

# Developmental Apraxia of Speech: III. A Subtype Marked by Inappropriate Stress

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Two prior studies in this series (Shriberg, Aram, & Kwiatkowski, 1997a, 1997b) address the premise that children with developmental apraxia of speech (DAS) can be differentiated from children with speech delay (SD) on the basis of one or more reliable differences in their speech. The first study compared segmental and prosody-voice profiles of a group of 14 children with suspected DAS to profiles of 73 children with SD. Results suggest that the only linguistic domain that differentiates some children with suspected DAS from those with SD is inappropriate stress. The second study cross-validated these findings, using retrospective data from a sample of 20 children with suspected DAS evaluated in a university phonology clinic over a 10-year period.

The present study is of particular interest because it cross-validates the prior stress findings, using conversational speech samples from 19 children with suspected DAS provided by five DAS researchers at geographically diverse diagnostic facilities in North America. Summed across the three studies, 52% of 48 eligible samples from 53 children with suspected DAS had inappropriate stress, compared to 10% of 71 eligible samples from 73 age-matched children with speech delay of unknown origin.

Discussion first focuses on the implications of stress findings for theories of the origin and nature of DAS. Perspectives in psycholinguistics, neurolinguistics, and developmental biolinguistics lead to five working hypotheses pending validation in ongoing studies: (a) inappropriate stress is a diagnostic marker for at least one subtype of DAS, (b) the psycholinguistic loci of inappropriate stress in this subtype of DAS are in phonological representational processes, (c) the proximal origin of this subtype of DAS is a neurogenetically specific deficit, (d) the distal origin of this form of DAS is an inherited genetic polymorphism, and (e) significant differences between acquired apraxia of speech in adults and findings for this subtype of DAS call into question the inference that it is an apractic, motor speech disorder. Concluding discussion considers implications of these findings for research in DAS and for clinical practice.

**KEY WORDS:** apraxia, phonology, speech, children, disorders

**F**indings from a local ascertainment study support the clinical functionality of the term *suspected developmental apraxia of speech (DAS)* (Shriberg et al., 1997a). In two following studies of the conversational speech of children with suspected DAS, 43% of 14 children and 58% of 20 children had inappropriate phrasal stress (Shriberg et al., 1997b). Measurement and conceptual considerations supported the validity of the stress deficit as a candidate diagnostic marker for DAS, pending external cross-validation. The present study provides the external cross-validation support that was deemed crucial for the claim

that inappropriate stress may be a diagnostic marker for suspected DAS. We report speech and prosody-voice findings from a sample of children with suspected DAS as defined by investigators at five clinical-research sites in North America. Following a presentation of results, which include summary analyses of the two prior studies and the present study, we consider five hypotheses about the nature and origin of suspected DAS. In the final section we consider implications of the stress findings for research in DAS and for clinical practice.

## Method

### Procedures

Six persons with clinical-research programs in developmental apraxia of speech were contacted for their assistance in a study of developmental apraxia of speech. Researchers were asked to select conversational speech samples from 2–4 children who, by the researcher's criteria, warranted the classificatory term *developmental apraxia of speech*, or any preferred variant of this term (e.g., *developmental verbal dyspraxia*). The two inclusionary constraints were that (a) the child's cognitive status was within normal range and (b) there was no known developmental or acquired disorder affecting the speech-hearing mechanism, cognitive functioning, or psychosocial processes. The researchers were asked to attempt to provide samples of both younger (3- to 6-year-old) and older (7- to 16-year-old) children with suspected DAS.

Five of the six researchers contacted were able to forward speech samples and complete subject forms within the requested time period. A total of 21 tapes and completed subject information forms, including repeated samples for 2 children, were returned within 6 months of receipt of the request package. All audiocassette tapes sent to Wisconsin were either original recordings made on high-quality tapes provided to each researcher, original recordings made on tapes of comparable quality, or copies of original high-quality tapes made on the tapes provided. The conversational speech samples were transcribed and prosody-voice coded by the same transcriber who completed all transcription tasks for the audiocassette recordings in Study I. She was provided with only the gender and age of the subject on each tape. Because the transcriber had been involved in many studies of children with developmental phonological disorders, she was accustomed to transcribing children of all severity levels. A research assistant error-checked and entered the phonetic transcripts and prosody-voice codes into the suite of analysis programs described for Study I in Shriberg et al. (1997b).

## Subjects

Table 1 is a summary of the age, gender, cognitive-language status, and classification rationale for the 19 children with suspected DAS. For continuity with the two studies reported in the prior paper in this series (Shriberg et al., 1997b) this sample is referred to as Study III. The children in Study III represented children with suspected DAS from clinical-research sites in Iowa, Massachusetts, Ohio, Ontario, and Texas. As shown in Table 1, researchers provided conversational speech samples from 2 to 6 children, with two researchers providing samples of a child at two points in time. Ages of the 19 children, 10 girls and 9 boys, ranged from 4 years 7 months to 14 years 4 months. Using test data available at each site, researchers classified children's cognitive level as *Within Normal Limits (WNL)* or *Low Normal (LN)*, and receptive and expressive language levels as *Within Normal Limits*, *Low Normal*, or *Below Age Level (BAL)* (nominally, below one standard deviation from age level on a standardized test).

The rightmost column in Table 1 includes summaries of classification information for each child provided by each of the five researchers. As expected, the basis for classifying a child as suspected DAS varied considerably across research sites, reflecting the range of deficits in speech, prosody-voice, and nonspeech domains seen on symptom checklists described in the first paper in this series (Shriberg et al., 1997a). However, within each research group, a set of characteristics was used fairly consistently to identify samples for this project.

Table 2 is a summary of the speech status data for the 19 children in Table 2, divided by age into younger and older subgroups of children with suspected DAS. The younger subgroup of children with suspected DAS included 11 samples from children aged 4 years 7 months to 6 years 6 months, with a mean age of 5 years 3 months. The older subgroup of children with suspected DAS included 10 samples from children aged 7 years 11 months to 14 years 4 months, with a mean age of 10 years 9 months. Thus, children in the older group were approximately twice as old as children in the younger group. As indicated in Table 2, samples labeled Child 7 and Child 14 are from the same child, as are samples labeled Child 11 and Child 17. In the statistical analyses to follow, the second samples from these 2 children are excluded.

The Percentage of Consonant Correct scores and corresponding PCC severity level adjectives for children in the younger group with suspected DAS in Table 2 ranged from Mild to Severe, averaging Mild-Moderate (PCC:  $M = 67.1\%$ ,  $SD = 12.3\%$ ). SDCS classifications indicated that 7 of the 11 speech samples met developmental error criteria for speech delay (SD) and two met criteria for questionable speech delay (QSD). Three children also marginally met criteria for speech delay+

**Table 1.** Age, gender, cognitive-linguistic information, and classification rationale for 19 children with suspected DAS provided by five researchers in DAS.

Researcher Child <sup>b</sup>	Age at sample (yrs;mos)	Gender	Cognitive-language status <sup>a</sup>			Basis for classification as suspected DAS
			Cognitive language	Receptive language	Expressive language	
A	1 <sup>c</sup>	M	WNL	WNL	WNL	(1) Phonemic inventory significantly less developed than phonetic inventory; (2) numerous vowel errors; (3) unusual and persisting speech errors; (4) intonation and stress inconsistencies; (5) poor performance on diadochokinetic tasks. Same features (1)–(5) as Subject 1 above plus inappropriate loudness.
	9	M	WNL	WNL	BAL	Same features (1)–(5) as Subject 1 above, plus oral groping movements.
	12	M	WNL	LN	BAL	Same features (1)–(5) as Subject 1 above, plus inappropriate loudness, oral groping movements, and poor progress in speech/language treatment.
	21	F	WNL	WNL	BAL	(1) Multiple sound errors; (2) inconsistent productions; (3) unable to imitate oral movements; (4) clinical diagnosis.
B	3	F	WNL	WNL	BAL	(1) Multiple sound errors.
	6	F	WNL	BAL	BAL	(1) Multiple sound errors; (2) inconsistent productions; (3) groping movements.
	8	F	WNL	WNL	BAL	(1) Multiple sound errors; (2) inconsistent productions; (3) groping movements.
	11 <sup>c</sup>	F	LN	BAL	BAL	(1) Diagnosed as DAS at preschool age.
	16	F	WNL	BAL	BAL	See Subject 11 above—second sample from same child.
	17	F	LN	BAL	BAL	(1) Severely unintelligible speech; (2) unable to imitate oral motor movements; (3) groping movements of the articulators; (4) inconsistent speech errors; (5) unusual speech errors such as metathetic errors.
C	19	M	WNL	BAL	BAL	Does not meet criteria of one of three other types of speech disorders, and oral movements requiring changing planes are inadequate for age.
	4	F	WNL	LN-WNL	BAL	Meets same criteria as for Subject 4 above.
D	5	M	WNL	WNL	BAL	Meets same criteria as for Subject 4 above.
	10 <sup>c</sup>	M	WNL	WNL	BAL	(1) Variable and inconsistent errors; (2) vowel distortions; (3) voicing errors; (4) increase in errors and decrease in intelligibility with increase in length of stimuli or utterance; (5) imprecise and slow DDK rates—speech and nonspeech; (6) “islands of intelligibility;” (7) nasal emissions; (8) problems with volitional oral movement: oral apraxia. Co-occurring problems: poor sucking as infant; drooling; some problems with choking on foods and liquids; slow gross and fine motor development; slow development of speech skills.
E	2	M	LN	LN	LN	Same features (1)–(5) as Subject 2 above, plus metathetic errors—syllable reversals, consonant reversals within syllables; decrease in accuracy with increase in effort; “groping” observed; addition errors; repetition errors. Co-occurring problems: academic problems (reading); word-retrieval problems.
	15	F	WNL	WNL	BAL	Same features (1)–(5) as Subject 2 above, plus metathetic errors; oral apraxia; prosody “off” at times; variable performance across remedial sessions. Co-occurring problems: academic difficulties (reading); family with many members exhibiting significant speech problems.
E	18	F	LN	LN-WNL	LN	Same features (1), (3)–(4) as Subject 2 above, plus difficulties with volitional oral movement; oral apraxia; difficulties sequencing phonemes and syllables.
	20	M	LN	BAL	BAL	(1) Receptive-expressive gap; (2) lack of consonant production in babble and early speech; (3) effortful, groping, unintelligible speech with pauses; (4) poor phonotactics.
E	7	M	WNL	WNL	BAL	Same features (1)–(3) as Subject 7 above, plus very idiosyncratic words; unable to imitate or approximate others.
	13	M	WNL	WNL	BAL	See Subject 7 above—second sample from same child.
E	14	M	WNL	WNL	BAL	See Subject 7 above—second sample from same child.

<sup>a</sup>WNL: Within Normal Limits; LN: Low Normal; BAL: Below Age Level.

<sup>b</sup>Identifying numbers were assigned by increasing age (see Table 2).

<sup>c</sup>Prosody-Voice Screening Profile could not be completed due to technical or content constraints on the conversational speech sample.

**Table 2.** Speech status of 19 children with suspected DAS provided by five researchers in DAS.

Child	Gender	Age at assessment		Percentage of Consonants Correct (PCC)		
		Months	Yrs;mos	PCC	Severity level	SDCS <sup>a</sup>
<b>Younger subgroup</b>						
1	M	55	4;7	68.4	Mild-Moderate	QSD
2	M	55	4;7	69.5	Mild-Moderate	SD
3	F	56	4;8	71.9	Mild-Moderate	QSD
4	F	57	4;9	68.8	Mild-Moderate	SD
5	M	62	5;2	77.4	Mild-Moderate	NSA-/SD
6	F	64	5;4	52.5	Moderate-Severe	SD[+]
7 <sup>b</sup>	M	64	5;4	79.3	Mild-Moderate	SD
8	F	65	5;5	59.9	Moderate-Severe	SD[+]
9	M	67	5;7	42.8	Severe	SD[+]
10	M	67	5;7	62.2	Moderate-Severe	SD
11 <sup>c</sup>	F	78	6;6	85.7	Mild	NSA-/SD
Subtotal:	M	62.7	5;3	67.1		
	SD	6.9	0;7	12.3		
<b>Older subgroup</b>						
12	M	95	7;11	60.0	Moderate-Severe	SD
13	M	98	8;2	80.6	Mild-Moderate	NSA-/SD
14 <sup>b</sup>	M	101	8;5	78.3	Mild-Moderate	NSA-/SD
15	F	110	9;2	89.9	Mild	[RE-2]
16	F	126	10;6	72.7	Mild-Moderate	RE-2
17 <sup>c</sup>	F	139	11;7	96.6	Mild	[RE-1]
18	F	143	11;11	84.8	Mild-Moderate	[RE-2]
19	M	148	12;4	89.1	Mild	RE-1
20	M	162	13;6	88.2	Mild	[RE-2]
21	F	172	14;4	68.1	Mild-Moderate	RE-2
Subtotal:	M	129.4	10;9	80.8		
	SD	27.6	2;4	11.3		
Total:	M	94.5	7;10	73.6		
	SD	39.1	3;3	13.5		

<sup>a</sup>Speech Disorders Classification System (Shriberg, 1993). See text for discussion of classification entries.

<sup>b</sup>These data are from the same child at two points in time (see text).

<sup>c</sup>These data are from the same child at two points in time (see text).

(SD[+]), indicating that their conversational speech samples included uncommon clinical distortion errors in 10%-20% of words. Two children were classified as NSA-/SD, indicating intermediate status between speech delay and normal (here, normalized) speech. PCC scores for the older group ranged from Mild to Moderate-Severe, averaging approximately 13 percentage points higher than the PCC scores for the younger group (PCC:  $M = 80.8\%$ ,  $SD = 11.3\%$ ). One child in the older group was classified as SD using the SDCS system, and the remaining 2 children under age 9 were classified as NSA-/SD. Of the 7 remaining children 9 years of age and older, 2 were classified as having nonmarginal or marginal residual errors-1 (RE-1), which indicates that

they had only common clinical distortions on fricatives and/or liquids. The remaining 5 children were classified as nonmarginal or marginal RE-2, which indicates they had both common clinical distortions and omissions/deletions termed imprecise speech.

The repeated measures for Child 7/14 and Child 11/17 were included in Table 2 to illustrate an important observation about the course of normalization for children with suspected DAS. Child 7/14 made virtually no gains in his speech development during a 3-year period, as shown by his PCC at 5 years 4 months (79.3%) and again at 8 years 5 months (78.3%). For Child 11/17, however, his PCC of 85.7% at 6 years 6 months (and SDCS classification of NSA-/SD) probably represents progress

toward normalization of his earlier speech delay. When assessed again at 11 years 7 months, he has essentially normalized omission and deletion errors (i.e., PCC = 96.6%), but marginally retains common clinical distortion errors (i.e., [RE-1]). These longitudinal data confirm the alternative normalization histories that may occur in DAS, assuming that at least at one time either or both children were true positive DAS. Later discussion will focus on such questions relative to information on these children's prosody-voice status. To maintain independent degrees of freedom, the data for Child 14 and Child 17 are excluded from the older group in all of the following analyses.

## Results

### Speech

#### Speech Severity Measures

Table 3 provides descriptive and inferential statistics for five speech severity indices for children with suspected DAS in the younger and older groups and the SD comparison groups. For the younger group comparisons, children in the DAS group scored significantly lower than controls in the SD group on the Intelligibility Index ( $W = 222.5$ ,  $p < .01$ ). Scatter plots of scores of

children in the DAS and SD groups indicated overlapping values at all levels of the total and subdivided indices. Thus, as found in Study I, severity of speech involvement was not a discriminating diagnostic feature.

#### Speech Error Target and Error Type Analysis

Each of the error target and error type analyses described in Study I were also obtained separately for the younger and older subgroups of children with suspected DAS and their comparison groups of children with SD. These comparisons included analyses of the 15 natural phonological processes and analysis of error targets, error types, and error consistency for consonants and vowels by sound, developmental class, and features. Glide errors were of particular interest, relative to Study I findings. With the exception of one statistically significant finding in all these analyses, the error targets, error types, and error consistency of children with DAS did not differ from those of children of comparable age with speech delay. The one significant finding was for younger children with suspected DAS to have lowered error consistency on one of the class features, obstruents (DAS: 72.6%, SD: 83.9%). Although none of the other error consistency comparisons were significant, even when

**Table 3.** Severity of involvement of 19 younger and older children with suspected DAS and age-matched comparison groups of children with speech delay (SD).

Severity metric	Younger					Older				
	DAS (n = 11)		SD (n = 64)		W <sup>a</sup>	DAS (n = 8)		SD (n = 9)		W
	M	SD	M	SD		M	SD	M	SD	
<b>Percentage of Consonants Correct (PCC)</b>										
Singletons	69.8	12.8	66.3	8.5	517.5 ns	79.8	11.2	83.1	6.6	69.0 ns
Clusters	57.5	17.8	50.3	12.8	515.5 ns	76.5	12.0	71.6	11.2	82.0 ns
Total	67.1	12.3	62.7	8.2	517.5 ns	79.2	11.1	80.0	7.2	73.0 ns
<b>Percentage of Consonants Correct-Adjusted (PCC-A)</b>										
Singletons	71.3	11.8	71.7	9.7	422.0 ns	81.9	10.7	88.9	4.7	55.5 ns
Clusters	60.8	17.5	60.6	16.2	427.5 ns	80.3	13.5	80.7	9.9	73.0 ns
Total	69.1	11.2	69.3	10.2	408.0 ns	81.8	11.0	86.8	5.7	62.5 ns
<b>Percentage of Consonants Correct-Revised (PCC-R)</b>										
Singletons	72.4	11.2	74.5	9.2	380.5 ns	83.6	8.5	89.6	4.6	56.0 ns
Clusters	62.8	17.3	64.5	16.1	399.0 ns	80.4	13.4	81.8	9.0	71.5 ns
Total	70.3	10.7	72.4	9.8	372.0 ns	83.1	9.3	87.5	5.4	61.0 ns
<b>Percentage of Vowels Correct (PVC)</b>										
	92.8	5.2	91.4	3.6	504.5 ns	95.1	2.0	92.5	4.0	88.0 ns
<b>Intelligibility Index (II)</b>										
	82.1	12.5	91.7	7.9	222.5 †	91.0	8.9	97.5	2.1	55.5 ns

<sup>a</sup>Wilcoxon-Mann-Whitney (Siegal & Castellan, 1988)

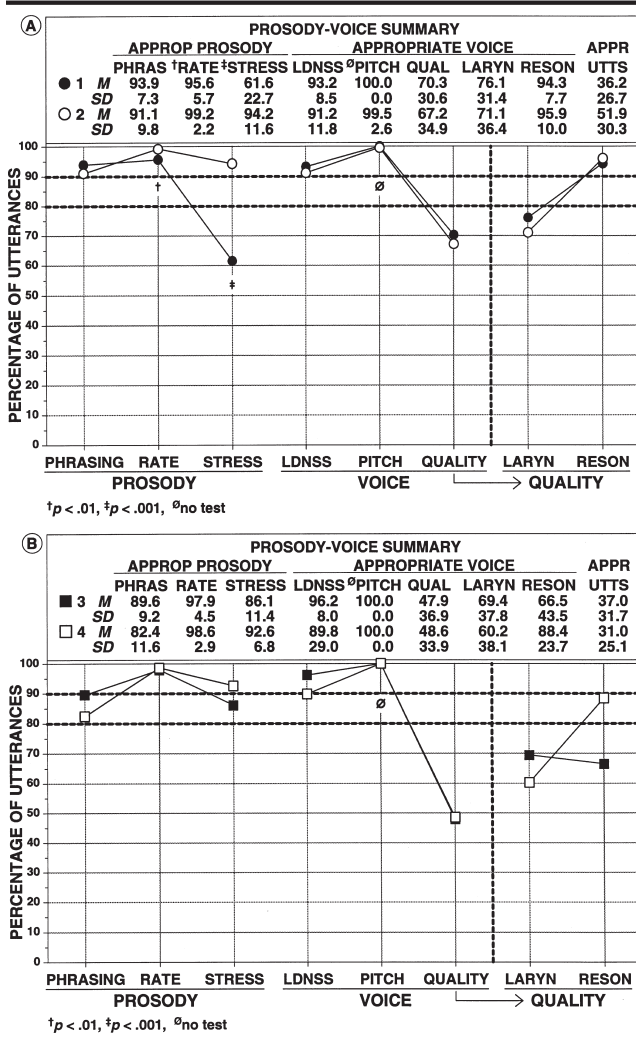
† $p < .01$

tested with parametric statistics with and without arcsin transformations, there was a notable trend for the younger children with suspected DAS to have lower error consistency scores than children with SD.

### Prosody-Voice

Figure 1 includes prosody-voice profile comparisons for the younger (Panel A) and older (Panel B) subgroups of children with suspected DAS and with SD. For the younger groups, children with suspected DAS had significantly lower average scores on two prosody-voice variables, rate (DAS: 95.6%, SD: 99.2%,  $p < .01$ ) and stress (DAS: 61.6%, SD: 94.2%,  $p < .001$ ). Examination

**Figure 1.** Prosody-Voice Profile comparison of Study III children with suspected developmental apraxia of speech (DAS) and children with speech delay (SD). Panel A data are for the younger children in each group (Groups 1 and 2, respectively) and Panel B data are for the older children in each group (Groups 3 and 4, respectively). Only the data indicating percentages of utterances with appropriate prosody-voice are shown (see text).



of the prosody-voice codes indicated that inappropriate rate was mostly associated with PV Code 9: Slow Articulation/Pause Time, and to a lesser extent, PV Code 10: Slow/Pause Time. Inappropriate stress was almost entirely associated with PV Code 15: Excessive/Equal/Misplaced Stress. As shown in Figure 1, Panel B, there were no statistically significant differences for any of the comparisons for older children with suspected DAS and with SD.

Group-level findings for Study III are interpreted as support for the conclusion from Study I and Study II that inappropriate stress may be a diagnostic marker for DAS. Additional analyses of Study III data are incorporated in the following combined examination of findings from the three studies.

## Combined Analyses of Studies I, II, and III

### Analyses of Children With Suspected DAS Who Have Appropriate and Inappropriate Stress

Table 4 is a summary of the stress findings from Studies I, II (Shriberg et al., 1997b) and Study III. A conservative approach was used to dichotomize stress into appropriate (80% or above, which includes questionable and appropriate) and inappropriate (below 80%). Cross-tabulations in Table 4 allow an examination of the percentages of children with appropriate and inappropriate stress by diagnostic group (suspected DAS, SD), age (younger, older), gender (male, female), and research site (Study I, Study II, and Study III: Researchers A-E). The following sections summarize findings for each variable.

### Analyses by Diagnostic Group

The intersect of the rows and columns in Table 4 titled *Combined* provides summative comparison of the percentage of children with suspected DAS and with SD who have inappropriate stress. Of the total of 48 children with suspected DAS whose stress could be assessed, 25 (52%) had inappropriate stress and 23 (48%) had appropriate (or questionable) stress. Moving to the right in Table 4, these data can be compared to the 71 children with speech delay whose stress was assessed by the same procedures and personnel. For this comparison group, 7 (10%) had inappropriate stress and 64 (90%) had appropriate or questionable stress. These differences in percentages were statistically significant ( $\chi^2 = 25.972$ ,  $df = 1$ ,  $p < .001$ ).

A useful way to characterize these differences for both theoretical and clinical considerations is to consider the sensitivity and specificity of the stress assessment

**Table 4.** Summary of stress findings in Studies I, II, and III, and the comparison groups of speech-delayed children.

Group	Age/Gender	Children with suspected DAS <sup>a</sup>																					
		Study I			Study II			Study III			Combined			Children with Speech Delay <sup>a</sup>									
		Inapprop.	Approp.		Inapprop.	Approp.		Inapprop.	Approp.		Inapprop.	Approp.		Inapprop.	Approp.								
n	%	n %	n	%	n %	n	%	n %	n	%	n %	n	%	n	%	n %							
Younger	M	1	16.7	5	83.3	5	55.6	4	44.4	3	75.0	1	25.0	9	47.4	10	52.6	4	10.3	35	89.7		
	F	—	—	—	—	3	100.0	0	0.0	3	75.0	1	25.0	6	85.7	1	14.3	3	13.0	20	87.0		
	All	1	16.7	5	83.3	8	66.7	4	33.3	6	75.0	2	25.0	15	57.7	11	42.3	7	11.3	55	88.7		
Older	M	4	66.7	2	33.3	3	60.0	2	40.0	1	25.0	3	75.0	8	53.3	7	46.7	0	0.0	8	100.0		
	F	1	100.0	0	0.0	0	0.0	2	100.0	1	25.0	3	75.0	2	28.6	5	71.4	0	0.0	1	100.0		
	All	5	71.4	2	28.6	3	42.9	4	57.1	2	25.0	6	75.0	10	45.5	12	54.5	0	0.0	9	100.0		
Combined	M	5	41.7	7	58.3	8	57.1	6	42.9	4	50.0	4	50.0	17	50.0	17	50.0	4	8.5	43	91.5		
	F	1	100.0	0	0.0	3	60.0	2	40.0	2	40.0	4	50.0	4	50.0	8	57.1	6	42.9	3	12.5	21	87.5
	All	6	46.2	7	53.8	11	57.9	8	42.1	8	50.0	8	50.0	8	50.0	25	52.1	23	47.9	7	9.9	64	90.1
<b>Clinical research site</b>																							
	Study I	6	46.2	7	53.8																		
	Study II	11	57.9	8	42.1																		
	Study III	A	2	66.7	1	33.3																	
		B	2	40.0	3	60.0																	
	C	1	50.0	1	50.0																		
	D	2	50.0	2	50.0																		
	E	1	50.0	1	50.0																		

<sup>a</sup>Stress status could not be judged for one sample from the younger group in Study I, one sample from the older group in Study II, three samples from the younger group in Study III, and two samples from the younger comparison group of Speech-Delayed children.

procedure. If inappropriate stress is, in fact, a valid diagnostic marker for DAS, the sensitivity of the stress measure as a “test” of DAS was only approximately 52%. That is, if all 48 children with suspected DAS truly had DAS, only 52% were detected. Alternatively, if there are subtypes of DAS, a figure of 52% might be the estimated prevalence of this subtype of DAS within children with suspected DAS. A third possibility is that the remaining approximately 48% of children with suspected DAS are false positives for DAS, suspected to have DAS for reasons described in the prior review and the local ascertainment study (Shriberg et al., 1997a, Table 1).

In comparison with alternative interpretations of the sensitivity data, interpretation of the specificity data is straightforward. The comparison groups of children with speech delay are not suspected to have DAS, and the data in Table 4 indicate that 90% of children with speech delay had appropriate stress. Specificity values of 90% and above are considered adequate for diagnostic testing in the medical sciences, with a typical goal to achieve 90%–95% for both sensitivity and specificity. Considering possible improvements in the assessment of stress, as discussed in a later section, the finding of above 90% specificity on the current stress task is considered major support for theoretical and clinical perspectives on stress as a marker for at least one form of DAS. Subsequent discussion addresses this hypothesis.

### Analyses by Age and Gender

A second data element in Table 4 is the relationship of age and gender to inappropriate stress within children with suspected DAS. Considering the convergence in the per-study data above and cell size needs for statistical analysis, the combined data were used for  $\chi^2$  analysis. The statistical findings were mixed and nonsignificant. For younger children, 9 of the 19 (47%) males and 6 of the 7 (86%) females had inappropriate stress, a descriptively suggestive, but statistically nonsignificant difference in proportions ( $\chi^2 = 3.082$ ,  $df = 1$ ,  $p < .079$ ). For older children, 8 of the 15 males (53%) and 2 of the 7 females (29%) had inappropriate stress, also a nonsignificant difference in proportions ( $\chi^2 = 1.18$ ,  $df = 1$ ,  $p < .278$ ). Collapsing across these nonsignificant gender findings, 15 of the 26 (58%) younger children with suspected DAS had inappropriate stress, compared to 10 of the 22 (46%) of the older children with suspected DAS ( $\chi^2 = .715$ ,  $df = 1$ ,  $p < .398$ ). Thus, although trends in some subgroups suggested age and gender differences for inappropriate stress, trends were mixed and none of the comparisons were statistically significant at the conventional .05 alpha level.

### Analyses by Clinical-Research Site

The final variables to examine in Table 4 are the per-site percentages of children with inappropriate

stress, which, as allowed by findings above, are collapsed over age and gender. As shown in the bottom rows of Table 4, the percentage of children with suspected DAS who had inappropriate stress was approximately 46% for the site in Study I, 58% for the site in Study II, and, for the five research sites within Study III labeled A–E, respectively, 67%, 40%, 50%, 50%, and 50%. The distributional statistics for these seven estimates of per-site percentages (using all original values) are:  $M$ : 51.6%,  $SE$ : 3.2%,  $Mdn$ : 50%, and  $SD$ : 8.6%. Thus, seven independent estimates of inappropriate stress in children with suspected DAS converge on a median/mean of approximately 50%–52%, with the standard error of the mean and the 8.6% standard deviation suggesting that most estimates are within the boundary of approximately 40% to 60%. An inference from these data, and possible generalization to studies using similar ascertainment methods, is that approximately 4–6 children of every 10 referred with suspected DAS may have inappropriate stress.

### Speech Status of Children With Suspected DAS Who Have Appropriate and Inappropriate Stress

The final analysis series provides close examination of the speech of the younger and older children with suspected DAS with inappropriate stress.

### Error Types and Error Targets

Speech profiles similar to those shown in prior figures were obtained comparing the children with suspected DAS and inappropriate stress to the children with suspected DAS and appropriate stress and to their age-matched controls. Separate analyses were obtained using the speech profiles comparing consonants, consonant features, vowels, phonological processes, and the several profiles comparing error types at the level of diacritics. Other than a few statistically significant differences in the hundreds of comparisons, which were well within chance probability, there were no patterns of speech errors or error targets that differentiated the children with inappropriate stress from the other children with suspected DAS or from the comparison children with SD. As found in Study I, the few statistically significant differences in speech error type were associated with severity. The more severely involved younger children with suspected DAS and inappropriate stress had more omission errors than younger children with SD. To estimate whether the lack of significant findings was associated with the power of the nonparametric statistic, profiles were rerun using parametric  $t$  tests, both with and without arcsin transformations. These analyses yielded several more statistically significant



differences, but the findings were again unpatterned relative to the direction of findings and convergence within domains. Thus, the speech characteristics of children with inappropriate stress were not different in error targets or error types from other children with suspected DAS and appropriate stress or from children with SD.

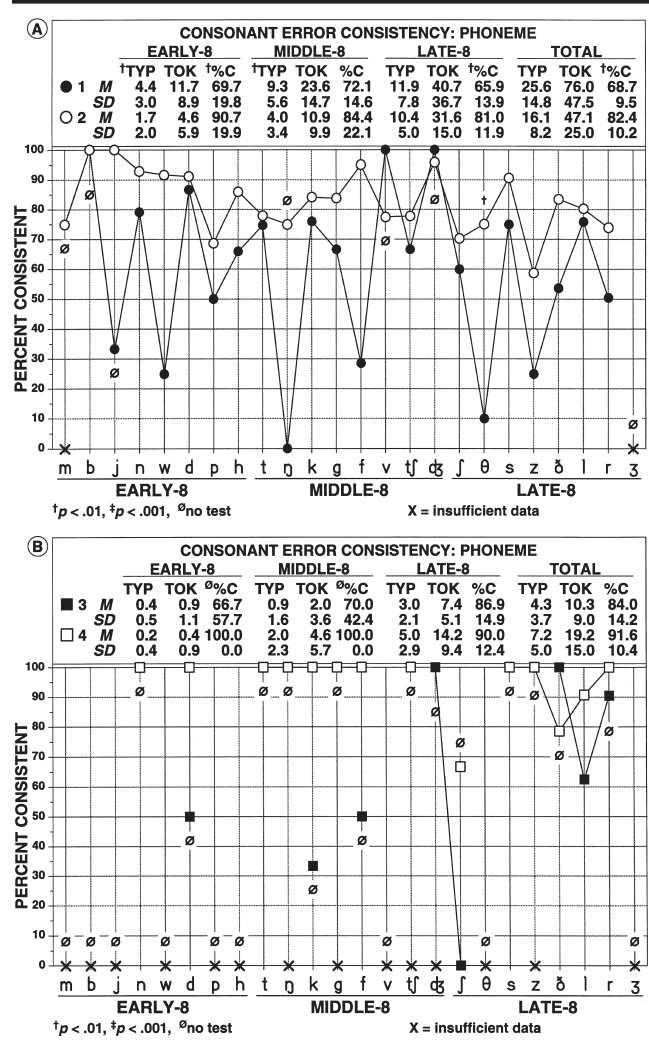
**Error Consistency**

The final analysis series assessed the error consistency status of children with suspected DAS who had inappropriate stress. Consistency of consonant and vowel errors for children with inappropriate and appropriate stress was assessed using the same procedure described for the data in Figure 5 of Shriberg et al. (1997b). Cell sizes and distributions met assumptions for *t* tests, which were performed with and without arcsin transformations of the error consistency percentages.

Figure 2 provides consonant error consistency data for the younger (Panel A) and older (Panel B) comparisons. Group 1 in Panel A includes the younger children with suspected DAS and inappropriate stress; Group 2 includes the younger control children with SD. Within Panel B, the Group 3 children are the older children with suspected DAS and inappropriate stress; Group 4 children are the older control children with SD. As indicated in the numerical panel of Panel A, there were two significant differences in the average number of word types used to calculate the error consistency percentages. For both Early-8 and Late-8 sounds, significantly more word types were used in the consistency calculations for Group 1 children compared to the average number of types for Group 2. As before, these significant differences are viewed as constraints on interpretation of any obtained differences in the error percentages calculated for each group.

Beginning with Panel A, younger children with suspected DAS and inappropriate stress had significantly lower error consistency percentages than younger SD children for the Early-8 sounds (Group 1: 69.7%, Group 2: 90.7%, *p* < .01), Late-8 sounds (Group 1: 65.9%, Group 2: 81.0%, *p* < .01), and total sounds (Group 1: 68.7%, Group 2: 82.4%, *p* < .01). There was also one statistically significant comparison at the consonant sound level: Group 1 children had significantly lower error consistency scores than Group 2 children on /θ/ (Group 1: 10.0%, Group 2: 75.0%, *p* < .01). Additional consistency analyses (not shown here) indicated that, in comparison to younger children with SD, children with suspected DAS and inappropriate stress also had significantly lower error consistency on obstruents (Group 1: 68.8%, Group 2: 84.1%, *p* < .01), voiceless consonants (Group 1: 67.5%, Group 2: 83.3%, *p* < .01), and the rhotic vowels (Group 1: 8.3%, Group 2: 65.0%, *p* < .01). There were no

**Figure 2.** Consonant error consistency comparison for children from Studies I and III. In Panel A, Group 1 includes younger children with suspected DAS and inappropriate stress; Group 2 includes the younger control children with SD. In Panel B, Group 3 includes the older children with suspected DAS and inappropriate stress; Group 4 includes the older control children with SD.



statistically significant differences on any of the error consistency analyses for comparisons involving the older children with suspected DAS and inappropriate stress and older children with SD.

**Discussion**

To address the complexity of issues relevant to the two questions posed at the outset of this research—diagnostic markers and the possibility that DAS may be genetically transmitted—discussion is structured in the form of five hypotheses about inappropriate stress and DAS.

## Five Hypotheses About Inappropriate Stress and DAS

### Hypothesis I: Inappropriate Stress Is a Diagnostic Marker for a Subtype of DAS

The first hypothesis addresses the primary question in this research. Pending additional cross-validation of the type discussed later in this paper, the hypothesis reflects a conclusion about three possible explanations for the stress findings.

*Interpretation 1: Inappropriate stress is a compensatory behavior in some children with DAS.* As reviewed in the prior paper in this series, stress differences similar to those found in this research have been reported in both adult and child apraxia literatures, with frequent explanatory appeal to the construct of *compensatory behavior* (e.g., Aram & Glasson, 1979; Chappell, 1984; Colson, 1988; Glasson, 1979; Ingram & Reid, 1956; Murdoch, Porter, Younger, & Ozanne, 1984; Odell, McNeil, Rosenbek, & Hunter, 1990; Rosenbek & Wertz, 1972; Shuster, Ruscello, & Haines, 1989; Velleman & Strand, 1994; Wertz, LaPointe, & Rosenbek, 1984; Yoss & Darley, 1974a). For example, Yoss and Darley (1974b) proposed that “durational lengthening, monotony of stress, and lack of speech sound blending can be explained as attempts to compensate for severe speech production problems” (p. 348). Velleman and Strand (1994) suggest that intermittent pauses between syllables, words, and phrases may “‘buy time’ for the organization and initiation of upcoming movement plans” (p. 126). Kent and McNeil (1987) noted that “apraxia of speech has a conspicuous dysprosody, although it is not clear if the dysprosody is an independent feature of the disorder or simply a consequence of a primary articulatory impairment” (pp. 214–215). Marquardt and Sussman (1991) also questioned whether prosodic disturbances are “an intrinsic part of the disorder or a compensatory strategy employed in response to motor programming problems” (p. 346).

Thus, one explanation for the source of the stress deficits in approximately 52% of the children with suspected DAS is that stress deficits are secondary to some primary deficit, perhaps in the service of intelligibility. This first interpretation of the stress findings would claim that inappropriate stress is an optional characteristic of DAS, and thus is neither a necessary nor a sufficient diagnostic marker for DAS or for a subtype of DAS. Three findings in the current data and anecdotal information from colleagues provide counter evidence for the “compensatory strategy” explanation of inappropriate stress.

First, if excessive-equal stress is a learned behavior to enhance intelligibility or perhaps acceptability—or to compensate for some deficit in selection-retrieval or

prearticulatory sequencing—a likely correlate for at least some of the children with stress deficits would be reduction in speech rate. As reviewed earlier, the criterion range used to code normal articulation rate in the prosody-voice instrument is 2–4 syllables per second for 3- to 7-year-old children and 4–6 syllables per second for older children. With the exception of the severely reduced rate for one child with suspected DAS in Study I, whose speech was considered dysarthric, the two codes for reduced rate (PV Code 9: Slow Articulation/Pause Time and PV Code 10: Slow/Pause Time) were infrequently used for children with suspected DAS in Study I and Study III. These findings are counter to literature reports of an impression of reduced rate in children with suspected DAS.

A second finding interpreted as counter evidence for the compensatory strategy interpretation is that inappropriate stress was unrelated to age. Specifically, 57.7% of the younger children with suspected DAS had inappropriate stress, compared to 45.5% in the older children with suspected DAS (see Table 4). If stress deficits were due to compensatory strategies, the more likely distribution of ranges would favor older children where one might expect to see the most evidence of long-term adoption of a compensatory pattern.

A third finding viewed as inconsistent with the compensatory behavior explanation is that inappropriate stress was observed in brief and simple utterances as well as long and complex utterances. For example, inappropriate stress was coded in the following seven examples from 5 different children: “Go fishing,” “And door,” “We go swimming,” “Spot on puppies?” “He only two,” “Put it on yellow,” and “It’s like a snake.” These utterances seem too short to require stress adjustments in the service of intelligibility. Moreover, inappropriate lexical stress (PV13: Multisyllabic Word Stress) has been observed in single-word utterances of children with suspected DAS. Two examples are a child who said “SidNEY” instead of SIDney and a child who said “sisTER” instead of SISTER. Such lexical stress differences also cannot readily be explained by an appeal to enhanced intelligibility.

A fourth observation appeals to anecdotal information on the course of normalization of children with suspected DAS. P. Hall (personal communication, 1995), who has extensive clinical-research experience with children with suspected DAS, reports that “a stress deficit in children with DAS...seems to be a continuing element regardless of how much improvement in sound production the children achieve.” If the stress changes are compensatory in the service of intelligibility or acceptability, additional mechanisms would have to be invoked to explain their persistence after the behaviors they compensate for have normalized.

*Interpretation 2: Inappropriate stress is the necessary and sufficient diagnostic marker for DAS.* An alternative interpretation of findings in the three studies is that inappropriate stress is a primary deficit in DAS, and more significantly, is the “necessary and sufficient” (cf. Deputy, 1984; Guyette & Diedrich, 1981; Love, 1992) diagnostic marker. This claim posits that DAS is a unitary entity, although the stress deficits and any other behaviors may vary in severity of expression. Formally, no one behavior can be a sufficient diagnostic marker of DAS because of the need for other inclusionary and exclusionary criteria. By definition, children with suspected DAS must have a speech disorder with its onset during the developmental period and other gross involvements must be excluded. Also, other obvious explanations for inappropriate stress (e.g., dialectal differences, dysarthria, autism) must be ruled out. However, assuming these inclusionary/exclusionary criteria for the sufficiency claim have been met, there also remains a problem with the concept of one necessary and sufficient deficit applied to the current findings. If there is only one form of DAS requiring stress deficits as the necessary and sufficient diagnostic feature, then what is the explanation for the approximately 48% of children with suspected DAS in the three studies who did *not* have stress deficits? Are they children who do not have DAS? Three possible explanations in support of this second hypothesis—that stress is a necessary and sufficient marker for DAS—are as follows.

First, as reviewed above, the failure to find inappropriate stress in all of the children with suspected DAS could reflect false negatives due to measurement error. This measurement explanation would claim that the sensitivity (i.e., ability to detect true positives) of the stress measure was only 52%, which is not consistent with the validity data reported for this measure (cf. Shriberg, Kwiatkowski, & Rasmussen, 1990; Shriberg, Kwiatkowski, Rasmussen, Lof, & Miller, 1992). Although some of the children with suspected DAS who tested normal on stress may have been borderline false negatives, it is unlikely that such a high percentage of the total number of children with suspected DAS (48%) actually have DAS but falsely tested negative.

A second possible explanation for the lack of inappropriate stress in approximately 48% of children with suspected DAS considers their relatively older age. Perhaps a significant number of children with suspected DAS had normalized stress at the time of assessment, whereas they did have measurable stress deficits at some earlier period of development. This explanation would be especially attractive if inappropriate stress had been observed primarily in the younger children with suspected DAS. However, because this was not the consistent finding in Study I and Study III, an appeal to early

normalization of stress deficits is not considered a likely explanation for the children with suspected DAS who had appropriate stress.

A third explanation for the finding of stress deficits in only some children with suspected DAS is that the remaining children with normal stress truly do not warrant the classificatory term DAS. This position suggests that a false positive rate for suspected DAS of approximately 50% (53.8% in Study I and 50% in Study III) may be an appropriate and generalizable estimate of referral and ascertainment statistics. As reported previously in the Local Ascertainment Study (cf. Shriberg et al., 1997a, Table 1), DAS is suspected when younger or especially older subjects with more severe speech involvement do not make typical progress in treatment.

Although any or all of these three considerations supporting the second interpretation may be compelling, parsimony suggests that it is more prudent to reject this interpretation of the stress findings in favor of a third interpretation discussed below—until more well-developed research (see Research Issues) can provide a better basis for deliberating these three interpretations.

*Interpretation 3: Inappropriate stress is a diagnostic marker for one subtype of DAS.* The claim here assumes that children with suspected DAS have a speech disorder in the developmental period and that other obvious explanations for inappropriate stress have been ruled out. Assuming these inclusionary/exclusionary criteria are met, a third explanation for the positive stress findings in approximately 52% of the children with suspected DAS is that there may be more than one form of DAS. This perspective posits inappropriate stress as a diagnostic marker for one of at least two subtypes of DAS, with the other subtype(s) requiring some other diagnostic marker(s). The attractive feature of this interim proposal is that it addresses the classificatory status of children with suspected DAS without stress deficits. As above, some to all of these subjects may have been false negatives for DAS due to lack of sufficient sensitivity in the measurement of stress. Alternatively, as posited by this third interpretation, their classification could remain suspected DAS, but not the subtype marked by inappropriate stress.

It is important to consider counterarguments for this view. If the remaining children with suspected DAS are valid candidates for one or more subtypes of DAS, what potential diagnostic criteria for subtyping remain? There were no significant speech findings distinguishing any of the children with suspected DAS—both with and without inappropriate stress—from children with speech delay. The strongest area of negative findings were the phrasing and error-consistency data, which failed to differentiate children with suspected DAS from their age-matched controls. That is, compared to children with

SD, children with suspected DAS did not have more frequent utterances coded as revisions or repetitions, nor did they have significantly lower error consistency percentages. A subtype claim would require a finding of significant phrasing or error-consistency differences in at least one of the three studies. No significant differences were obtained, and trends for the phrasing means were actually in the opposite direction.

A potential DAS subtype based on performance deficits on nonspeech tasks is also problematic. First, many perspectives argue that a speech apraxia must be documented by history or performance on a speech, rather than nonspeech, variable (Love, 1992). Second, although many of the subjects in the present study had performance deficits on nonspeech and oral volitional tasks, not all did, including those with the most severe speech deficits (cf. Table 1).

*Summary.* The present hypothesis of inappropriate stress as a diagnostic marker for a subtype of DAS reflects a conservative position pending future research that may document a speech or nonspeech marker for at least one other subtype. This view does not exclude the likely situation that DAS co-occurs with other disorders affecting cognitive-linguistic and motor-speech areas, such as dysarthria and the many syndromes in which apraxic-like disorders have been attested. For example, although one of the children in Study I was excluded from DAS because her extremely slow rate was more consistent with dysarthria, several children with inappropriate stress also had infrequent vocal tremors suggesting other motor-speech involvements.

## **Hypothesis II: The Proximal Origin of Inappropriate Stress in This Subtype of DAS Is Deficits in Phonological Representational Processes**

Five considerations support the hypothesis that the loci of inappropriate stress in this proposed subtype of DAS are in phonological representational processes. The first three considerations are based on evidence from the three studies, and the last two reflect diverse theoretical perspectives and empirical information about stress and DAS.

*Inference from self-monitoring data.* Self-monitoring of speech output is a central concept in models of speech acquisition and performance (e.g., Levelt, 1989). In speech disorders, self-monitoring concepts are most dramatically evident in stuttering, where speakers are acutely aware of differences between intended phonological forms and those realized as disfluencies. In the present data, if the observed inappropriate stress reflected deficits at motor programming (i.e., prearticulatory sequencing or articulatory execution) phases, rather

than at prior planning stages of linguistic organization of stress, children with suspected DAS would have been expected to evidence some behaviors consistent with self-monitoring. Specifically, they would try in successive efforts to match their output to their planned stress targets. The term *groping* (searching for the intended articulatory postures) is used to describe such prearticulatory behaviors in adults with AOS, and terms such as *sound*, *syllable*, and *word revisions* are used to describe post-articulatory self-monitoring, self-correcting behaviors.

There was a clear lack of support in the present data for behaviors indicating self-monitoring of speech (groping or revisions) at segmental or suprasegmental levels: (a) The anecdotal transcription data (Study I) does not contain evidence of groping, although such behaviors were expected to occur, (b) the subject information for Study III does not include groping for articulatory postures or silent posturing for a majority of the children with suspected DAS (see Table 1), and (c) the phrasing data for all three studies does not indicate that children with suspected DAS had more sound, syllable, or part-word repetitions than children with SD. Therefore, rather than implicating motor control deficits, the stress deficits observed in these studies are more consistent with deficits in underlying representations. Specifically, a child's lack of effort to self-correct inappropriate stress could be explained by lack of awareness that correction was needed—which would be the case if the stress deficit were at the level of representational planning processes rather than motor-speech programming processes.

*Inference from rate data.* A second observation concerns the negative findings for slowed speech rate, which, as indicated above, was not observed for children with inappropriate stress. In the acquired AOS literature, as reviewed previously, slow articulatory rates are a primary characteristic of AOS. Presumably, they reflect selection-retrieval and/or prearticulatory sequencing deficits (cf. Shriberg et al., 1997a, Figure 1) that are expressly not due to end-stage, articulatory execution deficits. In the present research, slow articulatory rates were not observed in the children with suspected DAS in the three studies. On the contrary, some of the older children had somewhat faster rates, with anecdotal comments indicating that the speech rates resembled the rapid rushes of speech described in cluttering. Interestingly, persons with cluttering profiles are also reported to lack self-awareness of speech errors (cf. Daly, 1986; Diedrich, 1984). Especially given the severity of involvement of these children, particularly the younger group in Study I, deficits posited in selection-retrieval and/or prearticulatory sequencing would be expected to be associated with reduced rate of speech. Lacking such association, the loci for inappropriate stress presumably are at one or more prior stages of speech processing.

*Inference from lexical stress data.* A third observation suggesting an association of inappropriate stress with representational processes concerns the lexical stress errors made by some children with inappropriate stress. As described previously, the transcription notes for Study I indicate that some children made word-level stress errors such as “SidNEY” and “sisTER.” Such errors have been observed only in children with suspected DAS in our clinical-research experience with hundreds of children with speech disorders of unknown origin. Lacking controlled data from well-developed protocols, the prevalence of such errors in children with suspected DAS compared to the prevalence in other types of childhood speech disorders (e.g., SD) cannot be estimated. However, if reliable stress errors on only certain lexical items can be documented in children with suspected DAS (i.e., stress errors on repeated tokens of only certain word types), the loci would specifically implicate representational processes, rather than selection-retrieval or prearticulatory sequencing deficits at later stages of speech production.

*Inferences from theoretical perspectives and ontogenetic data.* A fourth and primary rationale for locating stress deficits at the stage of underlying representations is based on theoretical accounts of stress in contemporary linguistic theories. At a time when connectionist models continue to challenge alternative theoretical accounts of speech-language organization, acquisition, and performance (cf. Goldsmith, 1993; Markey, 1995; Plunkett, 1995; see Gupta & Touretzky, 1994, for an explicit demonstration of the acquisition of 19 natural language stress systems by a perceptron), syllable stress plays an increasingly central role in linguistic theory. Diverse perspectives underscoring the central role of the syllable can be found in descriptive linguistics (cf. Goldsmith, 1995; Kaye, 1989; Selkirk, 1984), psycholinguistics (e.g., Fear, Cutler, & Butterfield, 1995; Ferreira, 1993; Gee & Grosjean, 1983), developmental linguistics (e.g., Gerken, 1991; Locke, 1983, 1993, 1995; Mandel, Jusczyk, & Kemler Nelson, 1994; Schwartz & Goffman, 1995; Vihman, 1996; Wijnen, Krikhaar, & Den Os, 1994), and clinical linguistics (e.g., Bernhardt & Stoel-Gammon, 1994; Bock & Loebell, 1990; Piggot & Kessler-Robb, 1994; Spencer, 1984; Weinert, 1992). Although differing in formalisms and substantive content, common to the accounts in these and other sources are hierarchical organizations that position stress assignment at the “highest” tier relating prosodic, syntactic, and segmental elements. For example, Ferreira (1993) presents psycholinguistic evidence for a model of sentence processing in which “a prosodic structure is created from a sentence’s syntactic structure but without knowledge of its phonemic content” (p. 234). The ontogenetic primacy of suprasegmental relative to segmental behaviors is well documented empirically and in several theoretical views

of normal speech acquisition (Edwards & Shriberg, 1983; Locke, 1983, 1993, 1995; Menn & Stoel-Gammon, 1995). Wijnen et al.’s (1994) paper relating children’s omission of unstressed closed class morphemes and unstressed syllables within words to a developmental rhythmical constraint—not a perceptual constraint—offers compelling theoretical discussion.

Closer to a clinical perspective, the model of Kent and McNeil (1987) cited in the first paper in this series (Shriberg et al., 1997a) also posits that stress assignment occurs at a level above motor speech programming. Although these authors claim that a “motor speech programmer” is the loci of the problem in acquired AOS, their model asserts that stress assignment is represented in the instructions sent for prearticulatory processing by this programmer. Revisiting the relevant section of the quote from Kent and McNeil:

We believe that, at the least, the prearticulatory representation contains information on syllable structure and segment composition. Because these two bodies of information are held separately, they are susceptible to separate loss or error. Furthermore, the syllabic and segmental specifications only gradually lose their separateness in motor control. Syllabic organization is a primary level of cohesion in which, (1) suprasegmental information is given form in the prosodic envelope of a syllabic sequence; and (2) segmental information is converted to movements (preferably compound trajectories defined by compatible sequential goals [Shaffer, 1982]). (p. 213)

Although not appealing to any one theoretical framework, the claim in the present work is that lexical and phrasal stress are reflected in the “prosodic envelope of a syllabic sequence” and that this information is organized at representational levels prior to segmental organization and prearticulatory sequencing. In the present context, when a child says a stressed vowel instead of an unstressed (schwa or lax) vowel—typical of the stress pattern identified in the present study—the change in vowel is not viewed as a deficit in selection-retrieval or prearticulatory sequencing processes. Rather, a nonlinear perspective on representation of the underlying syllable claims that the stressed vowel was selected to accord with prior stress assignment. Thus, the stress deficits of some children with suspected DAS are presumed to occur at representational levels, which are taken as reflecting “linguistic planning” rather than “motoric programming” processes.

It is important here to acknowledge the many elaborated models of speech processing and speech motor control, in comparison to the simplified framework in Figure 1 in the first paper. Kent, Adams, and Turner (1996)

provide recent detailed review of the most widely cited models, including schemas that posit representational levels for motor control processes. Dodd's (1995) recent clinically oriented model of the speech-processing chain also includes additional processing levels for phonological plans, stored routines, and motor speech programming. Whichever model may have appeal as the most highly valued, the central point here is that representations that serve only to buffer prearticulatory commands are not the representations appealed to as the loci of inappropriately marked stress. The several reasons for this claim are reviewed above. Again, among the considerations offered, strong support is the lack of groping and attempts at self-correction observed in the samples of children with suspected DAS in this study, compared to the pronounced groping and revisions that define the speech profile of adults with AOS. A compelling explanation for such differences is inherent in the linguistic assumptions made for developmental versus acquired speech-language disorders. In the latter, linguistic representations presumably remain intact, with a lesion affecting only the motor control representations and/or commands. For developmental speech problems, however, there is good support for the perspective that linguistic representations themselves may be inappropriate or underspecified, including underspecification of stress marking. As above, the deviant stress and lack of awareness reported for children with the clinical entity termed cluttering appeals to similar explanatory rationale.

*Compatibility with other features of DAS.* A final rationale for locating stress deficits within representational processes is the ability of such modeling to motivate the primary features and other deficits observed in children with suspected DAS. Late onset of speech, severe speech delay, long-term normalization, and associated language involvements have been described as the primary features of children with suspected DAS. Aram and Glasson (1979) report that many children with suspected DAS are nearly nonverbal until age 3 or 4. In comparison, children classified as late talkers (e.g., Paul, 1993) or speech delayed (e.g., Shriberg & Kwiatkowski, 1994) may have impoverished phonetic inventories, vocabulary size, and morphosyntactic development, but they talk. Moreover, at least in children with speech delay, severity of speech involvement does not appear to be a strong predictor of short-term versus long-term normalization (Shriberg, Kwiatkowski, & Gruber, 1994), whereas late onset of speech is associated with long-term normalization in case studies of children with suspected DAS (cf. Hall, Jordan, & Robin, 1993).

Contemporary clinical linguistic perspectives view the segmental deletions and substitutions observed in late talking and speech-delayed children as evidence of delays in both representational-level processes and

selection-retrieval processes. Placing the loci of inappropriate suprasegmental behaviors (i.e., the correlates of stress) at underlying representational levels for some children with suspected DAS provides a plausible explanatory source for the features that differentiate this form of DAS from late talkers or speech delay. Because prosodic development is fundamental to segmental development, receptive and/or productive stress deficits in underlying representations would provide a sufficient explanation for early, severe, and persistent phonological delay. An analogy would be to children who have a developmental form of aphasia, which results in a severe linguistic delay due to deficits in the ability to form appropriate underlying phonological representations. Consider how the following three sets of deficits might follow from stress or metrical-level deficits at a representational level of phonology.

First, a prelexical deficit in the ability to format stress assignment for syllables (cf. Bastiaanse, Gilbers, & van der Linde, 1994) could explain deficits on diadochokinetic tasks, multisyllabic word tasks, and other speech and oral volitional sequencing tasks used to assess children with suspected DAS. On-demand repetitions of even simple sequences such as "pa-ta-ka," which reportedly are difficult for many children with suspected DAS (cf. Hall et al., 1993), require a metrical constant for the rapidly changing articulatory movements. Thus, a crucial difference between sounds produced in isolation versus those produced in multisyllabic contexts is that the latter require reliable stress assignment for each syllable. (For some relevant cross-linguistic examples of interactions among prosody, speech, and language, see Matthews, 1994, particularly the discussion of *rendaku* in Japanese phonology [Fukuda & Fukuda, 1994]).

Second, placing the loci of stress deficits at a stage of processing prior to selection-retrieval and prearticulatory sequencing might also explain the more general language and learning problems that have been reported for children with suspected DAS. The assumption is that there is both a receptive and expressive aspect of the stress deficit, as specifically suggested in the studies of rhyming deficits in children with suspected DAS (Marion, Sussman, & Marquardt, 1993). Consider Chiat and Hirson's (1987) speculations about the centrality of rhythmic structures in a case study of a child with developmental aphasia:

Is output alone constrained in the ways observed, or is input subject to the same constraints? It could be that [the child's] processing of input matches the pattern observed in output, i.e., that there are limitations on her recognition and comprehension of phonological detail within a rhythmic structure, and that unstressed items preceding stress are especially vulnerable...so that [such

children] have precluded access to certain aspects of adult language. If [the child] has never been able to hold certain phonological details within a phonological structure, she will not have been able to identify certain phonological items (e.g., certain function words) and their syntactic-semantic role. (p. 54)

Third, the success of stress-based management procedures in children with suspected DAS might also be invoked as support for stress deficits at underlying levels. Guyette and Diedrich (1981) note that response to treatment can be a source of support for a proposed diagnostic category. Most of the major approaches to intervention involve direct or indirect work on prosody, specifically on stress assignment (see later section on Treatment Considerations). Although such programs may be thought to provide a form of motor-skills learning, they actually may share the common and crucial effect of helping children to develop appropriate and accessible stress forms.

### **Hypothesis III: The Proximal Origin of Inappropriate Stress in This Form of DAS is Likely a Deficit Affecting Neurologic Functioning**

Speculations about neurological substrates based solely on the present findings are clearly preliminary. However, for the purposes of the second goal of this study concerning the possibility of genetic transmission of DAS, it is appropriate to address briefly the etiological implications of Hypotheses I and II. Hypothesis II claims that the proposed subtype of DAS based on stress deficits has neurogenic origins.

Marquardt and Sussman (1991) posit two possible types of neurologic substrates for DAS: diffuse or focal brain damage or “a disturbance in normal neurological maturation—perhaps specific to cortical areas responsible for speech and language functions” (p. 343). Based on negative literature findings for diffuse or focal brain damage and results from their rhyming tasks studies, these authors conclude that DAS is associated with neurological immaturity. Findings from the present study, however, seem more consistent with the notion of a specific neurological delay or insult, rather than neurological immaturity. Support for the hypothesis of a specific neurogenic etiology, rather than a functional (i.e., learned or compensatory) origin of this form of DAS or a general neuromaturation delay is based on three observations.

First, stress differences of the type observed in over 50% of the children with suspected DAS represent a *qualitative* departure from normal speech acquisition, as well as from clinical profiles of children with developmental phonological disorders (cf. Shriberg &

Kwiatkowski, 1994). Unlike continuous variables reflecting speech-language development (e.g., Percentage of Consonants Correct), stress is treated as a nominal variable (appropriate, inappropriate) in the perceptual screening procedures used in this study because it is not distributed continuously in normally speaking and speech-delayed children. In a technical report on the prosody-voice screening procedure, 95.7% of 115 speech-normal children (3–18 years of age,  $M = 5$  years 5 months,  $SD = 2$  years 11 months) had appropriate stress, as defined specifically by their status on Prosody-Voice Code 15: Excessive/Equal Stress (Shriberg et al., 1992, Table 11). In the current study, inappropriate stress of any type occurred in only 9.9% of control children with speech delays. In the absence of some readily explainable environmental source of learning (e.g., an idiolect patterned after a caregiver’s model of equal-excessive stress) it is more parsimonious to assume that some intrinsic deficit accounts for this qualitative disorder in phrasal stress observed in 52% of the 48 speech samples from children with suspected DAS.

A second consideration consistent with a specific neurogenic perspective on the stress deficits is their persistence or lack of normalization. It is especially unusual to observe the types of inappropriate stress patterns observed in the older children with suspected DAS in this study, some as old as 14 years 11 months (cf. Lewis & Shriberg, 1994). A specific neurological deficit provides a compelling explanatory source for an otherwise inexplicable resistance to improvement in many children with suspected DAS.

The third and arguably strongest consideration in support of neurological involvement is the appeal to adult acquired apraxia of speech, in which neurological insults are clearly documented as the origin of apraxia of speech. As described previously, stress deficits essentially similar to those observed in the approximately 52% of children with suspected DAS in the present studies have also been observed in adults with AOS. The qualifier *essentially* is important because the topographies of the excessive-equal stress patterns are not exactly similar in each group. As noted previously, the excessive-equal stress patterns observed in the present studies are not characterized by the lengthened vowel durations, increased pause times, and reduced speech rates reported for adults with AOS (Kent & McNeil, 1987; Kent & Rosenbek, 1983; McNeil & Kent, 1990). These and other differences underlie the proposal of different psycholinguistic loci for DAS and AOS, which, in turn, call into question whether DAS is the correct nosological term for this disorder (see Hypothesis V). What is needed for detailed examination of these issues are comparative studies of AOS and DAS using similar methods to quantify stress deficits. Pending findings from such studies, the perspective suggested here is that

stress deficits, rather than any pattern of segmental error targets or segmental error types, most clearly associate DAS with acquired AOS.

As above, only preliminary comment is appropriate on the specific neurological deficits that may underlie the type of stress deficits observed in this study. Studies of hemisphere specialization for prosody find extensive laterality of prosodic processing, with right hemisphere associated with pragmatic, affective, and emotional uses, and left hemisphere associated with the comprehension of lexical stress (Cohen, Branch, & Hynd, 1994). Left hemisphere cortical involvement is therefore a candidate for the type of productive linguistic stress deficit observed in these studies. As of yet, however, it has not been demonstrated that left hemisphere lesions sustained by children result in linguistically based prosodic disturbances. Given the near complete recovery of other linguistic abilities following left hemisphere lesions in children (Aram, Ekelman, & Whitaker, 1986, 1987; Feldman, Holland, Kemp, & Janosky, 1992), it would be premature to argue that left hemisphere lesions in children fully account for the observed prosodic disturbances. Other sites clearly are involved in the acquisition and realization of phrasal stress. For example, Gracco (1990) describes the articulatory realization of stress, noting that the physiology involves “increases in the actions of all portions of the vocal tract rather than being focused on one specific articulator” (p. 9). Hird and Kirsner (1993) propose a model of prosody that involves cortical and physiological control processes.

The notion of a deficit in praxis underlying the stress deficit further complicates an eventual account of neural substrates. If the pattern of excessive-equal stress observed in the present studies is eventually associated with segmental deficits, the pathophysiology of speech apraxia will require additional explication. Currently, it is not clear whether there are common neurophysiological mechanisms underlying all praxic deficits or whether there are separate praxis systems: for example, one for planning and controlling limb gestures, another for planning and controlling orofacial movements, and another for speech (Dewey, 1993; Ochipa, Rothi, & Heilman, 1992). Moreover, developmental aspects of praxis deficits must be accounted for. In a review of 86 studies, Kools and Tweedie (1975) concluded that some forms of praxis are not fully developed until 5–6 years of age.

#### **Hypothesis IV: The Distal Origin of This Form of DAS Is an Inherited Genetic Polymorphism**

Hypothesis I addresses the primary goal of this research, and Hypothesis IV the second goal. Rationales for the hypothesis of a genetic origin for the stress

deficits identified in this study include four sets of considerations.

First, although not differentiated by the present stress findings, the familial aggregation information on children with suspected DAS is higher than even the familial aggregation data for developmental speech-language disorders. As reviewed in the first paper in this series (Shriberg et al., 1997a), a preliminary population prevalence estimate for DAS based on clinical referrals is 1–2 per thousand. Yet, a study-wise average calculated on the 8 eligible studies described by Hall et al. (1993, Table 5.1, pp. 88–90), indicates that approximately 60% of children with suspected DAS had one or more relatives demonstrating communication and/or academic problems. Several of these studies indicated that one or more siblings, parents, or other relatives had the same type of disorder (i.e., suspected DAS). A 60% familial aggregation estimate is higher than those from some methodologically comparable studies of children with speech-language impairments of unknown origin. Lewis, Cox, and Byard's (1993) literature review estimated familial aggregation for developmental speech-language impairment at 24%–46%. However, Shriberg and Kwiatkowski (1994) reported that 56% of 84 children with speech delay of unknown origin had one or more family members who, on parental report, had a history of a speech problem.

Second, the previously described male:female prevalence ratios of as high as 9:1 are also higher than average estimates for children with speech delays of unknown origin (3:1), strongly suggesting a sex-influenced transmission model. In the present study, inappropriate stress was found in 50% of the 34 boys with suspected DAS and 57% of the many fewer (14) girls. As suggested previously, some sex-influenced genetic transmission models predict that affected girls will be more severely involved than affected boys (i.e., a *threshold effect*; cf. Plomin, Defries, & McClearn, 1990). However, calculation of the average stress and PCC scores for the 25 children with inappropriate stress indicated that girls were not significantly more involved than boys on either variable.

A third rationale in support of a genetic origin for the disorder reported here refers again to the notion that inappropriate stress reflects a qualitative rather than a quantitative difference in speech acquisition. As indicated previously, the argument for viewing inappropriate stress as a qualitative disorder is based on two findings in developmental and clinical linguistics: (a) Most children have appropriate phrasal stress at the outset of speech acquisition, and (b) stress deficits seem to persist past the biosocial developmental period for speech acquisition (approximately 9 years of age). Although both qualitative and quantitative behavioral traits can be



genetically transmitted, qualitative traits are more commonly associated with a major gene locus (i.e., a polymorphism at one chromosomal site). For example, Hurst, Baraitser, Auger, Graham, and Norell (1990) describe a three-generation family in which 16 members have a "severe developmental verbal dyspraxia." Inheritance was autosomal dominant, with full penetrance (all persons estimated to have the DAS genotype expressed the phenotype). Saleeby, Hadjian, Martinkoski, and Swift (1978) reported defective speech termed "verbal dyspraxia" in 34 of 66 family members, 12 of whom were directly tested with a spontaneous speech sample. Stress deficits of the type observed in the present study were observed in some (but not all) affected family members tested. The inheritance pattern was consistent with autosomal dominant, but with incomplete penetrance, because it occurred in each generation and affected males had affected sons. More recently, Vargha-Khadem, Watkins, Alcock, Fletcher, and Passingham (1995) provide evidence for praxic involvement in approximately half of a 30-member, four-generational family, again consistent with monogenic, autosomal dominant transmission. Thus, as a qualitative rather than quantitative trait, the mode of inheritance in DAS is likely to involve one major genetic polymorphism (a version of a gene that codes for a disorder), rather than mixed models of transmission in which there is a major locus and other genes contribute fractionally to the expressed form of the disorder.

A fourth observation in support of a genetic origin is that apraxia is reported to occur in inherited metabolic disorders and other inherited syndromes. Examples include Angelman Syndrome (Penner, Johnston, Faircloth, Irish, & Williams, 1993), galactosemia (Nelson, Waggoner, Donnell, Tuerck, & Buist, 1991), Fragile X syndrome (Hanson, Jackson, & Hagerman, 1986; Newell, Sanborn, & Hagerman, 1983; Paul, Cohen, Breg, Watson, & Herman, 1984), Prader-Willi syndrome (Branson, 1981; Munson-Davis, 1988), Renpenning syndrome (McLaughlin & Kriegsmann, 1980), and Robinow's syndrome (Hall et al., 1993). A provocative example for genetic linkage studies is reported by Nelson et al. (1991), who studied 24 patients with galactosemia, a rare metabolic disorder involving a failure to convert galactose to glucose. Nelson et al. found that half of the subjects met their criteria for verbal dyspraxia, concluding that the findings "indicate the association of a specific and unusual speech defect with a specific and rare metabolic disorder" (p. 45). There also are case-study opportunities to pursue the possibility of nonmetabolic, noninherited genetic origins of DAS. For example, in Schiff-Myers and Weistuch's (1994) detailed report on a child with translocations of portions of chromosome 1 and 2, there is considerable support for SLI and severe apraxia of speech.

A genetic basis for the stress-based subtype of DAS proposed here mandates certain constraints on phenotype markers. Severe phonological involvement, as indexed by delayed onset and/or severely delayed development of speech, is frequently proposed as a diagnostic marker or at least an important consideration for the diagnosis of DAS. Because genetically transmitted disorders may vary in severity of expression, severity of involvement cannot itself be used as a phenotype marker for the disorder. If the transmission mode is monogenic (one major gene locus), the disorder should be characterized by less variability of expression than if transmission is by a relatively small (oligogenic) or a relatively large (polygenic) group of genes, each contributing additive variance to the expression of the disorder. Such issues bear on earlier discussions of DAS as a clinical entity versus the possibility of clinically and/or etiologically distinct subtypes. Specifically, there may be two or more DAS genotypes coding for two or more DAS phenotypes.

Finally, inappropriate stress may be an excellent candidate for an "ideal" phenotype marker, as discussed by Pennington (1986) and reviewed at the outset of the second paper in this series. Relative to Pennington's five criteria for ideal phenotype markers, stress (a) has *early onset* and *developmental persistence*; (b) reflects a *single*, rather than multivariate domain; (c) has a *bimodal* distribution, yet can be measured as a *continuous* variable in nonaffected relatives; and, (d) has a *logical and potentially causal* relationship to the set of speech features associated with DAS. Appropriate molecular genetic studies are needed to determine whether inappropriate stress meets Pennington's fifth criterion of a genotype—that it has *full penetrance*.

### **Hypothesis V. Significant Differences Between This Possible Subtype of DAS and Acquired Apraxia of Speech in Adults Call Into Question the Inference That It Is an Apractic, Motor Speech Disorder**

It is important to question whether the children with inappropriate stress in these studies warrant the diagnostic label DAS. As reviewed, other than the stress findings, there were no speech or nonspeech findings consistent with adult acquired apraxia of speech. Specifically, the children with inappropriate stress did not have inconsistent speech errors or slowed speech rates, and few had groping, revisions, or lowered nonvolitional oral performance consistent with an apractic, motor speech disorder. Crucially, the perspective taken on the loci of the stress deficits (Hypothesis II) differs from the most prevalent view of acquired AOS. In adults with AOS, problems of access to (selection-retrieval) or motor programming (prearticulatory sequencing) of long-

established underlying representations are more parsimonious explanations for the results of a lesion than inferring loss of the representations themselves. In children, however, the stress deficits identified in this paper can more readily be attributed to a developmental difference in the acquisition of underlying representations. Such deficits are not, of themselves, consistent with the construct of a pragmatic disorder affecting speech production (albeit they are consistent with more cognitive views of praxis, such as ideational praxis). An alternative or at least provisional perspective on the children identified in this study is that they have a *prosodic*, or more narrowly, a *stress* disorder of unknown origin (see relevant discussion in Vance, 1994). Wells's (1994) description of a child with a "junction" deficit who elsewhere might be labeled apraxic is a useful example of this typological alternative to diagnostic classification. The following section considers some relevant research issues toward study of this hypothesis and the other four.

## Research Issues

An eventual account of the nature and origin(s) of DAS requires better measurement approaches and more powerful designs than reported to date, including the many constraints on the methods used in the present three studies. Thus, additional comment on the stress findings is deferred, pending additional validation studies using better designs. Following are some specific methodologic suggestions.

### Measurement

#### Lexical and Phrasal Stress

Quantification and classification of inappropriate stress can be pursued in two ways. First is the collection of perceptual and acoustic information from conversational speech samples—as in the present studies. Alternatively, information can be obtained from controlled comprehension and evocation tasks. For both approaches, but particularly those involving spontaneous conversation, phonological analysis using emerging nonlinear frameworks should be productive for the subtype of DAS proposed in this report. Moreover, conversational speech sampling procedures such as those described by Chiat and Hirson (1987) and Wingate (1984) include methods to associate normal and inappropriate stress with other parameters within speech and language. Transcription issues will continue to pose an especially difficult problem in conversational speech sampling, where casual speech forms and dialectal variants require close attention in studies using broad as well as narrow phonetic transcription.

Development of conceptually and psychometrically appropriate tasks or tests for stress is an attractive alternative to conversational speech sampling. Ideally, parallel tasks to assess comprehension and production of lexical, phrasal, and emphatic stress would include lists of multisyllabic words and phrases reflecting varying lexical-semantic, morphosyntactic, and phonological contexts. Considerable developmental work would be needed to evolve and document instruments with demonstrated construct validity. Roy and Square-Storer's (1990) parametric analyses of variables affecting sequencing errors indicate the number of potentially relevant independent variables. For example, these authors found that "the impairment in sequencing with left hemispheric damage arises only when the sequence must be generated from memory" (p. 485). An array of extant analyses, tasks, and measures provide a starting point from which to elaborate a composite set of tasks (e.g., Blakeley, 1980; Chiat & Hirson, 1987; Gerken, 1994; Hargrove & McGarr, 1994; Hayden, 1994; Hird & Kirsner, 1993; Kaufman, 1984; Milloy, 1985; Panagos & Prelock, 1994; Shadden, Asp, Tonkovich, & Mason, 1980; Stark & Blackwell, 1995). Whichever the form of assessment, conversational speech sampling or controlled elicitation, a central need is to differentiate whether deficits in lexical stress always accompany deficits in phrasal stress, with implications for theory and intervention.

#### Other DAS Variables

There also is need for improved measurement of nonspeech variables associated with DAS, with ongoing work in adult AOS providing useful models. For example, Roy and Square-Storer (1990) have developed procedures that meet the need for behavioral descriptions of *groping* in adult AOS, using motor notation systems that describe the temporal unfolding of motor sequences in limb, oral, and verbal apraxias. McNeil and Kent (1990) and Munhall (1989) discuss methods to assess variability in rate and other speech dimensions. Emerging data and discussions on the neural substrates of phonological development (cf. Christman, 1995a, 1995b; Kent, 1995), children's speech rate, diadochokinesis, and oral-volitional movements (e.g., Ansel, Windsor, & Stark, 1992; Blackwell & Stark, 1993) should provide the information needed to document developmental biolinguistic validity.

### Research Designs

Several types of research designs can provide converging evidence on the nature and origins of DAS, including the following five approaches.

#### Longitudinal Studies

For both theoretical and applied needs, an eventual account of DAS will require life span information on

children with DAS. For example, Le Normand and Chevie-Muller (1991) and Stackhouse and Snowling (1992) each provide detailed speech data on case studies followed over several years. Stackhouse (1992) documents how a child had each of the common behaviors associated with DAS at some point in his development. Crucial predictive questions for clinical counseling can only be addressed in such designs. For example: Which behaviors associated with DAS may be expected to normalize during the developmental period or later, and which, if any, may be expected to persist, perhaps for life?

### Comparative Studies

Kelso and Tuller (1981) make a telling observation on how the lack of collaborative research has limited the knowledge base in apraxia research: "It is an interesting but perhaps distressing feature of science that different areas of study, each bearing a strong potential relationship to the other, can function independently, each in its own oblivion" (p. 224). Research in DAS could be markedly aided by comparing findings from studies of children with the type of stress problem identified here with speech-prosody findings in other clinical populations (e.g., stuttering, cluttering, aphasia).

One need cited previously is for studies in which children with suspected DAS and adults with AOS are assessed with the same methods by the same researchers. Considering the variability due to sampling, data reduction, and data analyses in the child and adult apraxia literatures, a study of both groups using the same protocols and procedures could test whether and the degree to which there are parallel profiles of speech and prosody-voice involvements.

A second useful source for comparative study is samples from other childhood speech disorders in which apraxia of speech has been reported or suspected, such as inherited and other syndromes referenced previously and other groups reported to have apraxias (e.g., clumsy children, Gubbay, 1975). Benefits of studying apraxia in children with other involvements include the greater availability of subjects and the candidate genetic loci that might be suggested by converging data across diverse syndromes.

A third source for comparative study is children with suspected DAS acquiring a first language that differs from English in its stress marking. English is a stress-timed language, whereas languages such as French are syllable timed (see Sato, 1994, footnote p. 47, for alternative perspectives). As with other cross-linguistic studies in child language, such studies can provide environmental controls on linguistic variables that are otherwise unavailable (see Matthews, 1994, for examples of cross-linguistic convergence on a research question).

### Behavioral and Molecular Genetics Studies

A third type of research design is behavioral and molecular genetic studies using different family members (i.e., siblings, twins, nuclear and extended relatives) to test associations between genotypes and phenotypes. Unlike speech, which typically normalizes in children with the subtype of DAS proposed here, the stress deficit should still be identifiable in older children and relatives using measures such as those discussed above. Thus, the phenotype for this subtype of DAS should be readily measurable in molecular genetics designs that have the potential to identify the genetic loci of inherited childhood speech disorders (Lander & Schork, 1994; Shriberg, 1993). Until such time as that promise might be fulfilled, including subsequent understanding of gene-to-behavior pathways, the following discussion considers several applied issues.

### Clinical Issues

#### *Nosological Considerations*

A perspective that follows from the brief discussion in Hypothesis V and that warrants initial consideration here is the effect of nosological terms in professional training and service delivery. DAS is taught and serviced as a motor speech disorder, which has both advantages and disadvantages to instructors, clinicians, caregivers, and children who are given this diagnostic label.

An advantage of DAS as a classification label, however tentative, is that this diagnostic term provides some measure of comfort as an explanation for speech errors that may be mild to profound, but resistant to change despite the best efforts of caregivers, clinicians, and the child. The subsequent and perhaps even greater advantage is that DAS or some variant of this term is a passport to clinical services that may not be available with alternative diagnostic labels. Specifically, maximal clinical services can be requested and typically are readily obtained for children classified as having a motor speech disorder, as opposed to a functional or educational disorder.

Possible disadvantages of the diagnostic label DAS include the assumption of more pervasive deficits than those associated with severe developmental phonological disorders. By reifying speech-motor involvement, the label DAS can be intimidating to the clinician with limited background in the disorder, setting the stage for possibly counterproductive assumptions about treatment, long-term prognosis, and the child's self-concept. The source of clinicians' perceived or real lack of preparation may be traced to academic programs that exclude

DAS from courses in developmental phonological disorders on the assumption that it is more appropriately placed within motor speech disorders. Depending on faculty germandering of curriculum in a training program, DAS often winds up not being taught or being given short shrift by instructors in adult neurogenics with little experience or interest in pediatric speech disorders.

As reviewed previously, there presently is a concern whether the form of a prosodic-speech disorder identified in this paper warrants the label DAS. A nosological position that seems appropriate at this point is to suggest clinical use of the term *suspected DAS* if assessment results warrant it (see below), and if the term meets criteria needed to obtain the appropriate amount and type of service delivery.

## Assessment

Although there are a number of useful diagnostic instruments to assist clinicians to assess children with suspected DAS, none enjoys widespread acceptance. Pending the availability of consensus on a diagnostic instrument, a suggestion is that clinicians consider three types of children who are suspect for DAS. As suggested below, only children meeting criteria for the first two types warrant the diagnostic term, *suspected DAS*.

The first type of child warranting classification as suspected DAS presents with obvious difficulties in several areas associated with adult AOS and DAS. Such behaviors include clearly documented nonspeech apraxias, clearly observed groping or other difficulties in speech onsets, or marked token-to-token inconsistencies (including deletions, substitutions, distortions) within both phonetically simple and phonetically difficult words. The classic model for this type of child would be the inconsistent repetition of multisyllabic words (e.g., *tornado*, *statistics*) attributed to adults considered to have AOS. Rate considerations are important in differentiating children in this possible subtype from those with dysarthria or other conditions. Kent and Rosenbek (1983) have noted that slow rate "probably contributes significantly to the perceptual impression of apraxic speech as effortful and groping" (p. 243). Rosenbek and Wertz (1972) reported that 26% of clinician referrals for suspected DAS had combinations of apraxia and dysarthria (16% had combinations of apraxia, aphasia, and dysarthria), underscoring the possibility of multiple involvements for children presenting with this array of diagnostic features.

A second subtype of suspected DAS is a child who clearly has inappropriate stress of the type identified in the present study. Within multisyllabic articulation test responses, attention should focus on the child's stress consistency and any deviation from appropriate lexical

stress. A procedure we have found useful to observe both segmental and suprasegmental consistency is to obtain two spontaneous and two imitated tokens of selected words on a standard articulation test, particularly multisyllabic words. Children with common speech delay generally improve within each mode, with the second spontaneous trial a chance to correct errors on the first trial, and the two imitative trials a chance to profit from the examiner's model and the previous two trials. For some children, however, including those with suspected DAS, there is a notable diminution of performance on both second trials, as though the enterprise had become confusing. Our tentative explanation is that efforts to produce correct stress can be detrimental, causing more errors or trade-offs in the errors produced. Stimulability testing of isolated continuants does not disclose this effect, whereas requests to imitate multisyllabic words with variable and unusual stress patterns evokes the inconsistency. For the assessment of phrasal stress, conversational speech samples can be used to obtain the same types of utterance-level tallies of inappropriate stress as described for the studies reported here.

A third type of child is one who, upon testing as above, has neither the clear segmental difficulties nor the inappropriate stress patterns observed in the present studies. Children who might only have one or a combination of the remaining characteristics thought to be associated with DAS (e.g., late speech onset, greatly reduced phonetic inventories, very low PCC scores, inconsistent errors, or even atypical speech errors) do not warrant the term suspected DAS. Although this tentative diagnostic term may be useful to obtain services, findings in the current study suggest that such children are not descriptively different from children who have been labeled late talkers or children with speech delay of unknown origin.

## Treatment

Guyette and Diedrich (1981) note that in medicine the diagnostic label is expected to indicate etiology, prognosis, and treatment, although useful even if only associated with effective treatment. Love (1992) also stresses the value of labels for intervention options: "One significant value in correctly diagnosing a child with [DAS] is that the diagnosis often radically changes the direction of therapeutic management and opens the door to a variety of techniques not usually employed with the typical child with developmental phonological disability or suspected developmental aphasia" (p. 98).

If the hypothesis of inappropriate stress as a DAS subtype is supported by additional research, the diagnostic label of DAS does suggest a marked change in the direction of therapeutic management. As noted

previously, inappropriate stress is a “continuing element” in children with suspected DAS, regardless of how much they improve in sound production. The intervention assumption is that such children need production and likely comprehension work on stress assignment, whereas children with other subtypes of DAS require focus on segmental aspects of prearticulatory sequencing. Both types of intervention foci differ from the presumed primary needs of children with common speech delays, for whom intervention typically focuses on selection and retrieval of correct segmental targets in increasingly complex phonetic, morphosyntactic, and discourse contexts.

Several established and emerging intervention approaches are consistent with an emphasis on stress assignment. Focus on stress is central to intervention approaches that involve imitation of syllable sequences, sometimes varying in other prosodic features. Some examples include variants of Melodic Intonation Therapy (MIT) (e.g., Albert, Sparks, & Helm, 1973; Grube, Spiegel, Buchhop, & Lloyd, 1986; Helfrich-Miller, 1984; Krauss & Galloway, 1982; Schumacher, McNeil, & Yoder, 1984) and various classic and new approaches that include contrastive stress drills and production practice using polysyllabic words (e.g., Chumpelik, 1984; Grube et al., 1986; Klein, 1981; Kusko, 1980; Liss & Weismer, 1994; Young, 1995).

Other contemporary approaches for children with suspected DAS emphasize the need to teach or refine a general rhythmic deficit. Sequencing activities involving whole body movements (e.g., Ballard, 1986) are based on the premise of common neurological substrates among praxis disorders, such that improved movement and praxis in nonspeech domains should have positive transfer to speech domains. Findings for the subtype proposed in the present study are more consistent with the view that the disorder is neither a praxic nor a sequencing deficit. Rather, the deficit is a linguistic problem reflected in the representation of stress assignment. Accordingly, rather than being addressable by generic therapeutic activities motivated by rhythmic, sequencing, or praxis needs, the deficit may require individually tailored intervention units specifically addressing each child’s inappropriate stress profile.

Some emerging procedures illustrate an emphasis on matching intervention specifically to children’s linguistic deficits, rather than following uniform “programs” for children with suspected DAS. Velleman (1994) presents two case studies of children with DAS illustrating the complex role of the syllable in these children’s phonological deficits. Chiat and Hunt’s (1993) case study indicates that speech variability cannot be traced to one level of lexical-semantics or phonology, with implications for intervention. Stackhouse and Snowling

(1992) reach a similar conclusion after detailed study of 2 children with suspected DAS, noting that “...the precise nature of their speech errors was related to their individual psycholinguistic strengths and weaknesses” (p. 51).

## Summary and Conclusions

The primary goal of the studies reported in this series was to determine if a diagnostic marker could be identified for the clinical entity termed (among several other labels) Developmental Apraxia of Speech (DAS). Secondary goals were to consider the level of support for DAS as a genetically transmitted disorder, and to discuss implications of findings for research and clinical practice. The following is a summary of findings and conclusions.

1. Conversational data from three samples of children with suspected DAS indicated that a deficit in phrasal stress was the only linguistic variable that statistically differentiated 52% of these children from age-matched comparison children with speech delay of unknown origin. Consideration of alternatives lead to the suggestion that the most useful interim interpretation is that such children represent a subtype of children with suspected DAS. It is likely that there also is at least one other form of DAS that is marked by one or more of the array of segmental deficits described in several diagnostic checklists.

2. Observations about the prevalence and course of inappropriate stress in children lead to the hypothesis that the disorder reflects a qualitative deficit of neurologic origin. Epidemiologic and other descriptive features suggest that this developmental biolinguistic deficit may be genetically transmitted. Pending confirmation in imaging or other studies, inappropriate stress can serve as a behavioral marker for research studies and applied needs.

3. In comparison to features reported to characterize adult AOS, the present findings suggest that the stress deficit in this form of DAS occurs within linguistic representational levels of phonology, rather than within prearticulatory sequencing. Because the disorder is, therefore, more consistent with a phonological as opposed to a speech-motor deficit, DAS may not be the appropriate term for the group of children identified in these studies. However, pending the needed research relating such deficits to praxic deficits, such children might be classified as having *suspected DAS*, which meets several important service delivery needs. Suggestions for research, assessment, and treatment of this proposed subtype of DAS emphasize the distinctions between phonological and motor-speech processes as targets for theoretical investigation and behavioral change.

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## **Developmental Apraxia of Speech: III. A Subtype Marked by Inappropriate Stress**

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