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Childhood Stuttering – Where are we and Where are we going?

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Abstract

Remarkable progress has been made over the past two decades in expanding our understanding of the behavioral, peripheral physiological, and central neurophysiological bases of stuttering in early childhood. It is clear that stuttering is a neurodevelopmental disorder characterized by atypical development of speech motor planning and execution networks. The speech motor system must interact in complex ways with neural systems mediating language, other cognitive, and emotional processes. During the time window when stuttering typically appears and follows its path to either recovery or persistence, all of these neurobehavioral systems are undergoing rapid and dramatic developmental changes. We summarize our current understanding of the various developmental trajectories relevant for the understanding of stuttering in early childhood. We also present theoretical and experimental approaches that we believe will be optimal for even more rapid progress toward developing better and more targeted treatment for stuttering in the preschool children who are more likely to persist in stuttering.

Introduction

Our first point related to developmental stuttering circa 2016 is this: Amazing progress has been made in the past two decades in understanding stuttering as a neurodevelopmental disorder. During this period, the fact that stuttering *is a neurodevelopmental disorder* has been clearly established on the basis of many experimental results. Any account of the *cause* of stuttering must explain the neurodevelopmental processes that generate an atypically functioning speech motor system that is vulnerable to breakdowns in the face of increased linguistic, other cognitive, and emotional demands. Major progress has been made on multiple experimental fronts expanding our knowledge of the onset and development of persistent stuttering. Groundbreaking work has been reported in the assessment of behaviors associated with stuttering (e.g.,[1]), physiological characteristics of stuttering near its onset, including linguistic and motor processes ([2], [3]) and in neuroimaging studies documenting the anatomical and functional neural correlates of childhood stuttering (e.g., [4]). These exciting results using many different experimental approaches provide a rich and promising foundation for future work. We believe the next 10 years will bring even greater expansion of our knowledge base concerning developmental stuttering. For the major research areas related to childhood stuttering, we suggest below how current and new experimental approaches are likely to lead to a clearer understanding of how stuttering emerges in early childhood. Also we explore current and future, potential approaches to delineating the factors contributing to the ultimate persistence of or recovery from stuttering.

Finally, we suggest that a critical future challenge for stuttering researchers and clinicians is to use our multileveled understanding of how stuttering emerges to treat young children who are stuttering. Our knowledge of the behavioral, peripheral physiological, and central neural correlates of stuttering ultimately will provide the optimal basis to generate new therapeutic approaches for young children. We envision a future in which the efficacy of early treatment for stuttering will be established both via behavioral and neurophysiological assessments. For example, we predict that newer, less invasive functional imaging techniques, such as fNIRS, will be used to determine if ongoing treatment is enhancing the development of more typical interactive neural networks supporting speech production.

Genetics/Epigenetics

We have known for many years that stuttering runs in some families. Families share genes and environments. Work from Dennis Drayna's laboratory has revealed that genes related to stuttering transmission can be identified within family trees with a high density of stuttering individuals ([5]). However, within different family trees, the genes related to stuttering vary, so that there is no single or consistent genetic basis for the propensity to stutter. Also it is clear that genes do not *cause* stuttering. Humans with identical genes at birth, that is monozygotic twins, are not destined to share the trait of stuttering or not stuttering. In fact monozygotic twins are only about 50% to 60% concordant for stuttering ([6,7]), which means that if one twin stutters, there is a 50 to 60% chance that his/her twin also stutters. Also, MZ twins may both begin to stutter, and one may recover while the other does not. Infants are not born programmed to stutter; rather stuttering behaviors emerge during development.

In recent years developmental scientists have increased focus on understanding epigenetic processes. Epigenesis refers to the way in which genes are expressed, including both the timing and intensity of their expression. The ultimate phenotype stuttering/not stuttering is a product of genetic and environmental interactions. Epigenesis is influenced by environmental factors, and therefore epigenetic processes provide the bridge between genes and environment ([8]). Future work on the genetic/epigenetic basis of stuttering will focus not only on the genes that are involved in transmission across generations of a family, but also on the way in epigenetic processes influenced by environmental factors contribute to a higher probability of the emergence of persistent stuttering.

Neural and Motor Correlates

Over the past two decades there have been hundreds of neuroimaging studies of adults who stutter, and more recently a few studies of children who are stuttering (e.g., [4]). Studies have examined anatomical differences between stuttering and normally fluent individuals and functional differences in brain activity during performance of a variety of tasks and at rest. This literature has produced many conflicting results both in the anatomical and functional details of the neural bases of stuttering. However, common to many investigations is the finding of atypical connectivity and functioning among left inferior frontal areas usually specialized for speech production and other areas involved in auditory and linguistic processing.

Thus these investigations clearly tell us that stuttering does not "live in" an isolated area of the brain. The neural networks implicated in stuttering are widespread. This makes sense, because speech production requires complex communications between many different parts of the brain. Fundamentally we know that stuttering is a disorder of speech production. The cardinal symptom of stuttering is a failure of the speech motor system to generate the neural commands to muscles necessary to produce flowing, fluent speech. Thus a key to understanding how stuttering develops is likely to arise from future work to determine how the complex neural nets active during language formulation/speech production develop the atypical connections and patterns of activation that result in stuttering.

From recent work in children as young as 3 to 5 years, there is now convincing evidence that atypical patterns of neural connectivity and activity characteristic of stuttering are already present near the onset of stuttering ([4]). Functional outcomes of these atypical neural growth patterns are also observable in the speech motor output of preschool children. Children who are stuttering at 4 to 5 years of age (many of whom will ultimately recover) differ in their speech motor characteristics, such that their speech motor coordination patterns and basic speech movement characteristics are immature relative to their normally fluent peers ([2]). Compared to girls, preschool boys who are stuttering show greater lags relative to their peers on speech motor development. Sex differences have also been documented in speech-related brain regions in boys and girls who stutter ([4]). These novel findings provide insight into a well known phenomenenon: Girls recover from stuttering at a significantly higher rate than boys. In the future such findings should be useful in establishing better treatment approaches for boys whose vulnerability to persistence of stuttering is higher.

One very promising technology for future stuttering research is functional near infrared spectrography (fNIRS). This is a noninvasive brain imaging method that can be used in infants and young children. The system involves the use of laser emitter and detector pairs, and it estimates the blood oxygenation level in targeted brain areas, and like fMRI is therefore an indirect measure of neural activity. A major advantage of fNIRS compared to other methods is that, in addition to being applicable in young children, it can be used during normal speech production. Past studies using fMRI and other methods for brain imaging have been limited to the investigation of single word production due to problems with movement artifact. Our colleague, Bridget Walsh, has just completed the first study using fNIRS to record neural activity during natural speech production (picture description task) in children ([9]). Her results are striking-- showing that school-age CWS have qualitatively different patterns of activation over left inferior frontal areas compared to CWNS, suggesting that speech-related left hemisphere functions are atypical in school-age CWS.

Language

In the language domain, children who stutter present with a wide range of profiles on standardized tests of linguistic proficiencies. One cannot make any generalization about "the" language abilities of stuttering children. In preschool children who are stuttering, there is a higher incidence of phonological disorders (e.g., [10]). In the Purdue Stuttering Project, the majority of CWS who have participated display speech and language skills within the normal range; however, of 130 preschool children who stutter recruited into the project, approximately 40% have scored at least 1 SD below the mean on phonological skills (BBTOP), and 20 % have were low (at least 1SD below mean) on a test of syntactic and morphosyntactic proficiency (SPELT-3). From a neurodevelopmental perspective, we know that rapid maturation of the speech motor system's capacities for coordinating fluent speech production ([11]) occurs during a period of rapidly expanding language ([12]) and phonological ([13]) abilities. The relationships between language proficiency, even within the normal range, and stuttering have long been and continue to be, recognized as a key for determining how language demands contribute to the development and maintenance of stuttering.

A significant advance in our understanding of linguistic processing and stuttering comes from results of investigations of children's brain activity during listening and reading tasks (in which no speech production is required). In these studies we have excluded CWS with frank language or phonological delays. This has allowed us to determine whether brain activity associated with stuttering is either delayed in maturation (i.e., is more similar to a younger child's responses in pattern) or atypical in some way (e.g., different in hemispheric distribution or in the presence/absence of specific ERP components). From these studies, it has become clear that for some aspects of language processing at specific times in development, CWS lag behind their normally fluent peers (CWNS). For example, CWS (who persisted) at 6-7 years displayed less mature ERP patterns for processing syntactic violations, and their ERPs resembled those recorded in 3-5 year old children in an earlier study ([14]). In a study of visual rhyme processing, compared to their peers 9-12 year old CWS showed immature ERP components associated with verbal working memory and silent rehearsal ([15]). As an example of atypical, rather than delayed neural activity, 4-5 year old CWS produced a right-lateralized response while processing syntactic violations, while CWNS showed a clearly left lateralized effect ([3]).

We have also been able to examine neural processing in linguistic tasks in older children who have recovered from stuttering (CWSr) and who have persisted (CWSp). In a study of phonological processing, 7-8 year old CWSr showed a right-lateralized ERP response while CWNS exhibited a bilateral response ([16]). Further, this particular component, typically linked to phonological processing, was absent in CWSp aged 7-8 years. Finally, it is important to note that the neural indices of delayed or atypical language processing are windows or snapshots taken at specific developmental time points. Our work indicates that differences in processing found at a particular age for a specific aspect of linguistic processing may not be observed at another developmental time point. For example, an ERP component that reflects response preparation (CNV) was markedly immature in 9-12 year CWS compared to CWNS, but this component was identical in AWS and fluent controls. These findings highlight the dynamic nature of the neurodevelopmental course of language and phonological processing, and these dynamically changing neural networks are co-developing with rapidly changing speech motor control networks. This is further evidence that stuttering arises when there are maturational lags or atypical development in multiple neural networks involved in language processing and speech production.

Emotion/Temperament

It is well-established that there is no “stuttering personality” characteristic of adults, but adults who stutter do have, understandably, higher levels of anxiety about communication ([17]). Historically clinicians and scientists suggested that near its onset, stuttering in young children was not associated with anxiety or other negative emotions and that such emotional reactions arose later in childhood, in the school-age and teenage years. Recent work, however, suggests that preschool children who are stuttering are aware of their stuttering behaviors and that they react negatively to them ([17]).

There also has been notable attention to the question of whether stuttering children tend to have certain temperamental characteristics, including lower, or in some cases, higher scores on temperamental dimensions such as self-regulation, inhibitory control, and emotional reactivity ([18]). Findings of behavioral studies of temperamental characteristics of stuttering children are mixed, as are results from a few investigations in which physiological indices of autonomic arousal have been used to investigate early childhood stuttering. Therefore, at this point in time, potential early emotional factors related to stuttering are unknown. Results of earlier studies and the clear clinical evidence of the importance of the feelings and attitudes of individuals who stutter suggest that this is an area well worth detailed exploration with both behavioral and physiological measures. Walden and colleagues ([19]) have proposed an interesting model of potential interactions between emotional and linguistic factors underlying stuttering, the Dual Diathesis-Stressor Model. This is an important step in the direction of assessing critical *interactions* among the complex, developing, behavioral systems involved in childhood stuttering.

Future Theoretical and Experimental Directions

What causes stuttering? We are now at a point in time when the answer to this question no longer must be “we don’t know.” It is now widely accepted that stuttering is a multidimensional disorder. It is also widely accepted that stuttering is a neurodevelopmental disorder, which means that it arises during development in childhood. We have made the case in recent presentations and in a manuscript now in preparation that stuttering should be viewed within a multidimensional, developmental, dynamic framework. We have called our theory Developmental Pathways to Stuttering (DPS). The central tenet of DPS is that to understand the development of stuttering, we must focus not solely on the developmental trajectory of stuttering behaviors, but on co-developing systems that potentially interact in critical ways with neural networks involved in speech production. During the time window when stuttering emerges, and recovery or persistence occurs, approximately 2 to 6 years of age, there are dramatic, rapid changes in many behavioral systems and the underlying neural processes supporting them. For example, there are remarkable changes in many dimensions of temperament, in language abilities, and extremely rapid improvement in speech motor skills occur. Thus, we argue it is critical to investigate stuttering within the developmental *context* within which it begins and follows its ultimate course to persistence or recovery.

Within the DPS framework, it becomes clear that finding group differences between stuttering and non-stuttering children or adults will not lead to an understanding of the causes of stuttering. Rather our conceptualization of the course of development of stuttering firmly recognizes the heterogeneity of pathways to stuttering onset and to ultimate recovery or persistence. There are common underlying factors that contribute to the emergence of stuttering: atypical development of speech motor planning and execution networks and the interaction of speech motor systems with networks mediating linguistic, emotional, and other cognitive processes, such as self-regulation. But the relative weights of various factors involved in stuttering vary among different individuals. Therefore it is essential to understand a particular child's multidimensional profile on the developmental trajectories likely to be related to stuttering.

Taking this theoretical approach, an important goal of future research becomes clear. We need much better tools to assess preschoolers who are stuttering to determine their risk of persistence. Research is needed to determine which factors can be used in a multifactorial equation to give us an estimate of likelihood of persistence in preschoolers who are stuttering; just as adults are given estimates of the probability of cardiovascular incident based on our body mass index, blood pressure, etc. Developing such a diagnostic battery is currently the goal of the Purdue Stuttering Project. Not only would such a tool help us to determine which children should have a high priority for early intervention, the multidimensional profile would aid in designing an optimal treatment approach. A child who is stuttering who has a concomitant phonological or language delay or disorder would likely benefit from a different treatment strategy than a child who scores within normal limits on tests of linguistic proficiency.

Also apparent from the theoretical perspective we have outlined is a real need for longitudinal investigations of stuttering in early childhood. This experimental approach will allow assessment of which characteristics present near the onset of stuttering aid in predicting persistence. For example in our project, we have preliminary evidence that early delays in speech motor skills and relatively low scores on nonword repetition tasks are early predictors of later persistence ([20]). In addition, cross-sectional studies of groups of children who are stuttering and matched, normally fluent peers must be conducted with larger numbers of participants than the numbers often included in investigations of stuttering. A clear example of the pitfalls of relatively small n studies of stuttering comes from our project. In a study of timing control in a bimanual clapping task in preschool children who stutter, we came to the wrong conclusion. Initially, we were excited about the potential diagnostic value of results of our first study ([21]) showing that 60% of 17 CWS had poorer clapping abilities than any of the 13 children who were not stuttering. We hypothesized that poor bimanual clapping skills signaled a generalized motor timing deficit in some children who stutter and that this might be predictive of persistence.

We followed up with much larger samples (70 CWS and 45 CWNS) and included longitudinal data and data from CWS grouped by ultimate persistence or recovery ([22]). Our results did not replicate the earlier finding. We did not find differences in timing performance between the preschool aged groups of CWS and CWNS; nor did we find that there was a large group of CWS whose timing control was outside the normal range. In fact, the children's abilities to "keep the beat" while clapping were identical for CWS and CWNS. In addition, we found no evidence that the children who ultimately persisted in stuttering were among the poor clappers when they were tested at age 4 or 5. We were misled due to undersampling the two populations in the initial report. Olander et al., by chance, included a disproportionate number CWNS who were better at the clapping task and in contrast, a large number of CWS who were less proficient at it. We note that the sample sizes in Olander et al. are traditionally viewed as reasonable for this type of research. The take-home message from our experience with the clapping experiments is that heterogeneity among children who stutter is real. We all know this, but we are guilty of publishing results and drawing conclusions about stuttering children as a group on the basis of data from relatively few participants. Obviously, it is extremely difficult for one investigator to recruit 50 or more stuttering children and matched controls. The future clearly calls for large-scale, multi-site, cross-sectional and longitudinal investigations of stuttering preschoolers and their matched controls.

Concluding Comments

Millions of children around the world start stuttering between the ages of 2 to 5 years. Fortunately, approximately 80% of these children will recover with or without treatment. The remaining 20% are in need of a better early predictor of their likelihood for persistence. Finding such a clinical tool should be a major goal for researchers and clinicians who work on stuttering. If we can treat the children most likely to persist with protocols that are specifically designed for their profile of lagging, advanced, or typical developmental trajectories, we have the best chance to help them recover. Recovery from stuttering means that the brain has established neural networks for speech motor planning and execution that are not unusually vulnerable to the demands of linguistic complexity, various speaking environments, or emotional state. From a neurodevelopmental perspective, our best opportunity to enhance the pathway to recovery from stuttering is during this highly dynamic developmental time window. We believe we have a great opportunity moving forward from 2016 to make substantive progress towards the goal of evaluating the risk of each child who is stuttering for persistent stuttering and devising more comprehensive and tailored treatment approaches to effectively address the individual profiles of these children. We predict that dramatically improved diagnostic and treatment strategies for preschoolers will contribute to a significant increase in the rate of recovery from stuttering.

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REFERENCES

1. Ambrose NG, Yairi E. Normative disfluency data for early childhood stuttering. J Speech Lang Hear R. 1999; 42(4):895–909.
2. Walsh B, Mettel KM, Smith A. Speech motor planning and execution deficits in early childhood stuttering. J Neurodev Disord. 2015; 7(1) doi: 10.1186/s11689-015-9123-8.
3. Weber-Fox C, Hampton Wray A, Arnold H. Early childhood stuttering and electrophysiological indices of language processing. J Fluency Disord. 2013; 38(2):206–221. [PubMed: 23773672]
4. Chang SE, Zhu DC, Choo AL, Angstadt M. White matter neuroanatomical differences in young children who stutter. Brain. 2015; 138(3):694–711. [PubMed: 25619509]

5. Drayna D, Kang C. Genetic approaches to understanding the causes of stuttering. J Neurodev Disord. 2011; 3(4):374–380. [PubMed: 21850444]
6. Felsenfeld S, Kirk KM, Zhu G, Statham DJ, Neale MC, Martin NG. A study of the genetic and environmental etiology of stuttering in a selected twin sample. Behav Genet. 2000; 30(5):359–366. [PubMed: 11235981]
7. Howie PM. Concordance for stuttering in monozygotic and dizygotic twin pairs. J Speech Hear Res. 1981; 24:317–321. [PubMed: 7197739]
8. Petronis A. Epigenetics as a unifying principle in the aetiology of complex traits and diseases. Nature. 2010; 465:721–727. [PubMed: 20535201]
9. Walsh B. Illuminating the speaking brain: An fNIRS investigation of speech production in children who do and do not stutter. In preparation.
10. Paden EP, Yairi E, Ambrose NG. Early childhood stuttering II: Initial status of phonological abilities. J Speech Lang Hear R. 1999; 42(5):1113–1124.
11. Smith A, Zelaznik HN. Development of functional synergies for speech motor coordination in childhood and adolescence. Dev Psychobiol. 2004; 45(1):22–33. [PubMed: 15229873]
12. Miller JF, Chapman RS. The relation between age and mean length of utterance in morphemes. J Speech Lang Hear R. 1981; 24:154–161.
13. Rvachew, S., Brosseau-Lapré, F. Developmental Phonological Disorders: Foundations of Clinical Practice. Plural Publishing; San Diego, CA: 2012.
14. Usler E, Weber-Fox C. Neurodevelopment for syntactic processes distinguishes childhood stuttering recovery versus persistence. J Neurodev Disord. 2015; 7(1) doi: 10.1186/1866-1955-7-4.
15. Weber-Fox C, Spruill JE III, Spencer R, Smith A. Atypical neural functions underlying phonological processing and silent rehearsal in children who stutter. Developmental Sci. 2008; 11(2):321–337.
16. Mohan R, Weber C. Neural systems mediating processing of sound units of language distinguish recovery versus persistence in stuttering. J Neurodev Disord. 2015; 7(28) doi: 10.1186/s11689-015-9124.
17. Iverach L, Menzies RG, O'Brian S, Packman A, Onslow M. Anxiety and stuttering: continuing to explore a complex relationghip. Am J Speech Lang Pathol. 2011; 20(3):221–232. [PubMed: 21478283]
18. Vanryckeghem M, Brutten G, Hernandez L. The KiddyCAT: A normative investigation of stuttering and nonstuttering preschoolers' speech-associated attitude. J Fluency Disord. 2005; 30:307–318. [PubMed: 16246410]
19. Jones R, Choi D, Conture E, Walden T. Temperament, emotion and childhood stuttering. Semin Speech Lang. 2014; 35(2):114–131. [PubMed: 24782274]
20. Walden TA, Frankel CB, Buhr AP, Johnson KN, Conture EG, Karrass JM. Dual diathesis-stressor model of emotional and linguistic contributions to developmental stuttering. J Abnorm Child Psychology. 2012; 40(4):633–644.
21. Spencer C, Weber-Fox C. Preschool speech articulation and nonword repetition abilities may help predict eventual recovery or persistence of stuttering. J Fluency Disord. 2014; 41:32–46. [PubMed: 25173455]
22. Olander L, Smith A, Zelaznik HN. Evidence that a motor timing deficit is a factor in the development of stuttering. J Speech Lang Hear R. 2010; 53:876–886.
23. Hilger A, Zelaznik HN, Smith A. Evidence that a motor timing deficit is not a factor in the development of stuttering. J Speech Lang Hear R. in press.