Clinical Linguistics & Phonetics, May 2012; 26(5): 445–482

ISSN: 0269-9206 print / ISSN 1464-5076 online
DOI: 10.3109/02699206.2012.655841

informa
healthcare

Encoding, memory, and transcoding deficits in Childhood Apraxia of Speech

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(*Received 6 September 2011; revised 3 January 2012; accepted 5 January 2012*)

Abstract
A central question in Childhood Apraxia of Speech (CAS) is whether the core phenotype is limited to *transcoding* (planning/programming) deficits or if speakers with CAS also have deficits in auditory-perceptual *encoding* (representational) and/or *memory* (storage and retrieval of representations) processes. We addressed this and other questions using responses to the Syllable Repetition Task (SRT) [Shriberg, L. D., Lohmeier, H. L., Campbell, T. F., Dollaghan, C. A., Green, J. R., & Moore, C. A. (2009). A nonword repetition task for speakers with misarticulations: The syllable repetition task (SRT). *Journal of Speech, Language, and Hearing Research*, *52*, 1189–1212]. The SRT was administered to 369 individuals in four groups: (a) typical speech–language (119), (b) speech delay–typical language (140), (c) speech delay–language impairment (70), and (d) idiopathic or neurogenetic CAS (40). CAS participants had significantly lower SRT competence, encoding, memory, and transcoding scores than controls. They were 8.3 times more likely than controls to have SRT transcoding scores below 80%. We conclude that speakers with CAS have speech processing deficits in encoding, memory, and transcoding. The SRT currently has moderate diagnostic accuracy to identify transcoding deficits, the signature feature of CAS.

Keywords: *apraxia*, *dyspraxia*, *genetics*, *motor speech disorder*, *speech sound disorder*

Issues, Findings, and Research Needs in Childhood Apraxia of Speech

Research in Childhood Apraxia of Speech (CAS) in the current century has begun to address descriptive–explanatory questions, using theory and methods from a number of new and emerging disciplinary perspectives. The following four sections provide overviews of issues, findings, and needs in four interdependent research areas in CAS: origins, neuromotor substrates, speech processes, and signs and correlates. A final section describes the research framework for CAS, underlying the study to be described.

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Distal origins of apraxia of speech

The two substantially independent research literatures in apraxia of speech, CAS and AOS (the conventional acronym for acquired apraxia of speech), are historically based on significant differences in etiological and temporal origins (i.e. distal causes). Congenital origins of CAS include individuals with genetic, epigenetic, and idiopathic CAS in syndromic and nonsyndromic contexts (see reviews in Shriberg, 2010b; Shriberg, Potter, & Strand, 2011). Congenital origins may be due to hereditary transmission of genomic variants or sporadic (*de novo*) genomic disruptions present at birth or shortly thereafter. In comparison, AOS consequent to neuropathology can occur any time during or after the neurodevelopmental period for speech–language acquisition, most commonly later in adulthood due to stroke (Duffy, 2005). In contrast to the relatively large body of descriptive–explanatory research in AOS during the past two centuries (reviews in Duffy, 2005; McNeil, Robin, & Schmidt, 2009; Robin, Jacks, & Ramage, 2008; Weismer, 2007), research in the origins of CAS has occurred primarily in the past two decades referenced as the genomic and current post-genomic periods. Some literature reviews of genetic research in verbal trait disorders that include speech-genetics research in CAS include Newbury, Fisher, and Monaco (2010), Newbury and Monoco (2010), Shriberg (2010b), Bishop (2009), Fisher and Scharff (2009), Grigorenko (2009), Ramus and Fisher (2009), Stromswold (2008), Caylak (2007), Fisher and Marcus (2006), and Lewis et al. (2006). In addition to extensive studies of the *FOXP2* gene, including research using mammalian and avian orthologs of this transcription gene, CAS has recently been associated with several genes including *CNTNAP2* (Vernes et al., 2008), *FOXP1* (Carr et al., 2010; Hamdan et al., 2010; Horn et al., 2010; Pariani, Spencer, Grahan, & Rimoin, 2009), *FOXG1* (Brunetti-Pierri et al., 2011), *ELP4* (Pal, Li, Clarke, Lieberman, & Strug, 2010), and *RAI1* (Kogan, Miller, & Ware, 2009). Detailed phenotype information on speech, prosody, voice, cognitive, language, affective, and other findings in association with CAS have been reported for four persons with *FOXP2* disruptions in two unrelated families (Rice et al., 2011; Shriberg et al., 2006; Tomblin et al., 2009) and in three siblings with CAS associated with a chromosome translocation (Shriberg, Jakielski, & El-Shanti, 2008). Two differences in the causal and temporal origins of CAS compared to AOS have implications for the issues and questions in the discussions to follow of speech processing and signs and correlates of apraxia of speech.

The primary difference between CAS and AOS is the neurobiology of CAS associated with neurogenetic and epigenetic origins, in contrast to the neuropathologies underlying AOS (e.g. trauma, infectious processes, and stroke). In neurogenetic CAS, cognitive and sensorimotor development may be affected in all brain regions and circuits in which gene expression is disrupted, including disruptions in genes regulated by other genes. For example, *FOXP2* is expressed bilaterally and widely in the infant and adult brain, with gene expression studies reporting involvement of many neural sites and circuits active in speech processing and other studies describing its regulatory role in expression of other genes (e.g. *CNTNAP2*, *ATP2C2*, and *CMIP*; see Newbury et al., 2010).

A second difference between CAS and AOS is the temporal consequences of neural impairments relative to speech–language acquisition. Congenital deficits affect both target systems and systems dependent on the integrity of prior growth and development. In a review of descriptive–explanatory studies with colleagues, using a variety of instrumental methods, Maassen (2010) has discussed the unifying concept of *developmental trajectories* in CAS. Maassen cites Karmiloff-Smith's (2006) perspective on the gradual emergence of the adult modular system and Bishop's (1997) observation that associations are the rule in developmental disorders (i.e. compared to the dissociations in acquired disorders used in

cognitive neuropsychology to support modularity concepts). As discussed in the following overviews, the pathophysiologies and temporal onsets of CAS, in comparison to those in AOS, present challenges to accounts of apraxia of speech as one clinical entity.

Proximal origins of apraxia of speech

The second explanatory loci relevant to the present study include neurocognitive and neuromotor constructs in speech processing imputed to be the proximal sources for apractic speech. Research in typical and especially atypical speech acquisition deconstructs speaking into four constituent processes, with the neural substrates for each process accomplishing a product necessary for articulate speech. These constructs include: (a) auditory-perceptual *encoding* processes that transform auditory input into phonemic, sublexical, and lexical representations; (b) *memory* processes that store and retrieve these representations; (c) *transcoding* processes that plan and programme the representations for the motoric gestures of manifest speech or other forms of communication such as signing, finger spelling, and typing; and (d) neuromotor *execution* processes. Success in speaking and in tasks that require fast, short-term processing of novel words (e.g. nonword repetition tasks) is dependent on the integrity of speech processing at each of these four "stages" (see Ellis Weismer & Edwards, 2006, for a discussion of interactivity among such putatively serial constructs). Specific mechanisms within each element and the influence of mediating and moderating variables (e.g. age, gender, phonological awareness, processing speed, and articulation rate) differ considerably within and among the many disciplines that use speech processing constructs in diverse psycholinguistic, neurocognitive, and speech motor control frameworks (e.g. Bock, 1982; Dell, 1986; Guenther, 1995; Levelt, 1989; van der Merwe, 2008; Stackhouse & Wells, 1997; Ziegler, 2006). Brief comment on encoding, memory, and transcoding findings in CAS are relevant to the findings to be reported in the present study.

Encoding and memory deficits. Unlike AOS, in which premorbid representations of words, syllables, and sounds are assumed to be intact, CAS presumably interferes with the rate if not type of acquisition of linguistic representations. Although there are many alternative causal accounts of encoding and memory deficits in primary language impairment and speech delay, there is general consensus that incomplete or poorly formed representations due to encoding and/or memory constraints are the proximal cause(s) of primary language impairment and speech delay. A wide-ranging literature has documented such deficits, using a number of nonword repetition tasks with sociodemographically diverse samples of children with speech–language impairment (reviews in Graf Estes, Evans, & Else-Quest, 2007; Shriberg et al., 2009). Within the CAS literature, there has been a consistent trend to interpret findings as supporting encoding deficits as either a core or correlative feature of apraxia of speech in children (see extensive review in Froud & Khamis-Dakwar, 2011; Nijland, 2009). Velleman (2011, p. 83) in a recent discussion of CAS notes:

> There is some evidence for cognitive–linguistic as well as motor planning [deficits in children with CAS] components of the disorder, though, in that they typically demonstrate other, higher-order linguistic deficits that depend upon fully-formed phonological representations: reduced perception and production of vowels (Maassen, Groenen & Crul, 2003), syllables (Marquardt, Sussman, Snow & Jacks, 2002), rhymes (Marion, Sussman & Marquardt, 1993) and phoneme sequences – especially in nonwords (Bridgeman & Snowling, 1988); deficits in word attack; and difficulties with spelling that do not necessarily relate to the child's current speech errors (Snowling & Stackhouse, 1983; Lewis et al.,

2004). The nature of these deficits observed in children with CAS suggests impoverished phonemic representations (Marquardt, Jacks & Davis, 2004).

Notably, as before, the statistical data in findings to date indicate incremental between-group differences on encoding and memory variables, rather than differences approaching bimodal distributions. The major question in the study to be reported, which includes larger and more diverse CAS and speech–language samples than reported to date, is whether participants with CAS in both idiopathic and neurogenetic contexts have substantially lower encoding and memory scores compared to children with speech-only and speech–language impairment.

Transcoding deficits. Although motor speech disorders share many features that discriminate these disorders from typical speech and from speech delay (e.g. slower rate, vowel errors, and prosodic deficits), only transcoding deficits, by definition, are specific among motor speech disorders for apraxia of speech. There is currently no consensus in the AOS or CAS literatures, however, on what types of speech or prosody behaviours index planning/programming deficits. Proposed signs are generally based on some type of processing delay reflected in spatiotemporal disruptions within segments, clusters, syllables, or morpheme boundaries (i.e. subsumed by terms such as *delayed transitions*). The study to be reported identified a unique speech behaviour that was viewed as indexing transcoding deficits in the context of a nonword repetition task.

Signs and correlates of apraxia of speech

The "circularity" constraint in all CAS research, articulated over three decades ago by Guyette and Diedrich (1981, p. 39) in their iconic summary – (Childhood Apraxia of Speech is) "a label in search of a population" – is the lack of standardized inclusionary/exclusionary criteria for true positive participants with CAS. The search for biomarkers and behavioural markers of CAS and studies of its genetic, neurocognitive, and neuromotor substrates requires a standardized assessment protocol to quantify its core phenotypic and endophenotypic features as they evolve over developmental epochs. Other than consensus on a few non-operationalized and nonstandardized speech and prosody indicants of CAS in speakers at some ages (reviews in American Speech-Language-Hearing Association [ASHA], 2007; Jacks & Robin, 2010; Shriberg & Campbell, 2003; Shriberg et al., 2003), Guyette and Diedrich's challenge to develop a gold standard for CAS remains unanswered.

In the AOS literature, the Mayo Clinic System remains the standard classification system for acquired motor speech disorders, notwithstanding critique based on alternative views of the values of classification systems versus taxonomies (Weismer, 2006; Weismer & Kim, 2010). Essentially, there are no comparable systems for pediatric motor speech disorders (see also Steinman, Mostofsky, & Denckla, 2010). There currently are no validated signs of CAS, although there is emerging consensus that deficits in vowels, phoneme distortions, distorted transitions, unstable errors, and stress are candidate signs to discriminate CAS (ASHA, 2007). As discussed elsewhere, key needs are to organize, operationalize, and standardize a set of signs of CAS that has at least 90% sensitivity and specificity, yielding positive and negative likelihood ratios of at least 10.0 and no greater than 0.10, respectively (Shriberg, Strand, Jakielski, & Lohmeier, 2012). Shriberg, Potter, et al. (2011) and Shriberg et al. (2010a) includes a set of 87 potential signs of motor speech disorders, including subsets for each of the three subtypes of motor speech disorders to be described in the next section.

A framework for research in speech sound disorders

Figure 1 places the previous review of issues, findings, and needs in CAS research in the larger context of research in speech sound disorders (SSD). This framework unites three domains within SSD: a clinical typology, the speech processes discussed above, and diagnostic markers.

Clinical typology for SSD. The clinical typology for SSD in Figure 1 is the Speech Disorders Classification System (SDCS) described in detail elsewhere (Shriberg et al., 2010a, 2010b). SSD are divided into three superordinate classes based on their presumptive distal causes and consequent proximal speech processing deficits. Briefly, as described in the SDCS papers cited, Speech Delay (SD) includes the three distal origins shown in Figure 1 and Speech Errors (SE) includes two subtypes. A third class of SSD, termed MSD, subsumes three subtypes: Motor Speech Disorder–Apraxia of Speech (MSD-AOS (generic term for both CAS and AOS in the present context)), Motor Speech Disorder–Dysarthria (MSD-DYS), and a placeholder classification for speakers suspected to have MSD, but who do not meet speech criteria for MSD-AOS or MSD-DYS, termed Motor Speech Disorder–Not Otherwise Specified (MSD-NOS).

Speech processes. The central descriptive–explanatory domain of SSD shown in Figure 1 includes the four speech processes discussed previously. The proximal deficits of two of the three classes of SSD, SD and SE, are presumed to be neurodevelopmental constraints in auditory-perceptual encoding and/or memory (Shriberg et al., 2010a). As indicated previously, the consensus in most to all discussions of MSD-AOS (i.e. CAS) is that its proximal cause is a deficit in transcoding (planning/programming). Finally, the proximal causes of MSD-DYS, including subtypes of MSD-DYS (e.g. spastic, ataxic), are posited to be deficits in execution consistent with those in congenital and acquired forms of dysarthria.

Diagnostic markers. The third element in the SSD research framework addresses the diagnostic issue that is one of the two goals of this report – the need for diagnostic signs with

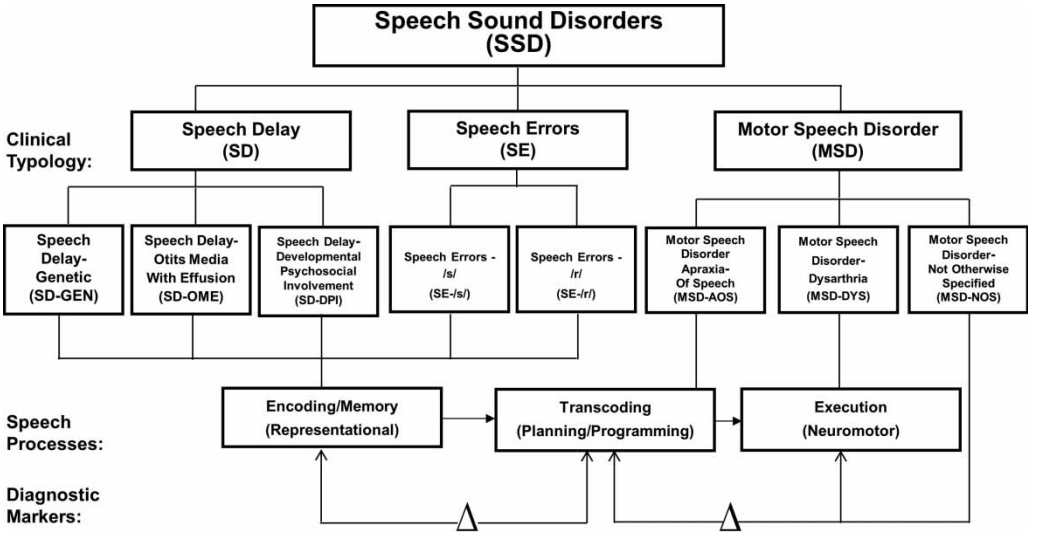

Figure 1. A framework for research in SSD.

sufficient accuracy to differentiate speakers with MSD from speakers with SD and that differentiate the three types of MSD from one another (i.e. diagnostic markers). The present report is part of a research programme to develop and validate behavioural signs as diagnostic markers of CAS by studying apraxia of speech in neurogenetic, neurologic, and idiopathic contexts in children and adults (Shriberg, 2010b).

The Greek *delta* symbol in Figure 1 denotes diagnostic markers meeting evidence-based criteria for conclusive research and clinical decisions, which in the behavioural sciences as indicated previously, need to exceed 90% sensitivity and 90% specificity to meet positive and negative likelihood ratio criteria (Dollaghan, 2007). Identification of behavioural signs that conclusively differentiate CAS from the several subtypes of dysarthria of currently unknown origin (MSD-DYS) can be addressed only when a methodologically robust clinical typology for idiopathic forms of dysarthria becomes available. In turn, such typologies should aid in the development of biomarkers. Many to most of the core features of CAS expected to be identified have been based on signs identified (but not validated) in the AOS literature. Others features are expected to be associated with genetic, epigenetic, and neurodevelopmental processes not present in speakers with later acquired CAS or with AOS.

Statement of the Problem

Emerging research on the genomic, neurobiology, and treatment of CAS continues to be limited by the lack of assessment procedures with sufficient diagnostic accuracy to conclusively identify true positives. Assessment procedures with high predictive value to rule out CAS (i.e. high specificity) are also needed for clinical decision-making, especially with young children with limited speech repertoires who currently are overdiagnosed as CAS (ASHA, 2007).

The research framework underlying the present study is based on the assumption that findings from studies of individuals with CAS in neurogenetic and neurologic contexts will inform descriptive–explanatory accounts of idiopathic CAS – the "…label in search of a population". Research using this strategy requires lifespan measures appropriate for participants at differing levels of cognitive, affective, and motor function whose speech output may be severely limited. Perceptual and instrumental scoring methods must have demonstrated reliability, requiring minimally demanding skills and time for data acquisition, data reduction, and interpretive analytics. The specific need is for valid quantitative information on each of the four speech processing domains in Figure 1.

A nonsense word task to be described, termed the Syllable Repetition Task (SRT; Shriberg & Lohmeier, 2008; Shriberg et al., 2009), was developed to meet the substantive, psychometric, and clinical efficiency needs just described. Nonsense word repetition tasks have played a major role both as diagnostic measures of verbal trait disorders (i.e. language, reading) and as endophenotypes in genomic research in speech, language, and reading. They provide culture-free information on a speaker's ability to acquire and produce real words. As described presently, they may also be informative about the speech processes underlying poor performance on such tasks.

The current study used information from four groups of participants' SRT scores to address two questions about speech processing deficits in CAS:

(1) *Are study findings consistent with a transcoding deficit-only account of CAS in neurogenetic and idiopathic contexts, or are they more consistent with a multiple domain account with deficits in encoding, memory, and transcoding processes?*

(2) *Do findings from SRT competence and processing measures contribute significant or conclusive diagnostic information to an assessment battery for CAS?*

Method

Comparison group participants

Table I includes summary information for the number, age, and gender of 369 study participants. Each of the 329 comparison group individuals (Groups 1–3) had participated in collaborative studies in speech sound disorders at research sites in several US cities. As part of the assessment protocols at each site, all comparison participants and 40 CAS participants (Group 4) in Table I were administered the SRT following procedural guidelines in Shriberg and Lohmeier (2008). The appendix includes the 18 SRT nonword stimuli and the following section summarizes relevant conceptual and methodological information for this measure.

Recruitment and assessment procedures for comparison participants were generally similar at each of the four research sites, indicated as A–D in Table I. Volunteer participants or their parents signed assent/consent forms approved by local institutional review boards to participate in 1 to 2 h speech–language assessment protocols at each site. Screening procedures and case history data were used to exclude participants for Groups 1–3 with frank cognitive, structural, sensory, motor, or affective disorders. Speech samples at each site were obtained in quiet rooms by trained examiners, using different high-quality digital recorders and matching external cardioid condenser microphones at a 44.1 kHz sampling rate with 16-bit resolution. All assessment protocols included a conversational speech sample obtained using a standardized procedure (Shriberg, Hersh, et al., 2008). Recorded stimuli for the SRT were presented using laptop computers and external speakers adjusted for comfortable listening.

Well-developed classification procedures for the SDCS (Shriberg et al., 2010a), the software running in the PEPPER (Programs to Examine Phonetic and Phonological Evaluation Records; Shriberg, Allen, McSweeny, & Wilson, 2001) environment were used to classify participants' speech status. As described in Shriberg et al. (2010a), classification of a speaker's speech status was based on speech data from a 5–10 min conversational speech sample. SDCS software classified participants as meeting criteria for Typical Speech (TS [termed Normal Speech Acquisition in the SDCS]) or Speech Delay (SD), the latter including a classification for marginal speech delay termed normal speech acquisition/Speech Delay. Participants' language status was classified as either typical language (TL) or Language Impairment (LI) based on whether their standardized scores on one or more language instruments used in each collaborative study met criteria for expressive or receptive–expressive language impairment. As shown in Table I, participants were divided into three comparison speech–language groups: Group I: Typical Speech–Typical Language (TSTL), Group 2: Speech Delay–Typical Language (SDTL), and Group 3: Speech Delay–Language Impairment (SDLI). Due to the focus on speech delay in the studies from which these participants were drawn, there were insufficient number of participants for a fourth potential comparison group of participants with Typical Speech-Language Impairment.

As shown in Table I, the number and types of participants obtained at the four research sites ranged from 148 participants from site A, approximately equally divided among the three comparison groups, to 20 participants from site D, all classified as SD. There were proportionately fewer Group 3 participants from site B, whereas site C participants were more proportionally divided among the three comparison speech status groups.

The ages of participants in the comparison groups ranged from 3 to 22 years, with sites A and D, including primarily preschool children, site B, elementary age children, and site C,

Table I. Descriptive information for the four study groups.

	Group 1: TSTL					Group 2: SDTL					Group 3: SDLI					Group 4: CAS				
	Age (yrs)				%	Age (yrs)				%	Age (yrs)				%	Age (yrs)				%
	n	*M*	SD	Range	Males	*n*	*M*	SD	Range	Males	*n*	*M*	SD	Range	Males	*n*	*M*	SD	Range	Males
Research site																				
A	52	4	1	3–5	38.5	53	4	1	3–5	71.1	43	4	1	3–5	67.4					
B	54	8	1	5–10	36.4	60	6	1	5–8	68.3	16	6	1	5–7	93.8					
C	13	15	5	5–22	23.1	9	14	4	8–18	66.7	9	11	4	5–18	77.8					
D	–	–	–	–	–	18	4	1	3–6	44.4	2	5	1	4–5	100.0					
Totals	119	7	4	3–22	36.1	140	5	3	3–18	66.4	70	5	3	3–18	75.7	40	11	8	4–50	60.0
CAS																				
Neurogenetic																				
Chromosome deletion or translocation																4	13	3	11–16	25.0
Copy number variations																2	11	5	8–15	100.0
FOXP2																4	24	19	4–50	25.0
Galactosaemia																8	9	4	5–16	75.0
Joubert syndrome																1	11	–	–	100.0
Prader–Willi syndrome																1	8	–	–	0.0
Neurogenetic subtotals																20	13	10	5–50	55.0
Idiopathic subtotals																20	9	4	4–19	65.0
CAS total																40	11	8	4–50	60.0

older youth. The wide age range of participants is considered a strength for generalizations from findings. To adjust scores for possible differences associated with age, the analyses to be reported include age as a covariate. The gender proportions for participants with SD at each site were generally consistent with the approximately 70% prevalence of males with SD. The proportion of males was somewhat closer to 50% for the TSTL participants in Group 1. Although significant gender effects for SRT data were not found in Shriberg et al. (2009), the analyses in this report included gender as a covariate.

It is useful to underscore the relatively large number and diversity of participants with speech delay in the two comparison groups in Table I, among the largest reported. With a few exceptions, the cell sizes, age ranges, and multiple geographic locations allow robust statistical and clinical comparisons to the data for the CAS participants to be described. In turn, the CAS participants to be summarized in the following sections constitute one of the largest reported samples and the most diverse in age range and diversity of origins.

Participants with CAS

Classification of Group 4 participants suspected to have CAS (to be described in a following section) as true positives for CAS was accomplished using the classification system developed

Table II. The third author's classification criteria for CAS (MSD-AOS) and dysarthria (MSD-DYS).

Linguistic and motor domains	MSD-AOS	MSD-DYS
Segmental		
Vowels	1. Vowel distortions	
Consonants	2. Voicing errors	
Vowels and consonants	3. Distorted substitutions	1. Sound distortions
	4. Difficulty achieving initial articulatory configurations or transitionary movement gestures	2. Reduced strength of articulatory contacts
	5. Groping	
	6. Intrusive schwa	
	7. Increased difficulty with multisyllabic words	
Suprasegmental		
Prosody		
Phrasing	8. Syllable segregation	3. Scanning speech
Rate	9. Slow rate	4. Slow rate
	10. Slow diadochokinetic rates	5. Irregular diadochokinetic rates
Stress	11. Equal stress or lexical stress errors	6. Equal stress
Voice		
Loudness		
Pitch		
Laryngeal quality		7. Strained or breathy phonation
Resonance		
Motor		8. Reduced range of motion
		9. Reduced respiratory support or respiratory incoordination
		10. Adventitious movements

Note: MSD-AOS requires vowel distortions and at least three of the listed characteristics in at least three of the MSAP tasks. MSD-DYS requires at least 3 of the 10 listed characteristics in at least 3 MSAP tasks.

by the third author shown in Table II. This system was used in a prior study of CAS in neurogenetic and idiopathic contexts (Shriberg, Potter, et al., 2011). The third author was first provided video or audio recordings of the assessment protocols completed by a number of participants in prior studies who had been suspected to have CAS. She used these samples to formalize quantitative criteria for the entries in Table II, most drawn from the adult AOS literature (Duffy, 2005). As shown in Table II, participants classified as positive for CAS met perceptual criteria for at least 4 of the 10 proposed signs of apraxia of speech, as each sign occurred in at least 3 of the speech tasks in the assessment protocol. Crucially, only one of the 40 participants with CAS also met the third author's minimum criteria for dysarthria shown in Table II. Interjudge reliability estimates for this procedure were reported in the study of CAS in participants with galactosaemia (Shriberg, Potter, et al., 2011). Classification agreement for the procedures was completed by a colleague of the third author with 30 years of clinical experience in pediatric and adult motor speech disorders. The system shown in Table II was used to classify a randomly selected 10 (25%) of the 40 samples. For these 10 samples, she listened to all the participants' recorded responses to either a preliminary or later version of the Madison Speech Assessment Protocol (MSAP; Shriberg et al., 2010a), a 2 h protocol that includes 15 speech tasks and several measures assessing hearing, cognition, language, and developmental history. The third author then made judgements regarding the presence of the speech, prosody, and voice signs in Table II and classified the samples as meeting or not meeting the criteria for CAS. Interjudge agreement with an experienced colleague trained on the classification procedure was 90%.

Returning to Table I, the lower section summarizes descriptive information for the 40 participants suspected to have CAS that had been obtained from local and collaborative studies in several states. As above, all participants had been assessed using the MSAP and all participants had been classified as true positives for CAS, using the third author's criteria shown in Table II. Half of the 40 participants with CAS had idiopathic backgrounds and the other half had a variety of neurogenetic backgrounds, including 8 participants with galactosaemia and 4 participants (from two unrelated families) with disruptions in *FOXP2*. Participants ranged in age at assessment from 4 to 50 years; however, there was only one participant older than 18 years in the subgroup with idiopathic CAS and three in the subgroup with neurogenetic CAS. The percentage of males in the neurogenetic subgroup (55%) was somewhat lower than in the idiopathic subgroup (65%) and lower than other studies of CAS reporting as high as 90% males (ASHA, 2007). The present finding of essentially equal gender prevalences of CAS in neurogenetic contexts replicates the approximately equal gender prevalence of possible CAS in the 55 cases in neurogenetic contexts reported in Shriberg (2010b) and recently obtained in a review of 23 cases of *FOXP2* disruptions and CAS (Palka et al., 2011), with implications for genomic pathway models of CAS.

Case history information and/or hearing screening indicated hearing within normal limits for 29 of the 40 participants, histories of screening failures and/or insertion of pressure equalization tubes for 7 participants, with no hearing information available for 4 participants due to technical problems. Intellectual assessment using several measures (Kaufman & Kaufman, 2004: *Kaufman Brief Intelligence Test-2*; Wechsler, 1997: *Wechsler Adult Intelligence Scale-III*; Wechsler, 1991: *Wechsler Intelligence Scale for Children-III*) indicated that 22 of the 37 participants with standardized test scores (59%) had composite or full-scale IQ scores below 85, with 40 being the lowest obtained standard score. Of the 22 participants, 12 were from the neurogenetic subgroup and 10 were from the idiopathic subgroup. As described later, covariance procedures were used to adjust SRT scores for differences in intellectual status, and other analyses were completed to assess the contribution of intellectual status to SRT competence and processing scores. All participants with CAS in Table I

had prior or persistent language impairment, as documented by standard language measures, conversational language sampling analytics, and/or case history data.

Participants: speech, prosody, and voice status

Table III includes information on the speech, prosody, and voice status of participants in the four study groups, providing information on their relative competence in these domains. The speech competence information in Table III is organized by the 10-linguistic domains analytic framework (leftmost columns) described in Shriberg et al. (2010a), with subtotals provided for participants divided into younger (3–6 years) and older (7+ years) speaker groups. This age-based division provides comparative information on speaker competence during the 3–6 year period of full expression of CAS and at later ages beginning at age 7 when segmental and/or suprasegmental signs of CAS may persist, perhaps for a lifetime. The data for each speech, prosody, and voice measure are percentage scores, with higher percentage scores, indicating greater competence. The term *competence* in the methods described in Shriberg et al. (2010a) is used in its conventional sense in communicative disorders to denote or quantify relative mastery (e.g. velopharyngeal competence, articulatory competence). Two observations about the competence data in Table III obtained from conversational samples using perceptual methods (narrow phonetic transcription and prosody–voice coding) support the internal validity of scores obtained from comparison group participants.

First, the data for Group 1 speakers, generally indicating 90% or above competence on all segmental and suprasegmental competence measures, are consistent with prior reference data for some of these same measures from typical speakers within the two age groupings (Austin & Shriberg, 1996). The speech, prosody, and voice competence percentages for the two SD groups (Groups 2 and 3) are notably similar to prior epidemiologic findings for speech delay with and without language impairment. Key statistics are the findings for the Percentage of Consonants Correct (PCC) measure, which in 3- to 6-year-old children with speech delay with and without language impairment has been found to average approximately 70% with a standard deviation of approximately 10% (Shriberg, Austin, Lewis, McSweeny, & Wilson, 1997). As shown in Table III, participants in Groups 2 and 3, respectively, had PCC means and standard deviations of 76.8 (11.6) and 70.2 (14.0).

A second observation is that the competence scores of Group 4 participants in Table III were generally lower at each of the two ages than scores for the two SD speaker groups, particularly for the suprasegmental measures and within the older speakers. The three exceptions occurred for the younger age group, which included only four participants. There are too few participant scores in other cells to assess trends with inferential statistics. Descriptively, however, competence scores on the segmental measures were generally lower for participants with neurogenetic CAS backgrounds than participants with idiopathic CAS. As noted previously, possible differences in the proportion of males within the two groups and differences in competence levels could be associated with differences in genomic pathways to CAS.

The SRT

The focus of this report is on the type and informativeness of diagnostic information for CAS available from responses to nonword repetition tasks. The following sections provide methodological and conceptual details on the nonword repetition task used to address the two questions in Statement of the Problem.

Table III. Speech, prosody, and voice competence statistics obtained from the conversational speech samples of participants in the four study groups.

Speech, prosody, and voice measures	Group 1				Group 2				Group 3				Group 4							
	TSTL				SDTL				SDLI				CAS-idiopathic (CAS-I)				CAS-neurogenetic (CAS-N)			
	3–6 years ($n = 64$)		7+ years ($n = 55$)		3–6 years ($n = 119$)		7+ years ($n = 21$)		3–6 years ($n = 59$)		7+ years ($n = 11$)		3–6 years ($n = 6$)		7+ years ($n = 12$)		3–6 years ($n = 4$)		7+ years ($n = 16$)	
	M	SD	*M*	SD	*M*	SD	*M*	SD	*M*	SD	*M*	SD	*M*	SD	*M*	SD	*M*	SD	*M*	SD
Segmental																				
Vowels																				
PVC	97.2	2.5	99.3	1.5	93.3	5.2	96.7	2.6	91.2	5.4	96.9	2.9	87.2	8.4	89.2	9.1	78.5	8.4	90.9	6.5
Consonants																				
PCC	90.4	5.6	96.3	6.6	76.8	11.6	86.1	9.2	70.2	14.0	87.5	6.8	69.0	5.5	74.2	14.2	47.2	5.7	80.9	9.8
Vowels and consonants																				
PPC	93.1	4.0	97.5	4.4	83.5	8.5	90.4	6.1	78.7	10.0	91.2	4.7	76.4	5.4	80.3	11.5	59.7	2.3	84.9	7.8
II	97.5	4.0	99.0	3.2	92.5	8.3	96.5	6.4	83.5	14.2	91.6	9.7	79.8	7.9	85.0	16.0	62.7	6.9	94.9	5.9
Suprasegmental																				
Prosody																				
Phrasing	89.8	10.5	85.2	9.5	87.5	10.9	85.3	11.9	91.1	8.8	92.0	8.2	88.3	16.0	88.4	7.7	98.3	2.9	83.4	11.8
Rate	99.3	2.4	96.5	10.1	97.0	8.0	98.9	3.5	98.7	4.1	99.2	2.5	81.3	14.1	83.7	15.7	71.2	29.4	70.6	30.4
Stress	92.6	9.3	94.6	8.3	91.2	7.7	91.7	10.4	90.9	8.0	92.0	15.4	75.8	16.5	76.6	16.6	90.3	9.5	67.9	23.1
Voice																				
Loudness	93.8	13.2	96.9	6.5	94.6	10.7	97.1	8.8	89.3	15.7	96.7	4.0	100.0	0.0	99.1	1.8	98.4	2.7	98.2	3.0
Pitch	96.6	6.6	98.8	6.8	97.4	7.7	99.0	3.2	97.1	8.3	100.0	0.0	100.0	0.0	96.3	8.2	100.0	0.0	98.4	2.1
Laryngeal quality	93.2	11.5	93.0	16.1	87.4	19.7	91.8	17.4	80.4	25.1	83.0	30.2	96.3	2.3	88.4	15.8	75.3	38.7	84.1	18.0
Resonance quality	97.6	8.8	98.8	7.4	95.8	12.8	97.7	6.3	92.6	14.7	97.0	7.5	97.1	2.8	69.0	41.4	93.6	7.2	58.1	42.9

Note: PVC, Percentage of Vowels Correct; PCC, Percentage of Consonants Correct; PPC, Percentage of Phonemes Correct; II, Intelligibility Index.

Overview. As reviewed in Shriberg et al. (2009), nonword repetition tasks have been used to study lexical acquisition, to identify children with primary language impairment, and as endophenotypes in genomic studies of verbal trait disorders. The SRT (Shriberg et al., 2009) was developed primarily for the third use, specifically for speakers with mild to severe speech sound disorders. It is an 18-item task typically administered and scored in less than 5 min. Briefly, the SRT provides a means to assess the integrity of processes underlying nonword repetition in a simple context that eliminates the scoring and interpretive confound when respondents have mild to severe articulation errors. The strategy was to construct stimuli that require respondents to have phonemic mastery of only 5 of the approximately 42 speech sounds: two early acquired nasal (/m/, /n/) and stop (/b/, /d/) consonants and the back vowel /ɑ/. SRT competence scores are calculated for the four consonant sounds as they occur in 50 target syllables, yielding subscale competence percentage scores for 2-syllable, 3-syllable, and 4-syllable nonwords and a total SRT percentage correct score termed the SRT Total.

Shriberg et al. (2009) and Shriberg and Lohmeier (2008) describe the development, validation, administration, scoring procedures, and psychometric characteristics of the SRT. A recent technical report (Lohmeier & Shriberg, 2011) provides tabular and graphic reference data on 23 SRT competence and speech processing scores obtained from several cohorts of children and youth and processed with computer routines. The appendix includes the 18 SRT stimulus items and annotations for the three reference citations.

Competence score. Studies using the SRT have indicated that in addition to overall and subscale speech competence scores (i.e. total percentage of correctly repeated consonant sounds and subscales percentages for the 2-, 3-, and 4-syllable items), the errors participants make on each item can be used to index deficits in speech processing. Using the framework shown in Figure 1, metrics have been developed to quantify respondents' *encoding*, *memory*, and *transcoding* processes. *Execution* processes, which are relevant for the neuromotor deficits in dysarthria, are beyond the scope of the present focus on apraxia of speech.

Encoding processes. As noted previously, children's and adults' responses to nonword repetition tasks reflect variance associated with their short-term representations of the nonword stimuli. Correct representations require, in addition to adequate peripheral hearing, accurate auditory-perceptual encoding of the salient features of each speech sound in the stimulus. There are numerous ways in which representations at segmental (e.g. place features) and suprasegmental (e.g. lexical stress) tiers can be missing, incomplete, or incorrect (see Ziegler, Staiger, & Aichert, 2010). Using only the information available from responses to the SRT, a metric termed the *Percentage of Within-Class Manner Substitutions* was created to quantify nonword repetition errors due to processing deficits in auditory-perceptual encoding of segmental features. The denominator for this metric is the number of a respondent's substitution errors for the 50 target consonants in the SRT (see the appendix). The numerator is the number of consonant substitutions with the same manner class as the target phoneme (i.e. a nasal for a nasal or a stop for a stop). Within-class manner, errors were posited to reflect at least partial representation (i.e. knowledge) of the SRT target sound, compared to out-of-class substitutions, which do not indicate partial knowledge. Preliminary analyses indicated that across the three SRT nonword syllable lengths, the mean percentage of the within-class manner substitutions occurring in the 3- and 4-syllable words provided the most sensitive of several pilot metrics of the encoding construct, including the alternative of within-place substitutions. Thus, higher scores on the encoding measure indicate higher competence in correctly encoding the manner features of SRT sound targets replaced by another sound.

Memory processes. The controversial topic of the type and relative contribution of memory processes in nonword repetition task responses is beyond the scope of the present report (Shriberg et al., 2009). Essentially, many to most investigators appear to view nonword repetition task responses as indexing the capacity of a participant's phonological loop because respondents repeat shorter words more accurately than longer words. In prior work, however, we found 38 of 156 (24.4%) participants had scores ranging from 0% to 50% correct on the 2-syllable SRT stimuli. Thus, a nontrivial percentage of children in Shriberg et al. (2009), approximately one-quarter of the sample, had difficulty in correctly repeating nonwords that required storage and retrieval of only two consonant singletons that they did not misarticulate.

The majority of these participants (33 of the 38 children, 87%) had speech delay, including 16 (42.7%) with speech delay and typical language, and 17 (44.7%) with speech delay and expressive language disorder. We suggested that at least for some children, failure to repeat consonant sounds correctly in simple CVCV forms is likely due to deficits in auditory-perceptual encoding, rather than in memory processes.

A series of analyses in the development of the SRT-assessed alternative procedures to quantify the relative contribution of memory processes to SRT scores. The most sensitive metric that emerged were ratios of the percentage of sounds correct within longer compared to shorter syllable-length items. Specifically, for each participant, the greater the difference in his or her scores on items with longer compared to shorter syllable lengths, the greater the contribution of memory to the total SRT competence score. In contrast, the SRT competence scores of respondents with lower ratios may be more associated with encoding and/or transcoding. Of the three possible ratios (4-syllable: 3-syllable, 4-syllable: 2-syllable, 3-syllable: 2-syllable), the most sensitive ratio in pilot analyses was the 3-syllable: 2-syllable ratio. All the memory processing SRT scores in this report use this ratio, although data for the 2-, 3-, and 4-syllable items are presented for completeness in several tables. To adjust for the wide range of obtained 3-syllable to 2-syllable ratios, the obtained ratios were transformed using their natural log. Also, to provide a more transparent score that would be directionally comparable to the percentage scores for other measures, these scaled log scores were transformed using the formula 100 * (1 + log value). Resulting scores below 0 and above 100 were truncated to each of these values, respectively, to avoid negative algebraic signs and scores above 100 for individual and between-group analyses. Thus, lower SRT memory scores in the forthcoming analyses indicate greater difficulty in accurately repeating 3-syllable compared to 2-syllable SRT items relative to typical speakers of the same age and gender.

Transcoding processes. Analyses of SRT competence errors indicated that many respondents added a sound to a correctly or incorrectly produced target sound (e.g. [bandɑ] for /bɑdɑ/). Most scoring systems for nonword repetition tasks score a response as correct as long as the correct sound was produced adjacent to the addition, but have some convention to keep track of the frequency of such sound additions (e.g. Dollaghan & Campbell, 1998). The SRT competence scores follow the same convention with all additions to correct sounds ignored in the scoring system (Shriberg & Lohmeier, 2008).

For the purposes of the present study, sound additions were viewed as consistent with the construct of an error in transcoding representations to speech. Essentially, they were viewed as plausibly reflective of planning/programming deficits in AOS and CAS (versus deficits in encoding or memory processes), consistent with the construct of atypical distortions. Transcoding scores were derived from the narrow phonetic transcription by utilities in the analyses software to be described. Transcoding scores were defined as the percentages of responses to

each of the 18 SRT items that contained one or more additions, with these scores subtracted from 100 for directional clarity (i.e. to make low scores denote less competent transcoding).

Data reduction

Experienced research transcriptionists used a system of narrow phonetic transcription symbols and conventions to transcribe responses to all speech tasks administered in the research protocols at each site and completed prosody–voice coding of the conversational speech samples. Procedural details for narrow phonetic transcription, prosody–voice coding, and formatting transcripts for analyses in the PEPPER environment (Shriberg et al., 2001) have been reported in prior papers and are described in detail in an unpublished laboratory manual (Shriberg, Hersh, et al., 2008). Shriberg et al. (2010b) includes extensive reliability data for narrow transcription and prosody–voice coding of speakers with SD and MSD, including samples from some of the participants with CAS in the present study. Point-to-point percentage of agreement findings for the two perceptual methods were as high or higher than reported in literature reviews and comparable to previous studies conducted using these methods. It is important to note that although narrow transcription is used for all other speech production tasks, broad phonetic transcription is sufficient for scoring responses to the SRT.

Associations among SRT measures

Table IV is a summary of Pearson correlation coefficients and coefficients of determination (r^2) for the four SRT scores. These associations were completed to determine the degree of collinearity among the processing scores and to assess the associations between SRT competence and each of the processing scores. The lower the statistical associations among processing scores, the stronger the interpretation of them as indexing independent constructs.

First order, nonpartialled correlations were obtained on all SRT scores unadjusted for age or gender. As the interest is in conceptual associations among the measures, rather than the statistical significance of the coefficients, focus is on the r^2 values as indices of shared variance. Bolded r^2 values indicate bivariate associations in the same direction with greater than 10% common variance, which for the cells sizes available to compute each coefficient also generally meet the conventional 0.05 level of significance.

Findings in the first three rows in Table IV indicate that the three SRT processing constructs are essentially unassociated with one another. None of the 12 r^2 values for the four study groups

Table IV. Correlational analyses of all SRT measures for each of the four study groups.

	Group 1: TSTL		Group 2: SDTL		Group 3: SDLI		Group 4: CAS		All groups	
SRT measures	*r*	r^2 (%)	*r*	r^2 (%)	*r*	r^2 (%)	*r*	r^2 (%)	*r*	r^2 (%)
Encoding–memory	0.20	4	0.26	7	0.19	4	−0.01	<1	0.247	6
Encoding–transcoding	−0.01	<1	0.10	1	−0.02	<1	−0.10	1	0.114	1
Memory–transcoding	0.22	5	0.17	3	−0.11	1	0.25	6	0.217	5
Competence–memory	0.59	**35**	0.59	**35**	0.60	**36**	0.47	**22**	0.624	**39**
Competence–encoding	0.36	**13**	0.43	**18**	0.36	**13**	0.07	<1	0.409	**17**
Competence–transcoding	0.46	**21**	0.22	5	−0.06	<1	0.36	**13**	0.338	**11**

Note: r^2 greater than 10% are bolded.

or the three values across all groups (rightmost column) met the arbitrary criteria of greater than 10% shared variance. Thus, the three SRT speech processing constructs are interpreted as essentially orthogonal to one another, with none sharing more than 7% common variance.

The last three rows in Table IV provide information on the strength of associations between SRT competence scores and the three processing scores. The r^2 percentages in the rightmost column in Table IV indicate that the strongest associations, respectively, are competence with memory (39%), encoding (17%), and transcoding (11%) processes. The r^2 values for SRT competence with encoding met the greater than 10% shared variance criterion for each of the three comparison groups (Groups 1, 2, and 3), but not for findings from Group 4 participants. Also, the association between SRT competence and transcoding scores did not reach the r^2 criteria for the two speech groups (Groups 2 and 3). Overall, these findings indicated that memory, encoding, and transcoding scores, respectively, are associated with SRT competence scores, with differences in the amount of shared variance associated with participants' speech–language classification.

SRT scores and speech–prosody scores analyses

Associations between SRT measures and speech and prosody measures. Table V includes Pearson coefficients and r^2 values, indicating the association of each of the four SRT scores with measures of speech and prosody competence. There are no entries in Table V for Group 1 participants because they had typical speech, and there are no Group 2 and Group 3 entries for the prosody measures because these participants did not frequently have prosodic errors in conversational speech (Table III). As in Table IV, r^2 values exceeding 10% shared variance are bolded.

Fourteen of the 72 r^2 values in Table V exceed the criterion of greater than the 10% shared variance (i.e. excluding the two coefficients in which measures were negatively correlated). As shown in the first three rows, 11 of the 14 associations indicate that SRT competence scores shared greater than 10% common variance with each of the segmental speech variables – Percentage of Vowels Correct (PVC), PCC, and intelligibility index (II). Another bolded coefficient indicated greater than 10% common variance between memory and II scores for Group 3 participants. Thus, with the exception of the lack of criterial association between PVC scores and SRT competence scores in Group 2, SRT competence was mildly to moderately associated with speech and prosody competence scores, with the largest shared variance for participants in Group 4. The other two associations meeting criteria in Table V indicated that among all of the 250 participants with speech delay (rightmost column), their transcoding scores were mildly correlated with their percentage of errors on the PVC and PCC.

Results

(1) *Are study findings consistent with a transcoding account of CAS in neurogenetic and idiopathic contexts, or are they more consistent with a multiple domain account with deficits in encoding, memory, and transcoding processes?*

Between-group comparisons

Table VI is a summary of pair-wise comparisons among the four participant groups on the four SRT scores, including descriptive statistics (centre section) and effect size statistics (right section). Descriptive findings include the unadjusted (raw) means and standard deviations and three statistics adjusted for the covariates of age and gender (marginal means,

Table V. Correlational analyses of SRT measures and speech and prosody measures for participants in Groups 2, 3, and 4.[a]

	Group 2: SDTL		Group 3: SDLI		Group 4: CAS		All groups	
SRT and speech–prosody measures	*r*	r^2 (%)	*r*	r^2 (%)	*r*	r^2 (%)	*r*	r^2 (%)
SRT competence with:								
PVC	0.21	4	0.35	**12**	0.50	**25**	0.43	**19**
PCC	0.36	**13**	0.55	**30**	0.53	**28**	0.54	**29**
II	0.41	**17**	0.44	**19**	0.57	**32**	0.51	**26**
Phrasing	–	–	–	–	−0.21	4	−0.12	1
Rate	–	–	–	–	−0.03	<1	0.11	1
Stress	–	–	–	–	−0.08	<1	0.12	1
SRT encoding with:								
PVC	0.18	3	0.31	10	0.12	1	0.25	6
PCC	0.23	5	0.23	5	0.16	3	0.25	6
II	0.29	8	0.12	1	0.08	<1	0.22	5
Phrasing	–	–	–	–	−0.01	<1	−0.09	<1
Rate	–	–	–	–	0.08	<1	0.09	<1
Stress	–	–	–	–	−0.05	<1	0.03	<1
SRT memory with:								
PVC	0.11	1	0.02	<1	−0.04	<1	0.21	4
PCC	0.11	1	0.09	<1	0.06	<1	0.24	6
II	0.10	1	0.33	**11**	0.08	<1	0.29	8
Phrasing	–	–	–	–	−0.07	<1	−0.07	<1
Rate	–	–	–	–	−0.34	12	0.03	<1
Stress	–	–	–	–	−0.01	<1	0.13	2
SRT transcoding with:								
PVC	0.31	10	−0.01	<1	0.29	8	0.40	**16**
PCC	0.31	10	−0.03	<1	0.28	8	0.35	**12**
II	0.09	<1	−0.06	<1	0.24	6	0.26	7
Phrasing	–	–	–	–	−0.33	11	−0.05	<1
Rate	–	–	–	–	−0.10	1	0.26	7
Stress	–	–	–	–	−0.18	3	0.20	4

Note: PVC, Percentage of Vowels Correct; PCC, Percentage of Consonants Correct; II, Intelligibility Index.
[a]Group 1: TSTL participants did not have appreciable variance on the speech and prosody measures. Group 2: SDTL and Group 3: SDLI did not have appreciable variance on the prosody measures.

standard errors, and standard deviations). The effect size findings for each of the four SRT scores include six pair-wise comparisons of the four study groups with one another. Negative effect sizes indicate that the second group in each pair-wise comparison had lower average scores adjusted for age and gender than the first group. One-tailed confidence intervals could be motivated, but to provide for more conservative tests treated family-wise, all significance tests in Table VI and elsewhere are two-tailed. Significant effect sizes are indicated with the conventional asterisk, with the adjectives for each significant comparison, scaling the relative magnitude of the effect size (Cohen, 1988). For ease of reading in the present and additional tables, effect size information is bolded for each of the comparisons of Group 4 with each of the other three groups. The descriptive and inferential statistical findings in Table VI can be summarized as follows.

First, the descriptive statistics within each of the four sets of SRT scores are consistent with the speech–language competence data in Table III. For each of the four sets of SRT

Table VI. Summary of the pair-wise comparisons for the four participant groups on the SRT scores.

	Group			Descriptive statistics					Effect size statistics				
				Unadjusted		Adjusted for covariates							
SRT Variable	No.	Speech–language status[a]	*n*	M	SD	M	SE	SD	Comparison	ES	Confidence interval	Significance[b]	Adjective[c]
Competence	1	TSTL	119	83.8	13.6	83.3	1.36	14.84	1–2	−0.40	−0.64, −0.15	*	S
	2	SDTL	138	76.1	15.4	77.4	1.27	14.92	1–3	−1.13	−1.45, −0.81	*	V
	3	SDLI	68	64.8	19.6	66.4	1.81	14.93	2–3	−0.73	−1.03, −0.44	*	M
	4	CAS	35	64.6	16.4	58.6	2.52	14.91	**1–4**	−1.65	−2.07, −1.23	*	V
									2–4	−1.25	−1.65, −0.86	*	V
									3–4	−0.52	−0.93, −0.10	*	M
Encoding	1	TSTL	109	63.4	28.6	64.3	2.38	24.85	1–2	−0.22	−0.48, 0.03		
	2	SDTL	133	58.3	24.1	58.7	2.15	24.80	1–3	−0.56	−0.87, −0.25	*	M
	3	SDLI	66	50.3	24.0	50.3	3.06	24.86	2–3	−0.34	−0.63, −0.04	*	S
	4	CAS	40	45.3	16.8	41.3	4.08	24.82	**1–4**	−0.92	−1.30, −0.54	*	L
									2–4	−0.70	−1.06, −0.34	*	M
									3–4	−0.36	−0.76, 0.04		
Memory	1	TSTL	119	87.2	22.2	87.8	2.70	29.45	1–2	−0.28	−0.53, −0.04	*	S
	2	SDTL	138	77.7	28.5	79.4	2.50	29.37	1–3	−0.67	−0.98, −0.36	*	M
	3	SDLI	68	66.3	42.8	68.0	3.57	29.44	2–3	−0.39	−0.68, −0.09	*	S
	4	CAS	38	60.1	30.1	47.4	4.98	29.46	**1–4**	−1.36	−1.76, −0.97	*	V
									2–4	−1.08	−1.46, −0.71	*	V
									3–4	−0.69	−1.10, −0.29	*	M

Transcoding	1	TSTL	119	94.4	9.4	93.9	1.19	12.98	1–2	−0.32	−0.57, −0.08	*	S
	2	SDTL	138	88.1	13.6	89.7	1.10	12.92	1–3	−0.51	−0.82, −0.21	*	M
	3	SDLI	68	85.1	14.9	87.2	1.57	12.95	2–3	−0.19	−0.48, 0.10		
	4	CAS	38	68.1	21.2	57.8	2.19	12.96	**1–4**	−2.77	−3.24, −2.29	*	E
									2–4	−2.46	−2.90, −2.01	*	E
									3–4	−2.25	−2.75, −1.75	*	E

Note: The means and standard deviations adjusted for the covariates of age, gender, and IQ were used in the effect size calculations.

[a]The following abbreviations are used for the speech–language groups: TSTL, Typical Speech–Typical Language; SDTL, Speech Delay–Typical Language; SDLI, Speech Delay–Language Impairment; CAS, Childhood Apraxia of Speech.

[b]Significant Hedges' corrected effect sizes are indicated by an asterisk.

[c]Effect size adjective abbreviations, adopted from Cohen (1988) and extended, are >0.2, Small (S); >0.5, Moderate (M); >0.8, Large (L); >1, Very large (V); >2, Extremely large (E).

Table VII. Diagnostic accuracy findings for the SRT competence and processing measures[a].

Pair-wise comparisons[b]	Cut-off percent[c]	Sensitivity	Specificity	Diagnostic accuracy	Predictive value of a positive test	Predictive value of a negative test	Likelihood ratio of a positive test	Likelihood ratio of a negative test	Odds ratio
Competence	65.0								
1–2		23.2	91.6	54.9	76.2	50.7	2.8	1.2	3.3
1–3		44.1	91.6	74.3	75.0	74.2	5.3	1.6	8.6
2–3		44.1	76.8	66.0	48.4	73.6	1.9	1.4	2.6
1–4		**52.6**	**91.6**	**82.2**	**66.7**	**85.8**	**6.3**	**1.9**	**12.1**
2–4		**52.6**	**76.8**	**71.6**	**38.5**	**85.5**	**2.3**	**1.6**	**3.7**
3–4		**52.6**	**55.9**	**54.7**	**40.0**	**67.9**	**1.2**	**1.2**	**1.4**
Encoding	46.9								
1–2		35.3	72.5	52.1	61.0	47.9	1.3	1.1	1.4
1–3		48.5	72.5	63.4	51.6	69.9	1.8	1.4	2.5
2–3		48.5	64.7	59.3	40.5	71.7	1.4	1.3	1.7
1–4		**55.0**	**72.5**	**67.8**	**42.3**	**81.4**	**1.9**	**1.6**	**3.2**
2–4		**55.0**	**64.7**	**62.4**	**31.9**	**82.7**	**1.6**	**1.4**	**2.2**
3–4		**55.0**	**51.5**	**52.8**	**40.7**	**65.4**	**1.1**	**1.1**	**1.3**
Memory	67.5								
1–2		29.7	86.6	56.0	71.9	51.5	2.2	1.2	2.7
1–3		48.5	86.6	72.7	67.4	74.6	3.6	1.7	6.1
2–3		48.5	70.3	63.1	44.6	73.5	1.6	1.4	2.2
1–4		**55.3**	**86.6**	**79.0**	**56.8**	**85.8**	**4.1**	**1.9**	**7.9**
2–4		**55.3**	**70.3**	**67.1**	**33.9**	**85.1**	**1.9**	**1.6**	**2.9**
3–4		**55.3**	**51.5**	**52.8**	**38.9**	**67.3**	**1.1**	**1.2**	**1.3**
Transcoding	80.0								

1–2	20.3	90.8	52.9	71.2	49.5	2.2	1.1	2.5
1–3	35.3	90.8	70.6	68.6	71.1	3.8	1.4	5.4
2–3	35.3	79.7	65.1	46.2	71.4	1.7	1.2	2.1
1–4	**73.7**	**90.8**	**86.6**	**71.8**	**91.5**	**7.9**	**3.4**	**27.5**
2–4	**73.9**	**79.7**	**78.4**	**50.0**	**91.7**	**3.6**	**3.0**	**11.0**
3–4	**73.7**	**64.7**	**67.9**	**53.9**	**81.5**	**2.1**	**2.5**	**5.1**
2/3–4	**73.7**	**74.8**	**74.6**	**34.0**	**93.9**	**2.9**	**2.8**	**8.3**

Note: See rationale in the text for the combined pair-wise comparison in the last row.

[a]See Kraemer (1992) and MacKinnon (2000) for details on the diagnostic accuracy statistics.

[b]The groups are as follows: 1, TSTL; 2, SDTL; 3, SDLI; 4 = CAS; and 2/3 = SDTL combined with SDLI.

[c]The cut-off percent in the first row of each set of findings was used for all pair-wise comparisons in the set. It was derived from the pair-wise 3–4 comparison (i.e. SDLI–CAS comparison) within each of the four sets of SRT score comparisons because the participants in Group 3: SDLI and Group 4: CAS have the closest speech–language status.

scores, Group 1 participants had the highest SRT competence and processing scores and Group 4 participants had the lowest SRT competence and process scores, with the two intermediate groups' scores in each comparison (Groups 2 and 3), lower for Group 3 participants. The standard errors of the adjusted means scores indicate that the estimated means for each SRT measure are reliable within less than 1 to approximately 5 percentage points.

Second, for the bolded effect size comparisons in Table VI, Group 4 participants had significantly lower SRT scores than comparison group participants on 11 of the 12 pair-wise comparisons. All SRT score comparisons between Group 4 and Group 1 participants were significant, with the lower scores of Group 4 participants associated with large (>0.80) to extremely large (>2.0) effect sizes. All Group 4 and Group 2 comparisons were also significant, with effect sizes ranging from moderate to very large. The only Group 4 comparison with a nonsignificant two-tailed effect size was on encoding, with Group 4 participants' scores not significantly lower than the average scores of the Group 3 participants.

Third, and central for the question posed, the most discriminating SRT processing measures for Group 4 participants were their transcoding scores. As shown in Table VI, these participants' adjusted mean transcoding score of 63% was associated with the three largest effect sizes of the 24 pair-wise comparisons. Specifically, comparisons with the average-adjusted scores of Group 1 (93.4%), Group 2 (89.3%), and Group 3 (86.8%) yielded significant effect sizes, respectively, of −2.25, −1.93, and −1.74.

Fourth, although not as strongly discriminative as transcoding scores, the effect size data in Table VI also indicate that participants in Group 4 had significantly lower adjusted mean SRT competence, encoding, and memory scores than at least one of the three comparison groups. In comparison with Group 1 participants, Group 4 participants had significantly lower scores on all three SRT measures, with effect sizes, respectively, of −1.71, −0.94, and −1.23. For the comparisons with participants in Groups 2 and 3, Group 4 had significantly lower SRT competence and memory scores. Group 4 participants' average encoding scores were significantly lower than Group 2 participants' scores, but were not significantly lower than Group 3 scores.

Finally, findings from the present large data set replicate and extend findings reported in Shriberg et al. (2009), indicating that the SRT scores discriminate speakers with speech–language impairment or speech delay alone from speakers with typical speech and language. As shown in Table VI, participants with both speech and language impairment (Group 3) had significantly lower scores on all four SRT measures than participants with speech delay and typical language (Group 2) and participants with typical speech and language (Group 1). Finally, participants with speech delay alone (Group 2) had significantly lower competence, memory, and transcoding scores than participants in Group 1, although effect sizes were small.

(2) *Do findings from SRT competence and processing measures contribute significant or conclusive diagnostic information to an assessment battery for CAS?*

Diagnostic accuracy analyses

The second question addressed in this study was to estimate the diagnostic accuracy of SRT competence and processing scores at the level of individual speakers. For the research and clinical needs discussed at the outset of this paper, the specific goal was to determine how well any of the SRT processing measures identified participants with CAS and excluded participants with typical speech or speech delay.

Table VII is a summary of the diagnostic accuracy statistics (Kraemer, 1992) obtained for each of the four SRT measures using software by MacKinnon (2000). The focus is on

findings for the CAS speakers (bolded) in comparison with speakers in each of the three comparison groups for continuing studies of the SRT in genetic and other research contexts. The cut-off score for each of the SRT measures in Table VII was the value that best discriminated participants in Group 4 from participants in Group 3, with the latter group closest in severity of involvement to participants with CAS. As shown in Table VII, that cut-off value was used in each of the six pair-wise comparisons. To increase statistical power and generalization to participants with Speech Delay irrespective of their language status, an additional pair-wise comparison was computed that combined Groups 2 and 3 (Table VII; bottom row). The diagnostic accuracy findings in Table VII can be summarized as follows.

First, of the four SRT measures, the SRT transcoding scores had the highest diagnostic accuracy (cut-point set at 80%) in discriminating participants meeting the criteria listed in Table II for CAS. Findings in the last row of Table VII indicated that transcoding scores were 74.6% accurate in discriminating CAS speakers from speakers with speech delay with or without language impairment (73.7% sensitivity; 74.8% specificity). These sensitivity and specificity values were not sufficient to produce the positive and negative likelihood ratios required for conclusive markers described previously. Note in Table VII, however, that values for the predictive value of a negative test (93.9%) and odds ratio (8.3%) were both high. The predictive value of a negative test finding in the present study indicates that an SRT transcoding score above 80% ruled out CAS with 93.9% accuracy. The odds ratio indicates that SRT transcoding scores of <80% were 8.3 times more likely from participants with CAS than from the participants with Speech Delay.

A second observation concerns the diagnostic accuracy of the SRT competence measure and the three processing measures among the three speech–language comparison groups. The present diagnostic accuracy findings for the SRT competence scores are consistent with those reported in Shriberg et al. (2009), providing statistical support for SRT competence scores to discriminate children with both speech and language impairment from children with typical speech–language acquisition. Specifically, the present findings indicated the competence, memory, and transcoding scores had at least 70% diagnostic accuracy in discriminating participants with speech–language impairment (Group 3) from participants with typical speech–language (Group 1). None of the SRT measures had high diagnostic accuracy in discriminating participants in Group 3 from participants in Group 2.

Figure 2 is a graphic summary of the diagnostic accuracy findings. The dashed horizontal lines in each of the four panels are the cut-off percentages for each of the SRT measures that best discriminated Group 4 participants from Group 3 participants on each of the four measures (see Table VII). The boxplots in each of the four panels include the median for each of the four SRT measures, the interquartile range, values higher and lower than 1.5 times the interquartile range (upper and lower "whiskers"), and values above and below the whiskers (i.e. outliers (asterisks) above and below 1.5 units from the interquartile range). The whiskers and outliers in each of the other three groups overlapping the transcoding scores of Group 4 participants (lower right panel) illustrate the specificity constraint just reviewed. Notice that even some participants with typical speech–language (Group 1) had transcoding scores in the range of participants with CAS (Group 4).

Transcoding analyses

An analysis series assessed whether CAS participants with low transcoding scores differed from those with high scores on one or more demographic, SRT, speech, or prosody variables, findings for any of which might be informative for descriptive–explanatory accounts of CAS. Participants in the four study groups were divided into low and high transcoding subgroups,

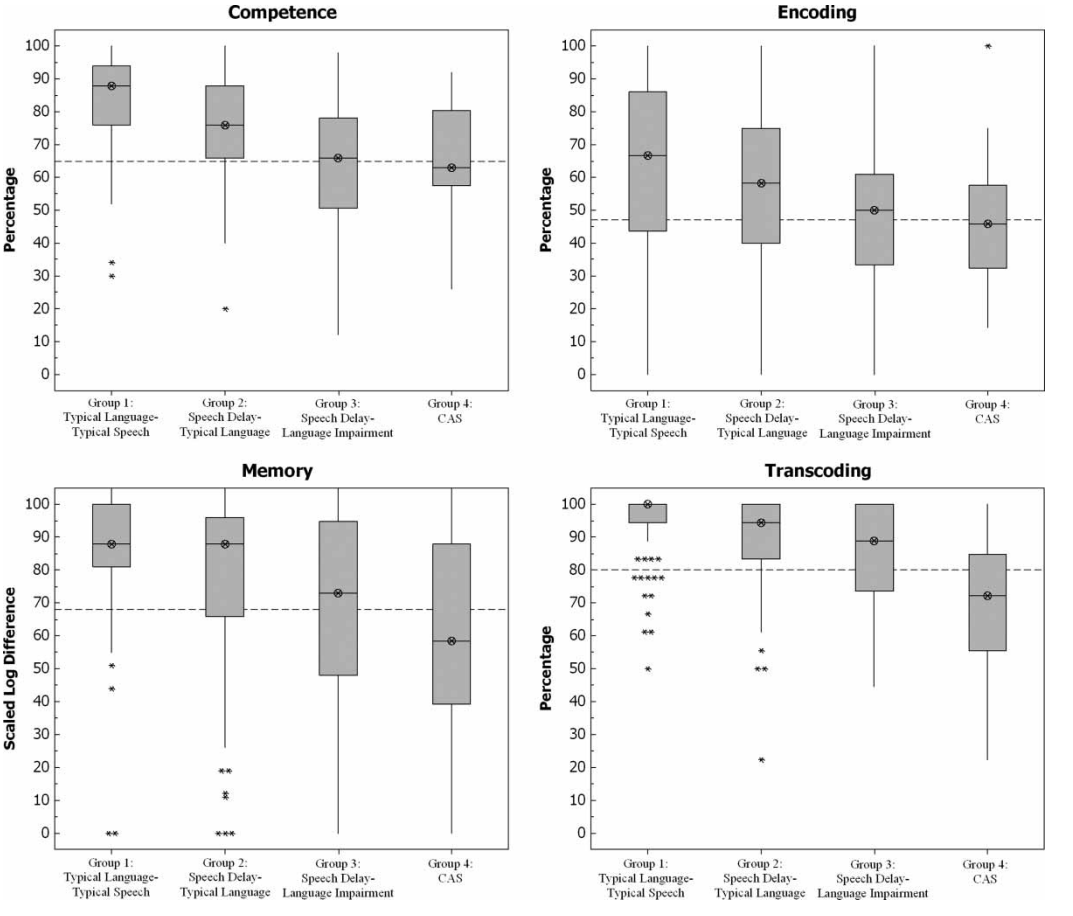

Figure 2. Graphic summary of the diagnostic accuracy findings. See text for description of the boxplot displays and the cut-off percentages indicated by the horizontal dashed lines.

using the 80% score cut-off described previously. Effect sizes were computed to determine significant means differences between subgroups. Table VIII includes findings for each of the four study groups.

Group 1. Findings from Group 1 were considered to be of central importance to understanding developmental perspectives on transcoding (i.e. planning/programming) processes. As shown in Table VIII, of the 119 participants ages 3–22 years with TSTL (Table I), the 11 participants with low transcoding scores were significantly younger (by approximately 2 years) than the remaining 108 participants whose transcoding scores were above 80%. Of the remaining variables in Table VIII, Group 1 participants with low transcoding scores also had significantly lower SRT competence scores than participants with high transcoding scores (averaging 13.5 percentage points lower), lower transcoding scores (averaging 26.7 percentage points lower), and lower PCC scores (averaging 4.4 percentage points lower). As discussed later, these findings are interpreted in the context of developmental aspects of speech motor planning/programming as indexed in the present study by the percentage of speech sound additions in nonword repetition responses.

Table VIII. Pair-wise analyses of variables potentially associated with high versus low SRT transcoding scores.

	High transcoding			Low transcoding			Comparative statistics				High transcoding			Low transcoding			Comparative statistics			
Variables	*n*	*M*	SD	*n*	*M*	SD	ES	CI	Significant[a]	Adj.[b]	*n*	*M*	SD	*n*	*M*	SD	ES	CI	Significant	Adj.
Group 1: TSTL											Group 2: SDTL									
Demographic																				
Age (years)	108	6.1	2.4	11	4.2	1.4	−0.82	−1.45, −0.19	*	L	112	5.0	1.4	28	4.4	1.0	−0.42	−0.84, −0.01	*	S
Gender (% male)		34.3%		54.5%			0.41	−0.69,1.55				67.0%			67.9%		0.19	−0.94, 0.90		
Background (% idiopathic)																				
SRT																				
Competence	108	85.0	13.3	11	71.5	11.3	−1.03	−1.66, −0.40	*	V	110	77.4	15.8	28	70.8	12.5	−0.43	−0.85, −0.02	*	S
Encoding	98	63.3	29.7	11	64.6	15.8	0.05	−0.58, 0.67			106	59.5	25.7	27	53.3	15.9	−0.26	−0.68, 0.17		
Memory	108	86.8	23.0	11	91.1	10.6	0.19	−0.43, 0.81			110	79.3	29.1	28	71.4	25.6	−0.28	−0.69, 0.14		
Transcoding	108	96.9	4.8	11	70.2	9.4	−4.96	−5.84, − 4.07	*	E	110	93.7	6.2	28	66.3	12.6	−3.45	−4.03, − 2.87	*	E
Speech																				
PVC	108	98.3	2.3	11	97.4	3.3	−0.35	−0.97, 0.27			112	94.5	4.2	28	91.1	7.1	−0.70	−1.12, −0.28	*	M
PCC	108	93.6	6.7	11	89.2	6.2	−0.65	−1.28, −0.03	*	M	112	79.7	11.3	28	72.3	11.9	−0.65	−1.07, −0.23	*	M
II	108	98.3	3.9	11	97.4	1.6	−0.23	−0.85, 0.39			112	93.4	8.2	28	91.9	8.1	−0.19	−0.60, 0.23		
Prosody																				
Phrasing	108	87.4	10.4	11	91.3	8.8	0.38	−0.24, 1.00			112	87.2	10.9	28	87.0	11.6	−0.01	−0.43, 0.40		
Rate	108	98.4	6.1	11	95.1	13.8	−0.46	−1.08, 0.16			112	97.8	5.6	28	95.1	12.4	−0.36	−0.78, 0.06		
Stress	108	93.5	9.1	11	93.9	6.0	0.05	−0.57, 0.67			112	91.3	8.4	28	91.1	7.0	−0.04	−0.45, 0.38		
Group 3: SDLI											Group 4: CAS									
Demographic																				
Age (years)	46	4.6	2.0	24	4.6	1.0	−0.03	−0.52, 0.47			11	12.5	6.2	27	10.7	8.4	−0.21	−0.92, 0.49		
Gender (% male)		67.4%			91.7%		0.63	0.35, 1.41	*	M		63.6%			59.3%		−0.90	−1.29, 1.19		
Background (% idiopathic)											2	18.2%		16	59.3%		0.88	0.40, 1.90	*	L
SRT																				
Competence	44	63.5	21.4	24	67.2	16.1	0.18	−0.31, 0.68			10	74.0	18.1	28	61.3	14.7	−0.80	−1.54, −0.06	*	L
Encoding	43	50.2	26.8	23	50.7	17.9	0.02	−0.49, 0.53			12	36.8	13.4	28	49.0	17.0	0.74	0.05, 1.44	*	M
Memory	44	60.5	48.9	24	76.8	26.5	0.38	−0.12, 0.88			10	71.8	19.5	28	55.9	32.4	−0.52	−1.26, 0.21		
Transcoding	44	94.7	5.4	24	67.6	10.1	−3.63	−4.41, − 2.84	*	E	10	92.8	5.8	28	59.3	17.3	−2.15	−3.02, − 1.28	*	E
Speech																				
PVC	46	92.0	5.9	24	92.4	4.8	0.06	−0.43, 0.56			11	91.7	6.8	27	87.2	8.7	−0.54	−1.25, 0.17		
PCC	46	72.6	15.5	24	73.7	12.9	0.08	−0.42, 0.57			11	77.6	17.9	27	71.7	12.8	−0.41	−1.11, 0.30		
II	46	84.1	14.5	24	86.2	12.6	0.15	−0.34, 0.64			11	90.9	13.1	27	84.0	14.4	−0.49	−1.20, 0.22		
Prosody																				
Phrasing	46	93.3	7.5	24	87.4	9.5	−0.71	−1.23, −0.19	*	M	11	84.3	11.7	27	87.8	11.5	0.30	−0.47, 1.07		
Rate	46	99.1	2.5	24	98.2	5.6	−0.24	−0.75, 0.27			11	77.2	25.7	27	75.3	24.9	−0.08	−0.84, 0.69		
Stress	46	91.4	10.3	24	90.6	7.7	−0.08	−0.58, 0.42			11	69.6	30.7	27	75.0	15.1	0.26	−0.51, 1.03		

[a]Significant Hedges' corrected effect sizes are indicated by an asterisk.
[b]Effect size adjective abbreviations, adopted from Cohen (1988) and extended, are >0.2, Small (S); >0.5, Moderate (M); >0.8, Large (L); >1, Very large (V); >2, Extremely large (E).

Groups 2 and 3. The Groups 2 and 3 transcoding analyses indicated, as expected, that transcoding scores for the low subgroups were significantly lower than those of the high transcoding subgroups. Of importance for descriptive–explanatory accounts of CAS, the average SRT transcoding scores of the Group 2 low scoring subgroup ($n = 28$; mean transcoding score of 66.3%) and Group 3 low scoring subgroup ($n = 24$; mean transcoding score of 67.6%) were within a standard deviation of the Group 4 low transcoding subgroup ($n = 28$; mean transcoding score of 59.3%). Thus, as discussed later, although proportionally more CAS participants had low transcoding scores, some participants within each of the three comparison groups had scores in the same range. As shown in Table VIII, Group 2 participants in the low transcoding subgroup were significantly younger than those in the high subgroup, had significantly lower SRT competence scores, and significantly lower PVC and PCC scores. Also as shown in Table VIII, Group 3 participants with low transcoding scores included a significantly higher proportion of males than females than in the high transcoding scores group and had significantly lower mean phrasing scores.

Group 4. In addition to their significantly lower mean transcoding scores, participants in the Group 4 low subgroup differed significantly from the CAS participants with high transcoding scores on three measures. Significantly more participants with low transcoding scores had idiopathic CAS (59.3%) compared to participants in the high transcoding scores subgroup (18.2%). Although the significant between-group difference suggests that the context for CAS may be important for eventual descriptive–explanatory differences, the small number of participants in each group prohibits additional analyses by subgroup. Also, CAS participants with low compared to high transcoding scores had significantly lower average SRT competence scores (61.3% compared to 74.0%), but had higher encoding scores (49.0% compared to 36.8%). This last finding seems to be due to the low encoding scores (36.8%) of the 12 Group 4 participants with high transcoding scores, a value that is lower than the values of all other entries for this measure in Table VIII and possibly a sampling error.

Finally, to determine whether intellectual status was associated with transcoding and each of the other three SRT measures, analyses were completed using a standard score of <85 as the cut-off point for low cognitive status. Participants in the low and typical intellectual status groups were examined relative to their scores on each of the four SRT measures, using the cut-off points for high and low scores on each of the measures shown in Table VII (i.e. cut-off points for competence = 65%; encoding = 46.9%; memory = 67.5%; and transcoding = 80%). The percentages of participants with low and typical intellectual status were approximately equal for each of the four comparisons, with none of the four comparisons statistically significant on chi-square tests for independent samples (0.05 alpha level). Specific to transcoding, for example, of eight Group 4: CAS participants with high (>80%) transcoding scores who completed intellectual testing, three (37.5%) had IQ within normal limits (>85). Of the 27 participants with low transcoding scores (<80%) and available IQ scores, 11 (40.7%) had IQ scores within normal limits. Thus, for the participants with CAS in the present study divided into IQ subgroups within and below the normal limits, intellectual status was not associated with performance on any of the four SRT measures.

Additions analyses

The final diagnostic analyses series focused on characteristics of the speech sound additions occurring before consonants in participants with CAS compared to participants in the other

three study groups. As described previously, additions to correctly produced target consonants were not scored as errors in the SRT scoring procedures, much as they are not considered errors in other nonword repetition tasks. We completed two analyses of the phonetically transcribed additions in the transcripts of the 70 participants in Group 3 and the 38 participants in Group 4. To reduce the interpretive confounds of increased word length on memory processes, tallies were completed for responses to four of the 2-syllable items and five of the 3-syllable items, but none of the 4-syllable items. To further reduce potential interpretive complexity, eligible addition responses for these analyses were limited to those in which the consonant target with the addition was repeated correctly and was the second or third consonant in the word (i.e. additions preceded by a vowel were more salient for phonetic transcription). Errors on the initial consonant and/or other consonants or in any vowel in the nonword were ignored. Additions that both preceded and followed the target consonant were included in the analyses.

Place analysis of additions. Inspection of the transcriptions indicated that the most frequent type of addition comprising 92.3% and 69.7% of all additions in Group 3 and Group 4, respectively, was the addition of a preceding nasal to a stop (e.g. [bɑndɑ] or [bɑmdɑ] for *bada*). The remaining additions in these words were of some other sound (e.g. [mɑrbɑ] for *maba*; [*nabavda*] for *nabada*). The question addressed was whether participants in the two groups might differ in the percentage of nasal additions that were homorganic (same articulatory place) versus heterorganic (different articulatory place) with the target consonant versus additions differing from the target consonant in both place and manner (e.g. mɑlbɑ for mɑbɑ). The hypothesis was that CAS participants might have higher percentages of heterorganic or other additions, reflecting planning/programming constraints that result in "unexpected" additions. In contrast, the additions of participants with speech delay might more often be homorganic to the target consonant, reflecting either assimilative processes associated with nasals elsewhere in the word or velar timing precision constraints due to the cognitive-linguistic encoding and memory resource demands in repeating nonsense words.

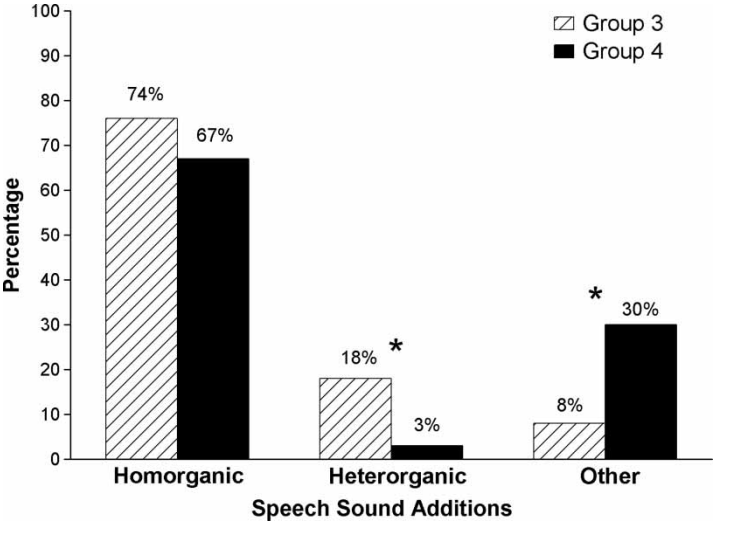

Figure 3. Graphic summary of the addition analysis findings.

Figure 3 is a graphic summary of the addition analysis findings. Cramer's V statistic, which assesses the strength of association between two categorical variables, was used to test the significance of differences among the three types of addition errors in the two groups. The asterisks in Figure 3 denote the comparisons with large standardized residuals following a statistically significant Cramer's V effect size ($p < 0.05$). As shown in Figure 3, there was no between-group difference in the percentage of homorganic additions for Group 4 (67.0%) compared to Group 3 (74.0%). However, there were significant between-group differences in the percentages of each of the other two types of additions. Group 4 had significantly lower percentage of heterorganic nasal substitutions (3%) than Group 3 (18.0%) and a significantly higher percentage of other additions (30.0% compared to 8.0%). Thus, nearly one-third of the additions in the SRT responses of participants with CAS were neither homorganic nor heterorganic nasals, the type of additions that occurred in 92.3% of the participants in Group 3. Additional, per-participant analyses of these data will be important to pursue for their possible insights into feature-level speech processing mechanisms in CAS.

Duration and relative amplitude analyses of nasal additions. A second series of addition analyses was completed, using acoustic methods to quantify the duration and relative amplitude of nasal additions in Groups 3 and 4. The rationale was that perhaps the nasal additions that did occur in CAS participants' responses differ acoustically in some way consistent with motor constraints. Specifically, the prediction was that the nasal additions of the CAS compared to speech–language impaired participants might be shorter, indicating that they were planned/programmed as part of the second rather than the first syllable (Umeda, 1977). Using conventional acoustic procedures for duration and relative amplitude in the computer environment described in Shriberg et al. (2010a, 2010b), duration analyses were completed for 13 speakers in Group 3 and 14 speakers in Group 4 and relative amplitude analyses were completed for 16 speakers in Group 3 and 14 Speakers in Group 4. Measures included duration and amplitudes of nasal targets and nasal additions to the second consonant, and the vowel preceding the nasal addition. To control for individual differences in articulation rate, three additional values were derived by dividing each raw duration by duration of the whole word. There were no between-group trends from these duration and relative amplitude analyses. That is, at the acoustic level of observations, there were no discernable differences associated with the production of nasal additions by participants in Group 4 compared to participants in Group 3.

Discussion

Discussion of findings is organized by the two questions posed in this study. Table IX includes for each question, a conclusion motivated by several findings and the source(s) for the descriptive and inferential statistical data for each finding.

(1) *Are study findings consistent with a transcoding deficit-only account of CAS in neurogenetic and idiopathic contexts, or are they more consistent with a multiple domain account with deficits in encoding, memory, and transcoding processes?*

Multiple domain account of speech processing

The first question posed in this study addresses a central question in speech processing accounts of the nature of CAS: is it predominantly a transcoding deficit or is it some type of representational deficit that is also a core feature? As summarized in Table IX, six findings

motivate the conclusion that a multiple domain account of speech processing deficits is more consistent with the data than a transcoding deficit-only account.

The first set of correlational findings summarized in Table IX indicated that the three SRT processing measures are not highly associated with one another, supporting their potential for independent contributions to descriptive–explanatory accounts of CAS (Table IV). This entry also indicates that scores from each processing construct accounted for greater than 10% of the variance in the construct of SRT competence (Table IV). The second Table IX entry indicates that SRT competence, in turn, was significantly associated with measures of productive speech competence, supporting common antecedents for the cognitive and speech processing constructs (Table V). As indicated in the third Table IX entry, although not all comparisons were statistically significant, the several group-based and individual-based diagnostic marker analyses indicated a clear trend for CAS participants to have lower SRT competence scores and lower processing scores on each of the three speech processing measures (Table VII, Figure 2). These findings are consistent with a multiple processing deficit account of CAS.

The fourth through sixth Table IX entries for this question also are consistent with multiple processing deficits in CAS. A transcoding deficit clearly emerged as the most significant of the processing deficits in participants with CAS. The most conceptually informative finding for transcoding, however, was that there were a substantial number of participants in Groups 1–3 with transcoding scores below the 80% cut-off point determined to best identify the CAS participants in this study. Notably as summarized in Table IX, these participants were significantly younger than participants with >80% transcoding scores and had significantly lower SRT competence scores and significantly lower PCC (i.e. speech production) competence scores, and were more frequently from idiopathic than neurogenetic backgrounds (Table VIII). Also, as described in the text, the additions analyses indicated that nasal additions of Group 1 participants with low transcoding scores (see Figure 3, Group 1 outlier data points) were more likely to be homorganic compared to the higher proportions of heterorganic nasals and other additions by the CAS participants (Figure 3) and that no unique acoustic feature differentiated the additions of the participants with CAS from those of participants in the other three groups.

These six findings summarized in Table IX suggest that rather than view speech sound additions as uniquely associated with motor speech processes, it is more parsimonious to view them as a response to the challenges presented by the SRT. That is, although the methods of the study do not delineate the neurocognitive or speech motor control locus or loci of additions made by participants in any of the groups, the high frequency of occurrence of additions by some participants in the non-CAS groups suggests that additions may be mediated by processing demands in addition to transcoding. A key finding is that additions occurred frequently on nonwords as short as 2syllables in length, consistent with findings in Shriberg et al. (2009), indicating a significant percentage of nonword repetition errors on the eight 2-syllable items. Token counts within and across study groups were not sufficient to complete inferential statistical analyses that might identify possible explanatory correlates of additions in each group. A need in future studies, using extended controlled stimuli is to include measures of participants' auditory-perceptual status, possibly using physiological measures (e.g. Froud & Khamis-Dakwar, 2011). On the central role of intact auditory representations in speech production, Weismer (2006, p. 318) cites Netsell's trenchant observation:

> The goal [of speech production] is to produce the appropriate acoustic patterns via flexible motor actions that are formed and maintained by "auditory images". These auditory

Table IX. Proposed answers to two questions posed in the study, motivated by the primary findings summarized and sourced in this table.

		Primary findings	
Question	Answer	Summary	Source
1. Are study findings consistent with a transcoding deficit-only account of CAS in neurogenetic and idiopathic contexts or with a multiple domain account with deficits in encoding, memory, and transcoding processes?	Findings are more consistent with a multiple domain account involving deficit in encoding, memory, and transcoding processes. Deficits in the first two speech processes, as indexed by the nonword repetition responses of both preschool and older participants with CAS, are viewed as core persistent deficits rather than developmentally secondary to motor speech constraints	1. The three SRT processing scores were not highly intercorrelated. For the entire sample, SRT competence scores shared 39% variance with scores indexing memory processes, 17% with encoding processes, and 11% with transcoding processes	Table IV
		2. SRT competence scores were significantly correlated with speech competence scores	Table V
		3. Participants in Group 4 had significantly lower competence, memory, and transcoding scores than participants in each of the three comparison groups, and significantly lower encoding scores than participants in Group 1 and Group 2	Table VII Figure 2
		4. Participants in Group 1 with <80% transcoding scores were significantly younger than those with >80% scores, had significantly lower SRT competence scores, and significantly lower PCC scores	Table VIII
		5. Participants in Group 4 with <80% compared to >80% transcoding scores were significantly more frequently from idiopathic than neurogenetic backgrounds and had significantly lower SRT competence scores	Table VIII
		6. The nasal additions of Group 1 participants with low transcoding scores were more likely to be homorganic, compared to the higher proportions of heterorganic nasals and other additions by the CAS participants. No unique acoustic feature differentiated the additions of the participants with CAS from those of participants in the other three groups	Figure 3

2. Do findings from SRT competence and processing measures contribute significant or conclusive diagnostic information to an assessment battery for CAS?	SRT scores contribute significant, but not conclusive diagnostic information. Participants with CAS had only incrementally lower competence and process scores and there was only modest support for between-group topologic differences in additions.	1. Of the four SRT measures, transcoding scores were associated with the largest effect sizes between Group 4 and Group 3 means, the highest diagnostic accuracy (74.6%), the highest prediction accuracy from a negative test (93.4%), and the largest odds ratio. SRT transcoding scores in this study did not meet the 90% sensitivity and 90% specificity criterion required for diagnostic likelihood ratios conclusive for CAS	Table VII Table VIII
		2. The predominant transcoding error by participants in each of the four groups was the addition of a nasal (/m/ or /n/) preceding a medial stop	Figure 3
		3. In comparison to participants with speech–language impairment, CAS participants had a significantly lower percentage of nasal additions heterorganic to the stop (i.e. different place) and a significantly larger percentage of other substitutions. The nasal additions of the two groups did not differ significantly in duration or relative amplitude	Figure 3

> images … become yoked to the motor and somato-afferent patterns used to generate them…

We suggest that in addition to indexing transcoding errors, addition errors on nonword repetition tasks such as the SRT may, in part, index auditory-perceptual encoding problems (i.e. nonveridical "auditory images").

To summarize, transcoding scores were moderately sensitive to CAS, with 74% of CAS participants scoring below the 80% cut-off point for this measure. However, 9.2% of participants with typical speech and 25.2% of participants with speech delay also scored below the 80% criterion, indicating that the processing source of addition errors is likely multiply determined.

Neurogenetic and idiopathic contexts for CAS

A second perspective on transcoding versus multiple domain speech processing accounts of CAS is Table IX finding for participants with idiopathic CAS compared to participants with CAS from neurogenetic contexts. As reported in Table VIII, a higher proportion of participants with idiopathic CAS had <80% transcoding scores. There were no age or speech differences in the two subgroups of CAS that might have moderated this finding (Table IX). Due to the diverse genomic histories of participants with CAS associated with complex neurodevelopmental disorders, the expectation was that they would have lower average transcoding score than scores from participants with idiopathic CAS. These findings are consistent, however, with prior findings on a subset of the present sample, indicating that participants with idiopathic CAS had a higher percentage of promising perceptual and acoustic markers of CAS than participants with neurogenetic CAS (Shriberg, 2010a).

(2) *Do findings from SRT competence and processing measures contribute significant or conclusive diagnostic information to an assessment battery for CAS?*

Findings for the second question posed in this study are interpreted as support for SRT scores as providing significant, but not conclusive diagnostic information to identify CAS. As summarized in the three entries in Table IX and reviewed in the preceding discussion, each of the SRT scores differentiated the CAS participants from those with typical speech–language development, but none had the diagnostic sensitivity or specificity for conclusive identification of CAS, which requires CAS to be differentiated from speech delay with or without language impairment. As summarized in Table IX, transcoding scores above the cut-off of 80% may be useful to rule out CAS if this cut-off value is cross-validated in larger samples of participants with neurogenetic and idiopathic CAS and larger samples of participants with significant SD. Additional analyses indicated that other cut-off points may be more discriminative, depending on the severity levels of the participants with both CAS and SD.

The key concept in the search for diagnostic markers to date is that no one diagnostic sign is likely to be conclusive to identify CAS at all ages, at all cognitive levels, at all levels of speech competence, and in all etiologic contexts. A primary clinical need is a set of tasks that provides conclusive signs of CAS in very young children with limited verbal output. As reviewed previously, we have reported candidate signs within the linguistic domains of vowels, phrasing, rate, and stress. The present findings add to the signs described to date, deficits in encoding, memory, and transcoding that exceed the range of deficits seen in severe speech delay. Lohmeier and Shriberg (2011) include reference data that can be

used for this purpose, including SRT reference data for a group of 91 Australian-English-speaking participants with speech delay.

Conclusions

Findings from this study are interpreted as support for three conclusions. First, findings are consistent with a multiple domain descriptive–explanatory framework for CAS in which auditory-perceptual encoding, memory, and transcoding deficits are core features of CAS in both idiopathic and neurogenetic contexts. This position is consistent with findings from a survey that included responses from 201 parents attending a national conference on CAS (Teverovsky, Bickel, & Feldman, 2009, p. 94); the investigators provide the following summary of survey findings:

> The most prevalent functional problems in addition to communication were attention (focus), vestibular function, temperament, fine hand use, maintaining attention, and learning to write. Four orthogonal factors accounted for 23% of the variance in functional problems: Cognitive and Learning Problems, Social Communication Difficulties, Behavioral Dysregulation, and Other Oral Motor Problems. Over half the sample had health, mental health, and developmental conditions. Almost all of the children used early intervention and speech/language therapy services. … The identified factors should guide the multidisciplinary team in conducting comprehensive evaluations, rehabilitation, and long-term follow-up of children with CAS.

A second conclusion is that the present findings are consistent with literature findings indicating primarily quantitative, rather than conclusive differences in the speech, prosody, and voice patterns of participants with CAS compared to participants with other moderate to severe speech sound disorders. With the exception of differences in the type and frequency of phrasing deficits (i.e. spatiotemporal disruptions and revisions that occur on challenging words), few conclusive signs of CAS have been documented. Until translational science yields a neurologic, biochemical, or pharmacologic biomarker for this disorder, behavioural diagnosis of CAS in different etiologic contexts will likely require a battery of individually appropriate, standardized measures and a validated algorithm, indicating the number and type of positive signs needed for conclusive diagnosis.

Finally, the present findings are interpreted as support for the etiological classification framework for speech sound disorders, including CAS that underlies this research (Figure 1) compared to other organizational proposals for congenital and environmental (e.g. Dodd, Holm, Crosbie, & McCormack, 2005) and acquired (e.g. Weismer & Kim, 2010) speech disorders based on taxonomic similarities in speech patterns (see also Damico, Müller, & Ball, 2010). We submit that taxonomic frameworks miss unifying biological and behavioural processes in complex neurodevelopmental disorders that can advance understanding of etiopathogeneses (e.g. the co-occurrence of oculomotor apraxia and CAS in Joubert syndrome cited previously). A framework for research and practice based on putative aetiology, epidemiologic information on risk and protective factors, and lifespan outcomes is viewed as best meeting the requirements of next-generation, personalized medicine (Shriberg et al., 2010a). Specifically, aggregating genotype–phenotype relationships and epidemiologic data by diagnostic classification placeholders is proposed to best position speech sound disorders such as CAS, the translational advances emerging in developmental neurobiology and the genomic sciences (Navon, 2011). Snowling and Hulme (2011) and Walsh (2011) provide useful discussions of classification theory and applications to two widely used

classification systems that include verbal trait disorders that are relevant to the common risk factor classification perspective advocated in the present report.

Acknowledgements

We thank the following colleagues and collaborators for their significant contributions to this study: Richard Boada, Thomas Campbell, Lisa Freebairn, Jordan Green, Heather Karlsson, Barbara Lewis, Jane McSweeny, Christopher Moore, Bruce Pennington, Heather Leavy Rusiewicz, Christie Tilkens, and David Wilson. Primary grant support was provided by the National Institute on Deafness and Other Communicative Disorders (NIDCD No. DC00496), by subcontracts with NIDCD No. DC00822, NIDCD No. DC00528, and NIMH No. 38820, a grant from the General Clinical Research Center at Children's Hospital of Pittsburgh (M01RR00084), and a core grant to the Waisman Center from the National Institute of Child Health and Development (HD03352).

Declaration of Interest: The authors report no declaration of interest. Primary grant support was provided by the National Institute on Deafness and Other Communicative Disorders (NIDCD No. DC00496), by subcontracts with NIDCD No. DC00822, NIDCD No. DC00528, and NIMH No. 38820, a grant from the General Clinical Research Center at Children's Hospital of Pittsburgh (M01RR00084), and a core grant to the Waisman Center from the National Institute of Child Health and Development (HD03352).

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Appendix. The SRT stimuli

1. bada	7. nada	13. banada
2. dama	8. maba	14. manaba
3. bama	9. bamana	15. bamadana
4. mada	10. dabama	16. danabama
5. naba	11. madaba	17. manabada
6. daba	12. nabada	18. nadamaba

In addition to the primary citation for the SRT (Shriberg et al., 2009), two technical reports and a PowerPoint presentation of the SRT stimuli can be downloaded without cost from the Technical Reports section of the Phonology Project website: http://www.waisman.wisc.edu/phonology/ (Shriberg & Lohmeier, 2008).

This technical report provides (a) psychometric data on the SRT, (b) statistical findings from several additional analyses of the SRT, (c) comparison data obtained from 70 typically speaking children from 4 to 16 years of age, (d) administration instructions, (e) scoring instructions, and (f) a form for manual scoring of the SRT (Lohmeier & Shriberg, 2011).

This technical report provides tabular statistics and graphic summaries of 23 SRT measures (i.e. including subscale data at each syllable length) obtained using a software utility. The primary measures, including SRT competence, encoding, memory, and transcoding scores, can be obtained using the manually analysed version of the SRT described in Phonology Project Technical Report No. 14. The tabular statistics and graphic displays include information from 3- to 17-year-old children and youth with typical speech–language and with speech delay assessed in several cities in the USA and Australia.