Speech, Prosody, and Voice Characteristics of a Mother and Daughter With a 7;13 Translocation Affecting *FOXP2*

Lawrence D. Shriberg
University of Wisconsin—Madison

Kirrie J. Ballard
J. Bruce Tomblin
University of Iowa, Iowa City

Joseph R. Duffy
Mayo Clinic, Rochester, MN

Katharine H. Odell
Meriter Hospital, Madison

Charles A. Williams
University of Florida, Gainesville

Purpose: The primary goal of this case study was to describe the speech, prosody, and voice characteristics of a mother and daughter with a breakpoint in a balanced 7;13 chromosomal translocation that disrupted the transcription gene, *FOXP2* (cf. J. B. Tomblin et al., 2005). As with affected members of the widely cited KE family, whose communicative disorders have been associated with a point mutation in the *FOXP2* gene, both mother and daughter had cognitive, language, and speech challenges. A 2nd goal of the study was to illustrate in detail, the types of speech, prosody, and voice metrics that can contribute to phenotype sharpening in speech-genetics research.
Method: A speech, prosody, and voice assessment protocol was administered twice within a 4-month period. Analyses were aided by comparing profiles from the present speakers (the *TB* family) with those from 2 groups of adult speakers: 7 speakers with acquired (with one exception) spastic or spastic-flaccid dysarthria and 14 speakers with acquired apraxia of speech.
Results: The descriptive and inferential statistical findings for 13 speech, prosody, and voice variables supported the conclusion that both mother and daughter had spastic dysarthria, an apraxia of speech, and residual developmental distortion errors.
Conclusion: These findings are consistent with, but also extend, the reported communicative disorders in affected members of the KE family. A companion article (K. J. Ballard, L. D. Shriberg, J. R. Duffy, & J. B. Tomblin, 2006) reports information from the orofacial and speech motor control measures administered to the same family; reports on neuropsychological and neuroimaging findings are in preparation.

KEY WORDS: apraxia, articulation, dysarthria, genetics, phonology

Research on the genetic bases of communicative disorders has been infused by landmark studies of a London family (*KE*), half of whose members have a point mutation on chromosome 7q31 that affects the transcription of ribonucleic acid (RNA) produced by a regulator gene, *FOXP2*. Several reviews are available summarizing genetic, language, neuropsychological, neurophysiological, and neuroimaging findings from studies of this family (e.g., Fisher, 2005; Fisher, Lai, & Monaco, 2003; Marcus & Fisher, 2003; Newbury & Monaco, 2002; Vargha-Khadem, Gadian, Copp, & Mishkin, 2005). More recent research by these investigators include studies of the "downstream" targets of *FOXP2,* including genes that develop the neural circuits underlying movements involved in speech production. Examples of associated research include studies of

how expression of *Foxp2* in avian vocal learners is associated with vocal plasticity (e.g., Haesler et al., 2004) and how disruption of *Foxp2* in murine (mouse) models affects ultrasonic vocalizations (Shu et al., 2005). The *Online Mendelian Inheritance in Man* (*OMIM*; McKusick-Nathans Institute for Genetic Medicine, Johns Hopkins University, and National Center for Biotechnology Information, National Library of Medicine, 2000) database provides up-to-date reviews of the *FOXP2* literature.

The primary communicative disorder in affected KE family members is described as "verbal dyspraxia" (i.e., *apraxia of speech*; hereafter, AOS). It is important that the behavioral phenotype used to classify family members as affected, to date, is atypically low performance on an orofacial apraxia task (Vargha-Khadem, Watkins, Alcock, Fletcher, & Passingham, 1995). The speech and orofacial disorders cosegregate completely; all family members reportedly affected for AOS have scores on the orofacial task that do not overlap scores of unaffected family members.

With the exception of brief case reports for 6 affected KE family members (Hurst, Baraitser, Auger, Graham, & Norell, 1990) and three informative studies of subsets of family members by Canadian researchers (Fee, 1995; Goad, 1998; Piggott & Kessler Robb, 1999), there have been no detailed clinical descriptions of the speech, prosody, and voice features that characterize the reported AOS in affected KE family members. AOS is not mentioned as a possible unifying clinical entity for the linguistic findings in these latter three articles; for a summary of linguistic findings, see Shriberg, Green, Campbell, McSweeny, and Scheer (2003). As discussed in our article, the available descriptions of the speech, prosody, and voice of affected KE members in these and other articles (e.g., Alcock, Passingham, Watkins, & Vargha-Khadem, 2000a, 2000b; Vargha-Khadem et al., 1995) include features that are both consistent with, and different from, descriptions of developmental (e.g., Duffy, 2003; Shriberg, Aram, & Kwiatkowski, 1997a) and acquired (e.g., Ballard, Granier, & Robin, 2000; McNeil, 2003) AOS.

Lack of detailed clinical information on the speech disorder segregating in affected KE members has made it difficult for speech researchers to develop models linking *FOXP2* regulation to neural processes underlying speech acquisition and disorders. The descriptive reports and video samples presented in research symposia (e.g., Vargha-Khadem, 2003, 2004) have suggested that, in addition to or instead of AOS, affected KE members may have some form of dysarthria as well as some type of craniofacial dysmorphology. Several sources of information support these possibilities. First, descriptions of AOS in the developmental and, in particular, the acquired neurogenic disorders literature include dysarthria as a frequent concomitant (cf. Crary, 1993; Duffy, 2005). Second, AOS with concomitant dysarthria and/or craniofacial dysmorphologies has been reported in many neurodevelopmental disorders, including epilepsy (e.g., Shuper, Stahl, & Mimouni, 2000), fragile X syndrome (e.g., Spinelli, Rocha, Giacheti, & Ricbieri-Costa, 1995), Rett syndrome (e.g., Bashina, Simashkova, Grachev, & Gorbachevskaya, 2002), and velocardiofacial syndrome (e.g., DeMarco, Munson, & Moller, 2004). As with studies of the KE family reviewed above, however, most of these case studies and reports have provided few quantitative (or qualitative) clinical descriptions of the speech, prosody, and voice features of the participants suspected to have AOS or have dysarthria.

Last, a number of reports have suggested genetic associations among *FOXP2,* AOS, and other speech, language, and craniofacial involvements. Tyson, McGillivary, Chijiwa, and Rajcan-Separovic (2004) described a child with a 7q31 deletion in the region of *FOXP2* who had "a bilateral cleft lip and palate, hearing loss, a language processing disorder, and mild mental retardation" (p. 254). Sarda et al. (1988) reported a child with a deletion in the region of the *FOXP2* gene whose main clinical features included "facial dysmorphy, psychomotor retardation, and absence of language" (p. 259). Zeesman et al. (2006) studied a child with a deletion of the *FOXP2* gene, who, in addition to having a facial dysmorphology similar to the child described in Sarda et al., had "oromotor dyspraxia and mild developmental delay" (p. 509). As with the child described by the Sarda group and a child (C.S.) with a translocation disrupting *FOXP2* reported in Lai et al. (2000), the child discussed by Zeesman et al. also was reportedly unable to voluntarily cough, sneeze, or laugh.

Note also that deficits associated with *FOXP2* may constitute a relatively small proportion of the distal causes for developmental AOS. Lewis and colleagues (personal communication, February 7, 2005) have failed to find any cases with *FOXP2* mutations in large samples of children with speech delay and suspected to have AOS. MacDermot et al. (2005) reported that of 49 children with verbal dyspraxia, only 1 child and his similarly affected sibling and mother had a mutation disrupting *FOXP2*. Xu, Zwaigenbaum, Szatmari, and Scherer's (2004) discussion of the possibility of different genetic subtypes of autism is instructive for our focus in this research:

> We hypothesize that there might be at least three types of autism susceptibility genes/mutations that can be (i) specific to an individual patient or family, (ii) in a genetically isolated sub-population and (iii) a common factor shared among different populations. The genes/mutations could act alone or interact with other genetic and/or epigenetic or environmental factors, causing autism or related disorders. (p. 347)

This case study examines speech, prosody, and voice findings from 2 family members who have a translocation disrupting the *FOXP2* gene. Although each family member had been diagnosed with AOS throughout their lives by speech-language clinicians at several clinical centers, their speech patterns initially impressed us as also consistent with spastic dysarthria (hereafter termed *S_DYS*). A suite of speech, prosody, and voice measures derived primarily from two conversational speech samples from each family member was used to describe similarities and differences in their segmental and suprasegmental profiles. To aid in interpreting the case study findings, the same set of measures and metrics (with some exceptions) were derived from speech samples of two comparison groups of adult speakers, one with acquired spastic or spastic-flaccid dysarthria and the other with acquired AOS. A second goal of this article is to illustrate how findings from perceptual and, eventually, acoustic measures of speech, prosody, and voice can contribute to phenotype sharpening in speech-genetics research.

Method

Assessment

The case study participants were a 50-year-old mother (B.) and her 18-year-old daughter (T.) referred to the third author by a geneticist (sixth author) in B. and T.'s home state. Clinical history obtained for these 2 speakers, termed the *TB* family, indicated that each had been diagnosed as having AOS associated with a de novo balanced 7;13 chromosomal translocation in the mother that was inherited by the daughter. The geneticist who made the referral suspected that the genotypes and phenotypes of these 2 individuals might be similar to those reported for affected members of the KE family. A review of their clinical records indicated that since early childhood, B. and, particularly, T., received extensive speech-language therapy, primarily in the public schools, for cognitive–language delays and severe AOS.

After an initial round of correspondence, B. and T. agreed to participate in an interdisciplinary study of their communicative disorder. The project was approved by the University of Iowa Internal Review Board. Speech, prosody, and voice characteristics of B. and T. were evaluated on two occasions separated by 4 months. Table 1 provides a summary of the measures and tasks administered in each session. Session 1 was completed in the participants' home by the first and third authors and two research assistants. Session 2 was completed by the first and second authors at the University of Iowa. Genetic, language, neuropsychological, and neurological protocols also were completed during the second assessment period by collaborators at the University of Iowa.

Both assessment sessions were audio- and video-recorded for later analysis. For Session 1, the audio system consisted of a Sony TCM-500EV audiocassette recorder, a University Sound 658L directional microphone, and high-quality audiocassette tapes. Lip-to-microphone distance was 6 in. (15 cm). The video system was an Hitachi VHS Video Camcorder, VM-7400A.

For Session 2, the audio system consisted of a Marantz PMD680 portable PC card recorder with a

Table 1. Speech, prosody, and voice assessment protocol.

Measure	Domains assessed		Sessions administered/ obtained		Administration		Approximate length (min)	Examples
	Speech	Prosody–voice	1st	2nd	Live	Computer		
Conversational speech sample	X	X	X	X	X		6–12	
Goldman Fristoe Test of Articulation—2[a]	X		X		X		3–5	
Nonword Repetition Task[b]	X		X	X		X	2	vætʃaɪp, teɪvɔɪtʃaɪg
Syllable Repetition Task[c]	X		X	X		X	2	madaba, banadama
Challenging word repetition tasks								
Multisyllabic Words: List 1[d]	X		X	X		X	2–4	helicopter, kangaroo
Multisyllabic Words: List 2[e]	X		X	X	X		1–2	municipal, skeptical
Stress tasks								
Lexical Stress Task[d]		X	X	X		X	1	bathtub, ladder
Emphatic Stress Task[d]		X	X	X		X	1	Bob MAY go home. May I see PETE?

[a]Goldman & Fristoe (2000). [b]Dollaghan & Campbell (1998). [c]Shriberg, Lohmeier, Dollaghan, & Campbell (2006). [d]Shriberg, Allen, McSweeny, & Wilson (2001). [e]Catts (1986).

unidirectional AudioTechnica ATM75 headset microphone. Video recordings were obtained with a Sony DCR-TRV11 Digital Handycam recording onto a miniDV tape.

Diagnostic Hypotheses

Table 2 contains a list of 13 speech, prosody, and voice variables derived from 18 metrics and analyses, which, in turn, were derived from 6 measures in the three domains. The last three columns in Table 2 include hypotheses about the descriptive features of S_DYS and AOS as well as 7 directional hypotheses (bolded) posited to be specific for each disorder. These descriptive features and diagnostic hypotheses are primarily based on five sources: McNeil, Robin, and Schmidt's (1997) influential review chapter on AOS, Duffy's (2005) review of the motor speech disorders literature, and findings in three widely cited reviews on developmental AOS (Davis, Jakielski, & Marquardt, 1998; Forrest, 2003; Murdoch, Porter, Younger, & Ozanne, 1984). Additional information was drawn from a study series on childhood AOS (Odell & Shriberg, 2001; Shriberg et al., 1997a; Shriberg, Aram, & Kwiatkowski, 1997b, 1997c; Shriberg, Campbell, et al., 2003; Shriberg, Green, et al., 2003; Velleman & Shriberg, 1999). As indicated previously, there is considerable debate on the speech, prosody, and voice behaviors that support diagnostic classification consistent with AOS. Therefore, rather than attempting to marshal extended lists of primary sources supporting each diagnostic hypothesis, in this and later sections, these secondary sources (particularly McNeil et al., 1997, and Duffy, 2005) should be understood to include the summative empirical bases for each hypothesis. In consideration of the histories of the present speakers, these features and hypotheses are based on findings reviewed for speakers with both acquired and developmental AOS. Consistent

Table 2. Matrix of domains (3), variables (13), tasks (6), metrics–analyses (18), diagnostic features (11), and hypotheses (7) assessed in the case study.

Domain	Variable	Task	Metrics–analyses	Diagnostic features and hypotheses[a]	Consistent with: Spastic dysarthria	Consistent with: Apraxia
Speech	1. Severity of speech involvement	CSS	Percentage of Consonants Correct Percentage of Vowels Correct Percentage of Consonants Correct by Manner Feature Intelligibility Index	Spastic dysarthria and apraxia of speech can range from mild to severe. Therefore, severity of involvement is not specific for either disorder.	—	—
	2. Error consistency	CSS Multisyllabic Words: List 2	Error target analysis Error type analysis Whole-word analysis Summative analysis	Inconsistent errors	?	?
	3. Error type	CSS	SODA analysis using severity-adjusted indexes Residual error analysis	Primarily distortion errors	?	?
	4. Error typicality	CSS Multisyllabic Words: List 2	EMA analysis	EMA errors		?
Prosody	5. Phrasing	CSS	PVSP Codes 2–8	**Inappropriate phrasing**		X
	6. Rate	CSS	PVSP Codes 9–12	Inappropriate rate	X	X
	7. Sentential stress	CSS	PVSP Codes 13–16 and subcodes	Inappropriate sentential stress	X	X
	8. Lexical stress	Lexical Stress Task	Lexical stress ratio	**Inappropriate lexical stress**		X
	9. Emphatic stress	Emphatic Stress Task	Emphatic stress ratio	**Inappropriate emphatic stress**		X
Voice	10. Loudness	CSS	PVSP Codes 17–18	**Inappropriate loudness**	X	
	11. Pitch	CSS	PVSP Codes 19–22	**Inappropriate pitch**[b]	X	
	12. Laryngeal quality	CSS	PVSP Codes 23–29	**Inappropriate laryngeal quality**[c]	X	
	13. Resonance	CSS	PVSP Codes 30–32	**Inappropriate resonance**[d]	X	

Note. CSS = conversational speech sample; SODA = substitutions, omissions, distortions, additions; EMA = epenthesis, metathesis, assimilation; PVSP = Prosody–Voice Screening Profile (Shriberg, Kwiatkowski, & Rasmussen, 1990).

[a]The seven diagnostic hypotheses are boldface. [b]Too soft and/or too low pitched. [c]Harsh voice. [d]Hypernasality.

with the descriptive goals of this case study, findings for B. and T. are presented in the Results section for all 13 of the variables listed in Table 2, with additional discussion focusing on the 7 directional hypotheses for S_DYS and AOS. Rationales for the assignment of markers to S_DYS, AOS, or both disorders are described in the following sections.

Speech Variables, Measures, Metrics, and Analyses

The following sections provide brief descriptions of the metrics used to assess each variable, with additional information on each metric provided in Results. Custom software in the Programs to Examine Phonological and Phonetic Evaluation Records suite (PEPPER; Shriberg, Allen, McSweeny, & Wilson, 2001) was used to generate the statistical and graphic information for most of the metrics and analyses.

Severity analyses. To assess "severity of speech involvement," the first speech variable in Table 2, PEPPER was used to produce 30 speech profiles from each participant that were based on their conversational speech. The profiles provided quantitative detail at the level of allophones (i.e., diacritic modifications), speech sounds, developmental sound classes (to be described), and place-manner-voicing features. To maximize generalizability and provide comparison standard deviation bars, B. and T.'s two conversational samples (i.e., Sessions 1 and 2) were averaged for all summary comparisons. Thus, the conversational speech data used in all the metrics in Table 2 are based on B. and T.'s total number of intelligible words, 446 and 447, respectively, in the two samples. The four measures selected to best describe B. and T.'s severity of speech involvement for the present study, each of which has been described elsewhere (Shriberg, Allen, et al., 2001; Shriberg, Austin, Lewis, McSweeny, & Wilson, 1997), were as follows: Percentage of Consonants Correct, Percentage of Vowels Correct, Intelligibility Index, and Percentage of Consonant Features Correct. As indicated in Table 2, severity of involvement was not hypothesized to be specific for S_DYS or AOS.

Error consistency analyses. Inconsistent speech errors are one of the core features proposed by many researchers to differentiate childhood AOS from several forms of dysarthria, including S_DYS (cf. Shriberg, Campbell, et al., 2003). Whereas the errors associated with S_DYS are reported to be relatively stable, that is, without "islands" of error-free speech, the planning or programming deficits underlying AOS are traditionally described as the source of inconsistent errors. In the developmental literature, these errors conventionally include variability at the phonemic level, such as unusual and variable consonant and vowel substitutions and additions. Based on contemporary models of speech motor control, however, such phoneme-level errors are placed at selection and sequencing levels that precede movement planning (McNeil, 1997, 2003; McNeil et al., 1997). Critical to such classification decisions is valid and reliable information on whether putative substitutions and additions are also distorted, as would be consistent with motor planning involvement (i.e., AOS). A constraint in the present data, as described in the following paragraph, is that they are based wholly on perceptual (transcription) methods, with the attendant problems of both reliability and validity (Shriberg & Lof, 1991). We have therefore taken the conservative position of using a question mark (see Table 2) to assign error consistency to both S_DYS and AOS.

As shown in Table 2, the software produced four consistency metrics that were based on speech tokens from conversational samples and responses to the Multisyllabic Word Task: List 2 (Catts, 1986). *Error target* consistency percentages are the averaged consistency of consonant targets produced at least once incorrectly in all repeated tokens of each word type in the sample (i.e., the percentage of times each such sound was said either correctly or incorrectly in repeated tokens of a word type). *Error type* consistency percentages are the averaged consistency of error types (i.e., the same phonemic-level error) on consonant targets produced at least once incorrectly in all repeated tokens of all word types in the sample. *Whole word* consistency percentages index the averaged consistency of all errors (i.e., same phoneme-level error on all vowel and consonant targets in the word) in all word types in which at least one sound in the word was produced incorrectly. Last, the software yields a *summative* consistency metric for each speaker, which is the simple average of scores from the three individual consistency metrics. Conceptually, the summative metric has the property of greater sensitivity to the construct of inconsistency, as reflected in contributions from any two or all three individual consistency metrics. Psychometrically, the use of the average of each participants' three consistency measures (i.e., error type, error target, whole word) generally reduces the observed between-subjects variance (i.e., standard deviations are smaller).

Error type analyses. As shown in Table 2, the software suite includes two sets of metrics and analyses to characterize a speaker's error types. The first is termed *SODA* analyses, the term for traditional analyses that divide speech errors into the four categories of substitutions, omissions, distortions, and additions. Our analysis, as described in the Results section, provides severity-adjusted percentages for each error type (a series of rules code additions as substitutions or distortions; Shriberg, Allen, et al., 2001). As indicated

previously, the literature on the differential prevalence of such error types in developmental AOS differs from more recent perspectives in acquired AOS. In developmental apraxia, the general perspective is that whereas omissions may be common to both apraxia and dysarthria, substitutions and additions (i.e., phonemic-level errors) are more consistent with apraxia and distortions (i.e., subphonemic-level errors) with dysarthria. In the adult literature, however, there is growing consensus that phoneme-level substitutions and additions are not consistent with the core planning or programming deficits proposed in speech motor control models of AOS (cf. Ballard et al., 2000; Duffy, 2005; McNeil, 2003; McNeil et al., 1997; Odell, McNeil, Rosenbek, & Hunter, 1990; Rosenbek & McNeil, 1991). As indicated previously, McNeil et al. and others conclude that adults with AOS predominantly produce distortions, whereas phoneme-level substitutions are proposed to be more associated with alternative or concomitant aphasia (i.e., paraphasias). We submit that this latter perspective presumes consensus on three methodological needs: (a) a list of the specific speech sound distortions that qualify as evidence for each subtype of dysarthria versus apraxia (i.e., specifying targets and the specific errors in place, manner, voicing, force, or duration), (b) sensitive, specific, and reliable methods for the detection of each distortion type, and (c) well-developed quantitative criteria for the frequencies and distributions of each error type required to classify speakers as making the criterial distortions. The perceptual methods and analysis tools available for the present study do not currently provide such information to differentiate among putative dysarthric versus apraxia distortion types. Hence, as with inconsistent errors reviewed previously, distortion errors are assigned as questionable markers of either dysarthria or apraxia (see Table 2).

A second error type analysis, termed *residual* error analysis, was used to describe the most frequent types of distortion errors B. and T. made in their conversational speech. Studies of children with prior speech delay of unknown origin have indicated that distortions of sibilants (/s/, /z/, /ʃ/) and rhotics (/r/, /ɝ/, /ɚ/) are the most frequent residual speech errors observed in life span data (Austin & Shriberg, 1996; Lewis & Shriberg, 1994; Shriberg & Kwiatkowski, 1994). The goal of the present analysis was to determine if B. and T.'s distortion errors were like those commonly observed in speakers with prior speech delay (e.g., dentalized sibilants, derhotacized liquids), were more consistent with problems of speech motor control (e.g., spirantized stops, epenthetic stops, or nasals), or included errors from both putative sources. The question addressed both the descriptive goals of this case study and the secondary goal of illustrating how detailed speech measures may be used to sharpen phenotypes used in speech-genetics research. No hypotheses were posited about the differential diagnostic significance of residual developmental speech sound distortions.

Error typicality analyses. Although researchers have not agreed on one diagnostic checklist for childhood AOS (cf. Shriberg, Campbell, et al., 2003), there is some consensus in this literature that three types of speech errors may have diagnostic specificity: epenthetic errors (additions of across-manner sounds; e.g., /ɔr͡kr̩ɪstrə/ for *orchestra*), metathetic errors (reversals of target sounds within words; e.g., /sɪmənɪm/ for *cinnamon*), and atypical assimilation errors (e.g., a target sound in a phonetically complex word changes to resemble exactly, or in salient features, another target sound in the word; e.g., /pɛrərɛl/ for *parallel*). We propose the cover term *EMA ĕrrors* (*e*penthesis, *m*etathesis, *a*ssimilation) to refer collectively to these error classes, included in Table 2, and as the metrics and analysis for error typicality.

In speakers who correctly produced the target sounds elsewhere in a corpus, EMA errors have been viewed as having face and construct validity as markers for developmental AOS. EMA errors are not included among the many natural phonological processes (with the exception of some forms of assimilation) that have been proposed to describe the deletion and substitution errors of children with typical and delayed-speech acquisition. However, as previously reviewed, EMA errors in adults with acquired disorders have been interpreted as consistent with the selection and sequencing deficits in aphasia (i.e., paraphasias). It is interesting that this is the classificatory perspective first proposed by Gopnik and colleagues (Gopnik, 1990; Gopnik & Crago, 1991; see also Watkins, Dronkers, & Vargha-Khadem, 2002) to account for the speech-language error patterns observed in affected members of the KE family. For the present study, we provide descriptive information on such errors but only provisionally assign EMA errors as support for AOS (see Table 2).

Prosody and Voice: Variables, Measures, Metrics, and Analyses

The Prosody–Voice Screening Protocol. Information for six of the remaining eight variables in Table 2 was obtained from a perceptually based analysis instrument termed the *Prosody-Voice Screening Protocol* (PVSP: Shriberg, Kwiatkowski, & Rasmussen, 1990). Table 2 includes the codes used for each variable, which are aggregated, percentaged, and profiled by the PEPPER analysis software. Figure 1 is a list of all exclusion and the primary prosody–voice codes, as described with audio exemplars in Shriberg et al. (1990) and in a reference data archive (Shriberg, Kwiatkowski, Rasmussen, Lof, &

Figure 1. The 32 exclusion codes and 32 prosody–voice codes used in the Prosody–Voice Screening Profile (Shriberg, Kwiatkowski, & Rasmussen, 1990). Copyright 1990 by Lawrence Shriberg, Joan Kwiatkowski, and Carmen Rasmussen. Reprinted with permission.

Exclusion Codes

Content/Context	Environment	Register	States
C1 Automatic Sequential	E1 Interfering Noise	R1 Character Register	S1 Belch
C2 Back Channel/Aside	E2 Recorder Wow/Flutter	R2 Narrative Register	S2 Cough/Throat Clear
C3 I Don't Know	E3 Too Close to Microphone	R3 Negative Register	S3 Food in Mouth
C4 Imitation	E4 Too Far from Microphone	R4 Sound Effects	S4 Hiccup
C5 Interruption/Overtalk		R5 Whisper	S5 Laugh
C6 Not 4 (+) Words			S6 Lip Smack
C7 Only One Word			S7 Body Movement
C8 Only Person's Name			S8 Sneeze
C9 Reading			S9 Telegraphic
C10 Singing			S10 Yawn
C11 Second Repetition			
C12 Too Many Unintelligibles			

Prosody-Voice Codes

Prosody

Phrasing	Rate	Stress
1 Appropriate	1 Appropriate	1 Appropriate
2 Sound/Syllable Repetition	9 Slow Articulation/Pause Time	13 Multisyllabic Word Stress
3 Word Repetition	10 Slow/Pause Time	14 Reduced/Equal Stress
4 Sound/Syllable and Word Repetition	11 Fast	15 Excessive/Equal/Misplaced Stress
5 More than One Word Repetition	12 Fast/Acceleration	16 Multiple Stress Features
6 One Word Revision		
7 More than One Word Revision		
8 Repetition and Revision		

Voice

Loudness	Pitch	Quality	
		Laryngeal Features	Resonance Features
1 Appropriate	1 Appropriate	1 Appropriate	1 Appropriate
17 Soft	19 Low Pitch/Glottal Fry	23 Breathy	30 Nasal
18 Loud	20 Low Pitch	24 Rough	31 Denasal
	21 High Pitch/Falsetto	25 Strained	32 Nasopharyngeal
	22 High Pitch	26 Break/Shift/Tremulous	
		27 Register Break	
		28 Diplophonia	
		29 Multiple Laryngeal Features	

Miller, 1992). The exclusion codes shown in Figure 1 are used to exclude utterances from PVSP coding that are due to assessment constraints, typically with very young children or children with cognitive, behavioral, or other challenges (cf. McSweeny & Shriberg, 2001). The codes are divided into those associated with content–context (Codes C1–C12), environment (Codes E1–E4), register (Codes R1–R5), and state (Codes S1–S10). The first 24 eligible (i.e., nonexcluded) utterances are coded with 1 of 32 PVSP codes used to classify utterances with inappropriate prosody or voice. Percentages above 90% appropriate for each of the 7 prosody and voice domains shown in Figure 1 are considered "pass" on this instrument, percentages from 80% to 89% are considered "questionable," and percentages below 80% are considered "fails." All technical information cited below is abstracted from the two reference citations.

Phrasing. Appropriate phrasing is defined as a flow of word and phrase groups that are appropriate for the speaker's age, emotional state, and the intended propositional content. As indicated in Figure 1, phrasing includes 7 PVSP codes that assess elements that disrupt phrasing, including part- and whole-word repetitions, revisions, and combinations of these behaviors in the same utterance. Such behaviors are posited to occur when speakers try to self-correct their errors (Shriberg et al., 1997c). As indicated in Table 2, inappropriate phrasing is posited to be specific for AOS.

Rate. The criterion for appropriate rate in PVSP analysis of adult conversational speech is 4–6 syllables per second. The four inappropriate rate codes differentiate between rates that are too slow because of articulation and/or pause time and rates that are too fast with or without accelerations (PV Codes 9–12). As indicated in Table 2, both speakers with S_DYS and AOS are posited to have slow rates.

Sentential stress. Appropriate sentential stress is coded perceptually in the PVSP using four primary codes and a series of secondary codes (not shown in Figure 1, but described later) that provide quantitative information on relevant subtypes of excessive-equal stress. Speakers with S_DYS and those with AOS are posited to have inappropriate sentential stress.

Lexical stress. The Lexical Stress Task (Shriberg, Allen, et al., 2001) was developed to provide acoustic data on a speaker's stress in imitation of prerecorded trochaic words (see examples in Table 1). The lexical stress ratio (LSR: Shriberg, Campbell, et al., 2003) is obtained by dividing a speaker's stress on the first syllable of each of eight trochaic words by stress on the second syllable, thereby normalizing for individual differences in intensity, frequency, and duration. A principal components analysis of a number of candidate variables to represent stress yielded weightings for three acoustic metrics for each syllable: amplitude area, frequency area, and duration. These weightings were applied to each speaker's scores on each syllable, yielding one dimensionless LSR value. LSR findings for 35 preschool and school-aged speakers with speech delay of unknown origin reported in Shriberg, Campbell, et al. (2003) have recently been cross-validated in an additional sample of 19 children with speech delay of unknown origin (Shriberg, McSweeny, Karlsson, Tilkens, & Lewis, 2006). Included in these two data sets of 54 total children were 17 speakers suspected to have AOS. Findings in each study indicated that a statistically greater proportion of the latter speakers' average LSR values on the eight words fell at each end of the distribution of LSR scores. These findings suggested that these speakers suspected to have AOS were either overstressing (high LSR values) or understressing (low LSR values) syllables in the trochaic words. As indicated in Table 2, inappropriate lexical stress is posited to be specific for AOS.

Emphatic stress. As shown in the examples in Table 1, the Emphatic Stress Task (Shriberg, Allen, et al., 2001) assesses a speaker's ability to imitate emphatic stress. This task, which was developed to be appropriate for the cognitive and speech constraints of young children with significant speech delay, consists of 2 four-word sentences repeated four times each. Emphatic stress shifts across each of the four words in each sentence on each repetition (e.g., BOB may go home, Bob MAY go home, etc.). Scoring is currently accomplished perceptually. Using consensus techniques, the transcriber and the first author scored each response as either matching or not matching the targeted stressed word in the recorded stimulus, yielding a maximum possible score of 8 for each task administration. As indicated in Table 2, inappropriate emphatic stress was posited as specific for AOS.

Loudness and pitch. Appropriate loudness and pitch were coded from the conversational sample. Six PVSP codes are used to classify utterances that are too loud or too soft and/or too low or too high pitched for the speaker's age and gender. Inappropriate loudness or pitch was not posited to characterize AOS, but lowered loudness and especially lowered pitch were posited to be consistent with S_DYS.

Laryngeal and resonance quality. Appropriate laryngeal and resonance quality was defined as vocal characteristics in conversation that were within the normal range for the speaker's age and gender. A series of 10 PVSP codes (Figure 1) were used to classify different types and combinations of laryngeal and resonance quality that were perceived as inappropriate, relative to the exemplars used in the training program completed by the research assistant who coded these data (see next section). Inappropriate laryngeal quality was posited as specific for S_DYS but not for AOS. Inappropriate resonance (in particular, consistent hypernasality) was posited as characteristic of S_DYS.

Comparison Data

To provide additional data on the questions addressed in this case study, we compared findings from B. and T. with data from two groups of adult speakers with acquired motor speech disorders. Odell and Shriberg (2001) reported data from 9 adults with S_DYS and 14 adults with acquired AOS. The conversational speech samples from all except 2 of the speakers with S_DYS met the prosody–voice coding requirement of 24 intelligible utterances, reducing to 7 the total number of participants in the present study with spastic or spastic-flaccid dysarthria. In Odell and Shriberg's study, audio recordings of conversational speech samples from each of the 21 eligible speakers were transcribed and coded for prosody–voice by the same transcriber who completed the transcription and prosody–voice coding of B. and T.'s samples in the present study. Approximately 61% of the present samples were transcribed and prosody–voice coded by consensus with another experienced research transcriber.

Table 3 is a summary of clinical information for the speakers in the comparison groups. All 21 participants were native speakers of American English, and with the

Table 3. Description of comparison speakers with spastic dysarthria (S_DYS) and apraxia of speech (AOS).

Variable	S_DYS (*n* = 7)					AOS (*n* = 14)							
	n	*M*	*SD*	Range	% male	*n*	*M*	*SD*	Range	% male	*Z*	*t*	*p*
Sex					100					85.7	1.53	1.53	.13
Age (years)		64.1	11.3	48–79			61.4	8.8	50–81			0.56	.59
Postonset (months)		33.2	19.2	4–60			50.4	55.2	1–180			1.03	.32
Etiology[a]													
Stroke	4					14							
MS	1					—							
TBI	1					—							
Type of dysarthria													
Spastic	4												
Spastic-flaccid	2												

Note. MS = multiple strokes; TBI = traumatic brain injury.

[a]Excluding the participant in the dysarthric group with cerebral palsy (congenital).

exception of 1 speaker with cerebral palsy (described below), none had premorbid histories of speech or language disorders. As shown in Table 3, speakers in both groups were predominantly male (S_DYS: 7 of 7, 100%; AOS: 12 of 14, approximately 86%); a between-groups test of proportions was nonsignificant. The speakers' ages ranged from 48 to 81 years; a *t* test for differences in mean age was nonsignificant. At assessment, participants in the two groups (excluding the participant with cerebral palsy) ranged from 1 to 180 months postonset of brain damage; a *t* test for differences in mean months postonset was nonsignificant.

Odell and Shriberg's (2001) research provided detailed information on the widely used cognitive and language tests administered to the two speaker groups. The fifth author, an American Speech-Language-Hearing Association (ASHA)–certified clinician with over 20 years of experience with neurogenic speech-language-voice disorders, used all available test information plus supplemental measures to cross-validate the referral classification. Classification criteria for acquired apraxia and subtypes of dysarthria followed the guidelines in Wertz, LaPointe, and Rosenbek (1984) and Darley, Aronson, and Brown (1975), respectively. As described for the speakers with AOS in Odell and Shriberg (2001) and confirmed for the speakers with S_DYS, none of these participants had substantial hearing loss, less than low–normal cognitive performance, or dementia. The primary diagnosis for 18 of the 21 speakers was one or more strokes, most commonly resulting in a unilateral, left-hemisphere lesion as documented by radiological reports or physician comments in the medical records. The speakers had been diagnosed by referring speech-language pathologists, all of whom were ASHA certified with 10–35 years of experience in the diagnosis and treatment of adult neurogenic speech-language disorders. Many of the speakers had participated in other research studies locally as well as research projects at several sites in North America. The 21 speakers had only very mild or no aphasia.

As indicated above, one of the speakers with S_DYS had cerebral palsy and was included in the present sample to allow for an inspection of developmental issues. This participant was within the same age range as the other speakers with S_DYS and approximately the same age as B. at the time his speech was assessed. Detailed inspection of this individual's speech, prosody, and voice profiles indicated that, with one exception discussed later, he did not substantially differ in severity or error type from the other 6 speakers with S_DYS.

Transcription and Prosody–Voice Coding

Procedures. All speech production tasks were transcribed and prosody–voice coded by a research transcriber who had over 12 years of experience using a narrow phonetic transcription system and the PVSP. The transcription system includes a set of diacritic symbols, transcription conventions, and formatting conventions for file processing in a software suite developed for research in speech sound disorders (Shriberg, Allen, et al., 2001; Shriberg & Kent, 2003). The conversational samples and all other speech tasks from Session 1 were transcribed and prosody–voice coded using one of several well-maintained Dictaphone Model 2550 analog playback devices used in prior studies of speech sound disorders. The digital video samples from Session 2 were played back using custom software for computer-based transcription (Shriberg, McSweeny, et al., 2005). Transcription of Sessions 1 and 2 were completed from the audiotapes and the digitally recorded videotapes,

respectively. Findings reported in Shriberg, McSweeny, et al. have indicated that transcription and prosody–voice data from digital compared with analog recording and playback systems are comparable.

Reliability. A total of 7 of the 25 (28%) conversational samples were randomly selected for individual retranscription and repeated prosody–voice coding by two transcribers. Ages of the 7 speakers—2 with acquired S_DYS, 3 with acquired AOS, and 1 each for B. and T.—ranged from 18 to 70 years. Three of the samples had been transcribed and coded initially by consensus between two research transcribers, and the remaining four samples by one (Transcriber 1) member of the team. All seven samples were retranscribed and recoded by both members of the consensus team. Point-to-point percentages of agreement for transcription and prosody–voice coding were provided by software utilities (Shriberg, Allen, et al., 2001; Shriberg & Olson, 1988).

Average intrajudge agreement (based on 1,480 words) for Transcriber 1, who transcribed all seven samples twice, was 98.2% for broad (i.e., disregarding diacritics in either transcript) phonetic transcription of consonants and 87.3% for narrow (i.e., including diacritics in either transcript) phonetic transcription. For vowels, average intrajudge agreement was 92.2% for broad and 82.8% for narrow phonetic transcriptions. Average intrajudge agreement for the second member (Transcriber 2) of the consensus team (based on 3 samples and 671 words) was 98.3% for broad transcription of consonants and 90.9% for narrow transcription. For vowels, average intrajudge agreement was 94.5% for broad phonetic transcription and 89.3% for narrow transcription. Last, average interjudge agreement calculated on four samples (772 words) was 95.5% and 89.4% for broad phonetic transcription of consonants and vowels, respectively, and 79.9% and 75.5% for narrow transcription of consonants and vowels, respectively.

Intrajudge prosody–voice agreement was based on 152 (Transcriber 1) and 72 (Transcriber 2) coded utterances from all seven randomly chosen conversational speech samples (Transcriber 1) and three conversational samples (Transcriber 2). Point-to-point percentages of agreement for each transcriber were 92.6% and 93.7%, respectively, on the basis of appropriate versus inappropriate prosody–voice codes for each of the six domains; agreements were 87.2% and 93.1% on the basis of exact agreement for both appropriate and all inappropriate codes. Intrajudge agreement for the five prosody–voice domains of interest in this study, including percentages calculated at the summative levels, was as follows for the two coders, respectively: phrasing: 98.7%/100%; rate: 87.5%/62.5%; stress: 88.7%/94.4%; laryngeal quality: 82.2%/98.6%; and resonance quality: 94.1%/100% (all utterances in the three samples were coded appropriately both times). Last, interjudge agreement at the summative level of prosody–voice coding (based on 80 coded utterances from four conversational speech samples) was 75.4% on the basis of appropriate versus inappropriate prosody–voice codes for each of the six domains and 73.2% on the basis of exact agreement for both appropriate and all inappropriate codes. These reliability findings were consistent with those obtained using comparable auditory perceptual systems for phonetic transcription and prosody–voice coding (cf. Shriberg & Lof, 1991; Shriberg, McSweeny, et al., 2005).

Results

Cognitive, Language, and Speech Status

Table 4 is a summary of findings from the cognitive, language, and articulation test instruments administered to B. and T., using the higher of the test scores for measures given twice. As indicated by the reference data in Table 4, B. and T. had scores ranging from the low–normal range of cognitive–language functioning to frank impairment. Although reference data on the phonological processing tasks were not available for speakers of this age, B. and T.'s percentages on these measures were lower than the approximately 90%–100% performance results obtained from typically speaking, older speakers' responses to the Nonword Repetition Task (Dollaghan, September 14, 2004, personal communication). B. and T. also had notably lower scores on the Syllable Repetition Task (Shriberg, Lohmeier, Dollaghan, & Campbell, 2006), a measure of phonological processing developed expressly for speakers with limited phonetic inventories. Last, B. and T.'s performance on the Sounds-in-Words subtest of the Goldman Fristoe Test of Articulation—2 (GFTA–2; Goldman & Fristoe, 2000), which assesses a speaker's phonetic inventory in citation forms, yielded standard scores of 88 and 94, respectively. Therefore, these values were somewhat lower for speakers of B. and T.'s age (using the oldest available GFTA–2 reference value for B.). Errors were primarily distortions of sibilants and rhotic consonants, as described in later analyses. A companion article (Ballard et al., 2006) provides detailed analyses of the cognitive–language findings, summarized in Table 4. For these reasons and space constraints, the present article does not include further discussion of B. and T.'s cognitive–language status.

Speech

Severity of Speech Sound Involvement

Findings. Figure 2 provides summary information on B. (dark-filled bars) and T.'s (light-filled bars) severity of speech sound involvement as assessed in the two conversational samples, including means and standard deviation bars. Findings for the comparison data, to be

Table 4. Summary data on mother's and daughter's cognitive, language, and speech status.

Domain	Measure	Mother ("B.")	Daughter ("T.")	Reference data	
				M	*SD*
Cognition					
Nonverbal	WAIS–III[a]				
	Picture Completion	9	6	10	3
	Digit Symbol–Coding	4	5	10	3
	Block Design	8	8	10	3
	Object Assembly	10	9	10	3
	Picture Arrangement	9	6	10	3
	Performance IQ	95	87	100	15
Verbal	WAIS–III[a]				
	Vocabulary	6	5	10	3
	Similarities	8	9	10	3
	Arithmetic	4	7	10	3
	Digit Span	5	8	10	3
	Information	11	5	10	3
	Verbal IQ	81	81	100	15
	Full Scale IQ	88	81	100	15
Language					
Receptive	PPVT–III[a]	83	79	100	15
	Grammaticality judgment[b]	76	82	99	—
Expressive	CELF–3: Recalling sentences	3	3	10	3
	EVT	4.6	4.6	10	3
Speech					
Phonological processing	Nonword Repetition Task	72%	75%	NA[c]	
	Syllable Repetition Task	66%	74%	NA[c]	
Production	GFTA–2 (2000)[a]	88	94	100	15

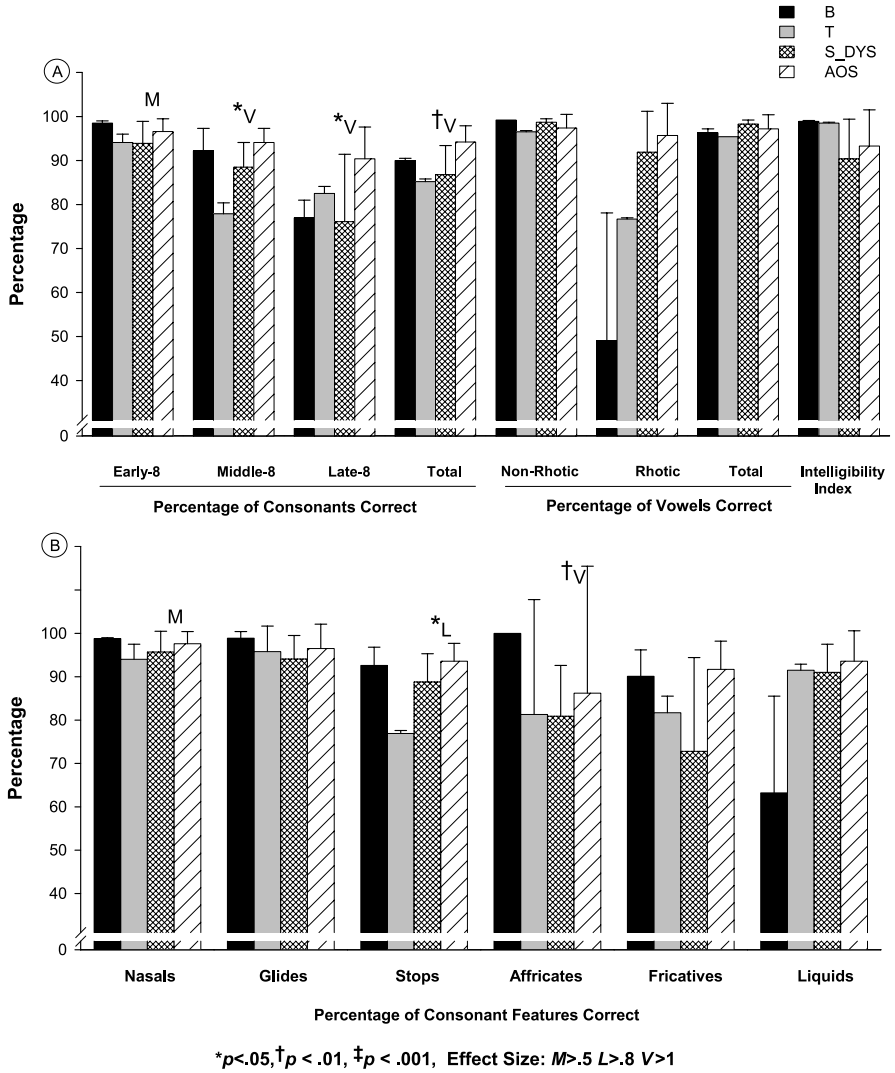
Note. WAIS–III = Wechsler Adult Intelligence Scale—Third Edition (Wechsler, 1997); PPVT–III = Peabody Picture Vocabulary Test—Third Edition (Dunn & Dunn, 1997); CELF–3 = Clinical Evaluation of Language Fundamentals—Third Edition (Semel, Wiig, & Secord, 1995); EVT = Expressive Vocabulary Test (Williams, 1997); GFTA–2 = Goldman Fristoe Test of Articulation—Second Edition (Goldman & Fristoe, 2000).

[a]Standard scores. [b]Percentage. [c]Data not available.

discussed in the following section, are also included in Figure 2, including means and standard deviations for the 7 adults with S_DYS (cross-hatched bars) and the 14 adults with acquired AOS (diagonal-striped bars). The legend for Figure 2 describes the speech metric comparisons in Panel A and Panel B. Reference data for these measures indicate that by 18 years of age or younger, typical speakers have scores approaching 100% on each metric (Austin & Shriberg, 1996). B. and T.'s percentages on the metrics shown in Figure 2 were generally lower than typical speakers.

As shown in Figure 2, Panel A, B.'s and T.'s total Percentage of Consonants Correct scores, respectively, were 90% and 85.2%, and their total Percentage of Vowels Correct scores were 96.3% and 95.4%, respectively. Their Intelligibility Index scores, however, were nearly perfect, at 98.9% and 98.5%, respectively. As organized by developmental sound class (Panel A: Early-8, Middle-8, and Late-8 consonants) and consonant manner features (Panel B: nasals, glides, stops, affricates, fricatives, and liquids), findings indicated that B. and T.'s error target patterns were consistent with the general order of English consonant acquisition, which in all languages, is presumed to reflect a gradient of increasingly difficult perceptual, cognitive, and articulatory demands. Specifically, in conversational speech, B. and T., respectively, had 98.5% and 94.1% correct articulation of the Early-8 consonant sounds (/m/, /b/, /j/, /n/, /w/, /d/, /p/, /h/), 92.2% and 77.9% correct Middle-8 consonants (/t/, /ŋ/, /k/, /g/, /f/, /v/, /tʃ/, /ʤ/), and 77.0% and 82.5% correct Late-8 consonants (/ʃ/, /θ/, /s/, /z/, /ð/, /l/, /r/, /ʒ/). As indicated by the consonant manner feature data in Panel B, B.'s errors were primarily on the liquids /l/ and /r/ (63.2% correct), whereas T.'s consonant errors were primarily on stops

Figure 2. Summary of findings from the severity of involvement metrics calculated from the conversational speech samples of mother (B.), daughter (T.), and the two comparison groups with motor speech disorders. Panel A is a summary of the findings organized by *developmental sound class* (Shriberg, Austin, Lewis, McSweeny, & Wilson, 1997). This ontogenetic construct divides the 24 English consonants into three subgroups (i.e., Early-8, Middle-8, and Late-8) that reflect the chronological order of consonant acquisition in typical and delayed speech development. Panel A also includes findings for the Percentage of Vowels Correct, which indexes the severity of involvement of vowels (divided into nonrhotic and rhotic [/ɚ/, /ɝ/]) and diphthongs, and the Intelligibility Index, which indicates the percentage of words in the conversational sample that were intelligible to the transcriber(s). Panel B provides severity data organized by manner features, arranged left to right to reflect their order of acquisition in typical and delayed speech development (Shriberg et al., 1997).



(76.9%), affricates (81.3%), and fricatives (81.7%). As indicated in Table 2, no diagnostic classification hypotheses were posited for severity of involvement findings because this variable is not specific for S_DYS or AOS.

Comparison data. Preliminary examination of the distributional moments for the data from the two comparison groups (Figure 2 and all following analyses) indicated that these data met criteria for parametric analyses without the need for transformations but warranted use of unpooled standard-deviation terms for the between-groups *t* tests. Descriptive and inferential statistical findings comparing speakers with S_DYS to those with acquired AOS, including Hedges-corrected effect sizes (Hedges & Olkin, 1985) are provided in Figure 2 and all following figures. As shown in the legend at the bottom of Figure 2, medium effect size ≥ 0.5, large effect size ≥ 0.8, and very large effect size ≥ 1.0. The confidence intervals bounding the effect sizes were generally large,

due, in part, to the small sample sizes. Significant effect sizes (i.e., similar to *t*-test values) at the .05 alpha level or larger are indicated by the conventional symbols. Because of low statistical power and consequent risk for Type II errors, alpha levels were not adjusted for family-wise testing within each panel, and the inferential statistical findings were used as supplementary support. In order of importance, interpretation of findings was influenced by (a) the number and coherence of obtained effect sizes of medium or greater magnitude per analysis, (b) the magnitudes of obtained effect sizes, and (c) the reliability of findings as estimated by the associated inferential statistics.

As indicated previously, Figure 2 includes information on the severity findings for speakers in the two comparative groups with S_DYS (cross-hatched bars) and acquired AOS (diagonal-striped bars). In 12 of the 14 comparisons shown in Figure 2, the speakers with S_DYS had lower average percentage correct scores than the speakers with acquired AOS. These differences were associated with large to very large effect sizes for 5 of the 12 comparisons, each of which was statistically significant.

Note that B. and T.'s scores on many of the severity metrics and subscales were closer to the average scores of the comparison-group speakers with S_DYS. The strongest findings were for the Late-8 sounds (the most challenging sounds), in which B. and T.'s standard deviation bars did not overlap those of the speakers with acquired AOS. As indicated previously, however, although B. and T.'s severity levels were considerably lower than the approximately 100% expected for typical speakers of their age, these findings cannot be viewed as support for a greater dysarthric component in their speech patterns because increased severity was not specific for S_DYS.

Error Consistency

Findings. The three consistency analyses described in the Method section assessed the stability of B. and T.'s speech sound errors in conversational speech and in response to the more difficult of the two challenging word repetition tasks (see Table 1, List 2). There were two constraints on the analyses. First, because of the large standard error of measurement at the level of narrow phonetic transcription of consonants and vowels–diphthongs (cf. McSweeny & Shriberg, 1995; Shriberg et al., 1997b; Shriberg & Lof, 1991), consistency analyses were not psychometrically appropriate at the phonetic (i.e., diacritic) level of analysis. Second, with the exception of the whole-word consistency analysis, there were too few occurrences of phoneme-level, vowel or diphthong errors to complete vowel or diphthong consistency analyses. The left side of Table 5 shows a summary of findings for B. and T. for the three consistency metrics. Each analysis was based on all repeated tokens of all word types (e.g., all tokens of the word *dog*) in the speech samples. In the combined two conversational samples for each speaker, B. had 11 eligible tokens of 5 types and T. had 45 eligible tokens of 16 types.

The primary consistency findings are that B. and T.'s consistency scores differed from one another, both as sampled in conversation and by responses to the challenging multisyllabic word task. As shown in Table 5, B.'s individual and overall consistency scores were low, averaging 55.3% in conversational speech and 61.4% in repetitions of the challenging multisyllabic words. In contrast, T. had over 20% higher individual and overall consistency scores, averaging 72.9% in repeated tokens of the same word type in conversation and 86.9% in repetitions of the challenging multisyllabic words. Relative to the potential value of such information for

Table 5. Summary of findings from three types of consistency analyses completed on the conversational speech samples and response to the challenging word repetition task, List 2.

		Comparison groups											
		B.		T.		S_DYS		AOS		*t* test		Effect size (ES)[a]	
Sample	Consistency metric	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*t*	*p*	ES	Adj.
Conversation	Error target	41.0	10.8	64.0	13.0	54.2	11.7	43.9	21.2	1.59	.329	0.54	Medium (*ns*)[b]
	Error type	83.3	23.6	84.7	9.8	87.6	18.1	58.8	40.3	1.88	.150	0.81	Large (*ns*)
	Whole word	41.7	11.8	70.0	20.4	70.0	22.9	45.6	39.5	1.56	.142	0.67	Medium (*ns*)
	Summative[c]	55.3	24.2	72.9	10.7	70.6	16.7	49.4	8.2			1.76	Very large[d]
Multisyllabic Word Task 2	Error target	60.0	4.2	88.7	16.0								
	Error type	87.1	2.0	97.1	4.1								
	Whole word	37.0	5.2	75.0	35.4								
	Average[c]	61.4	25.1	86.9	11.2								

[a]Hedges corrected (Hedges & Olkin, 1985). [b]*ns* = nonsignificant. [c]Summative = sum of 3 tasks divided by 3; *SD* computed from 3 task scores. [d]Significant: confidence interval = 0.71–2.81.

differential diagnosis, these consistency findings support an apraxic component in B.'s speech error pattern and possibly an apraxic component for T. if they included distortions. As indicated above, reliability concerns prohibit analysis of findings at the diacritic level of narrow phonetic transcription. However, the obtained differences between B.'s and T.'s error consistency, which in this case was independent of severity of involvement (B. averaged higher Percentage of Consonants Correct scores; see Figure 2), appear to underscore the individual differences in speech error patterns that may occur in speakers suspected of having a similar genetically based motor speech disorder.

Comparison data. Consistency data for the comparison groups were available only from the conversational speech samples. First, as shown for the conversational speech analysis findings in Table 5, the speakers with S_DYS had higher mean consistency scores than the speakers with acquired AOS on each of the three consistency metrics as well as on the summative consistency metric. The latter comparison was associated with a very large and statistically significant effect size. The between-groups comparisons for the individual consistency metrics were not statistically significant, but each comparison was associated with a medium or large effect size. These findings were viewed as providing mixed support for the conventional view that inconsistent speech errors are specific for AOS, because the speakers with S_DYS also had lowered consistency of errors, although not as low as the speakers with acquired AOS. That is, although these comparative data indicate that inconsistent speech errors may be a feature of both acquired and developmental AOS, the lowered consistency data for the speakers with S_DYS suggest that the specificity of inconsistency as a marker of AOS may not be as robust, if not conceptually untenable.

To summarize, using the means and standard deviations from the two speaker groups as comparison data, B.'s lowered error consistency scores were similar to the values found for a sample of speakers with acquired AOS. In contrast, T.'s lowered consistency scores were more similar to the values obtained for a sample of speakers with acquired S_DYS.

Error Type Analyses

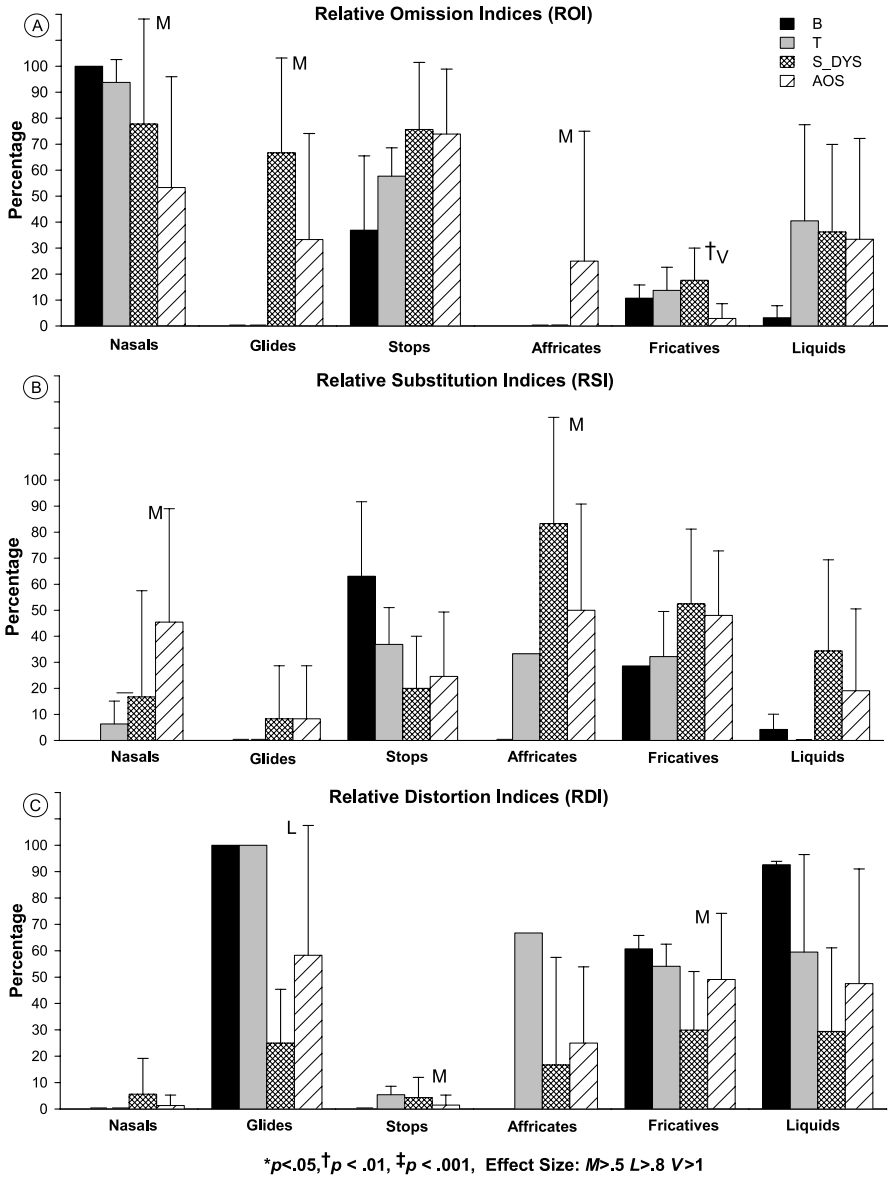
Findings. Figure 3 is a summary of the error type findings for B. and T. and the two comparative groups using SODA classifications proportioned within the six English consonant manner features. The phonetic transcription system and keyboarding procedures for computer analyses included conventions for treating some speech sound additions as substitutions and others as distortions (cf. Shriberg & Kent, 2003; Shriberg, Allen, et al., 2001). To adjust for differences in each speaker's severity of involvement, these analyses were based on metrics termed the Relative Omission Index (ROI), Relative Substitution Index (RSI), and Relative Distortion Index (RDI). The denominators for each of the three metrics (ROI, RSI, RDI) were the total number of errors on a target sound or target sound class, and the numerators were the number of errors obtained meeting requirements for each error type. Thus, as shown for B. and T. in Figure 3, the sum of their ROI (Panel A), RSI (Panel B), and RDI (Panel C) percentages equals 100% (i.e., 100% of the speech errors).

B.'s and T.'s errors on nasal targets were almost entirely omissions, whereas their errors on glide targets were always distortions. Their error types differed from one another on the remaining four manner types, particularly on stops, affricates, and liquids. Thus, B.'s and T.'s severity-adjusted error indexes at the level of manner features were not similar to one another.

Comparison data. Beginning with the comparison-group findings in Figure 3, Panel A, speakers with S_DYS had higher average ROIs than speakers with acquired AOS on five of the six manner types. These differences were associated with medium effect sizes for the nasals, glides, and affricates, and a very large effect size and a statistically significant difference ($p < .01$) for the fricative comparisons. The pattern of findings for the RSI (Panel B) comparisons across the six manner features was less clear. The speakers with S_DYS averaged higher RSI values than the speakers with acquired AOS on the affricates (medium effect size), fricatives, and liquids, but lower values on the nasals (medium effect size) and stops. The RDI findings (Panel C) were somewhat more consistent across manner classes, with speakers with S_DYS averaging lower RDI values on glides (large effect size), affricates, fricatives (medium effect size), and liquids.

Thus, using medium or larger effect size as the criterion for an empirically meaningful difference between the speakers with S_DYS compared to acquired AOS, the speech sound errors of speakers with acquired AOS were more often substitutions for nasals and distortions of glides and fricatives. The speech sound errors of those with S_DYS were more often omissions of target nasals, glides, and fricatives; substitutions for affricates; and distortions of stops. On the basis of transcription and analysis conventions used in the present comparison, these severity-adjusted findings do not support a perspective in the conventional childhood AOS literature that substitution errors predominate in speakers with apraxia and distortion errors predominate in speakers with dysarthria. Regardless of their diagnostic significance, these severity-adjusted error type findings for the adult speakers supported the present findings indicating that B.'s and T.'s errors included both distortions and other types of errors. Using the perceptually based SODA

Figure 3. Summary of findings from the error type metrics calculated from the conversational speech samples of mother (B.), daughter (T.), and the two comparison groups with motor speech disorders.



analyses, the types of speech errors made in conversation by B., T., and the speakers with acquired motor speech disorders appeared to have been dependent heavily on the manner feature of the target consonant.

Phoneme-Level Analyses

Findings. Phoneme-level analyses of B.'s errors indicated that her most frequent error sounds were the three rhotics. Her percentages correct on Targets /r/, /ɝ/, and /ɚ/ in conversational speech, respectively, were 44.6%, 71.4%, and 43.5%. The diacritic-level data indicated that nearly all of B.'s error productions were derhotacized. In comparison, T.'s percentages correct on rhotics were 86.2%, 87.5%, and 64.3%. T.'s most frequent errors were distortions and within-manner substitutions on the sibilants /s/ (74.6%), /z/ (68.6%), and /ʃ/ (40.0%), sounds on which B. had percentages correct of 86.2%, 85.0%, and 100%, respectively. A total of 83.1% of T.'s errors on these sibilants were distortions, including not only dentalized tokens but also palatalized, retroflexed, and lateralized distortions, suggesting inconsistent, imprecise lingual placement for these sounds. Such behaviors are consistent with both abnormal lingual tone (i.e., S_DYS) and motor planning issues (i.e., AOS), with all generalizations limited by the reliabilities of perceptual data reviewed previously.

Comparison data. For the comparison groups with S_DYS (excluding the participant with cerebral palsy) and acquired AOS, distortion errors on /s/ and /z/ accounted for 85.2% and 96.4% of all errors, respectively, in the conversational speech samples. For both the comparison groups, palatalized distortions of /s/ and /z/ predominated, although there were also examples of dentalized, retroflexed, and lateralized distortions. Means for the two comparison groups (minus the speaker with cerebral palsy) on the three rhotic sounds, respectively, were 94.8%, 97.2%, and 85.2% for the S_DYS group and 92.9%, 94.9%, and 95.7% for the AOS group. Thus, the two groups of speakers with acquired speech sound disorders made some errors on these two classes of speech sounds but not as many as observed in B. and T., who were presumed to have had a motor speech disorder since they began talking.

Recall that 1 speaker in the group with S_DYS had cerebral palsy. His data were retained for the present study specifically for the purpose of the present analysis. This 49-year-old speaker at assessment had received speech-language services for cerebral palsy during the developmental period. He had an error pattern not observed in B. and T. or in other speakers in the two comparison groups. His frequent errors on the sibilants /s/, /z/, and /ʃ/ consisted of 61.1% substitutions (mostly substitutions of stops), 29.6% omissions, and only 9.3% distortions. This error pattern was widely distributed across word types and phonetic contexts, suggesting that the substitutions were well established, perhaps as early learned compensatory behaviors in the service of intelligibility.

Findings from these phoneme-level analyses indicated that B., T., and the speaker with cerebral palsy have residual speech errors, that is, errors that may date back to their earliest attempts at correct articulation of ambient speech. We suggest that differentiating such individual developmental errors from those associated with their primary motor speech disorder (i.e., those shared with speakers with acquired disorders) is particularly important in speech-genetics research that uses quantitative speech phenotypes. Essentially, residual distortion and substitution errors may reflect a number of nongenetic contributions to overall severity. As such, they may require individual statistical handling in quantitative phenotypes, which ideally reflect the maximal genotypic contribution to severity of expression (cf. Shriberg, 2003).

Error Typicality Analyses

The fourth speech analysis variable as described in the Method section provided closer examination of the typicality of B.'s and T.'s speech errors, relative to the widely held view in the childhood AOS literature that atypical speech errors are specific for AOS. Table 6 provides examples of EMA errors made by B. and T. in conversational speech and on the challenging word repetition tasks. Table 6 also includes examples of EMA errors made in conversational speech by speakers in the two comparison groups. Quantitative summaries of EMA errors from either type of sample were not appropriate because the length and topics of the conversational speech samples were not equivalent and the number of imitations of each multisyllabic word was not controlled.

The number and diversity of EMA examples from B. and T. in Table 6 would classically be interpreted as providing strong support for persistent developmental AOS. As noted previously, such errors are not prevalent in speech samples from speakers of any age with other putative etiological subtypes of speech delay of unknown origin (cf. Shriberg, 2004). Moreover, it may not seem parsimonious to ascribe them to a developmental classification such as childhood aphasia. As reviewed previously, until such errors can be analyzed instrumentally, no position is taken on their diagnostic significance as support for AOS.

Prosody and Voice Analyses

Figure 4, Panel A, shows a summary of prosody and voice findings for B.,T., and the two comparison groups of adults with motor speech disorders. Overall, B. and T.'s prosody–voice profiles included scores below 80% (i.e., fail) in 5 of the 7 prosody–voice domains. For the following presentations and discussions of results, it was efficient to interleave case study findings with findings from the comparison groups.

Phrasing

B.'s and T.'s lowered appropriate phrasing scores (75% and 66.7%, respectively) were notable. Typical adult female speakers average over 85% appropriate phrasing (SD = 10.9%) in conversational speech (Lewis & Shriberg, 1994). B.'s and T.'s phrasing scores were more than 1 SD below those of the speakers with S_DYS (M = 89.9%, SD = 5.5%) and within 1 SD of the lowered mean scores for speakers with acquired AOS (M = 55.3%, SD = 25.8%). As shown in Figure 4, there was a significant difference between the average scores of the two comparison groups (effect size [very large] = 1.54, p =.004). Thus, the phrasing data supported an apraxic component for both B. and T.

Analyses of the phrasing findings at the level of PVSP subcodes were informative. Although they had lowered summative scores, B. and T. did not make the types of phrasing errors observed in speakers with acquired AOS. As shown in Figure 4, Panel A, approximately 55% of the utterances of the speakers with acquired AOS included one or more part- or whole-word

Table 6. Examples of epenthesis, metathetic, and assimilative errors.

	Stimulus					
	B.			T.		
Error	Orthographic	Phonetic	Production	Orthographic	Phonetic	Production
Epenthesis	assembler	əsɛmblɚ	əksɛmbəlɚ	municipal	mjunɪsəpl̩	_junɪʃənpːl
	bicyclist	baɪsəklɪst	baɪskəkəl ɪst[a]	skeptical	skɛptɪkl̩	skɛptɪd^{r}tkl̩
	consciousness	kantʃəsnəs	kantʃənsnəs	statistics	stətɪstɪks	s_ə^{s}tɪstɪk_
	consequence	kansɛkwɛns	kansɛnkwɛnt			
	orchestra	ɔrkɪstrə	ɔrkrɪstrə			
	statistics	stʌtɪstɪks	s_ʌstɪstɪks			
			s_ʌstɪstɪks[b]			
			stʌstɪstɪks			
Metathesis	bicyclist	baɪsəklɪst	baɪskəlɪst	bicyclist	baɪsəklɪst	baɪskəlɪθ_,
						baɪskəlɪs_
	cinnamon	sɪnəmɪn	sɪmənɪm	cinnamon	sɪnəmɪn	sɪmənəm
						sɪnənɪm
	municipal	mjunəsəpl̩	mjənusəbl	Colorado	kɔləradoʊ	kɔdəral^{ə}toʊ
			n_əmːɪsəbl			
	permanent	pɝmənɪnt	pɝnəmɪnt˺	skeptical	skɛptʌkl̩	skɛp^{t}kʌtl̩
	skeptical	skɛptəkl̩	stɛpkəkl̩			
Assimilation	Colorado	kalɚadoʊ	kadɚadoʊ	associate	əsoʊʃɪɛt	əʃoʊʃɪɛt˺
	mobilize	moʊbəlaɪz	moʊboʊlaɪz	caterpillar	kætɚpɪlɚ	kæləpɪlɚ
	municipal	mjunɪsəpl̩	n_unːɪsəbl̩	Colorado	kaləradoʊ	kaləralʊ
	parallel	pɛrəlɛl	pɛrɚrɛl	octopus	aktəpʊs	astəpʊs
	skeptical	skɛptəkl̩	stɛptəkl̩	orchestra	ɔrkəstrə	ɔrkɚstrə
	symphony	sɪmfənɪ	sɪnfənɪ	philosophy	fəlɔsəfɪ	fəlɔsəsɪ
				skeptical	skɛptəkl̩	skɛptətl̩
	S_DYS			AOS		
Epenthesis	and	æn	^{n}æn	America	əmɛrɪkʌ	ə^{v}mɛrɪkʌ
	in	ɪn	dɪn	another	ənʌðɚ	ɑlnʌdɚ
	the	ðə	d^{r}ɚ	are	ar	^{g}ar
	yeah	jæ	^{3}jæ	because	bəkʌz	pʌzkʌz
				divorce	dɪvɔrs	dɪvɔrs^{t}
				every	ɛvrɪ	ɛvrɪ^{z}
				had	hæd	hæd^{n}
				his	hɪz	hɪns
				hotel	hoʊtɛl	hoʊ^{t}tɛl^{d}
				international	ɪnɚnæʃənəl	ɪnɚnænʃənəl
				it	ɪt	jɪt˺
				know	noʊ	noʊ^{n}
				laundry	landrɪ	glandrɪ
				lunch	lʌntʃ	lʌntʃ^{t}
				my	maɪ	maɪ^{n}
				see	si	si^{z}
				the	ðə	dɚ^{d}
				then	ðɛn	ðɛn^{ð}
				to	tə, tu	tə^{r}, tu^{d}
				Wagner	vægənɚ	vækt^{ə}nɚ
				well	wɛl	wɛl^{d}
				you	jə	jəd

(table continues)

Table 6 *(continued).*

	Stimulus					
	S_DYS			AOS		
Error	Orthographic	Phonetic	Production	Orthographic	Phonetic	Production
Assimilation	a lot	əlat	əlal	calendars	kælʊndɚz	pæ_ʊnzɚz
	behavior	bihe͞ɪvjɚ	bihe͞ɪbjɚ	customers	kʌstəmɚz	tʌstəmɚz
	Berkeley	bɝkəlɪ	bɚkɚ_ɪ	dog	dɔg	gɔg
	father	faðɚ	faðɚ	enjoyed	ɪnʤɔ͞ɪd	ɪnʤo͞ʊn
	graduated	grædʒuē͞ɪtəd	grædʒɚe͞ɪtə_	microprocessor	mā͞ɪkro͞ʊprasɛsɚ	mā͞ɪkro͞ʊprasɛsɚ
				microprocessors	mā͞ɪkro͞ʊprasɛsɚz	mā͞ɪkroprasɛsɚz
				Montgomery	mantgʌmərɪ	məŋkgʌmᵖənɪ
				portfolio	pɔrtfo͞ʊlɪo	pə_pfo͞ʊlɪə
				Saturdays	sætɚdē͞ɪz	sæ_ɚzɪz
				symphonic	sɪmfanɪk	sɪmfanɪŋ
				vacation	vē͞ɪkē͞ɪʃɪn	vē͞ɪfē͞ɪʃɪn

Note. For B. and T., examples are primarily from the challenging word repetition tasks. Only one word (B:*assembler*) that met criteria for an EMA error occurred in B. and T.'s conversational speech samples. For the two comparison groups, examples were available only from the conversational speech samples. No examples of metathesis were found in the conversation samples of the two comparison groups.

[a]Also meets definition for metathesis. [b]Also meets definition for assimilation. [c]Also meets definition for epenthesis.

repetition or revision (i.e., had inappropriate phrasing). Content analyses indicated that unlike findings for B., T., and the speakers with S_DYS, many of the repetitions and revisions in the speakers with acquired AOS appeared to be the expected attempts at self-correction or articulatory modification. As annotated by the transcriber, the more severely involved speakers with acquired AOS appeared to be "revising repetitions and repeating revisions." As noted previously, such behaviors have been described conventionally as a primary characteristic of AOS. Thus, although B.'s and T.'s utterances contained frequent repetitions and revisions, anecdotally, they did not seem to have the quality of attempts to self-correct that was clearly evident in the speakers with acquired AOS. Odell and Shriberg (2001) and Shriberg et al. (1997b, 1997c) reported comparable findings: Children suspected to have developmental AOS in several study samples did not make the types of self-monitoring repairs that were dramatically evident in the conversational speech of the present adults with acquired AOS. Discussion of such differences has invoked differences in children's representational levels of phonology as possible explanatory variables (cf. Odell & Shriberg).

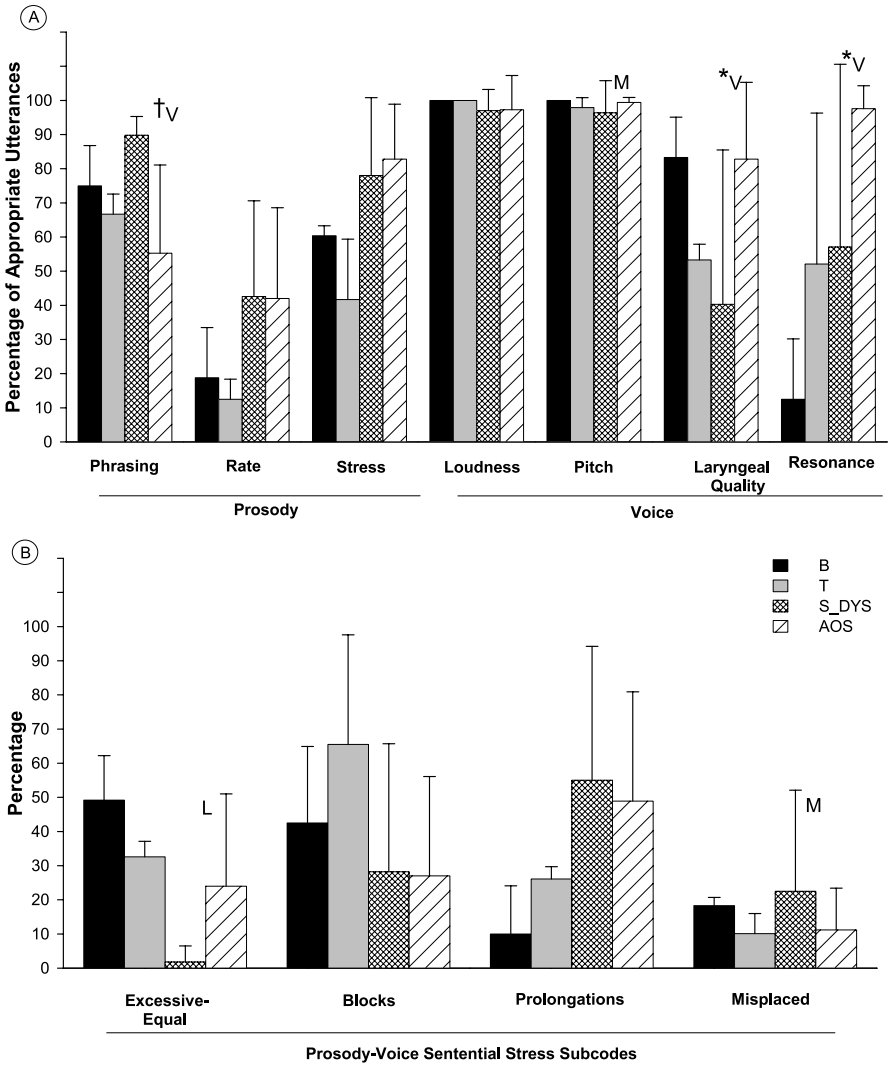
Rate

B. and T. both had notable challenges in speech rate, with only approximately 20% of their utterances coded as having appropriate rate. Examination of the rate subcodes indicated that almost all inappropriate utterances were coded as *too slow* in both speech time and pause time components (i.e., compared with the subcode used for slowness for pause time only; see Figure 1). B. and T.'s average rates (in syllables/second) for utterances coded with inappropriate rate from the first and second conversational samples (separated by a slash) were timed as follows: B.—$M = 2.6/2.8$, $SD = 0.6/0.6$, minimum = 1.6/1.4, maximum = 3.8/3.8; T.—$M = 2.7/2.4$, $SD = 0.7/0.7$, minimum = 1.4/1.3, maximum = 4.2/3.8. These rates are slower in general than the values for all utterances from normal speakers of comparable ages reported by Duchin and Mysak (1987). As indicated in the list of diagnostic features in Table 2, slow rates, quite similar within and across the present two conversational speech samples separated by 4 months, have been reported for adults with both acquired S_DYS and with acquired AOS (cf. Duffy, 2005). (Note in Figure 4, Panel A, that the comparison groups also had over 50% of utterances that were inappropriate in rate, also subcoded as *too slow* because of lengthened speech and lengthened pause times.) Thus, B. and T.'s slow rates are consistent with extant characterizations of both acquired S_DYS and acquired AOS.

Stress

The assessment protocol (Table 1) included two stress tasks, motivated by promising findings in prior studies indicating that inappropriate stress may be a sensitive and specific diagnostic marker for childhood AOS (Shriberg et al., 1997b, 1997c; Shriberg, Campbell, et al., 2003). The following sections review findings from the stress domain subcodes for sentential stress in conversational speech and from the Lexical and Emphatic Stress Tasks (see Figure 1).

Figure 4. Panel A: summary of findings from the seven prosody and voice domains coded perceptually from the conversational speech samples of mother (B.), daughter (T.), and the two comparison groups with motor speech disorders. Panel B: summary of findings from the four stress subcodes coded perceptually from the conversational speech samples of mother (B.), daughter (T.), and the two comparison groups with motor speech disorders.



Sentential stress. As shown in Figure 4, Panel A, B. and, particularly, T. had lower percentages of utterances with appropriate sentential stress compared with average stress percentage for the speakers in the two adult comparison groups. Figure 4, Panel B, includes findings for the four most frequent types of behaviors underlying B.'s and T.'s inappropriate sentential stress and averaged values for speakers in the two comparison groups. The four most frequently coded stress error subcodes were as follows: (a) *excessive-equal* stress (the percept of syllable-timed speech due to over/understressing syllables), (b) *blocks* (an articulatory block, usually on a stop consonant, that calls undue attention to the word), (c) *prolongations* (a prolongation on a continuant sound that calls undue attention to the word), and (d) *misplaced stress* (stress on a typically unstressed word in the utterance). These four percentages for each speaker group do not sum to 100% because the coding procedures allowed for more than one inappropriate stress code and/or subcode per utterance.

As shown in Figure 4, Panel B, the speakers with S_DYS and acquired AOS had generally similar error type patterns, with approximately half of their stress errors coded as prolongations of consonants and vowels and the remaining errors approximately equally divided among excessive-equal stress, blocks, and misplaced

stress. There were two comparisons associated with medium to large effect sizes; however, none of the between-groups differences in subtypes of sentential stress were statistically significant. Compared with the percentages for the speakers with S_DYS, proportionally more of the utterances with inappropriate stress from speakers with acquired AOS had excessive-equal stress (S_DYS: M = 1.8%, SD = 4.7%; AOS: M = 24.0%, SD = 27.0%; effect size = 0.97 [large]; p = .058). In contrast, proportionally more of the inappropriate stress codes from speakers with S_DYS were due to misplaced stress (S_DYS: M = 22.5%, SD = 29.6%; AOS: M = 11.2%, SD = 12.2%; effect size = 0.53 [medium]; p = .278).

B.'s and T.'s stress error type proportions differed somewhat from one another and from findings for the two comparison groups summarized above. Approximately 50% of B.'s stress errors were classified as excessive-equal stress, with an additional approximately 43% coded as blocks. T.'s stress errors included approximately 65% blocks and approximately 30% prolongations (multiple occurrences per utterance can yield more than 100%). Thus, in contrast to the more frequently occurring prolongation errors in the speakers with S_DYS and acquired AOS, B.'s and, in particular, T.'s stress errors were more frequently blocks. These findings were somewhat unexpected, but consistent with T.'s expressed concern with her lifelong problem with "blocking" on sounds (we did not attempt to differentiate such blocks from true disfluencies). Clearly, the perceptual definition of "blocks" used in the present study requires examination at the articulatory level using instrumental approaches. As indicated in Table 2, we theorized that inappropriate sentential stress would be a feature of both S_DYS and AOS.

Lexical stress. Inappropriate lexical stress was posited to be specific for AOS on the basis of findings that indicated that it may be a possible diagnostic marker of childhood AOS (Shriberg, Campbell, et al., 2003). LSR values were obtained for B. and T. using the procedures described in the Method section. Values from Session 1 were analyzed and compared with those of the 54 children assessed with the Lexical Stress Task and analyzed using the acoustic procedures reviewed in the Method section and described fully in Shriberg, Campbell, et al. (2003) and Shriberg, McSweeny, et al. (2006). T.'s average LSR value fell near the middle of the distribution of stress values for the 54 speakers with speech sound disorder, including some suspected to have childhood AOS. However, B.'s average LSR value was tied for the third highest ratio among the 54 values, considerably beyond 1 SD from the mean. Although the 54 speakers were younger than B., the LSR normalizes for differences in the three parameters of stress; moreover, T.'s LSR values were within the distributions obtained on children averaging approximately 7 years of age. For the present purposes, we concluded that these LSR findings supported classifying B. but not T. as having a deficit in lexical stress. In planned additional analyses of the acoustic data from this task, we will attempt to identify the source of B.'s stress difference (i.e., in intensity, frequency, and/or duration), particularly in relation to the suggestion of a core timing deficit in affected KE family members (Alcock et al., 2000b).

Emphatic stress. B. scored 6 out of 8 (75%) correct stress on both the first and second assessment-session administrations of the Emphatic Stress Task. T. scored 7 out of 8 (87.5%) and 6 out of 8 (75%) in the two administrations. Thus, although the task was designed for very young children, with typical speakers getting 100% correct scores, B. and T. had some difficulty imitating emphatic stress. In addition to production-level difficulties, problems with their perception of emphatic stress cannot be ruled out, particularly in view of the role grammar plays in rapid and successful imitations of the words emphasized in these brief sentences, in relation to B.'s and T.'s grammatical deficits (Table 4).

The more general question of the role of perception in inappropriate emphatic stress is a topic of some interest (cf. Alcock et al., 2000b; Maassen, Groenen, & Crul, 2003; Marion, Sussman, & Marquardt, 1993). As with the Lexical Stress Task findings described previously, acoustic findings on the source of stress leveling (i.e., in amplitude, frequency, and/or duration), should contribute to an eventual account of the neurolinguistic substrates mediating appropriate emphatic stress in AOS. For the present focus, because B. and T. were correctly able to imitate emphatic stress on nearly all of the items, we interpreted the findings conservatively as providing only marginal support for the hypothesis of AOS.

Voice: Loudness, Pitch, Laryngeal Quality, and Resonance

Loudness and Pitch

B.'s and T.'s loudness and pitch were perceptually within normal limits on nearly all utterances. T.'s pitch was perceived as questionably low but rated as within the normal range for most of the 24 utterances (see Figure 4). As described in the PVSP manual, perceptual coding of loudness and pitch are only reliable when markedly inappropriate and instrumental measures are needed to quantify fine-grained differences. On the bases of these perceptually based findings, B.'s and T.'s appropriate loudness and appropriate pitch did not support the hypothesis of S_DYS (Table 2).

Laryngeal Quality

Typical adult female speakers average over 83% appropriate laryngeal quality (SD = 28.7%) in conversational speech (Lewis & Shriberg, 1994). Thus, findings

for B. were considered inconclusive because 83% of her utterances were coded as appropriate. However, we viewed the findings for T., who had appropriate laryngeal quality on only 53% of utterances, with most of the inappropriate utterances coded as *rough,* as consistent with S_DYS (Figure 4).

Compared with the speakers in this study who had acquired AOS and whose average values were similar to those of typical speakers, the speakers with S_DYS had significantly lower percentages of utterances with appropriate laryngeal voice quality (S_DYS: M = 40.3%, SD = 45.2%; AOS: M = 82.8%, SD = 22.4%; effect size = 1.3 [very large]; p = .001). Rough quality was also the subcode used most often to characterize the inappropriate laryngeal quality of the speakers with S_DYS. Comparable adjectives have been used to describe this quality as a major vocal feature of S_DYS (Duffy, 2005), although other vocal features (e.g., *strain-strangle*) may have higher specificity.

Resonance

Typical adult female speakers average over 95% appropriate resonance (SD = 13.0) in conversational speech samples (Lewis & Shriberg, 1994). Both B.'s and T.'s frequently inappropriate resonance (particularly B.'s; see Figure 4) was perceived as hypernasal. The speakers with S_DYS also had a significantly lower percentage of utterances with appropriate resonance (S_DYS: M = 57.1%, SD = 53.5%) compared with the speakers with acquired AOS (M = 97.6%, SD = 6.7%; effect size = 1.27 [very large]; p = .013). Thus, the resonance findings for B. and T. suggest the possibility of underlying deficits in velopharyngeal function. Ballard et al. (2006) addressed associated questions in a review of perceptual and instrumental findings from the speech mechanism exam and speech motor tasks.

Summary

We interpret the findings for 13 speech, prosody, and voice variables assessed in this case study as support for characterizing B.'s and T.'s speech deficit as a mixed disorder that includes features of both S_DYS and AOS. Table 7 provides a summary of findings organized by the variables described in Table 2. For the 2 variables posited to be characteristic of both S_DYS and AOS (i.e., inappropriate rate [No. 6] and inappropriate sentential stress [No. 7]), findings for B. and T. (both 2/2 = 100%) supported a mixed disorder. For the 4 voice and resonance variables posited to be specific for S_DYS—inappropriate loudness (soft voice; No. 10), inappropriate pitch (low pitch; No. 11), inappropriate laryngeal quality (harsh; No. 12), and inappropriate resonance (consistent hypernasality; No. 13)—findings were somewhat stronger for T. (2/4 = 50%) than B. (1/4 = 25%). For the 3 variables posited to be specific for AOS—inappropriate phrasing (No. 5), inappropriate lexical stress (No. 8), and inappropriate emphatic stress (No. 9)—findings were somewhat stronger for B. (3/3 = 100% than for

Table 7. Summary of case study findings.

No.	Variable: brief description (see Table 2)	Consistent with		Findings			
				B		T	
		Spastic dysarthria	Apraxia of speech	S_DYS	AOS	S_DYS	AOS
2	Inconsistent errors	?	?	(Yes)	(Yes)	(Yes)[a]	(Yes)[a]
3	Primarily distortion errors	?	?	(No)	(No)	(No)	(No)
4	EMA errors		?		(Yes)		(Yes)
5	**Inappropriate phrasing**		X		Yes		Yes
6	Inappropriate rate	X	X	Yes	Yes	Yes	Yes
7	Inappropriate sentential stress	X	X	Yes	Yes	Yes	Yes
8	**Inappropriate lexical stress**		X		Yes		No
9	**Inappropriate emphatic stress**		X		Yes[a]		Yes[a]
10	**Inappropriate loudness**	X		No		No	
11	**Inappropriate pitch**	X		No		No	
12	**Inappropriate laryngeal quality**	X		No[a]		Yes	
13	**Inappropriate resonance**	X		Yes		Yes	
	Total % yes						
	Without parentheses			50	100	67	80
	Including parentheses			50	88	63	75

Note. The seven diagnostic hypotheses are boldface.

[a]Marginal support.

T. (2/3 = 67%). Finally, for the first three more controversial diagnostic features in Table 2 and Table 7—inconsistency, distortions, and EMA errors—B.'s and T.'s data (in parentheses) ranged from 50% to 67% support for each motor speech disorder. Summing over both the provisional and nonprovisional entries for S_DYS and AOS, as shown at the bottom of Table 7, findings for both speakers indicated 50% to 100% support for involvement in each disorder. These summary findings are consistent with the first five coauthors' clinical impressions of a mixed disorder in both speakers.

Discussion

Methodological Constraints

Several methodological limitations in the present design should be noted before a discussion of some perspectives. First, as indicated previously, findings are limited by the sensitivity and reliability of the auditory–perceptual systems used to characterize speech and prosody–voice. Particularly for the voice and resonance variables, we consider these data to be conservative estimates of involvement, pending additional information from instrumental measures. Second, as suggested in discussion of the emphatic stress findings, missing from the assessment protocol for this case study were tasks assessing auditory processing and other encoding aspects of speech and prosody that have been studied in relation to motor speech disorders. Third, the design did not include comparison groups who could have provided important information on subtypes of dysarthria. Lacking such data (i.e., findings differentiating spastic from ataxic dysarthria), it is not appropriate to speculate on possible neurological correlates from the present array of findings. Last, any differences between B.'s and T.'s developmentally based deficits and those of the adults with acquired motor speech disorders were confounded by B.'s and T.'s lifetime of treatment and their individual adjustments to early and persistent communication difficulties.

Descriptive Perspectives

The primary findings of this study—that B. and T. have a mixed speech disorder consistent with S_DYS and AOS—were unexpected, on the basis of the information provided in the KE studies and, more generally, on the basis of studies in the literature on developmental (i.e., nonacquired) AOS. As reviewed previously, there are some case studies in the medical literature on neurodevelopmental disorders and in the emerging *FOXP2* literature that suggest comorbid apraxia and dysarthria, but few clinical data support such claims. One suggestion, at least in the interim, that is based on the present findings is that *sensorimotor disorder* (e.g., McNeil, 1997) may be a more useful cover term for the phenotype associated with *FOXP2* and other emerging genetic findings. This perspective might have the heuristic value of encouraging inclusion of more detailed clinical protocols in speech-genetics research, reserving use of more specific classificatory terms such as AOS (i.e., verbal dyspraxia) to findings that include the relevant types of perceptual and instrumental assessments for differential diagnostic classification.

Notwithstanding the several methodological constraints reviewed above, the heterogeneities observed within and between B. and T. underscore the complex of genetic and nongenetic variables that likely have contributed to their current profiles. As in research with other neurodevelopmental disorders, B.'s and T.'s genotypic similarity provides only a starting point toward explanatory accounts linking genotype differences to proximal neural substrates to communication profiles. These individual differences illustrate why it has been so difficult for researchers to agree on the diagnostic features that characterize the putative developmental form of AOS. One possibility is that there may be subtypes of sensorimotor disorders associated with different forms of *FOXP2* involvement, as well as multiple causal genetic loci. On this issue, Vargha-Khadem et al.'s (2005) conclusions on the possibility of a core impairment observed in affected KE family members are particularly relevant, because, as described in Ballard et al. (2006), B. and T. do not appear to have an orofacial apraxia. Vargha-Khadem et al. (2005) stated,

> The extensive behavioral data on the KE family, combined with the success of linkage analysis, support the proposal that there is at least one core deficit—orofacial dyspraxia—underlying the speech and language disorder of the affected members. However, it is unclear whether their associated grammatical, semantic and other cognitive impairments are all secondary consequences of this fundamental deficit, or whether they point instead to the existence of additional core deficits. (p. 132)

Forthcoming reports that are based on neuroimaging findings for B. and T. will address related issues.

Research and Clinical Perspectives

These case study findings address the second goal of this article, which was to underscore the implications for *phenomics* (phenotype–genotype relationships) of assessment and analyses methods in speech-genetics research. It is clear that an eventually successful search for the genetic contributions to speech sound disorders, such as childhood AOS, will require well-developed phenotypes. In the present data, for example, B.'s rhotic and T's sibilant distortion errors illustrate the complexity of

speech processing domains that require description in speech-genetics research. Differentiating such errors in the proband, nuclear, and extended family members is important in research in childhood AOS as well as in genetics research in other subtypes of childhood speech sound disorders. Such potentially important conceptual differences in error typology may often go unexamined in studies that do not use at least narrow phonetic transcription or instrumentation or do not reference obtained behaviors to relevant findings in both the developmental and acquired speech sound disorder literature.

In addition to disorders likely to have monogenic origins, such as the motor speech disorders associated with *FOXP2* in the present 2 participants, susceptibility regions for speech sound disorders likely to have polygenic origins have recently been reported (cf. Smith, Pennington, Boada, & Shriberg, 2005; Stein et al., 2004; see *Online Mendelian Inheritance in Man, Speech Sound Disorder,* see %608445). Research on genetically based speech disorders, including those occurring in the context of other neurodevelopmental disorders, will require new assessment methods to enable investigators to exploit the power of continual advances in genetic bioinformatics. For example, as illustrated in this article, phenotypic metrics with high sensitivity and specificity are needed to differentiate speakers with (a) nongenetic, developmental speech sound errors; (b) genetically based developmental speech sound errors; (c) genetically based forms of apraxia, dysarthria, and aphasia; and (d) acquired forms of apraxia, dysarthria, and aphasia. The present study and associated subtyping research using perceptual (Shriberg, Lewis, et al., 2005), acoustic (e.g., Karlsson, Shriberg, Flipsen, & McSweeny, 2002; Shriberg, Flipsen, Karlsson, & McSweeny, 2001), and automatic speech recognition (e.g., Hosom, Shriberg, & Green, 2004) technologies suggest that it may be possible to develop valid, reliable, and efficient instruments for such research and its eventual applied goals (i.e., to sharpen genetic-based and other phenotypes).

Last, from a clinical perspective, B.'s and T.'s persistent speech, prosody, and voice challenges highlight the need for assessment and treatment methods for children with both AOS and clinical or possibly subclinical forms of dysarthria. Among other implications, the long-term consequences of regulator genes such as *FOXP2* are important considerations in clinical decision making. Normalization trajectories for the two types of motor involvements contrasted in the current case study could be regulated by different genetic substrates, with implications for the timing of treatment focused on apraxic versus dysarthric speech processes. For its eventual contribution to such clinical needs—information that has not been available to date to guide B.'s and T.'s clinical services—speech-genetics research is likely to yield valuable findings.

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Contact author: Lawrence D. Shriberg, Room 439, Waisman Research Center, University of Wisconsin—Madison, 1500 Highland Avenue, Madison, WI 53705.
E-mail: shriberg@waisman.wisc.edu