WHAT FACTORS CONTRIBUTE TO THE EMERGENCE OF STUTTERING?
What causes stuttering?

• Amazing progress over past few decades

• Many of us very excited about all the new work from clinicians and researchers

• Outline:
  – My answer to the question at hand (preamble to that, levels of explanation)
  – Three major domains of inquiry relevant to stuttering
  – Discuss complex developmental pathways to stuttering, the Developmental Dynamics Model
What causes stuttering? A Caveat to Start...

Wrong question to ask with regard to a complex highly heterogeneous human behavior.

By asking “what causes stuttering?” we encourage a linear thinking perspective-
A cause, a time before stuttering was caused
Then something happened and stuttering started

Better question “what are the factors that contribute to the emergence of stuttering?”

Developmental Stuttering reflects just that- A Developmental Process. It does not exist at birth, rather it emerges in the preschool years due to complex interactions of speech motor and other neural systems.
Stuttering is a neurodevelopmental speech production disorder with typical onset between the ages of 2 and 5 years. Neurodevelopmental disorders arise during childhood due to atypical development of the central nervous system. The primary symptoms of stuttering are involuntary disruptions in the forward flow of speech. These observable disruptions in fluent speech production clearly reflect an underlying speech motor disorder, but also critical in the development and persistence of stuttering are the developing neural networks mediating language and psychosocial processes and their interactions with speech production systems. (Smith and Weber, in progress).
From this statement, clear that different levels of explanation are required.

- One level that is relevant for understanding all complex human behaviors and/or disorders, from diabetes to stuttering. At this level, we need to understand that human behaviors arise from an interaction of genes, the environment (internal and external), and epigenetic processes.

- Another level-specific to stuttering, 3 major domains of inquiry relevant to understanding the onset in development of stuttering: motor, linguistic, and psychosocial.
  - describe some of the critical new data concerning these domains in preschool and school-age children who stutter.
  - discuss how the interaction of complex developmental trajectories in these domains potentially leads to recovery from or persistence of stuttering.
Recent Advances - Multifactorial, *Neurodevelopmental* Disorder – Emerges through Epigenetic Processes

What is a neurodevelopmental disorder?

Arises during childhood due to atypical growth and development of the CNS

Neurodevelopmental disorders include autism, Fragile X Syndrome, Turner Syndrome, 22q Deletion Syndrome, Prader-Willi Angelman Syndrome, Williams Syndrome, dyslexia, and specific language impairment. **And STUTTERING**

We know stuttering is a neurodevelopmental disorder, because there are consistent anatomical and functional differences in the brains of AWS and CWS. During disfluent speech the brain is not generating the correct command signals to the muscles involved in speaking.

**Leads to the questions: how does brain development typically occur? And what is atypical in the development of stuttering?**

Brain Development – Genes, Epigenesis, Experience

• Structure of the brain at any point in time is a product of interactions among genetic, environmental, and epigenetic factors (Lenroot & Giedd, 2006).
  – Genes – keys on the piano, fixed at birth
  – Environment = both outside environment and the internal physiological milieu
  – Epigenetics – timing and intensity of gene expression throughout life, the “playing the piano keys” influenced by the environment.
Brain “Wiring” - Synaptic Density & Start of Active Pruning or “Fine Tuning” of Brain Functions

Adapted from Huttenlocher & Dabholkar, 1997, *Journal of Comparative Neurology*
Between 3 – 72 months

Dendritic branching growth spurts and retractions

Left & Right Hemisphere follow different growth patterns

At 3 months Right speech pre-motor > Left

At 1 year patterns reverses

By 42-72 months, Left Broca’s has structural dominance in dendritic branching.

Great deal of evidence point to atypical anatomy and function of pre-motor and motor speech areas in CWS and AWS.

Simonds & Scheibel, 1989, Brain and Language
Typical and Atypical Epigenesis

Typical Development, Same child at
4 months  4.6 yr  10.8 yr

- Neonate cortex does not have localized functions at birth, allows interaction with the environment and epigenetic processes to play a crucial role in gene expression and in the ultimate phenotype (stuttering vs. not stuttering). Epigenesis is not predetermined, rather it is probabilistic (Karmiloff-Smith, 2007).

- Synaptic pruning leads to specialization, and in **atypical epigenesis, specialization is compromised**. Therefore one should see more widespread atypical activity.

- this is the case in stuttering.

Atypical epigenesis
Annette Karmiloff-Smith

*Developmental Neuroscience*

*Developmental Science 10:1 (2007), pp. 84-88*
Atypical Epigenesis

- Stuttering does not emerge due to an impaired speech motor module in the brain, or an impaired language center, or an impaired auditory monitoring system. All of these systems interact dynamically as the brain is developing speech motor skills.

- Stuttering emerges as these areas undergo synaptic pruning and grow in interconnectivity. This ongoing remodeling of the brain is \textit{experience-dependent}.

- The probability of a child developing persistent stuttering changes over the pre-school years.

The dorsal pathways of the language network in newborn infants, 7-year-old children, and adults. For newborns (A), no connection between the IFG and temporal regions is observed. Rather they only show a connection terminating in the premotor cortex.

Dorsal and ventral pathways in language development

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Twin Studies — Clues about the interactions of genes and environment and the role of epigenesis

Two persons with identical genes (MZ twins) may develop different phenotypes, e.g., one may stutter and one may not.

*This points to the critical role of epigenetic factors.*

Genes are not destiny. Child is born with some probably of stuttering based on his/her genes.
Twin Studies-
Evidence from Concordance Rates for MZ vs. DZ Twin Pairs

Adapted from P. Miller (2012) National Geographic
Three Major Domains of Inquiry (and their interactions) Critical for Understanding Stuttering

- **Stuttering is a speech motor disorder. If the motor system does not produce atypical fluency patterns, it is not stuttering. Occurs only when individual is speaking, and speaking is always linked to language – not found in sign languages, though SLI.**

- **But**... its “causes” have been located in many different systems, from linguistic to emotional.

- Easy to understand why (Other systems studied because they clearly affect the nature and frequency of motor disruption), but involuntary disruptions in speech motor control processes are **the essential, defining symptom of stuttering.**
Typical Development: Interarticulatory Coordination Indices. Oral motor Coordinative space gets smaller with maturation. Articulatory patterns become highly stable and overlearned motor programs. New evidence that CWS lag TD peers in speech motor development. And AWS have relatively unstable motor programs for speech both from articulatory kinematic studies and CNS functional studies.
From earlier work (Smith & Zelaznik, 2004) know that typically developing 4 and 5 year old boys lag girls on the lip aperture variability index.

Sex has a significant effect on CWS as well, with CWS boys showing more variability than CWS girls at age 4-5 years.

One factor for greater vulnerability of males to persistent stuttering.

Journal of Neurodevelopmental Disorders, 2015
Speech motor planning and execution deficits in early childhood stuttering. Walsh B\textsuperscript{1}, Mettel KM\textsuperscript{2}, Smith A\textsuperscript{1}
Longitudinal Analysis of Simple Sentences
Cross Sectional Analyses based on recovery status

Averaged Simple Sentence Data from 4-5 Year Olds

Averaged Simple Sentence Data from 7-8 Year Olds

From Walsh & Smith, In preparation.
Influence of Length and Syntactic Complexity on Speech Motor Performance

One of the critical features of the neurodevelopmental, epigenetic account of stuttering is the idea - in developing specialized regions/connections for speech/language functions, neural systems are highly interactive. Thus the imprint of a ND disorder is not confined to one brain region. Thus my colleague Chris Weber conducted a series of experiments on neural processing of language in AWS and CWS in tasks that required no speaking.
AWS exhibited reduced negative ERPs to function words (N280), content words (N350), and semantic anomalies (N400) compared to AWNS (Weber, 2001).

Clear evidence that even when AWS do not speak, their brains have atypical language processing functional networks. Again, is this an early component of dev. Stuttering?
Preschool CWS Display Syntactic P600 Effect over Right Hemisphere

For CWNS difference in ERP for Syntactic violation vs. correct sentence observed LH. For CWS, it is in the RH.

Preliminary evidence of laterality differences in language processing CWS already at age 4-5 years.

Weber-Fox, Hampton Wray, & Arnold, 2013, Journal of Fluency Disorders
Phonological Processing in 7-8 Year Olds
Recovery vs. Persistence

All Groups Show Robust Central-Parietal ERP Rhyme Effect, N400 larger for non-rhyme relative to rhyme
Rhyme facilitation distinguishes 7-8 year old children who have either recovered or persisted in stuttering.

The groups differed on ERPs over frontal areas; left differences in latency of effect, right differences in amplitude.

3. Emotion. To determine if preschool CWS differ from their normally fluent peers on measures of temperament. We will test the hypotheses that high or low scores on certain temperamental dimensions and a high degree of sympathetic reactivity to the child's own stuttering behaviors are associated with an increased probability of persistent stuttering.

*We include the KittyCat in our test battery and The Children's Behavior Questionnaire to assess temperament and physiological measures of ANS arousal during speech and nonspeech tasks.*

These Data Suggest....

- Clearly there are some remarkable behavioral and physiological differences between 4 and 5 year-old CWS and their ND peers.

- Evidence that the stuttering behavior we observe reflects atypical development of the brain, including motor and language networks.

- Stuttering is a symptom in young children of some marked differences in the way the brain is forming neural connections during development.

- At this age, the ultimate phenotype is unknown, 50% will recover. Atypical neural structures and functions can be compensated for – recovery.
Such findings lead us to propose:

*Developmental Dynamics Model of Stuttering*

- Typical onset 22-60 (mean = 33) months when dramatic and asynchronous neurodevelopment occurs across many different neural systems.

- Nature of onset is highly variable, 40% of cases have sudden (1-3 days) onset, 32% intermediate (1-2 weeks), 27% gradual (3-6 weeks) from Yairi et al.

- Question is, why does stuttering start? Why does it continue in some cases, and not in others?

- What are potentially important factors influencing neurodevelopment when stuttering typically starts? Why does it start when it does, after generally typical developmental history? *One of our major goals in specifying this DDM is to contextualize the emergence of stuttering within a more general developmental framework.*
Stuttering Onset and Recovery

Adapted from Yairi & Ambrose, 2005 and Reilly et al., 2013, *Pediatrics*
Stuttering Onset Coincides with Rapid MLU Growth

Adapted from Miller & Chapman, 1981, *JSLHR* and Rice et al., 2010, *JSLHR*

Dramatically Increasing Demands for Linguistic Processing and Motor Planning and Execution
Stuttering Onset and Recovery Period Contains Very Rapid Changes in Children’s Abilities to Inhibit Behavioral Responses

![Graph showing percent correct inhibition trials vs age in months.]

Development of self-regulatory systems is also changing rapidly in the preschool years. One dimension of temperament that may be important in the development of stuttering, as suggested by Choi, Conture and their colleagues is behavioral inhibition. Ability to inhibit behavioral responses, in this case in a Simon Says task, 36 month old children correctly inhibited their behavioral responses just 22% of the trials, while children just 1 year older inhibited correctly with 91% accuracy.

Adapted from Jones, Rothbart, & Posner, 2003, The Development of executive action in preschool children. *Developmental Science {elephant/bear simon says task}*
Stress on preschooler with unstable and limited speech production and language networks.

If demands > capacities, triggers compensatory responses. If occurs frequently, can change the structure of the brain over time.

This change may be adaptive-recovery.

Or if the mismatch between capacities and demands is too great, the compensations may lead to greater atypical growth pattern (PSD).

DEVELOPMENTAL DYNAMICS MODEL OF STUTTERING

RECOVERY 80%

PERSISTENCE 20%
Implications of Developmental Dynamics Model

- Dramatic and asynchronous neurodevelopmental changes across neural systems, with significant individual differences

- Must look across domains for each child to ascertain individual profile in various domains, language, motor, psychosocial.

- Stuttering emerges when the speech motor system produces atypical disfluencies, SLDs, reflecting unstable neural commands for speech. With development, goal is to help CWS to find fluent speech coordinative operating space. Most do!
What Causes Stuttering?

• For Parents,
  – Talking is a complicated process that requires the brain to do many things at once, from thinking to activating the muscles for speech. We humans are not born with nervous systems that do these things effortlessly (as we do when we are adults). Each child has to grow the nerve connections between many different brain areas and then ‘prune’ or ‘fine-tune’ those connections before speech becomes effortless and automatic – just like they have to learn to ride a bike. When the child is 2-3 years old and just learning to say longer utterances (along with learning a multitude of other things!), many different systems in the brain have to cooperate, translating thoughts and emotions to speech. This requires intricate timing. Some preschoolers start stuttering during this time, and for most, the neural systems come back into synergy, and the stuttering stops. Others have more difficulty and need therapy to help them find ways to produce fluent speech.
Purdue Stuttering Project

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http://www.purdue.edu/stutteringproject/