NEURAL CORRELATES OF PHONOLOGICAL ABILITIES IN CHILDREN WHO STUTTER

By

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ABSTRACT

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Stuttering is a complex neurodevelopmental disorder that is likely caused by an interaction of multiple factors, including genetics, linguistic ability, motor development, temperament, and neural development (Smith & Weber, 2017). Previous research has reported that children who stutter exhibit lower phonological abilities than children who do not stutter. In addition, it is commonly reported that children who stutter, as a group, exhibit decreased white matter integrity along the left dorsal language pathway (Chang, Garnett, et al., 2018; Neef et al., 2015), which is critical in supporting phonological processing and production.

In this study the phonological abilities were examined in children who stutter (N = 24) and children who do not stutter (N = 31) using whole-word measures that were produced during spontaneous connected speech. These phonological scores, phonological mean length of utterance (PMLU) and proportion of whole-word proximity (PWP), provide a quantitative measure of phonological complexity and accuracy. Examining whole-word segments instead of smaller segments of speech considers the fact that children's individual words become longer and more complex as vocabulary increases (Ingram, 2002). In addition to the analysis of phonological skills, the current study examined how phonological skills are associated with white matter integrity differences in children who stutter relative to children who do not stutter.

Results from this study show that children who stutter have similar phonological skills as children that do not stutter, but the association between phonological ability and white matter integrity is significantly different between the two groups. The children who stutter exhibited a

greater association between PWP and white matter integrity in the bilateral dorsal language pathways than children who do not stutter. Notably, these areas were along the arcuate fasciculus, and included the right IFG (BA 45/47), right insula, and left insula/Rolandic operculum. In addition, children who do not stutter showed robust white matter integrity scores in bilateral dorsal and ventral pathways associated with age, which was not observed children who stutter.

Overall, major findings from this investigation indicate that: 1) children who stutter do not differ from children who do not stutter in terms of phonological complexity as measured from spontaneous speech samples, 2) there is greater association between PWP and increased white matter integrity in bilateral dorsal tracts in children who stutter relative to children who do not stutter, 3) children who stutter exhibit an overall attenuated age-related white matter integrity growth in areas overlapping with those that showed heightened PWP-white matter integrity associations. The greater association between PWP and the bilateral dorsal tracts in children who stutter in the face of attenuated age-related increases in the same areas, suggest that stronger links between PWP and dorsal pathway integrity may be needed in children who stutter to achieve comparable phonological development to children who do not stutter. These results provide novel insights into phonological development in children who stutter, and the behavior – neural development associations relevant to phonological development that seem to differ in children who stutter.

This dissertation is dedicated to the memory of my late Father, Donald J. Spray, who always encouraged me to follow my dreams, regardless of how crazy they might be. This work is also dedicated to the memory of my Grandmother, Donna M. Taylor, who first inspired my passion for speech-language pathology and the brain.

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KEY TO ABBREVIATIONS

PMLU	Phonological mean length of utterance
DTI	Diffusion tensor imaging
SLD	Stuttering-like disfluency
OD	Other disfluency
MDP	Multifactorial dynamic pathways
IPSyn	Index of productive syntax
ERP	Event-related potential
MRI	Magnetic resonance imaging
CSF	Cerebrospinal fluid
BOLD	Blood-oxygenated-level dependent
fMRI	Functional magnetic resonance imaging
rsfMRI	Resting state functional magnetic resonance imaging
PET	Positron emission topography
IFG	Inferior frontal gyrus
fNIRS	Functional near-infrared spectroscopy
STG	Superior temporal gyrus
AF	Arcuate fasciculus
SLF	Superior longitudinal fasciculus
ILF	Inferior longitudingal fasciculus
IFOF	Inferior fronto-occipital fasciculus
ECFS	Extreme capsule fiber system

FA	Fractional anisotropy
MD	Mean diffusivity
AD	Axial diffusivity
RD	Radial diffusivity
BA	Brodmann area
DIVA	Directions into velocity of articulators
UF	Uncinate fasciculus
GFTA-2	Goldman-Fristoe Test of Articulation, Second Edition
KLPA-2	Khan-Lewis Phonological Analysis, Second Edition
BBTOP	Bankson-Bernthal Test of Phonology
PCC	Percentage of consonants correct
APP-R	Assessment of Phonological Processes-Revised
PWP	Proportion of Whole-Word Proximity
BBTOP-CI	Bankson-Bernthal Test of Phonology, Consonant Inventory
EEG	Electroencephalography
tPMLU	Target phonological mean length of utterance
aPMLU	Actual phonological mean length of utterance
CWNS	Children who do not stutter
CWS	Children who stutter
F	Female
SSI	Stuttering Severity Instrument
IRB	Institutional Review Board
CBQ	Children's Behavior Questionnaire

WPPSI-III	Wechsler Preschool and Primary Scale of Intelligence, Third Edition
KiddyCAT	Communication Attitude Scale for Pre-K and Kindergarten Children Who Stutter
TOCS	Test of Childhood Stuttering
SSI-4	Stuttering Severity Instrument, Fourth Edition
CELF-P:2	Clinical Evaluation of Fundamentals-Preschool, Second Edition
CELF-5	Clinical Evaluation of Fundamentals, Fifth Edition
EHI	Edinburgh Handedness Inventory
PPT	Purdue Pegboard Test
MLU	Mean length of utterance
NDW	Number of different words
VOCD	Vocabulary diversity
%SLD	Percentage of stuttering-like disfluencies
%OD	Percentage of other disfluencies
CLAN	Computerized Language Analysis
M	Male
SD	Standard deviation
CHAT	Codes for the Human Analysis of Transcripts
XML	Extensible markup language
IPA	International Phonemic Alphabet
ICC	Intraclass correlation coefficient
TTR	Type-token ratio
TR	Time of repetition
TE	Time of echo

FOV	Field of view
kHz	Kilohertz
mm	Millimeters
cm	Centimeters
s/mm2	Seconds per millimeters square
b	Beta
DW	Diffusion weighted
DARTEL	Diffeomorphic image registration algorithm
SPM	Statistical Parametric Mapping
FWHM	Full width at half maximum
AFNI	Analysis of Functional NeuroImages
LH	Left hemisphere
RH	Right hemisphere
RO	Rolandic operculum
L	Left
R	Right
MTG	Middle temporal gyrus
MFG	Middle frontal gyrus
IFG (operc)	Inferior frontal gyrus (opercularis)
n.s.	Not significant
SLI	Specific Language Impairment
ADHD	Attention deficit hyperactivity disorder
DOB	Date of birth

INTRODUCTION

Stuttering, or childhood onset fluency disorder, has been reported to affect 5 to 8% of preschool-aged children in the United States, and persists in approximately 1% of the adult population (Bloodstein & Bernstein Ratner, 2008; Yairi & Ambrose, 2013). The onset of stuttering occurs typically early in development, while children are beginning to create two-word utterances and short phrases. Approximately 80% of children who stutter eventually recover from the disorder (Yairi & Ambrose, 2013); however, for roughly 3 million Americans, this is not the case. As stuttering persists, these individuals may experience emotional reactions during speaking, such as fear, embarrassment, anger, and helplessness, which impact overall quality of life (Yaruss, 2007).

Although a rich literature exists within the field of stuttering, it has become evident that stuttering is a complex disorder that does not appear to be caused by a single etiology. Many researchers and clinicians agree that the cause of stuttering may be due to an interaction between many factors, including genetics, linguistic ability, motor development, temperament, and neural development (Smith & Weber, 2017). With regards to linguistic ability, many studies have reported differences in expressive and receptive language scores between children who stutter and children who do not stutter (Ntourou et al., 2011; cf. Nippold, 2012). In addition, research has suggested possible links between phonological development and stuttering (Arndt & Healey, 2001; Blood et al., 2003; Louko et al., 1990; Paden et al., 1999; Spencer & Weber-Fox, 2014; Wolk et al., 1993; Yaruss et al., 1998; cf. Nippold, 2002). Despite this evidence, it is still difficult to determine whether subtle differences in phonological ability are comorbid with stuttering or are associated with the onset of stuttering, because the nature of the link is not clear (for review, see Bloodstein & Bernstein Ratner, 2008).

More recently, efforts have been made to study neurophysiological development of children who stutter and children who do not stutter to determine whether underlying neural functions can provide additional information about the onset and course of stuttering. It has become increasingly apparent that children who stutter exhibit differences in underlying neuroanatomical structure and function (Beal et al., 2013; Chang, Angstadt, et al., 2018; Chang et al., 2008, 2015; Chang & Zhu, 2013; Chow & Chang, 2017; Garnett et al., 2018; Hampton Wray & Spray, 2020; Mohan & Weber, 2015; Usler & Weber-Fox, 2015; for review, see Chang, Garnett, et al., 2018). One aspect of those differences involves the neural structures underlying language formulation and speech production. Specifically, it has been proposed that speech perception and production involves the bilateral ventral and dorsal language pathways (Hickok & Poeppel, 2004; Saur et al., 2008). Evidence has suggested that the dorsal pathways are critically involved in mapping sound onto articulatory-based representations (Hickok & Poeppel, 2004) via the arcuate fasciculus and superior longitudinal fasciculus. Compared to children who do not stutter, children who stutter are commonly reported to exhibit decreased white matter integrity along the dorsal language pathway in the left hemisphere (Chang, Garnett, et al., 2018).

While the dorsal language pathway has been hypothesized to play a critical role in mapping sound to articulation, researchers have yet to examine the relationship between phonology and white matter integrity in children who stutter. The objective for this study was to determine whether an aberrant association exists between phonological abilities and white matter integrity in children who stutter compared to children who do not stutter. Unlike traditional phonology assessments, which use a segmental approach, the current study utilized a novel whole-word measure, phonological mean length of utterance (PMLU) to determine whether children who stutter exhibit differences in phonology compared to children who do not stutter. These whole-word measures were correlated with a measure of white matter integrity (fractional anisotropy, derived from diffusion tensor imaging (DTI)) in the dorsal language pathway to examine whether differences exist between children who stutter, and children with no history of stuttering. The examination of whole-word phonological abilities in relation to white matter structural brain imaging has not been attempted before, in part due to difficulties encountered when scanning very young children. Findings from this study shed light on questions that have yet to be addressed within the current literature, including differences in whole-word phonology, which might be sensitive in detecting phonological complexity differences, in addition to phonological development differences between children who stutter and children who do not stutter. The findings from this study are also expected to provide information regarding possible differences in brain structure associated with phonology.

CHAPTER 1:

LITERATURE REVIEW

Stuttering, or childhood onset fluency disorder, is a complex neurodevelopmental disorder (Smith & Weber, 2017) that involves disruptions in the forward flow of speech (Van Riper, 1971). Stuttering typically manifests itself as involuntary occurrences of sound or syllable repetitions, prolongations in speech, or prolonged articulatory postures. These may be accompanied by avoidance and struggle behaviors. Stuttering behaviors may also be accompanied by concomitant physical symptoms such as eye blinks, physical tension in the lips/face, or jerking of the head (Van Riper, 1971). In recent years, the definition of stuttering has expanded beyond the overt perceptual characteristics to include factors such as resulting anxiety about speaking, or limitations in a person's ability to perform daily activities or restrictions in a person's ability to participate in social or occupational endeavors (American Psychiatric Association & American Psychiatric Association, 2013). Because stuttering is observed during overt speech, and its onset coincides with a time of rapid expansion in linguistic abilities, an extensive body of research has investigated the link between language and stuttering, while also examining the relationship between grammatical complexity and the likelihood of stuttering (Bloodstein & Bernstein Ratner, 2008). The results of studies examining linguistic proficiency in children who stutter provide evidence that these children exhibit a range of language abilities that are quite similar to children who do not stutter (Bernstein Ratner, 1997; Ntourou et al., 2011; R. V. Watkins et al., 1999; R. V. Watkins & Johnson, 2004; R. V. Watkins & Yairi, 1997). Researchers have suggested that syntactic and linguistic complexity may contribute to the likelihood of stuttering (Bernstein Ratner & Sih, 1987; Buhr & Zebrowski, 2009; MacPherson &

Smith, 2013; Melnick & Conture, 2000; Ratner, 1995); however, the relationship between stuttering and language remains unclear, and no single theory has accounted for the diverse nature of the disorder.

The diagnosis of stuttering is typically determined by examining the frequency of stuttering-like disfluencies (SLDs) in continuous speech (Ambrose & Yairi, 1999; Schwartz & Conture, 1988; Zebrowski, 1991). Stuttering-like disfluencies (SLDs) consist of three observable speech elements: 1) repetitions of part words, 2) repetitions of single-syllable words, and 3) dysrhythmic phonation (i.e., sound prolongations and blocks). Other disfluencies (ODs) are those that commonly occur in typical speakers as well as people who stutter, which include: phrase repetitions, revisions, and interjections. The distinction between typical disfluencies and stuttering is based on evidence that SLDs occur more frequently in people who stutter than in people who do not stutter. Ambrose and Yairi (1999) reported that people who stutter produced significantly more SLDs than fluent speakers; however, these two groups produce approximately the same number of ODs during spontaneous speech (Ambrose & Yairi, 1999). Based on these findings, it has been suggested that children near stuttering onset who present with 3 or more SLDs per 100 syllables should be suspected as exhibiting stuttering (Ambrose & Yairi, 1999). In addition, because stuttering severity fluctuates across various contexts, conversations, and even utterances, it is recommended that speech samples include at least 1000 syllables when evaluating young children who may stutter, and occur in a number of contexts, which include: reading, storytelling/monologues, and conversations with a parent, clinician, and/or friend (Ambrose & Yairi, 1999).

Researchers have estimated that developmental stuttering affects 5-8% of preschool-age children, with typical onset between 30-48 months (Bloodstein & Bernstein Ratner, 2008; Yairi

& Ambrose, 2013). Recent studies investigating stuttering onset have suggested that stuttering typically begins between 24-35 months of age, with 95% of onsets occurring between 24-48 months of age, and is found throughout the world, across all races, ethnicities, and social classes (Bloodstein & Bernstein Ratner, 2008; Yairi & Ambrose, 2013). Stuttering is more prevalent in males than females (Yairi & Ambrose, 2013), with a male-to-female ratio of approximately 2 to 1 (Yairi & Ambrose, 1992; Yairi & Ambrose, 1999). The prevalence of stuttering in adult populations is approximately 1%, with a male-to-female ratio at approximately 3 or 4 to 1 (Yairi & Ambrose, 2013). Given the lower prevalence rate among adults, it is suggested that approximately 80% of children naturally recover from stuttering (Yairi & Ambrose, 2013), with studies reporting recovery rates ranging from 68% (Ryan, 2001) up to 94% (Månsson, 2000, 2005). At present, researchers and clinicians do not have objective markers to determine with confidence which children will persist in stuttering and which children will recover. Research has sought factors that may help predict which children will recover from the disorder (Yairi & Ambrose, 1999). These factors include familial history, sex, age of onset, time since onset of stuttering, language development, and more.

The Multifactorial Dynamic Pathways Model of Stuttering & the Brain

For the vast majority of speakers, speech production is a seemingly effortless task that allows people to convey information in a fluid manner. That said, speech production is a complex process that requires the coordination of multiple neural systems that support cognitive and language cortical areas, which interface with speech motor control regions that encompass cortical and subcortical areas. Speech production requires timely and precise coordination of approximately 100 muscles in the face, larynx, and respiratory system (Chang, Garnett, et al., 2018). The ability to sequence and articulate speech syllables, while also regulating pitch, rhythm, and prosody, requires efficient communication between a number of different cortical and subcortical regions as well as between the two hemispheres. Theories of stuttering have proposed that language and temperament contribute to stuttering (Adams, 1990; Starkweather & Gottwald, 1990; for review, see Bloodstein & Bernstein Ratner, 2008). The multifactorial dynamic pathways (MDP) theory of stuttering has advanced the notion that increased linguistic complexity and/or temperament factors may place additional stress on neural systems as they interact with unstable, aberrantly functioning speech motor networks, leading to breakdowns in speech fluency (Smith & Weber, 2017).

The MDP model suggests that instability within the speech motor system may be due to a reduced ability to develop and establish stable motor programs, even during fluent speech (Smith & Weber, 2017). Previous studies have reported that the inter-articulator coordination patterns of adults and children who stutter are significantly more variable than those seen in speakers who do not stutter during complex sentence production and nonword learning tasks (MacPherson & Smith, 2013; Smith et al., 2012; Walsh et al., 2006). These studies found that while fluent controls performed at or near ceiling levels, the adults and children who stutter required additional training opportunities to increase proficiency. Such findings suggest that the speech motor learning process in adults and children who stutter is less mature or less stable than that of speakers who do not stutter. This may lead to breakdowns in speech production when in the presence of emotional and linguistic stress. These studies have indicated that children as young as 4 to 5 years of age present with atypical speech motor development (MacPherson & Smith, 2013; Walsh et al., 2015).

Given the variability of inter-articulator coordination patterns observed in complex sentence production, researchers have examined the association between language production and stuttering. Previous studies have found interactions between syntactic complexity and the exacerbation of stuttering for children and adults who stutter (MacPherson & Smith, 2013; Smith et al., 2012). Leech and colleagues (2017) reported that growth in productive syntax, as measured by Index of Productive Syntax (IPSyn), was predictive of stuttering persistence or recovery, with children who recover exhibiting greater growth between 57 and 81 months of age. Stuttering onset occurs during a period that is marked with rapid expansion of linguistic skills (Bloodstein & Bernstein Ratner, 2008; Wagovich et al., 2009; R. V. Watkins et al., 1999; Yairi et al., 2005). Together, these studies, along with many others, lend support to the notion that stuttering may have a connection to language development.

The study of speech perception has greatly expanded over the past twenty years, using various approaches to examine the relationship between language processing and stuttering via electrophysiological measures. Event-related potentials (ERPs) utilize time-locked stimuli to examine synchronous activity of neuronal networks elicited by specific stimuli (Luck, 2005). These studies have provided evidence that the neural processes mediating semantic, syntactic, and phonological processing differ between children who do and do not stutter (Hampton Wray & Spray, 2020; Mohan & Weber, 2015; Usler & Weber-Fox, 2015; Weber-Fox et al., 2013; Weber-Fox & Hampton, 2008). For example, Weber-Fox and colleagues (2013) reported that preschool-aged children who stutter exhibit atypical lateralization of hemispheric functions related to language processing. More specifically, children who stutter were found to process syntactic violations using the right hemisphere while children who do not stutter engaged the left hemisphere (Weber-Fox et al., 2013). Together, the results of ERP studies suggest that children who stutter engage different neural systems relative to their fluent peers for language processing, and this may reflect less mature language development in children who stutter relative to children who do not stutter. Although these studies have reported differences in neural processing, it is important to note that many of the aforementioned studies failed to find behavioral differences. This suggests that the neural differences observed between the two groups may be related to trait differences as well as compensatory processes that allow for similar behavioral performance between the groups.

Functional Neuroanatomical Differences in People Who Stutter

The following section will provide an overview of magnetic resonance imaging, and how these images are acquired. Second, functional neuroimaging studies related to stuttering will be discussed, followed by a review of structural neuroimaging studies in children who stutter. The functional and structural neuroimaging sections provide an overview of findings that suggest anomalous structure and function in areas that auditory-motor integration. Namely, these areas include the left inferior frontal gyrus, ventral premotor cortex, ventral motor cortex, and the posterior superior temporal gyrus.

Magnetic Resonance Imaging

Magnetic resonance imaging (MRI) is one of the most commonly used non-invasive technique to examine brain anatomy and function. It utilizes a strong magnetic field and radiofrequency and capitalizes on the fact that the human body contains vast stores of hydrogen atoms, which differ in concentration depending on different tissue types (e.g., gray matter, white matter, cerebrospinal fluid). When a person is placed parallel to the bore of the MRI scanner, the hydrogen atomic nuclei begin to precess around an axis that is either parallel (low-energy state) or antiparallel to the magnetic field, which is a high-energy state (Huettel et al., 2009). Thus, while the person lies within the bore of the scanner, many of the nuclei are in a low-energy state, resulting in a net magnetization parallel to the magnetic field (Huettel et al., 2009). This is called longitudinal magnetization. In contrast, high-energy nuclei are associated with transverse magnetization. Radiofrequency coils are used to send electromagnetic waves to move low-energy nuclei into a high-energy state, thereby converting the longitudinal magnetization into transverse magnetization (Huettel et al., 2009). This process is known as excitation. Additional coils, known as gradient coils, are used to create spatial variation in the strength of the magnetic field. In total, there are three gradients (representing x, y, z axes), which allow for the spatial encoding needed to resolve an image (Huettel et al., 2009). After radiofrequency coils are turned off, two types of changes occur in the MR signal over time (i.e., relaxation): a recovery of longitudinal magnetization (T1), and a decay in transverse magnetization (T2). In addition, transverse relaxation can be combined with changes in the local magnetic field (T_2*), which is used for blood-oxygenated-level dependent (BOLD) contrast used for functional MRI (Huettel et al., 2009).

Unlike positron emission topography (PET), which utilizes radioactive tracers to examine neural activity, functional MRI (fMRI) does not require radioactive material to acquire the images (Huettel et al., 2009). fMRI makes use of the hemodynamic response function to measure regional changes in blood oxygenation over time, which allows researchers to localize neurological activity using T_2* (Huettel et al., 2009). Hemoglobin has magnetic properties depending on whether it is oxygenated (diamagnetic) or deoxygenated (paramagnetic). When neurons become active, the vascular system responds by supplying an excess amount of oxygenated hemoglobin to that particular region (Huettel et al., 2009). The abundance of oxygenated hemoglobin results in a decrease in the amount of deoxygenated hemoglobin, which, in turn, causes a decrease in the signal loss (Moseley & Glover, 1995). This process of increased oxygenation is known as the hemodynamic response, and is the basis for the signal change linked to brain activity that is detected and reported in fMRI studies (Huettel et al., 2009). Most fMRI studies have examined brain activity patterns associated with a task (e.g., listening to tones, speech sounds, visual tasks, finger movement tasks, etc.). Many resting state functional MRI (rsfMRI) studies have been conducted to examine temporally correlated changes in spontaneous blood-oxygenation level dependent (BOLD) signal oscillations while an individual is asked to relax and keep his/her eyes open or closed without falling asleep (Greicius et al., 2009). These temporal correlations are thought to reflect intrinsic functional connectivity among distinct neural networks as well as underlying structural connectivity (Greicius et al., 2009; Hampson et al., 2002; Obrig et al., 2000).

Functional Imaging Studies in Stuttering Using fMRI & Positron Emission Tomography

In early twentieth century, stuttering was hypothesized to be caused by abnormal laterality of the brain (Orton, 1927). Significant advances in the field of neuroimaging that encompassed fMRI and PET in the last two decades have led to a surge of studies in the field of stuttering to examine functional neuroanatomical differences in people who stutter compared to fluent speakers. Some convergent findings based on meta-analyses of functional imaging studies of adults who stutter include increased activity in the right premotor areas, and abnormally reduced activity in auditory association areas during speech tasks (Belyk et al., 2015; Braun et al., 1997; Brown et al., 2005; Budde et al., 2014; Chang et al., 2009; Fox et al., 2000). These findings lend support to theories of stuttering that have posited that core deficits in stuttering is associated aberrant integration of auditory feedback into the speech motor program (Belyk et al., 2015; Max et al., 2004).

Over the past 10 years, a number of studies have also reported findings based on examining children who stutter relative to age-matched children who do not stutter. Similar to adults who stutter, functional differences in brain regions supporting auditory-motor integration (e.g., left premotor, motor, and auditory cortical areas) were found in children who stutter (Chang, Angstadt, et al., 2018; Chang et al., 2009; Chang & Zhu, 2013; for review, see Chang, Garnett, et al., 2018). In addition, white matter pathways responsible for interconnecting these cortical areas are also found to show reduced integrity in children who stutter (Chang et al., 2008, 2015; Chow & Chang, 2017; Connally et al., 2014), which is discussed in more detail in a later section.

During speech production, perisylvian cortical structures, such as the inferior frontal gyrus (IFG), ventral premotor cortex, ventral motor cortex, and posterior superior temporal gyrus serve a critical function of integrating auditory feedback into the speech motor program. This process is known as auditory-motor integration. Within the context of speech production, the left IFG supports speech planning, but also updates articulation plans based on sensory feedback (Guenther & Hickok, 2016). Convergent findings indicate that people who stutter exhibit subtle functional differences among these regions supporting auditory-motor integration (Braun et al., 1997; Chang et al., 2009; Chang & Zhu, 2013; Fox et al., 1996; K. E. Watkins et al., 2008). For example, PET and fMRI studies demonstrate that people who stutter exhibit reduced activity between motor and auditory areas in adults who stutter (Braun et al., 1997; Fox et al., 1996). In addition, people who stutter have been found to exhibit reduced functional connectivity between motor and auditory areas within the left hemisphere (Neef et al., 2016; K. E. Watkins et al., 2008). Children who stutter also exhibit decreased connectivity in the left hemisphere between the posterior superior temporal gyrus and the insula, supplementary motor area, and the IFG (Chang & Zhu, 2013).

In contrast to the reduced activation and function connectivity observed in the left hemisphere, it is commonly reported that people who stutter exhibit an overactivation within the right hemisphere (Belyk et al., 2015; Brown et al., 2005; Budde et al., 2014; Fox et al., 1996; Preibisch et al., 2003). These overactive regions are within frontal motor areas, and encompass the primary motor cortex, premotor cortex, the pre-supplementary area, the IFG, the insula, and the frontal and Rolandic operculum (Neef et al., 2016). This same overactivation in children who stutter has not been reported to the same extent while using fMRI. On the other hand, studies using ERPs and functional near-infrared spectroscopy (fNIRS) have reported that children who stutter exhibit greater right lateralization while processing syntactic phrase structures (Weber-Fox et al., 2013) and phonemic distinctions (Sato et al., 2011), respectively.

The aforementioned studies provide evidence that the overactivation observed within the right hemisphere may serve as a compensatory mechanism for attenuated connectivity and activation in the left hemisphere. Because children who stutter do not exhibit the same overactivation in the right hemisphere, this may develop as a child ages and stuttering persists. Taken together, mounting evidence suggests that auditory-motor integration may be less efficient among people who stutter because of functional deficits in the left hemisphere. In addition to these functional anomalies, anatomical differences have also been found among children and adults who stutter.

Structural Imaging Studies in Adults & Children Who Stutter

Similar to functional neuroimaging studies, accumulating evidence has shown that adults who stutter exhibit anomalous gray and white matter volume differences among speech planning and auditory-motor integration structures of the left and right hemispheres (Beal et al., 2015; Chang et al., 2009; Cykowski et al., 2008, 2010; Fox et al., 2000; Kell et al., 2009; Sommer et al., 2002). For example, morphometry studies of adults who stutter have report both increases (Beal et al., 2007; Lu et al., 2010; Song et al., 2007) and decreases (Kell et al., 2009; Lu et al., 2010) in gray matter volume. Other studies have also reported no differences between adults who stutter and adults who do not stutter (Jäncke et al., 2004). Adults who stutter have been reported to exhibit increased white matter volume underlying the STG, IFG, precentral gyrus (Jäncke et al., 2004). Beal and colleagues (2007) also reported increased white matter volume underlying the right inferior frontal gyrus, but also extended findings to include the right insula. The mixed gray matter findings in adults who stutter highlights the importance of examining children who stutter to determine whether gray and white matter structure differ in people who stutter, become apparent over time.

Thus, morphometric studies have also examined children who stutter, and have consistently reported anomalous structure within left hemisphere speech networks (Beal et al., 2013; Chang et al., 2008; Chang & Zhu, 2013). A recent study by Garnett and colleagues (2018) found that children with persistent stuttering had decreased cortical thickness in motor and premotor areas in the left hemisphere when compared to fluent controls and children whose stuttering eventually subsided. Previous studies have reported that children who stutter have decreased gray matter volume in critical speech motor areas such as the left IFG and the motor and pre-motor cortices (Beal et al., 2013; Chang et al., 2008).

Taken together, previous studies that have reported aberrant functional and structural differences in adults and children who stutter. During speech production, children who do not stutter exhibit significant activation over the left dorsal inferior frontal gyrus and left premotor cortex while children who stutter showed decreased activation (Walsh et al., 2017). The structural and functional anomalies reported in left perisylvian area known to support speech suggest that these differences may impede the formulation and transmission of speech motor plans required for fluent speech. These distant cortical regions are interconnected via white matter pathways, which show decreased structural integrity within the left hemisphere. These structural differences within the left hemisphere might lead to structural and functional differences that are observed within adults who stutter.

White Matter Integrity Underlying Language & Stuttering

The formulation and fluent transmission of speech motor plans is achieved via a number of cortical and subcortical structures that are interconnected via major white matter tracts. The arcuate fasciculus (AF), which forms a branch of the superior longitudinal fasciculus (SLF), inferior longitudinal fasciculus (ILF), inferior fronto-occipital fasciculus (IFOF), and the extreme capsule fiber systems (ECFS) are present in both hemispheres, connecting spatially distant intra-hemisphere regions that play a role in speech and language production and perception. To examine white matter structure at a microscopic level, researchers have utilized a technique called diffusion tensor imaging (DTI). Diffusion tensor imaging studies have revealed structural changes associated with major white matter tracts relevant to speech and language production in people who stutter. The following section will review how diffusion tensor images are collected, how white matter integrity changes across development and training, reflecting plasticity associated with these processes. Finally, major findings from DTI studies that examined children who stutter will be reviewed.

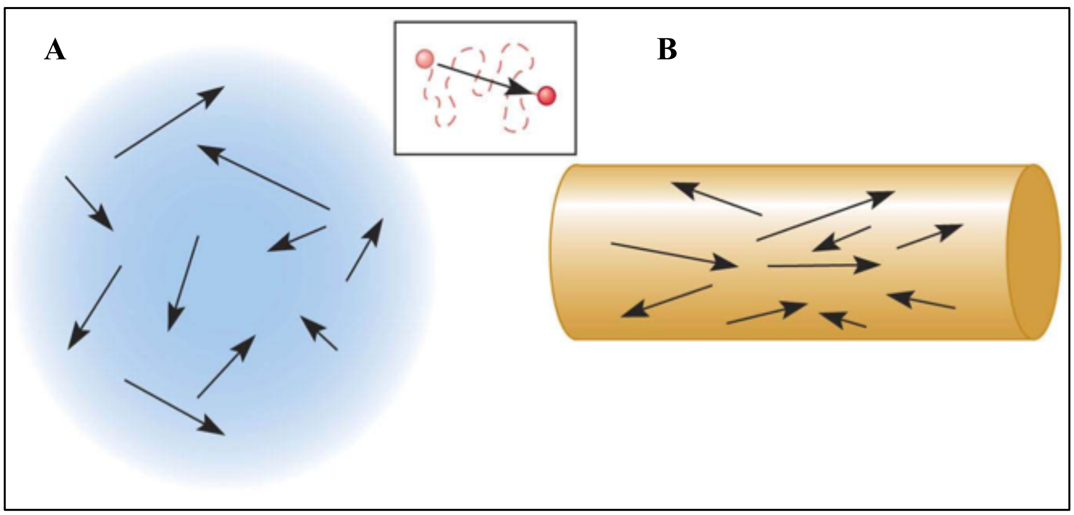
DTI Indices Reflecting White Matter Integrity

Diffusion tensor imaging (DTI) is an MRI technique that allows quantitative assessment of white matter microstructure within the brain. Although the technique provides visualization and microscopic characterization of white matter, it does not measure white matter structure directly (Huettel et al., 2009). DTI uses a special MRI sequence that tags movement of water molecules (Stejskal & Tanner, 1965). The acquisition of diffusion-weighted images requires the application of a strong gradient pulse in numerous directions (greater than six) to gain information about molecular mobility reflective of microstructure and infer orientation of axonal fibers (Dell'Acqua & Catani, 2012). A tensor model is used to describe diffusion in three-dimensional space (Basser et al., 1994). In addition, the diffusion tensor can be visualized as an ellipsoid, which provides a three-dimensional profile of the distance water molecules travel (Maffei et al., 2015).

Within the ventricles, water diffusion in cerebrospinal fluid occurs in equal magnitude in all directions (isotropic diffusion). This is known as Brownian motion (Feldman et al., 2010). In contrast, diffusion of water molecules present along white matter tracts are restricted by axonal membrane and the myelin sheath that encapsulates the axon. Water diffusion in white matter thus occurs in a direction parallel to the longitudinal axis of the white matter fiber, and less in the anti-parallel direction (anisotropic diffusion). Figure 1 provides an example of isotropic and anisotropic diffusion. Thus, this technique allows for quantification of the magnitude and direction of molecular diffusion of water molecules within a voxel, the basic unit of three-dimensional imaging scans (Le Bihan, 2003; Moseley et al., 1990).

Figure 1:

Isotropic & Anisotropic Diffusion Within the Brain



Note. Figure 1 provides examples of (A) isotropic diffusion and (B) anisotropic diffusion. Isotropic diffusion is commonly observed in cerebrospinal fluid, while anisotropic diffusion is observed in white matter pathways. Adapted from "Functional Magnetic Resonance Imaging (3rd ed.)," by S. A. Huettel, A. W. Song, and G. McCarthy, 2014, p. 140. Copyright 2014 by Sinauer Associates, Inc.

A commonly derived measure from DTI is called fractional anisotropy (FA), which reflects white matter coherence/integrity. FA is a scalar value that ranges between '0' and '1', indicating isotropic or anisotropic diffusion, respectively. For example, FA values near the maximum value of '1' represents almost perfect anisotropy, namely water diffusing along the same preferred axis (e.g., corpus callosum fibers), while FA values near '0' indicate almost perfect isotropy, or equal diffusion of water molecules in any direction, suggesting decreased white matter coherence. FA measures can be affected by factors such as axonal packing density, axonal diameter, amount of myelination, and/or crossing fibers within a voxel (Le Bihan et al., 2001). FA has been used quite extensively in adult and pediatric populations to gain a greater understanding of neural development, as well as clinical pathologies such as cerebrovascular accidents and traumatic brain injuries. FA generally increases during development (Barnea-Goraly et al., 2005; Giorgio et al., 2008; Neil et al., 2002); for instance it has been shown to show consistent increases with age in regions such as the arcuate fasciculus and superior longitudinal fasciculus (Lebel et al., 2008; Qiu et al., 2008; Rollins et al., 2010). Other values derived from DT, such as mean diffusivity (MD), axial diffusivity (AD), and radial diffusivity (RD) inform researchers about the diffusion of water in all directions, the diffusivity parallel to fibers, and diffusivity perpendicular to the fibers, respectively (Huettel et al., 2009).

Clinically, these measures can be used to inform physicians about any lesions in white matter. It has been shown to be sensitive to detecting pockets of white matter disease occurring in diseases such as multiple sclerosis (Filippi et al., 2001; Roosendaal et al., 2008), as well as detecting hemorrhagic cerebrovascular accidents. DTI has also allowed researchers to gain a greater understanding of white matter structure and function across brain development. Language development, skill acquisition, and intensive training/intervention, is known to create structural changes within white matter structure that can be captured with DTI (Bengtsson et al., 2005; Scholz et al., 2009).

White Matter Microstructure Changes Associated with Skill Acquisition

Action potentials occurring within the brain increase myelination (Demerens et al., 1996; Ishibashi et al., 2006; Mensch et al., 2015; Stevens et al., 1998, 2002; Zalc & Douglas Fields, 2000), which also influence white matter integrity measures. With this in mind, a number of studies have provided evidence that motor skill training increases white matter integrity (Halwani et al., 2011; Sampaio-Baptista et al., 2013; Scholz et al., 2009; Wang et al., 2014). For example, highly proficient musicians have been reported to exhibit greater white matter volume and integrity in the bilateral arcuate fasciculi compared to non-musicians (Halwani et al., 2011). This finding suggests that musicians learn to associate motor actions with auditory feedback, which in turn contributes to increased training of an auditory-motor feedback loop via the arcuate fasciculus. Neuroplastic changes following musical training are also found in young children (Hyde et al., 2009). This further suggests that white matter integrity can be facilitated with training across the in younger children, which also provides support for the notion that treatment facilitates structural changes within the brain.

In addition to motor skill training, changes in white matter integrity is associated with language acquisition. A number of studies have provided evidence that second-language learning, along with reading training, increases integrity of major white matter pathways, such as the arcuate fasciculus and superior longitudinal fasciculus (Wandell & Yeatman, 2013; Yeatman et al., 2011, 2012). These changes are observed in both adults and children, and highlight the neuroplastic changes that occur in response to short-term training (Hofstetter et al., 2013, 2017; Huber et al., 2018; Sagi et al., 2012; Sampaio-Baptista et al., 2013; Scholz et al., 2009). Huber and colleagues (2018) provided evidence that intensive reading intervention (occurring four hours per day, five days per week, for a total of eight weeks) improves reading skills and is associated with increased white matter integrity along the left arcuate fasciculus and inferior longitudinal fasciculus. These white matter pathways connect distant regions critical for reading function, such as the ventral occipito-temporal, superior temporal, and inferior frontal cortex. Collectively, training studies related to skill acquisition and remediation have provided clear evidence that neuroplastic changes occur in children and adults in response to various stimuli. Furthermore, training studies contribute to our understanding of white matter integrity as it relates to language ability.

White Matter Integrity Differences in Children Who Stutter

In relation to stuttering, DTI studies have examined white matter pathways relevant to speech, language, cognition, and motor control. Chang and colleagues (2008) were the first to examine the structural integrity of white matter in children with history of developmental stuttering and their age matched peers. This study reported that children who stutter exhibited significantly lower FA values in the left arcuate fasciculus underlying the laryngeal and tongue representation in the sensorimotor cortex, a finding that was previously reported in adults who stutter (Sommer et al., 2002). The finding of lower FA values along the left arcuate fasciculus in children who stutter has been replicated in a number of studies involving children and adults who stutter (Chang et al., 2008, 2015; Chow & Chang, 2017; K. E. Watkins et al., 2007; for review, see Chang, Garnett, et al., 2018). Lower FA has also been reported in the left SLF (Chang et al., 2015; Cykowski et al., 2010; K. E. Watkins et al., 2007).

More recently, a longitudinal study examined the developmental trajectories of white matter pathways to determine if children with persistent stuttering and children who recovered from stuttering differed from typically developing children (Chow & Chang, 2017). The results of this study indicated that, regardless of later persistence or recovery, children who stutter as a group exhibited decreased FA in the left arcuate fasciculus underlying the inferior frontal, inferior parietal, and posterior temporal areas, and the mid body of the corpus callosum (Chow & Chang, 2017). Namely, the two groups exhibited similar white matter deficits when compared to fluent peers. However, what differentiated the eventually persistent and recovered groups was the *growth rate* of FA in the same areas. That is, children with persistent stuttering exhibited a slower increase in FA growth in the left AF and corpus callosum, but normalized developmental trajectories in the same areas in children who recovered. Given the aforementioned studies, there is a general consensus that stuttering is associated with decreased white matter integrity along parts of the left arcuate fasciculus and superior longitudinal fasciculus in both children and adults who stutter (Chang, Garnett, et al., 2018; Neef et al., 2015; Smith & Weber, 2017). Collectively, the AF and SLF make up the dorsal language pathway, sometimes referred to as the dorsal auditory tract, and connect major perisylvian cortical structures involved in language processing and production (Hickok & Poeppel, 2004, 2007; Saur et al., 2008).

The Dual Stream Model

It has been proposed that speech-language processing occurs bilaterally within auditory regions (i.e., the superior temporal gyrus), and diverges into two broad streams (Hickok & Poeppel, 2000, 2004, 2007). The ventral stream supports speech comprehension bilaterally by mapping sound to meaning (Hickok & Poeppel, 2004; Saur et al., 2008). Together, the extreme capsule fiber system and the uncinate fasciculus make up the ventral language pathway. This ventral stream projects ventro-laterally and connects cortex in the superior temporal sulcus and the posterior inferior temporal lobe. It has been suggested that the ventral language pathway may correspond to the lemma level of representation proposed by Levelt (1989; 2001).

More germane to speech production is the dorsal language pathway, supports sensory-to-motor mapping (Hickok & Poeppel, 2004; Saur et al., 2008). This pathway consists of the arcuate fasciculus and the superior longitudinal fasciculus (Hickok & Poeppel, 2004). In adulthood, the dorsal language pathway contains two fiber bundles that are differentiated by their termination regions in the frontal cortex. One dorsal fiber bundle (D1) connects temporal regions with the premotor cortex while the other (D2) connects temporal regions to the IFG (Perani et al., 2011). The dorsal pathway connecting the temporal regions to the premotor cortex is present at birth (Bauer et al., 2013; Dubois et al., 2006, 2009, 2016; Perani et al., 2011). In contrast, the dorsal connection between the IFG and superior temporal gyrus is not myelinated at birth (Perani et al., 2011), or by 4 months of age (Leroy et al., 2011). This connection develops over the course of childhood from 3 to 10 years of age, as indicated by increased fractional anisotropy (Skeide et al., 2016; Skeide & Friederici, 2016). As the dorsal language pathway develops and shows increases in white matter integrity, that a leftward-asymmetry toward the language-dominant hemisphere exists during infancy (Dubois et al., 2009). Figure 2 below shows the dorsal and ventral language pathways.

The increased structural connectivity of the dorsal language pathway that develops across childhood suggests that changes also occur in functional connectivity during this time. As such, neuroimaging studies have examined functional connectivity in newborns to determine how specific regions of interest functionally connect to other regions of the brain. Perani and colleagues (2011) reported that adults exhibit functional connectivity between the left IFG (BA44) and the posterior superior temporal gyrus/superior temporal sulcus. In contrast, newborns exhibited a functional connection between the left IFG and right IFG (BA 44) while using what is called the same seed region (the left IFG). A seed region is a defined area of voxels that are used for data analysis (Huettel et al., 2009). The functional connectivity of the left posterior superior temporal gyrus also exhibited a functional connection to the right hemisphere homologue (Perani et al., 2011). These findings support the notion that increased structural integrity of white matter pathways in language regions might lead to increases in functional connectivity as well.

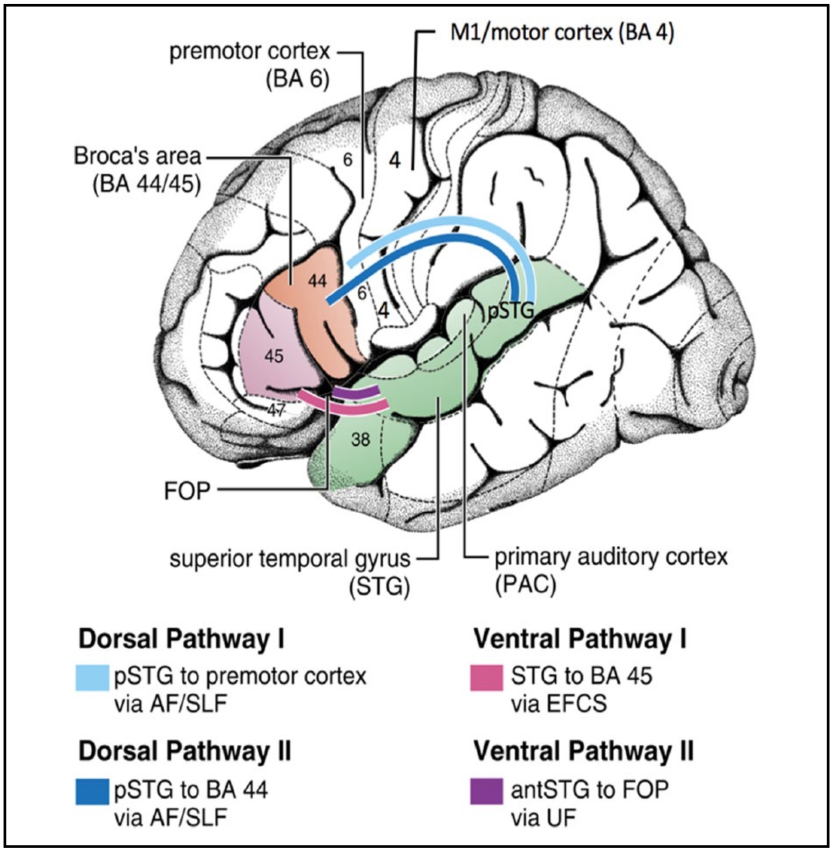
The dual stream model also suggests that the dorsal language pathway plays a role in speech acquisition. Hickok and Poeppel (2004, 2007) posited that because the dorsal language stream plays a role in auditory-motor integration, it is a critical component that helps a child learn to articulate speech sounds by providing a mechanism where: (1) sensory representations of speech can be stored, (2) articulatory attempts are compared against stored representations, and (3) the extent of mismatch revealed by this comparison can be used to correct/shape future articulation. The directions into velocity of articulators (DIVA) model has also proposed that speech acquisition occurs using both the auditory and motor systems (Guenther, 1994; Guenther

& Perkell, 2004; Guenther & Vladusich, 2012; Tourville & Guenther, 2011). The DIVA model posits that a tight mapping occurs between articulation and the acoustic outcome. As the children begin to acquire speech the mapping between these two systems become stronger and more efficient (Guenther, 1994, 1995; Guenther & Vladusich, 2012; Tourville & Guenther, 2011), which are likely supported by the dorsal language pathway.

Collectively, the dorsal language pathway has been suggested to play a critical role in sensory-to-motor mapping (Hickok & Poeppel, 2007; Saur et al., 2008). In addition, the dorsal pathway has also been reported to support syntax (Friederici, 2011; Friederici & Gierhan, 2013; Skeide & Friederici, 2016) and phonological awareness (Friederici & Gierhan, 2013; Hickok & Poeppel, 2007; Yeatman et al., 2011). It has further been suggested that complex production load may result in increased recruitment of dorsal language pathways during sentence generation (Ries et al., 2019). Evidence has also suggested that the left SLF and AF the play a role in combining phonemes into a sequence for words and sentences (Ries et al., 2019), and also support speech repetition (Hickok, 2012; Ueno et al., 2011). Adults with lesions along the dorsal language pathway are reported to exhibit phonological processing deficits and phonemic paraphasias following cerebrovascular accidents (Duffau et al., 2002, 2003, 2005). Taken together, there is strong evidence that points to the dorsal tract as playing a critical role in supporting speech and phonology. A question that arises is whether children with decreased white matter integrity in the dorsal tract may have subtle decreases in phonological developmental and ability.

Figure 2:

Dorsal & Ventral Language Pathways



Note. The dorsal and ventral language pathways interconnect major speech-language cortical areas. The ventral pathway (pink and purple) is comprised of the extreme fiber capsule system (EFCS) and uncinate fasciculus (UF), which map sound to meaning. The dorsal language pathway (blue and light blue) is made up of the arcuate fasciculus (AF), and the superior longitudinal fasciculus (SLF), which are responsible for mapping sound to articulation. From "The Brain Basis of Language Processing: From Structure to Function," by A. D. Friederici, 2011, *Physiological Reviews*, *91*(4), p. 1360 (https://doi.org/10.1152/physrev.00006.2011). Copyright 2011 by the American Physiological Society.

Phonological Development

Development of Phonological Skills

Phonological development is the process children use to acquire and use speech/sound patterns of a language. This process occurs quite rapidly, and requires children to learn the mapping between auditory speech inputs and the gestural output of speech production (Snowling & Hulme, 1994). At birth, infants have the unique ability to distinguish the phonemes used throughout the world; however, this unique ability is lost by one year of age (Hodson, 2007). As development occurs, frequently used sounds within the language environment become strengthened and stimulated, thereby resulting in the infant implicitly learning the rules that govern the sequencing of phonemes within a given language (Hodson, 2007). The ability to extract word boundaries from continuous speech, which occurs by approximately seven months of age (Jusczyk & Aslin, 1995) and serves a critical function during language acquisition according to some theorists (Hodson, 2007).

As the infant acquires the ability to distinguish word boundaries in continuous speech, phonological representations of words begin to form simultaneously (Goswami, 2000). These phonological representations are the mental representation of the various sounds and combinations of sounds, which join together to form words (Snowling & Hulme, 1994). Evidence has shown that as children begin to produce their first words, they initially organize speech gestures to approximate the syllable, but gradually organize their patterns of gestures in phonemic sized units as vocabulary increases (Nittrouer et al., 1989).

Phonological development and the underlying processes that contribute to its development, in fact, do not occur in a linear manner. Numerous theories attempt to provide an explanation for the acquisition and development of speech (e.g., Chomsky & Halle, 1968; Jakobson, 1968; Mowrer, 1960; Olmsted, 1971; Stampe, 1969; but see, Guenther, 1994; and Guenther & Vladusich, 2012). Many of these were developed based on the study of speech sound disorders. The acquisition and development of speech sounds and phonological skills is a complex motor and linguistic process beginning in infancy that continues throughout childhood. When a child fails to meet expected milestones, or produces speech with noticeable errors, it is necessary to investigate articulatory stimulability, while also conducting a comprehensive evaluation of phonology, as well as expressive and receptive language.

Assessment of Phonological Skills in Children

Many studies have investigated the phonological and articulatory abilities of children who stutter in comparison to children who do not stutter and those who have recovered (Anderson & Conture, 2000; Paden et al., 1999, 2002; Spencer & Weber-Fox, 2014; Yaruss & Conture, 1996). To examine phonological development, a number of studies have utilized standardized assessments such as the Goldman-Fristoe Test of Articulation, Second Edition (GFTA-2; Goldman & Fristoe, 2000), Khan-Lewis Phonological Analysis, Second Edition (KLPA; Khan et al., 2002), or Bankson-Bernthal Test of Phonology (BBTOP; Bankson & Bernthal, 1990), to probe a child's phonetic inventory at the word level. Although these assessments provide detail about a child's ability to produce each phoneme at initial, medial, and final position at word-level, they do little to explain how these skills generalize to conversational speech. Furthermore, many authors have suggested that children with phonological disorders may have little difficulty producing single-word responses but greater difficulty when producing these same words in connected speech (Glaspey & Stoel-Gammon, 2007; Ingram, 2002; Ingram & Ingram, 2001; Wolk & Meisler, 1998). For example, Morrison and Shriberg (1992) found that children produced a higher number of errors and types of errors during conversational speech than they did during standardized testing. With this drawback in mind, some researchers have utilized spontaneous speech samples to calculate the percentage of consonants correct (PCC; Shriberg & Kwiatkowski, 1982a, 1982b), or assessments such as the Assessment of Phonological Processes-Revised (APP-R; Hodson, 1986) to probe target words within the context of a structured speech sample to examine the correctness of individual phonemes. This increases the overall ecological validity of the assessment (Glaspey & Stoel-Gammon, 2007).

As children begin to acquire phonology of a specific language, they show little awareness of individual phonemes. Instead, they appear to acquire entire syllables and words as a single unit (Ferguson & Farwell, 1975; Menn, 1976). Ingram and Ingram (2001) introduced a whole-word approach to phonological analysis to account for whole-word complexity (i.e., phonological mean length of utterance; PMLU), in addition to the child's ability to approximate a target word during speech production (i.e., proportion of whole-word proximity; PWP). Examining larger units of speech instead of limiting analysis to only individual segments is necessary because, across development, a child's words become longer and more complex as vocabulary increases. Studies have shown that increased phonological complexity influences the articulatory accuracy of individual phonemes due to trade-off effects. These trade-off effects are defined as a child trading off complexity by simplifying or omitting consonants (Ferguson & Farwell, 1975; Lleó, 1996; Macken, 1978).

The use of whole-word measures to investigate a child's phonological abilities uniquely assesses phonological complexity. It also considers the variability of the child's production. Many standardized assessments only include a single opportunity for a child to produce a phoneme. If the production is incorrect, the child will receive a lower score. Alternatively, if the child accurately produces the target, it might be assumed that he/she has mastered the phoneme. Whole-word phonological measures consider a number of productions that occur across numerous contexts to determine overall accuracy.

Phonological analysis has been conducted using PMLU and PWP to investigate phonological acquisition of cross-linguistic/bilingual language acquisition, first words in English speakers, phonological acquisition in children with cochlear implants, and to study children with and without phonological impairments/speech-sound disorders (Arias & Lleó, 2014; Babatsouli et al., 2014; Ingram, 2002; Taelman et al., 2005). The use of percentage of consonants correct (PCC) allows researchers and clinicians to examine the accuracy of speech segments, specifically consonants, in a spontaneous speech sample, and is one of the most widely used measures. Flipsen and colleagues (2001), compared PMLU and PCC in children 2 to 5 years of age and found that PMLU was a better predictor of severity than PCC. More recently, Newbold and colleagues (2013) found that PWP was more sensitive than PCC in monitoring the articulatory abilities of children 4 to 6 years of age who had speech-sound disorders. Overall, whole-word measures such as PMLU and PWP offer a unique approach to investigating phonological development because of their applicability to spontaneous speech samples, which provide details about whole-word phonology, as opposed to traditional measures that investigate individual speech segments.

Phonological Differences in Children Who Stutter

A number of studies have investigated phonology as a potential contributing factor to stuttering, which is likely due to a rate of articulation/phonological deficits in children who stutter compared to that of the general population (Arndt & Healey, 2001; Blood et al., 2003; Louko et al., 1990; Wolk et al., 1993; Yaruss et al., 1998). Previous studies have estimated that approximately 30-40% of children who stutter present with a comorbid phonological disorder, which is significantly higher than that observed within the general population (Louko et al., 1990). While it is generally agreed that phonological impairments occur at a higher rate in children who stutter, the exact incidence of phonological disorders occurring in children who stutter has yet to be determined given differences in methodology (Yairi et al., 2005). Given the unique co-occurrence observed in a large proportion of children who stutter, researchers have investigated the potential relationship between stuttering and phonology/articulation skills.

Early investigations of phonological disorders in children who stutter eventually led to researchers examining whether phonological skills could serve as a predictor for recovery from stuttering. Paden and colleagues (1999) investigated phonological development to determine if phonological skills could be used near stuttering onset (within 6 months of onset) to predict stuttering persistence or recovery. The findings of the study demonstrated that children who persist in stuttering exhibit phonological performance scores *below* children who eventually recover and children who did not stutter, after weighting the Assessment of Phonological Processes-Revised (APP-R) by age (Paden et al., 1999). Utilizing a weighted calculation for age, the authors were able to more closely examine phonological development using the child's specific age in months. Paden and colleagues (1999) also reported that children with persistent stuttering produced a greater percentage of errors for basic patterns (e.g., nasals, glides, liquids, etc.), while also using cluster reduction to a greater extent than children who recovered from stuttering. This finding, which indicates that children who stutter have more difficulty acquiring consonant clusters, has also been supported by a number of other studies (Louko et al., 1990; Sambrookes, 1999; Wolk et al., 1993).

Phonological skills have been associated with later stuttering persistence or recovery, however, these differences become attenuated over time and eventually vanish (Paden et al., 2002). This pattern was reported by Paden and colleagues (2002) , who examined phonological development among 84 children who stutter (22 of which were children with persistent stuttering) by collecting data at three separate visits, which occurred roughly one-year apart, so the progression of phonological development could be examined. Findings of this study indicated that following the initial visit, children with persistent stuttering showed greater phonological development than children who recovered from stuttering. This suggests that phonological maturation may be protracted in these children (Paden et al., 1999). Finally, it is important to note that by the third visit, the two groups of children who stuttering were indistinguishable (Paden et al., 2002). This study provides evidence that children with persistent stuttering may develop phonology at a slower rate than children who do not stutter and children who recover from stuttering.

Although nonword repetition tasks are not traditionally used to investigate speech sound errors, these tasks have been utilized to examine the selection and preparation of sounds that form individual words. In addition, nonword repetition tasks have been used to investigate phonology because of the process required to successfully produce each word, which includes: processing speech stimuli, encoding phonological representations, storing phonological representations in phonological working memory, and speech motor planning and execution (Gathercole, 2006; Gathercole & Baddeley, 1990; Kent, 2000; Rispens & Baker, 2012). A number of studies have reported that children who stutter have lower nonword repetition abilities than children who do not stutter (Anderson et al., 2006; Anderson & Wagovich, 2010; Hakim & Bernstein Ratner, 2004). These tasks assess a person's ability to complete repetition of a whole word and provide an additional load on the phonological system. Still, they do not account for spontaneously produced speech, which includes conceptualization, preparation, encoding and execution to deliver a verbal utterance. The emphasis in nonword repetition tasks is primarily on phonological encoding. The reduced ability to complete nonword repetition tasks, which has been observed among children who stutter, has been linked to deficits in phonological encoding. These findings lend support for neurolinguistic theories of stuttering, which suggest that breakdowns and/or delays in phonological encoding may contribute to disfluencies in speech (Howell & Au-Yeung, 2002; Kolk & Postma, 1997; Perkins et al., 1991; Wingate, 1988). These articulatory abilities, as measured by nonword repetition tasks, have been useful in examining phonological processing.

Spencer and Weber-Fox (2014) retrospectively examined the articulation skills and nonword repetition abilities of children with persistent stuttering and those whose stuttering subsided. The results of this study were that children who persist in stuttering exhibited significantly weaker articulatory proficiency than children who recover, as measured by the Bankson-Bernthal Test of Phonology, Consonant Inventory subtest (BBTOP-CI; Bankson & Bernthal, 1990). The children who persisted in stuttering were found to perform less well than children who recover from stuttering on overall nonword repetition (Spencer & Weber-Fox, 2014). These findings are consistent with previous studies that report decreased nonword repetition abilities in children who stutter compared to children who do not stutter (Anderson et al., 2006; Anderson & Wagovich, 2010; Hakim & Bernstein Ratner, 2004). These breakdowns in speech repetition might suggest delays in phonological perception, encoding, and/or execution within children who stutter. While it is difficult to separate these processes, underlying neuroanatomical structure and function associated with linguistic function might provide additional information that helps to distinguish children who stutter from children who do not stutter.

Evidence suggests that phonological skills develop over a longer period in children who stutter. However, some authors posit that linguistic and/or phonological differences within these two groups are due to methodological shortcomings, which include: vast age ranges within a single study, the utilization of assessments used to detect disorders, or strict inclusion/exclusion criteria (Nippold, 1990, 2002, 2012, 2018). Reduced phonological differences between children who stutter and children who do not stutter may be subclinical, and too subtle to detect via traditional standardized measures. Unique tasks with the ability to speech planning and production must be employed, which afford the ability to examine the speech-motor planning process. With this in mind, a number of researchers have utilized electroencephalography (EEG) to record event related potentials (ERPs), which provide a direct measure of neural activity with high temporal resolution and are sensitive indicators of neural processes underlying language planning and phonology (Luck, 2005). ERPs measure populations of neurons firing in synchrony, and are sensitive indicators of language proficiency, regardless of an individual's ability to achieve scores within normal limits on standardized assessments. For example, a number of studies have reported that children who do and do not stutter perform similarly on behavioral tests and rhyming tasks, but the children who stutter exhibit atypical neural functions underlying phonological processing (Hampton Wray & Spray, 2020; Mohan & Weber, 2015). Collectively, these studies have suggested that a maturational lag might account for their findings. Alternatively, ERP differences may be indicative of differences in temporal activity due to anomalous structure and function of the brain.

Whole-Word Assessment of Phonological Skills in Children

As previously mentioned, PMLU and PWP serve as whole-word measures representing word length, complexity, and overall intelligibility. The PMLU measures the length of each word by assigning one point to each consonant and vowel that a child produces. The complexity of each word is further calculated by adding one additional point to each consonant that is produced correctly. Regardless of accuracy, each word the child attempts to produce can be examined to calculate a "target PMLU" (tPMLU), which provides information regarding the complexity of each word the child attempts. This tPMLU represents the maximum number score each whole word can receive, and can be used to calculate PWP, which directly compares the child's "actual PMLU" (aPMLU) and tPMLU (PWP = aPMLU/tPMLU). The PWP quantitatively describes not only how well a child can approximate each target word attempted; it also used to distinguish between two children that may have similar aPMLU, but differ in the complexity of target words (tPMLU). Overall, whole-word approaches to phonological analysis, such as PMLU and PWP have the potential to reveal phonological differences that may have otherwise been missed due to the focus on segments. These whole-word approaches have not been used previously to examine the abilities of children who stutter and children who do not stutter.

Statement of the Problem

Previous research has provided evidence that children who stutter exhibit decreased white matter integrity along the left dorsal language pathway relative to children who do not stutter. Subtle phonological differences have also been reported in children who stutter compared to children who do not stutter. On one hand, there is strong evidence supporting the role of the dorsal tracts in phonological processing and on the other hand, many previous studies reported subtle deficits in the same tracts in both adults and children who stutter. Thus there have been separate studies that examine dorsal tracts in relation to phonology, phonological differences in children who stutter, and white matter differences in children who stutter, but no studies to date have examined these aspects together to investigate how dorsal tract white matter integrity differs in children who stutter as a function of phonology. It is important to investigate whether dorsal tract white matter integrity associated with phonological skills differ in children who stutter, because the results from such an investigation are expected to lead to a clearer understanding of whether and how phonological development may be affected in stuttering, possibly reflected in relevant white matter tract development. Such findings would provide novel information about what regions of major white matter tracts might be relevant to phonological development in children who stutter, which may differ from that of children who do not stutter. Previous studies have provided evidence that phonological skills may differ in children who stutter using standardized assess, but whole-word phonological complexity and ability has yet to be examined in this population. The current study measures phonological production using whole words that were produced during spontaneous connected speech. In addition, the current study will examine how phonological production is associated with white matter integrity differences in children who stutter relative to children who do not stutter.

The Current Study

The current study examined the relationships between whole-word phonological skills of children who stutter and children who do not stutter in relation to white matter integrity along the bilateral dorsal language pathway. The aims of this study are two-fold: 1) to determine whether children who stutter, relative to children who do not stutter, exhibit poorer or delayed development in phonological skills as measured by PMLU and PWP, and 2) to examine whether children who stutter exhibit a potential relationship between anomalous white matter integrity measures (i.e., FA derived from diffusion tensor imaging (DTI) in the dorsal language pathway) associated with phonological skills using PWP. In line with these aims, the study will answer the following questions:

1. Do whole-word phonological skills in spontaneous speech differ between children who do and do not stutter as measured by PMLU (actual and target) and PWP?
2. Do children who stutter, or children who do not stutter, exhibit stronger associations between PWP and white matter integrity (i.e., FA) along the dorsal language pathway?

Based on previous studies, it is hypothesized that children who stutter, as a group, will perform less well than children who do not stutter on traditional measures of phonology (i.e., KLPA-2), which consider individual speech segments. Furthermore, it is hypothesized that children who do not stutter will exhibit greater phonological skills when examining whole-word measures of phonology. That is, children who do not stutter will produce words of greater phonological complexity (PMLU) and also approximate target words with greater accuracy (PWP) than children who stutter. In addition, it is hypothesized that children who stutter, relative to children who do not stutter, will exhibit a stronger association between PWP and white matter integrity along the right dorsal language pathway and a weaker association in the left hemisphere dorsal language pathway based on previous studies reporting decreased FA in the left hemisphere.

Results from the current study will, for the first time, determine if children who stutter and children who do not stutter differ on whole-word measures of phonology. This might serve as a measure to differentiate children who stutter from children who do not stutter, and also provide another perspective related to speech production and stuttering. In addition, the association between PWP and white matter integrity may provide additional evidence that the right hemisphere serves as a compensatory mechanism in people who stutter. Finally, results from this study might lend additional support to the idea that developmental stuttering is associated with white matter integrity differences along the dorsal language pathway, which might be associated by phonological development.

CHAPTER 2:

METHODS

Power Analysis

Planned tests of comparisons were between group t-tests for behavioral data, and linear regressions for white matter integrity and phonology using G*Power (Software 3.1; Faul et al., 2009). An a priori power analysis was conducted to estimate the sample size needed to achieve reliable data with an alpha = .05, effect size = 0.5, and a moderate beta of .70 to .80 to compute two-tailed independent sample t-tests. The results estimated that to detect significant between group differences for the phonological measures, a sample size of 51 to 64 children per group was needed. The second a priori power analysis was conducted based on data from Choo and colleagues (2016), who compared white matter integrity and behavioral proficiency of children who stutter (N= 34) and children who do not stutter (N= 35). The effect size in this study was 0.379, which is considered to be a small effect using the criteria established by Cohen (1988). With an alpha = .05 and power = .80, the estimated sample size needed to achieve an effect size of 0.4 is N= 22 in each group to detect significant between group differences in white matter integrity via FA measures.

Participants

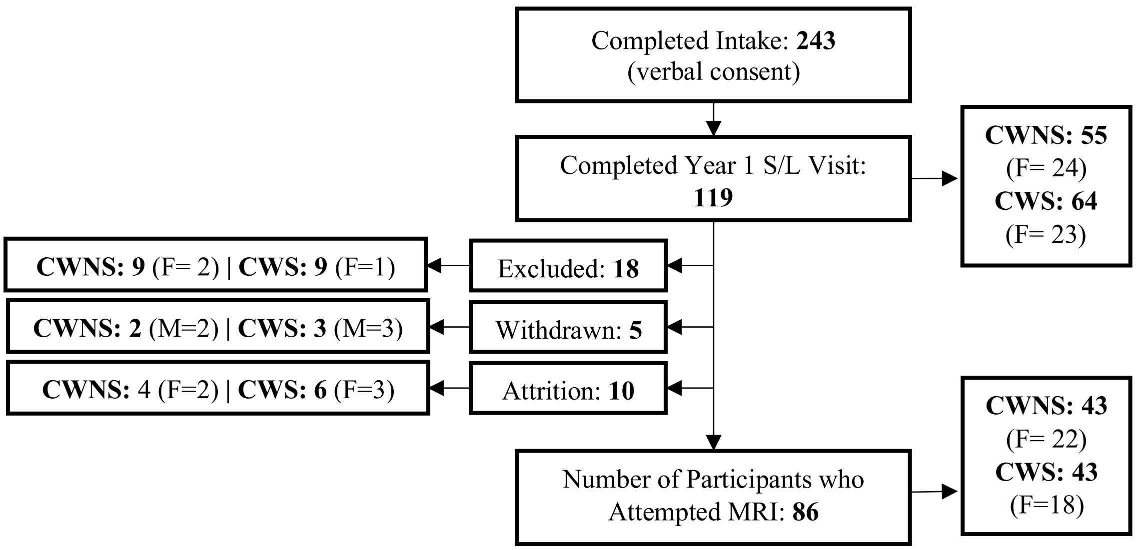
Children between the ages of 3 and 6 years of age with a history of stuttering were recruited as part on an on-going longitudinal neuroimaging study of developmental stuttering using community flyers, social media advertisements, and letters to speech-language pathologists, schools, parent groups, and daycares. An additional group of children who do not stutter were recruited for the longitudinal study to serve as a control group. A total of 243 children were screened as part of the longitudinal study. Prior to enrolling in the study, each family was asked to participate in a phone interview so eligibility could be determined (e.g., the child was a monolingual native North American English speaker, without a history of concussions or concomitant developmental disorders such as: dyslexia, attention deficit hyperactivity disorder, learning delay, Tourette's syndrome, or psychiatric conditions). A comprehensive list of questions asked during the phone screening can be found in Appendix 1.

Upon confirming that the child met basic inclusion criteria, the family was invited to participate in two study in two study visits: 1) a behavioral session that encompassed a battery of testing and speech sample collection, parent interviews, and 2) a MRI scanning session (more details about both sessions outlined below). Consent forms were signed by one or both parents for a total of 119 children (64 children who stutter and 55 children who do not stutter), and completed behavioral testing. Of the children who completed behavioral testing, 86 participated in the MRI scanning session (43 children who stutter and 43 children who do not stutter). Figure 3 shows details on how a child was determined to be eligible for the longitudinal study (also referred to as "the main study").

The 86 children who completed behavioral testing and participated in MRI scanning were eligible to participate in the current study if they met the following inclusion criteria: (a) standardized language scores exceeding -2 standard deviations of the mean and no history of articulation or phonological disorder; (b) a score greater than -1 standard deviation of the mean on full-scale IQ measures, (c) determined to be children who stutter (criteria outlined below) or children who do not stutter; (d) completed at least one successful DTI scan (i.e., no more than 11 volumes with greater than 1 mm of movement or other artifact); (e) within one year of behavioral testing at 3 to 6 years of age (36-83 months); and, (f) no siblings who stutter (this was only required for the group of children who do not stutter). To be classified as a child who stutters in the current study, three criteria were considered: (a) the child produced more than 3% SLD during a conversation sample with a clinician, (b) achieved a score of 10 or greater (very mild or greater) on the Stuttering Severity Instrument (Riley, 2009), (c) expressed parent concern and confirmation by a speech-language pathologist with many years' experience with stuttering. Of the remaining 86 participants, a total of 31 children failed to meet the established inclusion criteria (Figure 3). Thus, a total of 55 children participated in the study (children who stutter = 24, children who do not stutter = 31). Additional demographic data are provided below. Figure 4 provides detailed information on in/exclusion factors that influenced the final numbers of children included in the current study.

Figure 3:

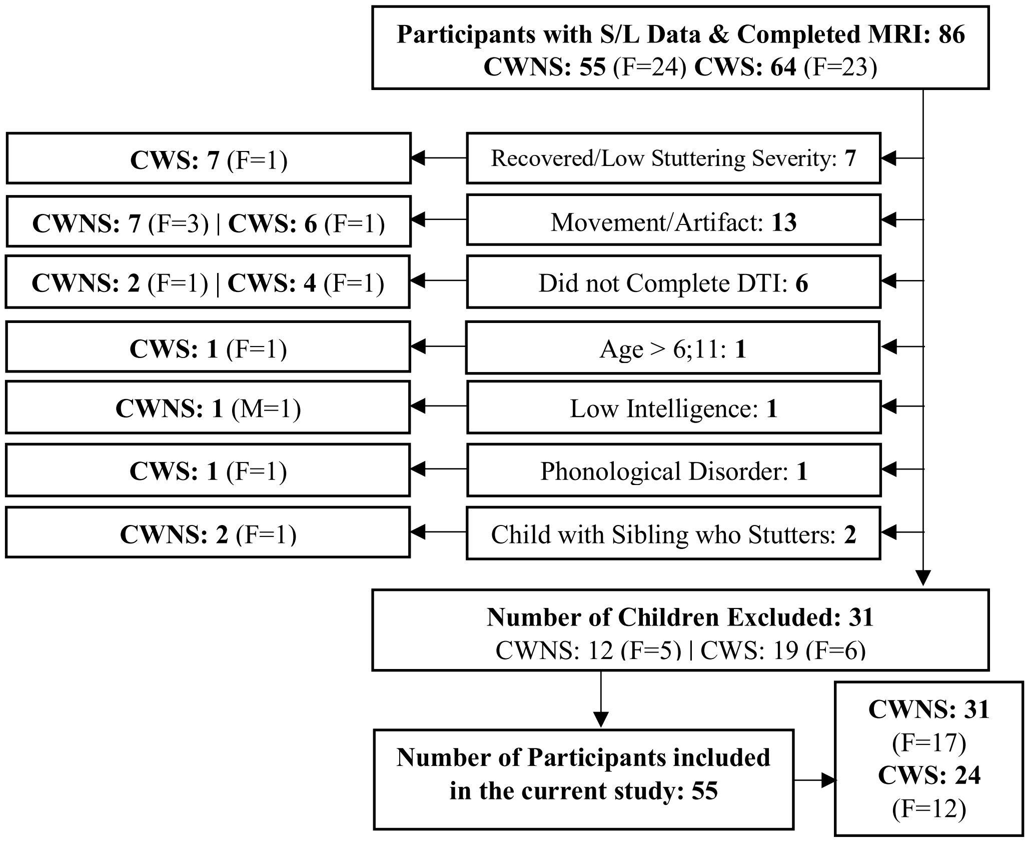
Detailed Inclusion Determination Tree for Longitudinal Study Participants



Note. A total of 243 participants completed a telephone screening. Of those participants, 119 children completed behavioral testing. Following the behavioral assessment, 18 children were excluded due to: low language scores (N = 3), family history of stuttering (N = 1), behavioral issues and/or poor tolerance of the mock MRI (N = 11), and, specific to children who stutter, low number of stuttering-like disfluencies (N = 3). Children who were considered to withdraw from the study if the study was notified that the participant no longer wanted to be enrolled in the study (N = 5), while attrition was attributed to participants who did not arrive to their scheduled MRI and/or failed to return calls and/or emails to the research study. Abbreviations: CWNS = children who do not stutter; CWS = children who stutter; F = females; S/L = speech/language.

Figure 4:

Participants for the Current Study



Note. Figure 4 displays the number of children who do not stutter (CWNS) and children who stutter (CWS) who completed the S/L session and the MRI scanning session. There were a total of 86 participants eligible for the current study before the exclusion criteria were applied. A total of 31 participants were excluded due to: recovery status/low SSI (N=7), movement/artifact in their MRI (N=13), failure to complete DTI scanning (N=6), chronological age was greater than 7 years (N=1), low intelligence scores (N=1), a phonological disorder (N=1), and/or the case history questionnaire revealed that a biological sibling was previously a person who stutters (N=2). A total of 55 participants remained eligible for the current study. Abbreviations: F = females; S/L = speech/language.

Procedures

During the first visit, the goals and procedures of the study were verbally explained to the parent and child by a certified speech-language pathologist. The parent signed the informed consent and child provided verbal assent that was approved by the Institutional Review Board at Michigan State University (IRB #09-810 LEGACY). The Four Factor Index of Social Status was used to collect data regarding each parent's level of education and occupation to determine socioeconomic status (Hollingshead, 1975). Each parent also completed the Children's Behavior Questionnaire (CBQ; Rothbart, 2000), in addition to completing a comprehensive case history form to gather information about the child's general health/development, language, musical training, and stuttering history (all questions can be found in Appendix 2). Description of all tests and questionnaires administered are summarized in Table 1. Prior to conducting statistical analyses, normality was assessed via the Shapiro-Wilk test (see Table 2). For each behavioral assessment that was normally distributed, independent t-tests were conducted; however, when the data exhibited a non-normal distribution, statistical analyses were conducted using the Mann-Whitney U-test.

All participants completed an oral-motor screening, along with an audiometric hearing screening to ensure typical hearing at 25 decibels (frequencies tested: 500 Hz, 1000 Hz, 2000 Hz, and 4000 Hz). The children were also tested on a comprehensive battery of standardized speech, language, and cognitive tests. The following questionnaires and assessments were used: the Edinburgh Handedness Inventory (Oldfield, 1971); the Purdue Pegboard Test (Tiffin, 1968); the Wechsler Preschool and Primary Scale of Intelligence (WPPSI-III; Wechsler, 2003); the Goldman-Fristoe Test of Articulation (GFTA-2; Goldman & Fristoe, 2000); the Communication

Attitude Scale for Preschool and Kindergarten Children Who Stutter (KiddyCAT; Vanryckeghem & Brutten, 2007); the Test of Childhood Stuttering (TOCS; Gillam et al., 2009); the Stuttering Severity Instrument (SSI-4; Riley, 2009); and the Clinical Evaluations of Language Fundamentals (CELF Preschool-2, Wiig et al., 2004, CELF-5, 2013). The TOCS was not utilized as part of the stuttering diagnostic criteria. Most participants were strongly right-handed (children who stutter = 21, children who do not stutter = 24); however, one child was left-handed, and nine children showed no-preference for handedness (children who stutter = 3, children who do not stutter = 6). Description of the participants is summarized in Table 3.

Table 1:

Protocols Utilized in the Longitudinal Study of Developmental Stuttering

Questionnaires & Assessments	Citation	Purpose
Parent		
Four Factor Index of Social Status	(Hollingshead, 1975)	Indexes maternal education for the present study, and provides information on socioeconomic status for the main study.
Children's Behavior Questionnaire (CBQ)	(Rothbart, 2000)	Provides a comprehensive assessment of reactive and self-regulative temperament in children, and is used in the main study.
Child		
Edinburgh Handedness Inventory (EHI)	(Oldfield, 1971)	This assessment is utilized in the longitudinal study to determine whether a child is right or left handed.
Purdue Pegboard Test (PPT)	(Tiffin, 1968)	The PPT is used in the main study to examine manual dexterity and bimanual coordination.
Wechsler Preschool and Primary Scale of Intelligence (WPPSI-III)	(Wechsler, 2003)	Used in year 1 of the main study to determine eligibility; measures intellectual abilities of children (2:6-7:3) by providing visual, performance, and full scale intelligence.
Goldman Fristoe Test of Articulation (GFTA-2)	(Goldman & Fristoe, 2000)	Measures the articulatory abilities of children within the main study. The current study calculated KLPA-2 scores using the GFTA-2.
Communication Attitude Scale for Pre-K & Kindergarten Children Who Stutter (KiddyCAT)	(Vanryckeghem & Brutten, 2007)	This assessment is used to measure awareness and/or speech-associated attitude of 3 to 6-year-old children who stutter using a 12-item questionnaire.
Test of Childhood Stuttering (TOCS)	(Gillam et al., 2009)	The TOCS identifies children who stutter and characterizes stuttering severity using four speech fluency tasks.
Stuttering Severity Instrument (SSI-4)	(Riley, 2009)	The SSI-4 is a norm-referenced assessment that measures overall stuttering severity using: frequency of stuttering, duration of stuttering, physical concomitants, and overall speech naturalness.
Clinical Evaluation of Language Fundamentals (CELF-P:2[a] & CELF-5[b])	(Wiig et al., 2004, 2013)	The CELF measures overall expressive/receptive language ability. If a child is >2 standard deviations outside of the mean, (s)he is excluded. The following subtests were used to calculate a Core Language Score.
Sentence Structure [a]		The child views four pictures, and is read a sentence, and then asked to identify the picture that best represents the sentence.
Word Structure [a, b]		This subtest examines a child's ability to apply morphological rules to identify inflections, derivations, and comparisons, while also using appropriate pronouns.
Expressive Vocabulary [a]		The child views pictures and is asked to verbally label each item. This subtest specifically investigates the child's lexicon.
Sentence Comprehension [b]		This subtest evaluates a child's ability to interpret sentences of increasing length and complexity via picture selection.
Formulated Sentences [b]		This subtest examines the child's ability to formulate complete, semantically and syntactically correct sentences of increasing length and complexity.
Recalling Sentences [b]		The child listens to spoken sentences of increasing length and complexity. The child must repeat each sentence without changing the semantics, morphology, or syntax.

Note. This table reports each assessment that each child completed as part of their comprehensive assessment.

KLPA-2 = Kahn-Lewis Phonological Analysis, second edition.

Table 2:

Shapiro-Wilk Test of Normality by Group & Total Sample

Variables	Children Who Stutter		Children Who Do Not Stutter		Total	
	Statistic	Sig	Statistic	Sig	Statistic	Sig
Age (behavioral session)	0.941	0.171	0.926	0.034	0.938	0.007
Age (MRI session)	0.948	0.242	0.918	0.022	0.940	0.008
Maternal education [a]	0.802	0.000	0.786	0.000	0.807	0.000
Full scale IQ	0.963	0.512	0.962	0.335	0.971	0.198
Performance IQ	0.924	0.073	0.976	0.707	0.966	0.127
Verbal IQ	0.971	0.684	0.965	0.384	0.982	0.555
Handedness	0.851	0.002	0.818	0.000	0.832	0.000
Core Language Score	0.964	0.525	0.970	0.525	0.981	0.511
GFTA-2	0.940	0.166	0.831	0.000	0.835	0.000
MLU (words)	0.933	0.113	0.956	0.231	0.957	0.049
MLU (morphemes)	0.934	0.123	0.957	0.242	0.956	0.041
NDW	0.896	0.018	0.970	0.528	0.968	0.146
VOCD	0.885	0.010	0.959	0.268	0.961	0.070
Phonological Variables						
KLPA-2	0.832	0.001	0.763	0.000	0.792	0.000
Target PMLU	0.908	0.031	0.948	0.139	0.945	0.014
Actual PMLU	0.958	0.397	0.976	0.700	0.972	0.217
PWP	0.953	0.311	0.936	0.064	0.946	0.016

[a] One child in the control group did not have maternal education reported, so the group mean was entered for this score.

Note. The above table displays results of the Shapiro-Wilk test for each group and the total sample. Abbreviations: Core Language Score is from the Clinical Evaluation of Language Fundamentals; GFTA-2 = Goldman-Fristoe Test of Articulation, second edition; ; SSI-4 = Stuttering Severity Index, fourth edition; KLPA-2 = Kahn-Lewis Phonological Analysis, second edition; PMLU = phonological mean length of utterance; PWP = proportion of whole-word proximity.

Table 3:

Participant Demographic Information & Behavioral Test Results

Variables	Children Who Stutter		Children Who Do Not Stutter		Test statistic	*p*	Cohen's *d*
	M	*SD*	*M*	*SD*			
Age (behavioral session)	4.96	1.21	4.91	1.13	356.5	*0.79*	0.042
Age (MRI session)	5.08	1.13	5.07	1.10	363.0	*0.88*	0.009
Maternal education	6.12	0.70	6.07	1.00	364.0	*0.88*	0.058
Full Scale IQ	110.63	11.66	110.74	9.72	0.041	*0.97*	0.010
Picture IQ	106.46	14.14	109.39	14.55	0.749	*0.46*	0.204
Verbal IQ	113.42	10.66	112.42	12.81	-0.308	*0.76*	0.085
Handedness	71.25	31.80	64.48	39.35	337.5	*0.55*	0.189
Core Language Score	110.88	14.32	109.90	12.29	-0.271	*0.79*	0.073
GFTA-2	106.67	7.56	100.00	14.55	298.5	*0.21*	0.575
MLU (morphemes)	6.37	1.87	5.87	1.62	309.0	*0.29*	0.286
NDW	52.79	7.73	49.90	8.14	281.0	*0.12*	0.364
VOCD	53.56	12.58	48.78	8.74	245.0	*0.03*	0.441
%SLD Clinician	7.34	4.47	1.24	0.80	23.0	*<0.01*	1.900
%OD Clinician	5.85	2.80	3.85	2.14	205.0	*0.01*	0.802
%SLD Parent	6.34	4.82	1.07	0.68	53.0	*<0.01*	1.531
%OD Parent	5.65	3.46	3.69	1.91	247.0	*0.03*	0.701
KiddyCAT	2.20	2.26	-			-	
SSI-4	19.29	5.31	-			-	
TOCS	65.45	11.87	-			-	

Note. Core Language Score is from the Clinical Evaluation of Language Fundamentals; GFTA-2 = Goldman-Fristoe Test of Articulation, second edition; MLU = mean length of utterance; NDW = number of different words; VOCD = vocabulary diversity; %SLD = percent of stuttering like disfluencies; %OD = percent of other disfluencies; KiddyCAT = Communication Attitude Test for Preschool and Kindergarten Children Who Stutter; SSI-4 = Stuttering Severity Index, fourth edition; TOCS = Test of Childhood Stuttering.

Determination of Stuttering

Stuttering severity was assessed using spontaneous speech that was elicited through storytelling and conversation with both a parent and certified speech-language pathologist during free-play. Each speech sample consisted of approximately 700-1,000 syllables, and took place in a quiet room with child-friendly furnishings. The evaluation room was equipped with a high definition video camera and microphone, which allowed each session to be video recorded so off-line transcription and fluency analysis could take place. Each speech sample was transcribed using Computerized Language Analysis (CLAN; MacWhinney, 2000) so additional analyses of language could be conducted as part of the on-going longitudinal study. After each speech sample was transcribed into CLAN, a certified speech-language pathologist calculated stuttering severity. The children who were classified as children who stutter were reported to display stuttering characteristics (e.g., repetitions, blocks, etc.) by both a parent and a certified speech-language pathologist working on the research study. Furthermore, the children classified as children who stutter exhibited 3 or more stuttering-like disfluencies per 100 syllables during the spontaneous language sample with the clinician. These children also exhibited 3 or more stuttering-like disfluencies during a monologue, which was elicited during a storytelling task using a wordless storybook, “Frog, Where are You?” (Mayer, 1969). In addition, the Stuttering Severity Instrument (SSI-4; Riley, 2009) was used to examine the frequency and duration of disfluencies occurring in the speech sample, including any physical concomitants associated with perceived moments of stuttering. Each participant within the stuttering group achieved a score of 10 or greater on the SSI-4, which corresponds to a severity rating of “very mild.” To confirm reliability of the SSI-4 scores, an intraclass correlation coefficient (ICC) was calculated based on two independent judges using a random subset (~44%) of children's speech samples. The ICC calculated based on the two judges' ratings was 0.98, indicating high reliability.

The average age of stuttering onset was 2.45 years (SD = 0.69; range 1.5 – 4.0 years), which is consistent with studies reporting the typical age of onset (Yairi & Ambrose, 1992, 2013). Information regarding time of onset was not available for two children (F=1, M=1) because the parent could not remember the precise timing of onset. Furthermore, all but two parents provided ages based on six-month intervals (i.e., 2.5 years old). The average time since stuttering onset was 2.53 years (SD = 1.28; range 0.33-4.83 years). Children who stutter also completed the KiddyCAT, which assesses communication attitudes of children who stutter, and averaged a 2.20 (SD = 2.26). During the spontaneous speech sample with a clinician, children who stutter exhibited an average of 7.34% SLDs per words (SD = 4.47), and an average of 5.85% ODs per words (SD = 2.80). The stuttering severity among the participants ranged from 'very mild' to 'severe,'; the children who stutter had an average SSI score of 19.29 (SD = 5.31; range 10–28) corresponding to 'moderate' stuttering severity. Appendix 3 shows the number of children in each severity group.

In addition to the speech-language and cognitive tests, all children participated in a mock MRI scanner training to familiarize them with the sights and sounds of the MRI environment. To increase the likelihood of acquiring quality MRI images, we utilized the "submarine protocol" (Theys et al., 2014) and also had each child view a short, interactive video that emphasized the importance of lying still while being in the MRI scanner. This video also provided an opportunity for children to discriminate between clear and blurry images of the brain that depended on the extent of movement during scanning. During this visit, a trained research assistant used a five-point scale (1= demonstrates fear; 2= cautious/reserved; 3= neutral (no adverse or positive reaction); 4= calm; 5= eager and excited) to rate the child's comfort on the following tasks: entering mock MRI scanner room; sitting on scanner bed; laying down on scanner bed; placing head within the head coil; reaction to sounds; tolerates in/out movement while going into the bore; laying still within the bore; and wearing ear plugs. These ratings helped determine a child's readiness to participate in the MRI scanning session. The mock scanner procedures were repeated as needed for some children.

The Michigan State University Institutional Review Board approved of all procedures used in this study. Children were paid $15.00 per hour for behavioral testing, and $25.00 for MRI testing. Each parent received a single $15.00 payment for his/her time and was also given additional payment if the family traveled greater than 60 miles one-way ($0.50/mile). In addition, each child was given a small prize for participating, and also received a printed image of his/her brain.

Phonological Assessment in the Current Study

For the purposes of this study, the 53 target words elicited from the GFTA-2 were used to compare the phonological processes of the children who do and do not stutter via the Khan-Lewis Phonological Analysis (KLPA-2; Khan & Lewis, 2002). The KLPA-2 provides a phonemic inventory, along with a phonological process summary, and allows clinicians and researchers to determine whether a phonological disorder exists. Because the KLPA-2 utilizes stimuli from the GFTA-2, individual phonemes can be examined in a number of phonetic contexts; however, the same child may not produce the same word accurate within the context of connected speech. Therefore, connected speech may be more representative of a child's overall production skills than traditional assessments such as the KLPA-2 (Morrison & Shriberg, 1992). After the GFTA-2 was administered, the scores were used to score the KLPA-2 to determine whether the phonological abilities of children who stutter and children who do not stutter were similar.

Speech Sample Transcription & Phonological Analysis

Each speech sample with the clinician was transcribed by a trained research assistant, and then coded by a certified speech-language pathologist or doctoral student proficient in language analysis to assure reliable parsing of utterance boundaries. An utterance was defined as any verbal utterance bounded by grammatical closure, terminal intonation contour, or a pause of more than 2 seconds. If discrepancies in coding existed, the transcribers reviewed the sample until agreement was reached by consensus. Transcripts were audited to ensure that any misspellings within the transcription were rectified so measures of vocabulary diversity were not impacted. All speech samples were transcribed using Codes for the Human Analysis of Transcripts (CHAT) so the mean length of utterance (MLU), number of different words (NDW), and vocabulary diversity (VOCD) could be analyzed using CLAN (MacWhinney, 2000).

Prior to conducting phonological analyses, each CHAT transcription was converted to an XML (extensible markup language) file using Chatter (version 1.0-alpha-154), which is one of the software utilities offered by CLAN (MacWhinney, 2000). After CHAT files were converted to XML files, phonological analysis was conducted using Phon version 3.0.6-beta.4 (Hedlund & Rose, 2019). Phon is a free and open-source software designed to facilitate the study of child phonology (Rose et al., 2006; Rose & MacWhinney, 2014b, 2014a), and can be used to calculate both the phonological mean length of utterance (PMLU), and the proportion of whole-word proximity (PWP). Each participant's speech sample file was opened in Phon so that phonological analysis could take place. Upon opening the speech sample file, the video recording of the speech sample was linked to the file so each word could be transcribed using the International Phonemic Alphabet (IPA). On average, 152 utterances were phonetically transcribed from each speech sample, with each sample requiring approximately 2.5-3 hours (with a minimum of 2 hours) to complete (137.5-165.0 hours in total). The graphical user interface of Phon displays the orthographical transcription of the speech sample in the "Orthography Tier" with "IPA Target" and "IPA Actual" appearing below. After each transcription was scanned to ensure proper conversion, the "IPA Lookup" function was used to automatically enter the IPA transcription into the "IPA Target" and "IPA Actual" tiers (see Figure 5). Once the IPA was automatically entered for each word within the speech sample, each word/utterance was reviewed within the speech sample to make changes within the "IPA Actual" tier, which reflect the child's actual

Figure 5:

Phon Transcription Window

Orthography	I	really	like	this	blue	color .
IPA Target	ˈaɪ	ˈɹiːliː	ˈlaɪk	ðəs	ˈbluː	ˈkɑləɹ
IPA Actual	ˈaɪ	ˈɹiːliː	ˈlaɪk	dəs	ˈbluː	ˈtɑlə

Note. Figure 5 displays the Phon transcription window that is used to examine phonological skills. The 'Orthography' tier shows where the speech transcript is typed. The 'IPA Target' and 'IPA Actual' tiers indicate where transcription in IPA takes place.

production. If a child's production of a target was incorrect, alterations were made on the "IPA Actual" tier. In some cases, the transcription appearing on the "IPA Target" tier was changed due to a difference between the child's dialect and the software default. After each child's production was entered into the "IPA Actual" tier, Phon automatically performed pairwise, phoneme-by-phoneme alignments between the "IPA Target" tier and the "IPA Actual" tier to determine the overall accuracy of each word produced. Ingram (2002) suggested that the calculation of PMLU and PWP should have high reliability; however, evidence is currently lacking. A second rater, who was also a certified speech-language pathologist, reviewed 16 transcriptions (29%) to ensure accurate IPA transcription. Results of an intra-class correlation coefficient revealed a high degree of reliability between IPA transcriptions. The average ICC was 1.00 with a 95% confidence interval from 1.00 to 1.00 ($F(15, 15) = 15667.35$, $p < 0.001$).

Calculation of PMLU

To calculate PMLU, one (1) point is assigned to each consonant and vowel produced within a word, and adds one (1) extra point for each consonant that is produced correctly relative to the target production. This calculation is completed on both the "IPA Target" tier, and the "IPA Actual" tiers to generate target PMLU (tPMLU) and actual PMLU (aPMLU), respectively. Once these calculations were completed for each word, the results were exported to a comma-separated values file for further analysis. A single output file was generated for each of the 55 participants in the study, and PMLU analysis was conducted using the rules established by Ingram (2002), which are outlined in Table 4. After restricting the number of words examined, aPMLU was calculated by summing together the child's PMLU of each word, and dividing it by the total number of words remaining in the corpus. The same procedure was conducted to calculate the child's tPMLU. Finally, PWP was calculated by dividing aPMLU by the tPMLU. The following examples demonstrate how PMLU and PWP are calculated:

PMLU = (Consonants + Vowels produced) + (Consonants Produced Correctly)
Example: /grin/ ("green") = 4 + 3 = 7 PMLU
/gwin/ ("gween") = 4 + 2 = 6 PMLU
/gin/ ("geen") = 3 + 2 = 5 PMLU

$$PWP = \frac{Child\ PMLU}{Target\ PMLU} \qquad \frac{/gin/\ 5}{/grin/\ 7} = 0.71$$

As described in Chapter 2, PMLU and PWP are measures used to estimate phonological ability at the whole-word level. More specifically, PMLU can be used to identify phonological stage of acquisition and evaluate the complexity of words produced by a child, while PWP quantifies how well a child approximates a target word, by comparing the aPMLU to the tPMLU (Ingram, 2002). The current study examines PMLU and PWP using connected speech from a language sample to determine whether phonological complexity and accuracy differ between children who do and do not stutter. To ensure IPA transcriptions were accurate, a second speech-language pathologist independently completed IPA transcriptions for 16 participants (8 children who stutter (F=4, M=4), and 8 children who do not stutter (F=4, M=4)). A high degree of reliability was found between the two raters. The average measure ICC was 1.00 with a 95% confidence interval from 1.00 to 1.00 (F (15, 15) = 15667.35, $p < .001$).

Table 4:

Rules for Calculating Phonological Mean Length of Utterance

Rule	Description
1. Sample-Size:	Select at least 25 words, and preferably 50 words for analysis, depending on sample size. If the sample is larger than 50 words, select a selection of words that cover the entire sample, e.g. every other word in a sample of 100 words.
2. Lexical-Class:	Count words (e.g. common nouns, verbs, adjectives, prepositions and adverbs) that are used in normal conversation between adults. This excludes child words, e.g. mommy, daddy, tata, etc. Counting child words can inflate the PMLU if a child is a reduplicator. Appendix 1 gives a sample list of words that are excluded from the PMLU count.
3. Compound:	Do not count compounds as a single word unless they are spelled as a single word, e.g. 'cowboy' but not 'teddy bear', i.e. 'teddy bear' would be excluded from the count. This rule simplifies decisions about what constitutes a word in the child's sample.
4. Variability:	Only count a single production for each word. If more than one occurs, then count the most frequent one. If there is none, then count the last one produced. Counting variable productions may distort the count if there is a highly variable single word.
5. Production:	Count 1 point for each consonant and vowel that occurs in the child's production. Syllabic consonants receive one point, e.g. syllabic 'l', 'r', and 'n'. (Some transcriptions may show these as two segments, i.e. a schwa plus consonant, e.g. 'bottle' [badəl], but it should be counted as one consonantal segment.) Do not count more segments than are in the adult word. For example, a child who says 'foot' as [hwut] has two consonants counted, not three. Otherwise, children who add segments will get higher scores despite making errors.
6. Consonants Correct:	Assign 1 additional point for each correct consonant. Correctness in vowels is not counted since vowel transcriptions are typically of low reliability. Syllabic consonants receive an additional point in the same way as nonsyllabic consonants. A child who applies liquid simplification, for example, will get 1 point for producing a vowel, e.g. 'bottle' [bado], but 2 points if the syllabic consonant is correct.

Note. This table demonstrates the rules utilized to calculate phonological mean length of utterance, as described by Ingram (2002).

Syntactic Abilities & Lexical Diversity Examined with CLAN

Language proficiency and expressive vocabulary are correlated with phonological complexity (Ingram, 2002); therefore, possible differences in MLU, NDW, and VOCD were examined in the two groups. With regards to morphosyntax, MLU was used to quantify the length of utterances using morphemes, expressed as a ratio of total number of morphemes produced divided by the total number of utterances (Brown, 1973). MLU has been commonly used to measure morphological and syntactic development in children, and has also been used as an index of grammatical complexity (Brown, 1973). In addition to the examination of syntactic abilities, lexical diversity was assessed using NDW and VOCD.

Number of different words (NDW) is a CLAN-derived measure that reflects the total number of different words utilized within a speech sample, and has been shown to be correlated with expressive vocabulary as measured with standardized assessments (Ukrainetz & Blomquist, 2002). Furthermore, NDW has been used to distinguish typically developing children from children diagnosed with specific language impairment (Klee, 1992). To compute NDW, CLAN utilizes only the first 100 words because the length of the speech sample can impact the variable. In addition, NDW was found to vary depending on the part of the speech sample that is analyzed (Malvern et al., 2004).

To ensure expressive language was considered using multiple approaches, the current study also examined VOCD because of its unique ability to measure vocabulary diversity while also taking into account the length of each speech sample. To measure VOCD, an algorithm is used that samples random groups of words 100 times to create a curve of the type-token ratio (TTR; Templin, 1957) against tokens (McKee et al., 2000). Briefly, TTR represents the total number of unique words (types) divided by the total number of words (tokens) used in a speech sample (Templin, 1957). Next, CLAN generates a best fit between the curve and theoretical curves calculated by the model. The resulting value ranges from 10 to 100, and is correlated with standardized measures of expressive vocabulary in children who stutter and children who do not stutter (Silverman & Bernstein Ratner, 2002).

As mentioned earlier, the above variables were examined to ensure any differences in phonological complexity were not driven by syntax or expressive vocabulary in spontaneous speech. Research studies examining syntactic abilities in spontaneous speech have reported mixed findings (Bloodstein & Bernstein Ratner, 2008). Furthermore, evidence suggests that children who stutter exhibit lower expressive language skills on standardized assessments than children who do not stutter. Interestingly, research investigating vocabulary diversity among children who stutter and children who do not stutter has failed to find significant group differences while examining spontaneous speech using NDW and VOCD (Luckman et al., 2020). Syntax and vocabulary diversity are outside of the scope of the current study; however, these measures were examined due to their relation to phonological complexity.

MRI Data Acquisition

All MRI scans were acquired on a GE 3T Signa® HDx MR scanner (GE Healthcare) with an 8-channel head coil. During each session, a set of 180 T1-weighted 1-mm3 isotropic volumetric inversion recovery fast spoiled gradient-recalled sagittal images (about 5 minute scan time), with cerebrospinal fluid (CSF) suppressed, was obtained to cover the whole brain with the following parameters: TE = 3.8 ms, TR of acquisition = 8.6 ms, time of inversion (TI) = 831 ms, TR of inversion = 2332 ms, flip angle = 8°, FOV = 25.6 cm × 25.6 cm, matrix size = 256 × 256, slice thickness = 1 mm, and receiver bandwidth = ± 20.8 kHz, and parallel acceleration factor = 2.

After T1-weighted images were acquired, DTI data were acquired with a dual spin-echo echo-planar imaging sequence for 9 minutes and 36 seconds with the following parameters: 48 contiguous 2.4 mm axial slices in an interleaved order, field of view = 22 × 22 cm, matrix size = 128 × 128, number of excitations = 2, echo time = 84.8 ms, repetition time = 12.8 s, 40 diffusion-weighted volumes (one per gradient direction) with b = 1,000 s/mm2, one volume with b = 4 and parallel imaging acceleration factor = 2.

During scanning, the children lay still in the MRI while viewing a movie via MRI-compatible goggles and headphones. To reduce the noise of the scanner, each child wore earplugs. Head motion was minimized using cushions within the head coil, and a strap across the child's forehead. In addition, a trained research assistant sat inside the scanner room next to the child at all times to monitor the child's comfort and to ensure cooperation during scanning.

DTI Analysis

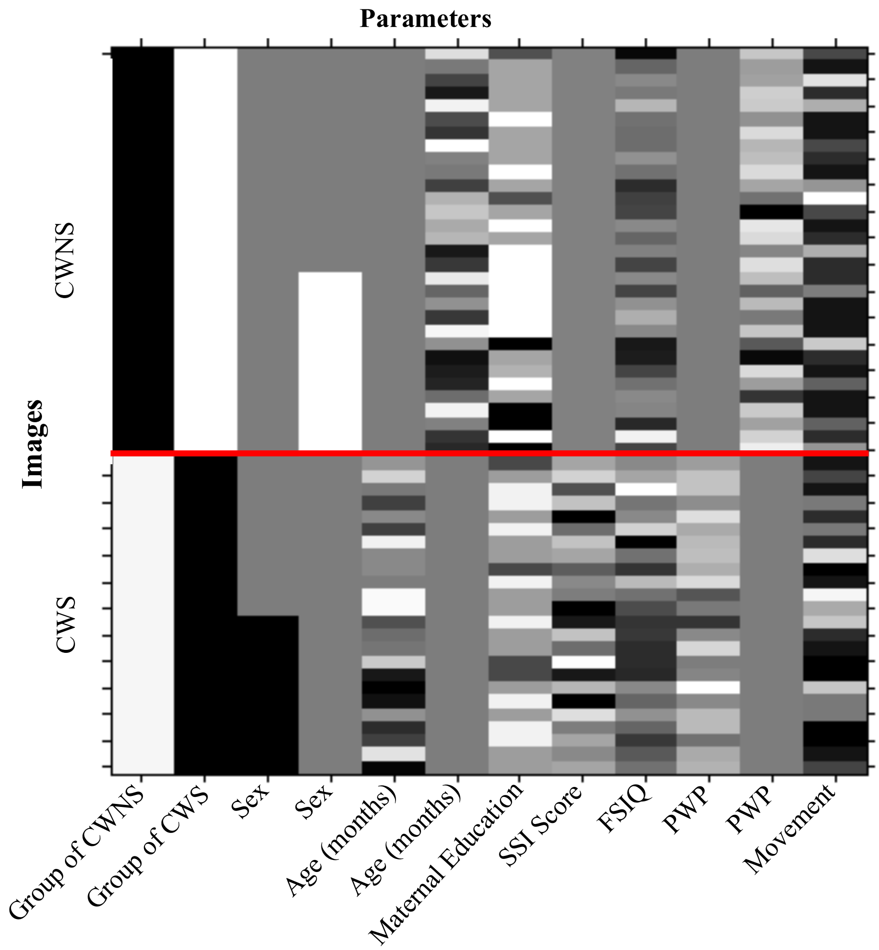
To ensure the DTI scans were of good quality, a technique similar to "Image Scrubbing" was used to remove movement artifacts (Power et al., 2012). For each DTI scan, slice-wise volume-to-volume head movements were estimated by coregistering each volume's slices to the slices corresponding to the first volume (B0). If a child's head movement exceeded more than 1 mm on more than 10 slices (20.5%) in a volume, it was considered susceptible to movement artifacts, and subsequently excluded from further analysis. To further ensure that the data were not contaminated by artifact, each scan was manually inspected to detect signal dropouts and image artifacts such as banding. After these initial quality control procedures were completed, the DTI scans were processed using MRtrix pipeline (www.mrtrix.org). Diffusion images were corrected for head motion, and eddy current artifacts using FSL eddy command (Andersson & Sotiropoulos, 2016). The eddy command was used because image distortions from eddy currents have the ability to blur boundaries between gray- and white-matter tissue. This blurring leads to a misregistration between individual diffusion-weighted (DW) images, and causes the miscalculation of the diffusion tensor. Diffusion tensors were estimated and individual fractional anisotropy (FA) maps calculated for group-level statistical analyses. To normalize FA maps into standard space, individual T1-weighted anatomical images were normalized using diffeomorphic image registration algorithm (DARTEL; Ashburner, 2007) implemented in SPM12 (http://www.fil.ion.ucl.ac.uk/spm/software/spm12/). After each individual T1 image was aligned with the DTI B0 image, the DARTEL algorithm segmented each T1 image into distinct gray and white matter tissue compartments using the unified segmentation algorithm (Ashburner & Friston, 2005). A customized template was generated using gray and white matter images from all participants. Next, DARTEL iteratively refined warping parameters for transforming individual subject space to the customized template, and then transformed the images from the customized template space to standard space (ICBM152). This two-step procedure was used in place of normalizing brain images directly to standard space because it has shown to be more robust in managing variations of brain sizes and morphometry (Yoon et al., 2009). Individual FA maps were transformed to standard space using the deformation fields generated from DARTEL, resampled to 2.5 mm isotropic voxel size, and spatially smoothed with a 3 mm full width at half maximum (FWHM) Gaussian kernel.

A voxel-based approach was used to analyze the normalized FA. Gray matter and gray/white matter boundaries were excluded when voxel-wise mean FA was calculated across subjects, and voxels with mean FA less than 0.25. A flexible factorial design was used in SPM 12 (https://www.fil.ion.ucl.ac.uk/spm/) to examine the effect of group and PWP on FA along white matter tracts while age, sex, SSI, maternal education, IQ, and head motion were entered as variables of no interest. A Pearson correlation was used to examine the correlation between PWP and age, which revealed a moderate correlation between PWP and age, r (53) = 0.55, $p < 0.01$. Therefore, PWP was adjusted to orthogonalize the two variables. Orthogonalization assigns the shared variability to one of the regressors, in this case PWP, and regresses one covariate on the other, and extracts the residual (Mumford et al., 2015). Figure 6 provides detail on the design matrix used in the analysis, and was generated in SPM 12.

Finally, independent t-tests were used to compare model estimates between groups. False positives due to multiple testing were controlled by a combination of p-value and a spatial extent (cluster size) threshold (k). The cluster size threshold was estimated using the recommended procedure of AFNI 3dClustSim (March 2016 version; http://afni.nimh.nih.gov/pub/dist/doc/program_help/3dClustSim.html). Unless otherwise specified, the thresholds were set at $p < 0.005$ and $k > 15$ voxels, corresponding to a corrected p-value of 0.05.

Figure 6:

Diffusion Tensor Imaging Regression Analysis in SPM12



Note. The above figure shows the design matrix that was generated in SPM12 and represents the parameters that were entered in the flexible factorial design analysis. The x-axis shows the parameters that were entered as variables of no-interest, and the y-axis displays each group of participants. CWNS = children who do not stutter; CWS = children who stutter; SSI Score = Stuttering Severity Index composite score; FSIQ = Full Scale Intelligence Quotient; PWP = Proportion of whole-word proximity; Movement = number of scans affected with head motion.

Statistical Analyses

Prior to conducting statistical analyses on the behavioral data, the normality of each standardized test was assessed via the Shapiro-Wilk test. We computed two-tailed *t*-tests for independent samples for each of the behavioral measures collected; however, the Mann-Whitney U-Test was used for each behavioral measure that was not normally distributed (Table 2). No tests for multiple corrections were needed because only composite scores were considered. Therefore, all results comparing demographic and behavioral characteristics were considered significant at $p < 0.05$, and can be found in Table 3.

CHAPTER 3:

RESULTS

The current study examined whole word phonological abilities in 3 to 6-year-old children who stutter and children who do not stutter. In addition, the behavioral measure of PWP was correlated with white matter integrity (FA) to determine whether children who stutter exhibit anomalous white matter integrity associated with phonological ability. It was hypothesized that traditional measures of phonology, such as the KLPA-2, which uses a segmental approach, along with whole-word measures of phonology, would differentiate the two groups. It was further hypothesized that the group of children who stutter would exhibit weaker correlations between PWP and white matter integrity along the left dorsal language pathway relative to children who stutter.

Behavioral Results

Standardized Assessments & Syntactic/Lexical Diversity Measures

The results of the present study failed to detect statistically significant group differences on the standardized assessments given (Table 4). In addition to standardized testing, spontaneous speech samples were analyzed using CLAN to determine whether the two groups had similar grammatical complexity (syntax) and expressive vocabulary (lexical diversity). Similar to the results of standardized testing, no significant group differences were found in MLU ($U = 295.0$, $p = .191$) and NDW ($U = 281.0$, $p = .122$), which index syntax and lexical diversity, respectively.

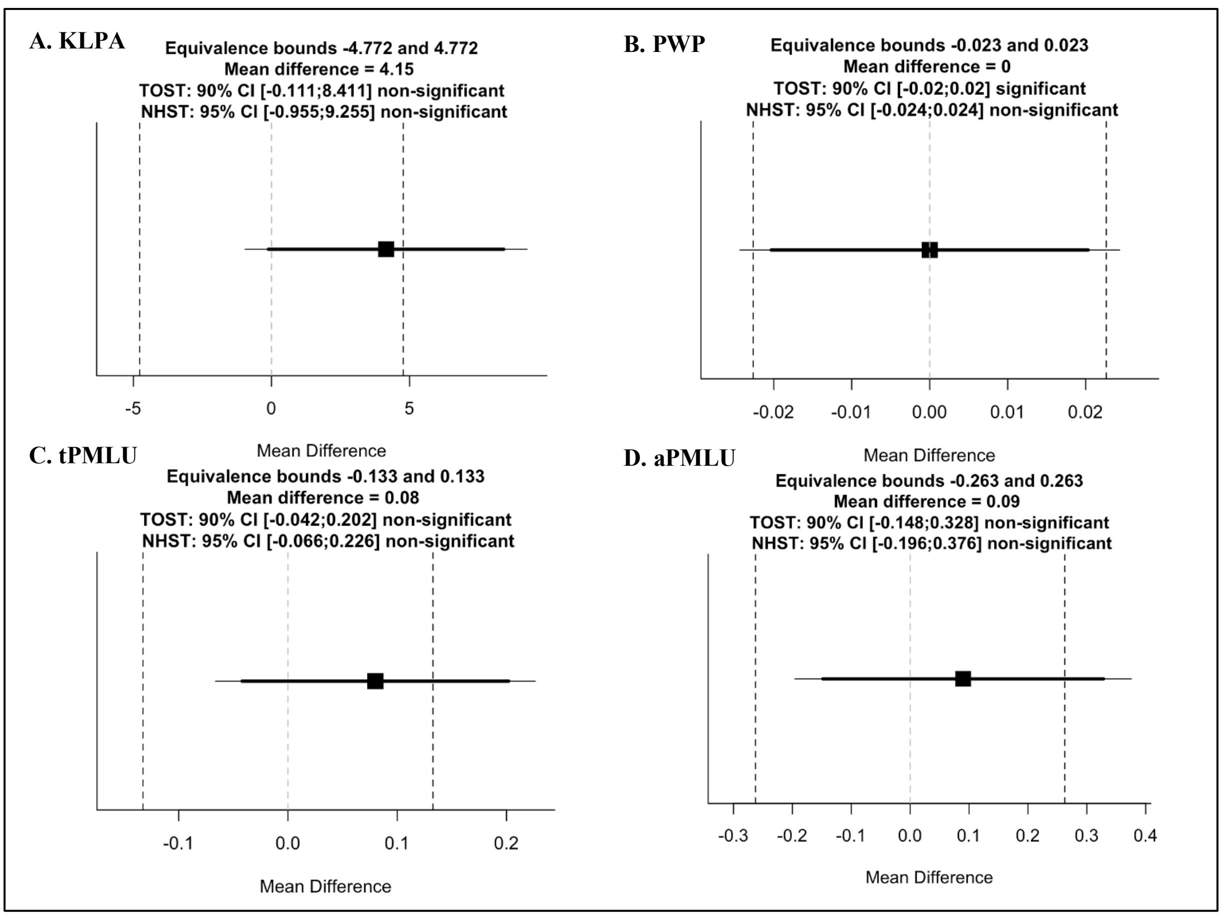
Interestingly, significant group differences were found for lexical diversity as indexed by VOCD (U = 245.0, $p = .031$), with children who stutter exhibiting greater lexical diversity skills than children who do not stutter.

Whole-Word Measures of Phonology do not Differentiate Children Who Stutter from Children Who Do Not Stutter

Although a number of studies have reported that phonological development differs between children who stutter and children who do not stutter (e.g., Paden & Yairi, 1996), there were no significant group differences found in the current study in phonological ability as indexed by the KLPA-2 (U = 275.0, $p = 0.10$). Furthermore, no differences in whole-word phonology were detected as measured by tPMLU (U = 313.0, $p = 0.32$), aPMLU ($t(53) = -0.62$, $p = 0.54$), or PWP (U = 365.0, $p = 0.91$), which is contrary to the original hypothesis. To better interpret these non-significant findings based on null-hypothesis significance testing (NHST), follow up equivalence tests (Lakens et al., 2018) were conducted to examine the extent of any potential meaningful differences in these measures between groups. Equivalence tests examine whether the presence of effects that are large enough to be considered meaningful can be rejected (Lakens et al., 2018). The equivalence test consists of two one-sided tests to determine whether the observed effect is statistically larger than the lower equivalence bound and statistically smaller than the upper equivalence bound (see figure 7 below). In equivalence tests, the null hypothesis is that there *is* a true effect that is considered more extreme than a smallest effect size of interest, which is defined by the upper and lower equivalence bounds (Lakens et al., 2018). The alternative hypothesis is that the effect falls within the equivalence range, which is a range of effect sizes deemed equivalent to the absence of an effect that is worthwhile to examine (e.g., ΔL = -5 to ΔU = 5, in the case of KLPA below, where Δ is a difference that can be defined by either standardized differences, or raw differences such as 1 scale point or 50 milliseconds). The use of equivalence testing provides a way to determine whether null findings are due to the absence of a meaningful effect or inconclusive results (Lakens et al., 2018). Figure 7 shows that while NHST showed insignificant results for each phonological measure, different conclusions can be made based on equivalence testing. In the case of PWP (Figure 7B), the equivalence

Figure 7:

Equivalence Testing for Phonological Variables



Note. Figure 7 shows the results of equivalence testing that was conducted for the phonological variables (A) KLPA, (B) PWP, (C) tPMLU, and (D) aPMLU.

testing was significant, indicating that the difference observed was too small to be considered meaningful. Namely, in this case the conclusion may be that there was an absence of a meaningful effect, thus providing more confidence that the two groups did not differ in any meaningful way. In the case of KLPA, aPMLU and tPMLU, the equivalence test results were all non-significant, indicating that although the NHST results showed nonsignificant results, the difference could be considered large enough to be considered meaningful based on equivalence testing. In these cases, more data would be needed to make a more conclusive determination of whether the two groups differ on these measures.

To examine whether group differences in VOCD affected comparisons for the phonological measures, one-way independent ANOVAs were conducted for each measure, with VOCD entered as a covariate. No differences between group differences were detected for tPMLU ($F(1, 52) = .043$, $p = .836$), aPMLU ($F(1, 52) = .038$, $p = .846$), or PWP ($F(1, 52) = .117$, $p = .734$). Table 5 contains behavioral results for the phonological variables.

The relationship between age (in months) and each whole-word phonology measure was examined using Pearson's correlation (used for aPMLU) or Spearman's correlation (used for tPMLU and PWP). Children who do not stutter exhibited a strong positive association between age and aPMLU (r=.719), and moderate associations between age and tPMLU (rho=.618) and PWP (rho=.617), which were all statistically significant ($p < .001$). Children who stutter exhibited a moderate association between age and aPMLU (r=.554), and age and PWP (rho=.579). The relationship between age and tPMLU was not statistically significant for the group of children who stutter. Scatterplots were created to further examine the relationship between age and whole-word measures of phonology (Figures 8-10).

Table 5:

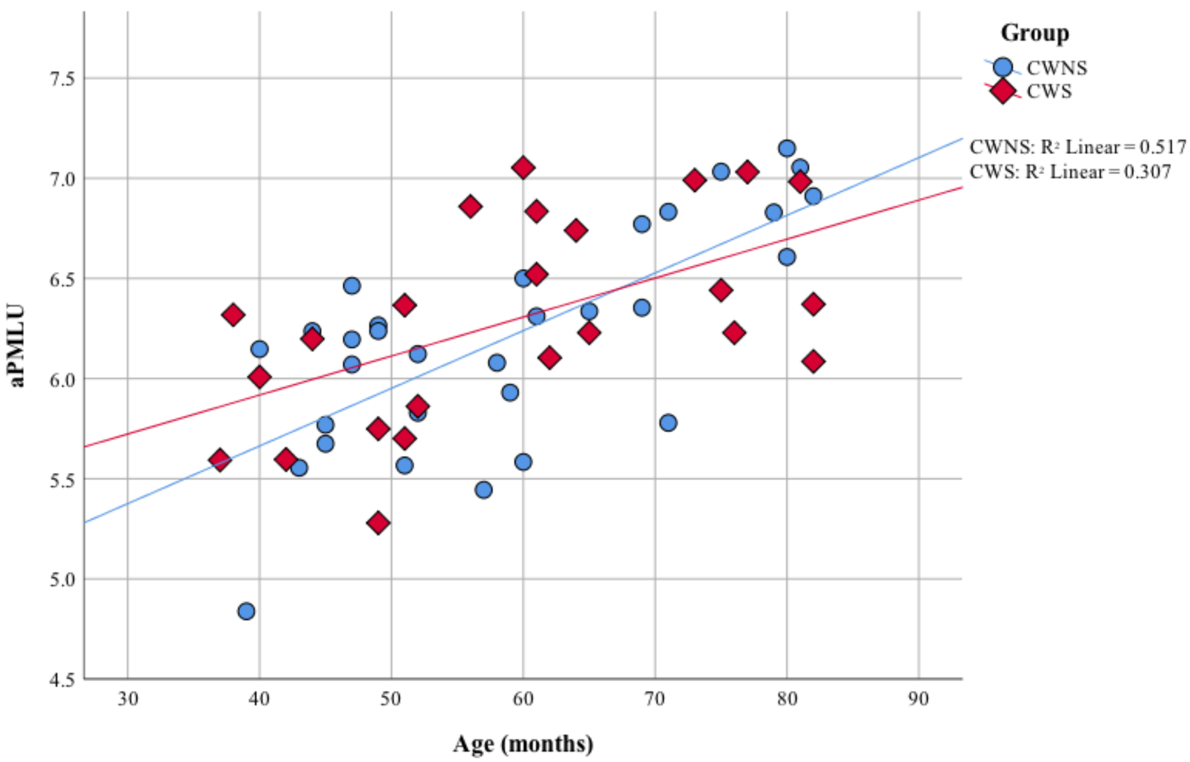
Phonological Measure Averages & Statistical Test Results

Variables	Children Who Stutter		Children Who Do Not Stutter		Test statistic	*p*	Cohen's *d*
	M	*SD*	*M*	*SD*			
KLPA-2	107.25	8.00	103.10	10.87	275.0*	0.10	0.435
Target PMLU	6.81	0.28	6.73	0.25	313.0*	0.32	0.301
Actual PMLU	6.30	0.51	6.21	0.54	-0.62	0.54	0.171
PWP	0.93	0.04	0.93	0.05	365.0*	0.91	0.043

Note. KLPA-2 = Kahn-Lewis Phonological Analysis, second edition; PMLU = phonological mean length of utterance; PWP = proportion of whole-word proximity. * indicates that a Mann-Whitney U-test was used. All other test statistics were based on independent *t*-tests.

Figure 8:

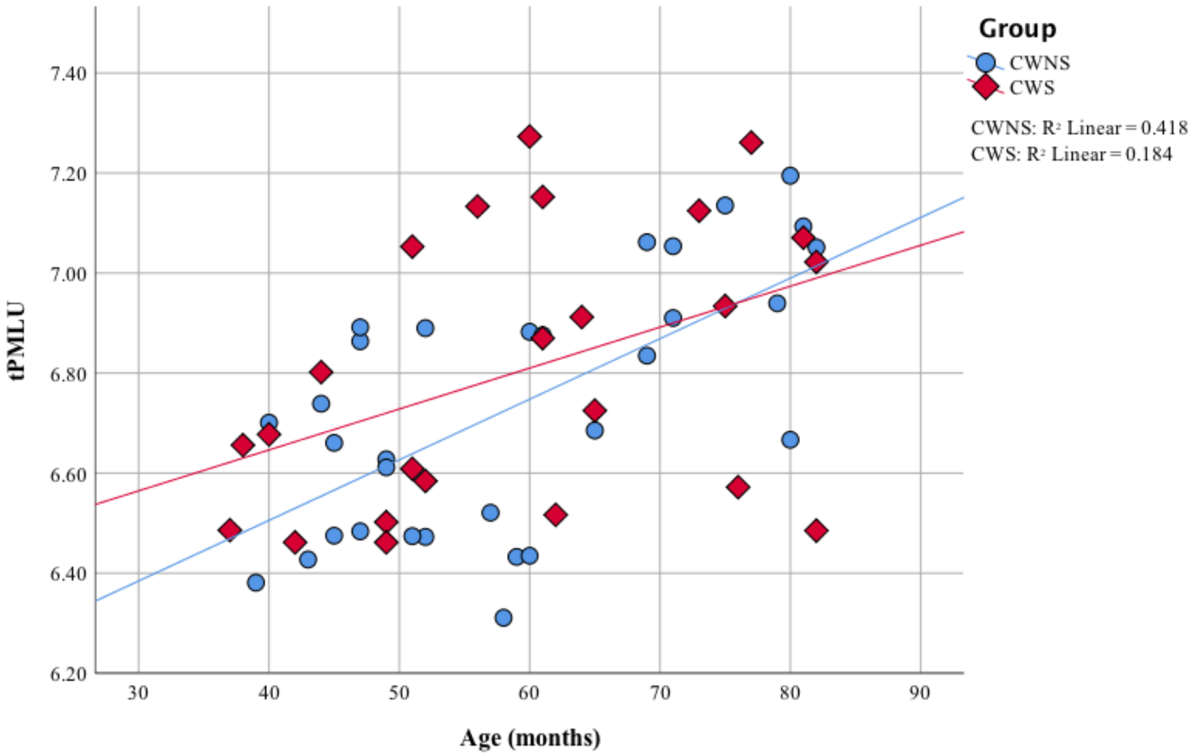
Scatterplot of Phonological Mean Length of Utterance (Actual) by Age for Each Group



Note. Figure 8 shows a scatterplot depicting aPMLU against chronological age for each group. There were no significant group differences. CWNS = children who do not stutter; CWS = children who stutter; aPMLU = actual phonological mean length of utterance.

Figure 9:

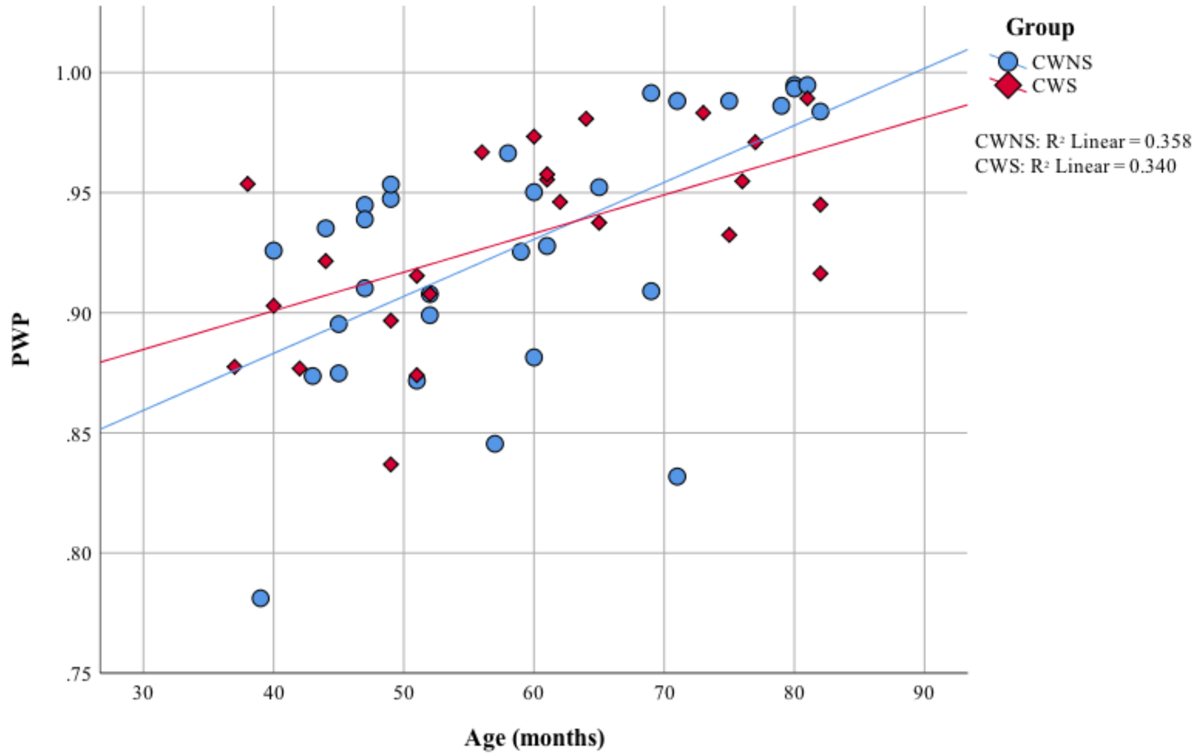
Scatterplot of Phonological Mean Length of Utterance (Target) by Age for Each Group



Note. Figure 9 shows a scatterplot depicting tPMLU against chronological age for each group. There were no significant group differences. CWNS = children who do not stutter; CWS = children who stutter; tPMLU = target phonological mean length of utterance.

Figure 10:

Scatterplot of Proportion of Whole-Word Proximity by Age for Each Group



Note. Figure 10 shows a scatterplot depicting PWP against chronological age for each group. There were no significant group differences. CWNS = children who do not stutter; CWS = children who stutter; PWP = phonological whole-word proximity.

DTI Results

Children Who Stutter Exhibit Widespread Associations Between White Matter Integrity & PWP Relative to Children Who Do Not Stutter

Associations between white matter integrity and PWP were examined separately for each group using voxel-wise $p = .005$, and cluster sizes > 15, which corresponded to $p = .05$ corrected for multiple comparisons. Within the group of children who stutter, a significant positive association existed between PWP and FA values along the bilateral dorsal and ventral language pathways. More specifically, the arcuate fasciculus, superior longitudinal fasciculus, inferior fronto-occipital fasciculus, and the corpus callosum exhibited a positive correlation between white matter integrity and PWP (Table 6; Figure 11).

In contrast to children who stutter, the children who do not stutter exhibited associations in the right dorsal language pathway only. Interestingly, the children who stutter exhibited a positive association between white matter integrity and PWP in the right arcuate fasciculus underlying the right IFG while children who do not stutter exhibited a negative association in this region. Furthermore, the children who do not stutter exhibited a positive association between PWP and white matter integrity in the right arcuate fasciculus underlying the right STG, which was not observed in children who stutter (Table 7; Figure 12).

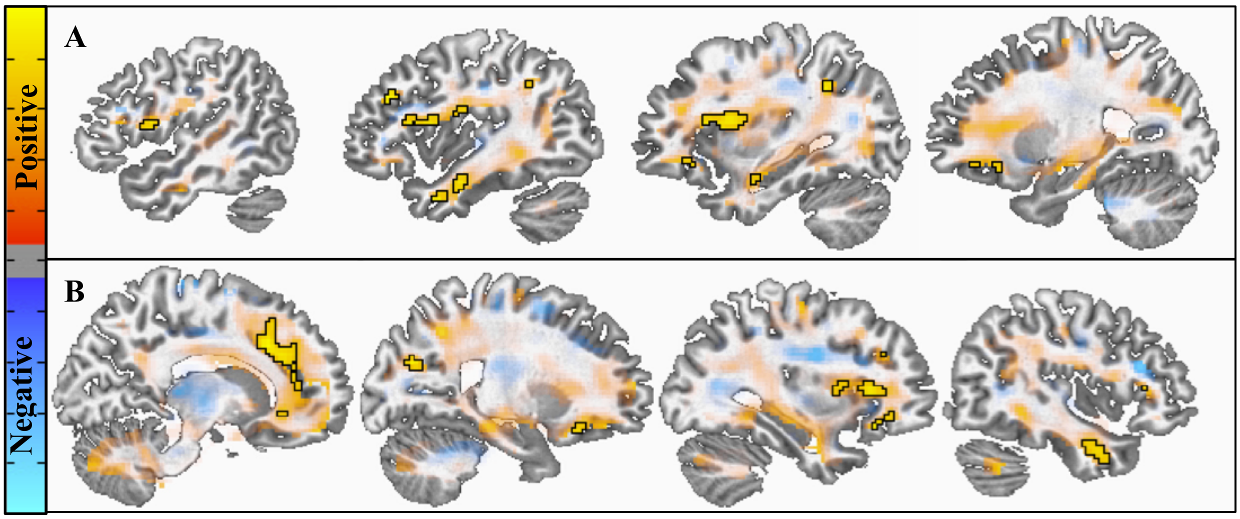
Table 6:

Significant Associations for PWP Modulation of Fractional Anisotropy in Children Who Stutter

White matter tracts	Regions (left/right)	Approx BA	Cluster size	max *t*	x	y	z
Cingulum	Cingulate gyrus (L)	32	140	3.90	-10	45	5
	Middle frontal gyrus (R)	8	106	4.38	12	22	32
Arcuate fasciculus	Insula (L)	13	134	3.92	-35	7	12
	Inferior frontal gyrus (R)	47	44	4.04	27	25	-17
	Inferior frontal gyrus (R)	45	37	4.07	35	25	7
	Insula (R)	13	25	4.09	37	7	10
	Middle frontal gyrus (L)	9	16	4.26	-37	30	27
Inferior longitudinal fasciculus	Inferior temporal gyrus (R)	38	43	3.78	40	-5	-27
	Inferior temporal gyrus (L)	38	43	3.81	-40	2	-32
Corpus callosum	Cuneus (R)	--	20	4.19	20	-72	22
	Middle frontal gyrus (R)	--	15	3.61	27	30	25
Superior longitudinal fasciculus	Angular gyrus (L)	39	18	3.81	-32	-50	32

Figure 11:

Significant Associations Between Proportion of Whole-Word Proximity & White Matter Integrity in Children Who Stutter



Note. Significant associations between PWP and FA in the dorsal and ventral language pathways in children who stutter are shown. The (A) left hemisphere and (B) right hemisphere are shown above in the sagittal plane. Significant clusters are displayed with a black outline. All clusters show a positive association. PWP = proportion of whole-word proximity; FA = fractional anisotropy.

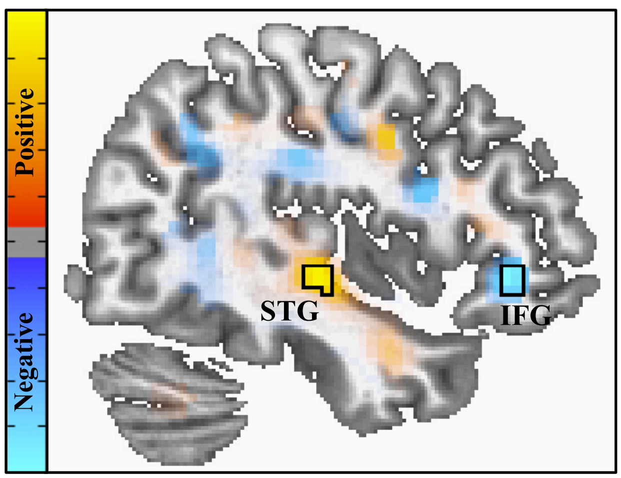
Table 7:

Significant Associations for PWP Modulation of Fractional Anisotropy in Children Who Do Not Stutter

White matter tracts	Regions (left/right)	Approx BA	Cluster size	max t	x	y	z
Arcuate fasciculus	Inferior frontal gyrus (R)	45	16	-3.94	45	32	-5
Inferior fronto-occipital fasciculus	Superior temporal gyrus (R)	21	15	3.73	42	-20	-5

Figure 12:

Significant Associations Between PWP & White Matter Integrity in Children Who Do Not Stutter



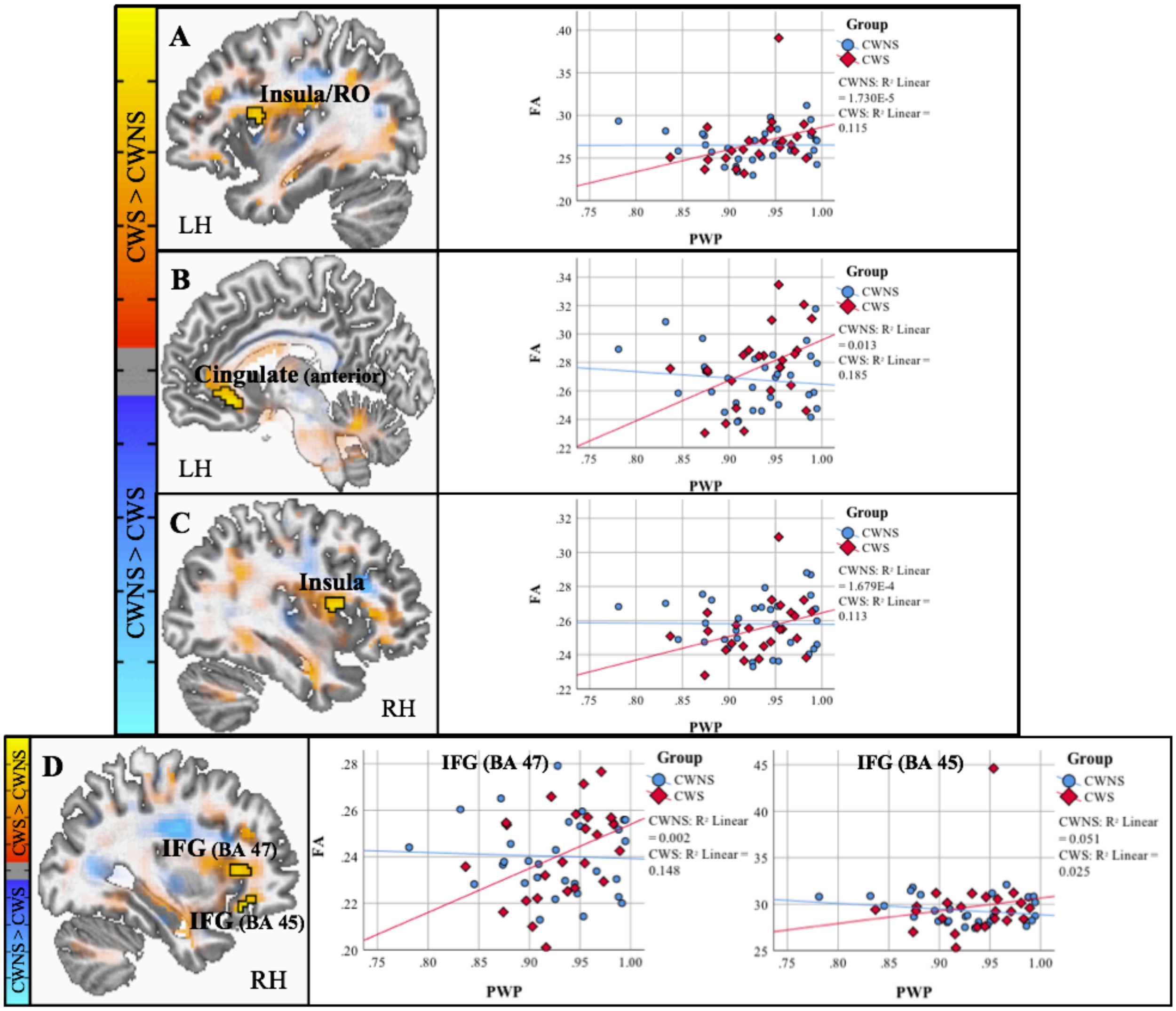
Note. Significant associations between PWP and FA in the right hemisphere are shown in the sagittal plane. Significant clusters are displayed with a black outline. Warmer colors represent a positive association between PWP and FA while cooler colors represent a negative association. PWP = proportion of whole-word proximity; FA = fractional anisotropy; IFG = inferior temporal gyrus; STG = superior temporal gyrus.

Group Differences in White Matter Integrity Associated with PWP

One primary aim of the study was to examine whether an association between white matter integrity and phonology exist among and whether this association differentiates children who stutter from children who do not stutter. Results from DTI analysis show that children who stutter exhibit greater associations between PWP and white matter integrity along the bilateral dorsal language pathways, which was not observed in children who do not stutter (Table 8; Figure 13). Children who stutter, relative to children who do not stutter, exhibited a stronger association between white matter integrity and PWP along the right arcuate fasciculus near the inferior frontal gyrus (BA47 and BA45) and the insula (BA13). Children who stutter also exhibited stronger associations than children who do not stutter in the left hemisphere arcuate fasciculus near the insula (BA13). However, these were smaller than those found in the right hemisphere. There was also a stronger association between PWP and white matter integrity along the left cingulum. The children who do not stutter exhibited a stronger relationship between PWP and FA values along the right superior longitudinal fasciculus underlying the precentral gyrus (BA3a; Figure 14). Fractional anisotropy values were extracted from each the six clusters where group differences for PWP modulation of white matter integrity were found, and are shown in the scatterplots below (Figures 13 & 14). A Fisher's r-to-z transformation was performed to test for potential differences between correlations. The results of these tests can be found in Appendix D and Appendix E.

Figure 13:

Group Differences for PWP Modulation of White Matter Integrity: Stronger Associations for Children Who Stutter than Children Who Do Not Stutter



Note. Significant group differences associated with PWP and FA underlying (A) the left insula, (B) the left anterior cingulate, (C) the right insula, and (D) the right IFG (BA 47) and (BA 45). Abbreviations: CWS = children who stutter; CWNS = children who do not stutter; PWP = proportion of whole-word proximity; FA = fractional anisotropy; LH = left hemisphere; RH = right hemisphere; IFG = inferior frontal gyrus; RO = Rolandic operculum.

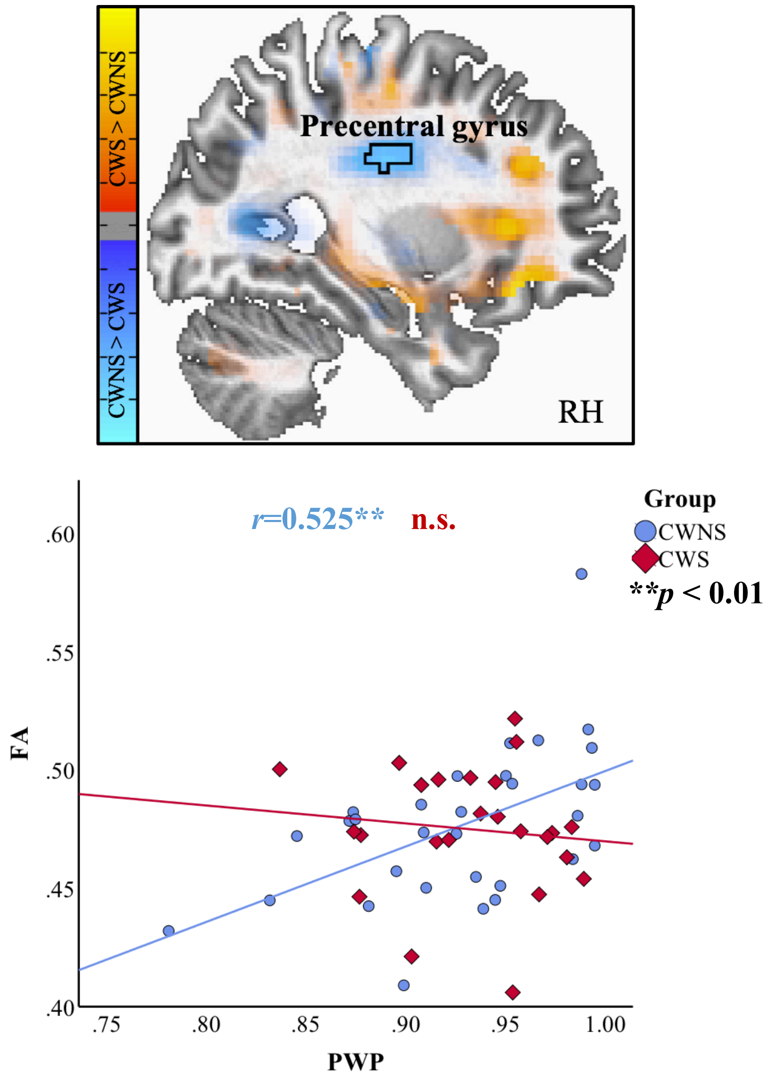
Table 8:

Significant Group Differences for PWP Modulation of Fractional Anisotropy

White matter tracts	Regions (left/right)	Approx BA	Cluster size	max *t*	x	y	z	Mean FA		Cohen's *d*
								CWS	CWNS	
Cingulum	Anterior cingulate (L)	24	81	4.23	-5	25	-10	0.276	0.268	0.361
Arcuate fasciculus	Inferior frontal gyrus (R)	47	47	2.93	32	27	-15	0.241	0.240	0.052
	Insula (L)	13	35	3.25	-35	7	12	0.268	0.265	0.128
	Inferior frontal gyrus (R)	45	26	3.02	23	25	5	0.297	0.293	0.154
	Insula (R)	13	18	3.39	37	7	10	0.255	0.258	0.183
Superior longitudinal fasciculus	Precentral gyrus (R)	3a	28	-3.35	30	-17	27	0.475	0.477	0.055

Figure 14:

Group Differences for PWP Modulation of White Matter Integrity: Stronger Associations for Children Who Do Not Stutter than Children Who Stutter



Note. Significant group differences associated with PWP and FA underlying the right precentral gyrus. CWS = children who stutter; CWNS = children who do not stutter; PWP = proportion of whole-word proximity; RH = right hemisphere; n.s. = not significant.

White Matter Integrity Associated with Age

Associations between white matter integrity and age were examined separately for each group using a voxel-wise $p = .005$, and cluster sizes > 15 ($p = .05$, corrected). Within the group of children who stutter, a significant positive association existed between age and FA values along the right corpus callosum underlying the precuneus, cuneus, and postcentral gyrus (Table 9; Figure 15). In contrast to children who stutter, the children who do not stutter exhibited significant positive associations in the bilateral dorsal and ventral language pathways (Table 10; Figure 16). In addition, the children who do not stutter exhibited associations between age and FA in the bilateral cerebellum and other subcortical structures such as the basal ganglia.

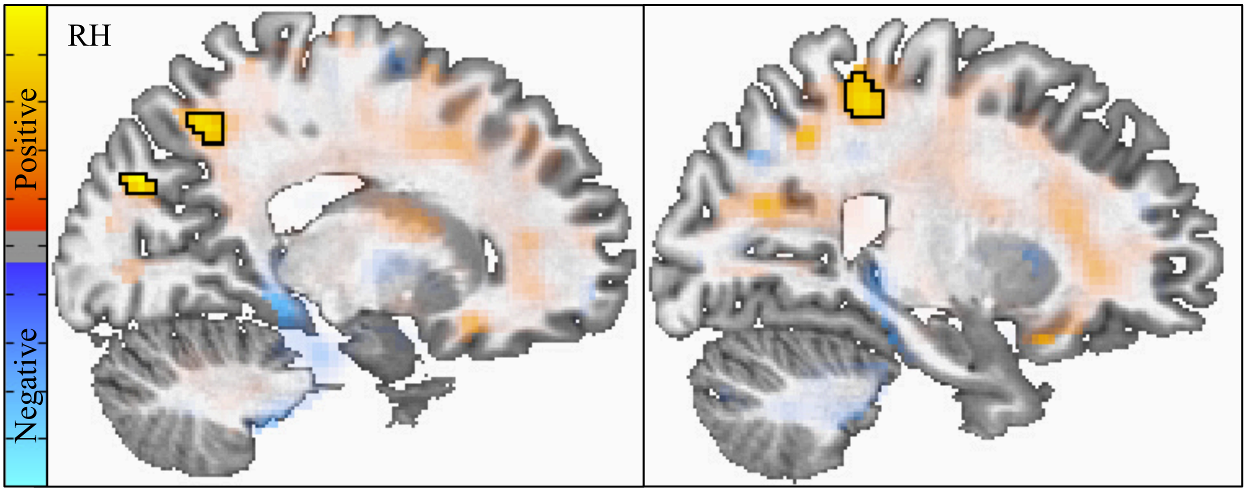
Table 9:

Significant Age Modulation of Fractional Anisotropy in Children Who Stutter

White matter tracts	Regions (left/right)	Approx BA	Cluster size	max t	x	y	z
Corpus Callosum	Precuneus (R)	7	26	4.47	20	-57	42
	Postcentral gyrus (R)	3b	23	3.82	25	-37	50
	Cuneus (R)	7	17	4.59	17	-77	27

Figure 15:

Significant Associations Between Age & White Matter Integrity in Children Who Stutter



Note. Significant positive associations between chronological age and fractional anisotropy in the right hemisphere are shown in the sagittal plane. The significant clusters are displayed with a black outline. RH = right hemisphere.

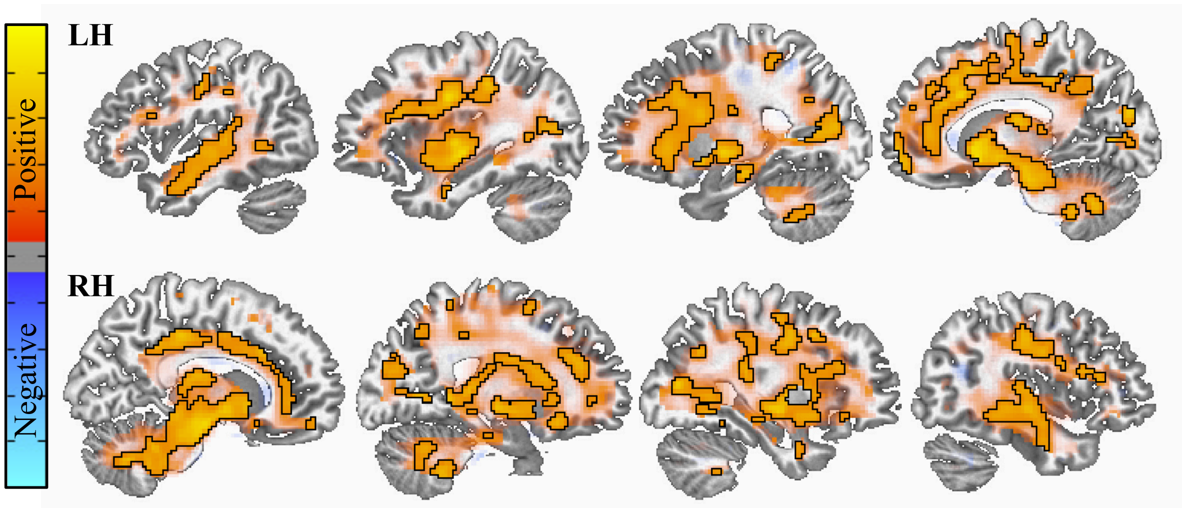
Table 10:

Significant Age Modulation of Fractional Anisotropy in Children Who Do Not Stutter

White matter tracts	Regions (left/right)	Approx BA	Cluster size	max t	x	y	z
Corpus callosum (frontal)	Superior frontal gyrus (L)	10	63	4.77	-10	57	-10
	Superior frontal gyrus (R)	6	35	4.36	15	2	60
	Superior medial gyrus (L)	9	19	3.85	-7	50	27
	Superior frontal gyrus (R)	6	16	5.15	12	-10	62
Corpus callosum (central)	Paracentral lobule (L)	31	197	4.71	-15	-35	45
	Middle frontal gyrus (R)	8	23	4.5	30	10	40
Corpus callosum (parietal-occipital)	Middle occipital gyrus (L)	30	296	6.43	-27	-75	10
	Postcentral gyrus (R)	5/6/31	188	5.48	17	-45	62
	Superior parietal lobule (L)	7	85	4.9	-15	-62	40
	Precuneus (R)	7/39	54	4.3	27	-62	30
Fornix/Copus callosum	Thalamus (L)	-	23	2.96	-17	-32	7
Cingulum	Cingulate gyrus (L)	24	18	3.24	-12	0	40
Inferior longitudinal fasciculus (posterior)	Middle temporal gyrus (L)	22	37	3.86	-35	-60	5
Inferior fronto-occipital fasciculus (anterior)	Middle frontal gyrus (L)	10/11	19	3.81	-27	47	-5
Corticospinal tract	Brainstem (L/R)	-	7499	7.14	-2	-20	-12
	Precentral gyrus (L)	4a/4p	16	3.53	-15	-30	62

Figure 16:

Significant Associations Between Age & White Matter Integrity in Children Who Do Not Stutter

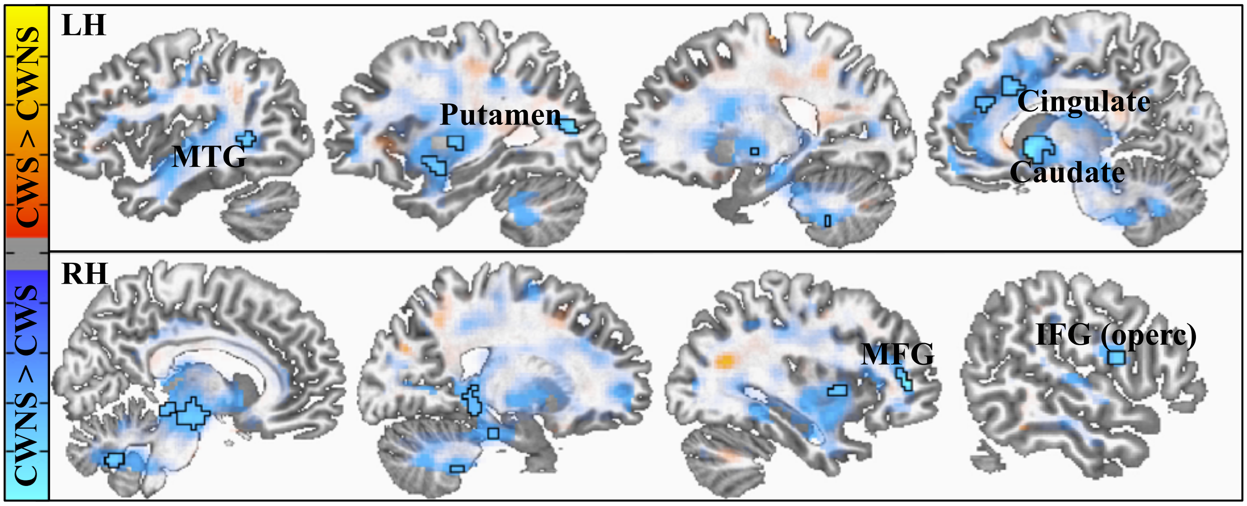


Note. Significant associations between chronological age and fractional anisotropy (FA) in the left hemisphere (LH) and right hemisphere (RH) are shown in the sagittal plane. Significant clusters are displayed with a black outline. Warmer colors represent areas where children who do not stutter exhibit a greater association between age and FA.

The whole brain analyses examining group differences in fractional anisotropy with age (i.e., Group x Age interaction) revealed that white matter integrity was higher in older children along the bilateral dorsal and ventral language pathways in children who do not stutter, but not children who stutter (Table 11; Figure 17). In addition, the children who do not stutter exhibited associations between age and FA in the bilateral cerebellum and other subcortical structures such as the basal ganglia. Children who stutter, however, only exhibited age-related FA increases along the right corpus callosum, underlying the postcentral gyrus, precuneus, and cuneus. Figure 18 shows significant correlations for each group for select regions found to exhibit group differences.

Figure 17:

Significant Group Differences for Age Modulation of White Matter Integrity



Note. Significant associations between chronological age and fractional anisotropy (FA) in the left hemisphere (LH) and right hemisphere (RH) are shown in the sagittal plane. Significant clusters are displayed with a black outline. Cooler colors represent areas where children who do not stutter (CWNS) exhibit a greater association between age and FA relative to children who stutter (CWS). MTG = middle temporal gyrus; MFG = middle frontal gyrus; IFG (operc) = inferior frontal gyrus (opercularis).

Table 11:

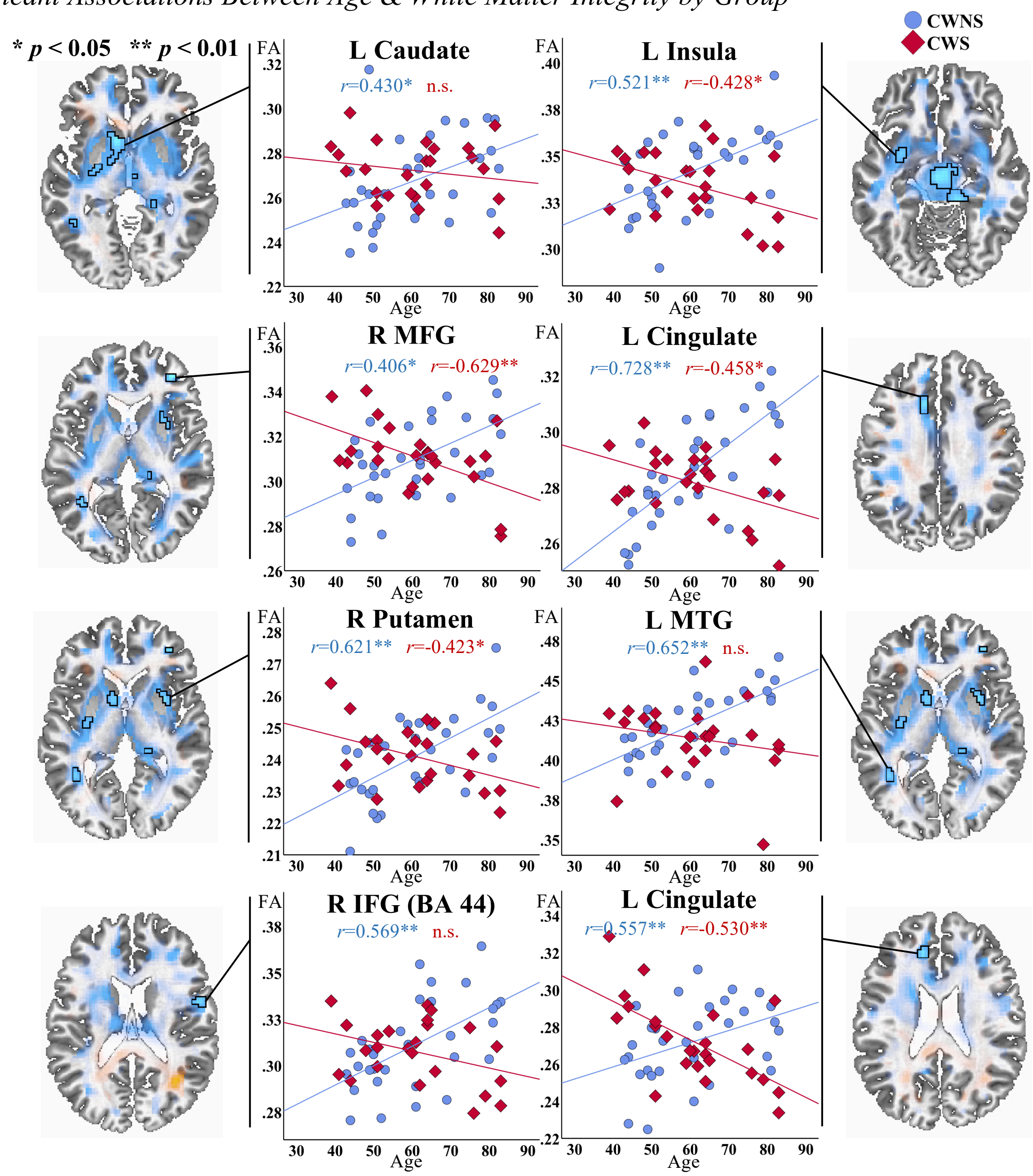
Significant Group Differences for Age Modulation of Fractional Anisotropy

White matter tracts	Regions (left/right)	Approx BA	Cluster size	max *t*	x	y	z	Mean FA CWS	Mean FA CWNS	Cohen's *d*
Internal capsule	Caudate nucleus (L)	-	123	-4.73	-10	10	-2	0.272	0.267	0.275
Cingulum [a]	Cingulate (L)	32	27	-4.33	-12	37	22	0.240	0.237	0.209
	Cingulate (L)	32	25	-4.22	-10	20	32	0.334	0.342	0.393
Cingulum [b]	Parahippocampal gyrus (R)	30	61	-4.68	15	-37	-10	0.281	0.287	0.232
	Parahippocampal gyrus (R)	28	15	-3.60	17	-25	-25	0.311	0.310	0.042
Arcuate fasciculus [c]	Middle temporal gyrus (L)	39	22	-3.78	-40	-55	2	0.282	0.286	0.274
Arcuate fasciculus [d]	Inferior frontal gyrus - opercularis (R)	44	19	-3.98	52	0	17	0.414	0.422	0.395
Inferior fronto-occipital fasciculus[a]	Putamen (R)	-	21	-4.08	32	5	2	0.241	0.241	0.000
	Middle frontal gyrus (R)	10	17	-5.05	35	45	5	0.307	0.311	0.179
Inferior longitudinal fasciculus	Middle occipital gyrus (L)	30/31	28	-4.18	-30	-80	10	0.272	0.272	0.000
Uncinate fasciculus	Insula (L)	36	37	-3.64	-30	-2	-15	0.399	0.408	0.293

[a] Anterior, [b] Posterior, [c] long segment, [d] anterior segment

Figure 18:

Significant Associations Between Age & White Matter Integrity by Group



Note. Scatter plots of average FA values extracted from significant clusters that differentiated the two groups (CWNS > CWS) in the extent of age effects on white matter integrity. In each scatter plot, children who do not stutter (CWNS) are represented in blue, while children who stutter (CWS) are indicated in red. L = left; R = right; FA = fractional anisotropy; MFG = middle frontal gyrus; MTG = middle temporal gyrus; IFG = inferior frontal gyrus; BA = Brodmann area; n.s. = not significant.

CHAPTER 4:

DISCUSSION

The overall aim of the current study was to compare whole-word phonological skills in spontaneous connected speech in children who stutter and children who do not stutter, and to examine the relationship between phonological skills and white matter integrity of major tracts that support phonological development. Unlike previous study that mostly recruited study participants through clinics, the current investigation involved recruitment of children who stutter from the community; therefore, children who stutter were comparable to children who do not stutter in all behavioral measures administered. The children who stutter did not exhibit comorbid speech-language deficits or other neurodevelopmental conditions that are commonly reported to be present in this clinical population. However, given the 2 standard deviation inclusion criteria, children may have, indeed, exhibited language deficits. The first aim of this study, guided by previous investigations, was to determine whether children who stutter exhibit phonological skills (as measured by PMLU and PWP) that lag relative to age-matched children who do not stutter. These measures of whole-word phonology reflected phonological complexity (PMLU) and the child's overall ability to approximate an intended target word (PWP). Previous studies have shown that children who stutter exhibit lower phonological ability than age matched peers near stuttering onset. Therefore, it was hypothesized that children who stutter as a group would perform less well than children who do not stutter on the Kahn-Lewis Phonological Analysis. Based on previous research examining speech production abilities in children who stutter, it was also hypothesized that children who do not stutter would exhibit greater whole-word phonological skills compared to the children who stutter. That is, children who do not stutter would produce words of greater phonological complexity (PMLU) while also approximating target words with greater accuracy (PWP) than children who stutter.

The second aim of the study was to examine whether children who stutter differ from children who do not stutter in white matter integrity in the dorsal language pathway associated with phonological skills (PWP). Based on previous studies reporting decreased FA in the left hemisphere in these tracts in children who stutter compared to children who do not stutter, it was hypothesized that children who stutter would exhibit a weaker association between PWP and white matter integrity of the left hemisphere dorsal language pathway relative to children who do not stutter. Guided by previous studies that have reported heightened right hemisphere homologue structural and functional increases in auditory-motor connectivity in people who stutter. This may indicate compensatory increases to weaker left dorsal tract connectivity. Therefore, it was further hypothesized that a stronger association between PWP and white matter integrity along the right dorsal language pathway would be observed in children who stutter relative to children who do not stutter.

Summary of Main Findings

There were no statistically significant differences between children who stutter and children who do not stutter in whole-word phonological ability as measured by tPMLU, aPMLU, or PWP. In the context of phonological development, tPMLU scores represents the phonological complexity a child attempts to produce, while aPMLU reflects the child's ability to accurately produce target words. Target PMLU and aPMLU can also be used to identify a child's stage of phonological acquisition. For example, lower PMLU scores, both target and actual, are indicative of a child producing less complex phonology. PWP is the ratio of aPMLU to tPMLU, and represents a child's ability to accuracy producing a target word, with 1.00 representing a perfect production of the target word. The current study also examined phonological abilities using traditional standardized assessment measures such as the Kahn-Lewis Phonological Analysis (KLPA-2). There were no significant differences between children who stutter and children who do not stutter in phonological ability as measured with KLPA-2, though an equivalence test indicated that the group difference exceeded equivalence (i.e., the two groups differed meaningfully given a lower and upper bound of effect sizes, with the stuttering group showing trends for increases relative to the non-stuttering group). A follow up study with larger sample sizes would help clarify these findings. The overall set of results from the behavioral measures of phonology are somewhat surprising given that previous studies reported that children who stutter, as a group, perform more poorly on standardized assessments of phonology near stuttering onset than children who do not stutter (Paden et al., 1999, 2002; Paden & Yairi, 1996). However, given the fact that some of the children had stuttered for greater than 1 to 2 years, the current study may have failed to detect group differences because these differences no longer present (Paden et al., 2002). Previous studies have provided mixed results regarding phonological differences between children who stutter and children who do not stutter (Bloodstein & Bernstein Ratner, 2008). However, earlier studies examining phonological differences in children who stutter and children who do not stutter utilized assessments that analyzed phonology using single words. While phonological assessments, such as the KLPA-2, prove to be useful in diagnosing frank phonological disorders, these assessments may not be sensitive enough to detect subtle differences between two groups of children whose overall language and articulation skills are similar. Finally, the participants included within the study were 37 to 82 months old, which might be too broad to differentiate the two groups (Paden et al., 2002). For example, Paden and Yairi (1996) reported that phonological skills differentiated children who do not stutter from children whose stuttering would become persistent, but not those who would recover from stuttering when each group was less than 4 years of age. In two follow up studies Paden and colleagues (1999; 2002) examined the phonological development in of children 5 to 6 years of age, and failed to detect differences in phonological development.

The above factors, and the fact that this study may have been slightly underpowered for this analysis, warrants caution in interpreting the present behavioral results. As previously mentioned, a sample size of 51 to 64 children per group would be needed to detect group differences in phonological variables with an alpha = .05, effect size = 0.5, and a moderate beta (.70-.80). Although the main longitudinal study has completed behavioral testing on 119 children (children who stutter = 64, children who do not stutter = 64), the current study's inclusion criteria significantly reduced the number of participants to a total of 55 participants (children who stutter = 24, children who do not stutter = 31). The equivalence tests provide more confidence that the two groups do not differ meaningfully in PWP (Figure 7B). For KLPA and PMLU measures however, the results are inconclusive and thus, additional studies will need to be conducted to determine whether children who stutter and children who do not stutter differ in these measures.

A whole-brain, voxel-based analysis was used to examine the association between PWP and FA along major white matter tracts. The effect of age on white matter integrity was also examined. Lower FA values reflect less robust development and structural connectivity among the network of regions that support rapid interaction required for speech production. The current study found that children who stutter exhibit a greater association between PWP and white matter integrity in the bilateral dorsal language pathways than children who do not stutter. Notably, these areas were along the arcuate fasciculus, and included the right IFG (BA 45/47), right insula, and left insula/Rolandic operculum. Relative to controls, children who stutter exhibited decreased association between PWP and white matter integrity along the right SLF underlying the precentral gyrus. These results suggest that increased white mater integrity within the bilateral dorsal language pathway may allow children who stutter to perform at a comparable level in terms of phonological complexity as children who do not stutter.

Previous research found that lower FA in children who stutter may reflect less robust development and connectivity among regions supporting auditory-motor integration (Chang, Garnett, et al., 2018; Neef et al., 2015), which is critical in supporting fluent speech production. Our results demonstrate that there were robust associations between white matter integrity and age along the dorsal and ventral language pathways in children who do not stutter, but such relationships were not found in children who stutter.

There were no significant differences between children who stutter and children who do not stutter on PWP, though the children who stutter exhibited greater association between PWP and white matter integrity along the bilateral dorsal pathways. Children who do not stutter showed robust white matter integrity scores in bilateral dorsal and ventral pathways associated with age, while children who stutter did not. The greater association between PWP and the bilateral dorsal tracts in children who stutter in the face of attenuated age-related increases in the same areas, suggest that stronger links between PWP and dorsal pathway integrity may be needed in children who stutter to achieve comparable phonological development to children who do not stutter. That is, heightened white matter integrity of the dorsal tracts might be particularly important to children who stutter for maintaining typical phonological development.

Behavioral Findings

Children Who Stutter do not Differ from Children Who Do Not Stutter in Phonological Ability as Measured by Whole-Word Phonological Measures

The association between phonology and stuttering has been studied extensively using surveys (Arndt & Healey, 2001; Blood et al., 2003), articulation assessments (Anderson & Conture, 2000; Ryan, 1992), and phonological assessments (Spencer & Weber-Fox, 2014). Results of a range of studies have yielded complex and contradictory findings (Bloodstein & Bernstein Ratner, 2008). For example, previous behavioral studies have reported that the articulatory/phonological abilities of children who stutter are equivalent to children who do not stutter (Ryan, 2001). Other studies have found that children who stutter have an increased rate of co-occurring phonological disorders (Arndt & Healey, 2001; Blood et al., 2003; Louko et al., 1990) and may have less efficient phonological skills than children who do not stutter (Paden et al., 1999; Paden & Yairi, 1996). Phonological skills have also been examined near stuttering onset to determine whether differences in phonology (e.g., using single word productions that examine individual phonemes to gain understanding of a child's phonological skills) is useful in predicting stuttering persistence (Paden et al., 1999; Spencer & Weber-Fox, 2014).

Previous studies examining phonological skills have utilized standardized assessments to identify error patterns as a child produces a specific speech target. The current study examined whether phonological complexity differed between children who stutter and children who do not stutter, and whether these whole-word targets were less accurate among children who stutter based on spontaneous speech samples. There were no statistically significant differences between children who stutter and children who do not stutter on measures of phonological complexity (aPMLU or tPMLU). Furthermore, the current study did not find a significant group difference between the two groups of children in their ability to accurately approximate target words, as indexed by PWP.

Many of the previous studies have reported subtle differences in the articulatory and phonological abilities of children who stutter by examining phonological processes measured with standardized assessments (Anderson & Conture, 2000; Coulter et al., 2009; Pellowski & Conture, 2002; Spencer & Weber-Fox, 2014; St. Louis & Hinzman, 1988). Therefore, the current study also used a standardized assessment to assess phonological processes, to complement findings for whole-word measures. Here, the results showed that there were no statistically significant differences between children who do and do not stutter on both the Goldman-Fristoe Test of Articulation or the Khan-Lewis Phonological Analysis. These findings are inconsistent with previous studies of children who stutter that found differences in articulation and phonology (Anderson et al., 2005; Anderson & Conture, 2004; Byrd et al., 2007; Louko et al., 1990; Ryan, 1992).

Although there were no statistically significant differences between the two groups based either on standardized assessments or whole-word measures of phonology, it might be due to the fact that children ranged between 3;1 (years; months) and 6;10 in the current study. Earlier studies have examined phonological processes in single words and spontaneous speech to determine whether differences exist between typically fluent children and children who stutter (i.e., increased errors indicating reduced mastery of phonological processes, such as cluster reduction). However, unlike these earlier studies, the current study did not examine differences in phonological processes between the two groups. Rather, spontaneously produced speech was used to examine the whole word phonological abilities of children. The present findings add to the present body of literature suggesting that phonological differences between children who stutter and children who do not stutter may be too subtle to detect using conventional measures of phonology (i.e., KLPA-2). It has been suggested that advanced proficiency in one domain (i.e., phonology, language, etc.) might require decreased performance in other domains of development. In the current study, children who stutter demonstrated greater lexical diversity than children who do not stutter; however, these words were not found to be more phonologically complex (as measured by PMLU). Developmental tradeoffs of this nature (i.e., greater vocabulary diversity) might require limitations in areas that extend to the production of fluent speech.

The current results indicate that whole-word phonological analysis may not be a sensitive measure for differentiating children who do and do not stutter. In addition to possible power issues, the results may have failed to detect differences because the children enrolled in the study were 3 to 6 years of age and produced many of the spontaneous utterances with accuracy. Ingram (2002), who investigated whole-word phonology in children 2 to 4 years of age, suggested that whole-word measures may be useful for placing children in developmental stages. Other studies have utilized whole-word phonological measures to examine speech production abilities in children with specific language impairments (SLI) between 4 to 5 years of age (Kunnari et al., 2012). The results of that study showed that children with SLI exhibited reduced phonological mean length of utterance and reduced PWP scores. Because the children within the current study were not diagnosed with phonological disorders, the measure may not be sensitive at this age range.

Exploratory Findings in Children 3 to 4 Years of Age

Exploratory analysis was conducted on a small number of participants 3 to 4 years of age to determine whether phonological differences existed between children who stutter (N = 11) and children who do not stutter (N = 17). Statistically significant differences existed between children who stutter and children who do not stutter on the GFTA-2 ($t(26) = -2.154$, $p = .041$; $d = .88$) and the KLPA-2 ($U = 41.50$, $z = -2.46$, $p = .013$; $d = .94$). Contrary to the main hypothesis, this group of children who stutter achieved higher articulation and phonological scores on standardized tests than children who do not stutter. No statistically significant differences existed between the two groups for any whole-word phonological measure (aPMLU, tPMLU, or PWP) or any other measure of language (MLU, NDW, VOCD). These findings, in addition to the exploratory analysis of children 3 to 4 year of age, corroborate findings of Luckman and colleagues (2020), who found that significant differences exist on standardized tests, but do not differ on spontaneous language measures of lexical diversity (Table 12).

Table 12:

Speech & Language Test Results Among Children 3 to 4 Years of Age

Variables	Children Who Stutter		Children Who Do Not Stutter		Test statistic	*p*	Cohen's *d*
	M	*SD*	*M*	*SD*			
GFTA-2	109.27	6.96	101.00	11.40	-2.15	*0.04*	0.88
KLPA-2	111.36	5.45	104.82	8.21	41.50*	0.01	0.94
Target PMLU	6.68	0.23	6.59	0.18	74.00*	0.38	0.44
Actual PMLU	5.96	0.45	5.91	0.40	-0.31	0.76	0.12
PWP	0.90	0.04	0.91	0.05	0.18	0.86	0.22
MLU	5.46	2.24	5.11	1.10	87.00*	0.78	0.20
NDW	50.73	9.47	50.76	8.24	0.01	0.99	0.00
VOCD	45.62	13.45	44.43	7.73	-0.27	0.79	0.11

GFTA-2 = Goldman-Fristoe Test of Articulation, second edition; KLPA-2 = Kahn-Lewis Phonological Analysis, second edition; PMLU = phonological mean length of utterance; PWP = proportion of whole-word proximity; MLU = mean length of utterance; NDW = number of different words; VOCD = vocabulary diversity. * indicates that a Mann-Whitney U-test was used. All other test statistics were based on independent *t*-tests.

Previous studies have examined phonological development in young children who stutter within one year of stuttering onset, and found that, within six months of stuttering onset, children who stutter achieved scores that were significantly lower than children who do not stutter. After one year, the phonological abilities between children who stutter and children who do not was no longer significantly different (Paden et al., 2002). This suggests that as children who stutter age, accelerated growth in phonology may occur, thereby eliminating the difference observed between the two groups. The current study examined children 3 to 6 years of age. Thus, lack of significant group differences might be attributed to examining children in a wide rage range that may have washed out the group findings. That is, it is possible that some children who stutter may have "caught up" with children who do not stutter after an earlier period of developmental delay. Alternatively, PMLU, along with PWP, may not be sufficiently sensitive to detect subtle differences in phonology when used in spontaneous speech. For example, if children utilize language skills familiar to them (e.g., using simple clauses and phrases), it is likely that the speech mechanism is not being taxed. Previous studies have provided evidence that people who stutter exhibit increased variability of speech motor coordination with increased syntactic and phonological complexity, which further indicate an impact of language demands on speech motor stability (MacPherson & Smith, 2013; Smith et al., 2012). However, it is important to note that the findings of MacPherson and Smith (2013) found higher variability regardless of syntactic complexity. Group differences may have been better captured if greater demands on language formulation were required. One way to tax speech motor abilities may be utilizing non-word repetition tasks that increase in syllable length and structure, which would increase the task complexity and might create breakdowns in articulation.

DTI Findings

Children Who Stutter Exhibit Positive Associations Between PWP & White Matter Integrity Along the Dorsal Language Pathway

In the current study, it was hypothesized that, compared to children who do not stutter, children who stutter would exhibit a weaker association between PWP and white matter integrity in the dorsal language pathway of the left hemisphere (i.e., the arcuate fasciculus and superior longitudinal fasciculus), and a greater association in the right dorsal language pathway. Contrary to expectations, the results of the study revealed that children who stutter, relative to children who do not stutter, exhibit a greater association between PWP and white matter integrity along the bilateral dorsal language pathway, specifically in the anterior segment of the arcuate fasciculus. For children who stutter, significant associations between PWP and white matter integrity were found in the arcuate fasciculus underlying the right IFG (BA 45/47), and the bilateral insula. Increases in white matter integrity provide the necessary structural support to allow for the efficient and rapid interaction among various brain regions, which in turn support well-functioning and synchronized neural circuits. Increased white matter has also been found to change across development (Perani et al., 2011), and is associated with skill acquisition and training (Chenausky et al., 2017; Han et al., 2009; Salminen et al., 2016; Sampaio-Baptista & Johansen-Berg, 2017). Convergent findings have shown that stuttering is associated with decreased FA along the left dorsal language pathway in both children and adults who stutter (Chang et al., 2008; Cykowski et al., 2010; Sommer et al., 2002; K. E. Watkins et al., 2008). Children who stutter exhibit decreases in fractional anisotropy within left hemisphere regions underlying sensorimotor regions along the dorsal language pathway (Chang et al., 2015). It was also shown that when children who stutter age, as a group, exhibit stagnant growth in the dorsal language pathway underlying the left IFG while children who do not stutter show linear FA increases with age (Chang et al., 2015).

Adults who stutter have been reported to exhibit anomalous activity in the brain, such as reduced activity in left hemisphere areas required for speech production with an over-activation in homologous areas of the right hemisphere, including Broca’s homologue (Braun et al., 1997; Chang et al., 2009; Fox et al., 1996; Kell et al., 2009). This overactivation within the right hemisphere has been hypothesized to be an attempt to compensate for functional and structural deficits within the left hemisphere (Kell et al., 2009; Preibisch et al., 2003). Interestingly, evidence has shown that this over-activation is corrected following treatment to reduce stuttering severity (Kell et al., 2009).

Importantly, sound-to-articulation mapping has been associated with the dorsal language pathway within the language dominant left hemisphere (Hickok & Poeppel, 2004, 2007; Saur et al., 2008), which is commonly reported to have decreased integrity among people who stutter (Chang, Garnett, et al., 2018). Moreover, increased white matter integrity along the left dorsal language pathway has been linked to consonant production within syllables (Chenausky et al., 2017; Saur et al., 2008). Thus, children who are likely to display reduced white matter integrity in the left dorsal language pathway, such as children who stutter, might also exhibit a less mature phonological skillset. Given this, the positive association between PWP and white matter integrity along the dorsal language pathway in children who stutter that goes beyond age-related increases, may indicate a mechanism that allows children who stutter to perform at a similar level as children who do not stutter. Alternatively, the observation of greater reliance on the dorsal tracts among the children who stutter may reflect phonological development occurring in earlier stages, when the speaker needs to consciously monitor the auditory outcome of one's own speech production (Hickok, 2014). In children who do not stutter, producing age-appropriate phonology may already be well-established as a group and thus may not rely as much on the integrity of the dorsal tracts. The current study also found that older children who do not stutter exhibited a stronger association between age and white matter integrity in bilateral white matter pathways, which was not observed in children who stutter. These results will be discussed below.

In sum, the current DTI results indicate that greater white matter integrity along the bilateral dorsal language pathways is associated with better phonology as measured with PWP in children who stutter. This might suggest that heightened involvement of the left dorsal tracts is particularly important for children who stutter to achieve comparable phonology as seen in children who do not stutter, while children who do not stutter, as a group, may not vary as much in their phonological skills to have significant associations with white matter integrity in the dorsal tracts. That is, the children who do not stutter may not rely on the integrity of the right hemisphere tract any longer to produce age-appropriate phonology. Because DTI only allows examination of structural changes, it would be important to examine with functional imaging to confirm whether heightened activity is present in regions connected via the left dorsal tracts in children who stutter that is associated with better phonology development.

Children Who Do Not Stutter Exhibit Associations Between White Matter Integrity & PWP in the Right Hemisphere

In addition to examining overall group differences related to white matter integrity and PWP, this association was also explored within each group. Similar to the above findings, children who stutter exhibited a positive association along the bilateral dorsal language pathways. In addition, the children who stutter also exhibited an association between PWP and white matter integrity along the bilateral ventral language pathways. However, the children who do not stutter exhibited PWP associations along the dorsal language pathway of the right hemisphere. Specifically, children who do not stutter exhibited a positive association underlying the right STG, and a negative association underlying the right IFG. The dorsal language pathway, specifically the superior longitudinal fasciculus, is responsible for connecting the posterior STG and the inferior frontal gyrus. This pathway shows protracted maturation across development, and exhibits increases in white matter integrity between the ages of 3 and 10 years

(Skeide et al., 2016). The positive association observed in the right STG might reflect that increased myelination and organization of the dorsal language pathway allows for more efficient communication with among structures supporting speech production accuracy. This increased association between age and white matter integrity underlying the STG might reflect increases from feedback monitoring during speech development. The posterior STG plays a critical role in sensorimotor integration whereby acoustic information is mapped to articulation, and the output of the speech production is monitored via the auditory system. Given the negative association observed along the right arcuate fasciculus underlying the IFG, this might be interpreted to mean that the right arcuate fasciculus is not required to support phonology in spontaneous speech. Alternatively, a more left-lateralized system might be more efficient and lead to better phonology. Thus, children who do not stutter might exhibit a negative correlation with the right IFG and PWP because greater white matter integrity underlying the right IFG might have a negative impact on phonological development.

These within-group analyses show more clearly how the two groups differ in the relationship between white matter integrity and phonological development. Despite showing comparable phonology scores (PWP), children who stutter, as compared to children who do not stutter, exhibited very different white matter integrity scores associated with PWP. Namely, greater white matter integrity in both bilateral dorsal and ventral tract was associated with higher PWP in children who stutter, while PWP was associated with mostly right dorsal tract changes in children who do not stutter. These results indicate that the structural integrity of the anterior bilateral dorsal pathway might allow children who stutter to produce phonology that is comparable to children who do not stutter.

Children Who Do Not Stutter Exhibit Age-Related Associations in White Matter Integrity

Previous research has shown that the dorsal language pathway continues to develop into adolescence in typically developing children (Barnea-Goraly et al., 2005; Hüppi & Dubois, 2006; Kochunov et al., 2012). The children who stutter did not exhibit robust significant associations between white matter integrity and age, which is common in other neurodevelopmental disorders, and has been reported in children with ADHD (Hamilton et al., 2008), developmental language disorder (Lee et al., 2020; Verly et al., 2019), and dyslexia (Yeatman et al., 2011). In the current study, children who do not stutter showed robust positive correlations between age and white matter integrity along the bilateral dorsal and ventral language pathways. These age-relevant increases in white matter integrity were not observed in children who stutter. Rather, children who stutter exhibited an association between FA and age in the right hemisphere corpus callosum. These results indicate that children who stutter may not exhibit the same associations between age and white matter integrity, thereby demonstrating a more protracted development of white matter integrity than children who do not stutter. Previous studies examining FA among adults who stutter have also reported white matter integrity differences between adults who stutter and fluent controls.

The results of the current study are in agreement with previous studies showing that children who do not stutter exhibit greater FA increases with age along the dorsal language pathway compared to children who stutter (Chang et al., 2015). A longitudinal study examining white matter developmental trajectories in children who stutter found that children who stutter have slower FA growth rates with age along the arcuate fasciculus compared to children who do not stutter (Chow & Chang, 2017). Atypical development of white matter tracts connecting frontotemporal and motor areas may lead to unstable sound-to-motor mapping and decreased coordination of articulatory movements in people who stutter (Smith et al., 2010, 2012; Walsh et al., 2015). The current findings show that children who do not stutter were found to exhibit a negative correlation between PWP and white matter integrity underlying the right IFG despite showing age-related associations with white matter integrity. In addition, the children who do not stutter exhibit an association between age and greater white matter integrity along the bilateral dorsal language pathway, which is not observed in children who stutter. Furthermore, these correlations were found to be significantly different between the two groups (see Appendix E).

These findings lend further support to the notion that the significant association between PWP and white matter integrity in children who stutter might facilitate similar phonological ability. Because the current results are based on cross-sectional samples, the present findings will need to be confirmed with longitudinal analysis in the future to determine whether the relationship between phonology and FA change across development.

Implications for Neuroanatomical Theories of Stuttering

The evidence provided by the current study may suggest that children who stutter exhibit increased white matter along the bilateral dorsal language pathways to provide additional support for phonology. Previous studies have advanced the notion that individuals who stutter may have failed to acquire stable and correct mappings between motor programs and sensory consequences, which results in difficulty using these mappings for efficient sensorimotor control of speech (Max et al., 2004). The dorsal language pathway of the left hemisphere plays a critical role in sound-to-articulation mapping. However, children and adults who stutter are consistently reported to have decreased white matter integrity along this pathway (Chang, Garnett, et al., 2018), which may cause instability in the speech-motor system and create breakdowns in speech production. This is supported by behavioral evidence which has demonstrated that children and adults show variable articulatory coordination patterns during nonword learning tasks (Smith et al., 2010; Smith & Zelaznik, 2004).

The exploratory findings from the 3 to 4 year-old children who stutter within the study were found to score significantly higher than children who do not stutter on standardized assessments of articulation and phonology. However, this finding was not significant when measures of whole word phonology were examined. These results were somewhat surprising because spontaneous speech samples are considered to be the most useful for evaluation of language. For example, the elicitation of specific target words produced during the GFTA-2 allow for the production to be compared in a consistent manner; however, it is also acknowledged that children typically produce more errors during continuous speech than single-word assessments. The results of the current study might indicate that children who stutter produce language similar to children who do not stutter during spontaneous speech to overcome an increase in linguistic demand (i.e., syntax, semantics, etc.). In addition, studies reporting phonological weaknesses in children who stutter have utilized cross-sectional data to report on phonological abilities in children who stutter. However, the current study leveraged speech samples and MRI data that were collected as part of a larger longitudinal study that enrolled participants from a larger community, and did not utilize clinical-based samples. Thus, the participants in the current study may better represent the broader population of children who stutter.

The findings that children who stutter have a greater association between white matter integrity and phonology along the bilateral dorsal language pathway might represent a compensatory mechanism that allows children who stutter to produce accurate phonology despite instabilities in motor programming. In sum, the current study demonstrates that the phonological abilities of children who stutter are similar to children who do not stutter; however, the white matter structures supporting phonological abilities appear to differ between the two groups. Further studies are needed to investigate whether these differences continue into adolescence and adulthood. Moreover, studies should examine whether an increased phonological load (i.e., increased phonological complexity) reveals greater differences between the two groups. Nonword repetition tasks have been used to differentiate children who stutter and children who do not stutter. Calculating whole-word phonology scores for these tasks might reveal behavioral differences, while also providing additional details regarding the association between white mater integrity and phonological abilities.

Limitations

The current study used a novel method to assess phonological skills within continuous speech among children who do and do not stutter who were 3 to 6 years of age. Due to the relatively large age range, it is possible that children (especially children who do not stutter) may have performed near the ceiling making it difficult to observe group differences. Future studies should examine children at 3 years of age to determine if differences in whole-word phonology are present. The current study did not examine whole-word phonology using the single-word productions from the GFTA-2 that were used to calculate KLPA-2 scores. The calculation of aPMLU and PWP might reveal differences between the two groups. Moreover, some children included in the current study were not within one year of stuttering onset because their DTI scans were unable to be used due to excessive movement. Additional studies are needed to determine if children within one year of stuttering onset differ from children who do not stutter on whole-word phonological measures. It is unlikely that the results of individual DTI scans would significantly change within two years of stuttering onset; however, obtaining phonological scores near stuttering onset might result in differences in the association between PWP and white matter integrity. Furthermore, it will be important to determine with a longitudinal study to examine how PWP modulates changes in white matter integrity across development, and whether children who recover from stuttering exhibit associations between PWP and white matter integrity. Finally, because the children who stutter that were enrolled in this study were within three years of stuttering onset, stuttering prognosis (persistence versus recovery) may change, and may impact the results presented. However, because these children are enrolled in a longitudinal study, the chronicity of stuttering is able to be monitored. Previous studies have indicated that by 4 to 5 years of age, children who stutter have an estimated 50% to 60% likelihood of recovery (Yairi et al., 2005). Therefore, it is unlikely that the results of the current study are impacted by the stuttering status of the children enrolled.

Conclusion

This study was conducted to examine the whole-word phonological skills of children who stutter and children who do not stutter using phonological mean length of utterance and proportion of whole-word proximity (Ingram, 2002). In addition, the current study examinined the relationship between PWP and white matter integrity along the bilateral dorsal language pathway, which is critical in mapping sound-to-articulation (Hickok, 2012; Hickok & Poeppel, 2004). In short, the aims of the study were to 1) determine whether whole-word phonological skills in spontaneous speech differ between 3 to 6 year-old children who stutter and children who do not stutter, and 2) determine whether children who stutter or children who do not stutter exhibit stronger associations between PWP and white matter integrity along the dorsal language pathway.

In line with these aims, this study found that the phonological skills did not differentiate 3 to 6-year-old children who stutter from children who do not stutter. However, the relationship between PWP and white matter integrity was found to differentiate the two groups. Namely, the group of children who stutter exhibited stronger associations between PWP and white matter integrity along the bilateral dorsal language pathways, which was not found in the group of children who do not stutter. The significant group differences for PWP-white matter integrity associations were found in the context of age-related differences in white matter integrity. Specifically, while children who do not stutter exhibited widespread bilateral age-related increases in white matter integrity, the children who stutter showed were only found to exhibit positive associations between age and FA in the right hemisphere corpus callosum. These results indicate that bilateral increases in white matter integrity that goes above and beyond age-related increases support phonological skills in children who stutter, allowing them to perform similar to children who do not stutter in phonological ability. In addition to the exploratory findings, the results of the current study suggest that children who stutter near stuttering onset may exhibit comparable or better phonological skills relative to children who do not stutter, which is largely inconsistent with findings reported in earlier studies. Finally, the results of the current study indicate that the phonological abilities of children who stutter are associated with white matter integrity along the bilateral dorsal language pathways, which are beyond age-related effects. The children who do not stutter exhibited age-related white matter integrity increases in bilateral dorsal and ventral tracts, while this was attenuated in children who do not stutter.

Overall, major findings from this investigation indicate that: 1) children who stutter do may not exhibit significant differences from children who do not stutter in terms of phonological complexity as measured from spontaneous speech samples, 2) there is greater association between PWP and increased white matter integrity along the bilateral dorsal tracts in children who stutter compared to children who do not stutter , and 3) children who stutter exhibit an overall attenuated age-related white matter integrity growth in areas overlapping with those that showed heightened PWP-white matter integrity associations, indicating that such increased PWP-white matter integrity associations may be critical for children who stutter to produce phonology that is comparable to children who do not stutter. These results provide novel insights into phonological development in children who stutter, and the behavior – neural development associations relevant to phonological development that seem to differ in children who stutter.

Finally, the results of this novel study provide information about white matter language pathways that might be relevant to phonological development in children who stutter. Furthermore, the current findings suggest that increased white matter integrity along the bilateral dorsal language pathway may provide children who stutter with additional support to produce similar phonology scores as children who do not stutter in spontaneous speech. Follow up investigations utilizing a longitudinal study design will need to clarify whether the differences found in this study represent a developmental trajectory difference (e.g., normal, but lagged growth, or altogether distinct trajectories) relevant to phonological development in children who stutter. Furthermore, longitudinal investigations of phonology and structural neuroanatomy may provide additional insights into how the neural bases of phonological development differs between children who stutter and those who eventually recover from stuttering, thereby paving the way to a better understanding of the relationship between phonological development and stuttering.

APPENDICES

APPENDIX A:

PHONE SCREENING QUESTIONNAIRE FOR INCLUSION/EXCLUSION

1. Are you calling to see if your child qualifies for the research study?

2. Is your child right or left-handed?

3. Does s/he have any allergies to medications?

4. What medications does s/he take?

5. Has s/he had any injuries or surgeries to their head or neck?

6. Has s/he had any neurological problems (including tremor/seizure)?

7. Has s/he had or is being treated for any psychiatric illnesses?

8. Has s/he ever lost consciousness or fainted?

9. Was pregnancy and delivery normal? (length of pregnancy, medications, fetal distress during delivery, etc.)

10. Has his/her physical development been normal? (Has met all developmental milestones in a timely manner?)

11. Has s/he been diagnosed with any developmental disorders? (dyslexia, autism, Tourette's syndrome, learning disorder, ADHD)

12. Does your child stutter?
    a. If yes, then how long has s/he been stuttering?

13. IF THE CHILD STUTTERS, has s/he had any therapy for stuttering?
    a. If so, when and what kind of treatment?

14. Does s/he have any family members who stutter?
    a. If so, please list their relations:
    b. Maternal:
    c. Paternal:

15. Does s/he have any other speech-language or hearing difficulties?

16. Does s/he have normal hearing in both ears

17. Has the child had any other speech, voice, or language therapy?

18. Is English his/her first and primary language?

19. Does your child have any foreign language experience?

20. Does s/he have braces or any dental prosthesis?

21. Does s/he have any tattoos or any metal objects in their body?

22. Does s/he feel anxious in close, tight spaces?

APPENDIX B:

CASE HISTORY FORM FOR PARENTS OF CHILDREN PARTICIPATING IN MRI RESEARCH

Date: Child's name: __________________

DOB: ________________________

Parent(s)' names: Mother: ______________________________ Age: ______

Father: _______________________________ Age: ______

General Health/Development

1. Has the child met all of his or her developmental milestones within the typical time range? (speech/language and motor – walking, running, using scissors, etc.)

2. Has the child ever had middle ear infections? If so, when? How often? How were they treated? DO you think that there might have been significant periods of hearing attenuation due to the ear infection?

3. Has your child ever had any concussions resulting in lack of consciousness or fainting?

4. Is he/she doing adjusting and doing well at school?

5. How is your child's temperament? Would you say s/he is easily distracted, or tend to be anxious? Please describe your child's personality.

6. Are there any concerns at this time regarding your child's general health or development?

7. Has the child been seen by a medical professional for any reason?

8. Please list any medication that the child is taking at this time.

Language & Music Training

9. Has your child received any significant musical training?
    a. What instrument/s does s/he play?
    b. Approximately what date did they start/end?
    c. What type of training (private lessons/school/self-taught etc.) have they received?
    d. What is the intensity of their musical training?
    e. About how many hours do they practice per week?

MRI-Related

10. Has your child had any type of surgery? (ask about the possibility of surgical clips, staples, etc. in the body)
11. Does your child have braces or any similar orthopedic fixtures in the mouth?

12. Does your child have piercings in or on the body?
13. What language(s) are spoken at home?

14. Is English his/her first language

15. Has your child learned any foreign language? If so, in what type of context/setting? Is he/she fluent (able to carry on conversation) in that language?

Characteristics of Stuttering & Family History (for children who stutter only)

16. When did your child start to stutter **(onset)**?
    Month/Year_____________________ Age (Years:Months) __________________

17. Would you say that the onset was **SUDDEN** (started during a period within 2 weeks) or **GRADUAL**?
    - ☐ Sudden
    - ☐ Gradual
18. Can you think of any unusual incident that might have co-occurred with your child's onset of stuttering?

19. Describe the stuttering behavior. What types of disfluencies do you notice in your child's speech? (Give some examples by demonstrating a sound-syllable repetition, prolongation, etc.)
    - ☐ PW Repetitions
    - ☐ WW Repetitions
    - ☐ Prolongations
    - ☐ Blocks
    - ☐ Phrase Repetitions
    - ☐ Interjections
    - ☐ Repetitions
20. Does your child have any **TENSION** in the face and/or neck area during stuttering or any other unusual tension or bodily movements that occur associated with stuttering?

21. Is the severity of stuttering **CONSTANT** or do you see **FLUCUATIONS**?

22. Are there any particular situations that make stuttering worse? / better?

23. Is the child AWARE of his/her stuttering? If so, has he/she commented on it?
    - ☐ Yes
    - ☐ No
24. Has the child been TEASED for stuttering? If so, how was this dealt with?.
    - ☐ Yes
    - ☐ No
25. Has the child been seen by a speech-language pathologist, psychologist, or any special education related professional? Has the child ever been formally diagnosed?

26. Are there any family members that stutter **(family history)**?

☐ **<u>Maternal</u>**	☐ **<u>Paternal</u>**	☐ **<u>Sibling</u>**
☐ Mother	☐ Father	☐ Brother
☐ Grandmother	☐ Grandmother	☐ Sister
☐ Great Grandmother	☐ Great Grandmother	
☐ Aunt	☐ Aunt	
☐ Grandfather	☐ Grandfather	
☐ Great Grandfather	☐ Great Grandfather	
☐ Uncle	☐ Uncle	
☐ Cousin	☐ Cousin	

27. Has the child been **TEASED** for stuttering? If so, how was this dealt with?
    - ☐ Yes
    - ☐ No
28. Has the child been seen by a speech-language pathologist, psychologist, or any special education related professional? Has the child ever been formally diagnosed?

29. Has the child ever been through **SPEECH THERAPY**? If so, what type of therapy, from whom (school speech pathologist or a private clinician), for how many hours a week? For how long?
    1. Start of TX: __________ (month/year)
    2. Type of TX: Traditional Other
    3. Frequency of TX: _________________________
    4. Setting: School Private
    5. Still in TX? Yes No
    6. Date TX discontinued ___________________

30. What was the result of the therapy?

31. Is there anything else you would want us to know about your child that we haven't asked so far?

<u>Study Participation</u>

32. Would you be open to us contacting you in the future for any additional participation in research?
    - ☐ Yes
    - ☐ No
33. What are some expectations you have about participating in this research?

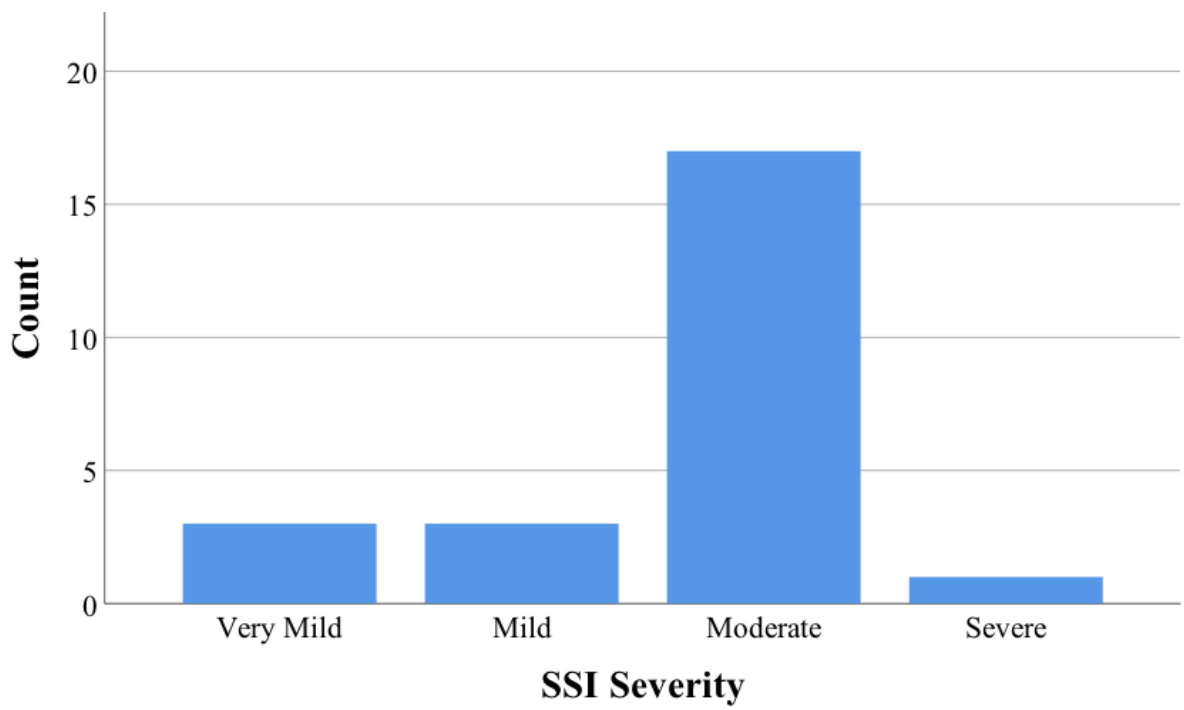
Please Sign Here: ____

APPENDIX C:

STUTTERING SEVERITY FOR CHILDREN WHO STUTTER BASED ON THE SSI-4

Figure 19:

Stuttering Severity for Children Who Stutter Based on the SSI-4



APPENDIX D:

SIGNIFICANT GROUP DIFFERENCES FOR PWP MODULATION OF FA

Table 13: *Significant Group Differences for PWP Modulation of Fractional Anisotropy Including SD*

White matter tracts	Regions (left/right)	Approx BA	Cluster size	max *t*	x	y	z	Mean FA (SD) CWS		Mean FA (SD) CWNS		Cohen's *d*
Cingulum	Cingulate (L)	24	81	4.23	-5	25	-10	0.276	(0.027)	0.268	(0.021)	0.361
AF	IFG (R)	47	47	2.93	32	27	-15	0.241	(0.020)	0.240	(0.016)	0.052
	Insula (L)	13	35	3.25	-35	7	12	0.268	(0.031)	0.265	(0.020)	0.128
	IFG (R)	45	26	3.02	23	25	5	0.297	(0.035)	0.293	(0.015)	0.154
	Insula (R)	13	18	3.39	37	7	10	0.255	(0.016)	0.258	(0.015)	0.183
SLF	PreCG (R)	3a	28	-3.35	30	-17	27	0.475	(0.027)	0.477	(0.033)	0.055

Table 14: *Correlations for Significant Group Differences in PWP Modulation of Fractional Anisotropy*

White matter tracts	Regions (left/right)	Approx BA	Cluster size	max *t*	x	y	z	Pearson's *r* (*p*-value) CWS		Pearson's *r* (*p*-value) CWNS	
Cingulum	Cingulate (L)	24	81	4.23	-5	25	-10	0.430	(0.036)	-0.115	(0.538)
AF	IFG (R)	47	47	2.93	32	27	-15	0.384	(0.064)	-0.044	(0.816)
	Insula (L)	13	35	3.25	-35	7	12	0.339	(0.105)	0.004	(0.982)
	IFG (R)	45	26	3.02	23	25	5	0.158	(0.460)	-0.226	(0.221)
	Insula (R)	13	18	3.39	37	7	10	0.337	(0.108)	-0.013	(0.945)
SLF	PreCG (R)	3a	28	-3.35	30	-17	27	-0.113	(0.598)	0.525	(0.002)

Table 15: *r to Z Transformation Results for PWP Modulation of Fractional Anisotropy*

White matter tracts	Regions (left/right)	Approx BA	Cluster size	max *t*	x	y	z	95% CI		Z	*p*-value
Cingulum	Cingulate (L)	24	81	4.23	-5	25	-10	0.005	0.983	-0.115	(0.538)
AF	IFG (R)	47	47	2.93	32	27	-15	-0.115	0.886	-0.044	(0.816)
	Insula (L)	13	35	3.25	-35	7	12	-0.209	0.809	0.004	(0.982)
	IFG (R)	45	26	3.02	23	25	5	-0.017	0.867	-0.226	(0.221)
	Insula (R)	13	18	3.39	37	7	10	-0.196	0.823	-0.013	(0.945)
SLF	PreCG (R)	3a	28	-3.35	30	-17	27	-1.076	-0.115	0.525	(0.002)

Note. AF = arcuate fasciculus; SLF = superior longitudinal fasciculus; IFG-operc = inferior frontal gyrus; PreCG = precentral gyrus; PWP = proportion of whole-word proximity; BA = Brodmann area; SD = standard deviation

APPENDIX E:

SIGNIFICANT GROUP DIFFERENCES FOR AGE MODULATION OF FRACTIONAL ANISOTROPY

Table 16: *Significant Group Differences for Age Modulation of Fractional Anisotropy Including SD*

White matter tracts	Regions (left/right)	Approx BA	Cluster size	max *t*	x	y	z	Pearson's *r* & *p*-value CWS	CWNS	Cohen's *d*
IC	Caudate (L)	-	123	-4.73	-10	10	-2	0.272 (0.013)	0.267 (0.020)	0.275
Cing [a]	Cingulate (L)	32	27	-4.33	-12	37	22	0.240 (0.014)	0.237 (0.014)	0.209
	Cingulate (L)	32	25	-4.22	-10	20	32	0.334 (0.018)	0.342 (0.022)	0.393
Cing [b]	PHIP (R)	30	61	-4.68	15	-37	-10	0.281 (0.018)	0.287 (0.030)	0.232
	PHIP (R)	28	15	-3.60	17	-25	-25	0.311 (0.015)	0.310 (0.018)	0.042
AF [c]	MTG (L)	39	22	-3.78	-40	-55	2	0.282 (0.012)	0.286 (0.019)	0.274
AF [d]	IFG-operc (R)	44	19	-3.98	52	0	17	0.414 (0.022)	0.422 (0.022)	0.395
IFOF [a]	Putamen (R)	-	21	-4.08	32	5	2	0.241 (0.010)	0.241 (0.013)	0.000
	MFG (R)	10	17	-5.05	35	45	5	0.307 (0.017)	0.311 (0.024)	0.179
ILF	MOG (L)	30/31	28	-4.18	-30	-80	10	0.272 (0.022)	0.272 (0.021)	0.000
UF	Insula (L)	36	37	-3.64	-30	-2	-15	0.399 (0.030)	0.408 (0.027)	0.293

Table 17: *Correlations for Significant Group Differences in Age Modulation of Fractional Anisotropy*

White matter tracts	Regions (left/right)	Approx BA	Cluster size	max *t*	x	y	z	Pearson's *r* & *p*-value CWS	CWNS
IC	Caudate (L)	-	123	-4.73	-10	10	-2	-0.187 (0.382)	0.430 (0.016)
Cing [a]	Cingulate (L)	32	27	-4.33	-12	37	22	-0.530 (0.008)	0.557 (0.001)
	Cingulate (L)	32	25	-4.22	-10	20	32	-0.458 (0.024)	0.728 (<0.001)
Cing [b]	PHIP (R)	30	61	-4.68	15	-37	-10	-0.391 (0.059)	0.517 (0.003)
	PHIP (R)	28	15	-3.60	17	-25	-25	-0.318 (0.130)	0.605 (<0.001)
AF [c]	MTG (L)	39	22	-3.78	-40	-55	2	-0.214 (0.316)	0.652 (<0.001)
AF [d]	IFG-operc (R)	44	19	-3.98	52	0	17	-0.368 (0.077)	0.569 (0.001)
IFOF [a]	Putamen (R)	-	21	-4.08	32	5	2	-0.423 (0.040)	0.621 (<0.001)
	MFG (R)	10	17	-5.05	35	45	5	-0.629 (0.001)	0.406 (0.023)
ILF	MOG (L)	30/31	28	-4.18	-30	-80	10	-0.113 (0.599)	0.645 (<0.001)
UF	Insula (L)	36	37	-3.64	-30	-2	-15	-0.428 (0.037)	0.521 (0.003)

Table 18: *r to Z Transformation Results for Age Modulation of Fractional Anisotropy*

White matter tracts	Regions (left/right)	Approx BA	Cluste r size	max *t*	x	y	z	95% CI		Z	*p* -value
IC	Caudate (L)	-	123	-4.73	-10	10	-2	-1.057	-0.075	-2.249	0.0245
Cing [a]	Cingulate (L)	32	27	-4.33	-12	37	22	-1.401	-0.609	-4.221	<0.0001
	Cingulate (L)	32	25	-4.22	-10	20	32	-1.486	-0.735	-4.916	<0.0001
Cing [b]	PHIP (R)	30	61	-4.68	15	-37	-10	-1.276	-0.393	-3.413	0.0006
	PHIP (R)	28	15	-3.60	17	-25	-25	-1.294	-0.418	-3.570	0.0004
AF [c]	MTG (L)	39	22	-3.78	-40	-55	2	-1.257	-0.368	-3.451	0.0006
AF [d]	IFG-operc (R)	44	19	-3.98	52	0	17	-1.300	-0.429	-3.576	0.0003
IFOF [a]	Putamen (R)	-	21	-4.08	32	5	2	-1.379	-0.557	-4.081	<0.0001
	MFG (R)	10	17	-5.05	35	45	5	-1.359	-0.559	-4.055	0.0001
ILF	MOG (L)	30/31	28	-4.18	-30	-80	10	-1.175	-0.262	-3.049	0.0023
UF	Insula (L)	36	37	-3.64	-30	-2	-15	-1.305	-0.440	-3.586	0.0003

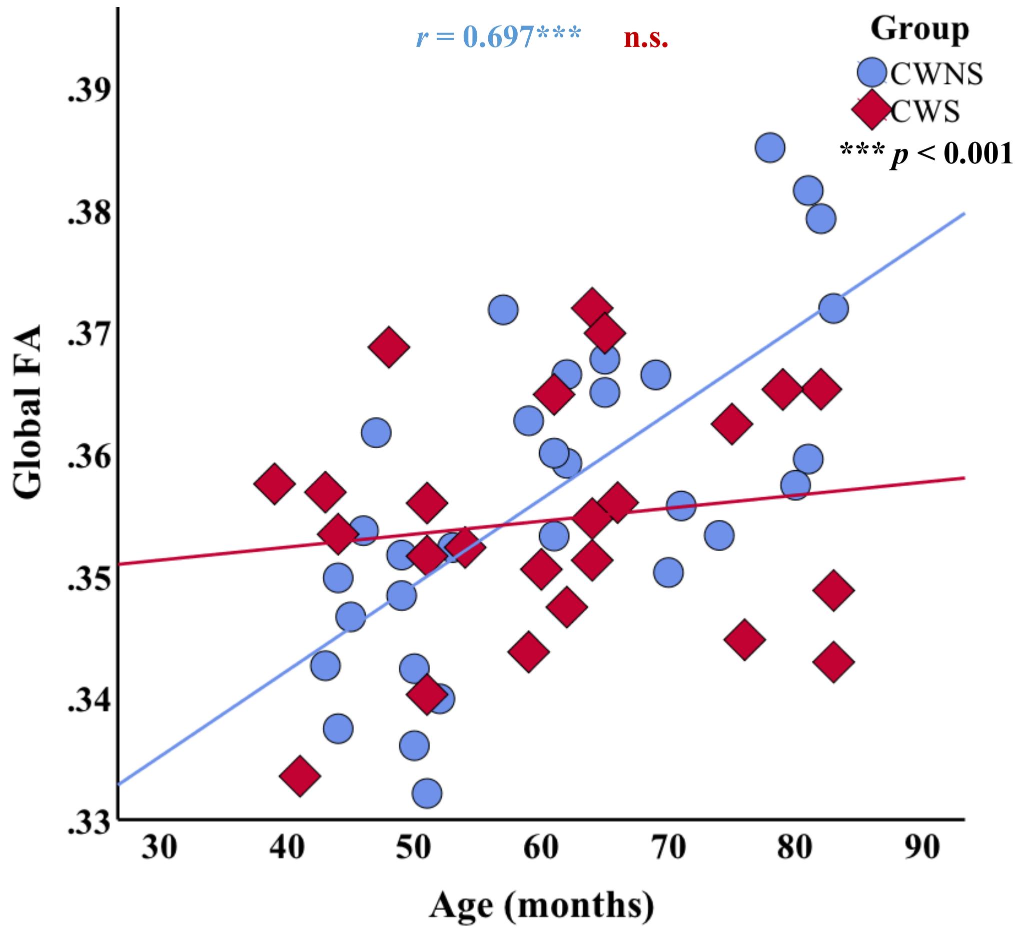
Note. IC = internal capsule; Cing = cingulum; AF = arcuate fasciculus; IFOF = inferior fronto-occipital fasciculus; ILF = inferior longitudinal fasciculus; UF = uncinate fasciculus; PHIP = parahippocampal gyrus; MTG = middle temporal gyrus; IFG-operc = inferior frontal gyrus-opercularis; MFG = middle frontal gyrus; MOG = middle occipital gyrus; BA = Brodmann area; SD = standard deviation; anterior [a]; posterior [b]; long segment [c]; anterior segment [d]

APPENDIX F:

SCATTERPLOT OF GLOBAL FA & AGE FOR EACH GROUP

Figure 20:

Global FA Association with Age for Each Group



Note. Correlation between chronological age and whole-brain FA for children who do not stutter (CWNS) and children who stutter (CWS).

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