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Vanderbilt SLP Journal Club: Executive Functions and Childhood Stuttering

Hatun Zengin-Bolatkale, PhD, CCC-SLP
&
Robin Jones, PhD, CCC-SLP

Moderated by:
Amy Natho, MS, CCC-SLP, CEU Administrator, SpeechPathology.com

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Vanderbilt SLP Journal Club: Executive Functions and Childhood Stuttering

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Disclosures

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- Non-financial disclosures:
  - None

7

CONTINUED

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Learner outcomes

1. Participants will be able to define and describe the core and higher order executive functions, with a focus on aspects of inhibitory and attentional control.
2. Participants will be able to explain how various components of executive functions relate to childhood stuttering.
3. Participants will be able to describe potential implications of executive functions for the clinical assessment and treatment of childhood stuttering.

8

CONTINUED

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Purpose

- The purpose of this presentation is to provide a brief overview on the construct of executive functions, in particular those that may relate to childhood stuttering.
- Empirical evidence on the role of executive functions in childhood stuttering as well as clinical implications for assessment and treatment will be discussed.

Key acronyms:

- CWS—Children who stutter
- CWNS—Children who do not stutter

9

CONTINUED

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Articles

- *Article 1*: Executive functions (Diamond, 2013)
- *Article 2*: Temperament dimensions in stuttering and typically developing children (Eggers, De Nil, Van den Bergh, 2010)
- *Article 3*: Attention regulation in young twins with probable stuttering, high nonfluency, and typical fluency (Felsenfeld, van Beijsterveldt, Boomsma, 2010)
- *Article 4*: Traits of attention deficit/hyperactivity disorder in school-age children who stutter (Donaher & Richels, 2010)

10

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Overview of talk

- **Executive functions:** What are they? How are they related to temperament? How may they relate to speech and language? (Article 1 & 2)
- **Executive functions, temperament, and stuttering:** One approach to measure executive functions and results in young children who stutter (Article 2)
- **Attention regulation and stuttering:** Examine the relation between attention and speech fluency as well as etiological overlap between attention and fluency (Article 3)
- **ADHD traits and childhood stuttering:** Are CWS more likely to exhibit ADHD symptoms? (Article 4)
- **Clinical implications of executive functions and stuttering**

11

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Article 1: Executive functions

Diamond, A. (2013). Executive Functions. *Annual Review of Psychology*, *64*(1), 135–168. https://doi.org/10.1146/annurev-psych-113011-143750

12

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Purpose

- The purpose of Diamond (2013) article is to provide an overview of core executive functions and a discussion of
  - the developmental progression of executive functions
  - the relation between executive functions
  - what impacts executive functions and whether executive functions can be improved with practice

13

CONTINUED

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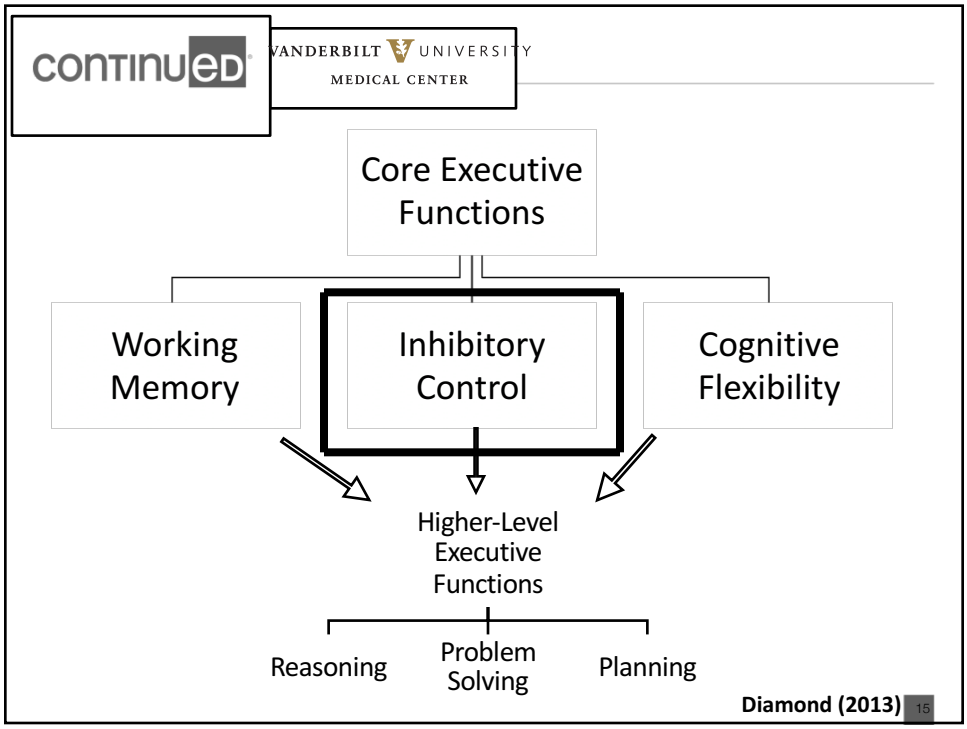
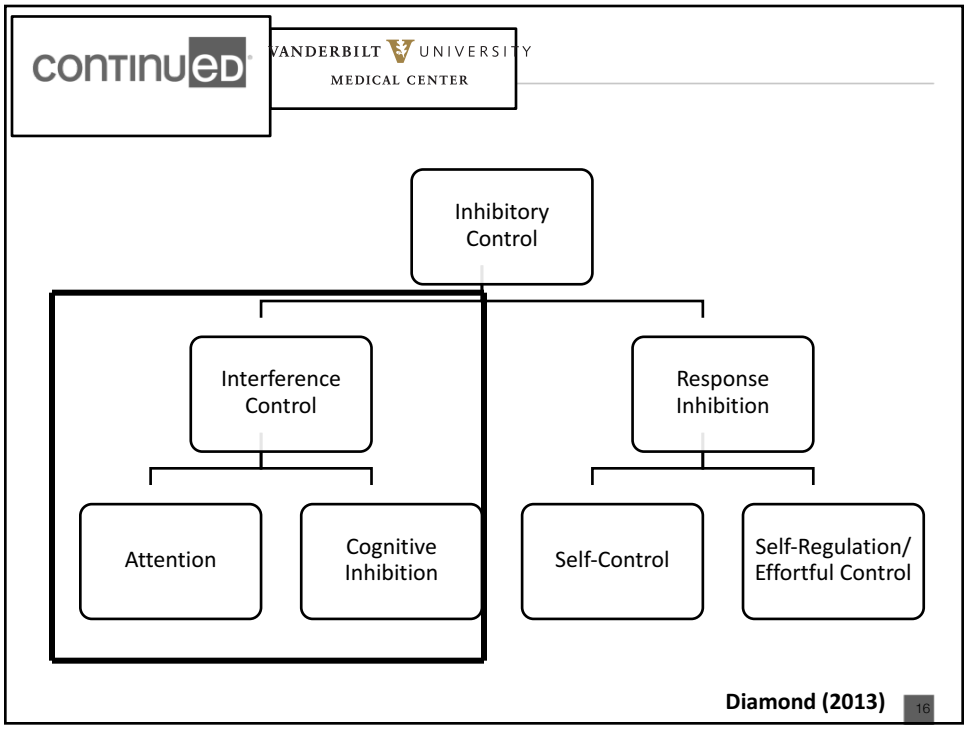
Executive functions

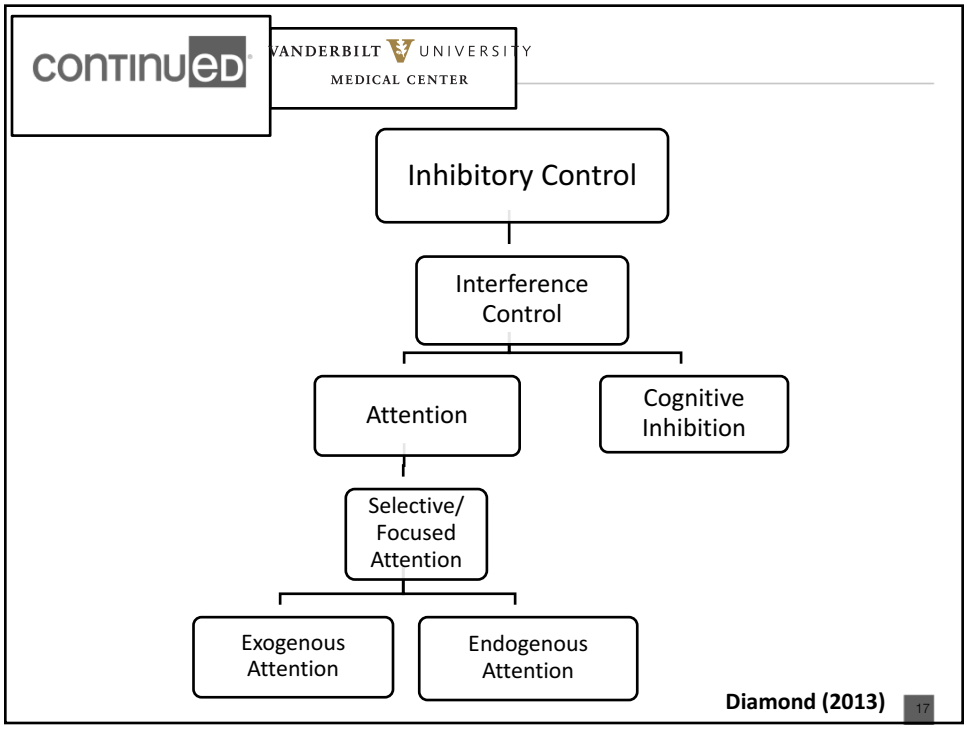
- "Executive functions refer to a family of top-down mental processes needed when you have to concentrate and pay attention, when going on automatic or relying on instinct would be ill-advised, insufficient or impossible" (p. 136).
- Executive functions are effortful; acting based on instincts or not resisting to temptation is easier.
- Executive functions are essential to physical and mental health, academic success, job success, cognitive, psychological and social development, and overall quality of life.

Diamond (2013)

14


Core Executive Functions
Working Memory
Inhibitory Control
Cognitive Flexibility
Higher-Level Executive Functions
Reasoning
Problem Solving
Planning
Diamond (2013)
15


Inhibitory Control
Interference Control
Response Inhibition
Attention
Cognitive Inhibition
Self-Control
Self-Regulation/ Effortful Control
Diamond (2013)
16


Inhibitory Control
Interference Control
Attention
Cognitive Inhibition
Selective/ Focused Attention
Exogenous Attention
Endogenous Attention
Diamond (2013)
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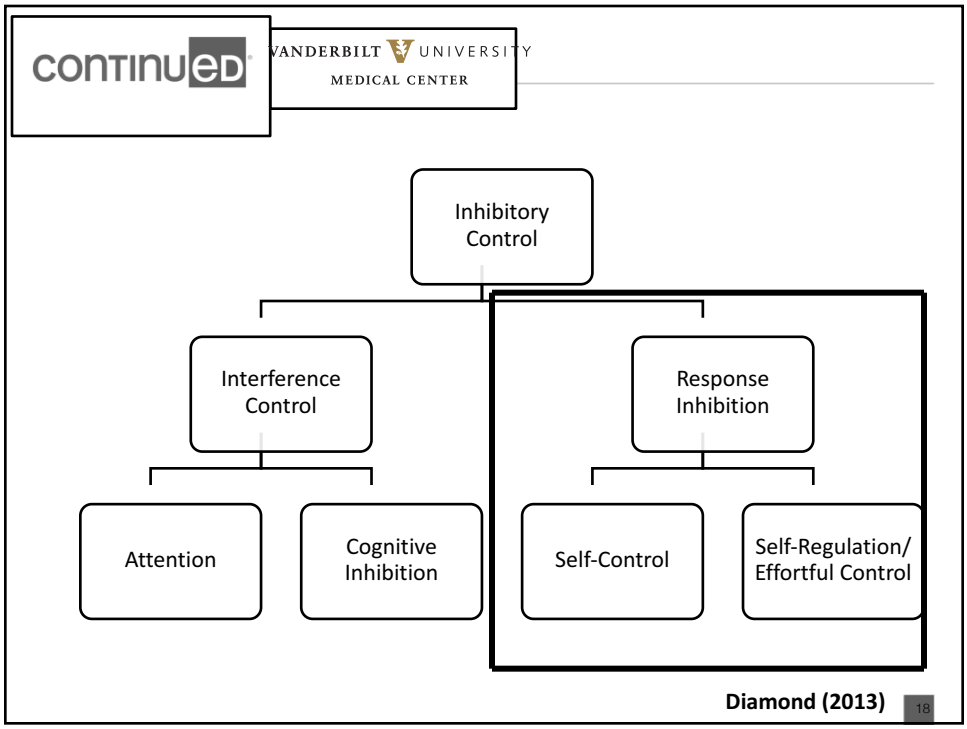
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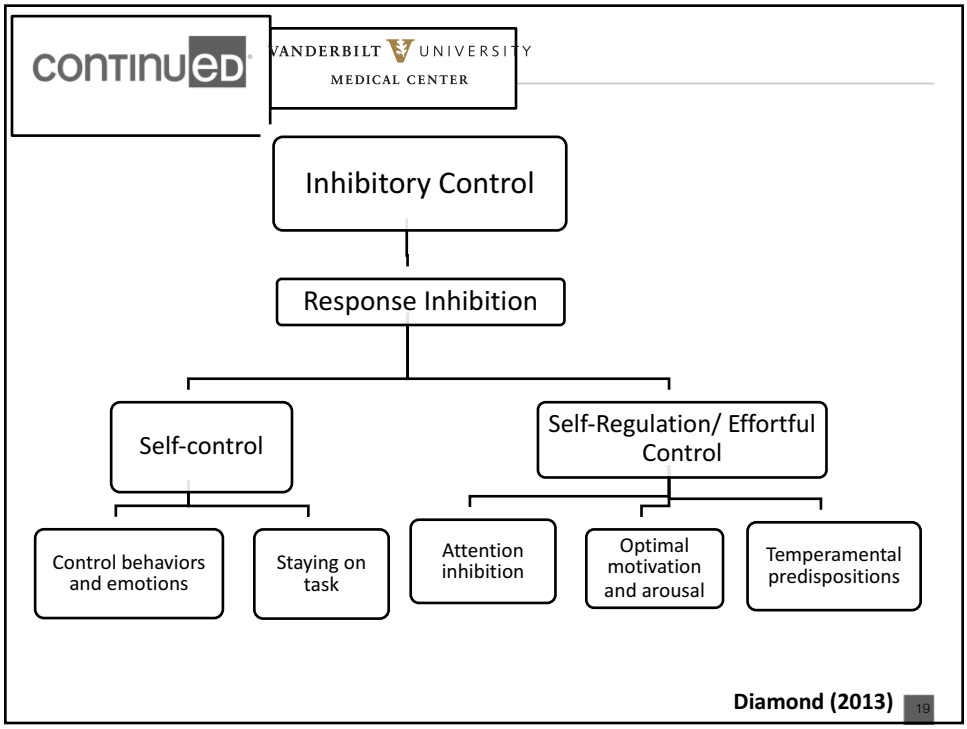
Self-Control
Self-Regulation/ Effortful Control
Diamond (2013)
18



Diamond (2013) 19

Development of Inhibitory Control

- Inhibitory control is far more difficult for young children than adults
  - inhibitory control tasks are far more difficult for children than working memory tasks; whereas the reverse is true for adults.
- Inhibitory control early in life is predictive of outcomes later in life.
  - Children with better inhibitory control at ages 3-11 were more likely to have better physical and mental health, earn more and be more law abiding adults 30 years later than those with worse inhibitory control skills, controlling for IQ, gender, social class (Moffitt et al., 2011, 2012 in Diamond, 2013).
- Inhibitory control shows a noticeable decline during normal aging.

Diamond (2013) 20

CONTINUED

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Relations between inhibitory control and working memory

- Inhibitory control and working memory are highly intertwined.
- Working memory supports inhibitory control.
  - Holding a goal, rules, or cues in mind are necessary for inhibitory control.
- Inhibitory control supports working memory.
  - Inhibition of external and internal distractions are necessary to focus on holding information in mind.

Diamond (2013) 21

CONTINUED

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Executive Functions

- It is not always helpful or beneficial to use executive functions or exert top-down control.
- "Canary in the coal mine":
  - Executive functions are the first to suffer and do so disproportionately under situations of stress, sadness, loneliness, sleep-deprivation, and lack of physical health and fitness.
  - Under these situations, a person may be incorrectly diagnosed with an executive function disorder.
- Executive functions can be improved with training and practice at any age, including the elderly and infants:
  - Repeated practice and continually increasing difficulty are key.

Diamond (2013) 22

CONTINUED

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Article 2:
Temperament dimensions in stuttering and typically developing children

Eggers, K., De Nil, L. F., & Van den Bergh, B. R. (2010). Temperament dimensions in stuttering and typically developing children. *Journal of Fluency Disorders*, *35*, 355–372. https://doi.org/10.1016/j.jfludis.2010.10.004

23

CONTINUED

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Purpose

- The purpose of this study was to investigate whether children who stutter (CWS) and children who do not stutter (CWNS) show differences in various aspects of temperament as measured by a parental temperament questionnaire: the Children's Behavior Questionnaire—Dutch (CBQ-D; Van den Bergh & Ackx, 2003).

Eggers et al. (2010)

24

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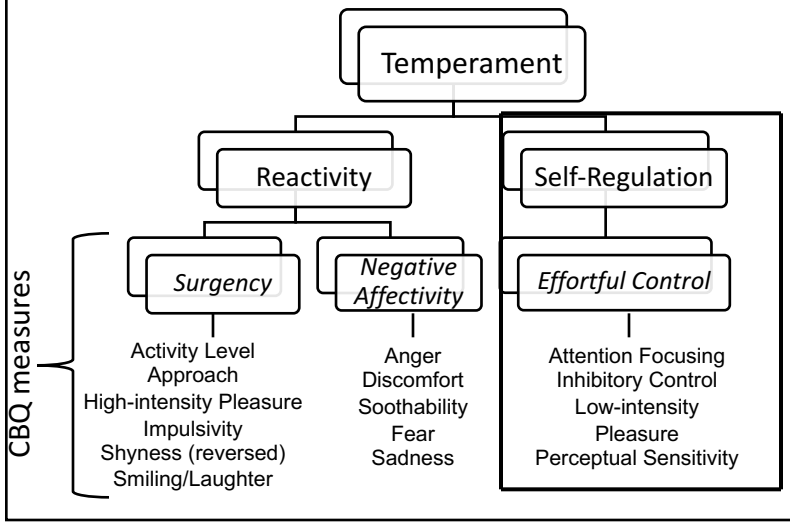
Temperament and Executive Functions

Rothbart's (2011) model of temperament:



25

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Temperament and Developmental Stuttering

- A review of literature on the development of behavioral disorders revealed that temperament characteristics play significant roles in *disorder onset*, *stress responses*, *conditioning processes*, and *treatment outcomes.*
- For developmental stuttering, theoretical perspectives and empirical evidence suggested that temperament might be importantly involved in the onset and development of stuttering (e.g., heightened reactivity and decreased regulation).
  - Eggers et al (2010) study was the first to specifically conceptualize both positive/negative reactivity and self-regulation in such a highly integrated manner.

Eggers et al. (2010)

26

CONTINUED

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Research Questions

- Do CWS, compared to CWNS, exhibit heightened scores on negative/positive reactivity and on some of the individual reactivity-related scales?
- Do CWS, compared to CWNS, exhibit lower scores on **effortful control** (i.e., self-regulation) and on some of the individual scales related to self-regulation?
- Is Temperament in CWS associated with length of therapy and/or stuttering severity?

Eggers et al. (2010) 27

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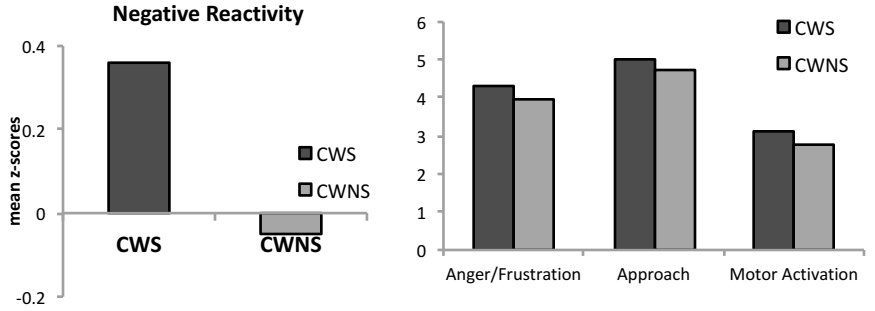
Methods

- Participants (matched for age and gender):
  - Children who do not stutter (n = 68)
  - Children who do stutter (n = 68)
  - Age: 3;04 – 8;11
- Temperament assessed via CBQ
- Stuttering severity assessed via Stuttering Severity Instrument
- Length of therapy was recorded for the CWS who had received treatment

Eggers et al. (2010) 28

Results

- Q1: Do CWS, compared to CWNS, exhibit heightened scores on negative/positive reactivity and on some of the individual reactivity-related scales?

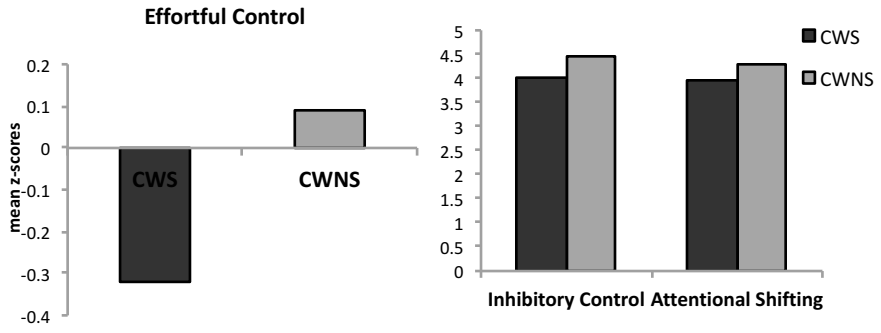


Eggers et al. (2010)

29

Results

- Q2: Do CWS, compared to CWNS, exhibit lower scores on **effortful control** (i.e., self-regulation) and on some of the individual scales related to self-regulation?



Eggers et al. (2010)

30

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Results

- Q3: Is Temperament in CWS associated with length of therapy and/or stuttering severity?
  - There were no significant correlations between temperament and therapy duration as well as temperament and stuttering severity.
  - Caveats:
    - Treatment or type of treatment was not directly controlled in this study, conclusions are "tentative at best".
    - Emotional reactivity and regulation was not assessed immediately prior to, during, or preceding communicative situations in which stuttering frequency/severity was assessed.

Eggers et al. (2010) 31

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Conclusions

- Overall, CWS, compared to CWNS, exhibit heightened negative reactivity and decreased self-regulation.
- CWS scored lower on Effortful Control, inhibitory control and attentional shifting:
  - They are less able to suppress inappropriate approach responses or to shift attention from one activity to another
  - The lower scores on these tests might be a result of a lower efficiency in the attentional network.
- Preliminary clinical implications, results:
  - May validate the use of desensitization (when age appropriate) to reduce reactivity to stuttering or other stimuli
  - Illustrate the use of parental guidance in working with young CWS

Eggers et al. (2010) 32

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Article 3:

Attention regulation in young twins with probable stuttering, high nonfluency, and typical fluency

Felsenfeld, S., van Beijsterveldt, C. E. M., & Boomsma, D. I. (2010). Attentional Regulation in Young Twins With Probable Stuttering, High Nonfluency, and Typical Fluency. *Journal of Speech Language and Hearing Research*, *53*, 1147–1166. https://doi.org/10.1044/1092-4388(2010/09-0164)

33

CONTINUED

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Purpose

- "Using a sample of 20,445 Dutch twins, this study examined the relationship between speech fluency and attentional regulation in children. A secondary objective was to identify etiological overlap between nonfluency and poor attention using fluency-discordant twin pairs." (p. 1147)

Felsenfeld et al. (2010)

34

CONTINUED

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Motivation attention-stuttering link

- The 3 to 5 year old age range represents the time period when ***significant development*** occurs in aspects of ***attentional control*** as well as early childhood stuttering.
- Attention regulation may play a significant role in speech-language planning and production.
  - *Speech production models* and *models of stuttering* have suggested that executive functions may be associated with speech (dis)fluency.
  - Attention and inhibitory control have been linked to speech-motor control and speech fluency in other populations.

Felsenfeld et al. (2010)

35

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Objectives of Felsenfeld et al.'s (2010) study:

- ***Objective 1:*** Determine whether there are differences in attention regulation between children with probable stuttering compared to highly non-fluent children and those with typical fluency.
  - ***Hypothesis 1:*** Children with probable stuttering would exhibit more problematic scores of attention than children with typical fluency (and highly non-fluent children who fall in between the groups).
- ***Objective 2:*** Determine whether there is etiological overlap between nonfluency and attention.
  - ***Hypothesis 2:*** Nonfluency and attention would share a common underlying genetic vulnerability.

36

CONTINUED

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Participants

Table 1. Criterion for fluency group membership.

Fluency group	Description	Criterion
PS *N = 826*	Children are reported to display one or more of the three "core" behaviors of beginning stuttering.	Mother reports that at least one of the following core stuttering behaviors occurs "often" or "very often": 1. Rapidly repeats part of a word (e.g., muh-muh-muh-may I go?"). 2. Blocks at the beginning or middle of a word (example: "I . . . go to school"). 3. Prolongs a sound within a word (example: "I go to sssssssschool.")
HNF *N = 547*	Children are reported to exhibit fluency that is chronically unstable but does not contain frequent occurrences of the core stuttering behaviors described above. These children's speech would be described as sounding disorganized, fragmented, and marked by the unnecessary repetition of words and phrases.	Mother reports that at least two of the following behaviors occur "often" or "very often": 4. Repeats a part of a sentence (example: "And then he..and then he came home"). 5. Slowly repeats a word in a sentence (example: "I. . .I. . .I go to school"). 6. Rapidly repeats a word in a sentence (example: "III go to school"). AND ➢ The three core stuttering behaviors described above are reported to occur "never," "rarely," or "sometimes."
TF *N = 19,072*	Children demonstrate speech fluency that is typical for age.	Approximately one third of these children (34%) had received ratings of "never" on all six of the fluency items.

Note. PS = probable stuttering; HNF = highly nonfluent; TF = typically fluent.

Felsenfeld et al. (2010) 37

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Measures of attention

- At 5 years old:
  - Deveruex Child Behavior scale (DCB; Spivak & Spotts, 1966; Van Beijsterveldt, Verhulst, Molenaar, & Boomsma, 2004)
    - Example items: "is distracted by others," "does not attend to an activity," and "jumps from one activity to another."
- At 7 years old:
  - Child Behavior Checklist/Ages 4-18 (CBCL/4-18; Achenbach, 1991)
    - Example items: "can't concentrate," "is impulsive," and "can't sit still"
  - Conners' Parent Rating Scale—Revised (CPRS-R; Conners, Parker, Sitarenios, & Epstein, 1998)
    - Subscales: 1) Cognitive Problems/Inattention, 2) Hyperactivity, 3) ADHD index

Felsenfeld et al. (2010) 38

Results: Between-group differences in attention

Table 3. Midparent *t* scores (*M*s and *SD*s) for the five attention measures by fluency group.

Measure	TF group			PS group			HNF group			
	N	*M*	*SD*	*N*	*M*	*SD*	*N*	*M*	*SD*	*F*
Age 5 years										
DCB Attention Problems	19,033	49.7	9.0	822	54.0[b]	10.4	546	54.1[b]	10.0	101.7[a]
Age 7 years										
CBCL/4-18 Attention Problems	12,219	49.8	9.4	482	54.5[b]	11.1	332	55.3[b]	11.2	79.7[a]
CPRS–R Attention Problems	5,927	49.8	9.4	214	53.8[b]	11.5	177	54.0[b]	11.1	14.0[a]
CPRS–R ADHD	5,948	49.8	9.4	214	54.1[b]	11.2	178	54.3[b]	11.0	16.6[a]
CPRS–R Hyperactivity	5,954	49.8	9.3	215	53.9[b]	12.2	178	53.3[b]	10.9	20.1[a]

Note. DCB = Devereux Child Behavior scale; CBCL4-18 = Child Behavior Checklist/Ages 4–18; CPRS–R = Conners' Parent Rating Scale—Revised.

[a]*F* value is significant at $p < .001$. [b]Affected group is significantly different from controls at $p \leq .002$.

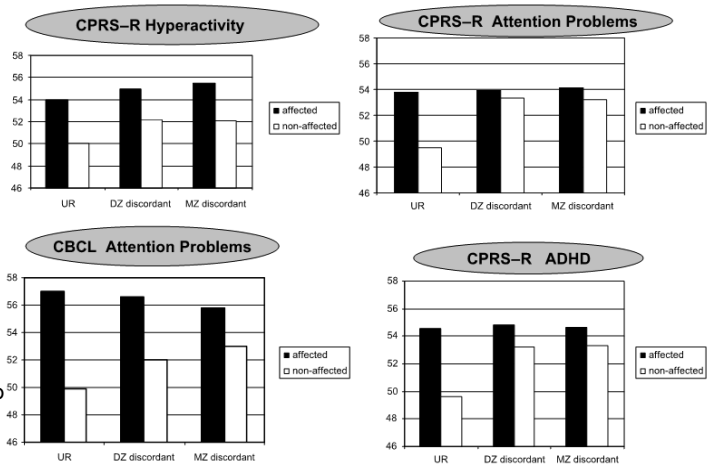
Takeaway: The probable stuttering group exhibited significantly higher (more problematic) attention scores than the typically fluent group. Further, the children in the highly non-fluent group exhibited significantly higher attention scores than the typically fluent children, similarly to the probable stuttering group.

Felsenfeld et al. (2010)

39

Results: Discordant twin analysis

Figure 1. Maternal attention *t* scores at age 7 years for the three fluency-discordant groups. CPRS–R = Conners' Parent Rating Scale—Revised; UR = genetically unrelated pseudopairs; DZ = dizygotic; MZ = monozygotic; affected = probable stuttering or highly nonfluent; non-affected = typically fluent; CBCL = Child Behavior Checklist; ADHD = attention-deficit/hyperactivity disorder index.



Takeaway 1: The probable stuttering and highly non-fluent children tended to receive higher attention scores than the typically fluent group.

Takeaway 2: Scores for the pairs that were genetically related were more similar to the unrelated pseudopairs.

Felsenfeld et al. (2010)

40

CONTINUED

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Discussion

- Present results indicate a ***salient association between attention and speech fluency*** that may be related to an "…important pathogenic mechanism for fluency development" (p. 1157).
- Both ***shared genes*** and ***shared environmental factors*** are involved with the association between unstable fluency and unstable attention.
  - Felsenfeld et al. interpret these results to indicate that the liability to express stuttering (or high nonfluency) and difficulties with attention/self-regulation may arise at least in part from "shared mechanisms."

Felsenfeld et al. (2010) 41

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Clinical implications

- Present results underscore the increased likelihood that clinicians may see children and adults who stutter who have **co-occurring attentional vulnerabilities** and that clinicians should ***adapt assessment and treatment*** approaches accordingly.
- Previously, executive function, attention, and self-regulation have been discussed as ***supporting the success of stuttering treatment*** in adults (for overview, see Finn, 2007) and children (Riley & Riley, 2000).

Felsenfeld et al. (2010) 42

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Article 4:
Traits of attention deficit/hyperactivity disorder in school-age children who stutter

Donaher, J., & Richels, C. (2012). Traits of attention deficit/hyperactivity disorder in school-age children who stutter. *Journal of Fluency Disorders*, *37*(4), 242–252. https://doi.org/10.1016/j.jfludis.2012.08.002

43

CONTINUED

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Purpose

- "The purpose of this study was to explore whether parents of CWS reported the presence of ADHD symptoms that would warrant a referral to a psychologist to rule out the disorder. This study also aimed to describe the characteristics of the sample in terms of gender, family history of stuttering, presence of neurological impairment, concomitant diagnoses, and stuttering severity." (p. 242)

Donaher & Richels (2012)

44

ADHD and stuttering background

- A number of studies have shown an association between the core symptoms of ADHD and difficulties with the efficient production of speech-language (e.g., Engelhardt, Corley, Nigg, & Ferreira, 2010).
  - Examples: excessive language production, poor topic maintenance, difficulty sequencing and organizing
- Donaher and Richels (2012) note the many similarities between stuttering and ADHD:
  - Significant genetic and environmental components
  - Both occur more often in boys (e.g., ~4 or 5 to 1 ratio)
  - Exacerbated by stress and change over development
  - Both have been related to neural circuitry of basal ganglia, which plays a role in motor behaviors, emotions and cognition

45

Research questions

1. Do parents of CWS routinely report clinically relevant symptoms of ADHD in their children?
2. Does family history of stuttering, history of neurological impairment, and concomitant diagnoses contribute to whether CWS meet criteria for referral for ADHD symptoms?
3. Do subtypes of ADHD interact with family history, neurological status, and concomitant diagnoses in CWS?

Donaher & Richels (2012)

46

Participants

- 36 CWS (32 males and 4 females)
  - Age: 3.9 to 17.2 years (M = 10.4, SD = 4)
  - Recruited from pediatric hospital setting (n = 18) and community based speech clinic (n = 18)

Donaher & Richels (2012) 47

Measures

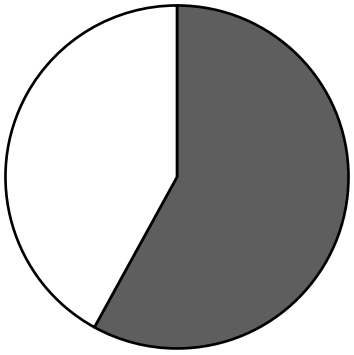
- *ADHD rating scale* (Power et al., 2001): 8 item questionnaire used to determine if children exhibit enough of the core characteristics of ADHD to warrant referral
  - 4-point scale ranging from 0 (never or rarely), 1 sometimes, 2 (often), 3 (very often)
  - ADHD-IA (inattentive), ADHD-HI (hyperactive-impulsive), ADHD-C (combined)
- Family history of stuttering, history of neurological impairment and concomitant diagnoses
  - E.g., stuttering (recovered or persistent), ADHD, Tourette's, anxiety, etc.

Donaher & Richels (2012) 48

Results

- ***Main finding***: 58% of CWS met criteria for referral for evaluation of ADHD symptoms.
  - **Important consideration**: This does NOT indicate that 58% of these children would be diagnosed with ADHD.
- ***Takeaway***: CWS present with high levels of parent reported ADHD symptoms, and future studies are necessary to explore the role of inattention, impulsivity, and hyperactivity in stuttering.

Percentage of participants that met criteria for referral



Donaher & Richels (2012)

49

Clinical implications

- Donaher and Richels (2012) suggest that CWS who also exhibit coexisting ADHD traits will require ***modifications to assessment and treatment***.
- Identification of ADHD traits may guide treatment approach:
  - Example 1: CWS with ADHD-HI may struggle with turn-taking, and therefore focusing on this may be effective for this child.
  - Example 2: A CWS with ADHD-IA may struggle with topic maintenance, and therefore may not benefit from turn-taking approaches.

50

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Executive functions and childhood stuttering: Clinical implications

- Assessment tools
- Case studies: Assessing and interpreting executive functions
- Treatment approaches
- Future directions

51

CONTINUED

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Children's behavior questionnaire (CBQ)

- CBQ (Rothbart et al., 2001) can be used for the comprehensive assessment of temperament in 3 to 7 year old children
  - The "full" version is 195 items. "Short" and "very short" versions are available.
    - Clinician may select and only administer scales of interest
  - Questionnaires available for all age ranges
- CBQ sample items from subscales focusing on executive functions:
  - Inhibitory control: *"Can easily stop an activity when s/he is told 'no'."*
  - Attention focusing: *"When picking up toys or other jobs, usually keeps at the task until it's done."*

52

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~~Behavior~~ Rating Inventory of Executive Function (BRIEF)

- BRIEF (Gioia et al., 2000) can be used to assess executive function in
  - Children 2 years to 5 years 11 months (BRIEF-P,)
  - Children 5 years to 18 years (BRIEF-2)
- BRIEF takes 10-15 minutes to administer and can be completed by parents and teachers
  - BRIEF self-report version is available for 11-18 years
- Multiple aspects of executive function can be assessed; scales include Inhibit, Shift, Emotional Control, Working Memory
  - Inhibit: *"Has trouble putting brakes on his/her actions even after being asked"*
  - Working Memory: *"Needs help from adult to stay on task"*

53

CONTINUED

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~~Case study~~

Name	Age	BRIEF Inhibit	BRIEF WM	GFTA	PPVT	TELD Recept	TELD Expres	% SLD	SSI	CBQ Att Fo	CBQ inhibit	CBQ Effort
Ellen	76	80	83	109	122	118	107	10.67	24	3.33	3.23	4.33
Chris	55	69	85	104	103	110	90	3.67	12	5.11	3.15	3.75
Patrick	70	69	76	108	98	105	88	4.67	14	3.89	3.46	4.46

BRIEF Inhibit
BRIEF Working Memory
Goldman Fristoe Test of Articulation
Peabody Picture Vocabulary Test
Test of Early Language – Receptive, Expressive

% Stuttering Like Disfluencies
Stuttering Severity Instrument
CBQ Attentional Focusing
CBQ Inhibitory Control
CBQ Effortful Control

54

CONTINUED

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Treatment approaches

- Assessment of a child's cognitive abilities and consideration re treatment approach has been employed in stuttering treatment (e.g., treatment approaches based on the Demands and Capacities Model, DCM; Adams, 1990; Starkweather & Gottwald, 1990).
- For a recent example of an empirically tested treatment approach that considers cognitive capacities and vulnerabilities, see the RESTART-DCM treatment approach (de Sonneville-Koedoot, Stolk, Rietveld, Franken, 2015).

55

CONTINUED

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Treatment approaches

- Potential profile of child with ***low inhibitory control and/or high impulsivity, hyperactivity***:
  - Child may not "know a stranger".
  - Talk nonstop without breaks for for pauses or turns.
- Communicative behaviors may be stressing cognitive, linguistic and motoric vulnerabilities for disfluencies.

Attribution: https://pixabay.com/en/children-fast-child-people-kid-2091275/

56

CONTINUED

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Treatment approaches

- Potential **treatment strategies** *for low inhibitory control and/or high impulsivity, hyperactivity:*
  - Provide appropriate ***modeling*** of communicative "manners" (e.g., turn-taking).
    - Play ***"turn-taking"*** game.
    - ***Use movement*** (e.g., "musical chairs").
  - Model pausing before responding/answering question.
  - "Play" with the concept of fast and slow (e.g., coloring with crayons).

57

CONTINUED

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Treatment approaches

- Potential profile of child with ***low attention***:
  - Child may have poor topic maintenance.
  - Child may exhibit increased disfluencies when talking about things not in the "here and now" or with increased linguistic complexity.
- Potential treatment strategies:
  - Find "clues" and put them together before solving a word "puzzle" and then describing it.
  - **Build** linguistic and conversational **complexity gradually** as child becomes better at pausing and taking turns.
  - **Model pausing** to think before answering a question.

58

CONTINUED

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Training inhibitory control

- Emerging evidence that executive functions ***may*** be a trainable resource (for review, see Diamond & Lee, 2011)
  - ***However***…many questions remain!
- Current evidence does NOT support executive function training as a part of stuttering treatment…
- Evidence is necessary that addresses:
  - Do benefits of inhibitory control training carry over to other executive functions?
  - Do benefits of inhibitory control training carry over to daily life activities?
  - Does inhibitory control training result in a reduction in disfluencies?
  - What are the best doses, durations, and frequency?
  - How long do benefits last?

59

CONTINUED

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Summary

- Executive functions, in particular attention and inhibition, appear to be associated with stuttering.
  - Executive functions may support fluent speech-language planning and production.
  - Children who stutter exhibit differences in attention, inhibition, and other measures of executive functions when compared to children who do not stutter.
- As clinicians, we can ***translate*** these findings to ***assessment*** and ***treatment*** and potentially enhance our treatment for children who stutter and their families.

60

CONTINUED

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Questions?

61

CONTINUED

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62

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63