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Prologue

# Research Priorities for Childhood Apraxia of Speech: A Long View

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ABSTRACT

This article introduces the *Journal of Speech, Language, and Hearing Research* Special Issue: Selected Papers From the 2022 Apraxia Kids Research Symposium. The field of childhood apraxia of speech (CAS) has developed significantly in the past 15 years, with key improvements in understanding of basic biology including genetics, neuroscience, and computational modelling; development of diagnostic tools and methods; diversity of evidence-based interventions with increasingly rigorous experimental designs; and understanding of impacts beyond impairment-level measures. Papers in this special issue not only review and synthesize the some of the substantial progress to date but also present novel findings addressing critical research gaps and adding to the overall body of knowledge.

A second aim of this prologue is to report the current research needs in CAS, which arose from symposium discussions involving researchers, clinicians, and Apraxia Kids community members (including parents of children with CAS). Four primary areas of need emerged from discussions at the symposium. These were: (a) What questions should we ask? (b) Who should be in the research? (c) How do we conduct the research? and (d) How do we move from research to practice? Across themes, symposium attendees emphasized the need for CAS research to better account for the diversity of people with CAS and improve the

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timeliness of implementation of high-level evidence-based practice across the lifespan. It is our goal that the articles and prologue discussion in this special issue provide an appreciation of advancements in CAS research and an updated view of the most pressing needs for future research.

The study of childhood apraxia of speech (CAS) as a field has come a long way since the 1980s when Guyette and Diedrich (1981) said CAS was "a label in search of a population" (p. 39). In the aftermath of that paper, the research community and the American Speech-Language-Hearing Association (ASHA) convened to establish a CAS research community of practice. Significant events in this timeline include the establishment of the Childhood Apraxia of Speech Association of North America in 2000, which later became Apraxia Kids (https://www.apraxia-kids.org); Research Symposia in 2002 and 2012; and the establishment of an ASHA working group on CAS, which published two seminal works in 2007—the ASHA Position Statement (ASHA, 2007a) and the ASHA Technical Report on CAS (ASHA, 2007b). The latter is one of the most cited documents in CAS (Google Scholar: 300+ times) as it laid the bedrock for the current definitions of CAS and clearly identified gaps in the research literature at that time, as shown in Table 1. The ASHA (2007b) CAS Technical Report remains a milestone in the advancement of the field; however, the time has come for it to be regarded as a historical document, replaced in part by the wealth of research that it spurred into existence, as shown in this special issue, and by a reevaluation of what research priorities exist today.

**Table 1.** Research needs identified in the 2007 Childhood Apraxia of Speech (CAS) Technical Report (ASHA, 2007b).

| Identified research needs |
|---|
| Basic research needs |
| 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. |
| Molecular genetic studies using contemporary genomic and bioinformatic resources to provide an eventual account of the developmental neurobiology of CAS. |
| 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 |
| 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. |
| 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. |
| 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. |

The purpose of this prologue is to introduce the papers featured in this special issue—cutting-edge research presented at the 2022 Apraxia Kids Research Symposium—and to report updated research priorities generated by symposium attendees through roundtable discussions interwoven with the papers of the special issue. Thus, we present a contemporary view of CAS research needs and contextualize the papers in the special issue against some of the progress that has been made in the more than 15 years since the ASHA Technical Report (2007b). It is our goal that this discussion will provide researchers and clinicians with an appreciation of the progress to date and a long view of the important work to come.

## 2022 Apraxia Kids Research Symposium

The symposium program was constructed around four broad topic areas: genetics and causes, diagnosis, treatment, and prognosis. An invited keynote presentation opened each topic, followed by 3–8 platform presentations selected by peer review (see Supplemental Materials S1 and S2 for the program and attendees, respectively). Presenters from around the world were invited to share their cutting-edge research in CAS. Generous time was allowed for discussion of each paper, not only with the speaker but also across the attendees. Each topic section concluded with a summary and moderated discussion. As the concluding event, symposium attendees participated in roundtable discussions to reflect on the program and collectively define future directions for CAS research, with an emphasis on short-term priorities. It should be noted that, although some topics were presented and discussed during individual sessions, what is reported here as research priorities are those topics and themes that emerged from the roundtable discussions and subsequent synthesis.

Research priorities were collected in three stages. First, attendees reflected on the question, "What are the top short-term priorities for apraxia research?" in small group discussions. Groups included researchers, graduate students, clinicians, and Apraxia Kids board members and staff, some of whom have children with apraxia (see Supplemental Material S2 for the list of attendees). Following the small group discussion, groups shared their collated priorities, which was followed by whole group discussion

to synthesize priorities for future CAS research, moderated by the first author. Oral discussions were audio- and video-recorded (all participants consented to recordings), and group notes were collected for later analysis. Some participants joined by Zoom and their group notes and chat contributions were also collected.

Following the symposium, an iterative process was followed to identify the key themes and subthemes from the group notes and recorded discussions. The first and second authors (P.M. and M.B.) reviewed video recordings to identify superordinate priority areas. The second author (M.B.) reviewed and coded the small group notes. Next, P.M. and M.B. reviewed the notes and video recordings to refine the themes. P.M. reorganized the themes, which was reviewed by M.B. To ensure accuracy, symposium participants were then invited to review the synthesized data and provide critical comments based on their personal notes and recollections of the discussions. Revisions were then made by the first and second author and all authors read and agreed to the final paper.

## *Research Priorities for CAS*

Four major themes emerged from the analysis: (a) What questions should we ask? (b) Who should be in the research? (c) How do we conduct the research? and (d) How do we move from research to practice? These key research needs are described below, including the subordinate priority areas and examples that were identified in the discussions at the symposium. A fifth theme, which does not relate to CAS research priorities, also emerged from the symposium discussions: "Non-Research Issues," which included discussion around advocacy, student and clinician education, and conference grants to parents. These are not discussed further but mentioned here for completeness.

### Theme 1: What Questions Should We Ask?

Critical questions related to CAS research fell into three subthemes: assessment, treatment, and outcomes (see Table 2). A common perspective emerged across subthemes—research questions should address the whole

**Table 2.** Major themes, subthemes, and examples from symposium discussions.

| Major theme | Subtheme code | Examples |
|---|---|---|
| What questions should we ask? | Assessment | Improve early identification (including genetic testing), understand how CAS changes over development, examine potential subtypes; who is vulnerable to CAS? How does it present from a whole-person perspective? |
| | Treatment | Develop knowledge and tools needed to conduct precision medicine; why do some treatments work better for some participants than others? Determine optimal treatment intensity; expand approaches for treatments for neglected populations (e.g., young children, adolescents, and adults). |
| | Outcomes | Emphasis on functional outcome measures (e.g., participation, intelligibility, social-emotional well-being); does the treatment impact quality of life in ways that are meaningful to the individual? |
| Who should be in the research? | Diversity | Studies involving speakers of non-English languages, and those from multilingual backgrounds; participants from a range of socioeconomic groups and geographic backgrounds. |
| | Lifespan | Need for longitudinal and epidemiological studies; attention to the full lifespan (infants and toddlers, adolescents, adults). |
| | Comorbidity | Examination of co-occurring conditions; involvement of neurodiverse populations. |
| How do we conduct the research? | Consumers as researchers | Young people with CAS co-designing a treatment study; funding bodies mandating inclusion of people with lived experience of CAS as members of the research team. |
| | Big data | Large scale and multisite studies, pooling data across sites. |
| | Standardized minimum data | Collection of the same data across studies and sites with the express purpose of comparison and secondary studies. |
| How do we move from research to practice | Access | Increase use of visuals/infographics to summarize research findings for lay audiences, open access publication, use of video abstracts for consumers and SLPs. |
| | Clinical pathways | Develop a single source (e.g., living handbook or clinical guide) describing current best practices for diagnosis and treatment; a standard set of clinical assessment measures. |
| | Translation and implementation | Use implementation science to plan uptake of new clinical approaches. Plan for and measure real-world effectiveness. |

*Note.* CAS = childhood apraxia of speech; SLP = speech-language pathologist.

person, with attention to complex communication profiles and functional impacts.

This marks a paradigm shift from the narrow and largely impairment-level focus of the Technical Report (ASHA, 2007b). It is of note that, while the symposium included presentations by researchers with credentials in neuroimaging or psycholinguistic modeling (e.g., researchers Cabbage, Lewis, Morgan, Preston, & Terband), the base mechanisms of CAS beyond genetics were not identified as research priorities by the symposium attendees nor do articles on these topics appear in the special issue. This is not to suggest that CAS basic science research is no longer needed or is unimportant, but that the symposium community prioritized research with immediate potential of clinical, social, or educational impact.

*Assessment.* The genetic origins and neurobiology of CAS appear more heterogenous and complex than was understood at the time of the Technical Report (ASHA, 2007b). In the past 2 decades, understanding of the genetic basis of CAS has vastly improved, and many candidate genes have been identified (see Morgan et al. 2024, for a review). Despite these advances, incorporation of genetics and genomics is not yet part of routine clinical practice (Lauretta et al., 2023, this issue). Symposium discussions emphasized the need for epidemiological studies—none of which have been conducted since the original call made by the Technical Report—and added the importance of including genetic testing in large-scale studies.

Valid and reliable diagnosis of CAS remains a challenge across clinical profiles (McCabe et al. 2024; Murray, 2022). Instead, a holistic view of the person, differentiating not only the speech impairment and determining comorbidities using a battery of tasks but also a broader view of the person's environment, characteristics, and participation is required. In this issue, Murray et al. (2023) examine the extent of agreement in expert CAS ratings—the current "gold standard" for CAS diagnosis. Poor interrater reliability highlights the need to further operationalize feature definitions, develop assessment tasks with adequate psychometric properties to detect those features, and develop a deeper understanding of how the diagnostic profile changes over time and across severity, and relates to other skills. As in the Technical Report (ASHA, 2007b), symposium attendees called for more research to improve early identification of CAS (see further discussion under the Lifespan subtheme) as well as the development of cross linguistic diagnostic tools and adaptive assessments to accommodate varying degrees of speech output and disorder severity.

*Treatment.* Maas (2024, this issue) provides a "bird's eye view" of the progress that has been made to date with respect to CAS (a) treatment efficacy/effectiveness; (b) comparative treatment efficacy/effectiveness and treatment optimization; (c) treatment candidacy, prognosis, and generalizability; and (d) treatment outcomes. As described in Maas' narrative review, substantial progress has been made in Areas 1 and 2, with considerably less research related to Areas 3 and 4 (Maas, 2024, this issue). These needs were echoed in those raised in symposium discussions. Attendees raised the need for research to develop the knowledge and tools required for precision medicine in CAS, so that the most efficacious treatments can be delivered at the ideal time and with the correct dosage. Attendees also expressed the need to expand intervention research to include individuals falling outside the school-age range (see further discussion in the Lifespan section).

This special issue includes several treatment studies that contribute toward these goals, including two investigations addressing treatment optimization. Preston et al. (2023, this issue) report findings from a randomized controlled trial examining the effect of intensive versus distributed treatment schedule and the impact of supplemental ultrasound biofeedback. As evidenced by previous research (e.g., Edeal & Gildersleeve-Neumann, 2011; Namasivayam et al., 2015), more intensive treatment resulted in greater treatment gains, and ultrasound biofeedback was useful to jumpstart treatment progress. In a single-case investigation in this issue, Thomas et al. (2023) highlight variability in CAS treatment response, demonstrating both the importance of intensive treatment and the need for further exploration of individual factors that may contribute to treatment response.

To date, very few studies have systematically investigated the role of individual-level factors on treatment response (see Maas [2024, this issue] for a review). In the only individual participant data meta-analysis in the CAS literature, Ng et al. (2022) pooled data across seven Rapid Syllable Transition Treatment (ReST) treatment studies to identify predictors of treatment outcomes. Children with greater expressive language skill, higher performance on standardized articulation testing, lower speech inconsistency, lower percentage of vowels correct, and higher baseline performance on treated targets collectively predicted better performance on treated targets. In this issue, Grigos et al. (2023) consider predictive factors related to performance of seven children who completed Dynamic Temporal and Tactile Cueing (DTTC; Strand, 2020) treatment. In contrast to the findings of Ng et al. (2022), Grigos et al. (2023, this issue) found that children who demonstrated the poorest pretreatment accuracy made the greatest treatment gains. These discrepant findings underscore the need for an investment in treatment candidacy research. It could be that participant differences in age and severity (i.e., younger, more severely impacted children in Grigos

et al. [2023, this issue]; older, more mildly impacted children in Ng et al. [2022]), or differences in the treatments provided (ReST vs. DTTC), could contribute to the conflicting findings and point to meaningful differences in treatment candidacy. This type of research is the beginning of a movement toward statistically driven precision prescribing for individuals. Much work remains to be done to achieve this goal.

*Outcomes.* In this issue, with respect to the type of outcome measures used in CAS treatment as well as their clinical significance and social validity, Maas (2024) poses the question, "What does 'work' mean, anyway?" Traditionally, relatively short-term change in speech accuracy as judged by trained listeners is the most widely reported measure of CAS treatment outcome (Murray et al., 2014). Maas (2024, this issue) suggests that while measures like speech accuracy align with the level of the impairment, changes have greater social validity if they lead to clinically significant improvement in the goals that are important to children and families. Symposium attendees advocated for increased uptake of functional outcome measures such as the Intelligibility in Context Scale (McLeod et al., 2012), which are thought to be more in line with client goals (e.g., communicative participation, intelligibility, social–emotional well-being). Several articles in this issue begin to address this call, reporting outcome measures beyond speech accuracy. In a single-case experiment exploring the feasibility of a hybrid speech and music therapy intervention for CAS, van Tellingen et al. (2023, this issue) include parent-reported changes in speech intelligibility and self-reported communication attitudes among the outcome measures. Also, Wang and Grigos (2023, this issue) examine changes in speech intelligibility following DTTC treatment as judged by unfamiliar listeners (as opposed to trained SLPs), who may better reflect the communication partners encountered by children with CAS in everyday life. Using the same sample, Grigos et al. (2023, this issue) report another analysis focused on the relationship between acoustic and kinematic measures of speech motor control with conventional auditory-perceptual ratings of word accuracy. Over the course of DTTC intervention, movement variability decreased alongside improvements in perceptual measures of word accuracy. This work contributes to improved understanding of the relationship between variability and motor skill learning and expands thinking around the standard methods for measuring treatment outcomes.

## Theme 2: Who Should Be in the Research?

To date, the preponderance of CAS research has involved a relatively homogenous group of preschool and early school age children, primarily monolingual English-speaking, who are mostly monocultural (or at least demographics in multiple sociocultural domains are unreported), and without multiple impairments or comorbidities. The key theme of "who should be in the research" highlights the need for a more inclusive research process and outcomes. Three subthemes emerged: (a) the need for greater *diversity* in terms of participant demographics (race, ethnicity, household resources, geography) and linguistic background; (b) studies addressing the full *lifespan*, with particular focus on infants and toddlers, adolescents and adults, and longitudinal studies; and (c) investigations of CAS in people with *comorbid* conditions.

*Diversity.* Almost without exception, the CAS research literature consists of single-language investigations, predominantly conducted with English speakers. Although a wide range of other European languages are represented in the literature (see Table 3), very few studies report non-European language examination of CAS (see exceptions including recent work in Cantonese [e.g., Wong et al., 2020; Wong, Wong, Velleman, et al., 2023] and Arabic [e.g., Aziz et al., 2010]).

**Table 3.** Examples of childhood apraxia of speech (CAS) literature in languages other than English.

| Language | Example citations | Topic |
|---|---|---|
| Arabic | Aziz et al., 2010 | Description of speech and language in CAS in Egyptian Arabic speaking children |
| Cantonese | Wong, Wong, Velleman, et al., 2023 | Lexical tone perception and production in Cantonese-speaking children with CAS |
| Danish | Skov, 2013 | A Danish CAS checklist |
| Dutch | van Tellingen et al., 2023 (this issue) | Speech and music therapy in CAS (case study) |
| Finnish | Martikainen & Korpilahti, 2011 | Melodic intonation and touch cue therapy (case study) |
| French | Meloni et al., 2020 | Description of CAS in French |
| German | Leonhartsberger et al., 2022 | comparison of high vs. low CAS treatment dose frequency |
| Italian | Fiori et al., 2021 | Neural changes induced by a speech motor treatment in CAS |
| Portuguese | Gubiani et al., 2021 | Validation of the DEMSS in Portuguese |
| Spanish | Olivares et al., 2020 | A Spanish CAS checklist |
| Swedish | Malmenholt et al., 2017 | CAS characteristics as described by SLPs |
| Turkish | Polat & Logacev, 2021 | Testing of CAS characteristics in Turkish-speaking children |

*Note.* DEMSS = Dynamic Evaluation of Motor Speech Skills (Strand & McCauley, 2019).

The lack of linguistic diversity is problematic for several reasons. First, clinical features of CAS may present distinctly in different languages, presenting a challenge for accurate diagnosis. For example, in Cantonese—a syllable-timed language—individuals with CAS display evidence of lexical tone errors, but not lexical stress errors as observed in English (Wong et al., 2020; Wong, Wong, Velleman, et al., 2023). Second, the lack of linguistic diversity in the present intervention evidence base makes it difficult for clinicians to determine whether a described treatment is generalizable to their clients (Murray, 2022).

Symposium attendees called for not only more single-language non-English investigations and multilanguage investigations but also development of cross-linguistic diagnostic criteria. To our knowledge, only one study has reported on cross-linguistic generalization (Gildersleeve-Neumann & Goldstein, 2015). Given that the vast majority of individuals around the world speak more than one language and many use multiple dialects, the relative absence of research describing CAS in bilingual and multilingual children is significant.

Disparities in diagnosis and access to services based on race, ethnicity, and language background occur to an alarming degree across childhood disorders, including speech and language impairments (e.g., Morgan et al., 2017). Although further study is needed to determine how this issue impacts children with CAS, it is likely that CAS is underdiagnosed or diagnosed later in children from traditionally underserved backgrounds. For example, self-report data collected by Apraxia Kids from almost 500 families revealed the average age of CAS diagnosis was lowest (3.5 years) for non-Hispanic White children—at least a year before children with other backgrounds (L. Moorer, personal communication, November 2022). Given the importance of early diagnosis and treatment for long-term communication outcomes (Highman et al., 2023, this issue), this topic warrants explicit study.

*Lifespan.* Despite the strong call in the Technical Report (ASHA, 2007b) for research into all aspects of CAS across the lifespan, to date, the focus of CAS research has heavily concentrated on the preschool and early school-aged years. As a result, apart from the work from the longitudinal Cleveland study (e.g., Lewis et al., 2023, this issue) little is known about how characteristics of CAS and related skills interact and change over the course of development. Likewise, evidence-based assessment and treatment methods are very limited for children and adults who fall outside the ages of 4–13 years.

As described in this issue by Highman et al. (2023), limited progress has been made regarding assessment, diagnosis, or treatment of infants and toddlers at risk of, or suspected to have, CAS. A notable exception to this critique is the series of studies describing Babble Boot Camp (e.g., Potter et al., 2023, this issue), which is an evidence-based intervention for infants who have classic galactosemia and are therefore at risk of CAS. Presently, preemptive treatment applies primarily to children with a known risk for CAS, as a prespeech CAS diagnosis cannot be determined. However, as Highman et al. argue, treatment can and should be provided to infants and children who are showing concerning signs of CAS before they have enough language to make a definitive diagnosis. Combined with the emergence of a clear genetic foundation to many cases of CAS (Highman et al., 2023, this issue) and the well-established value of early intervention, research involving infants and toddlers is of high priority.

Past the school-aged years, there is very little information available that describes CAS or its consequences in older adolescents or adults. Clinically, it has been believed that, with appropriate therapy, CAS resolves through adolescence. However, this is not supported by the research evidence, in particular from the singular CAS longitudinal cohort study, which has followed children from the ages of 3–6 years into adulthood (e.g., Lewis et al., 2021, 2023 [this issue]; Miller et al., 2019). There is increasing evidence that children with CAS go on to become adults with CAS, and that some of those adults continue to have speech disorders and adverse psychosocial effects (e.g., Carrigg et al., 2015, 2016; Cassar et al., 2023). From the available information, there is a pressing need to question the expectation that CAS will fully "resolve."

Prior to this issue, only three small case study reports (Preston et al., 2013; Rosenthal, 1994; Rusiewicz & Riviera, 2017) had described treatment for participants older than 14 years of age (combined $N = 5$). Preston et al. (2023, this issue) contributes an additional five adolescent participants, doubling the prior number and providing one of the first studies to address treatment optimization for adolescents with CAS. Given the mounting evidence of CAS as a lifetime disorder, further research involving adolescent and adult participants is required.

*Comorbidity.* Historically, CAS research has primarily been focused on speech production deficits; however, contemporary evidence suggests that many children with CAS likely present with co-occurring medical diagnoses and cascading developmental, social, and emotional consequences (e.g., Carrigg et al., 2016; Chenausky, Baas, et al., 2023; Nijland et al., 2015; Stein et al., 2020). CAS commonly co-occurs with language impairment (e.g., Chilosi et al., 2022; Murray et al., 2019), reading challenges (e.g., Stein et al., 2020), speech perception (e.g., Hitchcock et al., 2023, this issue; although potentially tied to language ability, see Zuk et al., 2018), and general

motor and coordination deficits (e.g., Iuzzini-Seigel, 2019). There is also a high rate of co-occurrence with other neurodevelopmental disorders, including galactosemia and Down syndrome (e.g., Shriberg, Strand, et al., 2019) as well as several genetic conditions (Morgan & Webster, 2018). Some autistic children present with CAS (e.g., Beiting & Maas, 2021; Chenausky, Baas, et al., 2023). Although the rate of co-occurrence among autistic children with more verbal skills does not appear to be elevated compared to nonautistic children (e.g., Shriberg et al., 2011), it is possible that those who use less speech may be impacted by CAS to a greater extent (e.g., Chenausky et al., 2019).

Presently, there is limited understanding of the ways in which clinical characteristics and conditions manifest and interact over the course of development. Among older children, it is possible that psychosocial comorbidities and literacy related challenges present as a consequence of the speech production difficulties encountered earlier in childhood (Cassar et al., 2023; Lewis et al., 2021). However, not all co-occurring challenges can be explained by motor speech deficits and may represent distinct disorders, originating either from coincidental or shared mechanisms (see Morgan & Webster, 2018, for a review). Symposium participants called for greater examination of complex CAS profiles, including individuals with multiple comorbidities and/or additional sources of neurodiversity (e.g., Murray, 2022). This knowledge is needed not only to design more effective tailored interventions, but also to improve understanding of CAS from a basic science perspective.

While CAS is understood as a disorder of speech production, impairments in speech perception (e.g., Hitchcock et al., 2023, this issue), language (e.g., Case & Hallin, 2023, this issue), phonological awareness (e.g., McNeill et al., 2009), literacy (e.g., Lewis et al., 2023, this issue), and nonspeech motor coordination (e.g., Iuzzini-Seigel, 2019) are also common in this population. The extent to which symptoms co-occur as well as the nature of potential causal relationships are currently unknown. For example, in this issue, Case and Hallin (2023) demonstrate that linguistic weaknesses (i.e., semantic and morphosyntactic errors) observed in narratives produced by children with CAS were not attributable to speech production errors. Likewise, Velleman et al. (2023, this issue) concluded that among children with a genetic condition, diagnosis of selective mutism and/or social anxiety disorder was not related to the severity of co-occurring speech sound disorders. To better understand the complex relationships (including the absence of associations) between commonly co-occurring conditions and symptoms, symposium attendees advocated for research conducted with a whole-person perspective.

### Theme 3: How Do We Conduct the Research?

Not only do we need to consider who is in the research and what research questions are important (see Table 2) but symposium attendees were also clear that we need to consider the method or the process by which research is conducted. Here, three themes emerged: engagement of consumers, the need to increase the scale of CAS research, and the need for a standardized minimum data set.

*Community engagement*[1]. For a long time, people with disabilities have demanded "nothing about us without us" (Charlton, 1998). This idea has been extended into consumer research engagement—people with lived experience of a condition such as CAS should be proactive research partners rather than solely research participants or recipients—and has gained traction internationally with a number of major funding bodies (Frank et al., 2020). To our knowledge, no CAS studies have used such a ground up approach, involving people with CAS and their families in the design, conduct, evaluation, implementation, and dissemination of the research. Exemplars of this type of approach can be found in the aphasia and brain injury literature (e.g., Power & Morrow, 2024).

The "community engagement" theme provides a call to action for both researchers and funding agencies. Researchers are called to move toward fully engaging consumers in research conduct, starting from the initial design and grant application phases. Funding agencies should adapt accordingly to incorporate true consumer engagement in their funding conditions. Researchers are encouraged to review the Involvement Matrix, which provides an explanation of this approach (Smits et al., 2020) and suggests research roles (from least to most complex) of listener, co-thinker, advisor, partner, and decision maker. In this regard, symposium attendees included not only researchers but also CAS specializing clinicians and Apraxia Kids board members, some of whom have children with apraxia (see Supplemental Material S2). This is a start on community-driven research goals.

Asking people with CAS, including children, young people, and adults with continuing or resolved CAS, for their research priorities, how the research should be conducted, and how the results should be interpreted, is a crucial step forward and will require a significant mind shift for many researchers. In recommending community-led research, we propose that Apraxia Kids, as the peak

[1]"Community" in this case would include not only people with lived experience of CAS but also clinicians who work with these people. These are two separate groups with distinct perspectives and research needs so for the purposes of this paper, we use community to mean both, and consumer to mean those with lived experience.

consumer organization in the United States and Canada, set up a registry for individuals, both with CAS lived experience and clinicians, who wish to partner in research. Such a registry would provide support for the uptake of this new model, particularly as funding bodies incorporate community engagement into funding criteria more broadly.

*Large data.* CAS is a relatively rare condition. In the most recent prevalence data available, CAS is present in approximately one to two in 1,000 children (Shriberg, Kwiatkowski, & Mabie, 2019; slightly less common than cerebral palsy; McIntyre et al., 2022), although it appears to be far more prevalent among children with complex neurodevelopmental disorders (Shriberg, Strand, et al., 2019). Because CAS is a relatively low incidence condition, much of the research to date has been conducted in small *N* studies, often fewer than 10 children, and the same people are used across studies (see Murray, 2022, and Maas, 2024, this issue, for reviews of diagnostic and treatment studies, respectively). While this type of research is valuable, to understand CAS as a disorder and to represent the wider community as described in Theme 1, larger scale studies are required. To this end, the symposium attendees recommended researchers create multisite, multinational studies, and establish and use large data sets, either independently or as part of existing data banks (e.g., TalkBank suite; PhonBank [https://www.talkbank.org/]). The advantages of a data bank are significant in providing opportunities to ask research questions without having to collect additional samples and for individual researchers to have access to many more people with CAS than they would through relying on local populations.

Large samples are imperative to answer the types of research questions that are presently of high priority (e.g., comparative treatment efficacy, treatment candidacy; as described by Maas [2024, this issue] and in the Treatment subtheme of Theme 1 above). The current evidence base includes sufficient evidence of treatment efficacy in controlled environments to substantiate investigations of real-world effectiveness (i.e., Phase 4 and 5 studies; Fey & Finestack, 2008) of approaches including ReST and ultrasound biofeedback (e.g., McCabe et al., 2023), and the Nuffield Dyspraxia Programme (e.g., McKechnie et al., 2020). Other study designs necessitating large data sets include (a) analyses of cost benefit (i.e., of one CAS treatment against another, or treatment against no treatment), (b) effectiveness of "usual care," and (c) comparison of one treatment to another with more diverse participants. Without investigations employing these design types, the CAS evidence base will be limited, in both the extent to which outcomes can be generalized, and whether precision prescription of CAS intervention can be achieved.

*Standardized minimum data.* Symposium participants recommended the development of a standardized minimum data set—that is, a universal set of measures collected and reported by each CAS study, regardless of their immediate utility to the study. There are a number of significant advantages in using a standardized minimum data set, including improved transparency and agreement on diagnostic profiles and the potential for future cross-study analyses (see Large Data section, above). The composition of such a set of measures would need to be widely agreed on (an e-Delphi study conducted by members of the Academy of Neurologic Communication Disorders and Sciences is in early stages; Maas et al., 2022), but is likely to include, for example, a given number of single words, connected speech, oral musculature, and hearing evaluation; diadochokinetic tasks including alternating motion rates (fast repeated production of a single syllable, e.g., "papapa") and sequential motion rates (fast repeated production of a syllable sequence, e.g., "pataka"), and repeated productions for calculation of consistency. The field may choose to include assessment of polysyllabic words and a short standard case history including language backgrounds, culture and ethnicity, and socio economic status to standardize the demographic information collected. To maximize generalizability cross-linguistically, cross-culturally, and across a wide range of ages, it is unlikely that specific standardized tasks would be included in the data set. A standard set of recording parameters, including, for example, sampling rate (e.g., 44 kHz) and notation of background noise (measured sound pressure level [SPL]) would facilitate cross-site acoustic analysis. Table 4 provides concrete suggestions as a starting point for consideration by the field.

Tests commonly used in clinical practice, such as the Dynamic Evaluation of Motor Speech Skill (Strand & McCauley, 2019) or the Verbal Motor Production Assessment for Children–Revised (Hayden & Namasivayam, 2021), are not included in their entirety in this suggested list. This is for two reasons. Firstly, by their nature, these tools are validated in specific languages or dialects and therefore may not be appropriate for use cross-linguistically. Secondly, the validation samples are limited in age range, severity, and the presence of comorbidity, all of which make them highly specific and increase the sensitivity but decrease the broader population applicability.

Meta-analysis is facilitated by increasing both study size and data standardization. To date, only ReST has been the subject of any form of meta-analysis (Ng et al., 2022). Collecting the same type of data locally and internationally will facilitate comparison across diverse populations, improve transparency in outcomes, and allow for effective re-analysis of studies as our understanding of CAS evolves.

**Table 4.** Suggestions for a standard minimum data set.

| Domain | Description | Comments |
|---|---|---|
| Single words | Elicited in (a) productive naming task and (b) imitation and/or dynamic assessment task with cueing | Include attempts at all phonemes in the language/dialect across multiple word positions. Include polysyllabic words with marked prosodic forms (e.g., wS stress patterns in English, lexical tones and tone sandhi in Cantonese) when possible given severity |
| Connected speech | Sample including a minimum of 75 different words (Wren et al., 2021) | Length of sample may be restricted given speech severity |
| Repeated productions of single words and/or nonsense words | Measure(s) allowing for token-to-token (whole word), phonemic, and phonetic consistency with sufficient complexity to tax the speaker's capacity | Although a number of methods have been reported for repeated productions (e.g., Dodd et al., 2002; Iuzzini-Seigel et al., 2017; Strand & McCauley, 2019), none is established as the gold standard and each measures a different construct |
| Oral motor evaluation | Speech and nonspeech tasks designed to evaluate speech related to cranial nerve function, respiration, phonation, and resonance; presence of possible oral apraxia | Thorough assessment is needed, not just a screening |
| Diadochokinesis (DDK) | Both (a) alternating motion rates (AMR; fast repeated production of a single syllable, e.g., "papapa") and (b) sequential motion rates (SMR; fast repeated production of a syllable sequence, e.g., "pataka") | Observation of breakdown in DDK has been shown to contribute to CAS diagnosis in school aged children (e.g., Murray et al., 2015) |
| Patient-reported outcomes | Measures addressing activity, participation, and environment | No presently available measures address all domains (intelligibility [e.g., Hodge et al., 2009; McLeod et al., 2012], participation [e.g., Thomas-Stonell et al., 2010], personal impact [e.g., McLeod, 2004]) |

*Note.* wS = weak strong stress pattern; CAS = childhood apraxia of speech.

## Theme 4: How Do We Move From Research to Practice?

The final major theme looks at moving from research to practice (see Table 2). It is often reported that change in practice on the ground takes more than 17 years from discovery to usual care (cf. Morris et al., 2011). Although this has not been formally investigated in speech-language pathology, recent survey research specifically in CAS (e.g., Gomez et al., 2019, 2022; Randazzo, 2019; Wong, Wong, & Velleman, 2023), and more broadly across the profession, suggests that it takes a number of years for clinical research outcomes to become practice. The symposium attendees were clear that this needs to change. In this issue, Lauretta et al. (2023) explored the qualitative evidence of faciliatory and inhibitory factors that influence SLP decisions to refer clients for genetic consultation. This is one step to reducing the research–practice gap in genetic testing of children with CAS.

*Improved access to research.* Potential barriers to timely implementation of research in clinical practice could be related to delivery format or accessibility of research findings or procedures. Symposium attendees recommended a number of practical steps to address these barriers, including more video, infographic, web-based, and social media research summaries specifically designed for consumers, and increased access to papers through researcher self-archiving. In the field at large, efforts to improve both the culture of research self-archiving and clinician knowledge of how to access research are underway, with a number of early career CAS researchers leading the way (e.g., http://www.csdisseminate.com; http://www.opencsd.com). Implementation of the previously described consumer-led research may also address this theme.

*Clinical guidelines.* The second subtheme was the development of clinical pathways and guidelines, which includes easily accessed and readily updated minimum care standards based on the current evidence. While several such sources exist (e.g., Apraxia Kids parent portal: https://parent.apraxia-kids.org/; ASHA Evidence Maps: https://apps.asha.org/EvidenceMaps/; McCabe et al., 2022), these may lack the rigor of formalized clinical guidelines. Symposium participants suggested that a single source document describing current best practices for diagnosis and treatment, a "living handbook," would be of high clinical value. As part of this recommendation, and the overlapping need for a standardized research data collection protocol, symposium participants recommended development of a standard set of clinical assessment measures. Although understanding of the critical tasks required for diagnosis continues to grow (see McCabe et al., 2024, for a commentary on current diagnostic challenges), enough empirical evidence has amassed for this to be possible (e.g., Chenausky et al., 2020; Murray et al., 2015).

**Table 5.** Childhood apraxia of speech–related implementation science examples of free resources and training.

| Resource | Link | Comments |
|---|---|---|
| Babble Bootcamp (e.g., Potter et al., 2023, this issue) | https://f1000research.com/articles/8-271/v5 and https://osf.io/yzht4/files/osfstorage/5efb520a9fceff01b882cbc1/?pid=yzht4 | Outline of the program and instructions for each activity |
| Dynamic Temporal and Tactile Cueing (e.g., Strand, 2020) | https://childapraxiatreatment.org/diagnosis-and-treatment-of-cas-online-course/ | Free initial online training course with video |
| Profile of Childhood Apraxia of Speech and Dysarthria (Iuzzini-Seigel et al., 2022) | https://doi.org/10.1044/2022_LSHSS-21-00164 | Open Access paper, checklist, and training materials to aid differential diagnosis of motor speech impairments |
| Rapid Syllable Transition Treatment (e.g., McCabe et al., 2023) | https://rest.sydney.edu.au/ | Free training materials, video, manual, treatment resources |
| Speech Motor Chaining (e.g., Preston et al., 2019) | https://chaining.syr.edu/SpeechMotorChaining/ | Free online resources, videos |

*Implementation science and motor speech.* Implementation science encompasses both a philosophy and a method that systematically studies how research becomes practice and, in doing so, it takes a very different set of assumptions to a traditional scientific-medical model research trajectory (Douglas et al., 2022). Importantly, implementation science addresses a broad range of issues, including research on methods that enhance uptake, research on designs that are conducive to community engagement and inclusion, and trialing strategies that facilitate implementation of new methods and that superseded ones are de-implemented. It is a fallacy to think that good science will trickle down to clinical practice—real effort needs to be made to design studies alongside clinicians and families that will ensure both implementation of better practices and, importantly, de-implementation of existing, less effective ones. To date, no motor-speech implementation studies have been conducted (Douglas et al., 2022). While formal implementation science investigations are exceedingly rare, CAS treatment researchers have been at the forefront in using some of the tools of implementation science (e.g., free and easy to access resources or online training; see Table 5). Symposium attendees offered additional strategies to improve research translation (see Improved Access to Research subtheme). The next step in CAS research is to make the required paradigm shift from the Technical Report's (ASHA, 2007b) emphasis on basic science to adoption of implementation science approaches, particularly in advancing assessment and treatment research.

## Conclusions

The ASHA (2007b) Technical Report's list of CAS research needs is now dated. This prologue and the associated special issue provide guidance for the most pressing short-term needs in CAS research. In particular, as a research and clinical community, we need to focus on the diversity of people with CAS and better, more efficient implementation of high-level evidence-based practice across the lifespan.

## Author Contributions

**Patricia McCabe:** Co-lead author. Lead contribution in conceptualization, writing of manuscript, and editing of manuscript. Co-guest editor of the special issue. Chair of the symposium (including a leadership role in designing the program and inviting keynote presentations) and peer review of presentations for the symposium. Participant in symposium discussions about research priorities leading to the paper.

**Molly Beiting:** Co-lead author. Equal lead contribution in conceptualization, writing of manuscript, and editing of manuscript. Co-guest editor of the special issue. Participant in symposium discussions about research priorities leading to the paper.

**Maria I. Grigos:** Substantial contributions to initial conceptualization of the work and critical revision of the drafted manuscript. Co-guest editor of special issue. Member of the symposium organizing committee (including a role in designing the program and inviting keynote presentations) and peer review of presentation submissions. Participant in symposium discussions about research priorities leading to the paper.

**Elaine R. Hitchcock, Edwin Maas, Amy Meredith, Angela T. Morgan, Nancy L. Potter, Jonathan L. Preston:** Substantial contributions to initial conceptualization of the work and critical revision of the drafted manuscript. Member of the symposium organizing committee (including a role in designing the program and inviting keynote presentations) and peer review of presentation submissions. Participant in symposium discussions about research priorities leading to the paper.

**Laura Moorer:** Substantial contributions to initial conceptualization of the work and critical revision of the drafted manuscript. Organizer of the symposium (including

a role in designing the program and inviting keynote presentations), and participant in symposium discussions about research priorities leading to the paper.

All other authors (listed in alphabetical order): Substantial contributions to initial conceptualization of the work through discussion contributions at the symposium and critical revision of the drafted manuscript.

All authors provided approval of the final version of the manuscript and agreed to be accountable for all aspects of the work.

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## Data Availability Statement

There are no data associated with this paper.

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