we extended the paradigm from Jackson et al. (2019) downwards in age to a small sample of CWS.

The network of cortical regions that underlies overt speech production has been well documented, and includes the inferior frontal gyri (IFG), precentral and postcentral gyri (preCG/postCG), pre-/supplementary motor area (preSMA/SMA), supramarginal gyri (SMG), and superior temporal gyri (STG) (Bohland & Guenther, 2006; Price, 2012; Riecker et al., 2008; Shuster & Lemieux, 2005; Sörös et al., 2006). Most studies of neural function in stuttering speakers have focused on adults. AWS exhibit atypical activation relative to unimpaired controls (CON) during overt syllable and word production throughout the speech network, including reduced activation across many regions of the left hemisphere speech network (Chang et al., 2011; De Nil et al., 2000; Fox et al., 2000; Jiang et al., 2012; Neumann et al., 2004) and increased activation in the right hemisphere speech network dependent on the tasks and contrasts employed (De Nil et al., 2000, 2004; Kell et al., 2009; Loucks et al., 2011; Neumann et al., 2004; Preibisch et al., 2003; Sakai et al., 2009). Several studies point to atypical cerebral dominance in

adults who stutter (Foundas et al., 2003, 2004; Travis, 1931), suggesting that atypical higher activation in the right hemisphere is associated with a compensatory neural mechanism for left hemisphere deficiency (De Nil et al., 2004; Foundas et al., 2004; Fox et al., 2000; Kell et al., 2009; Preibisch et al., 2003). Only one study has examined the functional neural correlates in CWS during a speech production task. Using functional near-infrared spectroscopy (fNIRS), Walsh et al. (2017) found that CWS aged 7-11 years exhibited reduced activation in the L-IFG relative to CON during a picture description task, a result consistent with the atypical functional organization for speech in AWS and indicative that atypical left hemisphere speech network activity may be a core deficit in developmental stuttering. Also consistent with the left hemisphere deficit theory is evidence that the anatomical developmental of L-IFG is atypical across boys and men who stutter relative to controls (Beal et al., 2015), as are the white matter tracts connecting this region to the broader neural network for speech-motor control (Chang et al., 2015; Misaghi et al., 2018).

Broadly, speech production involves planning and execution processes. Planning refers to the spatiotemporal specification of articulatory movements (Mooshammer et al., 2012), whereas execution refers to the implementation of these movements. From this perspective, the sequencing of articulatory configurations for simple utterances (e.g., words, short phrases) is established during planning, whereas execution load is determined via length of utterance. (e.g., three- versus one-syllable words). Studying the neural substrates of planning and execution requires isolating these processes because they may overlap in time. This is especially the case with neuroimaging techniques that measure changes in blood flow concentrations to estimate neural activation changes because the hemodynamic response is slow relative to the speech tasks under study (e.g., peak activation occurs between four and six seconds post stimulus). For this

reason, Jackson et al. (2019) implemented two tasks, each with two conditions, that separately manipulated planning and execution loads in AWS. For planning, three-syllable non-words consisted of either the same syllables (low load) or different syllables (high load). In this task, planning load changed (same versus different syllables), while execution load was held constant (three syllables). It was assumed that the demands associated with sequencing speech movements manifest themselves during planning stages. For execution, stimulus words were produced either overtly or covertly while planning load was held constant. Our previous findings related to planning and execution in AWS revealed atypical left-hemisphere activation in response to increased planning load, and atypical right fronto-temporo-parietal and bilateral motor activation in response to increased execution load (Jackson, Wijekumar, et al., 2019). The relative contributions of planning and execution in CWS, however, have not been studied. Establishing an understanding of the neural processes for planning and execution in the context of one another, especially earlier on in the developmental course of persistent stuttering, may provide insight into the neurobiology of disorder onset and progression. In the current study, we used fNIRS and the planning and execution paradigm from Jackson et al. (2019) to collect preliminary data on the neural substrates of planning and execution processing in a small sample of 9-12 year-old CWS and CON.

2. Methodology

2.1. *Participants*

This study was approved by the University of Iowa Institutional Review Board on Human Subjects. The CWS were recruited through UISPEAKS, a week-long camp for CWS; the CON were recruited through the University of Iowa community. Participants were right-handed, monolingual speakers of English and included six CWS (three female, $M = 11.71$, $SD = 1.36$),

and six CON (three female, $M = 11.21$, $SD = 1.07$). The CWS were included if they were rated greater than 1 on the 0-7 Iowa Scale of Severity of Stuttering (Johnson et al., 1963) by a speech-language pathologist, and either (1) described their speech as containing stuttering-like disfluencies as defined by Yairi and Ambrose (1992) in a communication history questionnaire, or (2) received a diagnosis of stuttering from an American Speech-Language-Hearing Association certified speech-language pathologist. Parent severity ratings on the 0-7 Iowa Scale included: one rating of 2, three ratings of 3, and one rating of 4. Clinician severity ratings on the 0-7 Iowa Scale included: three ratings of 3, two ratings of 4, and one rating of 6. Participants did not exhibit hearing or visual impairment based on screenings, and reported negative histories of other speech-language or neurologically-based disorders. Table 1 includes age, age of onset, parent education, parent and clinician severity ratings, and family history of stuttering, for both groups.

2.2. *Tasks and Stimuli*

2.2.1. *Speech Motor Planning Task*

The planning task was adapted from Lu et al. (2010), and is identical to that used in Jackson et al. (2019). It consisted of non-words that varied in their degree of planning load (high vs. low), but not execution load, which remained relatively constant (i.e., three syllables). High load non-words were comprised of three different syllables (different syllable non-words; DSN); low load non-words were comprised of the same three syllables (same syllable non-words; SSN). The phonotactic complexity of the DSN and SSN did not differ. There were 20 SSN and 20 DSN in total; 15 SSN and 15 DSN were produced twice, and 5 SSN and 5 DSN were produced once. In total, 35 SSN and 35 DSN were produced, in seven blocks of ten words each, and participants were given a brief rest between blocks. Appendix A includes the stimulus words for the planning

task. Fluency and accuracy judgments were determined by a speech-language pathologist (fourth author) who was also a doctoral student in Communicative Sciences and Disorders, who watched video of the sessions. Figure 1A depicts the timeline of the planning task.

2.2.2. *Speech Motor Execution Task*

The execution task was identical to that in Jackson et al. (2019). Participants produced real words aloud (overt) or silently without articulation (covert) during a picture-naming task. For the covert condition, participants were instructed to produce words silently, without moving their articulators (i.e., “Say it in your head”), which increased the probability that words were produced silently, and not inhibited. Stimuli included forty-two photographs from the Hatfield Image Test (Adlington et al., 2009). All pictures were nouns and developmentally appropriate (i.e., acquired by age five). Following a fixation period, the pictures were presented with a black border. After 1,500 ms, the border either turned green (overt naming) or red (covert naming). In total, there were 84 trials presented in seven blocks. In addition, eleven foils were included such that the overt-covert pairs consisted of different words, to reduce the predictability of the task. Appendix B includes the stimulus words for the execution task; figure 1B depicts the task timeline.

2.3. *fNIRS Recording and Processing*

2.3.1. *Instrumentation and cap design*

A TechEn CW6 system with 12 sources and 24 detectors and wavelengths at 830 nm and 690 nm were used (sampling rate 25Hz). The probe geometry was designed following the procedure described in Wijekumar et al. (2015). It consisted of 46 channels located between the sources and detectors covering parts of the frontal, motor, parietal, and temporal cortices. Regions of interest were selected based on the results of six published research reports that used

fMRI to examine speech production in AWS (Chang et al., 2009; Lu et al., 2009, 2010; Neumann et al., 2003; Preibisch et al., 2003; Watkins et al., 2008). Figure 2 shows the probe geometry. Four of the 46 channels served as short source-detector channels (highlighted in green). The Montreal Neurological Institute (MNI) atlas labels underlying each channel for the participants' probe geometries are shown in Table 2; MNI labels were obtained by determining the peak voxel of the sensitivity profiles of one participant. Scalp landmarks and positions of sources and detectors were digitized using a Polhemus 3D motion sensor.

2.3.2. *Pre-processing fNIRS data*

Data were processed using HOMER2 (www.nmr.mgh.harvard.edu/PMI/resources/homer2). Raw intensity values were converted to optical density units (function `hmrIntensity2OD`). Data were band-pass filtered (0.016 Hz and 0.5 Hz) and the modified Beer-Lambert law was used to convert optical density data to concentration units (functions `hmrBandpassFilt` and `hmrOD2Conc` respectively). Motion artifacts were identified and corrected using correlation-based signal improvement of the concentration changes (function `hmrMotionCorrectCbsi`) (Cui et al., 2010; Walsh et al., 2017). Data were then converted back to optical density units using function `hmrConc2OD`. The data was screened to check for any more motion artifacts using function `MotionArtifactbyChannel` function (`tMotion = 0.5`, `tMask = 1.0`, `StdevThresh = 50` and `AmpThresh = 5`). Any remaining artifacts were removed using function `enStimRejection` (`tRange = -1 to 11s`). Data was converted from optical density units to concentration (`hmrOD2Conc`). A block average was computed over a window of -1 to 11 s (`hmrBlockAvg`) to then identify outliers using `hmrFindHrfOutlier` (`tRange = -1 to 11s`; `stdThresh = 3`; `minNtrials = 0`). Separate general linear models (GLMs) were run on each task and chromophore (oxy-hemoglobin [HbO] and deoxy-hemoglobin [HbR]). A modified gamma

function was convolved with a square wave of duration 10 s to construct the regressors (hmrDeconvHRF_DriftSS; tRange = -1 to 11s; glmSolveMethod = 1; idxBasis = 2; paramsBasis: tau1 = 0.1, sigma1 = 3, T1 = 10, tau2 = 1.8, sigma2 = 3, T2 = 10). The onset of the regressor was speech initiation for the planning task, and the “go” signal—when the color of the picture frame changed from black to red or green—for the execution task. Short source-detector regression was performed within the GLM using channels with the highest correlation to remove any scalp artifacts (Gagnon et al., 2012). Beta estimates were obtained for each condition, channel, chromophore, and participant.

2.3.3. *Forward model for fNIRS image reconstruction*

The forward model for fNIRS image reconstruction was generated using the Colin adult template in HOMER2. The *3dSeg* function in AFNI (Analyses of Functional NeuroImaging) was used to segment a 12-year old T1 anatomical image into separate volumes for gray matter, white matter, cerebrospinal fluid, and scalp tissues. Then, AtlasViewerGUI in HOMER2 was used to convert the segmented volumes to an atlas. Digitized scalp landmarks, and source and detector positions from each participant were projected onto the atlas (see Figure 2B). Monte Carlo simulations were run with 100 million photons to create sensitivity profiles for each channel. The sensitivity profiles were summed together and thresholded such that voxels with an optical density of 0.0001 or greater were included (see Wijekumar et al., 2015 for justification). The resulting thresholded images were masked for each participant using *3dcalc*, and each mask was transformed to the space of the original atlas using *3dAllineate* in AFNI. A group mask was created by summing the participant-specific masks. The group mask was further thresholded such that only those voxels with data from all children were included, and transformed to Montreal Neurological Institute (MNI) space using *3dAllineate*.

2.3.4. *Image Reconstruction*

The methods used for image reconstruction in the current article have been described elsewhere (Eggebrecht et al., 2014; Hirsch et al., 2018; Perlman et al., 2016; Putt et al., 2017; Wijekumar, Huppert, et al., 2017; Wijekumar, Magnotta, et al., 2017). We describe them briefly here. After establishing the forward model and beta coefficients (above), the relationship between the hemodynamic response and delta optical density is as follows:

$$\begin{bmatrix} d \cdot \varepsilon_{HbO}^{\lambda 1} \cdot \beta_{HbO} + d \cdot \varepsilon_{HbR}^{\lambda 1} \cdot \beta_{HbR} \\ d \cdot \varepsilon_{HbO}^{\lambda 2} \cdot \beta_{HbO} + d \cdot \varepsilon_{HbR}^{\lambda 2} \cdot \beta_{HbR} \end{bmatrix} = \begin{bmatrix} \varepsilon_{HbO}^{\lambda 1} \cdot F^{\lambda 1} & \varepsilon_{HbR}^{\lambda 1} \cdot F^{\lambda 1} \\ \varepsilon_{HbO}^{\lambda 2} \cdot F^{\lambda 2} & \varepsilon_{HbR}^{\lambda 2} \cdot F^{\lambda 2} \end{bmatrix} \cdot \begin{bmatrix} \Delta HbO_{vox} \\ \Delta HbR_{vox} \end{bmatrix}$$

Here, F represents the channel-wise sensitivity volumes from Monte Carlo simulations. ΔHbO_{vox} and ΔHbR_{vox} represent voxel-wise relative changes in HbO and HbR concentrations. These changes are estimated using the image reconstruction approach. This equation can be re-written as:

$$Y = L \cdot X$$

where

$$Y = \begin{bmatrix} \beta_{dOD}^{\lambda 1} \\ \beta_{dOD}^{\lambda 2} \end{bmatrix}, L = \begin{bmatrix} \varepsilon_{HbO}^{\lambda 1} \cdot F^{\lambda 1} & \varepsilon_{HbR}^{\lambda 1} \cdot F^{\lambda 1} \\ \varepsilon_{HbO}^{\lambda 2} \cdot F^{\lambda 2} & \varepsilon_{HbR}^{\lambda 2} \cdot F^{\lambda 2} \end{bmatrix} \text{ and } X = \begin{bmatrix} \Delta HbO_{vox} \\ \Delta HbR_{vox} \end{bmatrix}$$

Voxel-wise maps of relative concentration changes for each condition, channel, participant and chromophore, were determined by solving for X . These maps were then transformed to MNI space and multiplied by the thresholded group masks.

2.4. *Group Analyses*

2.4.1. *Behavioral Analyses*

Accuracy and fluency judgments were made based on video recordings. Inaccurate and stuttered trials were not included in the fNIRS analyses. This is common practice in neuroimaging studies of stuttering so that neural differences observed between conditions can be

attributed to task manipulations (in the current study, planning or execution load), and not movement associated with stuttered versus non-stuttered speech. For the planning task, inaccurate trials included interjections, fillers, or revisions, as well as altered or omitted syllables or altered syllable sequences. In addition, trials were excluded if participants-initiated speech immediately after repeating the stimuli. For the execution task, inaccurate overt trials included those for which the item in the picture was not named accurately. Incorrect covert trials included those during which the participant produced speech. Stuttered trials (i.e., stuttering-like disfluencies) included part-word repetitions, prolongations, or blocks.

2.4.2. *fNIRS Analyses*

Four separate ANOVAs were run: planning HbO; planning HbR; execution HbO; execution HbR. Group served as the between-subjects factor (CWS vs. CON) and Condition served as the within-subjects factor (SSN vs. DSN for planning and overt vs. covert for execution). To constrain the family-wise error (FWE) rate, we used 3dClustSim in AFNI (voxel-wise threshold of $p < 0.05$, $\alpha < 0.05$, voxel size of 8 mm³). 3dClustSim employs Monte Carlo simulation of Gaussian noise to estimate the probability of false positive clusters. Based on the 3dClustSim simulations, we thresholded all main effect and interaction images using a cluster size of 38 voxels. Voxels with both significant interaction and group effects were interpreted at the interaction level. Voxels with both group and condition main effects were interpreted at the group level. Average beta values were extracted for each condition, participant, and chromophore. Paired t-tests were implemented for post-hoc comparisons for each cluster with a significant interactions for group and condition interaction. We focused on the Group main effects and the interactions between group and condition because we were most interested in the

CWS and CWS/CON comparisons. The data presented in this article are available from the corresponding author on reasonable request.

3. Results

3.1. Behavioral Results

Table 3 includes trial data for accuracy and fluency for both tasks. Mann-Whitney U tests were used to determine group differences for accuracy and fluency. For the planning task, accuracy was not significantly different between the CWS and CON ($W=17.5$, $p > .05$), but the between-group difference for fluency approached significance ($W=28.5$, $p = .07$). For execution, fluency was significantly different, with CWS exhibiting more disfluent trials than CON ($W=30$, $p < .05$); accuracy did not differ between groups ($W=19.5$, $p > .05$).

3.2. fNIRS Results

Significant clusters that survived family-wise correction are presented in Table 4. Interaction effects and subsequent posthoc tests are the focus of the current study. Group and condition main effects are not the focus of the current study, but are included in Table 4 for completeness. As is common reporting practice in the fNIRS literature, the results are presented such that increases in HbO or decreases in HbR are both referred to as *increases* in neural activation (e.g., Dravida et al., 2017; Strangman et al., 2002; Zhang et al., 2016, 2017). HbO and HbR are combined in all tables and figures, and only fluent and correct trials are included.

3.2.1. Planning

Significant interactions between group and condition were found for clusters in L-IFG, R-IFG, R-SMG, and L-preCG (Table 4). Post-hoc comparisons with Benjamini-Hochberg corrections revealed that the CON exhibited greater activation in L-IFG during the high vs. low planning load condition ($t=-2.99$, $df=5$, $p<.1$, approaches significance), but the CWS did not

exhibit this pattern (Figure 3, left). In addition, the CWS exhibited greater activation during the low vs. high planning load condition in R-IFG ($t=-2.72$, $df=5$, $p<.05$) and R-SMG ($t=3.80$, $df=5$, $p<.05$) (Figure 3, center and right, respectively), but the CON did not exhibit these patterns. No other post-hoc comparisons yielded significant results.

3.2.2. Execution

Significant interactions between group and condition were found for clusters in fronto-temporo-parietal regions including bilateral preCG and IFG, R-SMG, and R-STG (see Table 4). Post-hoc comparisons with Benjamini-Hochberg corrections revealed that the CWS exhibited lesser activation in L-preCG during the overt vs. covert condition ($t=4.08$, $df=5$, $p<.05$), and that the CON exhibited greater activation than the CWS in L-preCG during the overt condition ($t=4.33$, $df=10$, $p<.01$) (Figure 4). Both groups demonstrated an increase in activation in R-preCG (different clusters) during the overt vs. covert condition (CWS [$t=-2.44$, $df=5$, $p<.1$, approaches significance]; CON [$t=-4.15$, $df=5$, $p=.05$]), but the CWS exhibited greater activation than the CON during the overt condition ($t=-3.39$, $df=10$, $p<.05$) (Figure 4). Further, the CWS exhibited greater activation in L-IFG ($t=3.27$, $df=5$, $p<.05$) and R-IFG ($t=3.53$, $df=5$, $p<.05$), and lesser activation in R-STG ($t=-3.45$, $df=5$, $p<.05$) and R-SMG ($t=-3.77$, $df=5$, $p=.0585$, approaches significance), during the overt vs. covert condition (Figure 4). The CON did not exhibit these patterns.

4. Discussion

The purpose of this preliminary study was to determine potential neural substrates of speech planning and execution in CWS. Compared to CON, the CWS exhibited atypical neural activation underlying both planning and execution processing. For planning, the CWS exhibited atypical activation in bilateral IFG and R-SMG. For execution, activation patterns were atypical

in motor cortex and bilateral IFG, as well as other areas within the speech motor network (i.e., R-SMG, R-STG). The CWS exhibited some activation patterns for planning and execution that were similar to those reported for AWS in our previous study (Jackson, Wijekumar, et al., 2019). Planning was associated with atypical activity in L-IFG in both CWS and AWS, and execution was associated with atypical activity in L-preCG R-IFG, R-SMG, and R-STG. However, possible group differences also emerged. Planning was associated with atypical activation in R-IFG and R-SMG in CWS but not AWS. Execution was associated with atypical activation in bilateral IFG in CWS, but only R-IFG in AWS. The CWS also exhibited atypical activation in R-preCG while CON did not (CON exhibited a similar pattern in R-postCG), and the pattern exhibited by the CWS in L-preCG was opposite to that of AWS. These similarities and differences, as well as possible mechanisms involving sensorimotor integration and inhibitory processing during planning and execution are discussed in the following sections.

4.1. Neural Substrates of Planning in Children Who Stutter

The CWS exhibited atypical activation in L-IFG in response to the shift from low to high planning load—that is, the CWS did not exhibit the same increase in activation as the CON in the high planning load condition. The L-IFG, which includes Broca’s area, is a potential site for the neural representations of speech sounds, or the phoneme and syllable store, and has been implicated in tasks reliant on phoneme-to-articulatory coding (Glasser & Rilling, 2008; Indefrey & Levelt, 2004; Petrides et al., 2005). Further, anatomical development of L-IFG is atypical in CWS (Beal et al., 2013; Chang et al., 2008; Garnett et al., 2018), which has led researchers to implicate L-IFG as a core etiological deficit associated with stuttering (Chang et al., 2018). Presumably, the shift from low to high planning load requires the speaker to up-regulate activation in L-IFG. Our data indicate that the CON demonstrated this up-regulation in L-IFG,

but the CWS did not. Thus, CWS may not be engaging this critical node of the speech neural network in the same way as their peers. This result is in line with Walsh et al. (2017) who found – also using fNIRS – that CWS exhibit lesser neural activation in L-IFG during speech production. While the CWS in the current study did not exhibit a difference between the low and high planning load conditions, they did not exhibit the same up-regulation of activation in this area as their peers. This result is also in line with previous studies that showed atypical neural activity associated with speech planning in AWS (Chang et al., 2009; Jackson, Wijekumar, et al., 2019; Lu et al., 2010). Notably, Jackson et al. (2019) showed with the same task that AWS exhibit the same pattern as the CWS in the current study (i.e., no up-regulation in L-IFG when planning load increased), suggesting that this pattern is present from at least as young as the older school-age years or possibly even before. This claim warrants testing with a larger and younger sample of CWS and CON.

The planning contrast also revealed that the CWS exhibited greater activation in R-IFG during the low versus high planning load condition, but the CON did not exhibit this pattern. Increased activation in R-IFG has been characterized as a “neural signature” of stuttering (Brown et al., 2005; Budde et al., 2014). It has been proposed that increased activation in the right hemisphere speech network of AWS, particularly R-IFG, is compensatory for left hemisphere deficiency (De Nil et al., 2003; Foundas et al., 2004; Fox et al., 2000; Kell et al., 2009; Neef et al., 2018; Preibisch et al., 2003). The basis for these claims has been studies focusing on AWS primarily during speech production. The current results indicate that atypical activation in R-IFG may begin during the planning phase, which is in line with Lu et al. (2010) who found that during speech planning, AWS exhibit atypical activation in this area. It may be the case that the increase in planning load interferes with the ability of CWS to recruit the compensatory neural

resources in R-IFG necessary to produce speech as their fluent peers. Jackson et al. (2019) did not find this same pattern in AWS suggesting that by adulthood, speakers who stutter adapt to the demand of increases in planning load.

The CWS also exhibited greater activation in R-SMG during the low versus high planning load condition, but the CON did not exhibit this pattern. R-SMG is a key region associated with verbal working memory and speech motor control (Guenther, 2016), and is likely the site of the phonological store (Jonides et al., 1998; Paulesu et al., 1993). The R-SMG and R-IFG are directly linked by the arcuate fasciculus, a white matter pathway implicated in persistent stuttering (Chang et al., 2018). It may be the case that the CWS are susceptible to increases in planning load such that a higher planning load interferes with the ability of CWS to recruit the neural resources required to facilitate functioning during these phonological stages. It has been proposed that SMG also comprises somatosensory error cells, which effectively compare expected with actual sensory feedback during speech (Bohland & Guenther, 2006; Guenther, 2006). It also may be the case that the higher planning load interferes with the abilities of CWS to integrate sensory information during the planning of speech movements, which is reflected as reduced activation in this area. However, our previous study with AWS (Jackson, Wijekumar, et al., 2019) did not reveal the same pattern, suggesting that similar to R-IFG, AWS adapt to these suspected deficiencies. All of these claims warrant further testing with a larger sample of CWS, AWS, and CON.

4.2. Neural Substrates of Execution in Children Who Stutter

Arguably, the most common findings in the stuttering literature are reduced activation in left hemisphere and elevated activation in right hemisphere speech motor regions. Most of these studies focused on AWS. In the current study, the CWS exhibited less and greater activation than

the CON during the overt condition in L-preCG and R-preCG, respectively. The CWS also exhibited greater activation in L-preCG during the covert versus overt condition whereas the CON did not exhibit this pattern, suggesting that the increase in execution load interfered with the ability of the CWS to recruit neural resources in this area comparable to what CON recruit for speech production. These findings suggest that the atypical cerebral laterality long reported in stuttering research is due to execution processing and is present in CWS at least as young as 9-12 years old. However, the CWS exhibited a pattern in L-preCG opposite to the AWS in our previous study, such that the AWS exhibited greater activation in L-preCG for the overt compared to covert condition (Jackson, Wijekumar, et al., 2019). This may indicate that speakers who stutter adapt to execution load increases in adulthood, after the ages of 9-12 years. Further, both groups exhibited increased activation in R-preCG during the overt versus covert condition, but as stated above, the CWS exhibited greater activation than the CON during the overt condition. Thus, the neural processes that support the execution of fluent speech in CWS appear to be characterized by increased activation in R-preCG, potentially as a coping mechanism to adapt to the increase in execution load.

Similar to the planning contrast, the CWS exhibited greater activation in R-SMG during the low versus high execution load condition (i.e., covert), but the CON did not exhibit this pattern. This suggests that the increased load may have interfered with the ability of CWS to recruit neural activation similar to that of the CON in this area. It may also be that, similar to planning, the CWS exhibit difficulty with sensorimotor integration underlying execution processing. This result is in line with the AWS in Jackson et al. (2019), suggesting that this difficulty, that manifests in R-SMG, is present throughout development and into adulthood.

The CWS exhibited the same pattern in R-STG, that is, greater activation during the covert versus overt condition. STG is a key node in the speech network primarily responsible for the processing of speech sounds (Guenther, 2016). Reduced activation in R-STG during fluent speech production has also been described as one of the neural signatures of stuttering (Brown et al., 2005; Budde et al., 2014). STG and SMG are both connected to IFG via underlying white matter pathways, and both are critical for processing speech sounds; STG is involved in the perception of speech sounds whereas SMG is thought to parse the speech signal into sound features and meaning (Hickok & Poeppel, 2004), highlighting the critical role of SMG in sensorimotor integration (S. Cai et al., 2014; Daliri et al., 2018; Loucks et al., 2007; Loucks & De Nil, 2006). Differences in R-SMG and R-STG between CWS and CON during speech planning and execution may reflect a reduced ability to sufficiently activate these nodes when more complex sensorimotor integration is required. Interestingly, R-IFG appears to serve a different function during planning versus execution. The pattern in R-IFG during planning reflects under-activation due to increases in planning load, whereas during execution, the CWS exhibit over-activation in R-IFG with increased load. This may reflect compensatory processing in R-IFG to support fluent speech production. To bring clarity to these proposed interpretations, further investigation with larger sample sizes and downwards extensions in age of CWS is warranted.

That the CWS exhibited greater activation in R-SMG during the low versus high execution load condition and AWS exhibit the same pattern (see Jackson, Wijekumar, et al., 2019), suggests a potential difference in sensorimotor integration that begins in childhood and persists throughout adulthood in speakers who stutter. The CWS and AWS exhibited lesser activity during the overt versus covert condition suggesting under-activation in these

somatosensory areas during speech production, which may result from decreased sensitivity. Alternatively, these differences may reflect an under-reliance on somatosensory feedback, which contrasts with Max et al. (2004) who proposed that AWS demonstrate an overreliance on somatosensory feedback.

4.3. Inhibition in Stuttering

It has been proposed that the inhibitory network plays an important role in stuttering through error monitoring or conflict-induced slowing (Arenas, 2012; Aron et al., 2007). R-IFG and R-SMG comprise a network that underlies response inhibition and execution (Aron et al., 2004; Aron & Poldrack, 2006; W. Cai et al., 2014; Hartwigsen et al., 2018). For execution processing, the CWS exhibited lesser activation in R-SMG but greater activation in R-IFG, with high versus low execution load. It is possible that the increase in activation in R-IFG reflects inhibition when execution load increases, but that there is a mismatch within this network because R-SMG shows a reduction in activation when execution load increases. Perhaps more interesting is that the CWS exhibited greater activation in both of these areas for low versus high planning load. AWS do not show differences in these areas for planning (see Jackson, Wijekumar, et al., 2019), indicating that activity related to this inhibitory network during planning processing may be present in older school-age CWS but resolves in adulthood.

An alternative interpretation is that inhibitory networks, particularly those involved in proactive inhibition (Vanderhasselt et al., 2013) are central to the phenomenon of stuttering anticipation (Jackson, Wijekumar, et al., 2019; Neef et al., 2018). Anticipation refers to the awareness that upcoming speech will be stuttered. It is pervasive in both CWS and AWS, and by definition occurs at the level of speech motor planning (Jackson et al., 2015, 2018). The speaker becomes aware that should they continue speaking during anticipation without altering the

speech plan, they will produce stuttered speech. It may be that speech breaks down during planning in L-IFG – as indicated by the increase in activation in CON but not CWS in this area during the high planning load condition – and that R-IFG and R-SMG underlie awareness and decision-making related to how to proceed after the speaker knows that they will stutter. Speech was perceptually fluent in the current study, and participants were not asked about whether they anticipated stuttering, and therefore this interpretation requires further testing. Future investigations might include the extension of previous methods to elicit anticipation during neuroimaging (den Ouden et al., 2013; Jackson, Gracco, et al., 2019; Wymbs et al., 2013), which would allow for a comparison between anticipated and unanticipated (and stuttered versus non-stuttered) productions.

4.4. Limitations

The planning task in the current study may not have fully isolated planning processes. Specifically, two sets of 3-syllable non-words were produced: one set included the same syllables (e.g., “nauknauknau”) and the other included different syllables (e.g., “grassbrellna”). Our logic followed that of Lu et al. (2010): because the number of syllables was held constant, execution load was held constant. However, it is possible that execution load changed as well due to the sequencing of different articulatory configurations required for the different-syllable non-words. Importantly, our view of planning versus execution follows that of Mooshammer et al. (2012), in which articulatory configurations are specified during planning, and thus, the load associated with sequencing the actual movements is very low if not non-existent during execution. We assert that this is especially the case for simple words such as those presented to participants in this study.

Regarding the analysis, a 10 s. square block regressor was used for all trials, despite changing trial lengths. Thus, the data from each trial was convolved with block regressors of the same length even though trial lengths changed. This may reflect a limitation of the CBSI method which could have led to false positives. Further, Novi et al. (2020) pointed out that the CBSI method for artifact correction may be limited due to forcing anticorrelations in the data. HbO and HbR might not necessarily be anticorrelated, or at least not temporally aligned, and thus the CBSI approach could have led to false positives. The data were not corrected for possible autocorrelation (Huppert, 2016), though we acknowledge that this may have impacted the results.

4.5. Conclusion

The current study reveals possible neural substrates of speech planning and execution in 9-12 year-old CWS. These findings point to potential similarities and differences within the speech motor networks of CWS and AWS, though a direct comparison between age groups was not made and therefore, interpretations about group differences should be considered speculative. Nonetheless, these differences may be related to processes involving sensorimotor integration and inhibitory control. Future research should aim to directly compare different analytical approaches (e.g., for motion processing) across relevant experimental tasks, age-groups and tasks in stuttering individuals to reconcile differences in findings from previous literature. All results are based on a small sample, and are therefore subject to Type 1 and 2 errors and should be considered preliminary. Still, this study provides a basis for further investigation into speech planning and execution in CWS.

Figure Captions

Figure 1. Visual depiction of one trial from the planning (top) and execution (bottom) tasks. For the planning task, participants viewed the fixation mark, heard the non-word, produced the non-word, and rested for the jittered inter-stimulus interval of two, four, or six seconds. For the execution task, participants viewed the fixation mark followed by the picture, were presented with the “go” signal to either produce the word overtly (green frame) or covertly (red frame), and then rested for the jittered inter-stimulus interval of two, four, or six seconds. With permission: Jackson, E. S., Wijekumar, S., Beal, D. S., Brown, B., Zebrowski, P., & Spencer, J. P. (2019). A fNIRS investigation of speech planning and execution in adults who stutter. *Neuroscience*, 406, 73-85.

Figure 2. (A) Probe Geometry with channel numbers and layout. Red indicates source optodes, blue indicates detector optodes. Green lines indicate short source channels. (B) Digitized scalp landmarks and source and detector positions.

Figure 3. Significant interactions between group and condition clusters for planning task, and follow-up posthoc tests including averaged beta values for each participant. Oxygenated hemoglobin, pink = same syllable non-words (less complex), red = different syllable non-words (more complex); Deoxygenated hemoglobin, light blue = same syllable non-words (less complex), dark blue = different syllable non-words (more complex); L = left; R = right; IFG = inferior frontal gyrus; SMG = supramarginal gyrus.

Figure 4. Significant interactions between group and condition clusters for execution task, and follow-up posthoc tests including averaged beta values for each participant. Oxygenated hemoglobin, pink = covert (less complex), red = overt (more complex); Deoxygenated hemoglobin, light blue = covert (less complex), dark blue = overt (more complex); L = left; R = right; preCG = precentral gyrus; IFG = inferior frontal gyrus; STG = superior temporal gyrus; SMG = supramarginal gyrus.

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