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ABSTRACT

Recent studies report prevalence, phenotype, and persistence findings for a paediatric motor speech disorder in addition to childhood dysarthria and childhood apraxia of speech termed Speech Motor Delay (SMD). The aim of the present study was to determine if there is a frequent acoustic sign of SMD, with implications for theory, assessment, and treatment.

We examined the frequency of 19 acoustic signs of SMD in audio recordings of continuous speech and word-imitation tasks in three groups of speakers with SMD: 50 children (mean age 5.1 years) with idiopathic speech delay from 6 USA cities; 87 children, adolescents, and adults with eight types of complex neurodevelopmental disorders; and 9 children (mean age 8.8 years) with persistent idiopathic speech delay from a population-based study of children in the South West of England. The 19 acoustic signs of imprecise or unstable speech, prosody, and voice were standardized on typical speakers of the appropriate dialect. The criterion for a frequent acoustic sign was that it occurred in at least 50% of participants with SMD in each of the three groups.

Findings indicated that lengthened mid-vowels and diphthongs was the one sign that met criteria, occurring in 64.4% of the 146 participants with SMD, including 71% of the 87 participants with complex neurodevelopmental disorders. Findings are interpreted to support the potential of this acoustic sign, and possibly several others associated with temporal dimensions of speech sound development, to inform explication of the neuromotor substrates of SMD.

Key Words: acoustics, ALSPAC, motor speech disorders, speech sound disorders

1

A clinical entity termed Speech Motor Delay (SMD) has recently been proposed as a classification for speakers with a delay in the development of speech motor control (Shriberg, Kwiatkowski, & Mabie, 2019). Initial studies of SMD, using the provisional classification *Motor Speech Disorder-Not Otherwise Specified* (Shriberg, 2017), described a measure to identify children with Speech Delay and concurrent SMD termed the Precision-Stability Index (PSI: Shriberg et al., 2010a). In a study using the PSI to estimate the prevalence of SMD in 415 children with idiopathic Speech Delay in six USA cities (mean age: 5.1 years; standard deviation: 1.7 years), the group-averaged prevalence of SMD concurrent with Speech Delay was 12% (Shriberg, Kwiatkowski, & Mabie, 2019). In a study using the PSI to estimate the prevalence of SMD in 346 children, adolescents, and adults with eight types of complex neurodevelopmental disorders (CND), the group-averaged prevalence of SMD in the eight CND was 25.1% (Shriberg, Strand, Jakielski, & Mabie, 2019). In a retrospective longitudinal study of 14 participants in treatment for idiopathic Speech Delay, 11 (78.6%) participants normalized SMD before 9 years of age, and SMD persisted until at least late adolescence in the remaining 3 (21.4%) of participants (Shriberg, Campbell, Mabie, & McGlothlin, 2019). The latter study provided the longitudinal support for replacing the provisional classification term Motor Speech Disorder-Not Otherwise Specified with the present term, SMD. Specifically, most children in the retrospective longitudinal study resolved SMD without direct treatment of the motor speech component, supporting the construct of a developmental delay in speech motor development. A study by Namasivayam et al. (2018) is the first to report treatment effects using principles of motor learning with a group of children classified descriptively as having SMD.

The SMD study by Shriberg, Campbell, et al. (2019) cited above also included an analyses of the 10 most frequent perceptual or acoustic signs of SMD. The 10 signs that met the

frequency criterion (over 50% of participants positive on the sign) included age-inappropriate

precision and stability behaviours in speech, prosody and voice domains, with 7 of the 10 most

frequent signs involving speech production. Of present interest is that 4 of the 10 signs used

acoustic procedures (Shriberg, Campbell, et al., 2019, Table 4). Automated acoustic assessment

methods in speech pathology have several attractive measurement features relative to perceptual

methods, including sensitivity to continuous variables, reliability, and efficiency (cf. McAllister

& Ballard, 2019).

Statement of Purpose

The purpose of the present initial study was to determine if there is one or more prevalent

acoustic signs of SMD that could be informative in studies to explicate core motor speech

processes underlying SMD. A prevalent speech motor sign of SMD was defined as one on which

at least 50% of speakers with SMD have a z-score greater than 1.25 standard units from the mean

of typical speakers of the same age and sex. We address the question using audio-recorded

speech samples from three studies, with each study having the potential to provide alternative

sources of internal and external validity support for one or more prevalent signs of SMD.

<p align="center">METHOD</p>

Participants

Table 1 includes descriptive information for three groups of participants whose audio-

<p align="center">Insert table 1 about here</p>

recorded speech samples were analysed for the present study, and three groups of typically-

developing speakers used to standardize participant scores on the measures to be described. All

participants were assented and/or consented using procedures approved by Internal Review

Boards at the University of Wisconsin-Madison, the Mayo Clinic-Rochester, Minnesota, the

<p align="center">3</p>

University of Bristol (ALSPAC Ethics and Law Committee and the Local Research Ethics Committees), and collaborative research sites in five other USA cities (Cincinnati, Cleveland, Dallas, Denver, and Pittsburgh).

  USA group. Participants in the USA group were identified from a database of 415 children recruited for research in Speech Delay in six USA cities (Shriberg, Kwiatkowski, & Mabie, 2019). All participants met criteria for idiopathic Speech Delay using Speech Disorders Classification System (SDCS) software (Shriberg, 1993, Appendix). As shown in table 1, 50 of these participants also met criteria (to be described) for concurrent SMD and 341 participants from this database used as a control group met criteria for No Motor Speech Disorder. Participants' average ages in the USA participant and control groups, respectively, were 5.1 years (1.7) and 4.8 years (1.7) years. All participants had General (i.e., mainstream) American dialects. The approximately two-hour assessment protocol for the six samples included a five to eight minute audio-recorded conversational speech task. For participants in 3 of the six USA cities, the protocol included two audio-recorded vowel/diphthong word-imitation tasks presented by computer. The first task included pictures of words containing the four corner vowels (beet, bat, boot, pot) in four randomized lists, with each picture accompanied by an audio recording of each word spoken by an experienced female speech pathologist. The second task included 11 CVC words containing six mid-vowels (bit, pet, putt, Bert, put, bought) and the 5 diphthongs (bite, bait, boat, boy, pout) presented in four randomized lists accompanied by an audio recording of each word spoken by the same speech pathologist. Stimuli were presented free field at a comfortable loudness level. Participants were asked to repeat each word exactly the way the woman said it.

Complex Neurodevelopmental Disorders group. A total of 87 participants with SMD were identified from a database of 346 children, adolescents, and adults with one of eight types of CND recruited for speech-genetics and behavioural speech research (Shriberg, Strand, et al., 2019). As shown in table 1, 181 of the 346 participants met criteria for No Motor Speech Disorder. Participants' average chronological age was 15.4 years (11.2) in the CND SMD group and 11.7 years (10.6) in the CND No Motor Speech Disorder group. All participants had General American dialects. CND participants completed assessment protocols similar to the protocol used for participants in the USA group, which included a conversational speech sample for all participants. Because the vowel tasks were developed relatively recently, only some participants in three of the smaller size CND groups had been administered the two word-imitation tasks.

Avon Longitudinal Study of Parents and Children group. The Avon Longitudinal Study of Parents and Children (ALSPAC) dataset (Wren, McLeod, White, Miller, & Roulstone, 2013; Wren, Miller, Peters, Emond, & Roulstone, 2016; Wren, Roulstone, & Miller, 2012) provided an opportunity to examine acoustic signs of SMD in children with idiopathic Speech Delay who differed in several ways from the participants in the USA and CND groups. ALSPAC is a large-scale longitudinal prospective population study which has followed the health and development of a cohort of children from pre-birth into adulthood. Pregnant women resident in the geographical area previously known as Avon in the South West of England, UK, with expected dates of delivery 1st April 1991 to 31st December 1992 were invited to take part in the study. The initial number of pregnancies enrolled was 14,541. Of these initial pregnancies, there was a total of 14,676 foetuses, resulting in 14,062 live births and 13,988 children who were alive at 1 year of age (Boyd, Golding, Macleod, Lawlor, Fraser, Henderson, Molloy, Ness, Ring, Davey Smith, 2013; Fraser, Macdonald-

Wallis, Tilling, Boyd, Golding, Davey Smith, Henderson, Macleod, Molloy, Ness, Ring, Nelson & Lawlor, 2013). The study website contains details of all the data that is available through a fully searchable data dictionary and variable search tool and references the following webpage: http://www.bristol.ac.uk/alspac/researchers/our-data/

At age 8, all children in the cohort were invited to attend a clinic to undertake a battery of assessments measuring health and development across a number of domains, including speech and language. Audio-recorded speech samples were available from 156 children who met ALSPAC investigators criteria for Persistent Speech Disorder from the total of 7390 children who attended the 8-year assessment clinic. Thus, participants in this cohort differed from the USA cohorts in that they were sampled from a population-based study, spoke a different dialect of English than General American, had been classified as having idiopathic Persistent Speech Delay using a research protocol administered and transcribed by a different group of speech-language researchers, and were approximately 4 years older at assessment than participants in the USA group.

The assessment data for the ALSPAC study included audio-recorded continuous speech from three picture description activities (Wren et al., 2013). For the first picture, the child was asked to describe a community scene as if to someone who was not present and therefore could not see the scene. For the second picture the child was shown a map and asked to describe the shortest directions from one location to another. For the third task which also used pictures, the child was asked to explain the sequence of steps in putting batteries into a flashlight (torch). For the present study, as described in following sections, a file that included the three continuous speech samples was used to classify participants' status on the acoustic items of the measure used to identify speakers with SMD.

Reference databases. The database used to standardize measures for USA participants included 150 3- to 18-year-old typically-developing children with mainstream General American dialects (Potter et al., 2012). The reference data for the CND group included the 150 children and adolescents, plus data from 50 20- to 80-year-old adults with General American dialects, including approximately 10 participants per decade (Scheer-Cohen et al., 2013). The assessment protocols for both standardization samples included a conversational sample and the two word-imitation tasks. The database used to standardize the three continuous speech samples from the ALSPAC group included audio-recordings of the three picture description activities by 25 8-year-old typically-developing speakers from the same Avon, UK region (Wren et al., 2013).

Data Reduction

Four research specialists completed narrow phonetic transcription, prosody-voice coding, and acoustic analyses of the audio-recorded conversational speech samples and the two word-imitation tasks in the USA and CND groups using procedures developed for research in speech sound disorders (Shriberg et al., 2010b). For the three types of data reduction, randomly selected, 20% intrajudge and interjudge point-to-point percentages of agreements for 26 perceptual and acoustic variables ranged from 74.9% to 97.3% for the USA samples (Shriberg, Kwiatkowski, & Mabie, 2019) and 69.7% to 97.5% for the CND samples (Shriberg, Strand, et al., 2019).

Because pilot studies indicated that dialectal differences precluded reliable narrow phonetic transcription and prosody-voice coding of the ALSPAC picture task data by the American research specialists, only data from the acoustic analyses of the ALSPAC files were processed for the present study (to be described). An interjudge reliability estimate reported previously was obtained for the four British transcribers who transcribed all eight-year assessment data by having a transcriber who participated in previous years' assessments

7

transcribe 48 randomly selected files from the eight-year assessment (Wren et al., 2013). Chronbach's alpha coefficients were 0.78 for the Percentage of Consonants Correct-Adjusted and 0.87 for the Percentage of Consonants Correct Late-8.

Speech and Motor Speech Classification

USA and CND groups. Participants in the USA and CND groups met perceptual-based criteria for Normal Speech Acquisition, Speech Delay, or Persistent Speech Delay (if older than 9 years of age), using a prior version of the Speech Disorders Classification System (Shriberg, Austin, Lewis, McSweeny, & Wilson, 1997, Appendix A). Participants in both groups also met perceptual and acoustic criteria for one of five concurrent, mutually exclusive motor speech classifications described in a previous paper in this issue (Shriberg, Kwiatkowski, & Mabie, 2019): No Motor Speech Disorder, Speech Motor Delay, Childhood Dysarthria (CD), Childhood Apraxia of Speech (CAS), or concurrent Childhood Dysarthria and Childhood Apraxia of Speech (CD & CAS). The Speech Disorders Classification System software for motor speech disorders uses findings from three standardized perceptual-acoustic measures — the Precision-Stability Index, the Dysarthria Index, and the Pause Marker (Shriberg et al., 2017a) — to classify children into one of the five motor speech classifications. A Supplement for this research series provides detailed information on classification methods and measures [   ].

Figure 1 is a manual copy of the PSI assessment form, the measure used to identify

Insert figure 1 about here

speakers with early or persistent SMD. The PSI includes 13 perceptual and 19 acoustic signs of imprecise and/or unstable speech, prosody and voice in continuous speech. As shown in figure 1, the 32 signs are subsumed within ten linguistic domains. Performance on a sign greater than 1.25 standard deviations in the negative direction from typical speakers of same age and sex is coded

as positive for the sign. Speakers with more than 30% positive signs are classified as SMD. Speakers who are positive for CD and/or CAS may also be positive for SMD. For such outcomes, the classification software assigns the motor speech classification to the putatively more severe motor speech classification (i.e., CD and/or CAS). Thus, the 50 participants in the present USA group and 87 in the present CND group met motor speech classification criteria for only SMD.

   ALSPAC group. Due to the reliability constraints on the narrow phonetic transcription and prosody-voice coding of the British speech samples by the USA transcribers, the speech classification of the 156 ALSPAC speakers used findings reported in Wren et al. (2013). Specifically, classification of participants as Persistent Speech Disorder was based on the speech assessment measures and criteria described in Wren et al. (2013) and Wren et al. (2016), and classification of the delay as idiopathic was based on the extensive developmental data obtained for each participant in the same publications.

   Acoustic analyses of the 156 ALSPAC participants with Persistent Speech Disorder had concurrent SMD. First, each participant's responses to the three picture tasks were combined into one digital file. Second, two of the four USA research specialists completed acoustic analyses of the 19 acoustic items of the PSI (figure 1) using the same procedures as used for acoustic analyses of participants in the USA and CND groups. ALSPAC participants who were positive on more than 30% of the acoustic signs (i.e., who had a PSI score lower than 70% on the 19 signs) using the Bristol reference data (table 1) were classified as positive for SMD. It is important to note that due to this accommodation (i.e., using only the 19 acoustic signs in the PSI) the number of ALSPAC participants with SMD (n=9) is not an estimate of the prevalence of SMD concurrent with idiopathic Persistent Speech Disorder in this population-based sample.

A Frequent Sign of Speech Motor Delay

Figure 2 includes findings for the question posed: is there is one or more frequent

Insert figure 2 about here

acoustic signs of SMD that could be informative in studies to explicate the core motor speech

processes underlying SMD? The numbers below each bar in figure 2 correspond to the numbered

signs in figure 1, and the rounded percentage above each bar is the percentage of participants in

each group positive on the sign. For the present purposes, the 19 acoustic signs are aggregated

into eight speech, prosody, and voice domains, with each domain including 1-3 acoustic signs.

The left-to-right sequence of the eight domains and the signs within each domain are ordered

approximately by the highest percentage of total participants in the three groups or in just the

USA and CND groups positive on the domain.

As shown in figure 2, the most frequent of the 19 acoustic signs of SMD in the

continuous speech of participants in the three groups was PSI Sign No. 5: Increased Duration of

Mid-Vowels (Monophthongs) and Diphthongs (cf., figure 1). This acoustic sign was the only one

of the 19 on which over 50% of participants with SMD in the three groups were positive. As

shown, 71% of participants with SMD in the CND group had longer durations averaged over the

11 phonemes in the continuous speech samples than typical speakers of their chronological age

and sex. Inspection of the CND samples indicated that this was the case for 12 of the 12 (100%)

participants with Down syndrome, 7 of the 7 (100%) participants with Galactosemia, 5 of the 6

(83.3%) participants with Autism Spectrum Disorder, 4 of the 5 (80%) participants with 22q11.2

deletion syndrome, 22 of the 30 (73.3%) participants with 16p11.2 deletion, and 5 of the 8

(62.5%) participants with fragile X syndrome. Other signs of SMD in figure 2 (to be discussed)

met the >50% criteria for one or two of the three groups, including other vowel signs and signs associated with reduced rate, but PSI Sign No. 5 was the only sign meeting the three-group criterion for a frequent acoustic sign of SMD.

Vowel/Diphthong Durations in Conversational Speech and in a Word-Imitation Task

Table 2 includes descriptive and inferential statistical findings for PSI Sign No. 5:

Insert table 2 about here

Increased Duration of Mid-Vowels and Diphthongs for participants in the two groups (USA and CND) in each of the speech tasks (continuous speech and word-imitations). Unlike the word-imitation task in the Madison Speech Assessment Protocol that assesses corner vowels (Vowel Task 1), the word-imitation task that assesses mid-vowels and diphthongs had been administered to only three of the CND groups yielding only 11 participants with SMD in each group with available data on this task.

As indicated in table 2, three of the four comparisons of PSI Sign No. 5 findings yielded significant differences in the average duration of vowels/diphthongs, as determined by confidence intervals that did not cross 0.00. The primary finding in table 2 is that when sampled in continuous speech, the average durations of the 11 mid-vowels/diphthongs were significantly longer for the participants with SMD than for participants with No Motor Speech Disorder in both the USA (effect size = 0.63) and CND (effect size = 0.43) groups. A secondary finding was that as sampled in the word-imitation task, participants in the CND group with SMD had significantly longer average durations of the 11 mid-vowels/diphthongs than participants in the CND group with No Motor Speech Disorder (effect size = 1.16).

Figure 3 provides a graphic summary of the averaged percentage of increased durations

Insert figure 3 about here

11

of the six vowels and five diphthongs from the continuous speech samples of participants with

Speech Delay and SMD, compared to durations of these phonemes from the continuous samples

of participants with Speech Delay and No Motor Speech Disorder. The plotted values are the

averaged percentage of increased duration for each phoneme (i.e., Mean Duration for

participants with No Motor Speech Disorder – Mean Duration for participants with SMD/Mean

Duration for participants with No Motor Speech Disorder x 100). The dashed line at 20% is an

arbitrary visual aid that demarcates the four phonemes with the longest durations from the data

points for the remaining seven phonemes. As shown, participants with SMD averaged 39.6% and

21.8% longer durations on the two lax front vowels /ɪ/ and /ɛ/, respectively, 25.6% longer

durations on the stressed mid vowel /ɝ/, and 28.6% longer durations on the non-phonemic

diphthong /oʊ/.

DISCUSSION AND CONCLUSION

Findings

The primary finding for the question posed is that compared to participants with no motor speech

disorder, increased duration of mid-vowels and diphthongs was a frequent sign of SMD in

children with idiopathic speech delay. The finding was supported in participants with some types

of CND. Findings suggest that additional research focused on the durations of the front vowels,

/ɪ/ and /ɛ/ may be particularly informative for theory. Unlike the maximal spatial positions of the

jaw, tongue, and lips required for the four tense corner vowels (PSI Sign No. 4) and the complex

tongue and lips configurations required of the rhotic /ɝ/ and spatiotemporal requisites of the

diphthong /oʊ/ (figure 3), the lax monophthongs /ɪ/ and /ɛ/ are made with neutral tongue, jaw,

and lip positions (Shriberg, Kent, McAllister, & Preston, 2019). Because the spatial requisites of

/ɪ/ and /ɛ/ are not difficult in continuous speech, research on their temporal control in motor

speech development may provide insights into core speech motor control processes in SMD. One

possibility is that the increased durations of vowels in speakers with SMD is due to difficulties in

planning the phonological phrase. As reported in a study of childhood apraxia of speech by

Nijland et al. (2003), whereas children with typical speech learn to shorten the duration of an

unstressed syllable in a prosodically weak position, children with childhood apraxia of speech

(and perhaps SMD) have substantial difficulty acquiring the appropriate durations of unstressed

syllables (e.g., Arcuili & Ballard, 2017; Kehoe, Stoel-Gammon, & Buder, 1995). Comparative

research is needed to determine if the increased durations of /ɪ/ and /ɛ/ in children with SMD are

similar to the increased durations of these phonemes in the speech of children with CAS, perhaps

reflecting developmental constraints in feedforward processes in speech motor development

(Terband, Maassen, Guenther, & Brumberg, 2014). As described in Shriberg, Kwiatkowski, and

Mabie (2019) and Shriberg, Campbell, et al., (2019), a speculation is that false positive

diagnoses of CAS could be due, in part, to such overlaps in the phenotypes of childhood apraxia

of speech and SMD.

      Although they did not meet the arbitrary criterion for a frequent sign of SMD (i.e., >50%

occurrence in participants in all three research groups), other findings in figure 2 support the

potential deficits in temporal processes implied in the significant findings for PSI Sign No. 5. A

total of 58% of participants with CND were positive on PSI Sign No. 4: Increased Duration of

Corner Vowels and 50%-56% of participants in the USA and CND studies were positive on PSI

Sign No. 21: Increased Syllable Length and/or PSI Sign No. 20: Reduced Syllables per second (without pauses). Decreased articulation rate (i.e., speaking time minus pause time) can be a function of increased vowel and consonant lengths. The remaining PSI signs on which over 50% of participants in at least one group were are positive do not suggest immediate interpretation. Notably, for the widely studied domain of vowel space in the motor speech literature, neither of the two larger of the three groups had over 50% of participants positive on any of the three PSI signs of decreased vowel space.

Research and Clinical Directions

Studies to replicate and extend the present initial findings with participants selected to control for relevant threats to internal and external validity would appear to be warranted. Specifically, replication studies of PSI Sign No. 5 should be undertaken in studies with sufficient statistical power to control for possible associations of SMD with relevant demographic, sensory, cognitive, speech, language, and treatment history variables. If the present and/or additional acoustic signs are cross-validated, clinical directions for assessment and treatment of SMD include the possible development of automated assessment applications.

Conclusion

As proposed in the first three articles in the present research series, the precision and stability deficits in SMD are posited to result from processing constraints at the execution stage of speech production (Shriberg, Kwiatkowski, & Mabie, 2019, Figure 1). The present findings, however, are more consistent with the conclusion in Shriberg, Campbell, et al. (2019) that there likely are common neurodevelopmental pathways underlying the four SDCS classifications of motor speech disorder phenotypes—SMD, CD, CAS, and CD & CAS. Prevalence estimates in Shriberg, Strand, et al. (2019) indicated that CD & CAS was at least as prevalent as CAS in

some of the study groups, suggesting common genomic and neurodevelopmental pathways. In the present study, the high percentage of children with SMD with increased durations of vowels that are not difficult to articulate could reflect transcoding (planning/programming) deficits associated with stress that are similar to the transcoding deficits proposed in CAS (Shriberg et al., 2017b). The likelihood of identifying causal genes and copy number variants for SMD that confer risk for pleiotropic effects within and among speech, language, and motor domains is consistent with contemporary trends in genomic research in verbal trait disorders (e.g., Lewis, Iyengar, & Stein, 2018; Miscmarra et al., 2007; Peter, Matsushita, & Raskind, 2012; Smith, Pennington, Boada, & Shriberg, 2005; Stein et al., 2006; Stein et al., 2004; Truong et al., 2016).

ACKNOWLEDGMENTS

DECLARATION OF INTEREST

The authors report no declarations of interest.

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Figure 1. The Precision-Stability Index (PSI).

Figure 2. Acoustic signs of Speech Motor Delay in three study groups. The groups are USA = United States of America, CND = Complex Neurodevelopmental Disorders, and ALSPAC = Avon Longitudinal Study of Parents and Children.

Figure 3. Average duration (ms) of the 11 phonemes in PSI 5: Increased Duration of Mid-Vowels and Diphthongs in the continuous speech tasks from participants with idiopathic Speech Delay and Speech Motor Delay.