Motor Speech Disorder-
Not Otherwise Specified:
Prevalence and Phenotype

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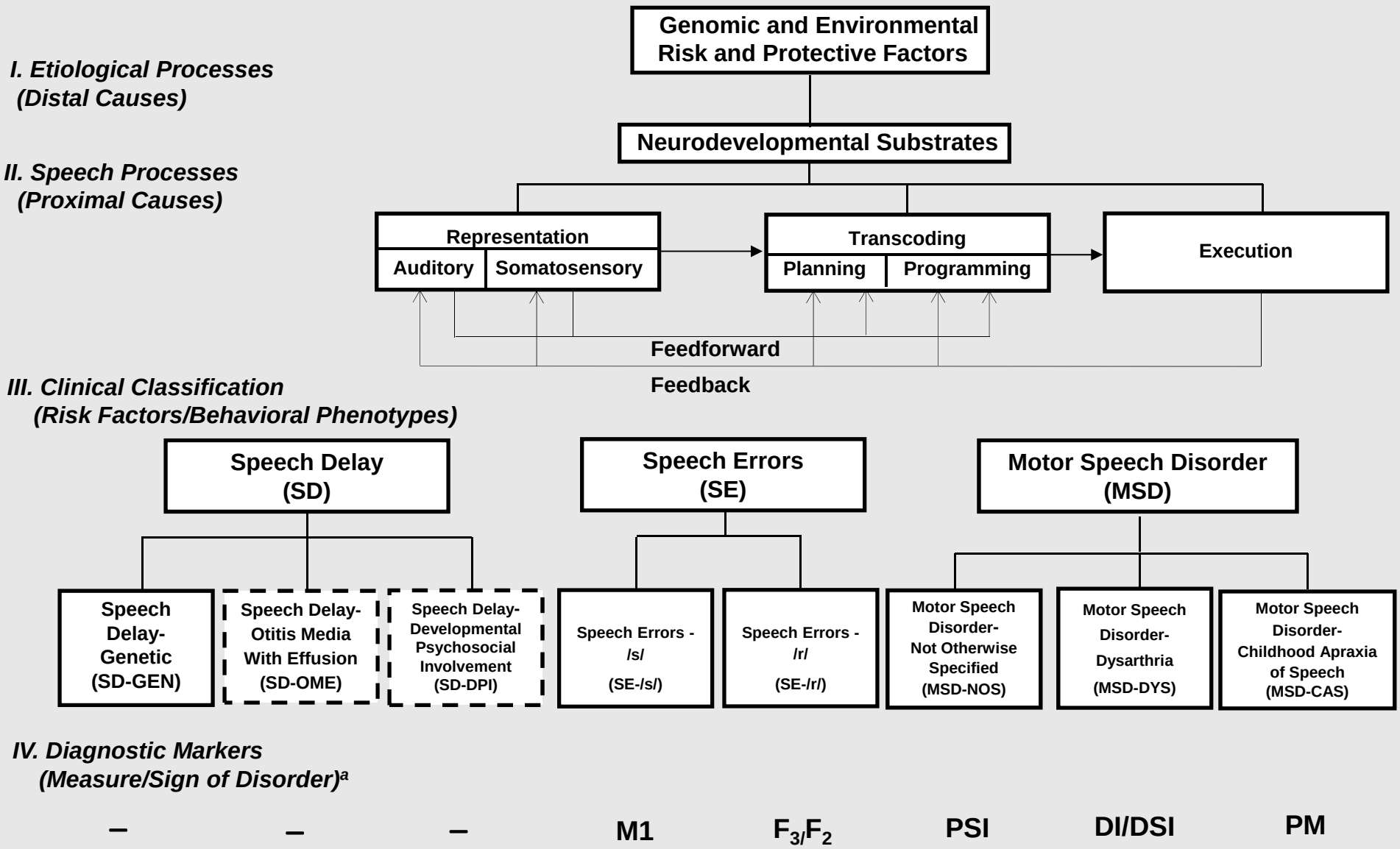
A Third Type of Childhood Motor Speech Disorder?

In addition to childhood dysarthria and childhood apraxia of speech, research suggests a need for a third motor speech classification for children with speech delay who also have a delay in the production of precise and stable speech, prosody, and voice.

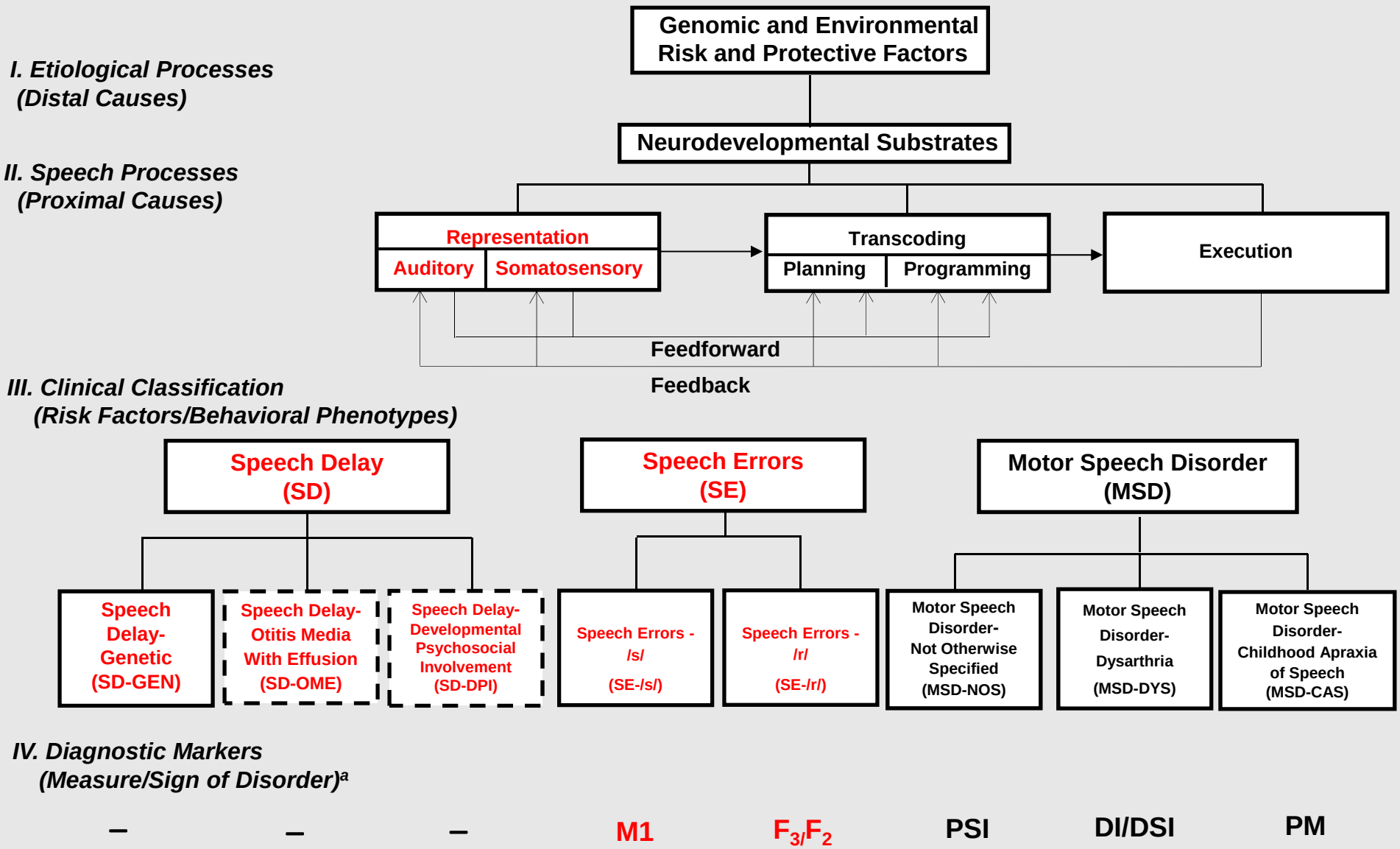
Some examples of relevant research findings and discussions: (Archibald & Alloway, 2008; Bishop, 2002; Cermak et al., 1986; Cheng et al., 2009; Flipsen, 2003; Gaines & Missiuna, 2007; Zelaznik & Goffman, 2010; Goffman, 1999; Goozée et al., 2007; Hill, 2001; Richtsmeier & Goffman, 2015; Lewis et al., 2011; Newmeyer et al., 2007; Nip et al., 2011; Owen & McKinlay, 1997; Powell & Bishop, 1992; Rechetnikov & Maitra, 2009; Visscher et al., 2010; Webster et al., 2006)

Placeholder Classification: Motor Speech Disorder-Not Otherwise Specified (MSD-NOS)

- Research needs
  - Develop and validate a measure of MSD-NOS
  - Estimate its prevalence in idiopathic speech sound disorders and in complex neurodevelopmental disorders
  - Characterize its behavioral phenotype
  - Characterize its biotype



[a] M1: First Spectral Moment; F_3/F_2: Format 3/Formant 2; PSI: Precision-Stability Index; DI/DSI: Dysarthria Index/ Dysarthria Subtype Indices; PM: Pause Marker



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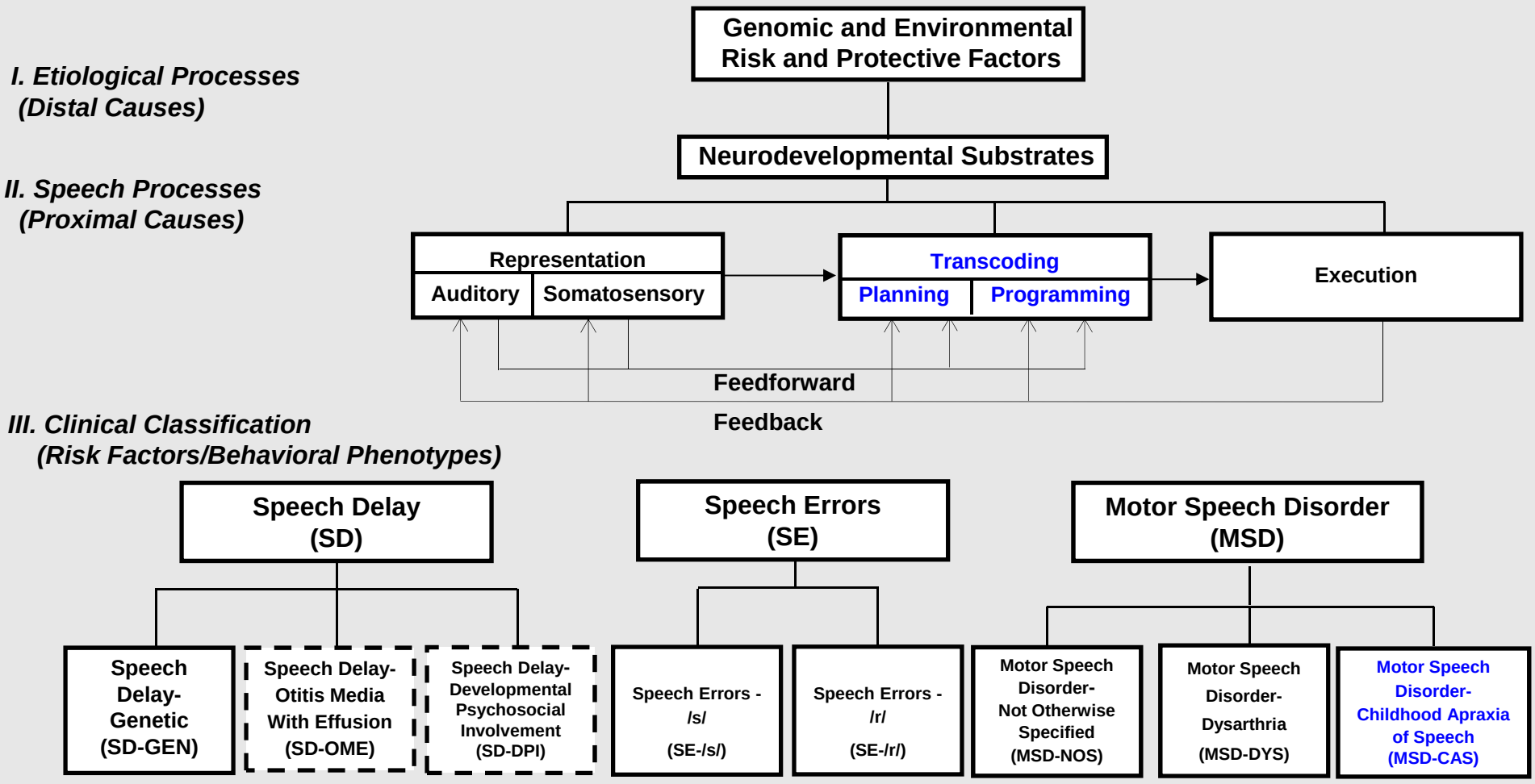
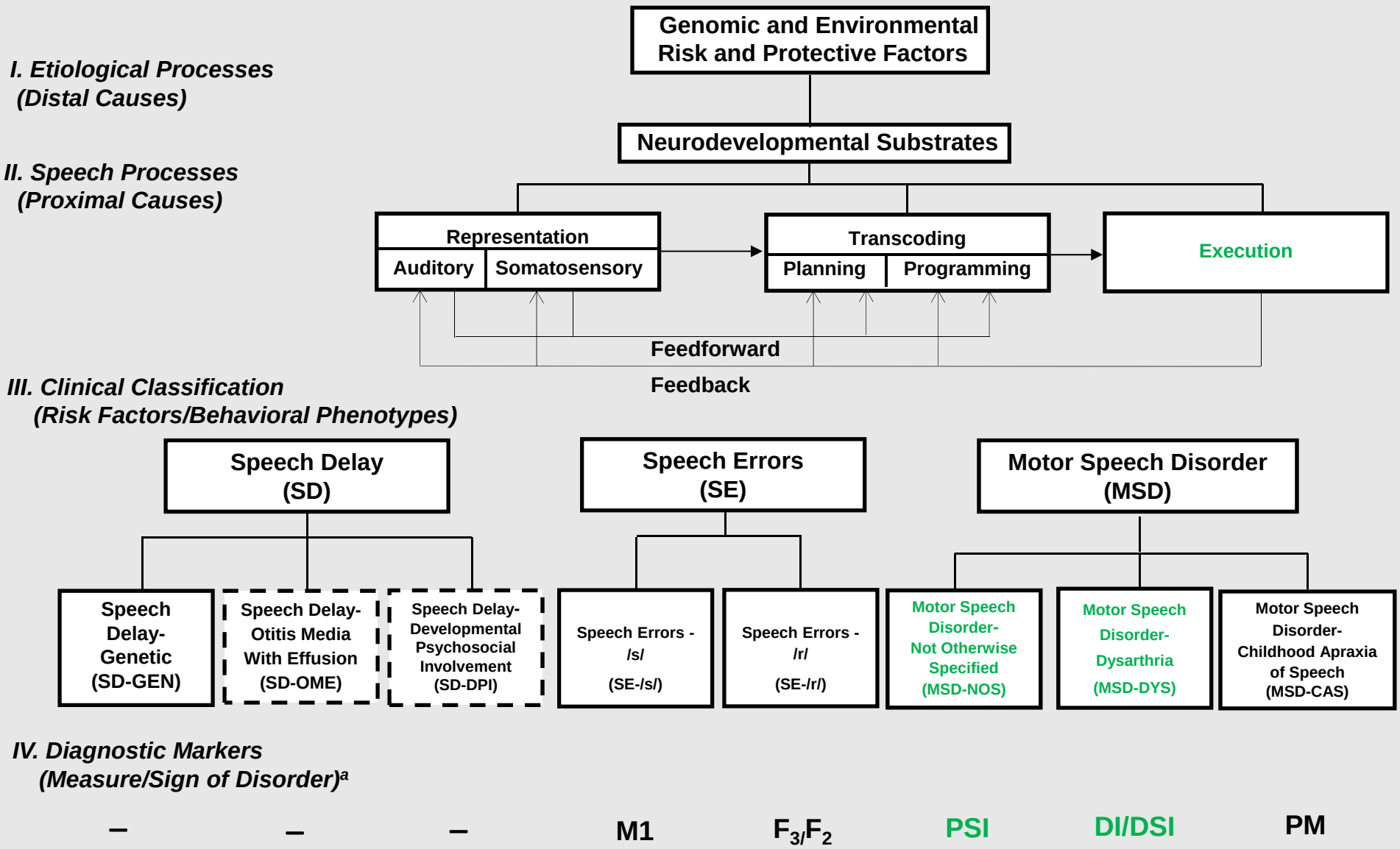


IV. Diagnostic Markers
(Measure/Sign of Disorder)[a]

–	–	–	M1	F_3/F_2	PSI	DI/DSI	PM

[a] M1: First Spectral Moment; F_3/F_2: Format 3/Formant 2; PSI: Precision-Stability Index; DI/DSI: Dysarthria Index/ Dysarthria Subtype Indices; PM: Pause Marker



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The Precision-Stability Index (PSI)

- 32 operationalized and standardized signs of speech, prosody, and voice precision and stability obtained from a conversational speech sample
- Perceptual and acoustic data reduction methods
- Reference database: 200 typical speakers; ages 3 to 80 yrs.
- Method
  - PSI signs greater than 1.25 standard deviation units from speakers of same sex and approximately same age are coded as imprecise/unstable
  - PSI score < 70% = MSD-NOS+ (i.e., positive for MSD-NOS)
  - Speakers meeting criteria for MSD-NOS and one of the other MSD classifications are classified as having the more severe disorder (i.e., as MSD-DYS, MSD-CAS, or MSD-DYS & CAS)

The Precision-Stability Index (PSI): Individual Form

		PSI Sign			PSI sign values		
Linguistic Domain	No.	Description	Assessment Mode[a]		Value	Z-score[b]	Code[c]
Vowels			P	A			
	1	Reduced Dispersion of Corner Vowels from Center		X			
	2	Reduced Dispersion of Corner Vowels from ʌ		X			
	3	Reduced Average Pairwise Distance for Corner Vowels		X			
	4	Increased Duration for Corner Vowels		X			
	5	Increased Duration for Diphthongs and Middle Vowels		X			
	6	Reduced % Vowel Phoneme Target Consistency	X				
	7	Reduced % Vowel Target Consistency	X				
Consonants							
	8	Reduced % Correct Glide	X				
	9	Increased Relative Distortion Index Sibilant	X				
	10	Reduced Relative % of Sibilants Dentalized	X				
	11	Increased Relative Distortion Index for Early Consonants	X				
	12	Decreased 1st Moment for /s/ Initial Singleton		X			
	13	Increased Sqrt 2nd Moment for /s/ Initial Singleton		X			
	14	Increased Sqrt 2nd Moment for /s/ initial, final /z/ final singleton		X			
	15	Increased All Consonant-Consonant Duration		X			
Vowels and Consonants							
	16	Increased % DMI Class Place	X				
	17	Increased % DMI Class Duration	X				
	18	Increased % of Epenthesis Errors	X				

The Precision-Stability Index (PSI): Individual Form

Phrasing							
	19	Increased % of Addition, Breath, Repeat or Long	X				
Rate							
	20	Reduced Average Syllable Artic Rate (without pauses)		X			
	21	Increased Average Syllable ms (without pauses)		X			
Stress							
	22	Increased % of PV15/16 EE codes of all coded utterances without fast/acceleration. (uncircled & circled)	X				
	23	Increased % of PV15/16 EE codes of all PV15/16 codes. (uncircled & circled)	X				
Loudness							
	24	Decreased Intensity Difference dB Fricative+Vowel		X			
Pitch							
	25	Decreased F0 for all Vowels & Diphthongs		X			
	26	Decreased Range of Characteristic F0 Vowels		X			
Laryngeal Quality							
	27	Increased % Jitter for Vowels		X			
	28	Increased % Shimmer for Vowels		X			
	29	Decreased HNR dB for Vowels		X			
Resonance Quality							
	30	Increased % Inappropriate Resonance	X				
	31	Decreased F1 /ɑ/ (Nasal)		X			
	32	Decreased F2 for High Vowels (Nasopharyngeal)		X			

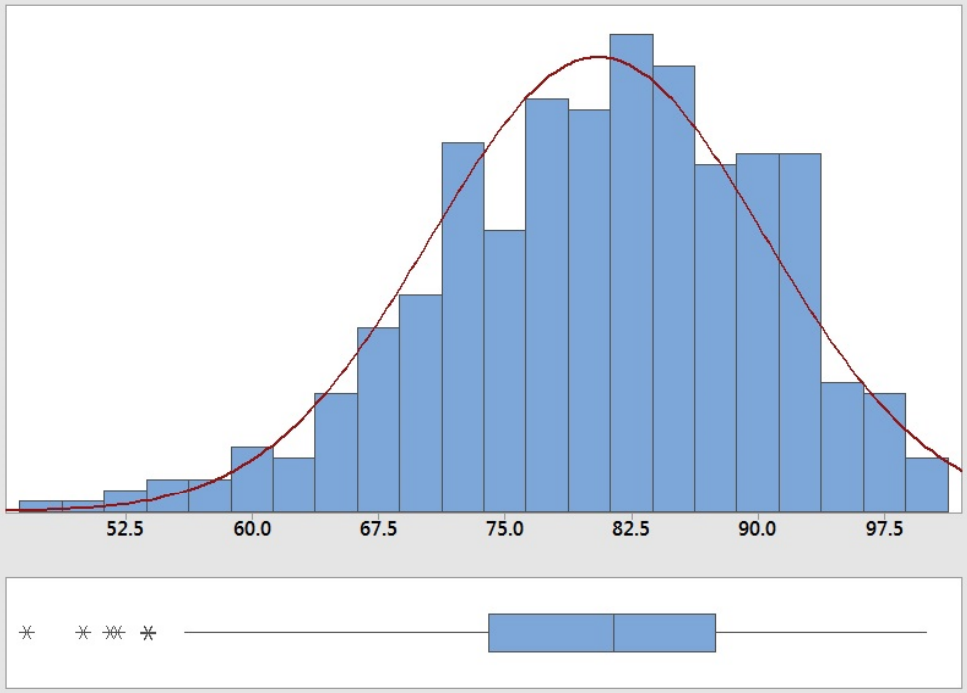
No. of positive signs	
No. of signs with data	
Average Sign Z-score	
% Positive Signs (PSI Score)	

[a] A: Acoustic; P: Perceptual

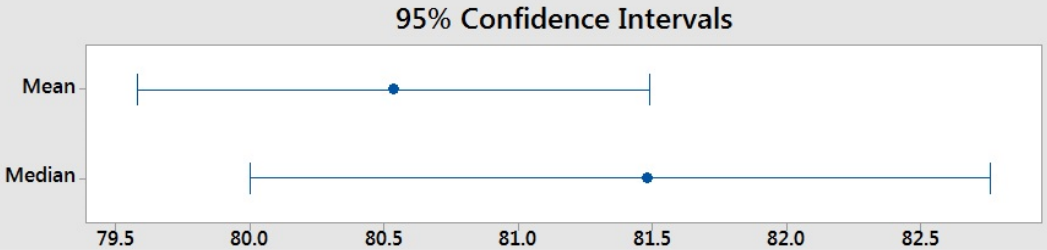
[b] Z-scores referenced to age-sex matched typically developing speakers (Potter et al., 2012; Scheer-Cohen et al., 2013)

[c] Code: 0 = not positive on variable; 1 = postive on variable (Z-score $\leq$ -1.25). Z-scores reversed for Increased.

PSI Scores of 415 children ages 3 to 16 Years of Age Recruited for Idiopathic Speech Delay: Psychometric Information



Anderson-Darling Normality Test	
A-Squared	1.43
P-Value	<0.005[a]
Mean	80.535
StDev	9.885
Variance	97.703
Skewness	-0.488570[b]
Kurtosis	0.083194
N	415
Minimum	46.670
1st Quartile	74.070
Median	81.480
3rd Quartile	87.500
Maximum	100.000
95% Confidence Interval for Mean	
79.581	81.489
95% Confidence Interval for Median	
80.000	82.760
95% Confidence Interval for StDev	
9.255	10.607



[a] Not normally distributed

[b] Skewed to left as expected

Prevalence Estimate for MSD-NOS in Children Recruited for Speech Delay

Estimate 1

164 children ages 3 to 5 years recruited and assessed for Speech Delay in collaborative research in 3 cities within the U.S. (Shriberg, Strand, & Mabie, 2016)

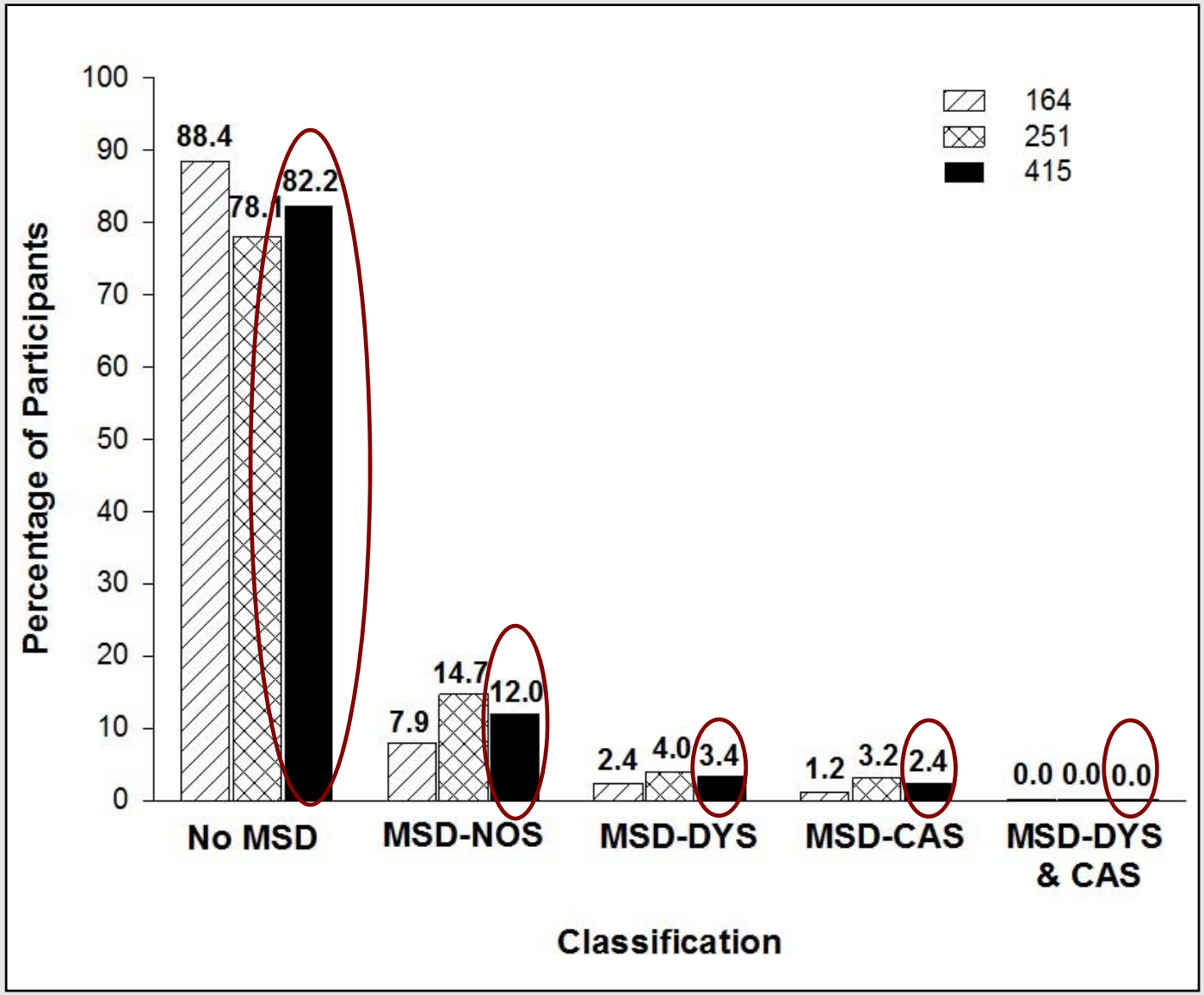
Estimate 2

251 children ages 3 to 16 years recruited and assessed for Speech Delay in collaborative research in 3 additional cities within the U.S.

Combined Estimate

415 children; 6 U.S. cities; 12 samples, 7 in same city

MSD-NOS Prevalence Estimates

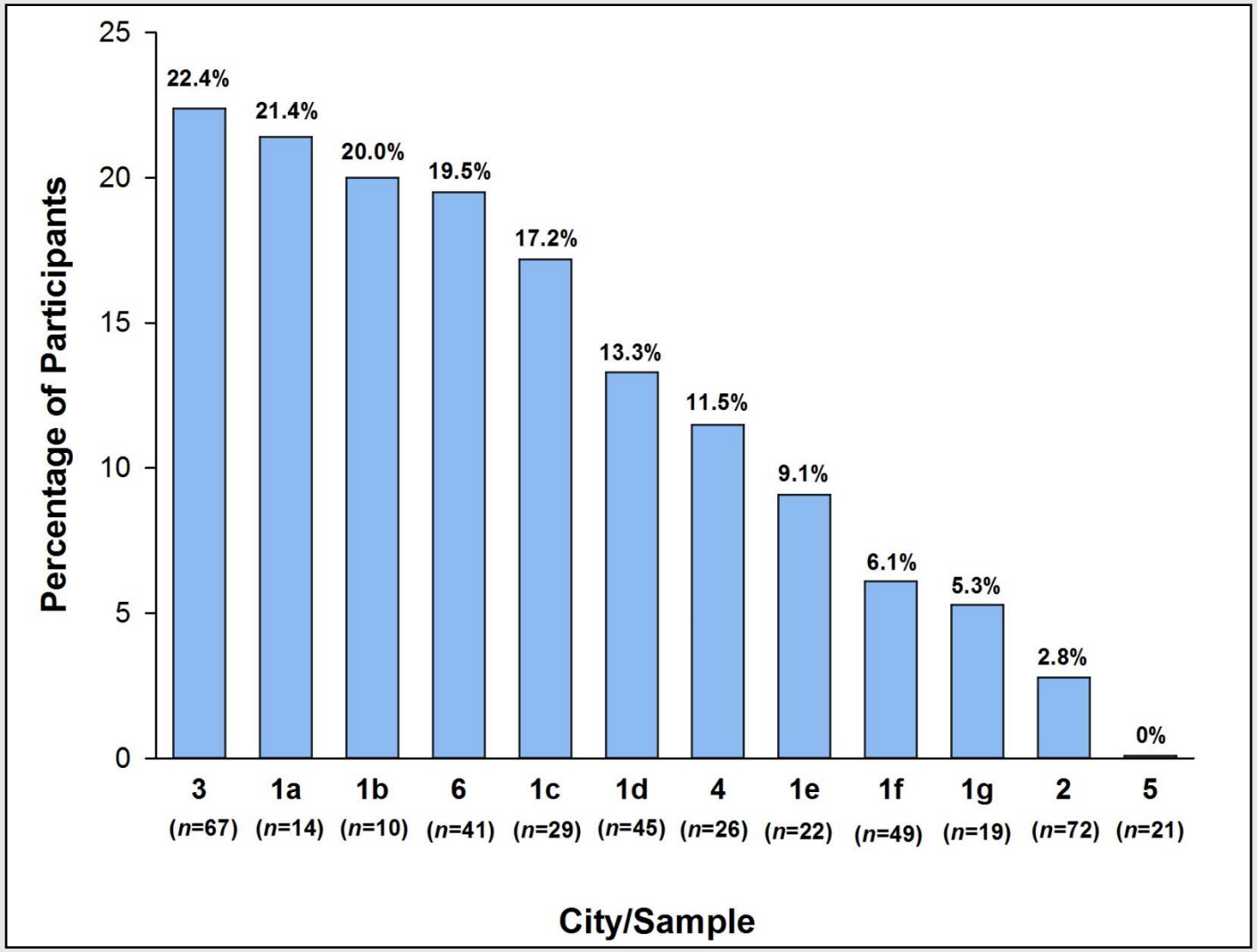


Percentage of Speakers with MSD-DYS or MSD-CAS Who Also Met PSI Criteria for MSD-NOS

- MSD-DYS
  - *n* = 14
  - MSD-NOS = 6
  - Percentage = 42.9%
- MSD-CAS
  - *n* = 10
  - MSD-NOS = 3
  - Percentage = 30.0%

- Interpretation: Most (i.e., over 50%) participants with MSD-DYS or MSD-CAS did <u>not</u> meet PSI criteria for MSD-NOS

Variability Among 12 Estimates of the Prevalence of MSD-NOS



Summary: MSD-NOS Prevalence Findings in Idiopathic Speech Delay Compared to Prior Prevalence Findings in Complex Neurodevelopmental Disorders

- Idiopathic Speech Delay
  - 415 children recruited for 12 research samples in 6 U.S. cities
  - Prevalence of MSD-NOS averaged over 12 samples:

    Mean: 12.4%

    SD: 7.8%

    Range: 0.0%-22.4%
- Complex Neurodevelopmental Disorders[a]
  - 340 participants recruited for research studies in 8 disorders
  - Prevalence of MSD-NOS averaged over 8 disorders

    Mean: 25.7%

    SD: 11.6%

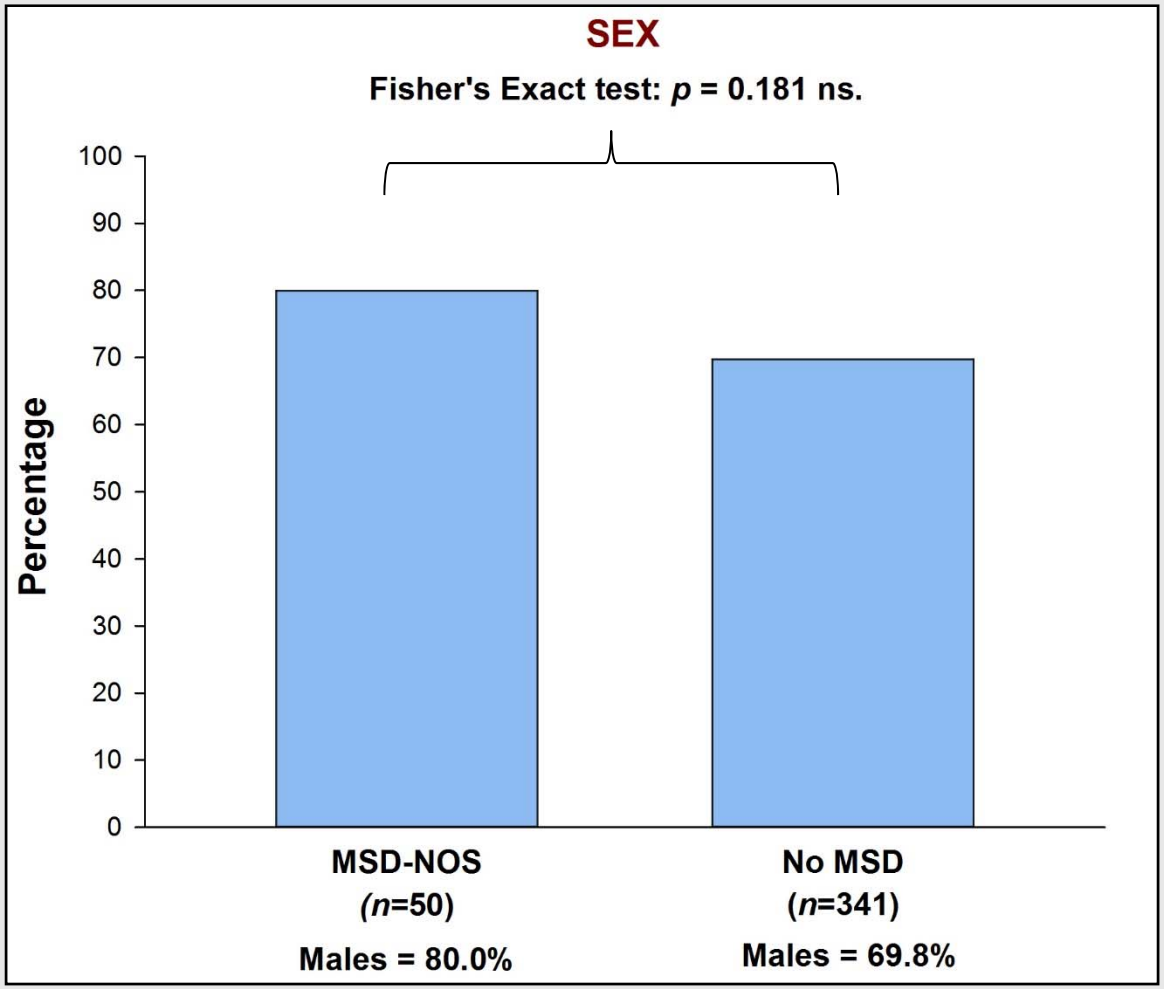
    Range: 8.7%-47.8%

[a] Shriberg, Strand, & Mabie (2016)

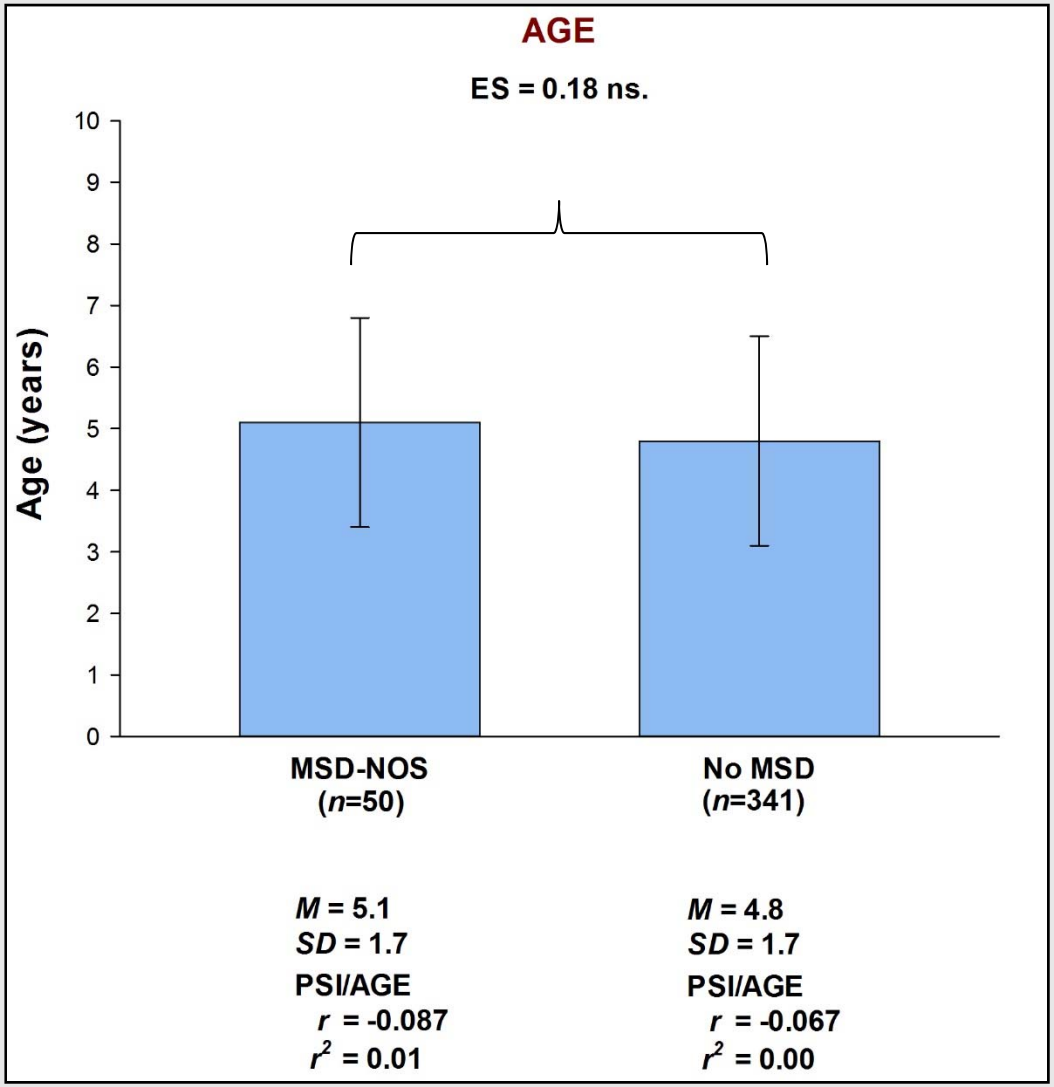
Phenotype Findings in Participants with MSD-NOS Compared to Participants With No MSD

- Demographic
  - Sex
  - Age
- Cognition-Language
  - Intelligence
  - Language
  - Nonword Repetition
    - Performance
    - Encoding
    - Memory
- Speech, Prosody, Voice
  - Consonants
  - Vowels
  - Intelligibility
  - PSI Item Analysis

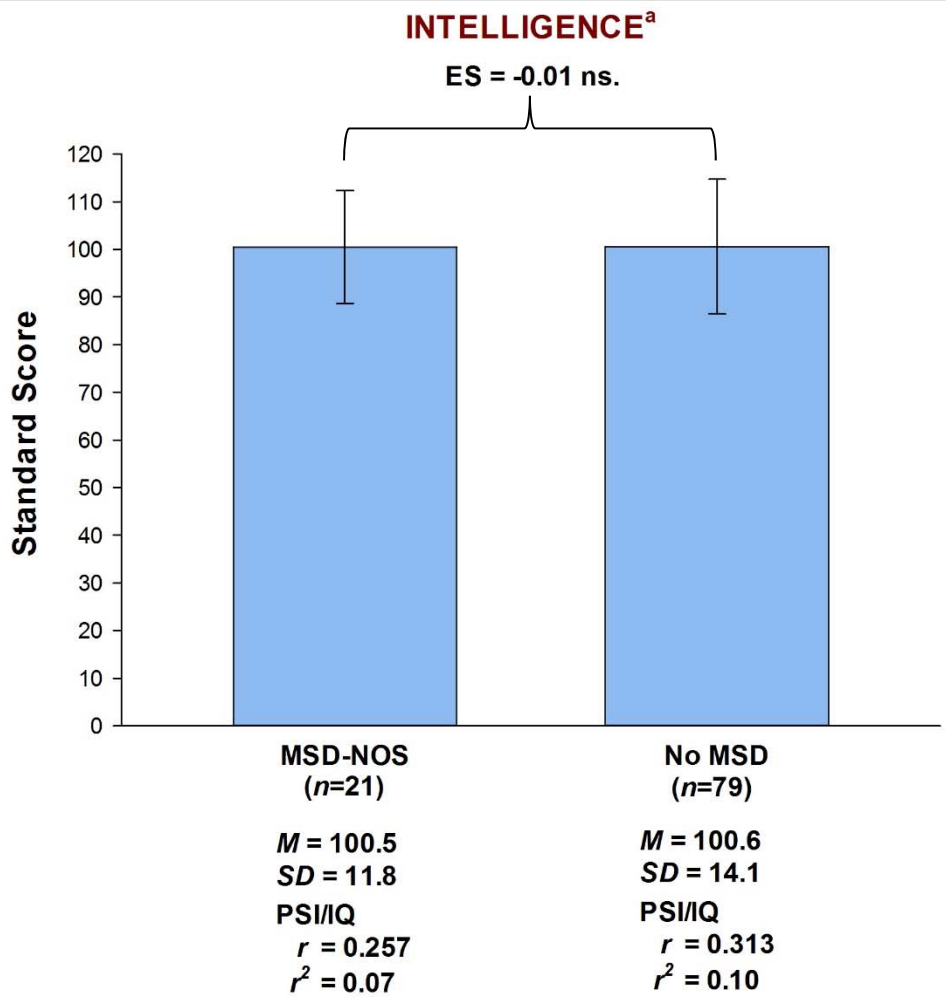
MSD-NOS: Demographic



MSD-NOS: Demographic

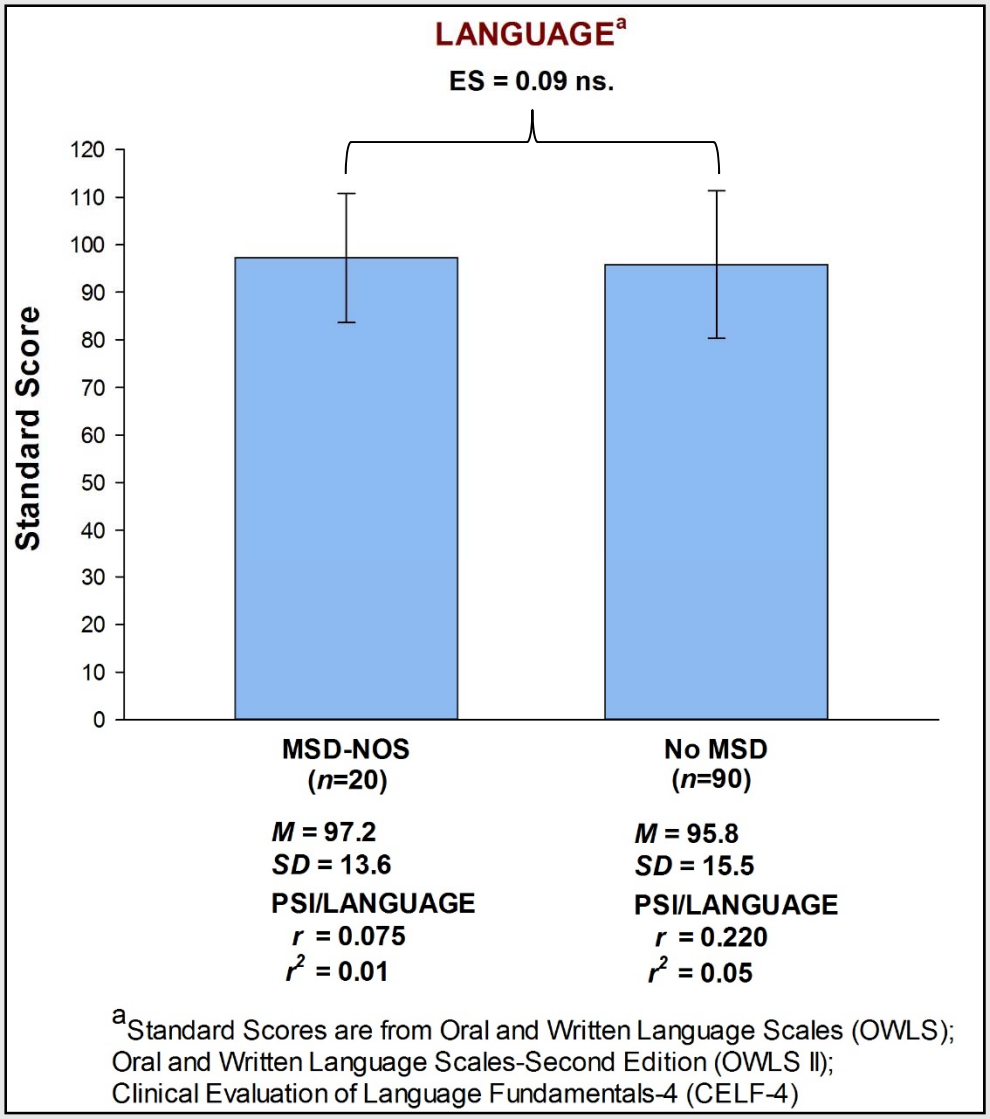


MSD-NOS: Cognition



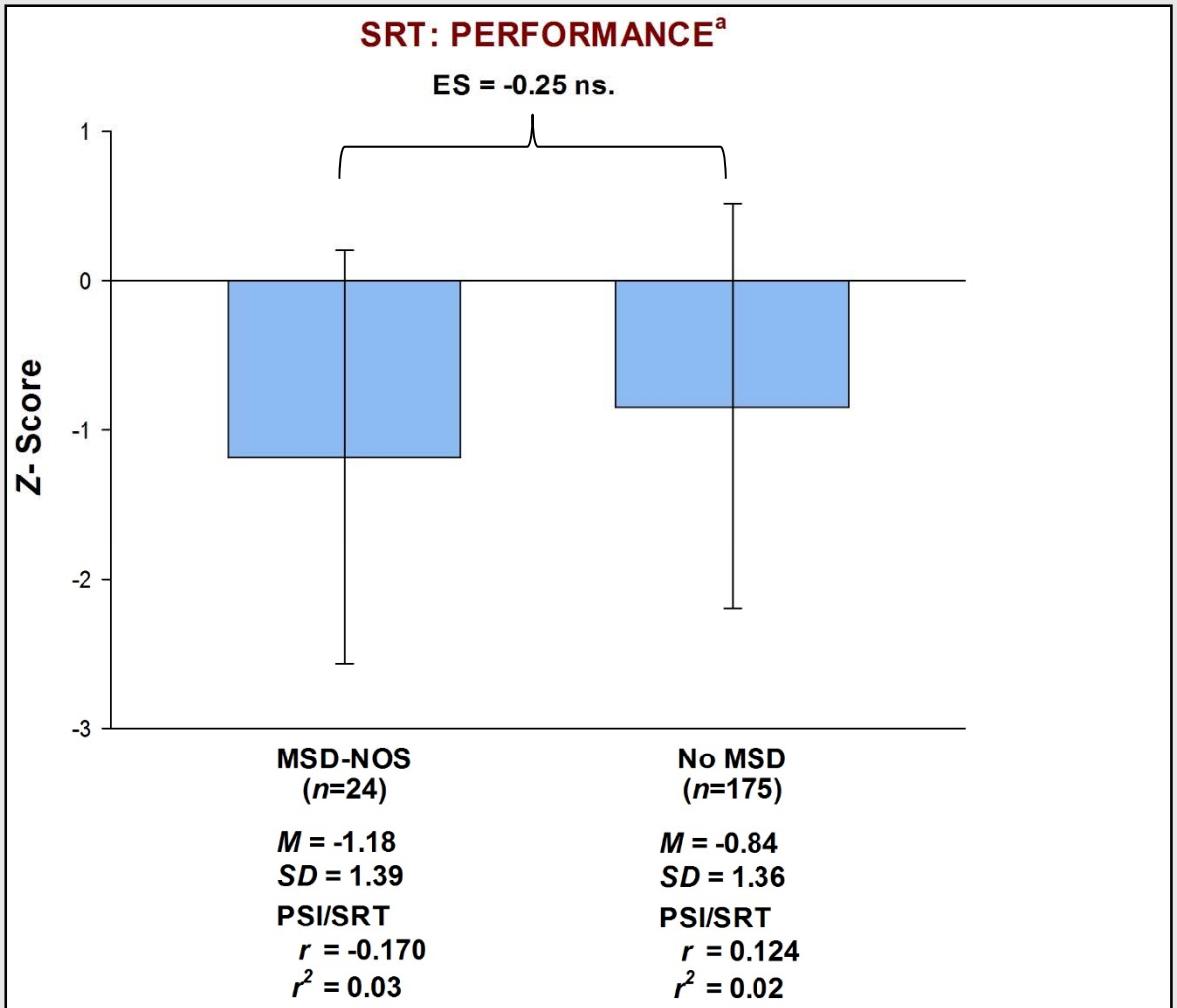
[a]Standard Scores are from Detroit Tests of Learning Aptitude-Primary: Third Edition (DTLA-P:3); Kaufman Brief Intelligence Test-Second Edition (KBIT-2); Weschler Abbreviated Scale of Intelligence (WAIS)

MSD-NOS: Language



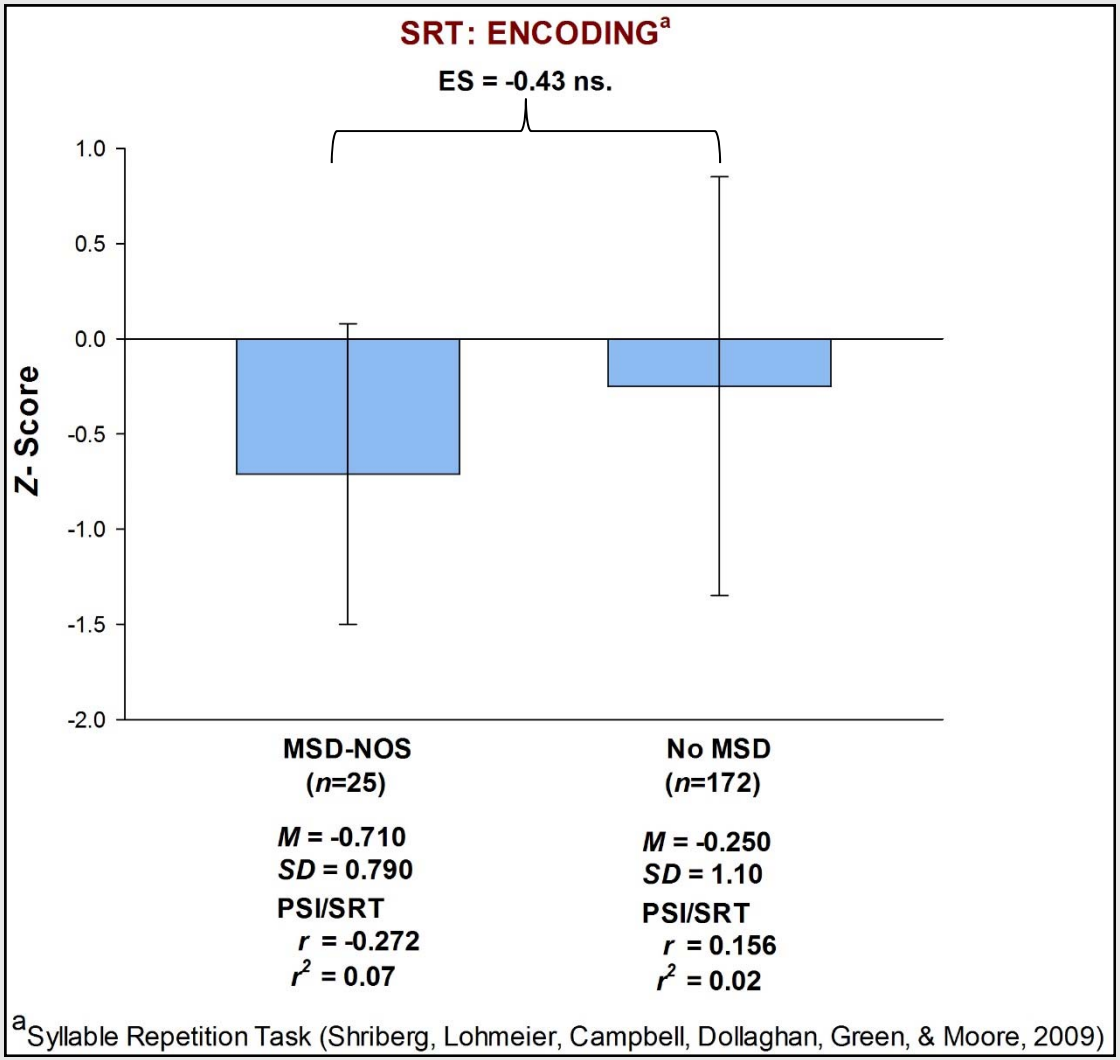
[a]Standard Scores are from Oral and Written Language Scales (OWLS); Oral and Written Language Scales-Second Edition (OWLS II); Clinical Evaluation of Language Fundamentals-4 (CELF-4)

MSD-NOS:
Nonword Repetition Task: Performance

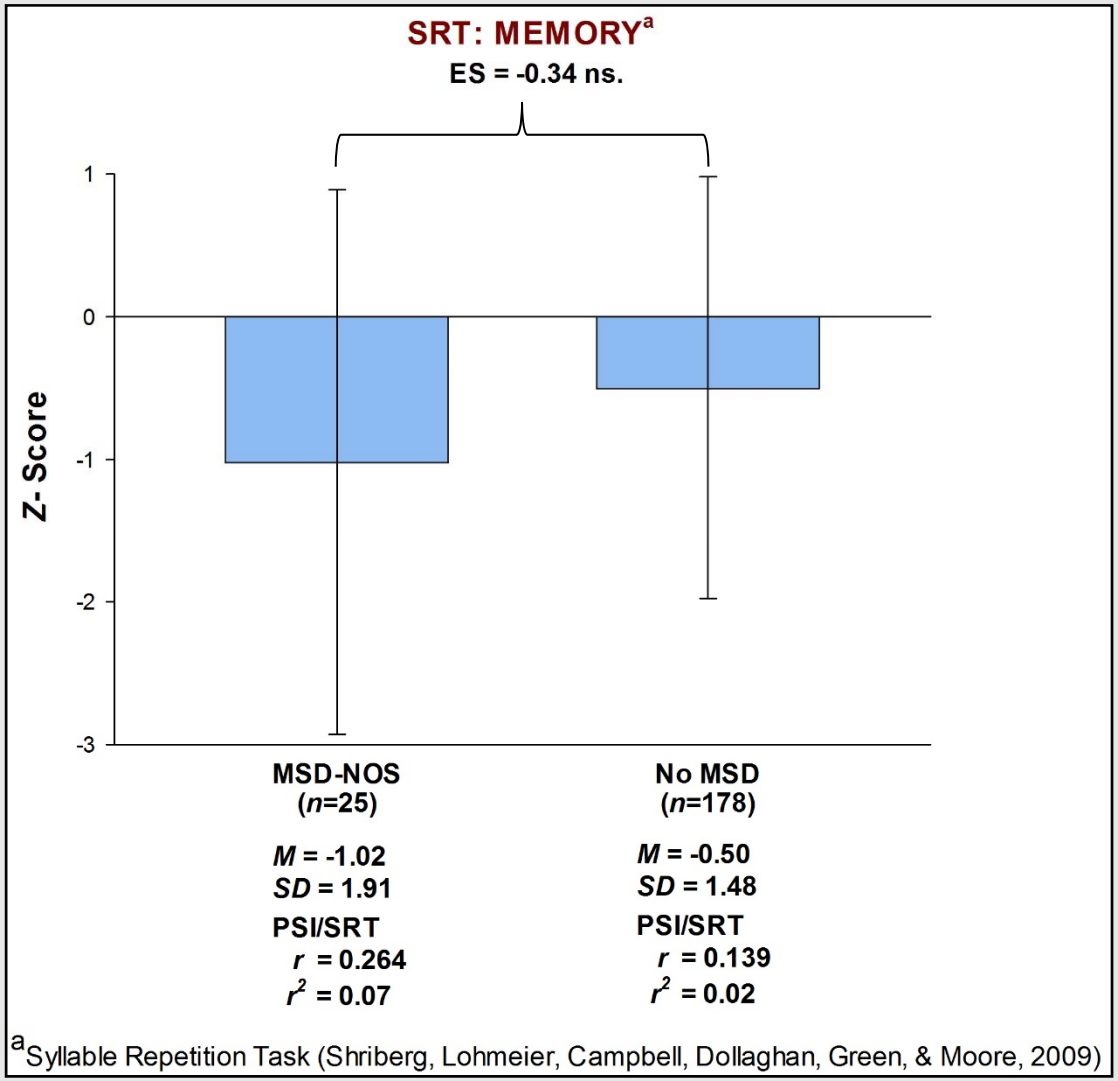


[a]Syllable Repetition Task (Shriberg, Lohmeier, Campbell, Dollaghan, Green, & Moore, 2009)

MSD-NOS:
Nonword Repetition Task: Encoding

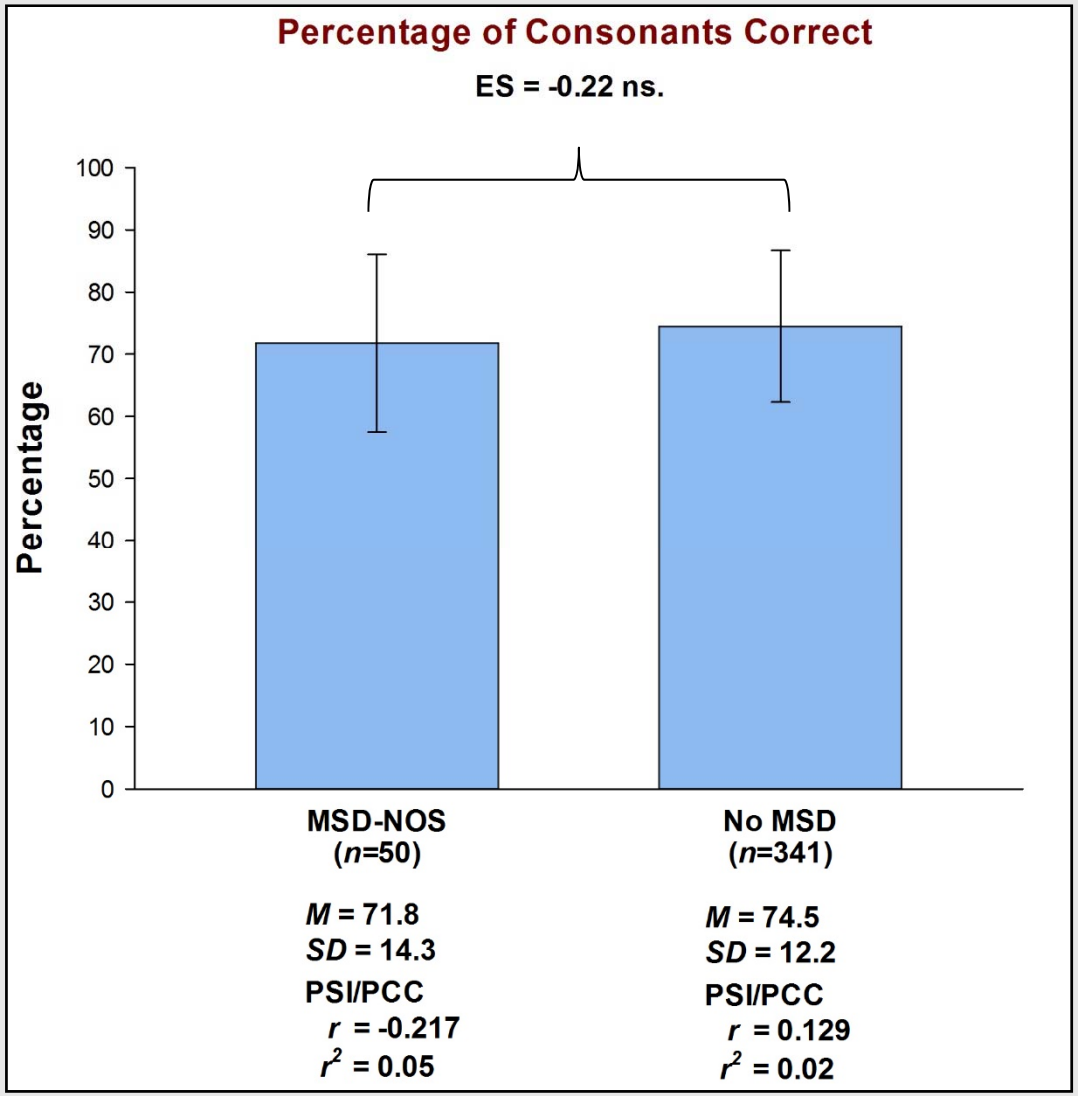


MSD-NOS:
Nonword Repetition Task: Memory

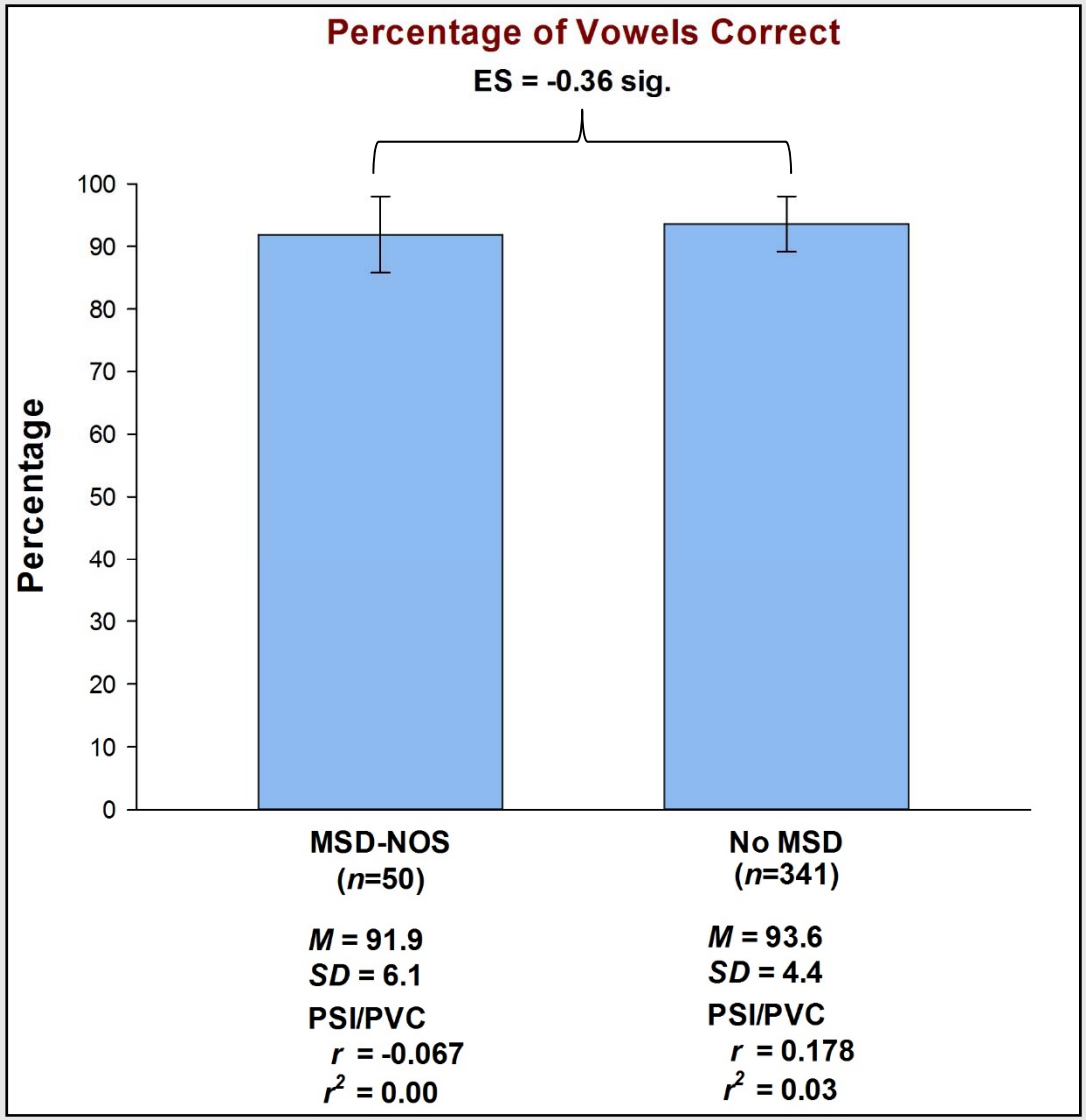


[a]Syllable Repetition Task (Shriberg, Lohmeier, Campbell, Dollaghan, Green, & Moore, 2009)

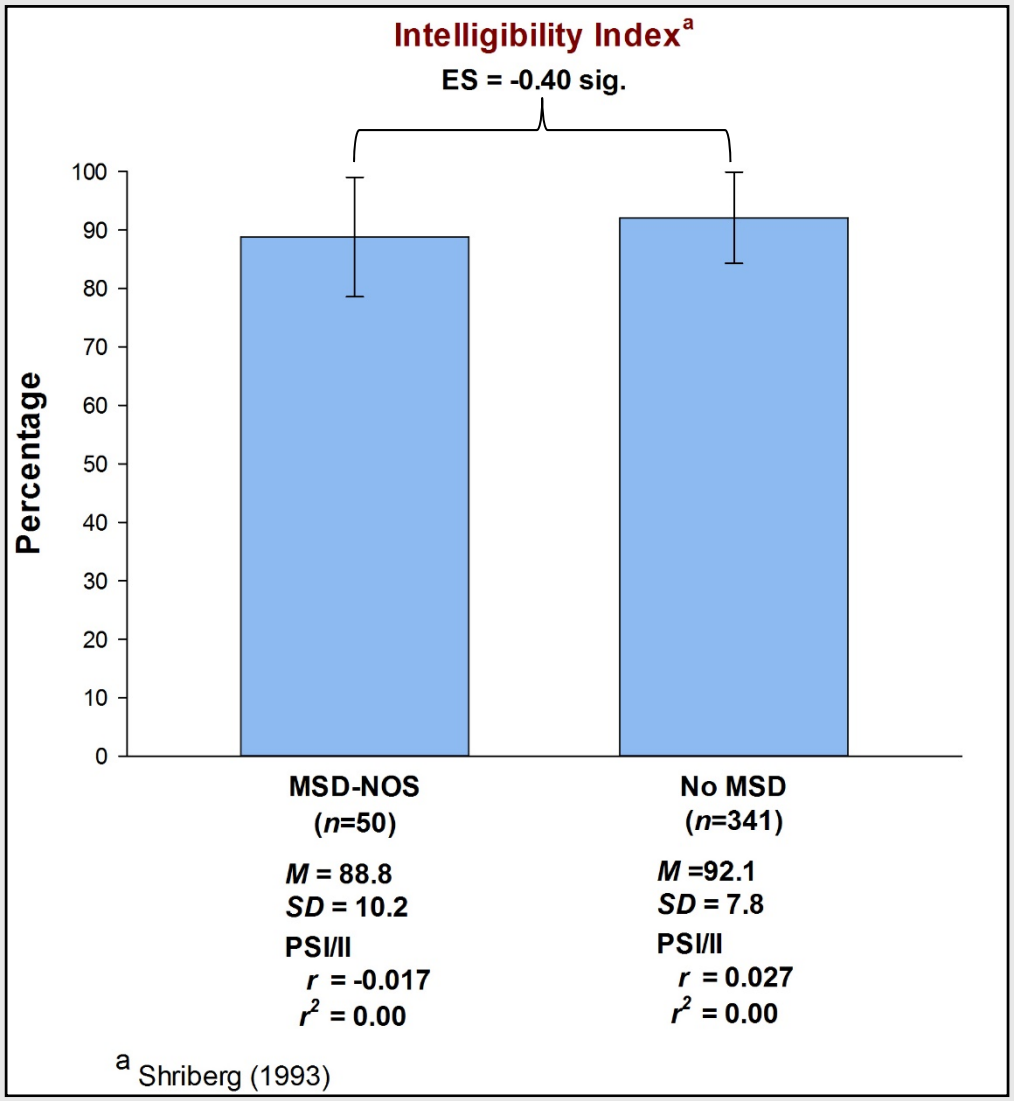
MSD-NOS: Percentage of Consonants Correct



MSD-NOS: Percentage of Vowels Correct



MSD-NOS: Intelligibility



[a] Shriberg (1993)

Item Analyses of PSI: 11 Most Frequently Positive Signs in MSD-NOS[a], Compared to the Frequency of the Same Signs in No MSD

Linguistic Domain	Item No.	Sign Description	MSD-NOS (n=50)	No MSD (n=341)	Test of Proportions
			%	%	p-value
Vowels					
	5	Increased Duration for Diphthongs and Middle Vowels	54.0	22.9	0.000
	6	Reduced % Vowel Phoneme Target Consistency	66.7	32.9	0.004
	7	Reduced % Vowel Target Consistency	68.2	29.1	0.001
Consonants					
	8	Reduced % Correct Glide	52.0	29.9	0.003
	12	Decreased 1st Moment for /s/ Initial Singleton	53.3	44.6	0.434
Vowels and Consonants					
	17	Increased % DMI Class Duration	54.0	24.9	0.000
	18	Increased % of Epenthesis Errors	60.0	30.5	0.000

[a] Signs on which 50% or more of the MSD-NOS participants were positive

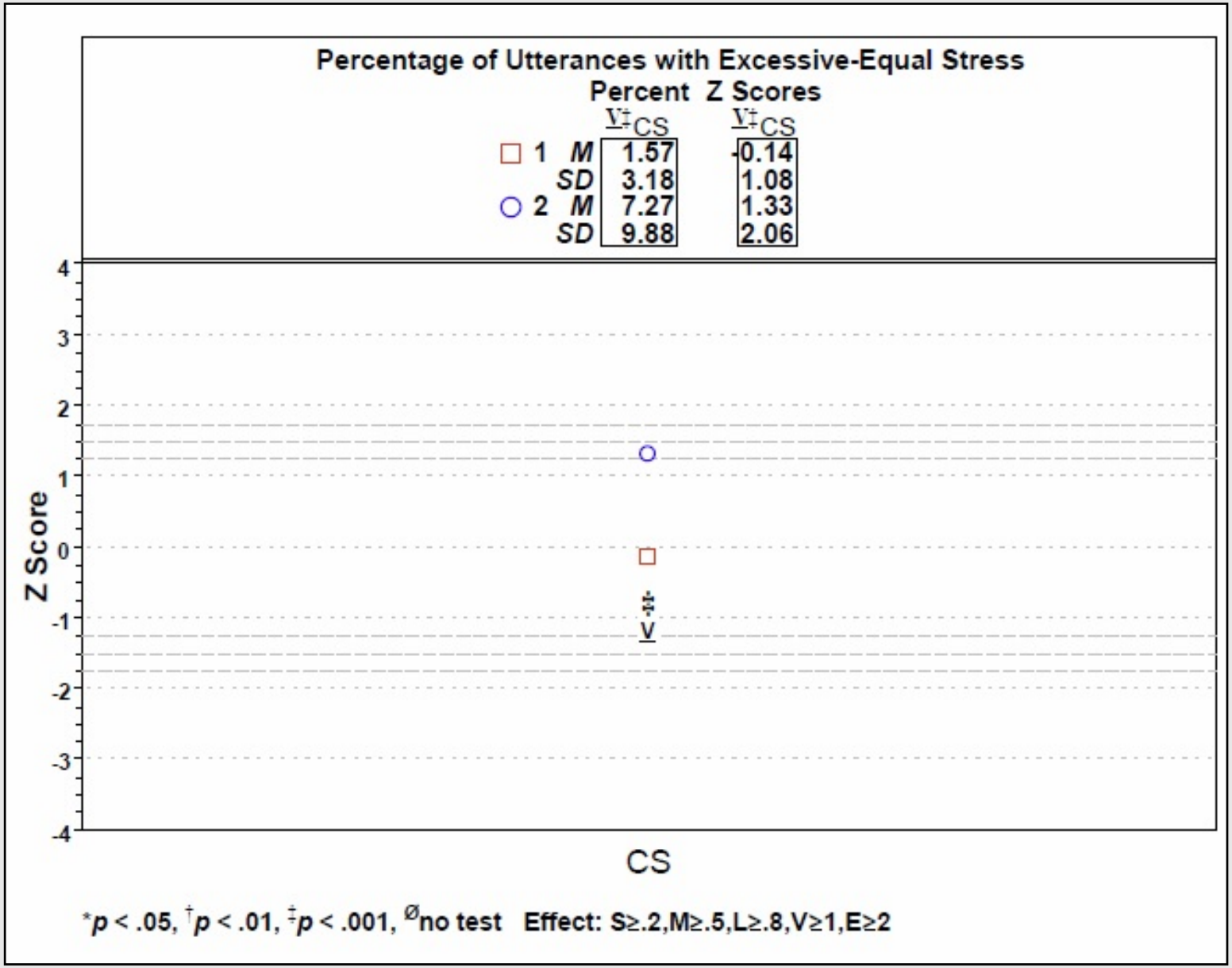
Item Analyses of PSI: 11 Most Frequently Positive Signs in MSD-NOS[a], Compared to the Frequency of the Same Signs in No MSD

Linguistic Domain	Item No.	Sign Description	MSD-NOS (*n*=50)	No MSD (*n*=341)	Test of Proportions
			%	%	*p*-value
Phrasing					
Rate					
	21	Increased Average Syllable ms (without pauses)	50.0	9.1	0.000
Stress					
	22	Increased % of PV15/16 EE codes of all coded utterances. without fast/acceleration. (uncircled & circled)	50.0	11.4	0.000
	23	Increased % of PV15/16 EE codes of all PV15/16 codes. utterances (uncircled & circled)	52.3	10.3	0.000
Loudness					
Pitch					
Laryngeal Quality					
	29	Decreased HNR dB for Vowels	54.0	19.7	0.000
Resonance Quality					

[a] Signs on which 50% or more of the MSD-NOS participants were positive

Some MSD-NOS Phenotype Findings

- Participants with MSD-NOS did not differ from participants with No MSD on demographic or cognitive-language variables
- Participants with MSD-NOS did not have the laryngeal quality deficits in some subtypes of childhood dysarthria
- Participants with MSD-NOS did have many of the precision and stability deficits in speech and prosody described in Childhood Apraxia of Speech
  - Are some children with MSD-NOS being classified as CAS (i.e., false positives for CAS)?


Percentage of Utterances with Excessive-Equal Stress
Percent Z Scores
V‡CS V‡CS
1 M 1.57 -0.14
SD 3.18 1.08
2 M 7.27 1.33
SD 9.88 2.06
Z Score
4
3
2
1
0
-1
-2
-3
-4
CS
*p < .05, †p < .01, ‡p < .001, Ø no test Effect: S≥.2,M≥.5,L≥.8,V≥1,E≥2

Sample Classification Summary Findings: 11-year-old previously positive for MSD-CAS

SDCS (Speech Disorders Classification System) Classification Summary (SCS): Individual								
Speech Classification		Motor Speech Classification					Totals	
Patient 1 (Rice et al., 2012)		No Motor Speech Disorder (NO MSD)	Motor Speech Disorder-Not Otherwise Specified (MSD-NOS)	Motor Speech Disorder-Dysarthria (MSD-DYS)	Motor Speech Disorder-Childhood Apraxia of Speech (MSD-CAS)	Motor Speech Disorder-Dysarthria and Childhood Apraxia of Speech (MSD-DYS & CAS)	n	%
Normal(ized) Speech Aquisition (NSA)[a]								
Speech Errors (SE) Persistent Speech Errors (PSE) (SE/PSE)								
Speech Delay (SD) Persistent Speech Delay (PSD) (SD/PSD)			X					
Totals	n							
	%							

[a] Includes children younger than 9 years old with age-appropriate distortions

Sample Classification Summary Findings: 19-year-old previously positive for MSD-CAS

SDCS (Speech Disorders Classification System) Classification Summary (SCS): Individual								
Speech Classification		Motor Speech Classification					Totals	
Patient 3 (Worthey et al., 2013		No Motor Speech Disorder (NO MSD)	Motor Speech Disorder-Not Otherwise Specified (MSD-NOS)	Motor Speech Disorder-Dysarthria (MSD-DYS)	Motor Speech Disorder-Childhood Apraxia of Speech (MSD-CAS)	Motor Speech Disorder-Dysarthria and Childhood Apraxia of Speech (MSD-DYS & CAS)	n	%
Normal(ized) Speech Aquisition (NSA)[a]			X					
Speech Errors (SE) Persistent Speech Errors (PSE) (SE/PSE)								
Speech Delay (SD) Persistent Speech Delay (PSD) (SD/PSD)								
Totals	n							
	%							

[a] Includes children younger than 9 years old with age-appropriate distortions

Sample Classification Summary Findings: 50-year-old previously and currently positive for MSD-DYS and MSD-CAS

SDCS (Speech Disorders Classification System) Classification Summary (SCS): Individual								
Speech Classification		Motor Speech Classification					Totals	
Participant B (Shriberg et al., 2006; Tomblin et al., 2009)		No Motor Speech Disorder (NO MSD)	Motor Speech Disorder-Not Otherwise Specified (MSD-NOS)	Motor Speech Disorder-Dysarthria (MSD-DYS)	Motor Speech Disorder-Childhood Apraxia of Speech (MSD-CAS)	Motor Speech Disorder-Dysarthria and Childhood Apraxia of Speech (MSD-DYS & CAS)	n	%
Normal(ized) Speech Aquisition (NSA)[a]								
Speech Errors (SE) Persistent Speech Errors (PSE) (SE/PSE)						X		
Speech Delay (SD) Persistent Speech Delay (PSD) (SD/PSD)								
Totals	n							
	%							

[a] Includes children younger than 9 years old with age-appropriate distortions

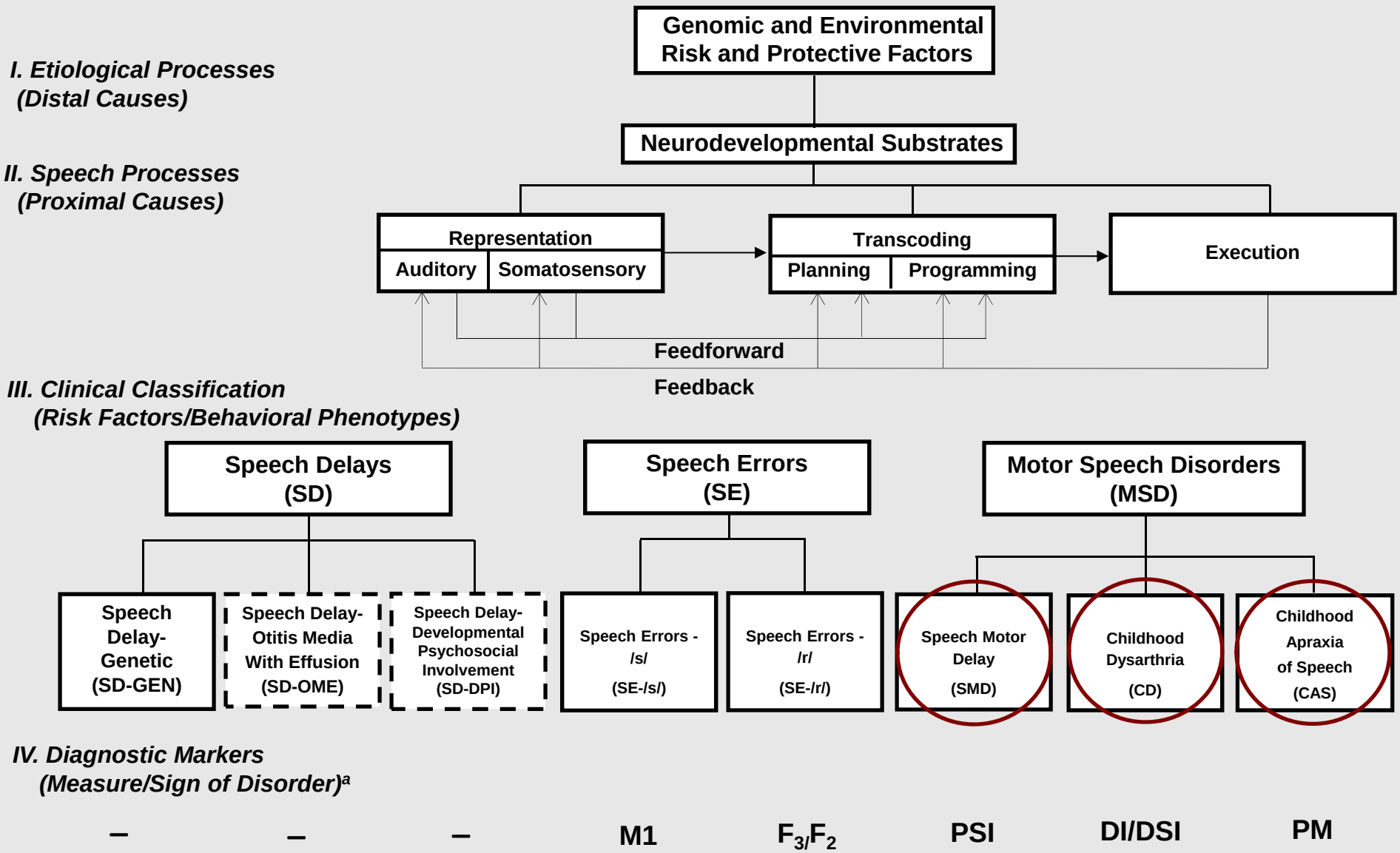
Conclusions

- Findings to date are interpreted as support for a third type of childhood motor speech disorder in addition to dysarthria and apraxia of speech
- The phenotype is consistent with a delay in neuromotor precision-stability (i.e., lower tail of speech motor development)
- Some speakers with dysarthria and/or apraxia of speech also meet requirements for this motor speech disorder (implications for pleiotropic motor genes).

Conclusions

- Research to be reported includes additional phenotype findings from speech tasks and persistence findings from longitudinal samples

- The current placeholder term has been replaced by the term "Speech Motor Delay (SMD)"
  - Adjective: Consistent with other motor skills terms i.e., gross motor, fine motor, oro-motor, "speech motor"
  - Noun: Consistent with other developmental deficits i.e., Language Delay, Speech Delay, "Speech Motor Delay"



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Acknowledgments

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