Childhood Apraxia of Speech: Technical Report

Ad Hoc Committee on Apraxia of Speech in Children

This technical report was developed by the American Speech-Language-Hearing Association (ASHA) Ad Hoc Committee on Apraxia of Speech in Children. The report reviews the research background that supports the ASHA position statement on Apraxia of Speech in Children [DATE]. Members of the Committee were Lawrence Shriberg (chair), Christina Gildersleeve-Neumann, David Hammer, Rebecca McCauley, Shelley Velleman, and Roseanne Clausen (ex officio). Celia Hooper, ASHA vice president for professional practices in speech-language pathology (2003–2005), and Brian Shulman, ASHA vice president for professional practices in speech-language pathology (2006–2008), served as the monitoring officers. The Committee thanks Sharon Gretz, Heather Lohmeier, Rob Mullen, and Alison Scheer-Cohen, as well as the many select and widespread peer reviewers who provided insightful comments on drafts of this report.

INTRODUCTION AND OVERVIEW

The goal of this technical report on childhood apraxia of speech (CAS) was to assemble information about this challenging disorder that would be useful for caregivers, speech-language pathologists, and a variety of other health care professionals. Information on CAS has often been the most frequent clinical topic downloaded by visitors to ASHA's Web site. This report addresses four questions most often asked about CAS: (a) Is it a recognized clinical disorder? (b) What are its core characteristics? (c) How should it be assessed? and (d) How should it be treated?

To address these four questions, the Committee undertook a review of the scientific foundations of CAS and trends in professional practice. A preliminary survey of the literature indicated that it would not be feasible to complete a systematic review consistent with evidence-based practice. The primary barriers to such a review were unresolved controversies about the quality rankings for commonly used research designs, as proposed in several evidence-based practice systems. The Committee therefore elected to complete narrative reviews restricted to peer-reviewed literature published since 1995, with additional sources consulted as needed for coverage of certain topics. We developed a template to summarize each study and consensus procedures to evaluate the strength and quality of evidence for research findings in relation to the four questions posed above. Findings from reviews and the consensus evaluation procedures were synthesized to form the bases for the information provided in this document, including recommendations on several key professional issues. The final document incorporated extremely useful information from select and widespread reviewers who responded to invitations to review preliminary drafts of this document, including a draft posted on ASHA's Web site.

In this initial section of the report, we introduce terms and concepts, consider issues associated with the definition of CAS, and discuss scientific and professional information related to the reported increased prevalence of CAS.

Terms and Concepts

Childhood Apraxia of Speech Versus Developmental Apraxia of Speech

The Committee recommends *childhood apraxia of speech* (*CAS*) as the classification term for this distinct type of childhood (pediatric) speech sound disorder. Beginning with the first word in this term, two considerations motivate replacing the widely used *developmental* with the word *childhood.* One consideration is that CAS support groups in the United States, the United Kingdom, and elsewhere have requested that *developmental*

not be used in a classification term for this disorder. Inclusion of this word is reportedly interpreted by service delivery administrators as indicating that apraxia is a disorder that children "grow out of" and/or that can be serviced solely in an educational environment (see relevant discussion on the Apraxia-Kids listserv: www.apraxia-kids.org/talk/subscribe.html). A second rationale for the use of *CAS* as a cover term for this disorder, rather than alternative terms such as *developmental apraxia of speech* (*DAS*) or *developmental verbal dyspraxia* (*DVD*), is that our literature review indicated that apraxia of speech occurs in children in three clinical contexts. First, apraxia of speech has been associated causally with known *neurological etiologies* (e.g., intrauterine stroke, infections, trauma). Second, apraxia of speech occurs as a primary or secondary sign in children with *complex neurobehavioral disorders* (e.g., genetic, metabolic). Third, apraxia of speech not associated with any known neurological or complex neurobehavioral disorder occurs as an *idiopathic neurogenic speech sound disorder*. Use of the term *apraxia of speech* implies a shared core of speech and prosody features, regardless of time of onset, whether congenital or acquired, or specific etiology. Therefore, *childhood apraxia of speech* (*CAS*) is proposed as a unifying cover term for the study, assessment, and treatment of all presentations of apraxia of speech in childhood. As above, *CAS* is preferred over alternative terms for this disorder, including *developmental apraxia of speech* and *developmental verbal dyspraxia*, which have typically been used to refer only to the idiopathic presentation.

Apraxia Versus Dyspraxia

Rationales for the second and third words in the classification term *CAS* reflect empirical findings for children suspected to have this disorder. The alternative terms—*apraxia of speech* versus *(verbal) dyspraxia*—each have established traditions in international literatures. *Apraxia of speech* is more widely used in the United States following the Mayo Clinic traditions (Duffy, 2005), whereas *verbal dyspraxia* is the preferred term in many other English-speaking countries. Differentiating between these alternatives based solely on etymological distinction (i.e., total [*a*] vs. partial [*dys*] absence or lack of function) is problematic when applied to CAS. Clinical experience indicates that although a child suspected to have CAS may have very limited speech, seldom is a child completely without mastery of some speech sounds. Notwithstanding this difference, and to parallel usage for the possible acquired form of this disorder in adults (i.e., AOS), the Committee recommends use of the affix *a* for this classification term.

The Apraxias Versus the Dysarthrias

Several other types of apraxia and several types of dysarthria play prominent roles in the scientific foundations of CAS. Physicians and researchers recognize *ideomotor and limb kinetic praxis problems* that may or may not be present in persons with apraxia of speech. As discussed in this report, orofacial and limb apraxias are of particular interest as the presence of one or both in a child suspected to have CAS may provide support for the diagnosis, particularly in prelingual children. Apraxia in other systems may also play important roles in treatment. For example, the presence of limb apraxia may preclude using manual signs for functional communication. Moreover, the presence of orofacial apraxia may support the need for either more aggressive or alternative approaches to the use of phonetic placement cues in speech treatment.

Concerning dysarthria, a neuromotor disorder presumed not to involve the planning or programming deficit in apraxia (see below), some forms of these two disorders may share common speech characteristics. As discussed in later sections, a significant research challenge is to determine the diagnostic boundaries between CAS and some types of dysarthria with which it may share several speech, prosody, and voice features.

CAS Versus AOS

Although the core feature of CAS, by definition, is proposed to be similar to the core feature of AOS in adults, this relationship does not preclude the possibility of important differences in associated features. For example, Maassen (2002) noted that "a fundamental difference between [adult] AOS and [CAS]...is that in [CAS] a specific underlying speech motor impairment has an impact on the development of higher phonological and linguistic processing levels" (p. 263). Despite a much larger and well-developed literature in AOS, including many chapter-length discussions of alternative theoretical frameworks, the Committee elected not to include reviews of theory and research on acquired apraxia of speech in this report. This decision was motivated by the view that the scientific foundations of CAS should be based on research directly concerned with this and related childhood speech sound disorders. However, as discussed in several places in this document, the Committee has attempted to anticipate likely parallels between acquired apraxia of speech in adults and CAS, a task made more difficult by differences in terminology used to describe them. Treatment guidelines for acquired apraxia of speech have recently been proposed by the Academy of Neurological Communicative Disorders and Sciences (Wambaugh, Duffy, McNeil, Robin, & Rogers, 2006a, 2006b).

Definitions of CAS

The Committee compiled a table of more than 50 definitions of CAS that have appeared in the research and clinical literature, primarily within the past 10 years. A few of the more widely cited definitions dating back to the early 1970s are provided in the table to sample the variety of perspectives on the nature of CAS among researchers, including some definitions found in secondary sources such as Web sites and professional organizations consulted by caregivers and health care professionals. We are keenly aware of the limitations of any definition of CAS until the behavioral correlates and neural substrates of this disorder have been identified and extensively cross-validated. Considering its value for children, caregivers, clinicians, researchers, and stakeholders, however, we viewed the scope of our task as including a working definition of CAS. Recognizing an almost certain need for revision based on emerging research findings, the Committee proposes the following definition:

> *Childhood apraxia of speech* (*CAS*) is a neurological childhood (pediatric) speech sound disorder in which the precision and consistency of movements underlying speech are impaired in the absence of neuromuscular deficits (e.g., abnormal reflexes, abnormal tone). CAS may occur as a result of known neurological impairment, in association with complex neurobehavioral disorders of known or unknown origin, or as an idiopathic neurogenic speech sound disorder. The core impairment in planning and/or programming spatiotemporal parameters of movement sequences results in errors in speech sound production and prosody.

Review of the research literature indicates that, at present, there is no validated list of diagnostic features of CAS that differentiates this symptom complex from other types of childhood speech sound disorders, including those primarily due to phonological-level delay or neuromuscular disorder (dysarthria). Three segmental and suprasegmental features that are consistent with a deficit in the planning and programming of movements for speech have gained some consensus among investigators in apraxia of speech in children: (a) inconsistent errors on consonants and vowels in repeated productions of syllables or words, (b) lengthened and disrupted coarticulatory transitions between sounds and syllables, and (c) inappropriate prosody, especially in the realization of lexical or phrasal stress. Importantly, these features are not proposed to be the necessary and sufficient signs of CAS. These and other reported signs change in their relative

frequencies of occurrence with task complexity, severity of involvement, and age. The complex of behavioral features reportedly associated with CAS places a child at increased risk for early and persistent problems in speech, expressive language, and the phonological foundations of literacy as well as the possible need for augmentative and alternative communication and assistive technology. It is useful to comment briefly on the core elements of this definition.

The Core Problem

As required of any proposed disorder classification, definitions of CAS have three elements that may be given in any order: description of the core problem, attribution of its cause or etiology, and listing of one or more diagnostic signs or markers. Definitions of CAS, such as the one above, invariably include the proposed core problem, whereas the other two elements may or may not be addressed. One of the major differences among alternative definitions of CAS is whether the core problem is proposed to include input processing as well as production, and if so, whether auditory, sensory, and prosodic aspects of perception may prefigure in the deficit. An example of a framework that might implicate the latter is the speech motor control model in development by Guenther and colleagues (e.g., Guenther, 2006; Guenther & Perkell, 2004). Whereas some of the definitions of CAS reviewed by the Committee view the core problem as one of planning and programming the spatiotemporal properties of movement sequences underlying speech sound production, others propose that the deficit extends to representational-level segmental and/or suprasegmental units in both input processing and production.

Etiology

Definitions of CAS have universally ascribed its origin to neurologic deficits, with alternative viewpoints differing with respect to specific neuroanatomic sites and circuits. There is also clear agreement that whatever the neural substrates of CAS, they differ from those underlying the several types of dysarthria. The definition of CAS proposed for this report is also clearly consistent with this neurogenic perspective.

Signs and Markers

In addition to the core problem and etiology, the third element in the proposed definition of CAS and those reviewed by the Committee is the inclusion of the key diagnostic features of the disorder. Three such features are included in the present definition with discussion of other candidate features reviewed in subsequent sections of this report. The three features in the present definition of CAS represent a consensus conclusion based on our evaluations of the clinical research and our evaluation of comments from reviewers of preliminary drafts of this report. A major conclusion of this report is that there presently is no one validated list of diagnostic features of CAS that differentiates this disorder from other types of childhood speech sound disorders, including those apparently due to phonological-level deficits or neuromuscular disorder (dysarthria).

Prevalence of CAS

As with several other complex neurobehavioral disorders (e.g., autism, attention deficit hyperactivity disorder), the prevalence of CAS has reportedly increased substantially during the past decade. For example, in a study of 12,000 to 15,000 estimated diagnostic outcomes for children referred with speech delay of unknown origin from 1998 to 2004, a staff of 15 speech-language pathologists in a large metropolitan hospital diagnosed 516 (3.4%–4.3%) of these children as having suspected CAS (Delaney & Kent, 2004). Much needed population prevalence data are not available, including information by race and ethnicity. One preliminary population estimate, based solely on clinical referral data, is that CAS may occur in one to two children per thousand (Shriberg, Aram, & Kwiatkowski, 1997a), a population rate that is much lower than the rate at which this

classification currently appears to be assigned. Although currently there are no epidemiologically sound estimates of the prevalence of CAS in the United States or elsewhere, several interacting factors likely contribute to clinical diagnostic figures as high as those reported by Delaney and Kent (2004).

Birth-to-Three Legislation

One potential source of the apparent increased diagnostic prevalence of CAS in the past one to two decades is the impact of legislative changes during this period. Since the passage of early intervention statutes, particularly the Individuals with Disabilities Education Improvement Act of 2004 (IDEA '04, Part C), speech-language pathologists are asked to evaluate and identify communicative disorders as early as possible in infants and toddlers. A major problem in classifying young prelingual children (i.e., children with severe delays in the onset of speech) is that a diagnosis of CAS must be based on variables other than speech itself. As discussed later in this report, findings claiming that behaviors such as difficulty in feeding or excessive drooling are pathognomonic (positive signs) of CAS are tentative at best. For children suspected to have CAS who do have at least a moderate inventory of speech sounds, their communication profiles can be similar to those of children with other speech-language disorders or neurobehavioral disorders (Davis, Jakielski, & Marquardt, 1998; Davis & Velleman, 2000). Thus, although we use the term *CAS* for children who are the focus of the research reviewed in this document, it should be understood that the lack of a gold standard for differential diagnosis requires that all such classificatory labels be considered provisional.

Increased Information

Increased information on a disorder may both reflect and contribute to increased prevalence. For CAS, the past decade has seen dramatic increases in both. Interest in CAS is readily apparent when reviewing the increased number of research symposia (e.g., Shriberg & Campbell, 2003), clinical workshops, and parent support groups on CAS. Although there have been no formal accounts describing the history of this clear trend, it appears to parallel similar development in other disorders. From an academic perspective, information about CAS has traditionally been embedded within undergraduate and graduate courses in speech disorders in children or, more typically, in motor speech disorders in children and adults. However, for many speech-language pathologists, applied information on this topic is typically learned in workshops presented by persons with varying research and clinical backgrounds and/or experience with children suspected to have CAS. The Committee's anecdotal observations are that such workshops are currently among the most widely advertised opportunities for continuing education credits.

The major source of readily available information on CAS is the Internet, including its numerous Web sites and electronic discussion forums that include information on this topic. As with other unregulated medical and health-related information sources, the accuracy and usefulness of information presented on the Internet varies substantially. Some sites available internationally provide excellent information, including detailed guidelines for caregivers seeking service delivery options.

Reimbursement Issues

Speech-language pathologists must be knowledgeable about reimbursement alternatives and insurance guidelines. Because insurance companies frequently require that a child have a medical diagnosis to approve coverage, there may be increased use of CAS as a diagnostic classification for a severe childhood speech sound disorder. However, insurance claims for children with this diagnosis may sometimes be denied due to the continuing controversial status of CAS as a clinical entity and its increased prevalence in diagnostic coding.

Lack of Diagnostic Guidelines

Clearly the major source of overdiagnosis of CAS is the inconsistent and conflicting behavioral features purported to be diagnostic signs of CAS (Shriberg, Campbell, et al., 2003; Shriberg & McSweeny, 2002). In addition to children who may be misdiagnosed as false positives (persons said to have a disorder who do not), diagnostic guidelines also may result in false negatives (persons said not to have a disorder who do). Later discussion addresses this fundamental issue.

<center>Summary</center>

On the first of four questions motivating this technical report—Is CAS a clinical entity?—the Committee concludes that the weight of literature findings support the research utility of this type of speech sound disorder. A primary research source for this position is the findings on apraxia of speech that occur as sequelae to a neurological disorder and within a number of complex neurobehavioral disorders, as noted later in this document. On the second question of the core features and behavioral markers of CAS, the Committee proposes a definition of CAS that classifies it as a neurological disorder affecting the planning/programming of movement sequences for speech. However, there currently are no lists of behavioral features that are validated as necessary and sufficient for the diagnosis of CAS, although three general characteristics are proposed as possible candidates based on our narrative review and consultation with peer evaluators. On the third and fourth questions, this report does not include specific guidelines for the assessment and treatment of CAS, primarily due to the lack of research support to date for such guidelines. In a section titled Professional Issues, we review general recommendations by experienced clinical practitioners, but specific guidelines for clinical practice are deferred to future ASHA policy documents. Finally, we have noted some issues that may be associated with the recent increase in the diagnosis of suspected CAS, including birth-to-three legislation, increased availability and accessibility of information on CAS, reimbursement issues, and the lack of diagnostic guidelines.

<center>SCIENTIFIC FOUNDATIONS:
OVERVIEW OF TYPICAL AND ATYPICAL SPEECH DEVELOPMENT</center>

We begin a review of the scientific foundations of CAS with an overview of typical and atypical speech acquisition, highlighting those segmental and suprasegmental behaviors that are frequently studied in CAS research. For example, we include prelinguistic speech development in each section because children suspected to have CAS are often reported to not babble at all, to babble less frequently than their typically developing peers, or to produce less mature, complex babble. Thus, a review of these foundational prelinguistic behaviors and their implications for later speech-language development seems warranted. In addition to delays in reaching developmental milestones, children suspected to have CAS may follow idiosyncratic developmental paths. For this reason, reference to typical milestones may be useful for diagnosis (i.e., atypical profiles may be suggestive of CAS).

<center>Motor Control</center>

A Note on Terms

In any discussion of speech motor control, or speech production generally, the terms *variable* and *inconsistent* are likely to arise. They are often used interchangeably and without precise definitions. This is of particular concern with respect to CAS, as some clinical investigators use *inconsistency* as a key classification criterion for the disorder. Some common uses of *variable* and *inconsistent* include the following:

1. differential use of a certain phoneme or sound class in different word positions (e.g., the child produces /r / accurately in final position but substitutes [ɶ]

for /r / in prevocalic position);

2. differential use of a certain phoneme or sound class in different word targets, even in the same word position (e.g., the child produces /ˑ / accurately in certain well-rehearsed words such as "mommy," but does not produce it accurately in similar or even seemingly easier words such as "moo");

3. differential use of a certain phoneme or sound class in multiple repetitions of the same word (e.g., the child produces "fish" once as "pish", once as "pit", once as "fit", and another time as "shiff"). This may include measures of the number of different errors the child made in the word (e.g., in the example above, errors consisted of stopping of /m /, stopping of /ʃ/, and metathesis) or measures of the frequency at which a given error type is used (e.g., in the example above, stopping was the most consistent error type because stopping was used four times, and metathesis only once). This type of inconsistency is sometimes referred to as "token-to-token variability" (Seddoh et al., 1996).

Except where specified otherwise within in this document, *inconsistency* refers to differences in multiple productions of the same target word or syllable (i.e., token-to-token variability). *Variability* is used elsewhere, when meaning 1 or meaning 2, or more than one of the above meanings, is included within the findings being reported or when parameters other than speech production (e.g., pitch) are being discussed.

Oral-Motor Development

Beginning this review with research on typical oral-motor development, studies indicate that jaw control is established by about 15 months, before control is established for the upper and lower lips (Green, Moore, Higashikawa, & Steeve, 2000; Green, Moore, & Reilly, 2002). Motor development is slower for structures, such as the lips, that have more degrees of freedom of movement (Green et al., 2002). Tongue development is also gradual, with extrinsic tongue movements necessary for swallowing and sucking developed prior to the intrinsic tongue movements required for fine motor control (S. G. Fletcher, 1973; Kahane, 1988). Such findings are hypothesized to account for the high frequency of occurrence of infants' production of syllables that can be articulated without changes in lip or tongue configuration—including labial consonants with low and neutral vowels, coronal (alveolar and dental) consonants with high front vowels, and dorsal (velar) consonants with high back vowels (Davis & MacNeilage, 1995; MacNeilage & Davis, 1990). The high prevalence of such syllables is claimed to be associated with infants' early ability to open and close the jaw, creating the consonant–vowel alternation necessary for the syllable, with the lower lip (for labials) or the tongue (for alveolars and velars) essentially "going along for the ride." Some clinical reports indicate that these immature patterns may persist in children suspected to have CAS (Velleman, 1994).

Through processes of differentiation and refinement, the slightly older child acquires independent control over individual articulators (lips, different portions of the tongue) and learns to produce more specialized configurations to grade movements, eventually sequencing these articulatory postures without extraneous movements (Davis & MacNeilage, 2000; Green et al., 2000). Thus, automaticity and flexibility develop over time. Both neuromotor maturation and practice are believed to underlie this developmental process, with vocal experience leading to the formation of specific neuronal pathways for finer levels of control (Green et al., 2000). Coarticulation that reflects poor temporal control or poor differentiation of structures decreases, whereas coarticulation that reflects language-specific efficiency increases, as the child becomes more adept (Nijland et al., 2002; Nijland, Maassen, van der Meulen, et al., 2003). One model of the role of perception in this process was provided by Guenther and colleagues (e.g., Guenther, 2006; Guenther & Perkell, 2004).

In a following section, we will see that these developmental changes may not occur spontaneously in children suspected to have CAS.

In the present context it is especially relevant to note that mastication and deglutition (swallowing) skills are not direct precursors to speech. Motor control of feeding functions is separate from motor control for vocalization early in infancy (Moore & Ruark, 1996), as is motor control for speech breathing versus breathing at rest (Moore, Caulfield, & Green, 2001). Although "the labiomandibular movement patterns established for feeding may influence initial attempts to coordinate these structures for speech" (Green et al., 2000, p. 252), this influence is more likely to be negative than positive, as feeding patterns involve tight linking of lips with jaw in a highly rhythmic stereotyped pattern. To produce a variety of syllables within varied prosodic patterns requires the child to overcome the interdependent inflexible patterns associated with sucking. Speech requires finer levels of coordination (Green et al., 2000) but lower levels of strength than are available for other oral-motor activities (Forrest, 2002). Thus, a consensus opinion among investigators is that nonspeech oromotor therapy is not necessary or sufficient for improved speech production (see also Professional Issues: Treatment).

When children reach middle school age and even beyond, their speech production continues to be more variable, less flexible, and less accurate than adult speech. Variability is especially noted during the initial portion of speech or speech-like movements, with more feedback required for unfamiliar speech tasks (Clark, Robin, McCullagh, & Schmidt, 2001). Furthermore, as discussed in Clark et al., children's speech may be constrained by resource allocation needs, such as the need to scale back the extent of a movement in order to complete it more quickly. For example, children between the ages of 5 and 6 years are able to partially compensate for the presence of a bite block between their teeth without an increase in variability or a change in coarticulation patterns, although vowel accuracy is decreased somewhat and segment durations are increased (Nijland, Maassen, & van der Meulen, 2003). Maximum performance rates have been shown to increase with age, with changes from 3.7 same syllable repetitions of /˘] / per second and only 1.3 repetitions of "patty-cake" at age 2;6–2;11 [years;months] to 5.5 same syllable repetitions and 1.6 repetitions of "patty-cake" at age 6;6–6;11 (Robbins & Klee, 1987). Maximum performance rates continue to increase with maturity, with young adult same syllable repetitions typically reported at average rates between 6 and 7 per second and 5.8 to 6.9 repetitions per second of /˘] Œ] k] / (Baken & Orlikoff, 2000). However, Williams and Stackhouse (1998, 2000) reported that rate of speech may be a less reliable measure of motor control in preschool children than accuracy and consistency of response. Again, many of the core questions about CAS address the possibility that children suspected to have CAS have different developmental trajectories on these and other motor control parameters.

Speech Production

Prelinguistic Period

Speech development begins long before the first word is spoken. Development of this system occurs as a child gains motor control of the speech mechanism and learns the phonological rules for production of the ambient language or languages. Prelinguistic perceptual and vocal experiences lay the groundwork for later speech and language. For example, the frequency of a child's vocalizations at 3–6 months is correlated with several later developmental milestones, including performance on the Bayley Verbal Scale at 11–15 months and expressive vocabulary size at 27 months (Stoel-Gammon, 1992).

One of the most important motor precursors to first oral words is canonical babbling, the rhythmic production of repetitive consonant–vowel (CV) sequences with

complete consonant closures and fully resonant vowels (Ejiri, 1998; Oller, 1986). The frequency of occurrence of "true" supraglottal nonglide consonants in babble is positively correlated with phonological development and even with language skills (Stoel-Gammon, 1992). Children who demonstrate consistent vocal motor schemes, or favorite babbles, tend to develop words earlier (McCune & Vihman, 1987). Children suspected to have CAS who are reported by their parents to have babbled very little or with very little phonetic diversity (Davis & Velleman, 2000) are at a linguistic disadvantage long before word production begins. The frequency and characteristics of early vocalizations also can be affected by perceptual factors such as early otitis media with effusion (Petinou, Schwartz, Mody, & Gravel, 1999; Rvachew, Slawinski, Williams, & Green, 1999), as well as by physiological and other factors (see Kent, 2000). Research suggests that the earliest stages of speech development in monolingual and bilingual speakers are highly similar regardless of language environment (Buhr, 1980; Davis & MacNeilage, 1995; Gonzalez, 1983; Kent, 1992; Locke & Pearson, 1992; MacNeilage & Davis, 1990; Oller & Eilers, 1982; Poulin-Dubois & Goodz, 2001; Thevenin, Eilers, Oller, & Lavoie, 1985; Zlatic, MacNeilage, Matyear, & Davis, 1997). Babbling includes stops, nasals, and glides at the labial and coronal places of articulation (Davis & MacNeilage, 1995; Kent & Bauer, 1985; Locke, 1983; Oller, Eilers, Urbano, & Cobo-Lewis, 1997), nonrounded vowels (Davis & MacNeilage, 1990; Kent & Bauer, 1985; Levitt & Aydelott-Utman, 1992), and simple CV and CVCV syllable shapes (Boysson-Bardies, Sagart, & Bacri, 1981; Buhr, 1980; Oller & Eilers, 1982; Vihman, Ferguson, & Elbert, 1986).

Linguistic Period

In the first linguistic stage, from 12 to 18 months, babbling decreases and word production increases. While slight differences in frequencies of sounds and word shapes are reported cross-linguistically (Boysson-Bardies & Vihman, 1991; Maneva & Genesee, 2002), the considerable cross-linguistic similarities observed in babbling also exist in first words. Children from various language environments mainly produce coronal and labial stops, nasals, and glides, and simple CV syllable shapes in their first words (Boysson-Bardies & Vihman, 1991; Eilers, Oller, & Benito-García, 1984; Gildersleeve-Neumann, 2001; Goldstein & Cintrón, 2001; Oller, Wieman, Doyle, & Ross, 1976; Teixeira & Davis, 2002; Vihman et al., 1986). In addition, limited research on English-learning infants and infants in other monolingual language environments suggests that low front, nonrounded vowels are most frequent in first words (Davis & MacNeilage, 1990; Gildersleeve-Neumann, 2001; Levitt & Aydelott-Utman, 1992; So & Dodd, 1995; Stoel-Gammon & Dunn, 1985; Teixeira & Davis, 2002). Research on sounds in the first words of simultaneous bilinguals is extremely limited; however, it appears that similar consonants (Keshavarz & Ingram, 2002) and word shapes (Kehoe & Lleo, 2003) predominate. Information is not currently available on possible cross-dialectal differences.

Children's early speech patterns include phonotactic errors such as reduplication (e.g., "wawa" for "water"), consonant harmony (e.g., "goggie" for "doggie"), and final consonant deletion (e.g., "da" for "dog") during their first 12–18 months of word production; these error patterns typically are markedly diminished by 3 years of age in children who are typically developing, although not, as reviewed later, in children suspected to have CAS. Apparent regression, in which the child produces a word less accurately but also with less variability than before, may also occur during the first year of word production as children systematize their phonologies (Vihman & Velleman, 1989). Individual sounds may be produced variably, even within the same word, although speech production patterns (i.e., frequent phonological processes) are consistent (Demuth, 2001; Ferguson & Farwell, 1975; Taelman & Gillis, 2002;

Velleman & Vihman, 2002).

Between the ages of 2 and 3 years, the speech sound system of typically developing children expands in complexity, resulting in productions of a greater variety of consonants, vowels/diphthongs, and word shapes. By this age, children in English-learning environments begin to produce the more complex sounds—velars, fricatives, affricates, and liquids—generally mastering the majority of sounds with these features by approximately 5 years of age (Stoel-Gammon & Dunn, 1985). The few studies that have examined vowel and diphthong development suggest that accurate production of all vowels and most diphthongs (but not rhotic vowels) is achieved by age 3 (Bassi, 1983; Larkins, 1983; Pollock & Berni, 2003). In Pollock and Berni's study, the average percentage of vowels correct for children between 18 and 23 months was 82%, increasing to 92% for 24- to 29-month-olds, 94% for 30- to 35-month-olds, and 97% by 36 months of age. As subsequently discussed, the picture is very different for children suspected to have CAS. For typically developing children, more complex word shapes become frequent during this early period, with many consonant clusters, final consonants, and unstressed syllables correctly produced, resulting in a large increase in accuracy and intelligibility (Stoel-Gammon & Dunn, 1985). Consonant clusters emerge by the first third of the fourth year (36–40 months), usually appearing first in final position in speakers of Mainstream American English (Kirk & Demuth, 2003). Typically developing children are reportedly 26%–50% intelligible by 2 years, 71%–80% intelligible by 3 years, and 100% intelligible by 4 years (Coplan & Gleason, 1988; Weiss, 1982). It is also after age 2 that the diverse effects of a child's ambient language become most apparent (see below; Goldstein & Washington, 2001; Johnson & Wilson, 2002; Walters, 2000).

In typical and most atypical, nonapraxic speech during this age period, earlier developing sounds tend to be substituted for later developing sounds that the child may not be able to produce as easily (e.g., stops substitute for fricatives and glides substitute for liquids). Children with nonapraxic speech sound disorders appear to be most successful at producing the correct voicing features of a segment and least successful at maintaining the correct place of articulation (Forrest & Morrisette, 1999). Although perceptual and articulatory constraints are the primary posited source of English-learning children's difficulty with affricates, fricatives, and liquids, the frequency of sounds in a particular environment also plays an important role in the age and order of phoneme mastery. Children from language environments with a greater frequency of occurrence of certain less universally common sounds (e.g., liquids, fricatives) tend to produce these sounds earlier and better, suggesting the early influence of the ambient language. For example, Russian children who are exposed to many palatalized consonants as well as nonpalatalized consonants typically master the palatalized ones first (Zharkova, 2004). Other research in non-English monolingual language environments has shown ambient language effects on the greater earlier accuracy of fricatives and affricates (Pye, Ingram, & List, 1987) as well as dorsal sounds and multisyllabic words (Gildersleeve-Neumann, 2001; Teixeira & Davis, 2002). In addition, children in other language environments may produce words with different error patterns. For instance, it is common for young Finnish children to have initial consonant deletions, an atypical phonological process in English-speech acquisition (Vihman & Velleman, 2000).

Ambient language effects on speech sound acquisition are also observed in bilingual children. Bilingual children may show an effect of each language on their productions within that language, such as reported for a Hindi–English simultaneous bilingual child who used predominantly monosyllables in English and predominantly disyllables in Hindi (Bhaya Nair, 1991; Vihman & Croft, in press).

Simultaneously bilingual children produce different segments and word shapes depending on which of their two language environments they are in (Holm & Dodd, 1999; Holm, Dodd, Stow, & Pert, 1999; Johnson & Lancaster, 1998; Kehoe & Lleo, 2003; Keshavarz & Ingram, 2002). Mixing of errors has been observed in the carryover of the phonetic and phonological properties of one language to the other, resulting in greater rates of error when compared to monolingual peers (Goldstein & Cintrón, 2001). Although bilingual children may follow the general developmental path, their speech patterns might still be expected to be influenced by the phonology/ies of their native language(s). Clearly, the large, cross-linguistic literature on typical and atypical speech sound acquisition provides a rich database for comparative research on speech development in children suspected to have CAS.

Prosody

Prelinguistic Period

Infants' early discrimination of prosody (see Speech Perception) is followed by production of language-specific prosodic patterns. By 6–12 months, their vocalization patterns reflect the dominant prosodic contours (e.g., falling vs. rising pitch; Whalen, Levitt, & Wang, 1991) of the ambient language.

Linguistic Period

English-speaking children use falling intonation contours first, then rising contours, to mark phrase and utterance boundaries (Tonkava-Yampolskaya, 1973). Typical English-learning children have been shown to use frequency, amplitude, and duration appropriately to mark sentential emphasis (Skinder, Strand, & Mignerey, 1999), as do children with speech delays (Shriberg, Aram, & Kwiatkowski, 1997b, 1997c). The primary period for the development of prosody occurs from approximately 5 to 8 years of age (Local, 1980; Wells, Peppe, & Goulandris, 2004). However, even typically developing children may not have adultlike comprehension and production of prosody until 10 or 12 years of age (Allen & Hawkins, 1980; Morton & Trehub, 2001). In English, later-developing prosodic functions include the production of compound words, rise–fall or fall–rise prosody on a single word to convey emotion, high rising pitch to request clarification, accent on a nonfinal word to convey emphasis (e.g., "I want a black bus"), and the comprehension of another person's use of accent to emphasize a certain part of an utterance (Wells et al., 2004).

Children's stress patterns parallel the dominant stress patterns of their languages in late babbling and early words (e.g., predominantly trochaic stress-first patterns in English; iambic stress-last patterns in French; Vihman, DePaolis, & Davis, 1998). By 2½ years of age, English learners' vowel durations differ appropriately in stressed versus unstressed syllables (Smith, 1978), and they are able to produce weak syllables in initial position (e.g., the first syllable of "giraffe") and between two stressed syllables (e.g., the second syllable of "telephone"; Gerken, 1994; Kehoe & Stoel-Gammon, 1997). Children with speech delay but not apraxia cease to delete such weak syllables by age 6 (Velleman & Shriberg, 1999); as reviewed later, children suspected to have CAS may persist in such patterns. By age 6, typically developing children have different coarticulatory and temporal patterns depending on the syllable structure of a word. For example, Dutch-learning children have coarticulation and duration patterns that differ with the metrical structure of the word (Maassen, Nijland, & van der Meulen, 2001; Nijland, Maassen, van der Meulen, et al., 2003).

Speech Perception

Prelinguistic Period

Between birth and 2 months of age, human beings are already able to discriminate among languages with different rhythmic patterns (Mehler et al., 1988), among words that

differ by number of syllables (Bijelac-Babic, Bertoncini, & Mehler, 1993), and among different vowels (Kuhl & Miller, 1975) and different consonants (Eilers, 1977; Eilers & Minifie, 1975; Jusczyk, Murray, & Bayly, 1979; Levitt, Jusczyk, Murray, & Carden, 1988). Some of these capacities may be innate, but others are learned through perceptual experience. For example, the neonate attends longer to her own mother's voice (DeCasper & Fifer, 1980) and to her own language prosody in conversational speech (Mehler et al., 1988). Speech perception skills become more and more language-specific as the child approaches 1 year of age. By 10 months, infants display preferences for stress patterns (Jusczyk, Cutler, & Redanz, 1993; Morgan, 1996; Weissenborn, Hohle, Bartels, Herold, & Hofmann, 2002), consonants, and sequences of consonants and vowels from their own language (Gerken & Zamuner, 2004; Jusczyk, Friederici, Wessels, Svenkerud, & Jusczyk, 1993; Jusczyk, Luce, & Charles-Luce, 1994). Furthermore, at 10–12 months, babies are less able than at earlier ages to discriminate segmental contrasts that are not relevant to their own languages (Werker & Tees, 1984).

Linguistic Period

At 4 years of age, children with nonapraxic speech ddisorders are significantly worse than their typically speaking peers at discriminating commonly misarticulated sounds from sounds that are generally substituted for them. There is a significant difference between the two groups' ability to identify whether a sound was produced correctly versus incorrectly within a word (e.g., [Œu Œ] vs. [r u Œ] for "cat") (Rvachew, Ohberg, Grawberg, & Heyding, 2003).

Language

Children with speech delay often also have language delays, especially in expressive morphology (Paul & Shriberg, 1982; Rvachew, Gaines, Cloutier, & Blanchet, 2005). Their morphological errors cannot be attributed to speech difficulty. For example, Rvachew et al. reported that children with speech delay omitted /∅/ and /ɛ / in final position more often in grammatical morphemes (plural, third person singular) than in uninflected words even though the phonetic complexity was the same in both contexts. Furthermore, frequency of omission of morphemes was correlated with mean length of utterance ([MLU] in words), not with articulatory skills.

A few studies have investigated profiles of children with speech delay only versus those who also have language delay. In a study of 15 children with speech delay only and 14 children with both speech and language delay, Lewis, Freebairn, Hansen, Iyengar, and Taylor (2004) reported that the speech patterns of the two groups were similar at school age (ages 8–10 years), with frequent liquid simplifications and distortion errors. The speech delay only group persisted in immature consonant harmony/assimilation errors, whereas the speech and language delay group produced frequent final consonant deletions, which, although not described in this study, may have been associated with morphological deficits. As part of a larger study, Nathan, Stackhouse, Goulandris, and Snowling (2004) followed 19 children with speech delay only and 19 children with both speech and language delay from preschool (age 4;6) through kindergarten (age 5;8) to school age (age 6;9). In preschool, the speech delayed only group performed better overall on articulation assessments as well as on language measures. At the two later ages, the children with speech delay only seemed to have normalized (caught up to typically developing peers), whereas the deficits of the speech and language delay group persisted. To date, there are no studies that have systematically compared specific language patterns (e.g., morphological vs. syntactic errors) in children with language delay only to those of children with both speech and language delay.

Metalinguistic/Literacy Skills

Studies indicate that at age 4, children with speech delay are at higher risk for impaired phonological awareness skills (e.g., rhyme matching, onset segmentation, onset matching) compared to children who are typically developing, although in one such study significant differences between the two groups' early literacy skills were not detected (Rvachew et al., 2003). Between the ages of 6 and 8 years of age, children without speech sound or language disorders develop the metalinguistic ability to explicitly identify the number of syllables in a word and the placement of individual sounds or clusters within words (Marquardt, Sussman, Snow, & Jacks, 2002). Children with a familial history of speech delay/disorder (including those with CAS, as discussed in the following section) are at higher risk for literacy difficulties, especially if they also demonstrate language delay (Bird, Bishop, & Freeman, 1995; Larrivee & Catts, 1999; Lewis et al., 2004; Nathan et al., 2004; Webster & Plante, 1992). In a study of 47 children with speech deficits only (reportedly including CAS), speech and language deficits, or no speech or language deficits, Nathan et al. reported that preschool language ability, especially for expressive language, is a strong predictor of later phonemic awareness skills. These investigators also found that persistent speech difficulties (beyond age 6;9) are strongly predicted by concurrent deficits in phonemic awareness. An ASHA document (ASHA, 2001) includes useful information about phonological awareness development and disorders.

<center>Summary</center>

The large literature on typically developing speech has been reviewed from the perspective of the key areas of possible developmental differences between children who are typically developing, children with nonapraxic speech sound disorders, and children diagnosed with CAS. The goal was to provide a reference basis for the review of CAS literature to follow. Notable areas of difference were found in the early and seemingly effortless development of vowels and prosody in children who do not have CAS. Important areas of overlap in the speech of typical learners and children suspected to have CAS include the gradual development of consonant repertoires and phonotactic structures (syllable and word shapes) and gradual decreases in both variability and inconsistency. Research indicates that children with any type of speech sound disorder are at increased risk for language and literacy difficulties, although the literature reviewed in the next section indicates that children suspected to have CAS may be at considerably greater risk.

<center>SCIENTIFIC FOUNDATIONS:
BEHAVIORAL RESEARCH IN CAS</center>

Studies of the developmental neurobiology of CAS are expected to provide an understanding of the relevant neural substrates and identify useful early diagnostic biomarkers. Even when such information becomes available, speech-language pathologists will still need to use behavioral tools (e.g., standardized tests, informal assessment measures, parental observations, reports from other professionals) to provide the individualized profiles needed to differentiate children suspected to have CAS from children with other types of speech-language disorders. To date, as previewed in the Introduction and Overview, no one test score or behavioral characteristic has been validated to differentially diagnose CAS (i.e., there are no necessary and sufficient markers). The present section provides an extended review of behavioral research in CAS.

<center>Overview</center>

In both research and clinical settings, the diagnostic challenge is to differentiate CAS from speech delay, dysarthria, and other speech sound disorders. Many of the speech and other behaviors (i.e., *signs*) thought to be associated with CAS are also found in children with more broadly defined speech sound disorders (McCabe, Rosenthal, & McLeod, 1998). The differentiation between apraxia and dysfluency (stuttering,

cluttering) is a less common clinical need, although there are some behavioral overlaps and children suspected to have CAS may go through periods of dysfluency (Byrd & Cooper, 1989). The question of differentiating language behaviors occurring in CAS from those in specific language impairment (SLI) is also highly challenging, a question that has only recently begun to be addressed in the clinical literature (Lewis et al., 2004).

Behavioral variables that have been studied in association with CAS can be divided into six major domains: nonspeech motor, speech production, prosody, speech perception, language, and metalinguistic/literacy. Within each of these domains, reference is made to core deficits in timing, programming, and sensorimotor coordination. However, due to the lack of a definitive diagnostic marker for CAS, conclusions from studies seeking to identify such markers are limited by issues of participant selection and circularity. When study participants are selected based solely on clinician referrals, it is difficult to determine which diagnostic criteria were used by individual clinicians, how clinicians differentially weighted their criteria, and the amount of agreement within and between clinicians. In fact, clinical agreement has not been demonstrated in recent studies. Davis et al. (1998) and Forrest (2003) reported high degrees of clinical disagreement among practicing speech-language pathologists in their criteria for diagnosing CAS. There are similar problems in research contexts. In a recent CAS study, two research teams were able to reach only 55% agreement on the assignment of 35 speech sound disordered study participants to CAS or non-CAS groups (Shriberg, Campbell, et al., 2003). This diagnostic uncertainty among both clinicians and researchers is the primary barrier to research on the underlying nature of CAS. As suggested by Strand (2001), another significant research constraint is the heterogeneity of children with CAS due to the co-occurrence of other disorders with CAS, as well as individual differences in compensatory behaviors that may be secondary to the primary deficits.

Definitional circularity is most evident when study participants are selected based on the presence of certain signs and those signs or derivatives of them are part of the study's descriptive findings. Because of the presumed low prevalence of CAS, it is difficult to conduct large scale studies of children with etiologically undifferentiated speech sound disorders hoping to identify speech and other characteristics unique to CAS. Moreover, there is increasing evidence that the signs of CAS not only vary among children with the disorder, but also change as children mature (Lewis et al., 2004; Shriberg, Campbell, et al., 2003). Thus, although there may be neural phenotypes that persist beyond the developmental period, it is likely that behavioral markers will need to be developed for several developmental epochs. CAS may be a complex of signs, with varying neurologic, motor, and behavioral characteristics that can be identified only by its unique profile over time (Ekelman & Aram, 1983). It is vital not to confuse descriptions with explanations; the varying behavioral consequences of a disorder can obscure as well as clarify its fundamental nature. Within each of the six behavioral domains listed above, we report findings supporting associations with CAS, followed by some perspectives on theories of CAS.

Behavioral Domains Studied in CAS

Nonspeech Motor Behaviors

Nonspeech motor behaviors are primarily used to differentiate children suspected to have CAS from children with various types of dysarthria, although there is some overlap between the two motor speech disorders. Nonspeech motor signs of CAS that are most commonly proposed in the literature (some of which are also cited as signs of dysarthria) include the following: general awkwardness or clumsiness, impaired volitional oral movements, mild delays in motor development, mildly low muscle tone, abnormal

orosensory perception (hyper- or hyposensitivity in the oral area), and oral apraxia (e.g., Davis et al., 1998; McCabe et al., 1998; Shriberg et al., 1997a). The nonspeech motor features typically listed for oral apraxia are impaired volitional oral movements (imitated or elicited postures or sequences such as "smile–kiss") and groping (e.g., Davis et al., 1998; McCabe et al., 1998; Shriberg et al., 1997a). Murdoch, Attard, Ozanne, and Stokes (1995) documented weaker lingual muscles and reduced tongue endurance in children who demonstrated oral apraxia than in typically developing children; a nonapraxic, phonologically disordered control group was not included in the study. Dewey, Roy, Square-Storer, and Hayden (1988) found that limb, oral, and verbal apraxia tend to co-occur in children. They highlighted the transition difficulties (moving from one action in a sequence to the next) exhibited by children "with a specific deficit in verbal sequences of consonant-vowel syllables" (p. 743) and noted that repetition of the same action was far less of a problem. They also stressed the volitional aspect of the disorder, as did Maassen, Groenen, and Crul (2003) and D. Nelson (1995). Specifically, Dewey et al. (1988) found that demonstrating the action of an object was a problem for their participants with CAS only when the children were miming the action without the object in hand. Crary and Anderson (1991) also noted that compared to children without a diagnosis of CAS, children with this diagnosis had slower rates and less accurate performance on sequences of hand and facial movements.

Speech Motor Behaviors

Motoric aspects of speech, especially *repetitions of syllabl*es (maximum repetition rate [MRR]) and *productions of alternating syllables* (diadochokinesis [DDK] or alternating motion rate [AMR]), are commonly used to diagnose CAS both clinically and for research participant selection. The utility of these measures has been verified in several research studies, including Davis et al. (1998), McCabe et al. (1998), Nijland et al. (2002), Thoonen, Maassen, Gabreëls, and Schreuder (1999), and Thoonen, Maassen, Wit, Gabreëls, and Schreuder (1996). Thoonen et al. (1996), for example, reported that maximum sound prolongation of vowels (e.g., producing /ɒ/ for as long as possible) and MRRs for single syllables (e.g., /˘] ɶ] k] / etc.) differentiated children with a diagnosis of spastic dysarthria from both children with a CAS diagnosis and those who were typically developing. Maximum sound prolongation of fricatives and maximum repetition rate of trisyllabic sequences (/˘] ɶ] k] /) differentiated children with apraxia from those who were typically developing. Thus, the differences between children with CAS and those who were typically developing were only significant for the more complex tasks (prolongations of more difficult consonant sounds; sequences of different syllables). Control groups of children with other speech sound disorders of unknown origin were not tested. Lewis et al. (2004) found significant differences between preschool and school-age children with CAS and matched children with non-CAS speech delay in their ability to repeat nonwords and multisyllabic words, with the CAS group performing more poorly. Children with CAS also had significantly lower Total Function scores on the Robbins and Klee (1987) oral-motor assessment, which includes DDK. Moreover, children with CAS had more difficulty on the Fletcher Time-by-Count test of DDK (S. G. Fletcher, 1978) at school age.

Lists of the speech behaviors proposed to characterize CAS abound in the research and clinical literatures. Frequent characteristics include some features that clearly are shared with other speech sound disorders (McCabe et al., 1998), including slow development of speech, reduced phonetic or phonemic inventories, multiple speech sound errors, reduced percentage of consonants correct, and unintelligibility. Commonly proposed characteristics (Davis et al., 1998; McCabe et al., 1998; Shriberg et al., 1997a) that are less likely to be found in children with nonapraxic speech sound disorders include

reduced vowel inventory, vowel errors, inconsistency of errors, increased errors in longer or more complex syllable and word shapes (especially omissions, particularly in word-initial position), groping, unusual errors that "defy process analysis," persistent or frequent regression (e.g., loss of words or sounds that were previously mastered), differences in performance of automatic (overlearned) versus volitional (spontaneous or elicited) activities, with volitional activities more affected, and errors in the ordering of sounds (migration and metathesis), syllables, morphemes, or even words. However, many of these features are found in children who do not fit the overall pattern of CAS (McCabe et al., 1998), leading some reviewers to question their diagnostic specificity for CAS (e.g., Macaluso-Haynes, 1978). Moreover, as discussed later, many of these posited features are not consistent with a deficit in praxis. For example, motor speech theories typically assign selection and sequencing of sounds, syllables, and words to a processing stage that *precedes* the planning and programming of movements needed to realize these units as manifest speech. Error patterns that are not consistent with a praxis deficit but are especially common in children suspected to have CAS need to be studied to understand whether or not they are causally related and, if they are, to identify the explanatory mechanisms.

Detailed studies of differences between children suspected to have CAS compared to those with typical development or with other subtypes of speech delay have sought to identify the diagnostic characteristics of CAS. As noted previously, all such studies have research design limitations due to the lack of certainty that the children suspected to have CAS indeed have this disorder. As just one of many examples, Maassen et al. (2001) reported that children with CAS have less predictable speech errors than children who are typically developing. These authors provided useful acoustic data documenting a lack of systematic effects of given phonetic contexts on certain sounds in the speech production of children with CAS. However, because of the absence of a control group of children with other speech sound disorders and because the only inclusionary criterion information provided was that "Clear cases of [CAS] were selected according to clinical criteria described by Hall, Jordan, and Robin (1993) and Thoonen et al. (1996)" (Maassen et al., 2001, p. 146), it is difficult to evaluate claims that variability of this type may be a unique feature of CAS.

Speech sampling methods may also be crucial to interpretation of findings. Shriberg et al. (1997b) reported that a group of children, chosen by five individual researchers as exemplars of these researchers' diagnosis of CAS, did not have any speech production errors in conversational speech that could be used to differentiate them from control children with speech delay of unknown origin. A potential constraint on these findings, as discussed more recently in Shriberg, Campbell, et al. (2003), is that these findings were based on conversational speech samples, rather than on children's responses to challenging speech production tasks designed to evoke more discriminative error patterns.

In a widely cited study of speech motor behaviors, McCabe et al. (1998) attempted to identify potential features of apraxia retrospectively (from clinic files) in a mixed group of 50 children with speech disorders, 9 of whom had been identified as having apraxia of speech by their speech-language pathologists. They described characteristics of CAS in some of the 50 children who had been classified as speech disordered (non-CAS). The characteristics most often identified in the total group were "changed level of awareness of own speech errors, problems with imitation of speech, breathing difficulties/asthma/allergies, decreased performance on DDK tasks, and presence of 'soft' neurological signs or minimal brain damage" (McCabe et al., 1998, p. 113). These were

also the most commonly reported symptoms in the subset of children who previously had been identified by their speech-language pathologists as having apraxia. However, McCabe et al. reported "inconsistent speech performance," vowel errors, and incorrect production of "lingual phonemes" (/ ʤ /, /— /) as best differentiating this CAS group from their other participants. Other differences that distinguished the two groups quantitatively included slow development of speech, idiosyncratic sound substitutions, and syllable omissions.

Lewis et al. (2004) compared a group of children suspected to have CAS to two other groups of children longitudinally: one group with non-CAS speech sound disorders only and one group with both speech and language disorders. The CAS group was selected based on both a diagnosis of CAS by the child's speech-language pathologist and on the child meeting at least four out of eight criteria for CAS. The CAS group differed from the speech disorder group, especially at school age, on syllable structures, sound sequencing, vowel and voicing errors, unusual types of errors, and the persistence of their error patterns. At school age, the children with CAS had more speech errors overall, more unusual errors, and more syllable sequencing errors in conversational speech than the children with both speech and language disorders (but see McNeil, Robin, & Schmidt, 1997, for an alternative interpretation of phoneme-level sequencing errors in AOS).

Ball, Bernthal, and Beukelman (2002) used a very careful procedure to identify participants with CAS, including diagnosis by a speech-language pathologist and administration of the Screening Test for Developmental Apraxia of Speech (Blakeley, 1980) and the Tasks for Assessing Motor Speech Programming Capacity (Wertz, LaPointe, & Rosenbeck, 1984). A panel of three speech-language pathologists then rated each child on a scale of 1 (*not CAS*) to 5 (*definitely CAS*) based on a list of 17 characteristics of CAS. The 36 children included in the study each had an average rating of at least 3. They also had other co-occurring language, social, and behavioral impairments. The purpose of the study was to attempt to identify more inclusive communication profiles of children with CAS. Following this identification procedure, an assessment battery of tests and measures was administered to the participants, and test results were subjected to cluster analysis to identify groups of participants who shared particular patterns of communication performance. Twelve of the participants who had been rated as having a high likelihood/severity of CAS had notable deficits in the following areas compared to participants in the other clusters: receptive language, vocabulary, MLU, percentage of consonants correct, intelligibility, and behavior.

Acoustic analyses have been used by several authors to characterize more precisely the speech production differences of children with CAS. Participants with CAS in these studies have demonstrated decreased differentiation of stop place of articulation (Sussman, Marquardt, & Doyle, 2000), decreased differentiation of vowels (Nijland et al., 2002), higher degrees of anticipatory coarticulation within syllables (Maassen et al., 2001; Nijland, Maassen, van der Meulen, et al., 2003), lack of impact of syllable boundaries or syllable shape on coarticulation (Maassen et al., 2001; Nijland, Maassen, van der Meulen, et al., 2003), lack of intersyllabic coarticulation, and variable idiosyncratic patterns (Nijland et al., 2002) that were less predictable acoustically in any given phonetic context (Maassen et al., 2001). Nijland, Maassen, van der Meulen, et al. (2003) further noted that children with CAS had higher scores than typically developing children on measures of coarticulation and vowel accuracy when a bite block was placed between their teeth. As noted previously, additional studies using control groups of children with other forms of speech delay would strengthen the claims of this carefully executed study series.

With respect to severity, McCabe et al. (1998) found that severity of speech impairment, as defined by the percentage of consonants correct, is correlated with the

number of features of CAS that a child exhibits even among children without this diagnosis and that CAS may be quantified on a continuum of severity as measured in this way. Relative to prognosis, Lewis et al. (2004) reported that, at school age, participants with CAS had more persistent difficulties in repeating nonsense words and sequencing syllables than participants who had previously been diagnosed with a non-CAS speech sound disorder.

Prosodic Characteristics

A consistent finding in the literature is that individuals suspected to have CAS have atypical prosody, including a variety of types of prosodic deficits (Davis et al., 1998; McCabe et al., 1998; Shriberg et al., 1997a). Also often noted are variations in rate, including both prolonged sounds and prolonged pauses between sounds, syllables, or words, which gives the listener the impression of staccato speech (*syllable segregation*), with sounds, syllables, or words produced as independent entities lacking smooth transitions to other structural units (Shriberg, Green, Campbell, McSweeny, & Scheer, 2003). As in other motor speech disorders, reduced range of or variable pitch, as well as reduced range of or variable loudness, gives the listener the impression of monotone, monoloud speech, respectively. Variable nasal resonance (sometimes hyponasal, sometimes hypernasal) has also been noted in the clinical research literature. Duration, pitch, and loudness combine to form the percept of stress in English; this, too, is commonly reported to be atypical in children suspected to have CAS. In a series of studies, Shriberg et al. (1997a, 1997b, 1997c) documented excessive-equal stress (all or most syllables in a word or sentence receiving prominent stress) in approximately 50% of each of three different samples of children suspected to have CAS. They noted that younger children with CAS were also rated as more involved than children with speech delay on perceptual measures of rate and resonance. However, excessive-equal stress was the only feature that reliably distinguished any of several CAS subgroups from control groups of children with speech delay of unknown etiology. Those children who exhibited excessive-equal stress also produced more distortions of early consonant sounds than the other children, but their error types (relative proportions of substitutions vs. omissions vs. distortions) and their severity and variability levels did not differ from those of the children with speech delay. The authors speculated that the children with a diagnosis of CAS who did not demonstrate excessive-equal stress may either have been incorrectly diagnosed or were possibly exhibiting another type of CAS. In a later article, Shriberg, Campbell, et al. (2003) suggested that the presence of stress errors may change over time within an individual with CAS. Odell and Shriberg (2001) further noted that prosodic disturbances may be different in adults with acquired apraxia of speech versus children with CAS. Children with CAS in their sample had excessive-equal stress, but, in contrast to the sample of adults with acquired apraxia of speech, did not have inappropriate phrasing or rate.

Velleman and Shriberg (1999) completed metrical analyses of the lexical stress patterns of children with CAS who had inappropriate stress, children with CAS who did not have inappropriate stress, and children with speech delays of unknown origin. They found that the pattern of stress errors of the children with CAS did not differ substantially from the error pattern of younger, typically developing children, suggesting that either the children with CAS were misdiagnosed or that such errors reflect prosodic delay rather than disorder. That is, participants with CAS who had inappropriate stress tended to either omit or overstress weak (unstressed) syllables, especially in the initial position of words, as do typically developing 2-year-old children. However, whereas the children with speech delay ceased to make such errors after the age of 6, lexical stress errors of this type had persisted into adolescence in the participants suspected to have CAS with inappropriate stress.

Stress differences in CAS have also been examined using acoustic analyses. Munson, Bjorum, and Windsor (2003) reported that the vowel durations, fundamental frequencies, vowel intensities and f_0 peak timing of stressed syllables produced by children with CAS were appropriate despite the fact that the children were perceived as producing inappropriate stress patterns. Skinder et al. (1999) also found that children with CAS marked stress in the same ways as children who were typically developing, although there was more variability within the CAS group. Again, listeners had judged the children with CAS as less accurate in their stress production than the typically developing children, but the acoustic measures used did not identify the source of these perceptions. Skinder et al. suggested that listeners were confused or distracted from attending to prosodic details by the higher number of segmental errors produced by the children with CAS. Shriberg, Campbell, et al. (2003), in contrast, reported that ratios based on acoustic measures of stressed versus unstressed syllables differed in children with CAS who perceptually were noted to produce excessive-equal stress compared to control children with speech delay. The acoustic differences were quantitative rather than qualitative. Thus, it may not be that children with CAS have uniquely different stress patterns. Rather, it may be their inability to fully contrast stressed versus unstressed syllables that leads to the impression of inappropriate stress patterns. Note that these and associated stress findings are consistent with findings reviewed earlier indicating that the phonetic distinctiveness of vowels/diphthongs and consonants is reduced in the speech of children suspected to have CAS.

Speech Perception Characteristics

A few studies have addressed the hypothesis that children suspected to have CAS have deficits in auditory perception, auditory discrimination, and/or auditory memory. Bridgeman and Snowling (1988) reported that compared to control children, children with CAS have more difficulty discriminating sound sequences in nonsense words. Groenen and Maassen (1996) found that children with CAS did not have difficulty identifying the place of articulation of a consonant but did have difficulty discriminating consonants with subtle acoustic differences associated with place of articulation. Furthermore, deficits in place discrimination were found to be correlated with deficits in accurate production of place. Maassen et al. (2003) also reported that compared to children with typically developing speech, children with CAS had poorer identification as well as poorer discrimination of vowels. Given the likelihood of phoneme-specific relationships between production and perception in children with other speech sound disorders (Rvachew, Rafaat, & Martin, 1999), an important research goal for theories of CAS is to determine if the speech perception deficits described above are replicable and whether they are unique to children with CAS.

Language Characteristics

There is general agreement in reviews of the literature that children suspected to have CAS typically also have significant language deficits (e.g., Crary 1984, 1993; Ozanne, 1995; Velleman & Strand, 1994). As with the perceptual findings reviewed in the previous section, a research challenge is to determine how such constraints are associated with the praxis deficit in planning and programming that defines CAS. One possibility is that language impairments are a consequence of having any type of disorder affecting neurological development (Robin, 1992). In response to another possiblity—that all expressive language deficits in children with CAS are due to their speech involvements— Ekelman and Aram (1983) documented language errors in a group of children with CAS that were clearly not due to the children's phonological deficits. Their participants made incorrect choices of pronouns and verbs. They also failed to invert auxiliary (helping) and

copula (*be*) verbs in questions. More recently, Lewis et al. (2004) found that language impairments were more significant and persistent in children with CAS than in children with non-CAS speech sound disorders. The authors concluded that language symptoms are "a key aspect of the disorder" (p. 131) based on the following observations: (1) gains in articulation did not eliminate language deficits (e.g., morphological omissions of plural, possessive, third person singular, and past tense markers are not simply due to an inability to produce final consonant clusters); (2) receptive as well as expressive language deficits were noted, although expressive language consistently lagged behind receptive language; and (3) there was a strong family history of language impairment in the families of the children with CAS (Lewis et al., 2004).

Language symptoms that might differentiate children suspected to have CAS from children with SLI have been implied in the literature. For example, Velleman and Strand (1994) modeled CAS as a disorder of hierarchical organization, which suggests that language errors should take the form of part–whole and sequencing difficulties. Lewis et al. (2004) failed to find language differences between children with CAS and children with a combined language and non-CAS speech sound disorder on standardized tests at the preschool level. As indicated above, they did identify more persistent receptive and expressive language difficulties among the children in the CAS group at school age; analyses of the children's spontaneous spoken and/or written language would have strengthened this claim. Overall, the literature remains inconclusive on whether there are differences in the language profiles of children with CAS versus children with SLI or with combined language and nonapraxic speech sound disorder.

Metalinguistic/Literacy Characteristics

Children with any sort of speech production deficit are at higher risk for difficulty with phonological awareness, which itself is a "critical element of literacy development" (Justice & Schuele, 2004, p. 378). Although CAS researchers frequently cite literacy and other academic difficulties as a characteristic of the disorder, few studies have explored this topic and some have been limited by the lack of a speech delayed comparison group. For example, Marion, Sussman, and Marquardt (1993) demonstrated that children with CAS have more difficulty perceiving and producing rhymes than do children with typically developing speech. Marquardt et al. (2002) similarly showed that children with CAS score lower than typically developing children on metaphonological (phonological awareness) tasks, such as tapping to count the syllables in a word and using blocks to represent the structure of a word (e.g., using black blocks to represent the consonants and white blocks to represent the vowels in the word *blue* [i.e., black black white]). Given that a history of speech delay puts a child at increased risk for phonological awareness deficits, it will be important to cross-validate such interesting findings with control groups who have speech sound disorders other than CAS.

Lewis et al. (2004), as cited previously, found that children with CAS had deficits in word attack, word identification, and spelling in comparison to children with speech disorders only. Their participants with CAS also scored significantly lower on tasks requiring them to spell unpredictable words, compared to scores from children with a combination of language and nonapraxic speech disorders.

Finally, it is useful to note that children suspected to have CAS have sometimes been described as having increased self-awareness of their own speech production limitations (McCabe et al., 1998; Velleman & Strand, 1994). This is a special type of metalinguistic awareness (the ability to reflect consciously about or comment on linguistic elements, structures, or processes) that, to date, has not been addressed in controlled research.

CAS research to date has almost exclusively focused on English-speaking participants in several countries, with the exception of several cohorts of Dutch children with CAS studied by Maassen and colleagues (Maassen et al., 2001, 2003; Nijland, Maassen, & van der Meulen, 2003; Thoonen et al., 1999). Although not exploring cross-linguistic similarities or differences between individuals with CAS who speak Dutch or English, these investigators have used CAS criteria from studies of English-speaking participants. Dutch and English are similar in phonetic and phonotactic properties and it appears that features of CAS may be similar in the two language environments. The Committee did not identify any studies that have compared aspects of CAS in individuals speaking different dialects of English or speaking languages that differ markedly from English in phonetic, phonemic, and phonotactic properties. Cross-linguistic studies of CAS could provide greater understanding of the effects of language and culture on its short- and long-term expression.

Theories of CAS

Theories about the nature of CAS are based on a limited number of observations that seem to be shared among most researchers. There appears to be general agreement that (a) the behaviors associated with CAS may vary from child to child and from time to time within the same child, (b) severity of expression may range from mild to severe, and (c) CAS is a symptom complex, rather than a unitary disorder (Dewey, 1995; Hall, 1989; Le-Normand, Vaivre-Douret, Payan, & Cohen, 2000; Lewis et al., 2004; Maassen, 2002; McCabe et al., 1998; Shriberg, Campbell, et al. 2003; Strand, 2001; Velleman & Shriberg, 1999). Beyond such observations, theories of the nature of CAS can be divided into the following two general categories: frameworks that focus on suprasegmental perspectives and those that emphasize sensorimotor perspectives.

Suprasegmental Perspectives

There appears to be widespread agreement that syllables and prosody are affected in more profound, distinctive ways in CAS than are other aspects of speech or phonology. Some researchers have hypothesized that deficits in the syllabic framework of speech result in prosodic symptoms (Davis et al., 1998; Maassen, 2002; Marquardt et al., 2002; Nijland, Maassen, van der Meulen, et al., 2003). Others have proposed the reverse: that fundamental prosodic deficits affect syllable and segment production (Boutsen & Christman, 2002; Odell & Shriberg, 2001). Other researchers have emphasized the critical roles of timing (e.g., Shriberg, Green, et al., 2003) and sequencing deficits (e.g., Thoonen et al., 1996) as core features underlying many of the other segmental and suprasegmental characteristics of CAS.

Sensorimotor Perspectives

Several theoretical frameworks for CAS posit that core deficits are in the relationship between perception or sensory processing and some aspect of motor processing. Maassen (2002), for example, proposed that deficient sensorimotor learning leads to weak prelinguistic articulatory–auditory mappings, which in turn fail to support full phoneme-specific mappings. He noted that "higher-level knowledge…must be acquired by the child via the problematic speech production and perception skills" (p. 265). Maassen suggested that unlike typically developing children, children with CAS seem to process real words more similarly to the way they process nonsense words. Maassen speculated that such processing renders their linguistic systems (e.g., lexical representations) less able to support online language processing tasks. Barry (1995a), Boutsen and Christman (2002), and Odell and Shriberg (2001) focused on the related issue of online self-monitoring and feedback systems. These investigators proposed that children with CAS may have weak sensorimotor feedback loops or decreased ability to respond to such feedback. Thus,

children with CAS may be unable to either benefit immediately from feedback in order to self-correct or to appropriately grade actions, or may be unable to use this feedback to alter incomplete representations or motor plans for future retrieval. Such a sensorimotor deficit could also underlie proposed difficulties in automating motor programs, such that each word production must be planned anew (Barry, 1995a; Nijland et al., 2002; Nijland, Maassen, van der Meulen, et al., 2003).

Deficits in the preprogramming, programming, and execution of speech motor events (Klapp, 1995, 2003) have each been proposed as a core deficit in CAS. Unfortunately, explicit definitions for each of these processes are, themselves, a source of debate in associated literatures. Most theoretical proposals place the source of the speech production difficulties in CAS further "upstream" than the actual execution of the motor plan. Marquardt, Jacks, and Davis (2004), for example, attributed high inconsistency levels in children with CAS to "lack of neural instantiation of phonemic representations" (p. 142) and unstable motor programs for word targets. They noted that increases in accuracy are associated with increases in stability (i.e., decreases in inconsistency), presumably reflecting more specific, stable motor plans for words. A further common theme in all such discussions is a deficit in integration or coordination across different levels proposed to be relevant to speech production (and, in some cases, speech perception as well). Such levels include syllabic, phonemic, or motor representations; motor plans and/or programs; and neuromotor group networks. Thus, multiple levels of speech motor processing, and the relationships among them, have been implicated in processing perspectives on CAS.

In prior decades, discussion of the core deficit(s) in CAS was often framed as a debate between linguistic/psycholinguistic perspectives versus motor perspectives. Currently, this opposition is more appropriately described as a debate between motor + linguistic versus motor-only views. As discussed previously, the primary question is how to reconcile the linguistic behaviors that have been associated with CAS in the research literature—differences in speech perception, phonological awareness, phonological patterns, and expressive language—with the core problem of praxis from which this disorder takes its name. In widely cited papers on AOS, McNeil and colleagues have argued on formal grounds that such deficits cannot be accommodated as core features of AOS (McNeil, 1997; McNeil et al., 1997). Rather, if present, they likely reflect secondary consequences of apraxia of speech. Thus, from a theoretical perspective, the lines are drawn fairly sharply. Following McNeil's rationale, if research validates deficits in speech processes that precede planning/programming of movement sequences for speech, reconsideration would have to be given to the appropriateness of the term *apraxia* for this clinical entity.

Summary

Studies of the behavioral features of CAS have been limited by methodological constraints and circularity in subject ascertainment criteria and by incomplete controls (e.g., comparing children with CAS only to children who are typically developing, rather than to children with other speech sound disorders). These limitations notwithstanding, there appears to be a research consensus that children suspected to have CAS often have deficits in any or all of the following domains: nonspeech motor behaviors, motor speech behaviors, speech sounds and structures (i.e., word and syllable shapes), prosody, language, metalinguistic/phonemic awareness, and literacy. Thus, at present, CAS presents as a complex of signs that varies across children and within the same child over time. An important corollary concept, however, is that many of these behavioral characteristics are also observed in children with other forms of speech sound disorders. Notably, although we restricted our search to literature published in English, we found few studies of children

with CAS who speak languages other than English. Finally, theories of the nature of CAS continue to reflect difficulty in explaining the relationship of a core deficit in motor planning and/or programming to deficits in other domains observed as part of the symptom complex seen in children with CAS.

SCIENTIFIC FOUNDATIONS:
GENETIC AND NEUROBEHAVIORAL RESEARCH IN CAS

One research approach that meets some of the needs discussed in the previous section is studies of children suspected to have CAS who share some common biological difference or disorder. This section reviews findings from two examples of this approach. One approach is to study families of children with *idiopathic* (i.e., a disorder of unknown origin) CAS to determine if affected family members share one or more genetic differences not found in unaffected family members. The second type of design is studies of children reported to have CAS as a secondary feature in a well-characterized complex neurobehavioral disorder, such as fragile X syndrome. In each of these two designs— studying children with idiopathic CAS and studying children with CAS as secondary signs within complex neurobehavioral disorders—information on the molecular genetics and developmental biology of the disorder can be used to develop an eventual explanatory account of CAS. Specifically, controlled investigations can be designed to study associations between the genotypic (genetic) characteristics of children suspected to have CAS and phenotypic (biobehavioral) manifestations of the disorder. Genotype/phenotype studies are widely reported in complex neurobehavioral disorders but have only recently begun to appear in the genetics literature on speech sound disorders.

CAS Research in the KE Family

A striking example of the productivity of studying genetic antecedents of CAS is the programmatic study series of a four-generation London family referred to as the *KE* family. Extensive research on this family, approximately 50% of whom have an orofacial apraxia, apraxia of speech, and cognitive-linguistic involvements, has had wide-ranging scientific impact in a number of disciplines in the life sciences. At the time this report was prepared, however, two constraints associated with the findings reviewed here have been perceived to limit the clinical impact of this study series on research and practice in CAS. First, due to the array of cognitive, language, motor, psychosocial, and possible craniofacial involvements in affected KE family members, researchers have questioned the value of generalizations from these findings to children with CAS. That is, the affected individuals in this family appear to have significantly more involved clinical profiles than reported for children suspected to have idiopathic CAS, as described in the previous section. Such differences may be more quantitative than qualitative, given that, as noted previously, most views of CAS characterize it as a *symptom complex*, which, by definition, suggests concomitant involvements in multiple domains. A second perceived limitation on generalization of findings from the KE studies to CAS is that the mutations in the gene identified in this family have not been found in many children suspected to have CAS or in children with other verbal trait disorders. Several published and unpublished molecular genetic studies of speech and language disorders have reported negative findings, although recent studies have implicated *FOXP2* deficits in some other families with speech problems apparently consistent with CAS: For reviews, see citations at the end of this introduction. Thus, in genetic epidemiology terms, the gene responsible for CAS in the affected KE family members appears to have low attributable risk in the general population.

As reviewed next, the Committee views studies of the KE family as a model of the type of programmatic research that may lead to an eventual understanding of one

class of etiological origins of CAS—CAS due to familial or new (sporadic) genetic differences. The following chronologically sequenced sections summarize findings for the KE family at four overlapping levels of methodological observation: descriptive-linguistic, genetic, neuropsychological, and neuroimaging. Table 1 provides additional technical details on findings in each of the latter three topics, including text relevant to the present focus on CAS excerpted from these primary sources. Extended syntheses of this large body of studies authored by the principal investigators are available in several excellent sources: Fisher, Lai, and Monaco (2003), Marcus and Fisher (2003), Newbury and Monaco (2002a, 2002b), and Vargha-Khadem, Gadian, Copp, and Mishkin (2005). The Online Mendelian Inheritance in Man (OMIM; 2007) database will continue to provide up-to-date reviews and bibliographies of associated genetic research.

Descriptive-Linguistic Findings

Despite the wide-ranging, cross-disciplinary impact of research on the KE family, there are few published descriptions of the segmental and suprasegmental error profiles of affected individuals. Hurst, Baraitser, Auger, Graham, and Norell (1990), the first paper by the U.K. (London and Oxford) researchers, included brief case summary paragraphs for 6 family members. These reports primarily described the speakers' apparent impairment in the organization of manual movements for signing and speech movements, with the clinical speech profile of affected family members interpreted by the researchers as consistent with CAS ("developmental verbal dyspraxia"; p. 352).

The Hurst et al. (1990) report was followed by a series of papers by Canadian researchers (primarily at McGill University) providing descriptive-linguistic analyses of selected affected KE family members. Using a variety of corpora, they interpreted their findings to suggest that the core deficit in these individuals was in their grammatical morphology (Gopnik, 1990a, 1990b; Gopnik & Crago, 1991; Matthews, 1994). More relevant for the present focus, Fee (1995) provided a perceptually based comprehensive description of the consonant errors of 8 affected family members sampled at two points in time. She reported that, even as adults, these speakers had deletion and substitution errors, especially on final consonants and consonant clusters. Goad (1998) provided a thorough analysis of the grammatical impairment in plurals in 5 affected adult family members, focusing on alternative theoretical explanations in prosodic versus morphological domains. Also, in a study assessing knowledge of lexical stress rules, Piggott and Kessler Robb (1999) reported that affected family members had considerably more incorrect and variable judgments of what constitutes appropriate lexical stress than unaffected family members. Although the investigators in this group did not use the term *apraxia* or *dyspraxia,* they reported that affected family members produced polysyllabic words that had "prominent pauses separating them" and that were "evenly stressed" (p. 61). Thus, prosodic impairment has been described as a key feature shared among affected family members.

Genetic Findings

As indicated in Table 1, the molecular genetic findings for the KE family began with the report by Fisher, Vargha-Khadem, Watkins, Monaco, and Pembrey (1998), which identified a region on chromosome 7 in affected family members that was subsequently narrowed to a susceptibility locus (i.e., a region of increased risk) on 7q31 termed SPCH1. This finding was the bridge between the earlier descriptive-linguistic characteristics summarized above and later identification of the *FOXP2* gene within the SPCH1 region. Two studies from the London/Oxford research group (Lai et al., 2000; Lai, Fisher, Hurst, Vargha-Khadem, & Monaco, 2001) provided information on this transcription gene, *FOXP2* (see Table 1). As a transcription gene, the protein products of *FOXP2* influence the function of other genes, which in turn reportedly may affect both linguistic and

sensorimotor aspects of speech and language acquisition. The U.K. group is conducting two large-scale projects to identify all genes "downstream" of *FOXP2* to determine how products of these genes may contribute to speech-language acquisition and disorder (cf. Marcus & Fisher, 2003).

A number of studies (e.g., Liégeois et al., 2001; MacDermot et al., 2005; Tyson, McGillivary, Chijiwa, & Rajcan-Separovic, 2004; Zeesman et al., 2006) have supported the association of *FOXP2* with apraxia of speech, as well as with a variety of other deficits first observed in affected members of the KE family. In addition, more recent research related to *FOXP2* and the KE family has also suggested possible mechanisms by which CAS and dysarthria may co-occur. Morgan, Liégeois, Vogel, Connelly, and Vargha-Khadem (2005) used FMRI and electropalatography (EPG) to study 5 affected members of the KE family and 5 sex-, age-, and handedness-matched controls. In addition to brain abnormalities reported previously in the motor cortex, the EPG data were reportedly consistent with speech sound distortions, with excessive variability in lingual-palatal contacts. Morgan and colleagues suggested that the *FOXP2* mutation in the KE family has disrupted the development and function of the brain regions involved in both planning and execution of speech movements (i.e., the latter process consistent with dysarthria). Finally, Shriberg et al. (2006) described a mother and a 19-year-old daughter who from early ages were treated for apraxia of speech associated with a balanced 7;13 translocation affecting *FOXP2*. Detailed speech and prosody analyses indicated that the mother's and daughter's speech profiles are consistent with both apraxia of speech and spastic dysarthria.

As indicated in the introduction to this section, findings from the KE family have prompted widespread interdisciplinary interest in the *FOX* family of genes. Examples at the time this report was in preparation include studies tracing the evolutionary history of *FOXP2* (e.g., Enard et al., 2002) and studies describing transcription processes and other molecular features of *FOXP2* and the larger family of *FOX* genes (e.g., Bruce & Margolis, 2002; Ferland, Cherry, Preware, Morrisey, & Walsh, 2003; Takahashi, Liu, Hirokawa, & Takahasi, 2003; Tamura, Morikawa, Iwanishi, Hisaoka, & Senba, 2003; B. Wang, Lin, Li, & Tucker, 2003; Zhang, Webb, & Podlaha, 2002). A major finding for definitional issues in CAS is that, as described in the studies cited immediately above, both *FOXP1* and *FOXP2* genes are expressed (switched on) widely in the brain. Importantly, these locations include many of the primary neuroanatomic sites that subserve speech-language development and processing. Additional work focusing on these genes and their counterparts in animals has suggested the potential to develop animal models for speech-language disorders; for example, Teramtizu, Kudo, London, Geschwind, and White (2004) in songbirds and Shu et al. (2005) in mice.

A number of studies in the emerging discipline of linguistic genetics (genetic studies of verbal traits and disorders) have sought to determine whether deficits in *FOXP2* are linked to other neurobehavioral disorders, including language impairment, dyslexia, and autism. As indicated previously, with the exception of a finding in autism (Gong et al., 2004), *FOXP2* deficits have not been found in children with other neurobehavioral disorders (see OMIM for current findings).

Neuropsychological Findings

Following their initial rejoinders to the Canadian group's interpretation of the communicative deficit in affected KE members (P. Fletcher, 1990; Vargha-Khadem & Passingham, 1990), the U.K. research group (i.e., Hurst et al., 1990) published four descriptive papers. The papers described findings from an extensive battery of neuropsychological and other measures given to family members and a number of control groups, including information on affected individuals' articulatory and prosodic

involvements. As shown in Table 1, Vargha-Khadem, Watkins, Alcock, Fletcher, and Passingham (1995) reported that the orofacial apraxia used as the phenotype for affected family members was accompanied by an array of deficits in other verbal and nonverbal domains, involving both comprehension and production. In particular, they summarized their alternative descriptive-explanatory perspective on the KE family as suggesting "a broad phenotype which transcends impaired generation of syntactical rules and includes a striking articulatory impairment as well as defects in intellectual, linguistic, and orofacial praxic functions generally" (Vargha-Khadem et al., 1995, p. 930).

Among other findings in two subsequent papers, Alcock and colleagues (Alcock, Passingham, Watkins, & Vargha-Khadem, 2000a, 2000b) reported that affected family members' deficits involve both comprehension and production of rhythms, as assessed using verbal and nonverbal (i.e., tapping) modalities. These authors speculated that a core problem in timing may underlie the performance deficits of affected family members on the diverse comprehension and production tasks included in the protocol.

The fourth paper (Watkins, Dronkers, & Vargha-Khadem, 2002) provided extensive neuropsychological information on cognitive-linguistic involvements in affected family members, who were compared to a sample of adults with aphasia. These findings are of particular interest for issues addressing similarities and differences in acquired adult AOS and CAS, as well as suggesting neostriatal (basal ganglion) involvement in praxic deficits in movement sequencing and procedural learning. If such findings are replicated in studies of other individuals with *FOXP2* or other genetic deficiencies, they will have important implications for assessment and treatment of CAS.

Neuroimaging Findings

As sampled in Table 1, a series of neuroimaging findings in affected members of the KE family has provided unprecedented information on neuroanatomic structures and circuits associated with this subtype of orofacial apraxia and apraxia of speech. Findings have possible implications for the developmental neurobiology of CAS symptoms and, more generally, speech-language acquisition.

In the first neuroimaging study of the KE family, Vargha-Khadem et al. (1998) reported that affected KE family members have diverse, bilateral neuroanatomic differences from unaffected members, primarily involving the neostriatum and associated neural circuits. Although the *FOXP2* gene was identified 2 years later as the gene deficit transmitted to affected family members, the authors' speculations about alternative genetic origins of these findings (e.g., see the following excerpts) remain relevant:

> Our data suggest that development of the neural mechanisms mediating the acquisition of fine oromotor coordination (both vocal and nonvocal) and of speech and language are interdependent, such that abnormality in the one will be associated with abnormality in the other. . .
>
> . . . a central abnormality affecting speech production could have a cascading effect resulting in intellectual defects . . .
>
> At this stage, we cannot discount the alternative possibility that the different components of the phenotypic profile are the consequence of abnormalities in several different neural networks resulting from disruption of either a single gene or even several contiguous genes (Vargha-Khadem et al., 1998, p. 12700)

As summarized in Table 1, the study by Watkins, Vargha-Khadem, et al. (2002) reported significant differences in white matter volumes bilaterally in affected compared to nonaffected KE family members and controls, with affected family members having both larger and smaller volumes at different neuroanatomic sites. These morphometric data underscore the complexity of the pathophysiology of CAS in these family members. Using

different neuroimaging methods, Belton, Salmond, Watkins, Vargha-Khadem, and Gadian (2003) provided additional neuroanatomic findings, again supporting bilateral involvements and morphological differences in areas that subserve both motor and language processing. Finally, Liégeois, Baldeweg, Connelly, Gadian, and Vargha-Khadem (2003) used both functional neural imaging methods and verbal processing measures to attempt to relate structural findings to behavioral findings in the affected individuals. The extended discussion of neural and neurocognitive findings in this paper provides a promising research agenda for studies in process on the genetic substrates of speech-language challenges.

<div align="center">CAS Research in Complex Neurobehavioral Disorders</div>

The many research reports indicating that CAS occurs in children with diverse neurologic disorders (e.g., as a sequela of neuronal migration disorders, infection, or trauma) were not reviewed in this document. The Committee did attempt to review a representative sample of studies reporting CAS in complex neurobehavioral disorders. Although both contexts for CAS provide promising avenues for research in all forms of CAS, most of the latter had disappointingly little technical information on the speech and prosody characteristics of the children reported to have CAS. However, these studies typically are rich in information on the neurophysiological pathways for each disorder, with implications for alternative descriptive-explanatory levels for an eventual understanding of CAS (e.g., genetic, biochemical, neuromotor). Table 2 includes brief descriptions of findings from a sample of such studies.

Autism

Limb apraxias, oral apraxia, and apraxia of speech have been frequently reported for children with autism or a pervasive developmental disorder (e.g., Boyar et al., 2001; Page & Boucher, 1998; Rogers, Bennetto, McEvoy, & Pennington, 1996; Seal & Bonvillian, 1997). Well-controlled studies are needed to test the hypothesis that apraxia of speech is more prevalent in autism than as occurs idiopathically in the general population. At the time this report was in preparation, several studies in process were studying this question using contemporary inclusionary/exclusionary criteria for both autism and CAS.

Epilepsy

CAS has been noted as comorbid with or a sequela of several forms of epilepsy, including benign rolandic epilepsy and autosomal dominant rolandic epilepsy, the latter of which is a rare form associated with more severe and long-term communicative disorders. Scheffer et al. (1995; see Table 2) provided interesting research hypotheses on the diagnostic significance of comorbid epilepsy and apraxia, again underscoring the value of studying apraxia in the context of well-characterized neurological and complex neurobehavioral disorders.

Fragile X Syndrome

Fragile X syndrome is a genetically transmitted complex neurobehavioral disorder in which speech and prosody deficits are associated with reduced intelligibility (Roberts, Hennon, & Anderson, 2003). Reports indicate that some of these deficits overlap with diagnostic criteria for CAS, but the measures used to assess the nature of speech and prosody involvement have typically not been well developed. At the time this report was in preparation at least one research study in process was attempting to replicate the Spinelli, Rocha, Giacheti, and Ricbieri-Costa (1995) findings (see Table 2) of apraxia of speech in 40% of a small sample of children with fragile X syndrome.

Galactosemia

Some form of CAS reportedly also occurs in 40%–60% of children with one of the several genetic forms of the metabolic disorder, galactosemia (Elsas, Langley, Paulk, Hjelm, & Dembure, 1995; Hansen et al., 1996; C. D. Nelson, Waggoner, Donnell, Tuerck, & Buist, 1991; D. Nelson, 1995; Robertson, Singh, Guerrero, Hundley, & Elsas, 2000;

Webb, Singh, Kennedy, & Elsas, 2003). At the time this report was in preparation, a study of CAS in galactosemia was in process using a relatively large sample of children with this disorder.

Rett Syndrome

Limb and speech apraxia are reportedly part of the sequence of neurological dysfunctions that characterize the degenerative course of expression of Rett syndrome. Because the apraxic disorder is so profound that children at this stage essentially do not speak (Bashina, Simashkova, Grachev, & Gorbachevskaya, 2002; Schanen et al., 2004), it is difficult to study speech apraxia in individuals with this neurobehavioral disorder. Genetic studies indicate that the molecular regions involved in Rett syndrome include susceptibility genes for a number of disorders reported to involve speech-language deficits (N. J. Wang, Liu, Parokonny, & Schanen, 2004).

Chromosome Translocations Involving Deletions and Duplications

One of the most active and productive areas of genetic research in complex neurobehavioral disorders involves the identification of persons with translocations that affect speech processing. The case study reported in Weistuch and Schiff-Meyers (1996) (see Table 2) illustrates the potential for CAS research in chromosomal translocations. Recall that it was a child with a translocation involving a breakpoint in chromosome 7 that helped the U.K. investigators identify the SPCH1 susceptibility region for the apraxic disorder found in the KE family. Somerville et al. (2005) reported a child with chromosome duplications affecting genes at 7q11.23 (the Williams-Beuren syndrome microdeletion locus) who has "severe delay in expressive speech." Kriek et al. (2006) also described a child with a duplication in the same region who reportedly also has significant speech delay (cf. Tassabehji & Donnai, 2006). These two papers have prompted a large-scale study now in process seeking to determine if duplications of this locus are present in children who reportedly have CAS. Lichtenbelt et al. (2005) described a child and 4 other reported cases with a rare supernumerary ring chromosome on 7q. All 5 cases have severely delayed expressive speech. Finally, Shriberg, Jakielski, Patel, and El Shanti (Shriberg, 2006) described 3 siblings with an unbalanced 4q;16q translocation whose speech and prosody profiles also are consistent with CAS and with dysarthria.

Summary

Research on the genetic bases of CAS is emerging in genetic studies of families in which CAS appears to be inherited and in genetic studies of individuals with chromosomal disorders that include CAS in a symptom complex. Although the complex of cognitive, linguistic, sensorimotor, and craniofacial involvements reported for some members of the KE family is not routinely observed in other children suspected to have idiopathic CAS, the extensive neuropsychological and neuroimaging findings from family members with deficits in the *FOXP2* gene have motivated widespread research efforts to understand the role of this gene in phylogenetic (in a species) and ontogenetic (in an individual) development of communication. Recent case studies are beginning to report other genomic regions of interest on chromosome 7 and on other chromosomes that appear to be associated with severe speech delay consistent with CAS. At the time this report was in preparation, a total of 35 cases (including 15 affected members of the KE family) had been reported in which severe speech sound disorder suspected to be CAS has been associated with genetic differences (Shriberg, 2006). There are only sparse research literatures, to date, on CAS in the context of neurological and complex neurobehavioral disorders. Such studies have the potential to contribute significant information to our understanding of the origins of this disorder and its pathophysiology.

SCIENTIFIC FOUNDATIONS:

ASSESSMENT RESEARCH IN CAS

Overview

Assessment is a broad construct encompassing many clinical decisions, including those related to diagnosis, severity of impairment, prognosis, and treatment focus. Diagnostic aspects of assessment serve as the focus of this section because diagnosis has a central role in clinical practice and in research, where it is fundamental to participant selection and description. Although of obvious importance for comprehensive treatment planning, the co-occurring problems that have been identified in persons with CAS (e.g., in expressive language and literacy) are not discussed here.

Several books published during the past decade have described diagnostic methods for CAS (e.g., Caruso & Strand, 1999; Hall et al., 1993; Velleman, 2003). Such resources and, in fact, every publication related to assessment identified for inclusion in this report have taken the position that the diagnosis of CAS falls within the professional responsibility of the discipline of speech-language pathology. Inspection of the more widely cited sources indicates that they typically address many of the issues and variables in assessment noted in the present discussion. However, because standardized tests for diagnostic assessment of CAS do not have the quality of evidence associated with peer reviewed research, review of these sources is outside of the scope of this report. The Buros *Mental Measurements Yearbook* series (e.g., Plake & Impara, 2001; Plake, Impara, & Spies, 2003) provides detailed reviews of several instruments developed primarily for diagnostic assessment of CAS.

The assessment literature was divided into three categories of peer reviewed articles: those using expert opinion for recommendations about assessment, those examining the methods currently used by clinicians and researchers, and those studying variables that may prove to be biobehavioral markers of the disorder, and thus potentially key indicators to diagnosis. The last of these categories is the most extensively studied; it also includes much that is controversial. As a group, peer reviewed articles consisting entirely of expert commentaries on CAS diagnosis are addressed briefly, but readers are cautioned to consider the potential subjectivity and the lack of transparency that is associated with expert opinion (ASHA, 2004).

Current Literature

Two articles since 1995—Crary (1995) and Davis and Velleman (2000)—included diagnosis as a major focus. Although each article offered lists of areas to examine, neither described specific decision rules linking observed behaviors with the final diagnosis nor even highly specified protocols. This is a frequent pattern in pedagogically oriented discussions of clinical methods. Notwithstanding the potential applied value of such discussions, they typically lead to considerable ambiguity when applied within a research context, where a premium needs to be placed on methodological replicability. They may also be subject to wide variability in clinical implementation.

Crary (1995) outlined a protocol intended to help clinicians identify dysarthria, oral apraxia, limb apraxia, and CAS. The protocol addresses five major areas: motor, motor speech, articulation/phonological skills, language, and an "other" category that included several additional areas. Discussion of some aspects of the protocol is relatively detailed. For example, specific suggestions are given for examining reflexes, sampling spontaneous language, and evoking and interpreting responses to diadochokinetic tasks. Specific guidelines for the identification of CAS are not provided. Rather, the assumption is that examining a child's performance on the array of recommended tasks will provide the speech-language pathologist with adequate information to arrive at a diagnosis and

engage in treatment planning.

Davis and Velleman (2000) discussed differential diagnosis within the context of a broader examination of many topics concerning CAS in infants and toddlers. Although there is considerable interest in this age group, Davis and Velleman was the only article the Committee identified that addressed the nature of signs in very young children suspected to have CAS. Their list of exclusionary and inclusionary characteristics is based on features they described as typical of older children diagnosed with CAS, but with the list modified to accommodate the more restricted language development and assessment data expected for very young children. Davis and Velleman's list of speech characteristics includes limitations in sound inventories (consonants and vowels), suprasegmental abnormalities, and variability in or lack of consistent speech patterning. They also listed six co-occurring characteristics related to the role of gestures in communication, gross and fine motor delays, clumsiness, volitional oral motor skills, diadochokinetic rates, and syntax. These recommendations notwithstanding, the authors urged extreme caution in reaching a diagnosis in very young children and suggested a period of trial intervention prior to diagnosis. As in the Crary (1995) article, these authors did not specify how the list of characteristics leads to a diagnosis, such as the number of characteristics required for diagnosis or a relative ranking of the importance of each characteristic in reaching the diagnosis.

<div align="center">Practice</div>

Two studies since 1995 have examined speech-language pathologists' perspectives on assessment of CAS, either as a primary (Forrest, 2003) or secondary (Davis et al., 1998) focus. Such studies provide insights into commonly held perspectives, developed from experiences in academic training programs, attendance at workshops and other postgraduate presentations on CAS, and from personal assessment and treatment experiences. These reports emphasize that as knowledge about the assessment of CAS accumulates from a scientific perspective, it will be important to disseminate it in ways that maximize effective clinical practice.

The Committee viewed it important to review the information from these two reports in some detail, beginning with the earlier study by Davis and colleagues (1998). Davis et al. proposed eight speech and three nonspeech characteristics for use in the diagnosis of CAS, a list that they developed from the existing research literature. The eight speech characteristics are limited consonant and vowel repertoire, frequent omission errors, high proportion of vowel errors, inconsistent articulation errors, altered suprasegmental characteristics, increased errors on longer units of speech output, significant difficulty imitating words and phrases, and predominant use of simple syllable shapes. Of these characteristics, the authors noted that several of these and other candidate features are also consistent with other types of severe speech sound disorders (i.e., "incomplete consonant repertoire, multiple speech errors, restricted production of word shapes, and poor performance on diadochokinesis"; Davis et al., 1998, p. 41).

To recruit participants for their descriptive study, Davis and colleagues (1998) described the eight characteristics listed above to practicing speech-language pathologists at conferences, asking clinicians to refer children diagnosed with this condition for possible participation in a longitudinal study. Although characteristics were described (possibly at some length) to the referring clinicians, it is unclear whether specific measures to quantify the characteristics were recommended. Further, it was not clear whether the referring speech-language pathologists were given guidance about referring children who had some, but not all, of the listed characteristics.

Twenty-two children were subsequently referred with a firm or tentative diagnosis

of CAS. Of those, only 4 (18%) were also identified as having CAS by the researchers. Findings for 5 children—4 for whom CAS was considered incorrectly diagnosed and 1 for whom CAS was considered correctly diagnosed—were described in detail to illustrate the ways in which misdiagnosed children failed to demonstrate the studied characteristics. The authors concluded that their study demonstrates the need for increased quantification of diagnostic indicators, with a focus on characteristics specific to CAS, rather than those found frequently among children with severe speech sound disorders. Three characteristics mentioned as potential candidates based on this kind of specificity are vowel misarticulations, variability of repeated productions, and suprasegmental differences, although the basis on which these three diagnostically relevant characteristics were selected was unclear. In addition, the authors warned consumers of the research literature to exercise caution when interpreting findings of previous studies in which clinician referral served as a primary basis for CAS diagnosis.

Forrest (2003) recruited as participants for her study 75 speech-language pathologists attending a workshop on CAS who indicated that they had had at least some experience with this disorder. Methodological constraints acknowledged by the author included a lack of detailed information about participant expertise and the nonrandom representativeness of the sample. Participants were asked to list three characteristics that they considered "necessary" for a diagnosis of CAS. This process produced a list of 50 characteristics, 6 of which accounted for about 51% of the responses. The 6 most frequently cited characteristics were inconsistent productions (14.1%), general oral-motor difficulties (9.3%), groping (7.9%), inability to imitate sounds (7.5%), increased errors with increased utterance length (6.6%), and poor sequencing of sounds (6.2%); the remaining 44 characteristics generated by the group were each cited by fewer than 4% of the participants. Forrest concluded that practicing clinicians may use widely varying and potentially contradictory criteria in the diagnosis of CAS. Although she did not address the extent to which clinicians may overdiagnose CAS in the course of their practice, as had Davis and colleagues (1998), Forrest's study documented likely inconsistencies in the clinical criteria used to diagnose CAS and underscored the need for research on this topic.

Behavioral Markers

The 16 studies reviewed next have yielded findings that may be informative for an eventual understanding of the behavioral characteristics suspected to define the disorder. As noted in Davis et al. (1998), not all proposed characteristics of CAS may be observed in every child suspected to have CAS, and some may be considerably more important for differential diagnosis. Perhaps that is why it has been difficult for proposed markers to meet standard statistical criteria, including high sensitivity (the proportion of true positives, or individuals with the target disorder for which the marker is positive) and high specificity (the proportion of true negatives, or individuals without the target disorder for which the marker is negative; see Sackett, Straus, Richardson, Rosenberg, & Haynes, 2000). By definition, an ideal diagnostic marker for CAS would be one that was perfectly sensitive and perfectly specific, a goal seldom met for any complex disorder. Few of the studies we identified have provided sensitivity and specificity estimates, and many have not provided inferential statistical tests to examine the likelihood that observed differences in groups were greater than chance. Other challenges posed in the search for diagnostic markers of CAS have been raised in several places in this report, including the likelihood that effective diagnostic markers may change over time (e.g., Lewis et al., 2004; Shriberg, Campbell, et al., 2003; Skinder, Connaghan, Strand, & Betz, 2000).

Table 3 is a summary of findings for 16 studies that compared the performance of a group of children suspected to have CAS to the performance of at least one other group of

children. This inclusionary criterion in our review was used because only those variables that can differentiate children suspected to have CAS from children with other closely related disorders are likely candidates for sensitive and specific diagnostic markers. Thus, although studies without comparison groups (e.g., Marquardt et al., 2002, 2004) have provided potentially valuable information about CAS—including variables that may turn out to be important diagnostic markers—they have been omitted from the present discussion. Moreover, Table 3 also does not include findings from otherwise informative studies that did not use inferential statistics to examine group differences (e.g., Barry, 1995a, 1995b; Betz & Stoel-Gammon, 2005; Peter & Stoel-Gammon, 2005) or that addressed comparisons between children and adults with developmental versus acquired forms of motor speech disorder (e.g., Barry, 1995a; Odell & Shriberg, 2001). The focus of Table 3 is on between-group differences even where these findings were not the major focus of the study. Note that findings from several of these studies were discussed from the perspective of the earlier reviews of behavioral correlates of CAS. The present emphasis is on the applied value of findings for assessment. Essentially, Table 3 provides a tabular summary of recent findings meeting the Committee's inclusionary criteria for a potential diagnostic marker of CAS.

Twelve of the 16 studies in Table 3 involve comparisons of children suspected to have CAS to participants in one other group of children. Among those 12 studies, the comparison group was children with typical speech development (8 studies) or children with speech delay (4 studies). The remaining 4 studies included two or three comparison groups. Three of these included a group of children with speech delay (Lewis et al., 2004; Shriberg, Campbell, et al., 2003; Thoonen et al., 1999), 3 included a group of children with typical speech development (Shriberg, Green, et al., 2003; Thoonen et al., 1996, 1999), 2 included a group of children with dysarthria (Thoonen et al., 1996, 1999), and only 1 included a group of children with speech and language disorders (Lewis et al., 2004). For purposes of differential diagnosis, studies that include children from other closely related disordered groups, as well as typically developing children, are obviously likely to be most helpful. In contrast, studies in which comparisons are made only to children with typical speech sound development may identify variables that distinguish between children with and without speech sound disorders of any kind (i.e., are sensitive for speech sound disorder), but are not specific for CAS.

The breadth of variables examined in these 16 studies mirrors the history of proposed underlying deficits, symptoms, and comorbid disorders in this controversial disorder (e.g., Crary, 1995; Yoss & Darley, 1974). Specifically, the major assessment domains in Table 3 include potential diagnostic markers in speech production (8 studies), prosody (4 studies), speech perception (2 studies), nonspeech oral-motor skills (1 study), and language and literacy skills (including both oral and written language; 1 study). Notice also that although half of these studies included non-English speakers, they were all children who speak Dutch, which as noted previously, is linguistically very similar to English. A critical need exists for studies identifying biobehavioral markers in children who are bilingual or monolingual in non-Germanic languages.

Although few specific findings were replicated within and across investigator groups, we note the frequency of the following two diagnostic findings for CAS in Table 3: lowered performance on tasks involving production of multiple syllables (e.g., diadochokinetic, nonsense word production, multisyllabic word production tasks) and differences or disorders on tasks involving a variety of related prosodic variables. On the first type of potential marker of CAS, the study by Thoonen et al. (1996) is unique for its findings indicating that multisyllabic word tasks differentiated CAS from dysarthria. On

the second class of potential markers summarized in Table 3, differences in the stress patterns of children with CAS were identified by Shriberg and colleagues in three studies (Shriberg et al., 1997a, 1997b; Shriberg, Campbell, et al., 2003) and in a fourth study by Munson et al. (2003). Each of these studies compared the performance of children with CAS to that of children with other speech sound disorders. Two of the studies (Shriberg et al., 1997b; Shriberg, Campbell, et al., 2003) included information on diagnostic accuracy (sensitivity, specificity) of the proposed stress markers.

Although no well-validated behavioral markers have emerged, the studies in Table 3 are interpreted as support for the perspective that children suspected to have CAS present unique patterns of difficulties in speech production, as well as in wider skill areas (e.g., areas related to nonverbal intelligence and literacy). In the present context, findings from these controlled studies suggest that many of the variables that have been proposed on the basis of clinical experience may eventually meet criteria for valid diagnostic markers. Note that these potential markers include variants of those reviewed previously in this section that are currently in use by practicing clinicians. Importantly, some of the CAS findings may reflect sequelae of underlying markers (e.g., literacy differences may reflect poor phonological foundations) rather than behavioral markers that tap core deficits.

<div align="center">Summary</div>

The literature reviewed on diagnostic assessment included recommendations cited in peer-reviewed literature, descriptions of current patterns of clinical practice, and findings from comparative studies in which between-group statistical differences suggest potential markers with high diagnostic accuracy. Currently, it appears that many of the features of CAS proposed by investigators and used by practicing clinicians overlap those of other severe speech and language disorders. Domains and measures that may have the greatest promise for sensitive and specific identification of CAS are maximal performance for multisyllabic productions and prosody. However, Williams and Stackhouse (1998) reported that rate changes far less between the ages of 3 and 5 in typically developing children than do accuracy and consistency. Such findings underscore the challenge of evaluating children in the toddler age range, at which time even in typically developing children, features such as multisyllabic productions and wide-ranging prosodic contexts are not as likely to occur.

<div align="center">SCIENTIFIC FOUNDATIONS:
TREATMENT RESEARCH IN CAS</div>

Although CAS is thought to require specialized and relatively intensive treatment (e.g., Davis & Velleman, 2000; Hall, 2000b; Strand, 1995; see later discussion), there are few recent articles that have addressed this topic and even fewer that have reported treatment efficacy findings. Methodological challenges include those described in preceding sections of this report—the lack of a standard definition for CAS, difficulties in differential diagnosis, likely significant heterogeneity in symptomatology, and changing symptomatology over time (cf. Le-Normand et al., 2000; Lewis et al., 2004). Some of the potential treatment domains that follow from the literature reviewed in the previous section include the areas of speech perception, speech production, nonspeech motor skills, prosody, language (including narrative and pragmatic skills), and metalinguistic/literacy skills. Notably, however, the few articles reviewed below, which comprise the recent treatment literature as well as selected older articles, have focused primarily on the overall communication skills of these children and on improvement in speech production. Most of the studies have been conducted within a clinical rather than school setting, making their generalizability to school practice as yet hard to gauge. Treatment involving oral-motor

exercises as a means of addressing speech-motor production was included as a small component of a multicomponent treatment approach in only one of the reviewed studies (Bahr, Velleman, & Ziegler, 1999; see Forrest, 2002, for a critique). To date, there is no treatment study in CAS that has focused on culturally and linguistically diverse populations.

As indicated in the Introduction and Overview, contemporary reviews of treatment have been heavily influenced by the emerging standards of evidence associated with evidence-based practice (ASHA, 2004; Reilly, Douglas, & Oates, 2004; Yorkston et al., 2001). Table 4 is an adaptation of the Scottish Intercollegiate Guidelines Network (SIGN) used in ASHA's 2004 technical report, *Evidence-Based Practice in Communication Disorders: An Introduction*. This system illustrates one of several currently used to classify levels of evidence for treatment studies. As this report was finalized, an amended version of the Oxford Centre for Evidence-Based Medicine system seems more likely to be adopted by ASHA.

Treatment Goals: Overall Communication and Language Skills—
Use of Augmentative and Alternative Communication (AAC)

Reduced intelligibility and comprehensibility (i.e., the ability to convey intended messages within communicative contexts; Yorkston, Strand, & Kennedy, 1996) are viewed as especially debilitating for many children with CAS (e.g., Hall, 2000a, 2000b). Treatment goals for such children have sometimes focused on facilitation of overall communication, with some studies using AAC. From the perspective of the World Health Organization's (WHO) *International Classification of Impairments, Disabilities, and Handicaps* (WHO, 1980) and *International Classification of Functioning, Disability, and Health* (WHO, 2001), interventions designed to directly improve overall communicative functioning may indirectly improve a child's ability to function within relevant social and educational contexts. This immediate focus on communicative effects differentiates AAC interventions from studies focused on behavioral deficits (e.g., speech production deficits). In a case study, Harlan (1984) described an intervention that simultaneously used manual signing to support a child's communication, while visual and tactile cueing were used to foster speech production goals. A similar case study was reported in Culp (1989).

Two relatively recent investigations (Bornman, Alant, & Meiring, 2001; Cumley & Swanson, 1999) have used case study methodologies to examine AAC interventions in a total of 4 children with CAS. Bornman and colleagues, who focused on a 6-year-old child, described use of an alternative digital voice output device. Cumley and Swanson, who studied 3 children of differing ages (preschool, elementary, and junior-high school age), used multimodal AAC that incorporated both a high-technology device and low-technology communication aids (e.g., context-specific communication board, remnant board, symbol dictionary) along with speech, gestures, and manual signs. Findings from both reports provide detailed descriptions of the implementation of AAC interventions for this population, suggesting the range of outcome behaviors that might be affected using these approaches (e.g., language, success in repairs of communication breakdown, level of communicative initiations). Both studies emphasized interdisciplinary and family involvement as important to successful implementation. Despite their descriptive value, however, these studies provide only a low level of support for the efficacy of AAC with children having CAS, due to the limited experimental control in case studies and the limited information on all measures (note the positioning of case studies at Level III in the SIGN evidence hierarchy; see Table 4).

Two additional recent studies (Binger & Light, in press; Harris, Doyle & Haaf, 1996) used the more rigorous methodology of single subject experimental designs to address the language and communication needs of participants, but studied children with

significant concomitant language disorders, developmental delay, or both. In the study by Harris et al., a 5-year-old boy with a "provisional diagnosis of developmental apraxia of speech" (p. 232) who also exhibited receptive language delays, served as the focus of a multiple baselines across communicative contexts (i.e., book reading and structured discourse) single subject design. The goal of the intervention was to teach the segmentation and combination of syntactic constituents to the child who primarily used messages consisting of a single symbol in his augmented communications. Although the child was described initially as using "multiple modalities of vocalization, gesture, facial expression and PCS [Picture Communication Symbols] to communicate" (p. 232), outcome data were limited to attainment of augmented communication goals. Over twenty-two 45-minute treatment sessions, positive effects of treatment on the use of multiple symbol communications were observed after baselining for both communicative contexts, with a greater effect observed in book reading than in structured discourse.

Binger and Light (in press) examined the effects of aided AAC modeling on the development of multisymbol messages in 5 preschoolers—2 of whom had diagnoses that included developmental delay and CAS. One of the latter 2 children had severe CAS. Both children used communication boards rather than devices with voice output, the mode used by the other 3 children in the study. A single subject multiple probe design was used. Symbol use was coded as having taken place whether the child used a graphic symbol on the AAC device, a manual sign, a consistently produced spoken "word," or a conventional head gesture (to indicate "yes" or "no," respectively). Other outcome measures examined were the number of different semantic–syntactic categories used as well as social validation measures. Both children with CAS showed positive gains across outcome measures.

Treatment Goals: Speech Production

Most treatment research has focused directly on improving speech production, using several approaches that are consistent with the prevalent views, reviewed previously, of CAS as a motor speech disorder. Writing in a professional journal, but aiming primarily at a nonprofessional audience, Hall (2000b) usefully classified CAS treatment approaches into four categories: linguistic approaches, motor-programming approaches, combinations of linguistic and motor-programming approaches, and approaches using specific sensory and gestural cueing techniques. Not included in Hall's classification, but of historical interest, are early and influential rhythmic approaches such as melodic intonation therapy (Helfrich-Miller, 1984, 1994), which was discussed in earlier treatment reviews appearing as a book chapter (e.g., Jaffe, 1984), as well as in more recent such reviews (e.g., Square, 1999).

Linguistic Approaches

Powell (1996) described a case study of a child who had been diagnosed with CAS and oral apraxia. Previous treatment at two different facilities, in which the child was typically seen for two 30-minute sessions per week, had yielded little progress. Treatment had reportedly appeared to focus on production of early developing sounds and those that were emerging in the child's phonetic inventory, as well as on the use of AAC. Powell initiated intensive treatment (four 1-hour sessions per week for a 3-month period in the summer) that included a significant focus on increasing the child's stimulability for sounds not appearing in his speech. The rationale for increasing stimulability, characterized as a phonologic approach, was based on findings from research (Powell, 1993) indicating that for children with speech sound disorders, "stimulable sounds are likely to be added to the speaker's phonetic inventory whereas nonstimulable sounds will continue to be excluded" (Powell, 1996, p. 319). This goal was supplemented by other components more typical of a

traditional articulation approach (Bernthal & Bankson, 2004), including stabilization of inconsistently used sounds in words and generalizations of known sounds to the conversational level. Over the 3-month period, the child's productive repertoire went from 11 to 17 phones, a 55% increase. The author suggested that these findings indicate the potential value of targeting stimulability in children with CAS. However, the overall lack of control, characteristic of a case study (see Table 4, Level III), provides only a low level of evidence for the findings. For example, the multiple components included in the rich intervention protocol prohibit clear assignment of the source of the findings to the stimulability activities.

Motor Programming Approaches

Motor programming approaches, which may also be termed *articulatory* or *phonetic* approaches, include integral stimulation (Strand & Debertine, 2000; Strand & Skinder, 1999) as well as a number of commercially available intervention programs (e.g., Dauer, Irwin, & Schippits, 1996; Kaufman, 1995; Kirkpatrick, Stohr, & Kimbrough, 1990; Strode & Chamberlain, 1993; Williams & Stephens, 2004). Of these examples, efficacy research has been reported only for the integral stimulation approach (Strand & Debertine, 2000; Strand & Skinder, 1999). Integral stimulation is a modification of a treatment approach developed for adults with apraxia by Rosenbek, Lemme, Ahern, Harris, and Wertz (1973). It incorporates principles of motor learning described by earlier authors in the field (e.g., Rosenbek, Hansen, Baughman, & Lemme, 1974) and also by researchers from outside of the field of speech-language pathology. Notable in the latter category is the research by Schmidt and colleagues (e.g., Schmidt, 1991; Schmidt & Bjork, 1992). Manipulation of parameters that affect motor learning, such as frequency and nature of practice opportunities and knowledge of results and performance, are fundamental elements of the integral stimulation approach. Strand and Debertine completed a multiple-baseline-across-behaviors single subject design to examine the efficacy of integral stimulation over 33 sessions (30 minutes, four times per week) for a girl (age 5;9) with low comprehensibility (10%–20%) and CAS. Speech production gains for a small number of functional one- and two-word phrases (e.g., "Hi, Dad", "Not now!", "No!") were observed when probe data for these treated items were compared against baseline and control measures.

The generalizability of Strand and Debertine's (2000) findings is limited by a lack of replication across subjects and because single subject experimental designs are not recognized within the SIGN hierarchy shown in Table 4. Nonetheless, such designs are thought to demonstrate a high degree of experimental control, especially for heterogeneous and rare participant populations for which randomized control trials may prove unfeasible or even ill-advised (Ylvisaker et al., 2002).

Combined Linguistic-Motor Programming Approaches

For the time period reported at publication of this review, approximately the past 12 years, the only published treatment study that can be readily classified using Hall's (2000b) category of combined linguistic-motor programming treatment is the research described in Bahr et al. (1999). This exploratory study described an inclusion classroom staffed by a speech-language pathologist and elementary school teacher as the context for treatment of 4 children diagnosed with suspected CAS (referred to as "oral motor impairments"; Bahr et al., 1999, p. 25), as well as 5 children with speech sound disorders. It may be described as a combined linguistic-motor programming approach because it was based on the work of Velleman and Strand (1994), who, as described in Bahr et al., proposed that CAS represents "an underlying impairment of the ability to generate hierarchical plans or sequences of behaviors—whether they be motor or linguistic or both" (p. 21). Speech production served

as a focus within a regular kindergarten curriculum in which 5 children with typically developing speech also participated. Classroom activities ranged from those focusing on oral-motor and oral sensory experiences (e.g., light touch and brushing of the face and articulators) to more phonological aspects of speech production focusing on phoneme practice. The latter activities made use of tactile cues as well as descriptive phrases to define important sound characteristics, such as those used in Metaphon (Dean & Howell, 1986). Sounds were practiced in varying phonetic contexts and, over time, in longer and more complex syllabic structures. Prosody also served as a treatment focus. If children were unable to complete activities within the group context, they received individual treatment on an as-needed basis. The authors' impression was that children made positive progress in speech production and intelligibility, but no data were provided to support those observations.

Approaches That Include Specific Sensory and Gestural Cueing Techniques

As with the class of approaches above, there have been no treatment methods during the review period that have focused on specific sensory and gestural cueing techniques, although this approach has been described as a component of intervention in many less recent papers (e.g., Bashir, Graham-Jones, & Bostwick, 1984; Chappell, 1974; Chumpelik, 1984; Hayden & Square, 1994; Klick, 1985). However, one recent paper that described what may be considered a sensory and gestural cueing approach consists of a literature review and case description concerned with the use of instrumentation to support children's speech production treatment (Gibbon, Stewart, Hardcastle, & Crampin, 1999). The authors introduced the use of electropalatography for children described as having persistent speech sound disorders, a category that can include children suspected to have CAS (although CAS was not considered as a diagnosis for the child described). Electropalatography is used to obtain detailed assessment of tongue movement and provides visual feedback regarding tongue contact with an artificial palate.

Summary

There have been few treatment studies of CAS since approximately 1995. Four treatment studies were identified, none of which met the highest level of evidence within both the hierarchy described in Table 4 and others that have been proposed (e.g., Robey, 2004). Examination of the earlier treatment literature (i.e., before 1995) failed to reveal a stronger evidence base. For example, Rosenbek et al. (1974) and Yoss and Darley (1974) reported on treatment studies falling virtually at the same level of evidence as the majority of more recent studies described above, due to their use of single- and multiple-case study methods. Earlier reported treatment studies shared current emphasis on the importance of practice, use of visual cues (ranging from mirror work to gestural cues to written words), early introduction of self-monitoring, and attention to stress production. These themes were noted in a review of this literature completed by Pannbacker (1988). Clearly, the limited evidence on treatment efficacy is one of the most clinically pressing needs in CAS research identified in this report.

PROFESSIONAL ISSUES

Earlier sections of this report have reviewed the scientific foundations for viewing CAS as a clinical entity. The Committee views the aggregate literature findings as support for the position that CAS should be included as a diagnostic entity within the class of childhood speech sound disorders. Findings also support viewing CAS as a symptom complex, with specific features varying in type and severity from child to child and over time. The available information indicates that unlike speech delay, the speech and prosody characteristics of CAS are likely to persist past the developmental period (Lewis et al., 2004). Moreover, language and metalinguistic/literacy deficits appear to often accompany

the motor speech constraints that are the core deficit in CAS. Although research to date has not provided sufficient information to support specific assessment and treatment guidelines, the following discussion includes general interim recommendations for assessment and treatment of this challenging neurobehavioral disorder.

Assessment

The Committee concludes that CAS is a communication disorder for which there is, at present, no certain genetic, neurobiologic, or behavioral marker. A well-trained speech-language pathologist with specific experience in pediatric speech sound disorders, including motor speech disorders, is the appropriate professional to assess and diagnose CAS. Referral to an occupational therapist for nonspeech, sensory-motor, or fine motor issues is often appropriate. Referral to a physical therapist is also warranted if gross motor skills or overall muscle tone are of concern, as is referral to a pediatric neurologist if neurological indicators (e.g., potential seizure activity) are present. As CAS may be a secondary diagnosis for children with autism, Down syndrome, and other widely recognized disorders with genetic and/or neurobehavioral bases, the speech-language pathologist may not be the first professional to assess the client. Whatever the history of identification or differential diagnosis, the evaluation and treatment of the child's speech sound disorder falls within the realm of clinical speech pathology. Thus, it is a speech-language pathologist who diagnoses CAS, not a neurologist or other medical practitioner. Speech-language pathologists who lack training or experience with this disorder should refer such cases to others or carry out assessments and interventions under the supervision of a speech-language pathologist with the appropriate background. Moreover, if an examiner lacks knowledge or experience in an allied area that is relevant for a particular child, such as AAC for the child with severe CAS, the examiner should make a referral to another speech-language pathologist with expertise in that area.

Overdiagnosis of CAS has become a widely discussed professional issue. As reviewed previously, speech-language pathologists appear to lack information about the key diagnostic characteristics of the disorder (Davis et al., 1998; Forrest, 2003) and research indicates that many of its features overlap with those of other speech sound disorders (McCabe et al., 1998). In view of the many diagnostic constraints reviewed in this document, it may be more appropriate in some diagnostic reports to use classification terms such as *CAS cannot be ruled out, signs are consistent with CAS,* or *suspected to have CAS*, rather than an unequivocal *CAS*. These cautions in classification apply especially to the challenges associated with diagnosis of younger children. Research has not addressed the question of the youngest age at which a diagnosis of CAS can be valid and reliable. Clinical guidelines on the appropriate minimum age for the diagnosis of CAS appear to range from under 2 years of age to under 4 years of age, including both children with idiopathic CAS and with CAS as a secondary symptom in neurological and complex neurobehavioral disorders.

Assessment of children for whom the diagnosis of childhood apraxia of speech is in question should include measures of all the domains described in this report: nonspeech oral-motor, speech production, prosody, voice, speech perception, language, and, for older children, metalinguistic/literacy skills. Of these domains, there is some consensus on the validity of the following three segmental and suprasegmental features of CAS: (a) inconsistent errors on consonants and vowels in repeated productions of syllables or words, (b) lengthened and disrupted coarticulatory transitions between sounds and syllables, and (c) inappropriate prosody, especially in the realization of lexical or phrasal stress. A thorough case history is vital. The cultural and linguistic background of each child must be fully understood and provisions made in assessment to address relevant sociolinguistic

needs.

Although research support for specific assessment procedures is limited by methodological variables discussed previously, clinically experienced researchers stress the diagnostic importance of certain key contrasts (Caruso & Strand, 1999; Davis & Velleman, 2000; Davis et al., 1998; Hall et al., 1993; Hodge, 1994; Skinder-Meredith, 2001; Thoonen et al., 1999; Velleman, 2003). Throughout an evaluation, emphasis should be on differentiating children's performance on functional/automatic versus volitional actions, single postures versus sequences of postures, simple contexts versus more complex or novel contexts, repetitions of the same stimuli versus repetitions of varying stimuli (e.g., sequential motion rates vs. alternating motion rates), and tasks for which auditory versus visual versus tactile versus combinations of cues are provided. Fluidity (smoothness) and rate as well as accuracy should be monitored, as there may be trade-offs among these variables (e.g., the child's productions are very smooth if slow compared to arrhythmic if rapid). Assessment should include performance in multiple contexts (e.g., spontaneous, elicited, imitation; syllable, single-word, phrase, sentence, discourse). At present, no standardized test incorporates all of these features and those that have been formally critiqued (e.g., Apraxia Profile [Hickman, 1997], Guyette, 2001; Screening Test of Developmental Apraxia of Speech-2 [Blakely, 2001], McCauley, 2003; Verbal Motor Production Assessment for Children [Hayden & Square, 1999], Snyder, 2005) have been found lacking in terms of important psychometric standards. Few of these recommendations have been studied in well-controlled designs, but some findings support their importance in differentiating children suspected to have CAS from those with speech delay.

Summary

Assessment and diagnosis of CAS are the responsibility of the speech-language pathologist with specialized knowledge, training, and skills in this area. The symptoms of CAS change over time and may be influenced by development in other behavioral domains. Although no single differential diagnostic marker with high sensitivity and specificity has been documented to date, there is some consensus among clinical researchers on three segmental and suprasegmental features observed in children suspected to have CAS.

Treatment

Intensity

Given the need for repetitive planning, programming, and production practice in motor speech disorders, clinical sources stress the need for intensive and individualized treatment of apraxia, especially for children with very little functional communication. There is emerging research support for the need to provide three to five individual sessions per week for children with apraxia as compared to the traditional, less intensive, one to two sessions per week (Hall et al., 1993; Skinder-Meredith, 2001; Strand & Skinder, 1999). Ideally, this should be done in as naturalistic an environment as possible to facilitate carryover and generalization of skills. Although home practice is critical for optimal progress, it cannot take the place of individual treatment provided by a speech-language pathologist who has expertise in motor speech skill facilitation. For the diverse backgrounds of children seen for early intervention, including their stages of psychological/emotional development, the Committee sees value in endorsing a treatment plan for optimum progress based on provision of intensive therapy.

Individual differences among children will also underlie rationale for changing the form, content, and intensity of treatment throughout the course of intervention. If toddler and preschool-age children are seen for early intervention that targets their speech-motor

deficits, the frequency of treatment may be able to be reduced over time. As long as the primary goal is to improve the motoric aspects of the child's speech production (i.e., more time for motor practice), individual therapy should be the preferred approach regardless of age. For children whose severity of involvement has decreased and whose treatment goals have begun to move toward language and pragmatic skills enhancement, a combination of both individual and small group therapy may also be optimal for some children, providing that a treatment focus is maintained on speech production.

For children with apraxia who also require other therapeutic services (e.g., occupational therapy, physical therapy), care must be taken to vary therapy activities to avoid fatigue. Collaborative decision-making is critical in such cases, where creative use of alternatives, such as co-treatment, should be considered (Davis & Velleman, 2000; Velleman & Strand, 1994).

Length of Treatment Sessions

In view of the Committee's information indicating that children are being enrolled for treatment of CAS at increasingly younger ages, careful consideration should be given to the length of the therapy session. If repetitive practice of speech-motor patterns is targeted in a therapy session, many children in the younger age ranges can remain engaged for only a maximum of 30 minutes per session. There are certainly those children for whom "adjustment" time is necessary prior to the introduction of more intensive treatment activities. Some service providers are allotted a certain number of minutes or hours per week of therapy time per child. Given the option between two 1-hour sessions and four 30-minute sessions, many clinical researchers strongly recommend the latter (e.g., "more sessions—less time per session"; Skinder-Meredith, 2001).

Treatment Strategies

The treatment literature in CAS indicates that the operating principles and strategies overlap those recommended for children with other speech sound disorders. Overall, the principles of motor learning theory and intensity of speech-motor practice appear to be the most often emphasized in an optimal treatment program. These recommendations include the need for distributed practice, in which speech-motor practice is carried out across a variety of activities, settings, and situations, and includes several exemplars per pattern (e.g., Strand & Skinder, 1999). Recall from the discussion above that speech requires more flexibility, less stereotyped rhythmicity, finer levels of coordination, and lower levels of strength than other nonspeech oral motor activities such as chewing, blowing, and the like. A systematic review addressing this topic is currently underway by an ASHA committee through its National Center for Evidence-Based Practice. Until the committee report is available, the consensus opinion is nonspeech oro-motor therapy is neither necessary nor sufficient for improved speech production. Another often-cited recommendation is to take advantage of other areas of strength for children with CAS by utilizing a multisensory approach to treatment. The use of sign language, pictures, AAC systems, visual prompts, and touch cues have been described as being extremely effective for children with CAS, providing functional communication while at the same time supporting and enhancing verbal speech production. Another important element for optimal progress and carryover is to involve as many important people in the child's life as possible, in a culturally appropriate manner, in understanding and completing therapy goals outside the treatment setting.

Funding Treatment

Although reimbursement and funding issues are a concern for all childhood speech sound disorders, insurance funding issues in CAS have become a topic of considerable interest. As reviewed, children with CAS are likely to require intensive services over a long

period of time. In a study on treatment outcomes from one large facility, Campbell (1999) reported that children with the diagnosis of CAS needed 81% more individual therapy sessions than children described as having a phonological disorder in order to achieve the same functional outcome. Of even more interest to funders are Campbell's findings indicating that the average cost of achieving the same functional outcome for a child with CAS was $11,325, compared to $2,000 for a child with a phonological disorder. This study reported that in addition to time needed for treatment of children with CAS, professionals needed additional time to identify the appropriate diagnostic codes for CAS, write reports, educate funders, and assist caregivers with advocacy needs in order to pursue reimbursement. Web sites such as that of the Childhood Apraxia of Speech Association of North America (http://www.apraxia-kids.org) have developed useful materials to help caregivers and others with the complex of resources to aid families and therapists in securing insurance funding.

Comorbid Conditions and Allocation of Resources

Recent research has continued to validate comorbid deficits that accompany the severe speech production constraints that characterize children with CAS. As reviewed, Lewis and her colleagues in a follow-up study reported that children with CAS are likely to have deficits in both expressive and receptive language as well as in academic areas such as reading, spelling, and written expression (Lewis et al., 2004). Lewis et al. proposed that speech-language pathologists should consider offering phonological awareness and preliteracy training as part of intervention for children with CAS at risk for language learning challenges.

For some children with CAS, therapy approaches that focus exclusively on oral output are inadequate, requiring augmentative and alternative modes of communication (Cumley & Swanson, 1999). AAC systems, of all types, require time for speech-language pathologists, other school personnel, and the child to learn and use in order to expand communication opportunities. Thus, in addition to their needs for intensive, individual speech and/or AAC treatment to maximize effective communication, children with CAS will also require therapeutic time to address a number of other issues—all of which contribute to these children becoming functional communicators who can be comfortable with and learn in the school environment. This level of need places demands on the speech-language pathologist, regardless of practice setting. It also requires that both private speech therapy providers and school-based clinicians work closely with one another and with families. Collaboration is required in order to optimally integrate the child's needs into the intervention time available and to assemble the best possible program of services for affected children.

Summary

Although the specific forms of treatment may change over time, the Committee recommends that children with CAS receive intensive services, especially in the earlier stages of intervention. The rationale for this recommendation is based on the assumption that the child's potential for normalization of speech and prosody may be substantially reduced if not addressed during early periods of growth and development. There are treatment constraints (e.g., limited funding, limited staff availability) in certain settings that make it challenging to secure intensive, individual therapy. Resources need to be made available to insurance companies, school districts, and specialized programs to provide children with CAS the best opportunity to develop functional communication. Sociodemographic issues should be addressed to ensure that all children with CAS receive the type and intensity of services needed to treat this complex motor speech disorder.

The Committee also underscores the responsibility of ASHA and its membership to

educate allied health care professionals on current perspectives in CAS so that timely referrals are made and appropriate therapeutic services are supported. This requires education at both local and national levels. More generally, speech-language pathologists must be adequately trained in areas such as differential diagnosis of childhood motor speech disorders, motor learning theory, cueing strategy usage, and other intervention techniques that clinical researchers have reported as effective. Such knowledge and skills training is the responsibility of academic training programs. New forms of partnerships must emerge among clinicians and across therapeutic settings to create intervention programs that maximize resources and address the multifaceted deficits presented by this clinical population. Finally, trends in the treatment literature indicate that professional education and collaboration are needed to enhance the resources and opportunities for children with apraxia of speech.

RESEARCH NEEDS AND COMMITTEE RECOMMENDATIONS

The Committee has attempted to provide a broad-based review of contemporary issues, findings, and directions in CAS research and practice. The summaries at the end of each section were designed to provide a digest of the key issues, findings, and directions that emerged from our review of the literature. We conclude this report with a consolidated list of research needs and the Committee's primary recommendations, the latter of which are also available in the companion document, *Childhood Apraxia of Speech: Position Statement*.

Research Needs

The Committee's primary conclusion is that, as with many other complex neurobehavioral disorders, research in CAS has not provided clear answers to the following five interdependent questions: (1) What are the biobehavioral origins of CAS? (2) What methods to diagnose CAS are valid and reliable for children of different ages and with co-occurring problems? (3) What is the prognosis for children with CAS? (4) What are the most effective ways to treat CAS? and (5) What might be done to prevent CAS and/or mitigate its impact on other areas of development? The Committee's most compelling single finding is the lack of consensus among investigators on the core diagnostic features of this disorder, thus limiting the utility of all research on optimum assessment and treatment. That is, lack of one or more necessary and sufficient diagnostic markers of CAS limits studies of the origins and neural substrates of CAS, and in turn, the scope and depth of our guidelines and recommendations for service delivery issues.

The Committee's second conclusion is that there is a need for large-scale, collaborative interdisciplinary research in CAS. CAS research clearly needs to expand to different, broader types of research models. Most of the CAS findings reviewed in this report reflect the research of individuals or relatively small groups of investigators using small numbers of participants suspected to have CAS. In comparison, emerging findings from research on such widely studied complex neurobehavioral disorders as autism, dyslexia, and language impairment reflect the research of many international, multidisciplinary collaborations. The only such research of this scope on CAS to date is the programmatic studies of the KE family.

The following list of six basic and applied needs is a brief outline of the Committee's recommendations for a CAS research agenda.

Basic Research Needs

1. Speech motor control and neurolinguistic studies using contemporary methods in such disciplines as neurophysiology, neurochemistry, neural imaging, kinematics, and acoustics to describe the pathophysiology of CAS.
2. Molecular genetic studies using contemporary genomic and bioinformatic resources to

provide an eventual account of the developmental neurobiology of CAS.

3. Epidemiological studies of CAS to delineate the gender-specific risk for this disorder in children reared in different countries, languages, races, ethnicities, and cultures.

Applied Research Needs

1. Cross-linguistic longitudinal studies to identify the core behavioral features of CAS and to develop clinically efficient diagnostic protocols for valid and reliable assessment of children at prelinguistic and later stages of CAS.

2. Studies to develop treatment programs that are appropriate for children of all ages and backgrounds with idiopathic CAS, as well as multidisciplinary studies to develop treatment programs for children with apraxia of speech occurring as the sequela of neurological deficits and within complex neurobehavioral disorders.

3. Randomized control trials and smaller-scale studies to test the efficacy of alternative treatment programs for children of all ages, types, and severities of expression of CAS, with findings enabling the development of guidelines for best practices.

<center>Committee Recommendations</center>

1. The Committee recommends that childhood apraxia of speech be recognized as a type of childhood (pediatric) speech sound disorder that warrants research and clinical attention.

2. The Committee recommends that *childhood apraxia of speech* (*CAS*) be recognized as the classification term for children with this disorder.

3. The Committee recommends the following definition for CAS:

> *Childhood apraxia of speech* (*CAS*) is a neurological childhood (pediatric) speech sound disorder in which the precision and consistency of movements underlying speech are impaired in the absence of neuromuscular deficits (e.g., abnormal reflexes, abnormal tone). CAS may occur as a result of known neurological impairment, in association with complex neurobehavioral disorders of known or unknown origin, or as an idiopathic neurogenic speech sound disorder. The core impairment in planning and/or programming spatiotemporal parameters of movement sequences results in errors in speech sound production and prosody.

> Review of the research literature indicates that, at present, there is no one validated list of diagnostic features of CAS that differentiates this disorder from other types of childhood speech sound disorders, including those due to phonological-level delay or neuromuscular disorder (dysarthria). Three segmental and suprasegmental features of CAS that are consistent with a deficit in the planning and programming of movements for speech have gained some consensus among investigators in apraxia of speech in adults and children: (a) inconsistent errors on consonants and vowels in repeated productions of syllables or words, (b) lengthened and disrupted coarticulatory transitions between sounds and syllables, and (c) inappropriate prosody, especially in the realization of lexical or phrasal stress. These features are not proposed to be the necessary and sufficient signs of CAS. As with other reported signs, they change in relative frequency of occurrence with task complexity, severity of involvement, and age. The complex of behavioral features reportedly associated with CAS places a child at increased risk for early and persistent problems in speech, expressive language, the phonological foundations for literacy, and the possible need for augmentative and alternative communication and assistive technology.

4. The Committee recommends that the American Speech-Language-Hearing Association (ASHA) adopt the position that although referrals to other professionals, including neurologists, occupational therapists, and physical therapists, may often be appropriate for associated, nonspeech issues, it is the speech-language pathologist who is responsible for making the primary diagnosis of childhood apraxia of speech and for designing,

implementing, and monitoring the appropriate individualized speech-language treatment program and/or augmentative and alternative systems and assistive technology.

5. The Committee recommends that careful consideration be given to the form and frequency of treatment for children suspected to have CAS, due to its potential to persist and to be associated with other verbal trait disorders.

Table 1. Studies of the KE family sequenced by area of study. All entries (research questions, findings, interpretations, conclusions) are quoted directly from the articles, with light editing (indicated by ellipses and brackets) used for brevity and clarity.

Area of study	Author (year)	Perspective
Genetics	Fisher, Vargha-Khadem, Watkins, Monaco, & Pembrey (1998, pp. 168, 170)	Chromosome 7 region identified that cosegregates with the speech and language disorder [in affected members of KE family], confirming autosomal dominant inheritance with full penetrance. Further analysis of microsatellites from within the region enabled us to fine map the locus responsible (designated SPCH1) to a 5.6-cM interval in 7q31. [These findings provide]...the first formal evidence for a single autosomal gene involved in speech and language disorder, and represent a major step towards its identification. … This gene is unlikely to be one specifically involved in grammar; nevertheless, it is clearly crucial for the normal acquisition of language skills...
Genetics	Lai et al. (2000) Lai, Fisher, Hurst, Vargha-Khadem, & Monaco (2001, p. 519)	Our previous work mapped the locus responsible, SPCH1, to a 5.6-cM interval of region 7q31 on chromosome 7 [Fisher et al., 1998]. We also identified an unrelated individual, CS, in whom speech and language impairment is associated with a chromosomal translocation involving the SPCH1 interval [Lai et al., 2000]. Here [Lai et al., 2001] we show that the gene FOXP2, which encodes a putative transcription factor containing a polyglutamine tract and a forkhead DNA-binding domain, is directly disrupted by the translocation breakpoint in CS.
Neuropsychology	Vargha-Khadem, Watkins, Alcock, Fletcher, & Passingham (1995, pp. 932, 933)	...the affected members were significantly more impaired on the simultaneous and successive movements than on the single movements. Thus, the praxic deficits of the affected members are not confined to articulation but also involve nonlinguistic oral and facial movements. Knowledge of neural and genetic correlates of this phenotype could provide important clues to underpinnings of the primary human faculties of speech and language as well as of the many other functions in which the affected members are also impaired.

Area of study	Author (year)	Perspective
Neuropsychology	Alcock, Passingham, Watkins, & Vargha-Khadem (2000a, pp. 17, 29)	Affected KE family members made errors on tasks requiring oral movements involving more than one group of muscles—marked impairment on tasks requiring either simultaneous or sequential movements. It is concluded that affected members of the KE family resemble patients with acquired dysphasia in having difficulties with oral praxis and that speech and language problems of affected family members arise from a lower level disorder.
Neuropsychology	Alcock, Passingham, Watkins, & Vargha-Khadem (2000b, pp. 42, 44, 45)	Affected family members were not impaired on any tasks involving musical intonation, but they were impaired on tasks involving the perception and production of rhythm. Because the tapping tests did not require oromotor coordination, impairment cannot be explained by either a language deficit or an oral praxis deficit.
		Deficits are consistent with neural findings in this family, including abnormalities bilaterally in the head of the caudate nucleus and many motor-related areas of the left hemisphere, including an area of functional underactivity in the supplementary motor area (SMA), the same area in which Halsband et al. (1993) found that lesions disrupted the production of rhythms.
		[The authors propose that] a timing and a sequencing deficit could account for deficits in both oral movements and tapping—these deficits could affect language, particularly difficulties in perceiving and producing morphemes of low phonetic substance (Leonard, 1989). [The authors consider a]...common underlying deficit explanation, versus possibility of several primary coexisting deficits, each related to a different structural or functional abnormality among the several that have now been identified in these individuals.
		When taken together with the impaired discrimination of rhythms, [the present findings are] best explained by a central deficit in the processing of timing.

Area of study	Author (year)	Perspective
Neuropsychology	Watkins, Dronkers, & Vargha-Khadem (2002, pp. 452, 454)	It is likely that both developmental disorders and acquired disorders of language have advantages and disadvantages for cognition; advantages of a developmental disorder over an acquired one are that there is presumably maximal brain plasticity and capacity for reorganization and compensation; [in contrast,] an acquired disorder could have advantages over a developmental one because of the pre-morbid period of normal development and normal use of language and other cognitive functions... We suggest that, in the affected family members, the verbal and nonverbal deficits arise from a common impairment in the ability to sequence movement or in procedural learning. Alternatively, the articulation deficit, which itself might give rise to a host of other language deficits, is separate from a more general verbal and non-verbal developmental delay.
Neuroimaging	Vargha-Khadem et al. (1998, pp. 12695, 12697, 12699)	Investigation of the three-generation KE family, half of whose members are affected by a pronounced verbal dyspraxia, has led to identification of their core deficit as one involving sequential articulation and orofacial praxis. A positron emission tomography activation study revealed functional abnormalities in both cortical and subcortical motor-related areas of the frontal lobe, while quantitative analyses of magnetic resonance imaging scans revealed structural abnormalities in several of these same areas, particularly the caudate nucleus, which was found to be abnormally small bilaterally. Although the mean scores of the affected members taken as a group fall significantly below those of the group of unaffected members on nearly every test used thus far to assess an aspect of their speech and language function and orofacial praxis, every one of the affected members is impaired individually on just three tests, namely, word repetition, nonword repetition, and simultaneous and sequential orofacial movements... On none of these three tests do the individudal scores of the affected members overlap with those of the comparison groups... [The data in this paper] confirm a major prediction derived from the affected members' phenopytpic profile and its persistence into adult life, namely, the presence of bilateral pathology in at least one and possibly other components of the motor system. Thus, the bilateral reduction in the volume of the caudate nucleas provides a plausible explanation for their orofacial dyspraxia which has persisted into maturity largely unchanged despite an origin in early development. Importantly, this same brain abnormality might also explain their verbal dyspraxia.

Area of study	Author (year)	Perspective
Neuroimaging	Watkins, Vargha-Khadem, et al. (2002, p. 465)	[The methods used] revealed a number of mainly motor- and speech-related brain regions in which the affected family members had significantly different amounts of grey matter compared with the unaffected and control groups, who did not differ from each other. Several of these regions were abnormal bilaterally. Affected family members had significantly more grey matter than controls [in some neuroanatomic areas] and significantly less grey matter than the unaffected members in others [see Liégeois, 2003, below, for summary]. The volume of the caudate nucleus was significantly correlated with the performance of affected family members on a test of oral praxis, a test of nonword repetition and the coding subtest of the Wechsler Intelligence Scale.
Neuroimaging	Belton, Salmond, Watkins, Vargha-Khadem, & Gadian (2003, pp. 194, 198, 199)	These results confirm that a point mutation in FOXP2 is associated with several bilateral grey matter abnormalities in both motor and language related regions... The association of the caudate nucleus with motor planning and seqencing, and with cognitive function...is suggestive of the role that this structural abnormlity may play in the phenotype of the affected members… In the case of bilateral abnormalities in these regions, reorganization would be compromised.
Neuroimaging	Liégeois, Baldeweg, Connelly, Gadian, & Vargha-Khadem (2003, pp. 1230, 1234)	Abnormally low levels of gray matter density have been found [in affected KE family members] in the inferior frontal gyrus, the head of the caudate nucleus, the precentral gyrus, the temporal pole, and the cerebellum, whereas abnormally high levels of gray matter density have been found in the posterior superior temporal gyrus (Wernicke's area), the angular gyrus, and the putamen. How these structural abnormalities affect brain function during language processing remains unclear. ... The aim of the present study was to determine the pattern of brain activation associated with the FOXP2 mutation in the KE family using functional magnetic resonance imaging (fMRI). We predicted that the regions that are morphologically abnormal bilaterally in the affected members would also be functionally abnormal, as evidenced by performance on language tasks. The unaffected family members showed a typical left-dominant distribution of activation involving Broca's area in the generation tasks and a more bilateral distribution in the repetition task, whereas the affected members showed a more posterior and more extensively bilateral pattern of activation in all tasks.

Area of study	Author (year)	Perspective
Neuroimaging	Liégeois et al. (2003, p. 1234)	Consistent with previously reported bilateral morphological abnormalities, the affected members showed significant underactivation relative to the unaffected members in Broca's area and its right homolog, as well as in other cortical language-related regions and in the putamen. The present findings demonstrate that the affected members of the KE family display highly atypical fMRI brain activation when performing both covert and overt verb generation tasks, as well as when repeating words... The FOXP2 gene may therefore have an important role in the development of a putative frontostriatal network involved in the learning and/or planning and execution of speech motor sequences, similar to that involved in other types of motor skills.

Table 2. Sample findings from studies of children with complex neurobehavioral disorders and reported apraxia of speech. All table entries are paraphrased summaries or quoted directly from the abstract or text of the articles, with light editing (indicated by brackets) used for brevity and clarity.

Disorder	Author (year)	Findings
Autism	Boyar et al. (2001)	Of 5 siblings with pervasive developmental disorder associated with an interstitial duplication of 15q11-q13 inherited from their mother, 4 had limb apraxia and apraxia of speech.
Epilepsy	Scheffer et al. (1995) Scheffer (2000)	Of 5 family members with benign rolandic epilepsy (BRE), all experienced oral and speech dyspraxia without prominent dysarthria; simple tasks (e.g., poking out the tongue) were difficult; they experienced difficulty with organization and coordination of high speed movements, impairing their ability to produce fluent and intelligent speech; receptive processing impairment affected the children more significantly than adults. [Authors suggest that] the findings of subtle speech disturbances in typical BRE is the key; autosomal dominant rolandic epilepsy (ADRESD) may represent a more severe manifestation of the same relationship; speech dyspraxia is intrinsically related to rolandic discharges; it is more difficult to explain why family members with BRE have longstanding difficulties of speech and language function; perhaps the impact of the epileptiform activity at a developmentally vulnerable stage results in damage.
Fragile X	Spinelli, Rocha, Giacheti, & Ricbieri-Costa (1995)	Of 10 participants with fragile X, 5 had word-finding difficulties, 1 had verbal paraphasias, and 4 had clearly dyspraxic speech. Participants with each disorder did not overlap; neither of the 2 females had clearly dyspraxic speech.
Galactosemia	Webb, Singh, Kennedy, & Elsas (2003)	Of 24 galactosemia patients consenting to formal speech evaluations, 15 (63%) had verbal dyspraxia.

Disorder	Author (year)	Findings
Rett syndrome	Bashina, Simashkova, Grachev, & Gorbachevskaya (2002)	The results of comparing clinical data and EEG traces supported the stepwise involvement of frontal and parietal-temporal cortical structures in the pathological process. The ability to organize speech and motor activity is affected first, with subsequent development of lesions to gnostic functions, which are in turn followed by derangement of subcortical structures and the cerebellum and later by damage to structures in the spinal cord. A clear correlation was found between the severity of lesions to motor and speech functions and neurophysiological data: the higher the level of preservation of elements of speech and motor functions, the smaller were the contributions of theta activity and the greater the contributions of alpha and beta activities to the EEG.
Translocations	Weistuch & Schiff-Meyers (1996)	[A case study is presented of a 5-year-old boy in whom] chromosomal studies revealed a de novo balanced translocation between first and second chromosomes. Results of the neurological, speech/language, cognitive, and play evaluations revealed a child with a severe expressive speech-language deficit but good nonverbal cognitive and communicative skills. Oral-mechanism examination appeared to be normal, but [the child] had difficulty performing oral motor tasks. The neurologist reported that the child could not smile or lateralize, elevate, or rapidly protrude tongue on command. Volitional nonverbal apraxia and apraxia of speech were well documented in this child.

Table 3. Findings for 16 studies of children with CAS compared with children in four types of comparison groups: typical speech (TS), speech delay (SD), dysarthria (DYS), and speech and language impairment (S/L). Findings that included diagnostic accuracy statistics are indicated by an asterisk; the remaining included only conventional inferential statistics. Within each major assessment domain, articles are arranged alphabetically by first author.

Major assessment domain	Author/year	Age of participants	Findings
Speech production	Nijland, Maassen, & van der Meulen (2003)	CAS: 5;5–6;10 TS: 5;0–6;10	CAS ($n = 5$) vs. TS ($n = 5$) CAS > TS in improvement in coarticulation and vowel quality in response to bite block condition, as measured using F2 values CAS < TS in compensation in response to bite block condition CAS > TS in within-subject variability of F2 frequencies
	Nijland, Maassen, van der Meulen, Gabreëls, Kraaimaat, & Schreuder (2002)	CAS: 4;11–6;10 TS: 4;9–5;11	CAS ($n = 9$) vs. TS ($n = 6$) CAS > TS in frequency of idiosyncratic coarticulation patterns, as measured using F2 CAS > TS in within-speaker variability of F2 frequencies in nonsense words CAS < TS in distinctiveness between midvowel F2 ratios, indicating less distinctiveness between vowels
	Nijland, Maassen, van der Meulen, Gabreëls, Kraaimaat, & Schreuder (2003)	CAS: 4;11–6;10 TS: 4;9–5;11	CAS ($n = 6$) vs. TS ($n = 6$) CAS > TS in degree of coarticulation effects, as measured using F2 values CAS < TS in change of durations related to syllable structure
	Shriberg, Green, Campbell, McSweeny, & Scheer (2003)	CAS: 3;5–8;0 TS: 3;7–5;8 SD: 3;5–5;5 Mean data	CAS ($n = 15$) vs. TS ($n = 30$) vs. SD ($n = 30$) CAS > TS and SD groups in the coefficient of variation ratio (i.e., the ratio of the variation of pause durations relative to the variation of speech segment durations)*
	Sussman, Marquardt, & Doyle (2000)	CAS: 5;9–6;8 TS: 5;9–6;9	CAS ($n = 5$) vs. TS ($n = 5$) CAS < TS in coarticulation effects (as measured using Locus equations of CV syllables for the consonants /b, d, g/ produced with 10 vowel contexts)

Major assessment domain	Author/year	Age of participants	Findings
Speech production (continued)	Thoonen, Maassen, Gabreëls, & Schreuder (1999)	CAS: 4;5–7;6 TS: 5;2–11;6 SD: 4;4–10;11 DYS: 5;3–16;5	CAS (*n* = 10) vs. TS (*n* = 11) vs. SD (*n* = 11) vs. DYS (*n* = 9) CAS < TS, SD, DYS in maximum rate of alternating sequences combined with maximum fricative prolongation*
	Thoonen, Maassen, Gabreëls, Schreuder, & de Swart (1997)	CAS: 6;2–7;11 TS: 6;0–7;11	CAS (*n* = 11) vs. TS (*n* = 11) CAS > TS in rate of singleton consonant errors and cluster errors CAS < TS in benefit to accuracy from real-word versus nonsense word status
	Thoonen, Maassen, Wit, Gabreëls, & Schreuder (1996)	CAS: 6;3–7;9 TS: 6;0–8;3 DYS: 6;4–10;3	CAS (*n* = 11) vs. TS (*n* = 11) vs. DYS (*n* = 9) CAS < TS in fricative prolongation, trisyllabic repetition rate, and 2 measures related to trisyllabic repetition (number of sequencing errors and number of attempts)* DYS < TS, CAS in monosyllabic repetition rate and vowel prolongation*
Prosody	Munson, Bjorum, & Windsor (2003)	CAS: 3;9–8;10 SD: 3;11–4;9	CAS (*n* = 5) vs. SD (*n* = 5) CAS < SD in matching of target stress contours during nonword repetitions, as judged by listeners despite no group differences in acoustic variables associated with stress
	Shriberg, Aram, & Kwiatkowski (1997a)	CAS: 3;3–10;10 SD: 3;4–12;0	Study 1: CAS (*n* = 14 [7 younger and 7 older]) vs. SD (*n* = 73) CAS > SD in use of inappropriate stress for a younger subgroup of participants Study 2: CAS (*n* = 20) vs. SD (*n* = 73) CAS > SD in frequency of inappropriate stress, including older as well as younger participants*

Major assessment domain	Author/year	Age of participants	Findings
Prosody (continued)	Shriberg, Aram, & Kwiatkoski (1997b)	CAS: 4;10–14;11 SD: 3;0–13;0	CAS ($n = 19$) vs. SD ($n = 73$) CAS > SD in frequency of inappropriate stress
	Shriberg, Campbell, Karlsson, Brown, Mcsweeny, & Nadler (2003)	CAS: 3;3–10;10 SD: 3;4–12;0	CAS ($n = 11$) vs. SD ($n = 24$) CAS > SD in frequency of abnormally high or low lexical stress ratio scores (composites of values obtained for 3 acoustic variables [amplitude area, frequency area, duration] for the strong syllable divided by values of those variables for the weak syllable in trochees)
Speech perception	Groenen & Maassen (1996)	CAS: 6;11–11;6 TS: 6;4–10;2	CAS ($n = 17$) vs. TS ($n = 16$) Experiment 1 CAS < TS in discrimination of place of production in stop consonants
	Maassen, Groenen, & Crul (2003)	Experiment 2 CAS: 6;9–9;5 TS: 7;0–9;7	CAS ($n = 11$) vs. TS ($n = 12$) CAS > TS in response variability in identification of stimuli from two vowel continua CAS > TS in size of just noticeable difference (*jnd*) in discrimination of stimuli from the same two continua CAS > TS in variability in *jnd* in discrimination of stimuli from the same two continua CAS > TS in measure derived from identification and discrimination measures[a]
Speech, oral and written language, and Performance IQ	Lewis, Freebairn, Hansen, Iyengar, & Taylor (2004)	Mean age at preschool testing CAS: 4;8 SD: 4;8 S/L: 4;7	CAS ($n = 10$) vs. SD ($n = 10$) vs. S/L ($n = 10$) CAS < SD at preschool testing on Goldman-Fristoe Test of Articulation (GFTA), Khan-Lewis Phonological Analysis, multisyllabic word repetition accuracy of phoneme production (MWR), nonsense word repetition (NWR) accuracy of phoneme production, oral and speech motor control protocol total functional score (TFS), Test of Language Development—Primary (TOLD–P)

Speech, oral and written language, and Performance IQ (continued)		Mean age at follow-up CAS: 8;7 SD: 8;6 S/L: 9;2	CAS < SD at school age follow-up on GFTA, NWR, MWR, Fletcher Count-by-Time of Diadochokinetic Syllable Rate, CELF–R (including Total, Receptive, and Expressive subscores), Test of Written Spelling–3, Woodcock Reading Mastery Tests—Revised, Wechsler Individual Achievement Test, selected WISC–III Performance subtests CAS < S/L at school-age follow-up on NWR; Fletcher Time-By-Count Test; Performance IQ; CELF–R Total Language, Receptive Language, and Expressive Language scores; and TWS–3 total score and unpredictable word score CAS < S/L and SD in change scores adjusted for preschool performance (residualized change) for CELF–R Expressive Language score, suggesting less change or later emerging weaknesses CAS < SD in change scores adjusted for preschool performance (residualized change) for CELF–R Total Language measure and NWR, suggesting less change or later emerging weaknesses CAS < S/L and SD groups at follow-up for the WISC–III Performance subtests: Coding, Block Design, and Block Assembly CAS < S/L and SD in change scores adjusted for preschool performance (residualized change) for CELF–R Expressive Language score, suggesting less change or later emerging weaknesses CAS < SD in change scores adjusted for preschool performance (residualized change) for CELF–R Total Language measure and NWR, suggesting less change or later emerging weaknesses CAS < S/L and SD groups at follow-up for the WISC–III Performance subtests: Coding, Block Design, and Block Assembly CAS < SD at follow up on WISC–III Performance subtests: Picture Completion and Picture Arrangements

Major assessment domain	Author/year	Age of participants	Findings
Nonspeech oral-motor skills	Murdoch, Attard, Ozanne, & Stokes (1995)	CAS: 8;8 (*M*) 2;4 (*SD*) TS: 8;2 (*M*) 2;6 (*SD*)	CAS (*n* = 6) vs. TS (*n* = 6) CAS < TS in maximum tongue strength, as measured by tongue pressures CAS < TS in ability to sustain maximum tongue pressures, as measured by pressure at onset, pressure at offset, area under the curve CAS < TS in repetition of maximum tongue movements, as measured by pressure at first repetition and pressure at tenth repetition, and across all 10 repetitions CAS < TS in pressure at last repetition and across all maximum force repetitions produced in 10 s

[a]Children with CAS were rediagnosed as showing some dysarthrias.

Table 4. Levels of evidence for studies of treatment eficacy, ranked according to the quality and credibility from highest/most credible (Ia) to lowest/least credible (IV). Reprinted from ASHA (2004, p. 2); adapted from SIGN.

Level	Description
Ia	Well-designed meta-analysis of >1 randomized controlled trial
Ib	Well-designed randomized controlled study
Iia	Well-designed controlled study without randomization
Iib	Well-designed quasi-experimental study
III	Well-designed nonexperimental studies (i.e., correlational and case studies)
IV	Expert committee report, consensus conference, clinical experience of respected authorities

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