From the
DEPARTMENT OF CLINICAL SCIENCE, INTERVENTION AND TECHNOLOGY, CLINTEC
Division of Speech and Language Pathology
Karolinska Institutet, Stockholm, Sweden

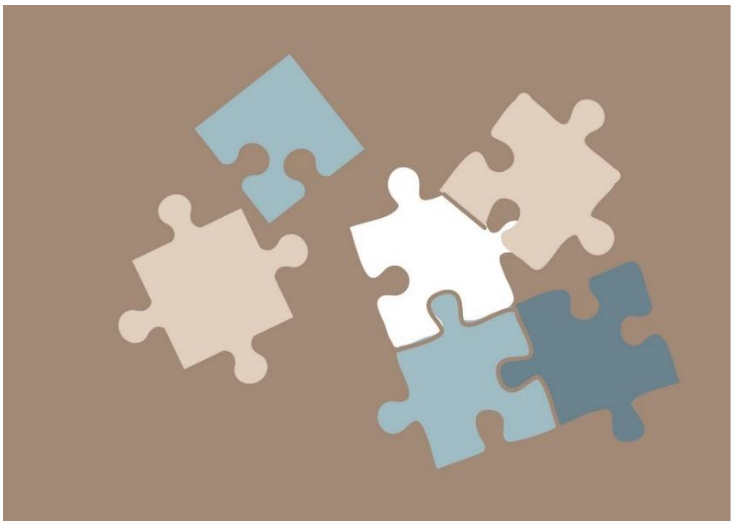
ASSESSMENT AND DIFFERENTIAL DIAGNOSIS IN CHILDHOOD APRAXIA OF SPEECH

Susanne Rex

Stockholm 2022

All previously published papers are published as open access.
Published by Karolinska Institutet.
Printed by Universitetsservice US-AB, 2022

ISBN 978-91-8016-634-8



Cover illustration by **Moa Edsö**

" It's not black or white. It is like a puzzle; you need several pieces to see the full picture"

Edythe Strand, 2009, speaking about the assessment of children with speech disorders

Assessment and differential diagnosis in Childhood Apraxia of Speech

THESIS FOR DOCTORAL DEGREE (Ph.D.)

By

Susanne Rex

which, by due permission from Karolinska Institutet, will be publicly defended in Birkeaulan 1, Forskningsgatan 51, Karolinska Universitetssjukhuset, Huddinge/Blickagången 9B, Stockholm, Sweden

June 17th, 2022, at 9.00 am

Principal Supervisor:
Associate professor Anita McAllister
Karolinska Institutet
Department of Clinical Science, Intervention, and Technology, CLINTEC
Division of Speech and Language Pathology

Co-supervisors:
Associate professor Kristina Hansson
Lund University
Department of Clinical Sciences
Division of Logopedics, Phoniatrics and Audiology

Emeritus professor Edythe Strand
Mayo College of Medicine,
Rochester, MN, USA

Opponent:
Dr Sara Wood
Queen Margret University, Edinburgh
School of Health Sciences
Division of Speech and Hearing Sciences

Examination Board:
Professor Christina Persson
University of Gothenburg
Institute of Neuroscience and Physiology
Department of Health and Rehabilitation

Associate professor Steven Lucas
Uppsala University
Department of Women's and Children's Health
Division of Paediatric Inflammation, Metabolism and Child Health Research

Associate professor Iris-Corinna Schwarz
Stockholm University
Department of Special Education,
Stockholm Babylab

ABSTRACT

Introduction and aims: Childhood Apraxia of Speech (CAS) is a disorder of speech motor planning and programming affecting the ability to transform the linguistic code into movements needed for intelligible speech. CAS is difficult to differential diagnose from other speech sound disorders (SSDs) and the diagnostic procedure involves many decisions based on outcomes from assessments before arriving at the appropriate diagnosis/es. Some children with CAS also have coexisting functional difficulties. Consequently, it is important for the clinician to describe and be aware of these difficulties. The overall aim of this project was to investigate speech performance and speech characteristics in 3-to-9-year-old children with typical and disordered speech, to facilitate more confident diagnostic decisions of Swedish-speaking children with CAS.

Material and methods: The project consists of four studies with a total of 139 participants in the age range 3:1 to 8:10 years (37 to 106 months). In **Study I**, a newly developed Swedish test for motor speech skills, Dynamisk motorisk talbedömning (DYMTA), adapted from Dynamic Evaluation of Motor Speech Skills (DEMSS) was described. 94 children (37 to 106 months old) with typical speech- and language development participated (45 boys/49 girls), including 17 bilingual children, and were assessed with DYMTA (DYMTA-A and DYMTA-B) as well as with a comprehensive test battery of language, speech, and orofacial function. In **Study II**, reliability and validity of DYMTA were analyzed to evaluate its ability to discriminate between children with a diagnosis of CAS from children with other SSDs. 45 children (31 boys/14 girls, 40 to 106 months) were assessed with DYMTA as well as with a comprehensive test battery covering language, speech, and orofacial function. In **Study III**, the number and types of characteristics associated with CAS were investigated in two different speech samples for each child, one using dynamic assessment (DA) and the other static assessment (SA). 33 children (22 boys/11 girls, 40 to 106 months) with a CAS diagnosis were included and a perceptual assessment of present and absent speech characteristics was performed. In **Study IV**, a parental questionnaire, specifically designed for this project asking about mental, sensory, motor, and voice functions, was given to caregivers of 33 children with CAS (the same group as in study III). An agglomerative clustering analysis was applied to the individual item-responses to identify groups of children with different patterns of parent-reported functional difficulties.

Results: The main result in **study I** was that Swedish children with typical speech- and language development performed well on tasks in DYMTA already at the age of three. Outcome measures on some subscores and subtests were close to ceiling from five years, while others showed a protracted refinement into early school-age. In **Study II**, intrarater reliability was found to be strong for the total scores and subscores in both DYMTA-A and DYMTA-B. Interrater reliability (point-by-point and ICC) was strong for total score and all subscores except for the ICC value of the subscore Prosody. The validity estimates showed that the results on DYMTA have good potential to differentiate children with CAS from children with other SSDs. In **Study III**, the most prominent CAS characteristics apart from inconsistency in the dynamic task were deviant transitionary movements, prosody error, and vowel error, and in the naming task vowel errors, voicing errors and prosody errors. More CAS characteristics were evident in the DA sample (on average 6.8), than in SA sample (5.2). In **Study IV** the number of parent-reported functional difficulties ranged from 1 to 27 across the participants. The hierarchical cluster analysis identified four groups of children with different functional profiles: 1A) few coexisting difficulties, 1B) coexisting difficulties mainly in voice domains, 2A) coexisting difficulties in mental functions (e.g., attention) and 2B) coexisting difficulties in diverse functions, including motor functions.

Conclusions: The findings from this thesis project support benefits of including several tasks and methods addressing speech motor skills in the diagnostic procedure of CAS. The preliminary evidence on validity and reliability of DYMTA showed acceptable results in separating children with CAS and other SSDs. Also, the investigation of detected speech characteristics associated with CAS supports current knowledge on the benefits of a dynamic assessment method, adding information on the child’s speech motor skills when cuing is provided. Apart from the results from assessments on speech performance itself, clinicians should also be aware of various functional profiles and coexisting difficulties evident in some children with CAS.

SAMMANFATTNING

Introduktion och syfte: Taldyspraxi hos barn är en motorisk talstörning med avvikelse i förmågan att planera och programmera de rörelser som produceras när vi talar, trots att talmuskulaturen fungerar som den ska. Taldyspraxi är ofta svår att särskilja, dvs. differentialdiagnostisera, från andra talstörningar och många testuppgifter behövs för att få fram de symptom som leder oss till diagnosen. En del barn med taldyspraxi har också andra samförekommande svårigheter vilka kan vara viktiga att beskriva och vara medveten om i det kliniska arbetet med barnen. Syftet med detta projekt var att bidra med mer kunskap om bedömning av talsymptom hos 3 till 9-åriga barn med talstörning och misstänkt taldyspraxi genom att undersökta prestationen hos typiskt utvecklade barn samt barn med talstörning på ett svenskt test för talmotorisk bedömning, undersöka talsymptom hos barnen med taldyspraxi och beskriva samförekommande funktionella svårigheter som rapporterats av föräldrar.

Material och metoder: Projektet innehåller fyra studier med totalt 139 barn i åldrarna 3 till 9 år (37 till 106 månader) som deltagare. I **studie I**, beskrevs ett nyligen utvecklat test för talmotoriska förmågor, Dynamisk motorisk talbedömning (DYMTA). 94 barn (37 till 106 månader gamla) med typisk tal- och språkutveckling deltog (45 pojkar/49 flickor), varav sjutton flerspråkiga barn. De bedömdes med DYMTA samt med ett testbatteri för språk, tal och oralmotorisk förmåga. I **studie II**, analyserades reliabilitet och validitet för DYMTA för att undersöka testets förmåga att särskilja barn med diagnosen taldyspraxi från barn med annan talstörning. 45 barn (31 pojkar/14 flickor, 40 till 106 månader) deltog och bedömdes med DYMTA samt med ett testbatteri för språk, tal och oralmotorisk förmåga. I **studie III**, undersöktes antal och typ av talsymptom som karaktäriserar talet vid taldyspraxi i två olika talmaterial för vart och ett av barnen, där det ena använde en s.k. dynamisk metod och den andra en statisk metod. Vid dynamisk bedömning ger man stöd och ledtrådar för att uppmuntra till maximal prestation. 33 barn (22 pojkar/11 flickor, 40 till 106 månader) med taldyspraxi deltog och en perceptuell analys av aktuella talsymptom från audio/ videoinspelningar utfördes. I **studie IV**, analyserades svaren från ett föräldraformulär, framtaget för detta projekt, gällande 40 samförekommande svårigheter inom funktioner såsom sensorik, uppmärksamhet och motorik hos de 33 barnen med taldyspraxi (samma barn som i studie III). En klusteranalys gjordes för att undersöka om det fanns grupper med barn med liknande fördelning av samförekommande funktionella svårigheter.

Resultat: Huvudresultatet i **studie I** var att svenska barn med typisk tal- och språkutveckling presterar väl på uppgifterna i DYMTA redan vid tre års ålder. Utfallsmått på vissa variabler och deltest nådde nära takeffekt från fem års ålder, medan andra variabler visade en successiv utveckling upp i tidig skolålder. Resultaten från beräkningar av reliabilitet för DYMTA i **studie II** visade på en övervägande god samstämmighet mellan bedömarna. Utifrån de olika undersökta måtten på validitet framkom att testet har en acceptabel förmåga att särskilja på barn med taldyspraxi och annan talstörning. I **studie III**, framkom att de vanligast förekommande talsymptomen i talmaterialet där dynamisk metod användes, förutom inkonsekvens, var avvikande talrörelsesekvenser, avvikande prosodi och vokalfel. I talmaterialet där statisk metod användes var vokalfel, svårigheter med distinktionen tonande-tonlös och avvikande prosodi vanligast. Fler talsymptom som pekar mot talplaneringssvårigheter visade sig vid bedömning med dynamisk metod (medel 6.8) än med statisk metod (medel 5.2). De föräldrarapporterade funktionella svårigheterna i **studie IV** varierade i antal från 1 till 27 hos de deltagande barnen. Fyra grupper med liknande funktionella profiler identifierades: de med få svårigheter, de med svårigheter huvudsakligen med röst och rytm, de med svårigheter i mentala funktioner (till exempel uppmärksamhet) och de med svårigheter i flera olika funktioner inklusive motoriska svårigheter.

Slutsatser: Resultaten från studierna stödjer fördelar med att introducera riktade testuppgifter för talmotorisk förmåga vid bedömning av taldyspraxi. Preliminära resultat av valideringen av testet DYMTA visar att de variabler som bedöms, dvs talrörelseprecision, vokalproduktion, prosodi och konsekvens, kan vara till hjälp i den diagnostiska proceduren att särskilja mellan barn med diagnosen taldyspraxi och barn med annan talstörning. Därutöver förefaller dynamisk testmetod, där support är tillåten under testning, vara till god hjälp att identifiera talsymptom som är associerade med taldyspraxi. Förutom att dessa talsymptom kan variera i antal och frekvens hos det enskilda barnet, kan olika profiler med samförekommande funktionella svårigheter förkomma och vara viktiga att ta hänsyn till vid omhändertagande av barn med taldyspraxi.

LIST OF SCIENTIFIC PAPERS

I. Rex, S., Hansson, K., Strand, E., & McAllister, A.
Performance of Swedish children on a dynamic motor speech assessment, 2021. *International Journal of Speech-Language Pathology*, 23:5, 453-464

II. Rex, S., Sand, A., Strand, E., Hansson, K., & McAllister, A.
A preliminary validation of a dynamic speech motor assessment for Swedish-speaking children with childhood apraxia of speech, 2021. *Logopedics Phoniatrics Vocology.*

III. Rex, S., Strand, E., Sand, A., & McAllister, A.
Observations of speech characteristics associated with Childhood Apraxia of Speech in static versus dynamic assessment. Manuscript, submitted, under review.

IV. Rex, S., Sand, A., Strand, E., Hansson, K., & McAllister, A.
Childhood Apraxia of Speech and coexisting functional characteristics. Manuscript.

CONTENTS

LIST OF ABBREVIATIONS

ADHD	Attention Deficit Hyperactivity Disorder
AOS	Apraxia of Speech
ASHA	American Speech-Language-Hearing Association
ASD	Autism Spectrum Disorder
AUC	Area Under the receiver operating characteristics Curve
CAS	Childhood Apraxia of Speech
CD	Childhood Dysarthria
DCD	Developmental Coordination Disorder
DEMSS	Dynamic Evaluation of Motor Speech Skills
DLD	Developmental Language Disorder
DTTC	Dynamic, Temporale and Tactile Cuing
DYMTA	Dynamisk Motorisk Talbedömning
EPG	Electropalatography
ESSENCE	Early Symptomatic Syndromes Eliciting Neurodevelopmental Clinical Examinations
FTF	Five To Fifteen questionnaire
GRAMBA	Grammatiktest för Barn
ICC	Intraclass Correlation Coefficient
ICF	International Classification of Functioning, Disability and Health
LINUS	Linköpingsundersökningen
nonCAS-SSD	SSD, but not CAS
NOT-S	Nordic Orofacial Test - Screening
OMS	Oral Motor Score
PCC	Percent Consonants Correct
PML	Principles of Motor Learning
PROMPT	Prompts for Restructuring Oral Muscular Phonetic Targets
PVC	Percent Vowels Correct
PWC	Percent Words Correct
ReST	Rapid Syllable Transitions Treatment

ROC	Receiver Operating Characteristic curve
SD	Speech Delay
SDCS	Speech Disorders Classification System
SIT	Språkligt Impressivt Test
SLP	Speech-Language Pathologist
SMD	Speech Motor Delay
SSD	Speech Sound Disorder
SVANTE	Swedish Articulation and Nasality Test
TD	Typical Development
TROG-2	Test for Reception of Grammar-2
TSD	Typical Speech Development
UTI	Ultrasound Tongue Imaging
VMPAC	Verbal Motor Production Assessment for Children

1 INTRODUCTION

1.1 GENERAL MOTIVATION

During my early years working as a Speech-Language Pathologist (SLP) at a general SLP clinic with children having speech disorders, the assessment and therapy focused on phonology (i.e., on the language system). However, in the clinical work with the children, it was noticeable that some of them also had motor difficulties. As I found the group of children with speech sound disorder (SSD) to be heterogenous, this also affected my thinking about what tasks to be used for assessment, to support the choice of a tailored intervention. One subgroup was especially of interest, the children who had difficulties in speech movement coordination and who had inconsistent speech productions, i.e., the children with Childhood Apraxia of Speech (CAS). Consequently, the need for an assessment tool in Swedish for these children, which was not available at the time, was apparent. At the same time, some of the children with speech motor difficulties seen at the clinic also showed difficulties in language, general motor abilities and oral motor skills. In this thesis project the intention was to put forward results that might contribute to a more confident diagnosis for children with speech motor difficulties.

CAS is a disorder suggested to affect 0.1 - 0.2 % of all children (Shriberg, Aram, & Kwiatkotski, 1997) or 2.4% in a group of children with idiopathic SSD (Shriberg, Kwiatkowski, & Mabie, 2019). However, this is a patient group difficult to diagnose and many clinicians and researchers have stressed the need for a careful differential diagnosis (Murray, Iuzzini-Seigel, Maas, Terband, & Ballard, 2021), and a more unified assessment procedure and clearer diagnostic guidelines (Iuzzini-Seigel & Murray, 2017). There is an ongoing international discussion on how to define SSD, how different speech disorders fit into the label and how to separate them (IALP, 2021). The assessment procedure for CAS is definitely complex. Arriving at a diagnosis takes time and involves many decisions along the way and some of them will be addressed in this thesis.

2 BACKGROUND

2.1 CHILDHOOD APRAXIA OF SPEECH

2.1.1 Definition

CAS is a motor speech disorder commonly organized under the umbrella term Speech Sound Disorders (SSD) (Bishop, Snowling, Thompson, & Greenhalgh, 2016; Caruso & Strand, 1999; Strand, 2017). The SSD term is currently used for a disparate group of children with speech disorders affecting phonology, articulation, or motor speech production including CAS. The SSD might be of known origin (e.g., cleft palate, sensori-neural hearing loss, other neurodevelopmental or genetic anomalies) or of unknown origin (idiopathic).

A great deal of research has focused on the definition of CAS, with the very first descriptions of apraxia of speech in children emerging in the 1950s by Morley, Court, Miller, and Garside (1955), followed by Rosenbek and Wertz (1972) and Yoss and Darley (1974). More than 50 definitions of CAS have been used in research and clinical literature over the years, but there is still ongoing work regarding the best classification criteria for identifying children with CAS. In the Technical report by the American Speech-Language-Hearing Association ASHA (2007a) a definition based on international consensus was presented:

> *"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 and 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" (ASHA, 2007, p. 3).*

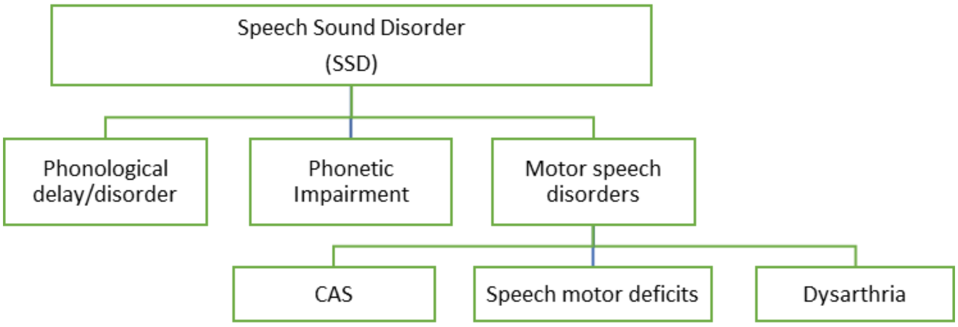
This definition has been globally accepted by most clinicians and researchers. The term CAS is used as a cover term for all presentations of apraxia of speech in childhood, whether developmental or acquired or whether associated with a specific etiology (ASHA, 2007b). The terms developmental verbal dyspraxia or developmental apraxia of speech typically refers to an idiopathic disorder and are sometimes still used in that sense. The term apraxia was originally used for disordered speech evident after a brain injury, separating it from the often cooccurring language impairment (aphasia) and neuromuscular deficit (dysarthria) (Darley, Aronson, & Brown, 1975). The acquired adult form of apraxia is referred to as apraxia of speech (AOS) (Duffy, 2013) defined as "a neurologic speech disorder that reflects an impaired capacity to plan or program sensorimotor commands necessary for directing movements that result in phonetically and prosodically normal speech" (Duffy, 2013, p. 4). Reports on notable discrepancies between voluntary and involuntary performance of simple speech and nonspeech tasks shown by some patients had been made already by Hughlings Jackson in 1866 (Head, 1915). For example, he described a patient not to be able to put his tongue out when asked to, but not having any problems with licking a crumb from his lips. Hence, the term apraxia is also used to describe inability to plan, organize and coordinate volitional movements in other areas, e.g., oral dyspraxia or limb apraxia.

The incidence for clinical occurrence of CAS in a Swedish study was found to be 5% (Malmenholt, Lohmander, & McAllister, 2017), and in English-speaking children it has been estimated to be 2.4% in children with idiopathic speech disorder (Shriberg, Kwiatkowski, et al., 2019) and 4.3% when associated with complex neurodevelopmental disorder (Shriberg, Strand, Jakielski, & Mabie, 2019). A sex ratio of 2:1 male:female ratio has been reported for idiopathic CAS (ASHA, 2007) and 1:2.51 across children with CAS from 34 studies reviewed by Murray et al. (2021)**.**

2.1.2 Classification

For the individual clinician the ongoing discussion on how to classify children with SSDs into subgroups is important, especially because separate theoretical systems are grouping the children with speech disorders differently and this may impact decisions on diagnosis and subsequent intervention planning (Waring & Knight, 2013). One classification system commonly referred to within the community studying and working with SSDs is the Speech Disorders Classification System (SDCS) (Shriberg et al., 2010; Shriberg & Strand, 2018). The SDCD is a clinical-research framework consisting of four levels including causal factors as well as clinical signs, all linked together. The level I "etiological processes", (distal causes such as genomic and neurodevelopmental factors) are linked to level II "speech processes" (the proximal causes which are the representation, transcoding, and execution of the speech production process), and then further linked to level III "the clinical typology" (the behavioral phenotype or subgroups of SSD) with diagnostic markers for these on level IV. The work on SDCD was recently finalized and the behavioral phenotypes included are Speech Delay (SD), three types of motor speech disorders (CAS, Childhood Dysarthria (CD), and Speech Motor Delay (SMD)), and Speech Errors for /r/ and /s/ (Figure 1). SMD has been introduced as a motor speech disorder to complement CD and CAS when the criteria for either are not met (Shriberg, Campbell, Mabie, & McGlothlin, 2019). The SMD is suggested to be used for children evidencing a motor component to their idiopathic SD, with symptoms such as imprecise and unstable speech, prosody, and voice. Studies on the phenotype and persistence (Shriberg, Campbell, et al., 2019) as well as findings from a recent treatment study (Namasivayam, Huynh, Granata, Law, & van Lieshout, 2021) support the construct validity of SMD. A gap in the diagnostic labels for children with a speech motor component, not meeting the criteria for CAS but with obvious motor involvement has also been acknowledged in the clinical setting (Mogren, 2021).

Figure 1. Schematic figure of SSD classification (author, based on Shriberg et al., 2010)



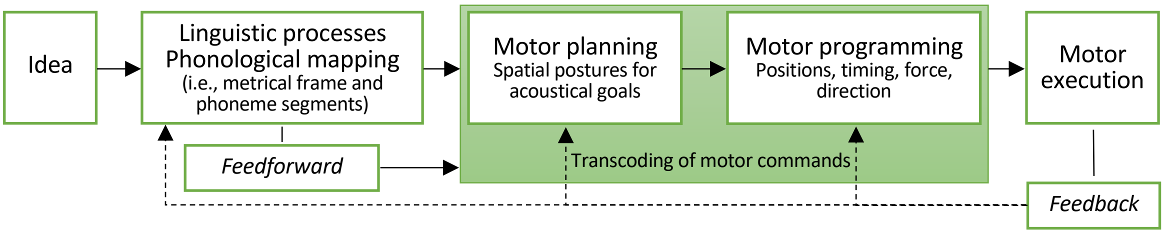
Additionally, The Model for Differential Diagnosis (Dodd, 2014) is a classification model with a descriptive-linguistic approach assuming that subgroup-specific processing deficits can be identified by surface speech characteristics. It has been argued to be easy to apply to the clinical population (Dodd, 2014) and has been validated (Ttofari Eecen, Eadie, Morgan, & Reilly, 2019). The diagnostic groups in this classification are Articulation disorder (phonetic disorder; substitutions or distortions of the same sounds in isolation and in all phonetic contexts), Phonological delay (phonemic; speech error patterns that are typical of younger children), Consistent atypical phonological disorder (phonemic; consistent speech error patterns that are unusual non-developmental errors), Inconsistent phonological disorder (phonemic; multiple error forms for the same lexical item) and CAS (motor speech disorder; difficulty sequencing articulatory movements, inconsistent). A disadvantage of this classification system might be that it only incorporates one subgroup for motor speech disorders (i.e., CAS).

A parallel work has also been made to identify and classify language impairments in children, the CATALISE project (Bishop et al., 2016). In the CATALISE project a panel of professionals was set to reach consensus on criteria for identification of language impairment, and although a high level of agreement was reached, residual disagreements were reported on, underlining the complexity of definitions in the field of speech- and language disorders. One of the final set of statements, regarding speech production: *"Assessment by a speech-language therapist will determine if a problem with speech production is linguistic, structural or motor in origin"* (p. 8, Bishop, 2016), highlights the interconnectedness between speech and language.

2.1.3 Speech production process

Research utilizing the different classification systems all provide insight into the nature of SSD. There is an agreement among research groups regarding the neuromotor processes underlying speech (Caruso & Strand, 1999; Nijland, Terband, & Maassen, 2015), although the sensorimotor speech processes have been modeled in many ways. All models implement a hierarchy of control and involves a preparatory process with the idea the person wants to communicate (Terband, Maassen, & Maas, 2016). For a schematic model see Figure 2. This idea will then be linguistically processed, retrieving words and mapping phonology for those words such as the metrical frame and the phoneme segments. Next, the transformation of the phonological map to speech movements involves motor planning. The speaker plans the spatial postures for the acoustical goal of the word. During the following stage the motor plans are applied in muscle-specific motor programs, specified in timing and the positions of structures, followed by the final speech output. In parallel there are online adjustments for correction of errors made. This self-monitoring takes place as feed-forward control (activated when a planned movement is detected to be incorrect before execution) and feedback control (used both for fast auditory and somatosensory feedback of articulation during speech) (Terband, Maassen, Guenther, & Brumberg, 2009). Although the models of the speech production processes schematically displaying the concepts separated from each other, there is an interaction between levels and these interdependencies cause the overlap of symptoms often seen in the different kinds of SSDs (Caruso & Strand, 1999; Terband, Maassen, & Maas, 2019). As stated in the definition, the overt symptoms detected in assessment of SSD should then be referred to the levels of planning and/or programming of speech movements for a diagnosis of CAS.

Figure 2. Schematic figure of the speech production process (author, based on Caruso & Strand, 1999).



2.1.4 Etiology

The neural network for the speech processing has mainly been based on studies of the adult brain (e.g., aphasia recovery and brain plasticity). Structural and functional neuroimaging data have revealed a language network divided into two anatomical streams with complementary functions (Hickok, 2012; Hickok & Poeppel, 2007). In the described dual route model, a bilaterally organized ventral stream, is engaged in processing speech signals for comprehension, i.e., mapping of sounds to meaning, while a dorsal stream is engaged in transcoding the acoustic signals into speech motor plans, i.e., integrating the sensory-motor information for verbal repetition. However, neurobiological research on children with language and speech disorders has increased, including children with CAS (Liegeois & Morgan, 2012). For example, structural and functional MRI data were analyzed in a study on a two-generation family with 13 individuals affected with CAS (Liegeois et al., 2019) showing bilateral white matter reductions in the arcuate fasciculus (i.e., an axon bundle connecting the temporal cortex to the inferior parietal cortex), indicating a disruption in the dorsal language stream (sound to motor speech transformations) as a novel phenotype of CAS. The same disruption was not found in the ventral language stream (sound/letter to meaning) or the motor tracts. It has also been suggested that an altered connectivity of the left corticobulbar tract could be a neural marker for developmental speech disorder (Morgan, Su, et al., 2018). MRI has also been used to observe improvements in speech after intervention (Fiori et al., 2021).

Historically, research on CAS has focused on idiopathic CAS. Recently the perspectives have broadened also embracing more complex phenotypes as well as various forms of comorbidity (Morgan, van Haaften, et al., 2018). Comorbidity does indeed seem to be common; in a review article on twelve cases, reported with neuroimaging data, CAS was found to be associated with epilepsy, metabolic disorders, syndromic disorders as well as idiopathic forms (Liegeois & Morgan, 2012). The authors of the review concluded that perisylvian and perirolandic cortices, the basal ganglia, and the cerebellum all play a major role in both speech planning and execution of speech in childhood.

The relationship between the genetic level and the phenotype has been found to be complex as the different genetic or biological deficits may be expressed as the same symptom, or the same biological deficit may be expressed as different phenotypes (Mei et al., 2018). Several genes have been associated to CAS, including FOXP2. This gene was the first found to be associated with severe motor speech disorder and identified in a three-generation family, the KE family (Lai, Fisher, Hurst, Vargha-Khadem, & Monaco, 2001; Marcus & Fisher, 2003). The affected KE family members showed coexisting oral-motor and verbal motor dyspraxia (CAS), language, and literacy impairments. Also, in an Australian study, eight families were recruited where the proband had CAS, and novel variations of FOXP2 were found in two probands (Turner et al., 2013). The findings supported earlier studies in that FOXP2 is associated with speech-, motor- and language deficits. Laffin et al. (2012), using a whole-genome-analysis in 24 children with a clinical CAS diagnosis, found copy number variants in half of the genes, with plausible neural consequences for cognitive, speech, language, and motor processes. However, a FOXP2 mutation was found only in one child. As a consequence of the extended and deepened knowledge of the genetic influence on speech processing, it has been suggested that when a severe and persistent diagnosis of CAS is confirmed, a referral for a genetic analysis should be done for optimized diagnosis and intervention (Morgan & Webster, 2018).

2.2 SPEECH CHARACTERISTICS ASSOCIATED WITH CAS

While the CAS core impairment in planning and programming spatiotemporal parameters of speech movement results in numerous overt characteristics or features, it is causing clinical challenges in differential diagnosis. Moreover, no feature alone has been found to be enough for a diagnosis of CAS (Allison, Cordella, Iuzzini-Seigel, & Green, 2020; ASHA, 2007a; Murray et al., 2021; Murray, McCabe, Heard, & Ballard, 2015) and overlaps of symptoms in children with CAS and other SSDs are reported, i.e., the features are not pathognomonic (Iuzzini-Seigel & Murray, 2017). Also, symptoms may change in their relative frequencies of occurrence with task complexity, severity, and age (ASHA, 2007b; Strand, 2019).

An internationally consensus on the core features of CAS was achieved in 2007, appointing three main features (ASHA, 2007b) (a) inconsistent errors on consonants and vowels in repeated productions of syllables or words (b) lengthened and disrupted coarticulatory transitions between sounds and syllables (c) inappropriate prosody. However, it was noted that *"these features are not proposed to be necessary and sufficient signs of CAS"* (ASHA, 2007a, p.2). Further, they were not operationally defined in a way that would make it possible for the researchers and clinicians to interpret and use them in a unified way. Iuzzini-Seigel and Murray (2017) presented a comprehensive feature list of characteristics associated with CAS containing operationally defined criteria initially suggested by Iuzzini-Seigel, Hogan, Guarino, and Green (2015), based on Mayo Clinic criteria (Strand's checklist) (Shriberg, Lohmeier, Strand, & Jakielski, 2012; Shriberg, Potter, & Strand, 2011). This checklist included vowel errors, consonant distortion, stress errors, syllable segregation, increased difficulty with multisyllabic words, groping, intrusive schwa, voicing errors, slow rate, difficulty achieving initial or transitionary movement gestures, resonance disturbance, and inconsistency. These characteristics associated with CAS are found to be used in several research studies (Centanni et al., 2015; Malmenholt, 2020; Mogren, 2021) and are occasionally extended with findings on limited phoneme inventory, speech sound deletions and substitutions (Grigos, Moss, & Lu, 2015; Liegeois et al., 2019). Hence, these extended checklists include speech characteristics traditionally referred to as phonologically patterned errors (e.g., velar fronting) common in a phonological disorder. However, findings from Electropalatography (EPG) (Gibbon, 1999) and ultrasound tongue imaging (UTI) (Cleland, Scobbie, Heyde, Roxburgh, & Wrench, 2017; Roxburgh, Cleland, Scobbie, & Wood, 2021) have detected evidence that some children with SSDs have difficulty with the articulatory gestures needed to clearly differentiate collapsed phonemes, e.g., alveolar, or velar sounds. Subsequently, it is important to be aware of that a characteristic typically referred to as phonological may have different origins, i.e., it could be linguistic or motor.

2.3 ASSESSMENT PROCEDURE AND DISCRIMINATIVE FEATURES

A comprehensive test battery on language and speech is commonly used in the assessment procedure of children with SSD and suspected CAS to collect information for further analysis (Iuzzini-Seigel, 2021; Murray et al., 2015; Strand, McCauley, Weigand, Stoeckel, & Baas, 2013). Suggested components are history uptake, connected speech sample, phonetic and phonemic inventories, expressive and receptive language testing, phonologic performance, orofacial examination of structure and function, and a motor speech examination (Strand & McCauley, 2019). However, the intended purpose of an assessment may differ in many ways depending on the situation. An assessment could aim to detect or confirm a problem, differential diagnose, classify, specify severity, determine treatment focus, make decisions on number and length of sessions, or measure change following treatment (Caruso & Strand, 1999). Depending on the specific assessment of a particular child and what diagnoses that must be distinguished between, different standardized tests are chosen. Further, when choosing test or measurements one should consider if there is evidence for reliability and validity (Dollaghan, 2007). However, guidelines for test selection are seldom available and the choice may influence interpretation and inconsistency in the use of a test (Daub et al., 2021). Daub et al. (2021) stress that it is the clinical decisions that should be validated, not the test itself. This imply that SLPs need to have access to a variety of tools to serve their different intended decisions (Daub et al., 2021). The validity evidence generally sought for are a clearly described rationale, procedure, and participants as well as information on psychometric measures such as likelihood ratios (Dollaghan, 2007).

There have, so far, been two reviews focusing on the validity and reliability of the diagnostic procedures used among children with CAS. In the first review, none of the included six tests of motor speech disorders in children with CAS was found to be sufficiently reliable and valid, or indeed include estimates of reliability and validity (McCauley & Strand, 2008). Only one test, the Verbal Motor Production Assessment for Children (VMPAC) (Hayden & Square, 1999), provided norms that were adequately described, but other information was absent and operationalized definitions were therefore not met. However, a revised version of the VMPAC (VMPAC-R, 2022 (https://vmpac-r.com/#!/)) has recently been published.

The second, more recent, review had a broader aim to include studies on tests and discriminative features that might contribute to differentiating CAS from other SSDs (Murray et al., 2021). The review included 53 studies investigating different discriminative measures categorized as perceptual, acoustic, and kinematic (several studies investigated more than one of those) and the types of the variable assessed varied (e.g., rate, speech movements, prosody, articulation). Of these 53 studies, seven met the reviewer's quality criteria based on study quality and reporting, and adequate diagnostic confidence. Eight more studies nearly met the criteria, among them a study on DEMSS (Strand et al., 2013). Discriminative diagnostic variables that were found to reliably differentiate children with CAS from other SSDs (studies with reported sensitivity and specificity over 90%) were maximum performance rate (CAS to CD) (Thoonen, Maassen, Gabreels, & Schreuder, 1996); a set of measures, combining lexical stress matches, syllable segregation score, Percent Phonemes Correct on single words and accuracy on diadokokinesis task (CAS to nonCAS-SSD) (Murray et al., 2015) and a set of measures combining inconsistency, segmental accuracy and prosody (CAS to nonCAS-SSD, but with lower sensitivity, 65%) (Strand et al., 2013). Discriminative individual markers were also evaluated, without any being sufficiently sensitive and specific, except one nearly meeting sensitivity and specificity of >90%, e.g., inappropriate pausing (the Pause Marker (87/99), (Shriberg et al., 2017). The overall conclusion in the end of the thorough review was that reliable differential diagnosis requires a combination of measures (Murray et al., 2021).

2.3.1 Dynamic assessment in the field of speech- and language pathology

Dynamic assessment (DA) is a test procedure with a close link between assessment and intervention, making it valuable for SLPs. The DA procedure will extend the information on current level of performance of the child to a potential capacity when given support, based on Vygotsky's thoughts about the Zone of Proximal Development (Vygotsky, 1978). This would allow for two individuals with the same initial performance to be differentiated by the amount of support needed through the assessment (Hasson & Joffe, 2007). In static assessment, assistance and feedback is not allowed and the scoring is binary. There are two main methods within DA sometimes referred to as the "sandwich" and "cake" formats (Sternberg & Grigorenko, 2001). The sandwich format has a pretest, a teaching phase and a posttest and often use standardized tests in the test phases, while the cake format integrates the support in the assessment procedure to a graduated prompt/successive cuing format. Both these DA formats have been adopted by SLPs in research and are slowly implemented in clinical practice.

The test-teach-retest-format has been used to discriminate disorder from difference and promising results were found in DA of word learning for accurately identifying bilingual children with DLD (Kapantzoglou, Restrepo, & Thompson, 2012). In another study on bilingual children the results from DA of narratives using the Frog story showed high classification accuracy (Petersen, Chanthongthip, Ukrainetz, Spencer, & Steeve, 2017). In the area of speech production, the successive cuing format has been used to evaluate learning potential, readiness for treatment and measure change over time. A standardized dynamic assessment of phonology was recently published (Glaspey Dynamic Assessment of Phonology (GDAP) (Glaspey, 2019) targeting phonemes, blends and sound classes of English using a progression of cues and different speech context complexity. However, most important for the present thesis project was the Dynamic Evaluation of Motor Speech Skills (DEMSS) (Strand et al., 2013). DEMSS is a dynamic test designed to identify children with difficulties in motor planning and programming for speech using a successive cuing format. It includes nine subtests with simple words of earlier developing phonemes varying in length, phonetic complexity, vowel content and prosodic content (Strand & McCauley, 2019). Each word (utterance) is judged regarding articulatory accuracy, vowel accuracy, prosodic accuracy, and consistency. A graded cuing hierarchy is included in the administration, and in response to an erroneous production of the word scoring depends on the child's responses to the cued repetitions. Validity and reliability evidence for DEMSS was overall good with values of how well the DEMSS total score and subscores discriminate between children being over 90% for all values except the prosody score. The rationale for the development of DEMSS was the need for a criterion-referenced tool for children who are young or with a severe speech disorder, a tool to facilitate differential diagnosis of children with SSD, a dynamic tool to facilitate judgement of severity and prognosis and to facilitate treatment planning and stimulus selection (Strand & McCauley, 2019).

2.4 SPEECH DEVELOPMENT AND PROCEDURAL LEARNING

Knowledge on typical speech development help us understand the complexity and course of typical development to relate to while identifying children at risk for disordered speech. Speech motor development has been found to have a protracted course, which extends into late adolescence (Schötz, Frid, & Löfqvist, 2013; Smith & Zelaznik, 2004). Motor speech control develops continuously from the first years (Grigos, 2009), through preschool years to adolescence, with lip control similar to that of adults acquired by the age of five (Iuzzini-Seigel, Hogan, Rong, & Green, 2015). Acquisition of speech sounds follows a relatively predictable order (Vick et al., 2012). In Swedish most vowels are found to be established by the age of three, except [ʏ], [ɪ] and [ɵ] established by the age of four, and all consonants by the age of five, with the motoric refinement of /s/ and /r/ by the age of six (Blumenthal & Lundeborg Hammarström, 2014). This is in parity with international research showing a more stable lip and jaw control by the age of six (Green, Moore, Higashikawa, & Steeve, 2000). Iverson (2010) argued that the developing motor system contributes to the development of language and speech sounds. In addition, it was shown that more complex oral movements were more closely related to language skills than simple oral movements, possibly because they have a more speech-like character (Alcock, 2006).

During development, the coordination of sequences of speech sounds typically develop seemingly automatically and effortlessly into stable units, so called functional synergies (Smith & Zelaznik, 2004). Equally, cognitive-linguistic and motor skills are implicitly learned and automatically produced, referred to as procedural learning (Sanjeevan & Mainela-Arnold, 2017). All motor sequencing involves learning, leading to establishment of effortless sequences of movements. For example, learning to ride a bike would need some initial practice, but after a while you would hop on your bike without thinking of how to sequence the movements of your feet. The possibility of a deficit in the procedural learning system in children with DLD (Sanjeevan & Mainela-Arnold, 2017) and CAS (Iuzzini-Seigel, 2021) has recently been studied. For example, Iuzzini-Seigel (2021) found that children with CAS, on group level, demonstrated grammatical and motor impairments and required an increased number of exposures to the procedural learning task, compared to children with SSD or TD. The results from the study showed partial support for a link between coexisting speech, motor, and cognitive-linguistic difficulties in the same individual (Iuzzini-Seigel, 2021).

2.5 COEXISTING FUNCTIONAL DIFFICULTIES

Many children with CAS have coexisting difficulties. For the minority of children for whom neurobiological or genetic markers of CAS are suspected, these markers are sometimes consistent with the coexisting difficulties. However, many children with CAS have coexisting difficulties that are subtle or are even reported to have no coexisting difficulties at all.

The reported coexisting difficulties range over a variety of functional areas. Difficulties in attention and impulse control were reported as prominent problems by parents in a study on functional characteristics in children with CAS (Teverovsky, Bickel, & Feldman, 2009), as well as in a study by (Newmeyer et al., 2009) relative to aged-matched children. In the study by Newmeyer et.al. (2009) the children with CAS were found to process sensory information (e.g., sensory seeking, oral sensory sensitivity) in a more passive way compared to typically developing peers. Further, awareness of oral motor function in children with CAS has been increasing. For example, Murray et al. (2015) found a subset of children with structural and neurological deficits that had not been detected by the referring SLP. Some of the children were as old as 12 years and had undetected structural and/or dysarthric symptoms. The authors stress that an oral motor assessment should always be part of a speech assessment. This is especially important for children with suspected CAS, knowing that cooccurrence with other motor deficits is possible. This has also been emphasized by several other researchers such as Caruso and Strand (1999), Morgan and Webster (2018), Strand and McCauley (2019) and Mogren (2021).

One frequently studied functional area in relation to CAS is fine and gross motor performance. In a literature review on children with language disorder, coexisting with fine and gross motor difficulties (also incorporating some studies on children with CAS), Hill (2001) found substantial comorbidity and suggested deficits in a general underlying cognitive process as a possible explanation. The idea of a common underlying deficit in the planning and processing of movements have also been suggested by other researchers (Iuzzini-Seigel, 2019; Tükel, Bjorelius, Henningsson, McAllister, & Eliasson, 2015). Correspondingly, non-verbal sequential functions have been found to be affected and to correlate with the severity of CAS (Nijland et al., 2015) as well as evidence of a global sequencing processing deficit in individuals with CAS (Peter, Lancaster, Vose, Middleton, & Stoel-Gammon, 2017).

Interdependencies between linguistic and motor levels were also reported by Highman, Hennessey, Leitao, and Piek (2013), examining eight at-risk children (i.e., having a sibling with a CAS diagnosis) from 9 months to 2 years of speech sound development, expressive and receptive language, social skills, and gross and fine motor skills. The findings showed an underlying weakness in the at-risk children in expressive language and speech motor skills (Highman et al., 2013).

A cooccurrence of DLD is often described in children with CAS (Iuzzini-Seigel, Hogan, & Green, 2017). Sometimes the presence or absence of cooccurring DLD is used to allocate participants into subgroups (Lewis, O'Donnell, Freebarin, & Taylor, 2002; Murray et al., 2015). This may contribute new knowledge on the interrelationship between speech and language during childhood. For example, those children who had CAS and cooccurring language disorder showed significantly poorer syllable discrimination abilities compared to children with CAS only and typically developing peers (Zuk, Iuzzini-Seigel, Cabbage, Green, & Hogan, 2018). Findings suggest that speech perception deficit is not a core feature of CAS but rather is cooccurring with language disorder in a subset of children with CAS (Zuk et al., 2018). This would also be supported by the dual language stream hypothesis described by Hickok (2012). Morphological errors are sometimes difficult to derive as being motor or linguistic based errors. This was investigated in a study of twenty-six 4-5-year-old children with CAS who evidenced morphological errors on a standardized test (Murray, Thomas, & McKechnie, 2019). The results showed that the speech motor disorder could not explain the morphological errors found, indicating expressive DLD coexisting with CAS and subsequently implicating the need to apply treatment methods for morphosyntax in addition to the speech motor treatment.

Language disorder, as well as CAS, persists through life for some children. Approximately 5% of school-aged children have communication difficulties (Eadie et al., 2015) In a recent study the impact of CAS on functional experiences were examined through caregiver reports (Rusiewicz, Maize, & Ptakowski, 2017). The parents of 40 children aged 3-16 years participated. Four key concerns emerged related to intelligibility, peer relationships, relying on parent to be the child’s “voice” and emotional responses. The biggest concern among caregivers was about the speech production itself (Rusiewicz et al., 2017). Persistent language, reading and spelling difficulties have also been described, affecting education and future occupation for children with CAS (Lewis, Freebairn, Hansen, Iyengar, & Taylor, 2004; Turner et al., 2019; Zaretsky, Velleman, & Curro, 2010).

The coexistence of disorders among children with communication disorders has been addressed in a study by Miniscalco, Nygren, Hagberg, Kadsjö, and Gillberg (2006). They found that as many as 62% of all children with suspected language impairment at age 30 months had a confirmed neurodevelopmental disorder at 7 to 8 years of age. This could indicate that language disorder usually is not isolated, but rather an early marker for a neurodevelopmental disorder in the child. In line with these findings Gillberg (2010) established a term for often co-existing disorders; ESSENCE (Early Symptomatic Syndromes Eliciting Neurodevelopmental Clinical Examinations), including ADHD, developmental coordination disorder (DCD), ASD and DLD. He pointed out that major problems in at least one ESSENCE domain before age 5 often signals persistent deficits in the same or overlapping domains.

The International Classification of Functioning, Disability and Health (ICF) (WHO, 2020) provides a framework that has been applied in several studies on children with speech difficulties and CAS to study effects of speech and language disorder on different aspects of a child's activities and participation (Sharynne McLeod, 2009; S. McLeod & McCormack, 2007; Rusiewicz et al., 2017). The ICF may be used for describing everyday function, irrespective of the underlying health condition. It provides a framework to describe children in a holistic manner considering how the impairment affects body structure, and function as well as activity and participation. This functional assessment of a person can be an important complement to the medical diagnosis. In a survey on functional problems in children with CAS, 42 domains from ICF were evaluated (Teverovsky et al., 2009). The most prominent difficulties in addition to the speech disorder were found to be attention, vestibular function, temperament, fine hand use and learning to write (Teverovsky et al., 2009).

2.6 RATIONALE FOR THE THESIS

Although the theoretical definition of CAS has been internationally established, the exact assessment procedure that should be used in diagnostic settings has not been decided. The recognized need for a test addressing speech motor skills as a part of the diagnostic procedure for children with SSD was getting more attention in Sweden about 15 years ago. The idea to develop/adapt a Swedish test for this purpose was motivated by the promising information on the DEMSS available at the time. The DEMSS was under development and had been introduced in the literature (Strand, 2009), in education/talks (e.g., Linköping, Sweden, 2009) and subsequently evidence on validity was published (Strand et al., 2013). Thus, the rationale for the thesis project was to describe and evaluate a Swedish test of speech motor skills, as well as add knowledge on Swedish children with CAS, also sparce at the time, knowledge that hopefully could contribute to diagnostic decisions.

3 RESEARCH AIMS

3.1 GENERAL AIM

The overall purpose of this project was to investigate speech performance and speech characteristics in children with typical and disordered speech, to facilitate more confident diagnostic decisions of Swedish-speaking children with CAS.

3.2 SPECIFIC AIMS

The specific aims were to

- investigate speech movement accuracy in children with typical speech-language development (study I)
- describe the design of the speech motor test DYMTA (study I)
- evaluate the reliability and validity of DYMTA (study II)
- investigate CAS speech characteristics in a group of children with CAS diagnosis (study III)
- describe dynamic assessment used for assessment of speech motor skills (study III)
- describe the inter-individual differences in parent-reported coexisting functional difficulties among children with CAS (study IV)

4 MATERIALS AND METHODS

4.1 PARTICIPANTS

This thesis includes data from a total of 139 children, in the age range of 3:1 to 8:10 years. The participating children were a clinical group of 45 children with speech sound disorder (SSD) suspected or identified to have speech motor deficits and a group of 94 children with typical speech- and language development (TSD). The age range (3 through 8) was selected as most referrals for assessment and differential diagnosis of CAS are made in the preschool- and early school years. Also, we wanted to include school-aged children because of recent clinical experience assessing several children in these older ages with speech motor difficulties, not previously assessed with a speech motor perspective.

The 94 children with TSD were 45 boys and 49 girls between 37 and 106 months, enrolled from (nine) preschools and (four) schools in five Swedish cities in 2014 - 2015. All children were Swedish-speaking and 17 children also spoke one or two more languages (e.g., Arabic, Bosnian, Danish, English, Finnish, German, Greek and Persian). The inclusion criteria were a) normal hearing b) typical speech- and language development c) Swedish-speaking d) between 36 and 107 months. Exclusions made were due to lack of participation (n = 19), presence of speech and language difficulties (n = 12) and technical problem (n = 1), which resulted in 94 children fully analyzed from the 126 children whose parents accepted to be included in the study. See Table 1 for the distribution of age and sex.

The 45 children with SSD were 31 boys and 14 girls between 40 and 106 months, consecutively recruited by the first author in a clinical setting at the Speech-Language Pathology Clinic, ENT Department, Skåne University Hospital, during a defined period of 1½ years in 2014-2015. The inclusion criteria were a) a referral of SSD with a specified question about possible speech motor involvement or second opinion about CAS diagnosis, b) between 36 and 107 months c) normal or adjusted-to-normal hearing d) Swedish-speaking. Exclusion criteria were a) structural deficits (e.g., cleft lip and palate) b) autism spectrum disorder (ASD) c) lack of participation on DYMTA assessments. See Table 1 for the distribution of age and sex.

Of the 45 children in study II, the 33 children with CAS diagnosis were included in study III and IV because we were specifically interested in investigating the speech characteristics (study III) and coexisting functional difficulties (study IV) in this group (Table 1).

Table 1. Participant distribution for age and gender in all included studies.

Age groups (Year; months)	**TSD** Study I	boys	girls	**SSD** (CAS + nonCAS-SSD) Study II	boys	girls	**CAS** Study III, IV	boys	girls
3-3:11	28	14	14	4	3	1	4	3	1
4-4:11	17	6	11	13	10	3	6	5	1
5-5.11	12	3	9	12	9	3	9	7	2
6-6.11	21	13	8	8	6	2	7	5	2
7-7:11	10	7	3	5	2	3	5	2	3
8-8:11	6	2	4	3	1	2	2	0	2
Total	94	45	49	45	31	14	33	22	11

4.2 PROCEDURE

4.2.1 Data collection

The data collection of the children with TSD was made in 2014 and 2015 as part of three separate degree projects by speech-language pathology students (Gustafsson & Johnsson, 2015; Harjuniemi & Malkić, 2014; Lundvall & Smetana, 2014). The students were trained in test administration prior data collection, including thorough training of the test procedure, and scoring of DYMTA. Each student assessed a minimum of two children with typical development, recruited through convenience among friends, to practice administration and scoring of DYMTA and to ensure fidelity. Feedback and discussions were provided during physical meetings, internet meetings and via email until the students were judged to have reached sufficient competence. The data collection took place in a secluded room at the pre-schools/schools of the participating children. The assessment took about one hour per child and the child was offered pauses when needed. Documentation was made with audio/video recordings using a Sony Handycam HDR-CX250E or Canon Camcorder Legria FS200.

The data collection of all the children with SSD was made by an SLP with long experience in developmental speech- and language disorders and motor speech disorders (the author) in a clinical setting. Most appointments took two hours and were divided into two sessions on separate days. Documentation was audio/video recorded using a Canon Camcorder Legria FS200 with an external microphone (Sony ECM-MS957).

4.3 MATERIAL

All participants completed a comprehensive test battery, composed to cover language-, speech- and oral motor abilities. The tests and tasks chosen were the same for both the TSD and SSD groups except for tests of orofacial function.

Table 2. Overview of materials and outcome measures in each of the four studies

	Study 1	Study 2	Study 3	Study 4
DYMTA			x	
Total score	x	x	D	x
Subscores	x	x		
LINUS			x	
PCC	D	x	D	x
PVC	D	x	D	
PWC		x	D	
CAS Speech characteristics			x	
VMPAC				
Sequencing score				x
Oral motor score		x	D	
NOT-S	D			
GRAMBA	D	D	D	
TROG-2	D	D	D	
SIT	D	D	D	
Parental questionnaire				x
Parental reported background data				D

Note: x = the test scores or outcome data used for results in the study D = results are displayed as descriptive data and not included in analysis presented as a result in the specific study

4.3.1 Speech assessments and outcome measures

LINUS

The phoneme test Linköpingsundersökningen (LINUS) (Blumenthal & Lundeborg Hammarström, 2014) has been used to generate several outcome measures in this project. LINUS is a standardized test for assessment of phonology containing all Swedish consonants and vowels and elicited through picture naming. The targeted words were chosen as they are expected to be in the vocabulary of children from three years of age and include different word structures, word lengths, syllable, and stress patterns. There are 107 words and the 40 first words are comprising a short version, used in this thesis. These 40 first words closely mirror the full version and includes one- to four-syllable words (Lundeborg Hammarström, 2019).

The outcome measures based on assessment with LINUS were percent consonants correct (PCC), percent vowels correct (PVC), percent words correct (PWC), consonant inventory and vowel inventory. LINUS was also used as a speech sample for the assessment on present CAS features. A narrow transcription was made from the audio recordings. The PCC and PVC measures were calculated according to guidelines in Shriberg, Austin, Lewis, McSweeny, and Wilson (1997) as number of correct consonants/vowels divided by number of correct plus incorrect phonemes times 100. The phonemes were scored as correct or incorrect, where distortions were scored as incorrect and allophones as correct. Questionable speech errors were scored as incorrect.

For the phoneme inventory, presented in this thesis, the consonant examination was addressing all 18 Swedish consonants targeted three to five times. A few words with the phonemes /j, ŋ, ɕ/ were added from DYMTA-B to add up to three representations. A consonant phoneme was counted as acquired if it was correct in 50% or more of the targeted words (following the SVANTE Manual (Lohmander et al., 2005). For the vowel examination 17 Swedish vowels were addressed at least three times each. A vowel phoneme was also counted as acquired if it was correct in 50% or more of the targeted words.

DYMTA

Dynamisk motorisk talbedömning (DYMTA) was developed and piloted the year before the project started and published 2016 (Rex, McAllister, & Hansson, 2016). The test is motivated from Dynamic Evaluation of Motor Speech Skills (DEMSS) (Strand et al., 2013). However, DYMTA was created for Swedish conditions, expanded with stimuli and subtests, and incorporating a second, more demanding, part. Words are assessed from a coarticulatory perspective. The first part, DYMTA-A, was developed for younger children or those with severe SSD and has a hierarchical structure from early developing syllables, and simple CV-shaped to multisyllabic words, in eight subtests. In total there are 55 words and 187 judgements. The second part, DYMTA-B contains of articulatory more demanding words in nine subtests and was designed for children with less severe SSD. See Table 3 for an overview of subtests in DYMTA. In total there are 71 words and 237 judgements in DYMTA-B. Words in both DYMTA-A and DYMTA-B are judged on repetition with respect to *articulatory accuracy*, *vowel accuracy, prosody,* and *consistency*. The test is managed using Dynamic Assessment (DA) where systematic cuing (i.e., visual cues, simultaneous production, or tactile cues) is added to incorrect speech productions, facilitating accuracy and support change and learning. For *articulatory accuracy* the scale for scoring is multidimensional, to reflect the cuing made through the repeated attempts producing the word. *Vowel accuracy* has a three-point scale whereas both *prosody* and *consistency* use a binary scale.

Table 3. An overview of the subtests in DYMTA-A and DYMTA-B

Subtests DYMTA-A	Subtests DYMTA-B
1. CV	1. Simple syllables
2. VC	2. Voice- voiceless
3. CVCV	3. Dental – velar
4. CVC_1	4. Stop – fricative
4. CVC_2	5. Consonant cluster
6. CV_1CV_2	6. Word stress
7. C_1VC_2V	7. Word tonal accent
8. Multisyllabic words and different stress patterns	8. Multisyllabic words
	9. Increased length of utterance

The results on DYMTA are used in all four studies in this project, with analysis of the Total score and the Subscores in Study I and Study II, the Total score in Study III and IV, and DYMTA was also used as a speech sample for the assessment of CAS speech characteristics in Study III.

CAS speech characteristics

Video of the children's performance was coded offline for speech characteristics associated with CAS. The checklist used (study III) was adapted from the checklist introduced by Iuzzini-Seigel et al. (2015) and later presented in a tutorial by Iuzzini-Seigel & Murray (2017). The characteristics in the list are commonly accepted and consistent with those listed by ASHA (2007). The checklist, with modifications for the present project is shown in Table 4. Token-to-token inconsistency is not included in the table but was assessed. The CAS feature *Increased difficulty with multisyllabic words* is difficult to assess objectively and in a pilot study (Hjalmarsson & From, 2018) no participant was found to evidence this feature applying the magnitude of change score described in Iuzzini-Seigel and Murray (2017). Therefore, it was not used in study III.

Table 4. Checklist for speech characteristics associated with CAS used in the present project

Speech characteristics	Definition
Vowel error	The vowel is distorted, (i.e., the production is recognizable as a specific vowel, but it is not produced correctly and, may sound like it is in between two vowels, or it is substituted for another vowel.) It is not considered an error if the vowel is substituted with another phoneme that is consistent with an identified dialect spoken by the child and/or the caregiver.
Consonant distortion	A consonant production error in which the production is recognizable as a specific consonant, but it is not produced correctly (e.g., an /s/ that is produced with lateralization or dentalization).
Prosody errors	An error in which the appropriate stress or tonal accent is not produced correctly. For example, ba'nan (banana) (weak–STRONG) and 'banan (the path) (STRONG–weak) have different stress patterns. It is considered an error if the stress is inappropriately equalized across syllables or placed on the wrong syllable. For word tonal accent, tòmten (Santa) and tómten (the garden) have different accents. It is considered an error if the tonal accent is inappropriate equalized or the one substituted with the other.
Syllable segregation	Brief or lengthy pause between syllables that is not appropriate.
Groping	Silent articulatory searching prior to onset of phonation, possibly in an effort to improve the accuracy of the production. This characteristic must be assessed live or from a video recording.
Intrusive schwa	A schwa (epenthesis) is added in between consonants. For example, it may be inserted in between the consonants in a cluster (e.g., /blu/ becomes /bəlu/). This is not considered a vowel error.
Voicing error	A sound is produced as its voicing or voiced cognate (e.g., a /p/ that is produced as a /b/ or vice versa). In addition, this could also describe productions that appear to be between voicing categories.
Slow rate	Speech rate is slower than expected. It is slower during production of part or the whole word.
Nasality disturbance	Sounds either hyponasal (not enough airflow out of nose/"stuffy"), hypernasal (too much airflow out of nose for non-nasal phonemes [e.g., plosives]) or alternate between hypo- and hypernasality.
Difficulty achieving initial articulatory configurations or transitionary movement gestures (for short: deviant articulatory transitions)	Initiation of utterance or initial speech sound may be difficult for child to produce and may sound lengthened or uncoordinated. Also, child may evidence lengthened or disrupted coarticulatory gestures or movement transitions from one sound, syllable, or word to the next.

Note: Definitions are adapted to Swedish from Iuzzini-Seigel, Hogan, Guarino & Green, 2015.

4.3.2 Assessment of orofacial function

NOT-S

Nordic Orofacial Test – Screening (NOT-S) (Bakke, Bergendal, McAllister, Sjögren, & Åsten, 2007; McAllister & Lundeborg Hammarström, 2014) is a test for orofacial function. It consists of two parts: a structured interview and a clinical examination. The interview part has 6 sections with questions on difficulties with oral sensitivity, breathing, habits, chewing, drooling and dryness of the mouth. The clinical examination part also includes 6 sections: face at rest, nose breathing, facial expression, masticatory muscle and jaw function, oral motor function, and speech. NOT-S was used in Study I to examine the participants with typical development, to make sure they were within typical range. It was administered and scored according to the instructions at the Mun-H-Center website (https://www.mun-h-center.se/siteassets/munhcenter/3-information-och-utbildning/4--not-s/not-s-manual_eng_090625.pdf).

VMPAC

The Verbal Motor Production Assessment for Children (VMPAC) (Hayden & Square, 1999) is an assessment of the neuromotor integrity of the speech production system using speech and non-speech tasks. The test items are systematically organized into the subtests *Global Motor Control*, *Focal motor control* and *Sequencing*, and two supplementary subtest which are *Connected Speech & Language control* and *speech characteristics.* The subtest *Sequencing* is used as one of the outcome measures in study IV. In this subtest the focus is sequence maintenance over items of both non-speech oromotor movement sequences and speech oromotor movement sequences, where the speech items are 18 versus five non-speech items.

The first 37 items of VMPAC were summarized as a total non-verbal oral motor score (OMS) comprising orofacial integrity and non-speech oromotor skills. The OMS includes observation on tone, phonation, reflexes, chewing, single oromotor movements of mandibular, labial-facial, lingual control and double oromotor movements. It also includes observations on anatomy and overall smoothness of movement. The OMS was used in study II and III.

4.3.3 Language assessments

Three tests were used for language assessment for both SSD and TSD children included in this project: GRAMBA (Grammatiktest för barn) (Hansson & Nettelbladt, 2010), Test for reception of grammar (TROG-2) (Test for Reception of Grammar – Second Edition) (Bishop, 2009) and SIT (Språkligt Impressivt test) (Hellquist, 2011). These tests were chosen to evaluate both expressive and receptive grammar. GRAMBA is a standardized test for different grammatical structures of Swedish (e.g., noun and verb forms as well as word order) with norms for children 3 to 6 years. TROG-2 is a standardized test for reception of grammatical contrasts marked by inflections, function words and word order, with Swedish norms from 4 to 16 years. Because of the inclusion of children from 3 years, we assessed this age group with SIT. SIT is a test for comprehension of Swedish with reference values from 3 to 6 years.

For the TSD children (Study I) the purpose of the language assessment was two-folded: 1. to exclude children with language impairments and 2. to present descriptive language data on the participants. For the first purpose we decided on a cut-off at below the 10th percentile to be nontypical. GRAMBA was used for all participants and for children from 6 years a cut-off was set to 1.25 SD from the mean to match the cut-off of percentile 10. Swedish TROG-2 has established norm data from 4 years and results were converted to percentiles according to the manual. For the SIT results we applied a cut-off at 1.25 SD from mean (13.8) for inclusion (within typical range) in accordance with the reference data (presented in the manual as a raw mean value of number of incorrect answers per age group which is 14 for 3- year-olds). For the SSD children in Study II and Study III the language data were used as descriptive data.

4.3.4 Parental questionnaire on body functions

A parental questionnaire on body functions was designed for the project and used in study IV. Our study on functional coexisting difficulties in children with CAS was inspired by the study by Teverovsky et al. (2009), using domains from the ICF (WHO, 2013). Forty statements on functions were chosen from six relevant chapters in the ICF: mental functions (b1), sensory functions (b2), voice and speech functions (b3), neuromusculoskeletal and movement related functions (b7), learning and applying knowledge (d1) and Interpersonal interactions (d7). Most questions (36) targeted the Body Function component in the ICF and four targeted the Activities and Participation component. The written questionnaire was designed specifically for this project and was administered to caregivers of all participants.

4.4 RELIABILITY

4.4.1 Study I

A point-by-point percent agreement was used for inter- and intrajudge reliability for DYMTA results of typically developing children, on a level of every single score. DYMTA-A has subtests with 55 words and a total of 187 judgements, whereas DYMTA-B has 71 words and 237 judgements. The scoring made by the research assistants collecting the data for this study, were compared with the scoring made by the first author, using the formula: agreements/ (agreements + disagreements) x 100. For the 25 (27%) randomly selected participants reliability was 96.5% for DYMTA-A and 96.3% for DYMTA-B. Intrajudge reliability was calculated on 28 (30%) of the participants and was 99%.

4.4.2 Study II

A point-by-point percent agreement and Intraclass Correlation Coefficients (ICCs) were used for inter- and intrajudge reliability of DYMTA results for the children with SSD. Twenty percent randomly selected children (n = 9) were included in the analysis and were checked for severity to make sure different degree of severity was represented, as reliability may be more difficult to obtain in a more severe impairment. The first and second rater, each with comprehensive experience of SSD and CAS, scored the administrations of DYMTA-A and DYMTA-B from video/audio-recordings. The point-by-point interjudge agreement was 0.91 and 0.87 respectively for total score and the ICC was 0.97 and 0.96 respectively for the total score. Intrajudge point-by-point agreement was 0.98 and 0.95 respectively for total score and for ICC 0.99 and 0.99 respectively for total score.

4.4.3 Study III

A point-by-point percent agreement was used for inter- and intrajudge reliability for the CAS characteristic ratings. The evaluations were made independently by the raters, for each of the two tests. Interjudge reliability for 13 (39%) randomly selected participants was 94.4% (range 0.818 – 1.0) for the DA sample and 90.9% (range 0.818-1.0) for the naming sample. The formula agreements/ (agreements + disagreements) x 100 was used.

4.4.4 Study IV

The answers from a parent questionnaire were the main outcome measure in this study not subject to a reliability analysis. Thus, DYMTA-B results for the children with CAS were used as one parameter in the analysis. All reliability data for DYMTA is described above (Study II).

4.5 STATISTICAL ANALYSIS

Descriptive statistics were used in all four studies to describe demographic information such as age and gender, and to present other variables such as results on language- and orofacial function tests. All statistical methods described in the paragraphs below are summarized in Table 5.

Table 5. Overview of statistical methods used in the thesis

Statistical method		Study 1	Study 2	Study 3	Study 4
Mann-Whiney U Test	Compare differences between two groups (nonparametric)	x			
Kruskal–Wallis Test	One-way ANOVA to compare three or more groups (nonparametric)	x			
Spearman's rho	Measure the strength of association (i.e., correlation) between two variables (nonparametric)	x			
Sensitivity	How well we identify those with the disease		x		
Specificity	How well we identify those without the disease		x		
Receiver operating characteristic curve (ROC)	Sensitivity and specificity pairs in function are plotted with different cutoff values of a parameter		x		
Area Under the ROC curve (AUC)	How well a parameter (or test) can distinguish between two diagnostic groups		x		
Likelihood ratio for a positive result (LR+)	How much more likely a child with disorder is identified by the test, than a child without the disorder is falsely identified.		x		
Likelihood ratio for a negative result (LR-)	How much more likely the test correctly identifies a child as not exhibiting the disorder, than misses a child with the disorder		x		
Intraclass correlation (ICC)	A reliability index that reflects both degree of correlation and agreement between measurements.		x		
Diagnostic odds ratio	The odds of a positive test in those with the disease relative to the odds of a positive test in those without disease (LR+/LR-)		x		
Dependent Student´s t-test for paired samples	Comparing differences between two samples for the same individuals			x	
Heatmap	A visualization of data via a graphical representation where values are illustrated by color.				x
Agglomerative clustering	How individuals are combined into clusters (groups) from similarity, often presented as a tree-like dendrogram				x
Boxplot	Visualization and comparing of median, minimum, and maximum values	x			x

4.5.1 Study I

Nonparametric tests were used because the samples showed negative skewness (DYMTA-A .92, DYMTA-B 1.07). A Mann-Whitney U Test was used for comparing sex and mono- vs multilingual children for the total group, followed by a Kruskal–Wallis Test for comparing within age groups. Spearman's rho was used for correlations. A significance level (alpha level) of 0.05 was used throughout.

Statistical Package for the Social Sciences (SPSS) (version 26.0, 2018) was used for the statistical analyses. Figures were made in R (R Core Team, 2017).

4.5.2 Study II

In study II we evaluated DYMTAs ability to discriminate between children with a diagnose of CAS from children with other SSDs in order to estimate its discriminative performance and validity. Several performance measures were therefore calculated: To evaluate how well DYMTA could separate groups as its discrimination threshold was varied, a graphical plot known as a receiver operating characteristic curve (ROC) was created. From this, DYMTAs overall ability to discriminate was calculated as the Area Under the ROC curve (AUC). Further, for a specific decision threshold, we also calculated DYMTAs sensitivity, the proportion of participants that are identified with the disorder (true positives), and specificity; the proportion of participants that are identified not having the disorder (true negatives). Further, likelihood ratios were also calculated to assess how much more likely a child with CAS would be detected by the DYMTA test and its subtests than a child without the disorder would be falsely detected (LR+) and how much more likely a child without CAS would be detected as not having CAS than missing a child having the speech disorder (LR-).

Inter- and intrajudge reliability were also thoroughly assessed in the study using a point-by-point percent agreement and Intraclass Correlation Coefficient (ICC). A point-by-point percent agreement compares every score between raters using the formula agreements/ (agreements + disagreements) x 100. The ICC has one more dimension than point-by-point agreement comparing the variability of different ratings of the same individuals to the total variation across all ratings and all individuals. In our study the ICC was calculated by using a random-intercept mixed-effect linear model, with participants as random effects and only an intercept as the fixed effect, following Strand et al. (2013). Data analysis was carried out in R (R Core Team, 2017).

4.5.3 Study III

In order to compare the performance of two assessment methods, one static and one dynamic, we calculated the number of noted speech characteristics found using each method. The average number of noted speech characteristics for each participant were then compared via a Student's *t*-test for pared samples as well as calculating a 95% confidence interval for the difference scores. The distribution of number of CAS speech characteristics per method was illustrated in a boxplot figure illustrating median, quantiles, and range values.

4.5.4 Study IV

The parent reported answers to the statements in the questionnaire were calculated for the relative frequency of disagree-responses to each individual item. An agglomerative hierarchical clustering procedure was then applied to identify groups of children showing similar types and/or number of functional difficulties. In an agglomerative clustering procedure, each individual starts as a separate cluster and are sequentially merged together, until all clusters have been merged into one. The result of this procedure was illustrated in a dendrogram. The individual item-responses per participant were also illustrated in a heatmap simply to visualize the identified patterns of individuals. The groupings found (based on functional difficulties) were also described graphically using boxplots to evaluate how they were presented on some speech outcome measures. Data analysis was carried out in R (R Core Team, 2017).

4.6 ETHICAL CONSIDERATIONS

The participants in this thesis have all been asked to participate and given their consent verbally and in writing. Although all the participants are children 3 to 9 years old and their parents have made the decision for them, they have all thoroughly been told about the test procedure and video recording. All the participants and their guardians have had the opportunity to withdraw from participation in the research study during or after the assessment session/s. This was the situation both for the typically developing group seen at their school as well as for the clinical group seen at the hospital.

The legal guardians of the children in the clinical group were all appreciative for being asked to participate and they were all interested in getting as much information as possible on their child's speech and language ability and impairment. The assessment procedure took up to two hours per child and all guardians were very supportive when the test leader encouraged the child to carry out all the tests and all the test items. If a child was tired or unfocused there was inset a break. The test leader made the climate of the sessions safe and comfortable for the child and the legal guardians.

The aim of the present study was to describe speech, oral motor and language skills in children aged three to nine to enhance the diagnostic procedure. The test procedure involved tasks where the child was looking at pictures for naming, telling, or pointing, repeating words given by the test leader or was asked to do voluntary movements with the mouth (i.e., tongue, lips, jaw). These tasks were not harmful or hurtful for the participant in any way. In two of the tests of the assessment procedure tactile cues were used as one of the cues available in a hierarchy. When a tactile cue was used the child was told beforehand what was going to happen, and the test leader sometimes also demonstrated on her own mouth or face to make the child feel comfortable. If the child in any way said or showed that he/she did not want to have this kind of guidance for speech movements, only verbal and/or visual cues were given, but most of the children participated in all tasks.

The handling of the data for the project was made with respect to confidentiality. This was made by using code names for all children and there is only one code key, kept in a locked safe. The data is processed and analyzed in an objective way. Ethical approval was obtained for the research project from the Regional Ethical Review board in Lund.

5 RESULTS

5.1 STUDY I

Included participants, all having typical speech development, performed well on the task items in DYMTA. The total score gave a mean of 98% (range 89 – 100) for DYMTA-A and 95% (range 81 – 100) for DYMTA-B for the total group of 94 children aged 3;1 to 8;8 (Figure 3). There was a significant correlation with age for both DYMTA-A (r = 0.49, p = 0.001) and DYMTA-B (r = 77, p = 0.001). There was no significant difference between gender (DYMTA-A U = 970, p = 0.306 and DYMTA-B U = 1098, p = 0.971), or between mono- and multilingual children (DYMTA-A U = 537, p = 0.147, DYMTA-B U = 622, p = 0.556).

The median results on the subscores for the measures *vowel accuracy* and *prosody* reached ceiling values across ages, whereas *articulatory accuracy* and *consistency* reached ceiling values from the age of five. The two latter subscores had a developmental tendency. When studying the subtests in DYMTA-A, it was found that all participants had a low variability and high scores already from the age of three. The median value was one hundred percent from the age of five. In DYMTA-B the results also showed low variability over subtests, although *multisyllabic words* had somewhat more variability. For this subtest and *word stress* a developmental tendency was also found. Other speech outcome measures reported on were PCC and PVC. The mean PCC for the group of the 94 TSD children was 94% and the mean PVC 99%.

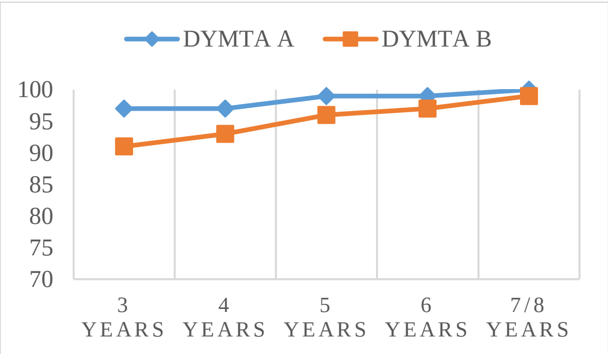


Figure 3. DYMTA-A (blue line) and DYMTA-B (red line) total mean score in TSD children across ages.

5.2 STUDY II

The Swedish DYMTA was developed to be used to facilitate identification of children with CAS in a group of children with different types of SSDs.

The results of the preliminary validation of DYMTA showed an overall good validity and reliability. The results on *intra*rater reliability agreement and ICC for total scores were very strong (DYMTA-A 98%; DYMTA-B 95%) as well as for most subscores. The *inter*rater reliability was also strong for total scores on both reliability measures: agreements of 91% and 87% and ICC values of 97% and 96%. For the separate subscores the *inter*rater agreement measurement was also strong across all subscores, as was ICC for all subscores except *prosody*.

A relatively small scoring difference across the two raters on two children drove the variability that generated the low ICC value for *prosody*.

The results on validity showed that DYMTA was discriminating between children with SSD and CAS with an AUC at 0.92 for DYMTA-A and 0.94 for DYMTA-B total score. The total score also had acceptable sensitivity (0.73 and 0.85) and specificity (0.92), and a high diagnostic odds ratio (29 and 62) for the cutoffs determined, using the positive likelihood ratio.

5.3 STUDY III

The three most prominent CAS characteristics in the dynamic task were (except inconsistency) *deviant articulatory transitions*, *prosody errors*, and *vowel errors*. The most prominent CAS characteristics for the naming task were *vowel errors* in all participants, followed by *voicing errors* and *prosody errors* (Table 6). The mean number of detected speech characteristics in the dynamic task was 6.85 (SD=1.33) (median value 7) and on average 5.24 (SD=1.48) (median value 5) in the static task. The mean number of characteristics over tasks were 7.24. Further, the frequency of occurrence of each CAS characteristics was investigated for this thesis and found to vary across participating children. Preliminary results based on observations over the two tasks are illustrated as a heatmap, in Figure 4.

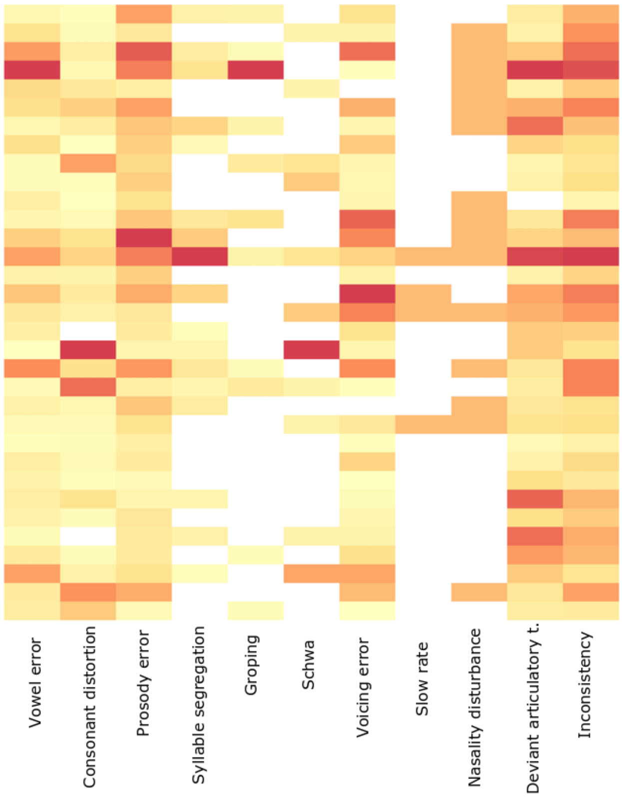


Figure 4. A heatmap to illustrate the relative frequency of each detected CAS speech characteristics (columns) for each participant (rows); darker red indicate relatively more occurrence of the characteristic. The frequency was scaled to express which child had the most frequent problem per CAS characteristic (column). Thus, the dark red cells illustrate that the child in that row had the most frequent occurrence of the CAS characteristic within that column. Orange cells as used for *Slow rate* and *Nasality disturbance* indicate that the characteristic is evident for the participant. White cells indicate that the child in that row had no signs of the problem (column).

Table 6. Number of children evidencing each CAS speech characteristics assessed, in the DA task, SA task and over the two tasks.

CAS speech characteristics	Dynamic task *n* (%)	Static task *n* (%)	Over tasks n (%)
Inconsistency	33 (100)	n.a.	33 (100)
Deviant articulatory transitions	33 (100)	16 (48)	33 (100)
Vowel errors	33 (100)	33 (100)	33 (100)
Prosody error	32 (97)	26 (79)	33 (100)
Voicing error	29 (88)	28 (85)	31 (94)
Consonant distortion	29 (88)	22 (67)	29 (88)
Nasality disturbance	19 (58)	14 (42)	19 (58)
Articulatory groping	17 (52)	8 (24)	18 (55)
Intrusive schwa	16 (48)	13 (39)	21 (64)
Syllable segregation	14 (42)	9 (27)	18 (55)
Slow rate	4 (12)	4 (12)	4 (12)

Note: CAS = Childhood Apraxia of Speech, n = number

5.4 STUDY IV

The five most frequently parent-reported functional difficulties were *understood by others* (82%), *retrieval of words* (67%), *trying new food* (67%), *being distracted by sounds* (55%), and *impulse control in body at rest* (52%). Least reported difficulties (under 10%) were regarding *smell*, *taste*, *salivating* and *orientation to person*. Reported difficulties were in the range of 1 to 27 in the individual participants. Also, the combination of items varied within the total participant group and after a cluster procedure two large, equally sized groups were identified: group 1 with relatively few coexisting difficulties, and participants in group 2 with more functional difficulties. These two groups were then further divided into two, resulting in four meaningful groups with separate profiles. Group 1A had few reported difficulties, group 1B had mainly difficulties in voice and speech rhythm functions, group 2A in functions such as attention and planning activities and group 2B in diverse functions, including motor functions. The four groups were also related to information from speech measures. A group difference (based on the functional domains) within each main group (1 versus 2) was found on results from speech measures, where a lower performance on DYMTA, PCC and sequencing could possibly be connected to coexisting deficits in the voice domain (within group 1) and motor difficulties (in group 2).

5.5 GROUP COMPARISONS

This thesis project did not include any studies on group comparisons of children with TSD and children with CAS. However, the results on DYMTA, PCC, PVC and PWC for both groups have been presented as main or background information in different studies in the thesis. The results on these speech outcome measures are summarized in Table 7. Additionally, results on consonant inventory and vowel inventory for each participant group are included in the table.

Table 7. Group level summary of mean values on some speech outcome measures for children with TSD and CAS.

Variable	**TSD n=94**	**CAS n=33**
Age, mean, in months	64	69
DYMTA-A, total in percent	98	73
DYMTA-B, total in percent	95	58
PCC	94	54
PVC	99	89
PWC	82	19
Consonant inventory (range, max 18)	17.2 (14-18)	11.8 (2-17)
Vowel inventory (range, max 17)	16.8 (15-17)	15.3 (8-17)

Note: DYMTA = Dynamisk Motorisk Talbedömning, PCC = Percent Consonants Correct, PVC = Percent Vowels Correct, PWC = Percent Words correct, calculated from LINUS (see method 4.3.1 for further explanation)

6 DISCUSSION

The overall purpose of this project was to investigate speech performance and speech characteristics in children with typical and disordered speech, to facilitate more confident diagnostic decisions of Swedish-speaking children with CAS.

The thesis project started with a newly developed Swedish test for speech motor skills in children with SSD and suspected CAS successively administered to both typically developing children and children with idiopathic speech disorder. The description of the test itself, and performance data on children without speech disorder, led to an investigation of psychometric evidence of validity and reliability in the second study. Since the speech motor test in focus in this thesis use a dynamic assessment approach, the benefits of dynamic assessment in a group of children with CAS were explored in the next study. In the fourth study coexisting functional difficulties for children with CAS was addressed. Findings from the studies in this project will be discussed below.

6.1 DYMTA AND TYPICAL SPEECH DEVELOPMENT

In the first study (study I) on 3 to 9 year old typically developing children, it was found that children with TSD performed with high scores, i.e., good performance, on tasks in DYMTA. Several children even reached ceiling level for some subscores across ages. This was observed in both DYMTA-A and DYMTA-B. From the literature reviewed, high performance on speech motor control was anticipated for the TSD children on the tested variables (i.e., the subscores of DYMTA): articulatory accuracy (Iuzzini-Seigel, Hogan, Rong, et al., 2015), vowel accuracy (Blumenthal & Lundeborg Hammarström, 2014), prosody (Samuelsson, Reuterskiold, Nettelbladt, & Sahlen, 2011) and consistency (Holm, Crosbie, & Dodd, 2007). Additionally, for articulatory accuracy in DYMTA-B we found a developmental tendency which is also supported by prior research (Schötz et al., 2013; Smith & Zelaznik, 2004). For example, variability of lip movements across repetitions of the same utterance decreased with age, from five years of age up to 30 years (Schötz et al., 2013) appointing a protracted time course for speech motor control.

Vowel performance was the one subscore that had high scores already from the age of three on both DYMTA-A and DYMTA-B, in line with earlier findings that most vowels in Swedish are established by the age of three (Blumenthal & Lundeborg Hammarström, 2014). Even the articulatory and prosodic more complex words in DYMTA-B did not seem to affect the performance on vowels in the participating children. The knowledge of early acquisition of vowels, also found for English (Pollock & Berni, 2003), may have informed on decisions when developing a recent new test on phonology, not including test items on vowels at all (Glaspey, 2019).

Consistency of speech production was found to be high on all targets from age 5 in both DYMTA-A and DYMTA-B. Reports on variability in TSD children vary from low (Holm et al., 2007) to high (Sosa, 2015). One factor for different results could be that the specific words or tasks used have an effect on the outcome. For example, in a study using words with consonant clusters the variability was found to be 53.7% (McLeod & Hewett, 2008) compared to 13% variability reported in a study where most words were not having clusters (Holm et al., 2007). The words in DYMTA-A and DYMTA-B were developed to differ in articulatory complexity (with later-acquired, more motorically complex phonemes and phoneme sequences, e.g., clusters, in DYMTA-B) and our findings showed that the child was more inconsistent in producing the words in DYMTA-B.

In DYMTA token-to-token (word) variability is used, but phonemic (speech error) variability would also be used for disordered speech. In a study using both token-to-token variability and phonemic variability it was found that token-to-token inconsistency of monosyllabic real words and the sentence *Buy Bobby a puppy*, were the best differential markers for children with CAS and SD (with moderate sensitivity and specificity) (Iuzzini-Seigel et al., 2017). Also, the authors suggested speech inconsistency to be a core feature of CAS as it could not be attributed to the coexisting language disorder evident in some participants with CAS. Inconsistent production has also been found to be important to assess according to survey studies (Forrest, 2003; Malmenholt et al., 2017).

It should be noted that DYMTA is standardized and provides a criterion-referenced measure, i.e., the examined behavior of a child is compared to the expected performance of that behavior. Typically, a comparison with a normative sample is not the main interest with criterion-referenced tests, but we found an interest in investigating the performance of typically developing children over the different age groups on the different variables in DYMTA. For phonological development, new reference data have recently been presented for Danish (Clausen & Fox-Boyer, 2017) as well as speech sound development in early childhood from a dynamic assessment perspective for English (Glaspey, Wilson, Reeder, Tseng, & MacLeod, 2022). In Sweden a couple of tests for phonology are available, but reference data have been sparce until normative data was published for Swedish in 2014 and 2017 (Blumenthal & Lundeborg Hammarström, 2014; Lohmander, Lundeborg, & Persson, 2017). Consequently, the results from English-speaking children have earlier been used as reference data in the diagnostic procedure for Swedish-speaking children. Although our participant group of 94 TSD children is too small to constitute a normative sample, the results could definitely be seen as reference data (e.g., as displayed in Table 7) and would also add to the increasing body of knowledge of cross-lingual speech performance in children.

Languages differ on several aspects which could have an impact on the clinical identification of speech sound disorders including CAS. Also, assessment procedures and test protocols are often developed for a specific language to be performed by a test leader of that same language. In a review by McLeod & Verdon (2014) 30 speech assessments in 19 languages were evaluated. They found that 70% were for use with monolingual speakers, 20% for one language of bilingual speakers and 10% for both languages of bilingual speakers. DYMTA was developed for Swedish-speaking children, but not specifically for monolingual speakers. As results were not indicating any significant difference between mono- and multilingual children on total scores or subscores it is possible that DYMTA would be useful for both populations.

6.2 DYMTA AND EVIDENCE OF VALIDITY

In the second study (study II) findings revealed acceptable to strong evidence for reliability and validity of DYMTA, for our clinical participant group of 45 children with SSD and CAS. The reliabilty (i.e., the degree of consistency of a measure) was strong for all subscores and the total score, except for interrater judgment on the prosody subscore (the ICC value). Even though it was only the judgement on two children that drove the lower results on the ICC for the prosody subscore, it is important to address the possible factors affecting a confident interpretation of Swedish prosody. One possible explanation would be that the prosodic system is quite complex, consisting of contrasts of vowel length, word stress, and word tonal accent (Bruce, 2007). It would also be likely that Swedish SLPs are not as trained to assess prosody as other speech variables, for instance phonological patterns (Samuelsson & Nettelbladt, 2004). Also, as the test items are single words, including several monosyllabic words, a lot of prosodic information from sentence level and connected speech could possibly be missed.

The evaluation of validity of DYMTA showed an overall acceptable to good ability for the test to discriminate between children with CAS and nonCAS-SSD. Findings from AUC values for most subscores and especially for the total score on both tests was high (92% in DYMTA-A and 94% in DYMTA-B). As AUC is a measure not influenced by the balance between affected and unaffected individuals in a sample and there are more children with CAS than children with other SSDs in our group, these values and high diagnostic odds ratio (29 and 62) indicate that adding DYMTA to our assessment battery could help facilitate our diagnostic decisions. However, when evaluating the separate subscores, (i.e., the separate measures of vowel accuracy, articulatory accuracy, prosody, and consistency) some estimates, especially for the prosody score, were not meeting recommended values for validly separating the groups. The combination of measures certainly seems to be most discriminative, in line with what was found in the recent review by Murray et al. (2021).

DYMTA with its dynamic approach, was developed with the intent to facilitate differential diagnosis, to measure change and determine severity. Assessment tools commonly are designed to specifically classify before making decisions about treatment (Dollaghan, 2007). However, the purpose of using a test may depend on the situation and what decisions the clinician has planned (e.g., differential diagnosis, measuring change or determining severity) (Daub, 2021). The question to ask would then be if the test is adequate for the (different) decision(s) needed to be made concerning the child, having the clinician find a test that possesses evidence on that.

6.3 OBSERVATION OF SPEECH CHARACTERISTICS FOR CAS USING DYNAMIC ASSESSMENT

In the third study (study III) on observations of CAS speech characteristics in two different speech samples, findings revealed a difference in number and type of detected speech characteristics for the same child. For 82% of the children more CAS characteristics were evident in the DA sample than in the SA sample with a median of 7 and 5, respectively. The type of most prominent characteristics also differed. The findings will be discussed in the following sections.

The introduction of dynamic assessment in a test for speech production in Swedish has been made through this project. However, the intention has not been for DA to replace traditional (i.e., static) assessments, but to be used in conjunction with it (Lidz, 1987). There are three central advantages with DA in the differentiation of motor speech deficits in children with SSDs (Strand & McCauley, 2019): the observation of specific speech characteristics, allowing judgement of severity and facilitating treatment planning. The first advantage, enabling observation of specific speech characteristics, was supported by the findings from the present study. More information on the child's difficulty reaching the articulatory goal was added from the outcome of the DA test, especially regarding the features *deviant articulatory transitions, groping* and, *syllable segregation*. These were behaviors not seen in the same degree in the naming task. DA may also facilitate judgements on severity and prognosis (Peña et al., 2006) from how much and what type of cuing is needed for the child to improve performance. Two children with the same initial response on a targeted word could need different types and number of cues helping the clinician to predict progress and get a sense of severity. This effect of cuing on the readiness for learning or change has also been found in language learning (Bain & Olswang, 1995). Information about the amount and types of cuing on each child is available in our raw data, but not yet analyzed suggesting a presumptive future project. Additionally, the dynamic assessment assists in treatment planning (Hasson & Joffe, 2007). Types of cues found to be helpful for the individual child, together with types of errors (e.g., specific movement transitions, syllable shapes, or prosodic aspects) would be considered when choosing words for the stimulus sets. Further, the dynamic approach has been described to be *"…appropriate for SLT use, where the relationship between therapist and child can enhance the performance of the child in both assessment and therapy, creating a feeling of greater competence and motivation in the learner"* (Hasson & Joffe, p. 12, 2007).

In children with idiopathic CAS, as in the present thesis project, there are typically no genetic or neurobiological information available, instead it is the behavioral symptoms that commonly lead us to the underlying speech processing level of breakdown (Terband, Maassen, et al., 2019). It is widely accepted that all three ASHA consensus-based core features should be met as a minimum for a CAS diagnosis (McCabe, Murray, & Thomas, 2020), even though the best way to capture them still is challenging (Terband, Namasivayam, et al., 2019). In our study we were using the Iuzzini-Seigel checklist (Iuzzini-Seigel, Hogan, Guarino, et al., 2015) for observable signs over two tasks, and all CAS features, except *slow rate,* were demonstrated in more than half of the participating children with CAS. Further, all children evidenced *deviant articulatory transitions, prosody errors* and were *inconsistent* thus meeting the ASHA consensus criteria. To use a combination of several features to ensure finding the signs corresponding to the three ASHA consensus criteria has been supported in previous studies. For example, observed CAS features from the Iuzzini-Seigel checklist (2015) in a group of children with CAS, were exploratory analyzed using a three-factor model and found to load on the factors prosody, coarticulation, and inconsistency (Chenausky et al., 2020). Additionally, several discriminative variables were found in studies reviewed by Murray et al. (2021) and it was summarized that *"Combinations of measures are more efficacious than singe diagnostic markers, […]"* (Murray et al., 2021, p. 17). These findings support the need to identify several speech characteristics associated with speech motor deficits, to be able find and describe the core deficit of planning and programming movements for speech. For Swedish, DYMTA is the first standardized test with targets aimed at assessing a combination of variables consistent with the core consensus-based features.

Our findings on the total number of observed CAS speech characteristics were in line with results from previous studies. The group of children with CAS in our project (including nine children with DLD) had an average of 7.24 features over tasks, comparable to 7.14 in children with CAS in a study by Centanni et al. (2015) and 8 features for CAS and 7 for CAS with DLD in a study by Iuzzini-Seigel et al., (2017). The number of different speech characteristics are used for group assignments in these studies.

However, every specific speech characteristic commonly is observed to a different degree in the individual child. Thus, a child with few occurrences of a feature will be compared to a child with multiple occurrences. Consequently, the use of number of CAS features may be one way to report on severity in children with CAS, but the frequency of occurrence of each feature could give additional value, as illustrated in a heatmap in this thesis (Figure 4). When severity of a speech disorder is discussed, several different measures is used in the literature. PCC has often been used (Waring, Rickard Liow, Dodd, & Eadie, 2022). Also, severity based on variability in reading, vocabulary, and articulation was found to define subgroups of children with CAS (Stein et al., 2020). The use of a measure indicating severity is thus dependent on the question asked. In assessments targeting speech motor skills, measures of sequencing and articulatory accuracy could be appropriate to use for a measure of severity. For example, the outcome measures from speech motor and oromotor assessments are measures that carry information on severity, such as VMPAC (Hayden & Square, 1999), or even measures of phoneme proficiency such as consonant and vowel inventory. These measures should then also be put in the perspective of intelligibility and the involvement of coexisting functional deficits (Chenausky et al., 2020). Consequently, to assess the complexity and effects of a speech disorder several aspects and measures are needed, and they all signal involvement of severity from different viewpoints.

As the focus of our study (III) was single-word speech samples, we did not report on CAS features evident in connected speech. However, perceptual assessment of observable speech characteristics in a connected speech sample (using the thematical picture included in the DYMTA test (the Buss picture, Rex et al., 2016)) was done in the larger project. The outcome showed fewer observable features than in the DA and naming samples for the same child. Although connected speech sampling could carry advantages such as efficiency and ecological validity, as well as permitting observations of phrasal aspects of prosody, the difference in amount of produced speech (e.g., some children did not say much about the picture, some used only single word utterances, and others produced a richer narrative) made it difficult to compare results across participants from this speech sample. However, outcome measures from connected speech samples have been found to be complimentary to single words (Glaspey et al., 2022).

6.4 CAS AND COEXISTING FUNCTIONAL DIFFICULTIES

In the exploratory study (study IV) on parent-reported functional difficulties in children with CAS we broadened our perspective to developmental domains other than speech (i.e., mental/cognitive functions (such as attention, language, memory), sensory functions, voice functions, motor functions and interpersonal relations). Our findings showed that the children ranged from having a few to many coexisting functional difficulties over the different targeted domains. This variability is in accordance with previous literature reporting on children with CAS as a heterogeneous group (Tükel et al., Iuzzini-Seigel, 2019). Further, we found that frequent functional difficulties were reported in about half of the participants, comparable to coexisting difficulties observed for language, attention, sensory functions, and fine/gross motor skills in about 50% of the children in several studies (Lewis, 2004, Tükel et al, 2015, Teverovsky, 2009, Iuzzini-Siegel, 2019).

From our results on the hierarchical cluster analysis, we identified four subgroups of children with different functional profiles: few coexisting difficulties (1A), coexisting difficulties mainly in voice and rhythm domains (1B), coexisting difficulties in mental/cognitive functions (e.g., attention) (2A) and coexisting difficulties in diverse functions, including motor functions (2B). These findings would be interesting to consider in the clinical setting. Although CAS is defined by its speech characteristics, other difficulties could impact the motivation and participation in treatment. For example, a child with a profile with subtle difficulties in many functions may need a lot of support getting started, maintaining attention and sit still, possibly affecting the planning of the treatment session regarding duration, what activity to choose or frequency of feedback (Maas, Butalla, & Farinella, 2012). However, the intervention method chosen would certainly be the same as for a child with few coexisting difficulties, i.e., based on Principles of Motor Learning (Maas et al., 2008).

Interestingly, while comparing the two identified functional profiles with several coexisting difficulties (i.e., 2A and 2B), it was found that children with several motor difficulties (2B) also had lower (worse) scores on assessed speech measures. This might indicate that this subgroup had more pronounced procedural learning deficits (i.e., difficulties with the ability to implicitly learn and program a variety of cognitive-linguistic and motor skills) (Sanjeevan & Mainela-Arnold, 2017), in line with findings from a study on procedural learning in children with CAS (Iuzzini-Seigel, 2020).

The speech profile of the child, parallel the functional profile, would guide us to optimized decisions on treatment and further interventions. However, our finding that the item *difficult to be understood by others* was the most prominent parent-reported item across children (82%)

makes us aware of the importance of accompanied knowledge on social interactions, psychosocial difficulties, and everyday activities in children with CAS. This type of information is essential for us to support the child and the family in the best way. For example, in a systematic review it was found that failure to accurately assess children's speech was linked to long-term negative impacts on children socially and academically (McCormack, McLeod, McAllister, & Harrison, 2009).

In the parent responses to questions regarding developmental milestones and heredity it was reported that 58% of the children was late talkers and 67% having a family member with speech-language disorder or known literacy disorder. These proportions are in line with phenotype data presented in a study by Laffin et al. (2012), where several children had delayed onset of speech-language (91%) and 70% of the children had a family member with verbal trait disorder (SSD, DLD, reading disorder, learning disorder). Also, background data presented in a study on orofacial dysfunction in a group of children with SSD (including children with CAS) showed the prevalence of family history of speech, language, and literacy disorders to be 67% (Mogren, 2021).

6.5 METHODOLOGICAL CONSIDERATIONS

Classification systems may impact differential diagnosis (Terband, Maassen, et al., 2019; Waring & Knight, 2013). In this thesis project, children in the clinical study group were classified as having CAS, mildCAS or nonCAS-SSD. The children with mildCAS evidenced characteristics associated with CAS as well as linguistic or phonetic deficits. These children all had five or more CAS speech characteristics, but the frequency of occurrence was mostly low. If the categories from the SCDS had been used and a higher cutoff had been set for the frequency of occurrence (in this study one occurrence for a feature was enough for it to be counted as present), it might have been more adequate to use the SMD label for these children.

In the preliminary estimation of validity (study II), the results from DYMTA were compared to a clinical diagnosis of CAS and nonCAS-SSD. Although expert judgement of perceptual assessment is still the most commonly used procedure in diagnosing CAS (Murray et al., 2021; Murray et al., 2015), a combination of perceptual and more objective methods such as acoustic and kinematic assessments has been proposed to best demonstrate the underlying deficits of motor planning (Nijland et al., 2015; Terband, Namasivayam, et al., 2019). Accordingly, future work on DYMTA will seek to accompany the clinical diagnosis based on a perceptual assessment with objective measures.

Linguistic complexity measures such as phonological neighborhood density, word complexity measure and word frequency were not judged in choosing the words in DYMTA and may constitute a limitation. Words were chosen from a coarticulatory perspective with movement patterns that varied with respect to motoric complexity of syllable shape and phonetic complexity. Neighborhood density refers to the number of words that differ from the target word by a single phoneme, i.e., similar-sounding words that are activated in memory during perception and production (Vitevitch, 2002). There are two contrasting hypotheses about if words with similar forms compete with each other or facilitate speech production, but most commonly it has been suggested that words with many phonological neighborhoods would facilitate easier access to the semantic representations (Vitevitch, 2002; Vitevitch & Luce, 2014). In preparation of the administration of DYMTA the child is always informed about the focus on repetition of words. Consequently, as there are no pictures and all items are assessed through repetition in the test, the words were not checked for the degree of neighborhood density. Word Complexity Measure (WCM) was developed for English by Stoel-Gammon in 2010 (Stoel-Gammon, 2010) , but a Swedish adaption (WCM-SE) was presented first in 2018 (Marklund, Marklund, Schwarz, & Lacerda, 2018) and therefore not present at the time of the development of DYMTA. The WCM-SE is a phonological complexity measure giving points for different complexity parameters adding up to a total score per utterance or word. As the score is a measure on phonology, articulatory demands may not be addressed (Marklund et al., 2018). However, a modified version of WCM for motoric complexity has been described for English and used in a recent study (Namasivayam, Huynh, Bali, et al., 2021). Word frequency data were not available in Swedish for children at the time for the construction of DYMTA, although commonly used and functional words for Swedish children were chosen.

The rationale for the fourth study was built on clinical knowledge and previous literature on coexisting functional difficulties in CAS, and the study by Teverovsky et al. (2009) especially informed our choices preparing the study (in 2013). In their study a parental questionnaire was set based on domains from the ICF (WHO, 2013). The authors described the ICF to be a useful tool for characterizing the functional difficulties cooccurring with CAS. In the manual (WHO, 2013) the ICF framework was explained as a multipurpose classification system to be used for describing and organizing health- and health related outcomes, providing a common language and systematic coding. Initially, already available parental questionnaires were sought after for use in our study. An early version of the Five-To-Fifteen (FTF) questionnaire (Kadesjö et al., 2004) was considered. Although the FTF examines abilities and behavioral patterns in children and youth of interest for our study, the FTF was found to be too extensive for our project with its 181 statements. Consequently, a parental questionnaire especially for this project was prepared with 40 items across functions, using the ICF as a frame. Thus, the ICF is not used in its full potential in this study, but as a framework to organize the investigated functions.

7 CONCLUSIONS

The results from the included studies in this thesis project have possible contributed to broadened knowledge on the assessment procedure of children with CAS. The results are in line with the rationale and aims of the project. The main findings and contributions are:

- The first Swedish assessment tool designed to facilitate the identification of children with Childhood Apraxia of Speech, DYMTA, has been evaluated and described

- Overall reliability evidence for DYMTA was found to be strong

- Evidence for the test DYMTA being accurate and validly separate children with CAS from nonCAS-SSD was found

- The task using dynamic assessment was adding information on speech motor characteristics in most participants, especially regarding deviant articulatory transitions and inconsistency, appointing DA to be a valuable complement in the diagnostic procedure of CAS.

- The children with idiopathic CAS were found to be a heterogenous group regarding the relative frequency of coexisting functional difficulties

- Different functional profiles were identified in children with CAS according to parental responses on a questionnaire of coexisting functional difficulties in their child. Four meaningful subgroups were revealed; few functional difficulties, few difficulties but some in the voice functions, more functional difficulties related to mental functions such as attention and planning activities, and a fourth group with diverse difficulties but specifically more in motor functions

- Children with typical speech development performed well on measures for speech motor performance in DYMTA already from the age of three, with some results showing a developmental tendency. The results are adding to the body of cross-lingual literature on speech development in children

8 CLINICAL IMPLICATIONS AND FUTURE PERSPECTIVE

DYMTA has been introduced to the Swedish SLPs and has been available to the SLP community for some years now (Rex et al., 2016). Implementing new tests or concepts in the clinical setting or education programs at universities is crucial to facilitate uptake and help develop SLPs skills in testing (Daub et al., 2021). The administration of DYMTA and results from studies in this thesis have been presented and taught in several courses during the last years, both to SLP colleagues and students in Sweden, Denmark, and Norway. Support in implementation was then in focus using numerous video-examples and hands-on information. The multidimensional scoring in DYMTA is valuable, but more dimensions make it more difficult (Strand et al., 2013), the reliability evidence of tests is commonly higher if scoring is dual. This adds to the responsibility of the test developer, teachers, and clinicians who use the instrument to get more training, such as participating in courses, carrying out training sessions together with colleagues, read available tutorials and attend to webinars.

As DYMTA was developed to be a part of a larger test battery for children with SSDs it would be important for the clinician to have knowledge on when and for what intended reason the test should be selected during the assessment procedure. Recent findings from a survey study on Swedish clinical practice of children with SSD showed that Swedish practice is variable, with some components assessed more frequent than others (Wikse Barrow, Körner, & Strömbergsson, 2021). The frequency and manner of seven assessment components were addressed in the survey, e.g., speech output, consistency, functional communication, intelligibility, speech perception, oral motor function and phonological awareness, and regarding the assessment of speech output and consistency some responders mentioned the use of DYMTA. The lack of national guidelines in Sweden for assessment of SSDs was recognized, highlighting the need for discussions on how to integrate research findings with best clinical practice, to form consensus on general guidelines (Wikse Barrow et al., 2021).

The validation evidence for DYMTA in this thesis project consists mainly of the discriminative ability of the test. A project focusing on content validation would be of interest in a possible future revision of DYMTA, since this would inform on what items (i.e., words) that best elicited speech productions contributing to the overall performance of the test or subscores. Another suggestion would be to use information on items to compile a screening version of DYMTA.

The invested time in a thorough assessment and diagnostic procedure is the platform for choosing the optimal treatment method. For the child with a motor based speech disorder, such as SMD or CAS, a treatment method based on principles of motor learning (PML) is preferrable chosen (Morgan, Murray, & Liegeois, 2018). PML has received increasing attention for intervention of motor speech disorders facilitating motor skill learning (Maas et al., 2008). The PML comprises prepractice to motivate the learner and several practice conditions (i.e., practice amount, practice distribution, practice variability, practice schedule, internal/external feedback, and practice complexity). Examples of motor-based therapies based on PML are Nuffield Dyspraxia Programme (Williams & Stephens, 2004), Prompts for Restructuring Oral Muscular Phonetic Targets (PROMPT) (Dale & Hayden, 2013), Dynamic Tactile and Temporal Cuing (DTTC) (Strand, 2019) and the Rapid Syllable Transitions Treatment (ReST) (Ballard, Robin, McCabe, & McDonald, 2010). In Sweden the Nuffield Dyspraxia Programme, DTTC and PROMPT have been introduced, but we still need to work for better conditions for them to be implemented and delivered in the clinical setting (e.g., resources and education).

Children with CAS may have different profiles of speech characteristics, but also have different profiles of coexisting functional difficulties as demonstrated in this thesis and found to be in line with an increasing body of evidence for coexistence of deficits (Gillberg, 2010; Mogren, 2021; Newmeyer, Grether, & ... 2007; Teverovsky et al., 2009). Cooccurring speech, language, motor, sensory, and behavioral difficulties leave a responsibility to the SLPs not only to have knowledge about the possible risk of difficulties in each of those functions but also to actually inquire about them, possibly using parental questionnaires with items representing different functions. The increased risk for learning difficulties, social-emotional and behavioral problems in children with SSD/CAS as described in many studies (Lewis et al., 2004; McCormack et al., 2009; Miller et al., 2019; Teverovsky et al., 2009) show the urge to work in multi-professional teams to ensure that intervention involves the total need of the child.

9 ACKNOWLEDGEMENTS

This project would not have been possible without the support from so many people. I sincerely want to send my gratitude to all of you.

First, I want to thank all the participating *children and their parents*. Thank you so much, without you there would not have been any project at all.

Anita McAllister, main supervisor who has believed in me throughout the whole PhD program. Thank you for planning this project with me and being so full of enthusiasm for research which has kept me going.

Kristina Hansson, co-supervisor who has been my support in 'the south'. Thank you for all important feedback on the manuscripts throughout the project, and the cooperation in supervision of student projects.

Edythe Strand, co-supervisor with extensive knowledge in the field. You have been an inspiration and a model, and so generously sharing your knowledge both in physical meetings in Sweden and via transatlantic Zoom-meetings.

Barbro Bruce, my mentor for good conversations and encouragement in moments of disbelief.

Anders Sand, co-author with excellent knowledge in psychometrics and statistics. You are a fantastic pedagogue and really have an eye for presentation of data. Thank you for your great engagement in this project.

Professor *Anette Lohmander* and *colleagues* at the Division of Speech-Language Pathology at KI who took me on board, and also included me in retreats as well as the trip to Canada and US in 2018.

Åsa Mogren and *Ann Malmenholt*, fellow PhD students for your friendship and fruitful discussions during this education. It would not have been the same to travel the 10 hours back and forth to Stockholm for courses and meetings, not also having a fika or dinner planned with you. *Ann Malmenholt*, thank you for the time-consuming consensus judgements.

All former and present *PhD students* at KI, thank you all for your company in discussions, seminars, and other events during the years. Thank you, *Jill Nyberg*, for being a great colleague and friend.

Ritwa Liljeblad, my former employer at the SLP clinic, Skåne University Hospital, who believed in me and enabled me to combine clinical work with the PhD program. *Caroline Sundberg,* my present employer who has been a good support during the last period of the PhD education.

Anna Petersson and *Agneta Wittlock*, at CLINTEC, for all your help with small and big administrative questions.

Ulf Thylander, Kunskapsutveckling i Lund AB, thank you for pleasant cooperation in the process of publishing DYMTA and for combining some of the meetings with a nice lunch in

Lund. I also want to thank *Anders Jeppson* for taking on the assignment to make the illustrations for DYMTA scoring form and manual.

Agneta Ivanov, SLP colleague and friend who loves working with children with CAS in the same way as I do. Thank you for your help with interrater judgments.

Mervi Harjuniemi, Lejla Malkić, Louise Lundvall, Linda Smetana, Johanna Gustafsson, Sanna Johansson, SLP students (at the time). Thank you so much for your involvement in collecting typical data in an early stage of the project. Thank you *Åsa Engström*, *Lisa Hjalmarsson* and *Ellinor From* for your engagement in the children with CAS, choosing this group of children as a subject for your degree projects.

Kicki Klintö, colleague and friend for valuable formal and informal meetings during the years, talking about research and life in general.

Former and present colleagues at Skåne University Hospital, Speech Language Department (Lund/ Malmö) for our many discussions about interesting cases of children with speech motor difficulties. I especially want to thank *Viktoria Olander* and *Maria Sporre* for all engaging time we spent together discussing patients with different speech and oral motor difficulties.

Lucina Schalén and *Malin Josefsson*, ENT specialists in otorhinolaryngology, Skåne University hospital, Lund/Malmö. Thank you for all the years with teamwork regarding children with orofacial and/or speech motor difficulties.

All the *friends and family,* who have been there during the years of the doctoral education, letting me engage in other things than work and research, refilling my battery. I am looking forward to seeing all of you more often again! I especially want to thank *Pia*, *Catharina*, *Charlotte & Stefan*, *Ingela & Calle*, *Ann I.,* and *Anders & Anna* for giving me energy to continue this project.

Thank you*, mom and dad*, for always being there, believing in me.

Titti, my dear sister, always so caring, and generous with love. And a special thanks to my sister-daughter *Moa*, who made the great illustration for the cover of this thesis.

Most importantly I want to thank my children *Alexander* and *Johannes* and my husband *Thomas*. I could not have done this project without the support from my closest family. *Alexander* and *Johannes*, you are the most generous, curious, warm, and smart children I could ever wish for. I am glad you want to share your lives with me. A special thanks to you *Alexander* for always being so helpful and professional with computer related matters, such as formatting and figures, when I need help.

Thomas, my dear husband who have put up with me during all these years of PhD studies. Thank you for supporting me in this project! Sharing my life with you makes me feel safe and extremely happy! I love you so much!

This research was financially supported by Sunnerdahls Handikappfond, Region Skåne FoU funds, Aina Börjessons foundation for Speech Language Pathology research and Stiftelsen Promobilia.

10 REFERENCES

Alcock, K. (2006). The development of oral motor control and language. *The journal of down syndrome, 11*, 1-8.

Allison, K. M., Cordella, C., Iuzzini-Seigel, J., & Green, J. R. (2020). Differential Diagnosis of Apraxia of Speech in Children and Adults: A Scoping Review. *J Speech Lang Hear Res*, 1-43. doi:10.1044/2020_JSLHR-20-00061

ASHA. (2007a). Childhood Apraxia of Speech [Technical Report]. *Rockville (MD): American Speech-Language-Hearing Association*, Available from: http://www.asha.org/policy/TR2007-00278/.

ASHA. (2007b). Childhood Apraxia of Speech: Nomenclature, definition, roles and responsibilietes, and a call for action [Position statement]. *Rockville. MD: American Speech-Language-Hearing Association*, Available from: http://www.asha.org/policy/PS2007-00277/.

Bain, B., & Olswang, L. B. (1995). Examining readiness for learning two-word utterances by children with specific expressive language impairment: Dynamic assessment validation. *American Journal of Speech-Language Pathology, 4* 81–92.

Bakke, M., Bergendal, B., McAllister, A., Sjögren, L., & Åsten, P. (2007). Development and evaluation of a comprehensive screening for orofacial dysfunction. *Swedish Dental Journal, 31*(2), 75-84.

Ballard, K. J., Robin, D. A., McCabe, P., & McDonald, J. (2010). A treatment for dysprosody in childhood apraxia of speech. *J Speech Lang Hear Res, 53*(5), 1227-1245

Bishop, D. (2009). TROG-2 Test for Reception of Grammar - Second Edition. *Pearson*.

Bishop, D., Snowling, M. J., Thompson, P. A., & Greenhalgh, T. (2016). CATALISE: a multinational and multidisciplinary Delphi consensus study. 1. Identifying language impairments in children. doi:10.7287/peerj.preprints.1986v1

Blumenthal, C., & Lundeborg Hammarström, I. (2014). LINUS – Linköpingsundersökningen. – [in Swedish]. A phonological test for children from 3 years of age. *Retrieved from http://liu. divaportal*.

Bruce, G. (2007). Components of a prosodic typology of Swedish intonation. *Tones and Tunes, 1*(T Riad and C Gussenhoven (eds), Berlin: Mouton de Gruyter), 113-146.

Caruso, A. J., & Strand, E. A. (1999). Clinical Management of Motor Speech Disorders in Children. *Thieme Publishing Group*.

Centanni, T. M., Sanmann, J. N., Green, J. R., Iuzzini-Seigel, J., Bartlett, C., Sanger, W. G., & Hogan, T. P. (2015). The role of candidate-gene CNTNAP2 in childhood apraxia of speech and specific language impairment. *Am J Med Genet B Neuropsychiatr Genet, 168*(7), 536-543. doi:10.1002/ajmg.b.32325

Chenausky, K. V., Brignell, A., Morgan, A., Gagné, D., Norton, A., Tager-Flusberg, H., . . . Green, J. R. (2020). Factor analysis of signs of childhood apraxia of speech. *Journal of communication disorders, 87*. doi:10.1016/j.jcomdis.2020.106033

Clausen, M. C., & Fox-Boyer, A. V. (2017). The phonological development of Danish-speaking children: A normative cross-sectional study. *Clinical Linguistics & Phonetics, 31*(6), 440–458. doi: https://doi.org/10.1080/02699206.2017.1308014

Cleland, J., Scobbie, J. M., Heyde, C., Roxburgh, Z., & Wrench, A. A. (2017). Covert contrast and covert errors in persistent velar fronting. *Clinical Linguistics & Phonetics, 31*(1), 35-55. doi:https://doi.org/10.1080/02699206.2016.1209788

Dale, P. S., & Hayden, D. A. (2013). Treating speech subsystems in Childhood Apraxia of Speech with tactual input: the PROMPT approach. *American Journal of Speech-Language Pathology, 22*(4), 644-661. doi:10.1044/1058-0360(2013/12-0055)

Darley, F. L., Aronson, A. E., & Brown, J. R. (1975). Motor speech disorders. *Philadelphia: W.B. Sounders Co.*

Daub, O., Cunningham, B. J., Bagatto, M. P., Johnson, A. M., Kwok, E. Y., Smyth, R. E., & Cardy, J. O. (2021). Adopting a Conceptual Validity Framework for Testing in Speech-Language Pathology. *Am J Speech Lang Pathol, 30*(4), 1894-1908. doi:10.1044/2021_AJSLP-20-00032

Dodd, B. (2014). Differential Diagnosis of Pediatric Speech Sound Disorder. *Current Developmental Disorders Reports, 1*(3), 189-196. doi:10.1007/s40474-014-0017-3

Dollaghan, C. A. (2007). The handbook for evidence-based practice in communication disorders. *Paul H. Brookes Publishing Co., Inc.*

Duffy, J. R. (2013). Motor Speech Disorders. Substrates, Differential Diagnosis and Management (Third edition). St. Louis, MO: Elsevier Mosby.

Eadie, P., Morgan, A., Ukoumunne, O. C., Ttofari Eecen, K., Wake, M., & Reilly, S. (2015). Speech sound disorder at 4 years: prevalence, comorbidities, and predictors in a community cohort of children. *Dev Med Child Neurol, 57*(6), 578-584. doi:10.1111/dmcn.12635

Fiori, S., Pannek, K., Podda, I., Cipriani, P., Lorenzoni, V., Franchi, B., . . . Chilosi, A. (2021). Neural Changes Induced by a Speech Motor Treatment in Childhood Apraxia of Speech: A Case Series. *J Child Neurol, 36*(11), 958-967. doi:10.1177/08830738211015800

Forrest, K. (2003). Diagnostic criteria of developmental apraxia of speech used by clinical speech-language pathologists. *American Journal of Speech-Language Pathology, 12*(3), 376-380.

Gibbon, F. E. (1999). Undifferentiated lingual gestures in children with articulation/phonological disorders. . *Journal of Speech, Language and Hearing Research, 42*(2), 382-397. doi:10.1044/jslhr.4202.382

Gillberg, C. (2010). The ESSENCE in child psychiatry: Early Symptomatic Syndromes Eliciting Neurodevelopmental Clinical Examinations. *Research in Developmental Disabilities, 31*(6), 1543-1551. doi:10.1016/j.ridd.2010.06.002

Glaspey, A. M. (2019). *The Glaspey Dynamic Assessment of Phonology.*, Academic Therapy Publications.

Glaspey, A. M., Wilson, J. J., Reeder, J. D., Tseng, W. C., & MacLeod, A. A. N. (2022). Moving Beyond Single Word Acquisition of Speech Sounds to Connected Speech Development With Dynamic Assessment. *J Speech Lang Hear Res, 65*(2), 508-524. doi:10.1044/2021_JSLHR-21-00188

Green, J. R., Moore, C. A., Higashikawa, M., & Steeve, R. W. (2000). The physiologic development of speech motor control: lip and jaw coordination. *Journal of Speech, Language and Hearing Research, 43*(1), 239-255.

Grigos, M. I. (2009). Changes in Articulator Movement Variability During Phonemic Development: A Longitudinal Study. *Journal of Speech and Hearing Research, 52*, 164-177. doi:10.1044/1092-4388(2008 /07-0220)

Grigos, M. I., Moss, A., & Lu, Y. (2015). Oral Articulatory Control in Childhood Apraxia of Speech. *Journal of Speech, Language, and Hearing Research*.

Gustafsson, J., & Johnsson, S. (2015). Hur presterar 3- och 6-åriga barn med typisk tal- och språkutveckling på DYMTA (Dynamisk Motorisk Talbedömning)? [In Swedish]. *Master's thesis in Speech Language Pathology. CLINTEC, Karolinska Institutet*.

Hansson, K., & Nettelbladt, U. (2010). *GRAMBA - Grammatiktest för barn [In Swedish]* (Vol. ISBN: 9789144069296). Lund: Studentlitteratur.

Harjuniemi, M., & Malkić, L. (2014). 3- och 5-åringars resultat på Dynamisk Motorisk Talbedömning - DYMTA: En åldersjämförelse. [In Swedish]. *Master thesis in Speech and Language Pathology. Institutionen för kliniska vetenskaper, Lund University*.

Hasson, N., & Joffe, V. (2007). The case for Dynamic Assessment in speech and language therapy. *Child Language Teaching and Therapy, 23*(1), 9-25. doi:10.1177/0265659007072142

Hayden, A., & Square, P. (1999). VMPAC, Verbal Motor Production Assessment for Children. *San Antonio, TX: The Psychological Corporation*.

Head, H. (1915). Hughling Jackson on aphasia and kindred affections of speech. *Brain, 38*, 1-190.

Hellquist, B. (2011). Nya SIT: språkligt impressivt test för barn [In Swedish]. *Bokus AB*.

Hickok, G. (2012). The cortical organization of speech processing: feedback control and predictive coding the context of a dual-stream model. *J Commun Disord, 45*(6), 393-402. doi:10.1016/j.jcomdis.2012.06.004

Hickok, G., & Poeppel, D. (2007). The cortical organization of speech processing. *Nature Reviews Neuroscience, 8*(5), 393-402. doi:10.1038/nrn2113

Highman, C., Hennessey, N. W., Leitao, S., & Piek, J. P. (2013). Early development in infants at risk of childhood apraxia of speech: a longitudinal investigation. *Dev Neuropsychol, 38*(3), 197-210. doi:10.1080/87565641.2013.774405

Hill, E. L. (2001). Non-specific nature of specific language impairment: a review of the literature with regard to concomitant motor impairments. *Int J Lang Commun Disord, 36*(2), 149-171. doi:10.1080/13682820010019874

Hjalmarsson, L., & From, E. (2018). Talmotoriska markörers relevans i bedömning av taldyspraxi. En studie om hur internationella kännetecken för taldyspraxi är applicerbara på svenskspråkiga barn. [In Swedish]. *Master's thesis in Speech and Language Pathology. Institutionen för kliniska vetenskaper, Lund University.*

Holm, A., Crosbie, S., & Dodd, B. (2007). Differentiating normal variability from inconsistency in children's speech: normative data. *International Journal of Language & Communication Disorders, 42*(4), 467-486. doi:10.1080/13682820600988967

IALP. (2021). What's in a name. from https://ialpasoc.info/third_slider/ialp-child-speech-committee-online-panel-series-2021-2022/

Iuzzini-Seigel, J. (2019). Motor Performance in Children With Childhood Apraxia of Speech and Speech Sound Disorders. *J Speech Lang Hear Res, 62*(9), 3220-3233. doi:10.1044/2019_JSLHR-S-18-0380

Iuzzini-Seigel, J. (2021). Procedural Learning, Grammar, and Motor Skills in Children With Childhood Apraxia of Speech, Speech Sound Disorder, and Typically Developing Speech. *J Speech Lang Hear Res, 64*(4), 1081-1103. doi:10.1044/2020_JSLHR-20-00581

Iuzzini-Seigel, J., Hogan, T. P., & Green, J. R. (2017). Speech Inconsistency in Children With Childhood Apraxia of Speech, Language Impairment, and Speech Delay: Depends on the Stimuli. *Journal of Speech Language and Hearing Research, 60*(5), 1194-1210. doi:10.1044/2016_JSLHR-S-15-0184

Iuzzini-Seigel, J., Hogan, T. P., Guarino, A. J., & Green, J. R. (2015). Reliance on auditory feedback in children with childhood apraxia of speech. *J Commun Disord, 54*, 32-42. doi:10.1016/j.jcomdis.2015.01.002

Iuzzini-Seigel, J., Hogan, T. P., Rong, P., & Green, J. R. (2015). Longitudinal development of speech motor control: Motor and linguistic factors. *Journal of Motor Learning and Development, 3*(1), 53-68. doi:10.1123/jmld.2014-0054

Iuzzini-Seigel, J., & Murray, E. (2017). Speech Assessment in Children With Childhood Apraxia of Speech. *Perspectives of the ASHA Special Interest Groups SIG 2, 2 (Part 2)*, 47-60.

Iverson, J. M. (2010). Developing language in a developing body: the relationship between motor development and language development. *J Child Lang, 37*(2), 229-261. doi:10.1017/S0305000909990432

Kadesjö, B., Janols, L. O., Korkman, M., Mickelsson, K., Strand, G., Trillingsgaard, A., & Gillberg, C. (2004). The FTF (Five to Fifteen): the development of a parent questionnaire for the assessment of ADHD and comorbid conditions. *European Child & Adolescent Psychiatry, 13*, 3-13.

Kapantzoglou, M., Restrepo, M. A., & Thompson, M. S. (2012). Dynamic Assessment of Word Learning Skills: Identifying Language Impairment in Bilingual Children. *Lang Speech Hear Serv Sch, 43*(1), 81-96. doi:10.1044/0161-1461(2011/10-0095)

Laffin, J. J., Raca, G., Jackson, C. A., Strand, E. A., Jakielski, K. J., & Shriberg, L. D. (2012). Novel candidate genes and regions for childhood apraxia of speech identified by array comparative genomic hybridization. *Genet Med, 14*(11), 928-936. doi:10.1038/gim.2012.72

Lai, C. S. L., Fisher, S. E., Hurst, J. A., Vargha-Khadem, F., & Monaco, A. P. (2001). A forkhead-domain gene is mutated in a severe speech and language disorder. *Nature, 413*, 519-523.

Lewis, B. A., Freebairn, L. A., Hansen, A. J., Iyengar, S. K., & Taylor, H. G. (2004). School-age follow up of children with childhood apraxia of speech. *Lang Speech Hear Serv Sch, 35*(2), 122-140. doi:https://doi.org/10.1044/0161-1461(2004/014)

Lewis, B. A., O'Donnell, B., Freebarin, L. A., & Taylor, H. G. (2002). Spoken Language and written expression - Interplays of delays. *American Journal of Speech-Language Pathology, 7*(66-73).

Lidz, C. S. (1987). *Dynamic assessment: an interactional approach to evaluating learning potential*. New York: Guilford Press; cop.

Liegeois, F. J., & Morgan, A. T. (2012). Neural bases of childhood speech disorders: lateralization and plasticity for speech functions during development. *Neurosci Biobehav Rev, 36*(1), 439-458. doi:10.1016/j.neubiorev.2011.07.011

Liegeois, F. J., Turner, S. J., Mayes, A., Bonthrone, A. F., Boys, A., Smith, L., . . . Morgan, A. T. (2019). Dorsal language stream anomalies in an inherited speech disorder. *Brain, 142*(4), 966-977. doi:10.1093/brain/awz018

Lohmander, A., Borell, E., Henningsson, G., Havstam, C., Lundeborg, I., & Persson, C. (2005). *SVANTE Svenskt artikulations- och nasalitets-test, Manual [In Swedish].* Lund: Studentlitteratur.

Lohmander, A., Lundeborg, I., & Persson, C. (2017). SVANTE - The Swedish Articulation and Nasality Test - Normative data and a minimum standard set for cross-linguistic comparison. *Clinical Linguistics and Phonetics, 31*(2), 137-154. doi:10.1080/02699206.2016.1205666

Lundeborg Hammarström, I. (2019). LINUS 2.0. – Linköpingsundersökningen [in Swedish]. A phonological test for children from 3 years of age. Manual. *Retrieved from* http://liu*. divaportal*.

Lundvall, L., & Smetana, L. (2014). Dynamisk Motorisk Talbedömning (DYMTA) - en referensstudie [In Swedish]. *Master's thesis in Speech Language Pathology, Uppsala University*.

Maas, E., Butalla, C. E., & Farinella, K. A. (2012). Feedback Frequency in Treatment for Childhood Apraxia of Speech. *American Journal of Speech-Language Pathology, 21*(3). doi:10.1044/1058-0360(2012/11-0119)

Maas, E., Robin, D. A., Austermann Hula, S. N., Freedman, S. E., Wulf, G., Ballard, K. J., & Schmidt, R. A. (2008). Principles of motor learning in treatment of motor speech disorders. *American Journal of Speech-Language Pathology, 17*, 277-298.

Malmenholt, A. (2020). *Exploring childhood apraxia of speech: Speech and language profiles in 5-year-olds with suspected apraxia of speech or cleft palate.* (Unpublished doctoral thesis), Karolinska Institutet, Stockholm.

Malmenholt, A., Lohmander, A., & McAllister, A. (2017). Childhood apraxia of speech: A survey of praxis and typical speech characteristics. *Logoped Phoniatr Vocol, 42*(2), 84-92. doi:10.1080/14015439.2016.1185147

Marcus, G. F., & Fisher, S. E. (2003). FOXP2 in focus: what can genes tell us about speech and language? *Trends in Cognitive Sciences, 7*(6), 257-262. doi:10.1016/s1364-6613(03)00104-9

Marklund, U., Marklund, E., Schwarz, I. C., & Lacerda, F. (2018). Introducing WCM-SE: The word complexity measure phonetically justified and adapted to Swedish. *Clin Linguist Phon, 32*(11), 1042-1053. doi:10.1080/02699206.2018.1493620

McAllister, & Lundeborg Hammarström, I. (2014). Oral sensorimotor functions in typically developing children 3 to 8 years old, assessed by the Nordic Orofacial Test, NOT-S. *Journal of Medical Speech - Language Pathology, 21*(1), 51-59.

McCabe, P. J., Murray, E., & Thomas, D. (2020). 2020 Evidence Summary: Childhood Apraxia of Speech. *Research Gate; Technical report.*

McCauley, R. J., & Strand, E. A. (2008). A Reveiw of Standardized Tests of Nonverbal Oral and Speech Motor Performance in Children. *American Journal of Speech-Language Pathology, 17*, 81-91.

McCormack, J., McLeod, S., McAllister, L., & Harrison, L. J. (2009). A systematic review of the association between childhood speech impairment and participation across the lifespan. *International Journal of Speech-Language Pathology, 11*(2), 155-170. doi:10.1080/17549500802676859

McLeod, S. (2009). An holistic view of a child with unintelligible speech: Insights from the ICF and ICF-CY. *Advances in Speech Language Pathology, 8*(3), 293-315. doi:10.1080/14417040600824944

McLeod, S., & Hewett, S. R. (2008). Variability in the production of words containing consonant clusters by typical 2- and 3-year-old children. *Folia Phoniatr Logop, 60*(4), 163-172. doi:10.1159/000127835

McLeod, S., & McCormack, J. (2007). Application of the ICF and ICF-Children and Youth in Children with Speech Impairment. *Seminars in Speech and Language, 28*(4), 254-264. doi:10.1055/s-2007-986522

McLeod, S., & Verdon, S. (2014). A review of 30 speech assessments in 19 languages other than English. *American Journal of Speech-Language Pathology, 23*(4), 708-723. doi:10.1044/2014_AJSLP-13-0066

Mei, C., Fedorenko, E., Amor, D. J., Boys, A., Hoeflin, C., Carew, P., . . . Morgan, A. T. (2018). Deep phenotyping of speech and language skills in individuals with 16p11.2 deletion. *Eur J Hum Genet, 26*(5), 676-686. doi:10.1038/s41431-018-0102-x

Miller, G. J., Lewis, B., Benchek, P., Freebairn, L., Tag, J., Budge, K., . . . Stein, C. (2019). Reading Outcomes for Individuals With Histories of Suspected Childhood Apraxia of Speech. *Am J Speech Lang Pathol, 28*(4), 1432-1447. doi:10.1044/2019_AJSLP-18-0132

Miniscalco, C., Nygren, G., Hagberg, B., Kadsjö, B., & Gillberg, C. (2006). Neuropsychiatric and neurodevelopmental outcome of children at age 6 and 7 years who screened for language problems at 30 months. *Journal of Developmental Medicine and Child Neurology, 48*, 361-366.

Mogren, Å. (2021). *Orofacial Function in Children with Speech Sound Disorders.* (Unpublished doctoral thesis), Karolinska Institutet, Stockholm.

Morgan, A. T., Murray, E., & Liegeois, F. J. (2018). Interventions for childhood apraxia of speech. *Cochrane Database Syst Rev, 5*, CD006278. doi:10.1002/14651858.CD006278.pub3

Morgan, A. T., Su, M., Reilly, S., Conti-Ramsden, G., Connelly, A., & Liégeois, F. J. (2018). A Brain Marker for Developmental Speech Disorders. *The Journal of Pediatrics, 198*, 234-239.e231. doi:10.1016/j.jpeds.2018.02.043

Morgan, A. T., van Haaften, L., van Hulst, K., Edley, C., Mei, C., Tan, T. Y., . . . Koolen, D. A. (2018). Early speech development in Koolen de Vries syndrome limited by oral praxis and hypotonia. *European Journal of Human Genetics, 26*(1), 75-84. doi:10.1038/s41431-017-0035-9

Morgan, A. T., & Webster, R. (2018). Aetiology of childhood apraxia of speech: A clinical practice update for paediatricians. *J Paediatr Child Health, 54*(10), 1090-1095. doi:10.1111/jpc.14150

Morley, M., Court, D., Miller, H., & Garside, R. (1955). Delayed speech and developmental aphasia. *British Medical Journal*(Aug. 20), 463-467.

Murray, E., Iuzzini-Seigel, J., Maas, E., Terband, H., & Ballard, K. J. (2021). Differential Diagnosis of Childhood Apraxia of Speech Compared to Other Speech Sound Disorders: A Systematic Review. *Am J Speech Lang Pathol, 30*(1), 279-300. doi:10.1044/2020_AJSLP-20-00063

Murray, E., McCabe, P., Heard, R., & Ballard, K. J. (2015). Differential diagnosis of children with suspected childhood apraxia of speech. *J Speech Lang Hear Res, 58*(1), 43-60. doi:10.1044/2014_JSLHR-S-12-0358

Murray, E., Thomas, D., & McKechnie, J. (2019). Comorbid morphological disorder apparent in some children aged 4-5 years with childhood apraxia of speech: findings from standardised testing. *Clin Linguist Phon, 33*(1-2), 42-59. doi:10.1080/02699206.2018.1513565

Namasivayam, A. K., Huynh, A., Bali, R., Granata, F., Law, V., Rampersaud, D., . . . Hayden, D. (2021). Development and Validation of a Probe Word List to Assess Speech Motor Skills in Children. *Am J Speech Lang Pathol, 30*(2), 622-648. doi:10.1044/2020_AJSLP-20-00139

Namasivayam, A. K., Huynh, A., Granata, F., Law, V., & van Lieshout, P. (2021). PROMPT intervention for children with severe speech motor delay: a randomized control trial. *Pediatr Res*. doi:10.1038/s41390-020-0924-4

Newmeyer, A. J., Aylward, C., Akers, R., Ishikawa, K., Grether, S., deGrauw, T., . . . White, J. (2009). Results of the Sensory Profile in children with suspected childhood apraxia of speech. *Phys Occup Ther Pediatr, 29*(2), 203-218. doi:10.1080/01942630902805202

Newmeyer, A. J., Grether, S., & ... (2007). Fine motor function and oral-motor imitation skills in preschool-age children with speech-sound disorders. *Clinical Pediatrics, 46*(604-611).

Nijland, L., Terband, H., & Maassen, B. (2015). Cognitive Functions in Childhood Apraxia of Speech. *J Speech Lang Hear Res, 58*(3), 550-565. doi:10.1044/2015_JSLHR-S-14-0084

Peña, E. D., Gillam, R. B., Malek, M., Ruiz-Felter, R., Resendiz, M., Fiestas, C., & Sabel, T. (2006). Dynamic Assessment of School-Age Children's Narrative Ability: An Experimental Investigation of Classification Accuracy. *Journal of Speech, Language and Hearing Research, 49*(5), 1037-1057.

Peter, B., Lancaster, H., Vose, C., Middleton, K., & Stoel-Gammon, C. (2017). Sequential processing deficit as a shared persisting biomarker in dyslexia and childhood apraxia of speech. *Clin Linguist Phon*, 1-31. doi:10.1080/02699206.2017.1375560

Petersen, D. B., Chanthongthip, H., Ukrainetz, T. A., Spencer, T. D., & Steeve, R. W. (2017). Dynamic Assessment of Narratives: Efficient, Accurate Identification of Language Impairment in Bilingual Students. *J Speech Lang Hear Res, 60*(4), 983-998. doi:10.1044/2016_JSLHR-L-15-0426

Pollock, K. E., & Berni, M. C. (2003). Incidence of non-rhotic vowel errors in children: data from the Memphis Vowel Project. *Clinical Linguistics & Phonetics, 17*(4-5), 393-401. doi:10.1080/0269920031000079949

R_Core_Team. (2017). R: A language and environment for statistical computing. R Foundation for Statistical Computing. http://www.R-project.org/.

Rex, S., McAllister, A., & Hansson, K. (2016). *Dynamisk Motorisk Talbedömning (DYMTA) Manual [In Swedish]* (Vol. ISBN: 978-91-976067-2-1). Lund: Kunskapsutveckling i Lund AB.

Rosenbek, & Wertz. (1972). A reveiw of fifty cases of developmental apraxia of speech. 23-33.

Roxburgh, Z., Cleland, J., Scobbie, J. M., & Wood, S. E. (2021). Quantifying changes in ultrasound tongue-shape pre- and post-intervention in speakers with submucous cleft palate: an illustrative case study. *Clin Linguist Phon*, 1-19. doi:10.1080/02699206.2021.1973566

Rusiewicz, H. L., Maize, K., & Ptakowski, T. (2017). Parental experiences and perceptions related to childhood apraxia of speech: Focus on functional implications. *Int J Speech Lang Pathol*, 1-12. doi:10.1080/17549507.2017.1359333

Samuelsson, C., & Nettelbladt, U. (2004). Prosodic problems in Swedish children with language impairment: towards a classification of subgroups. *Int J Lang Commun Disord, 39*(3), 325-344. doi:10.1080/13682820410001654874

Samuelsson, C., Reuterskiold, C., Nettelbladt, U., & Sahlen, B. (2011). Production and perception of metrical patterns in Swedish children with language impairment. *Logopedics Phoniatrics Vocology, 36*(1), 1-11. doi:10.3109/14015439.2010.506195

Sanjeevan, T., & Mainela-Arnold, E. (2017). Procedural Motor Learning in Children With Specific Language Impairment. *J Speech Lang Hear Res, 60*(11), 3259-3269. doi:10.1044/2017_JSLHR-L-16-0457

Schötz, S., Frid, J., & Löfqvist, A. (2013). Development of speech motor control: lip movement variability. *J Acoust Soc Am, 133*(6), 4210-4217. doi:10.1121/1.4802649

Shriberg, L. D., Aram, D. M., & Kwiatkotski, J. (1997). Developmental apraxia of speech: III. A subtype marked by inappropriate stress. *Journal of Speech, Language and Hearing Research, 40*, 313-337.

Shriberg, L. D., Austin, D., Lewis, B. A., McSweeny, J. L., & Wilson, D. L. (1997). The percentage of consonant correct (PCC) metric: extensions and reliability data. . *Journal of Speech, Language and Hearing Research, 40*(4), 708-722.

Shriberg, L. D., Campbell, T. F., Mabie, H. L., & McGlothlin, J. H. (2019). Initial studies of the phenotype and persistence of speech motor delay (SMD). *Clin Linguist Phon, 33*(8), 737-756. doi:10.1080/02699206.2019.1595733

Shriberg, L. D., Fourakis, M., Hall, S. D., Karlsson, H. B., Lohmeier, H. L., McSweeny, J. L., . . . Wilson, D. L. (2010). Extensions to the Speech Disorders Classification System (SDCS). *Clin Linguist Phon, 24*(10), 795-824. doi:10.3109/02699206.2010.503006

Shriberg, L. D., Kwiatkowski, J., & Mabie, H. L. (2019). Estimates of the prevalence of motor speech disorders in children with idiopathic speech delay. *Clin Linguist Phon, 33*(8), 679-706. doi:10.1080/02699206.2019.1595731

Shriberg, L. D., Lohmeier, H. L., Strand, E. A., & Jakielski, K. J. (2012). Encoding, memory, and transcoding deficits in Childhood Apraxia of Speech. *Clin Linguist Phon, 26*(5), 445-482. doi:10.3109/02699206.2012.655841

Shriberg, L. D., Potter, N. L., & Strand, E. A. (2011). Prevalence and Phenotype of Childhood Apraxia of Speech in Youth With Galactosemia. *Journal of Speech Language and Hearing Research, 54*(2). doi:10.1044/1092-4388(2010/10-0068)

Shriberg, L. D., & Strand, E. A. (2018). Speech and Motor Speech Characteristics of a Consensus Group of 28 Children with Childhood Apraxia of Speech - Technical Report No. 25, Phonology Project, Waisman Center University of Wisconsin-Madison.

Shriberg, L. D., Strand, E. A., Fourakis, M., Jakielski, K. J., Hall, S. D., Karlsson, H. B., . . . Wilson, D. L. (2017). A Diagnostic Marker to Discriminate Childhood Apraxia of Speech From Speech Delay: IV. The Pause Marker Index. *J Speech Lang Hear Res, 60*(4), S1153-S1169. doi:10.1044/2016_JSLHR-S-16-0149

Shriberg, L. D., Strand, E. A., Jakielski, K. J., & Mabie, H. L. (2019). Estimates of the prevalence of speech and motor speech disorders in persons with complex neurodevelopmental disorders. *Clin Linguist Phon, 33*(8), 707-736. doi:10.1080/02699206.2019.1595732

Smith, A., & Zelaznik, H. N. (2004). Development of functional synergies for speech motor coordination in childhood and adolescence. *Developmental Psychobiology, 45*(1), 22-33. doi:10.1002/dev.20009

Sosa, A. (2015). Intraword variability in typical speech development. *American Journal of Speech-Language Pathology, 24*(1), 24-35. doi:10.1044/2014_AJSLP-13-0148

Stein, C. M., Benchek, P., Miller, G., Hall, N. B., Menon, D., Freebairn, L., . . . Iyengar, S. K. (2020). Feature-driven classification reveals potential comorbid subtypes within childhood apraxia of speech. *BMC Pediatr, 20*(1), 519. doi:10.1186/s12887-020-02421-1

Sternberg, R. J., & Grigorenko, E. L. (2001). All testing is dynamic testing. *Issues in Education, 7*(2), 137-170.

Stoel-Gammon, C. (2010). The word complexity measure: Descripitons and application to developmental phonology and disorders. *Clinical Linguistics & Phonetics, 24*(4-5), 271-282. doi:10.3109/02699200903581059

Strand, E. A. (2009). Dynamic assessment of motor speech disorders in children. . In C. Bowen (Ed.), *Childrens's speech sound disorders.* (pp. 265-269): Oxford, England: Wiley-Blackwell.

Strand, E. A. (2017). Appraising Apraxia: When a speech-sound disorder is severe, how do you know if it’s childhood apraxia of speech? *The ASHA Leader, 22*(50-58). doi:10.1044/leader.FTR2.22032017.50

Strand, E. A. (2019). Dynamic Temporal and Tactile Cueing: A Treatment Strategy for Childhood Apraxia of Speech. *Am J Speech Lang Pathol*, 1-19. doi:10.1044/2019_AJSLP-19-0005

Strand, E. A., & McCauley, R. J. (2019). Dynamic Evaluation of Motor Speech Skill (DEMSS) Manual. *Brookes Publishing*.

Strand, E. A., McCauley, R. J., Weigand, S. D., Stoeckel, R. E., & Baas, B. S. (2013). A motor speech assessment for children with severe speech disorders: reliability and validity evidence. *Journal of Speech Language and Hearing Research, 56*(2), 505-520. doi:10.1044/1092-4388(2012/12-0094)

Terband, H., Maassen, B., Guenther, & Brumberg. (2009). Computational Neural Modeling of Speech motor control in Childhood Apraxia of speech (CAS). *Journal of Speech, Language, and Hearing Research, 52*, 1595–1609.

Terband, H., Maassen, B., & Maas, E. (2016). Toward a model of pediatric speech sound disorders (SSD) for differential diagnosis and therapy planning. *In Speech motor control in normal and disordered speech. Future developments in theory and methodology. ASHA press.*, 81-109.

Terband, H., Maassen, B., & Maas, E. (2019). A Psycholinguistic Framework for Diagnosis and Treatment Planning of Developmental Speech Disorders. *Folia Phoniatr Logop, 71*(5-6), 216-227. doi:10.1159/000499426

Terband, H., Namasivayam, A., Maas, E., van Brenk, F., Mailend, M.-L., Diepeveen, S., . . . Maassen, B. (2019). Assessment of Childhood Apraxia of Speech: A Review/Tutorial of Objective Measurement Techniques. *Journal of Speech, Language, and Hearing Research, 62*(8S), 2999-3032. doi:10.1044/2019_jslhr-s-csmc7-19-0214

Teverovsky, E. G., Bickel, J. O., & Feldman, H. M. (2009). Functional characteristics of children diagnosed with Childhood Apraxia of Speech. *Disabil Rehabil, 31*(2), 94-102. doi:10.1080/09638280701795030

Thoonen, G., Maassen, B., Gabreels, F., & Schreuder, R. (1996). The integrated use of maximum performance tasks in differential diagnostic evaluations among children with motor speech disorders. *Clinical Linguistics & Phonetics, 10*(4), 311-336. doi:10.3109/02699209608985178

Ttofari Eecen, K., Eadie, P., Morgan, A. T., & Reilly, S. (2019). Validation of Dodd's Model for Differential Diagnosis of childhood speech sound disorders: a longitudinal community cohort study. *Dev Med Child Neurol, 61*(6), 689-696. doi:10.1111/dmcn.13993

Turner, S. J., Hildebrand, M. S., Block, S., Damiano, J., Fahey, M., Reilly, S., . . . Morgan, A. T. (2013). Small intragenic deletion in FOXP2 associated with childhood apraxia of speech and dysarthria. *Am J Med Genet A, 161A*(9), 2321-2326. doi:10.1002/ajmg.a.36055

Turner, S. J., Vogel, A. P., Parry-Fielder, B., Campbell, R., Scheffer, I. E., & Morgan, A. T. (2019). Looking to the Future: Speech, Language, and Academic Outcomes in an Adolescent with Childhood Apraxia of Speech. *Folia Phoniatr Logop, 71*(5-6), 203-215. doi:10.1159/000500554

Tükel, S., Bjorelius, H., Henningsson, G., McAllister, A., & Eliasson, A. C. (2015). Motor functions and adaptive behaviour in children with childhood apraxia of speech. *Int J Speech Lang Pathol, 17*(5), 470-480. doi:10.3109/17549507.2015.1010578

Vick, J. C., Campbell, T. F., Shriberg, L. D., Green, J. R., Abdi, H., Rusiewicz, H. L., . . . Moore, C. A. (2012). Distinct developmental profiles in typical speech acquisition. *Journal of Neurophysiology, 107*(10), 2885-2900. doi:10.1152/jn.00337.2010

Vitevitch, M. S. (2002). The influence of phonological similarity neighborhoods on speech production. *J Exp Psychol Learn Mem Cogn, 28*(4), 735-747. doi:10.1037//0278-7393.28.4.735

Vitevitch, M. S., & Luce, P. A. (2014). Phonological Neighborhood Effects in Spoken Word Perception and Production. *Annual Review of Linguistics, 2*(1). doi:10.1146/annurev-linguistics-030514-124832

Vygotsky, L. (1978). Interaction Between Learning and Development. *Readings on the Development of Children*(Second Edition, Edited by Gauvain, M. & Cole, M), 29 - 36.

Waring, R., & Knight, R. (2013). How should children with speech sound disorders be classified? A review and critical evaluation of current classification systems. *Int J Lang Commun Disord, 48*(1), 25-40. doi:10.1111/j.1460-6984.2012.00195.x

Waring, R., Rickard Liow, S., Dodd, B., & Eadie, P. (2022). Differentiating phonological delay from phonological disorder: Executive function performance in preschoolers. *Int J Lang Commun Disord, 57*(2), 288-302. doi:10.1111/1460-6984.12694

WHO. (2013). International Classification of Functioning, Disability and Health (ICF).

WHO. (2020). International Classification of Functioning, Disability and Health (ICF).

Wikse Barrow, C., Körner, K., & Strömbergsson, S. (2021). A survey of Swedish speech-language pathologists' practices regarding assessment of speech sound disorders. *Logoped Phoniatr Vocol*, 1-12. doi:10.1080/14015439.2021.1977383

Williams, P., & Stephens, H. (2004). The Nuffield Centre Dyspraxia Programme. *[www.ndp3.org]*.

Yoss, K. A., & Darley, F. L. (1974). Developmental apraxia of speech in children with defective articulation. *Journal of Speech and Hearing Research, 17*, 399–416.

Zaretsky, E., Velleman, S. L., & Curro, K. (2010). Through the magnifying glass: Underlying literacy deficits and remediation potential in childhood apraxia of speech. *Int J Speech Lang Pathol, 12*(1), 58-68.

Zuk, J., Iuzzini-Seigel, J., Cabbage, K., Green, J. R., & Hogan, T. P. (2018). Poor Speech Perception Is Not a Core Deficit of Childhood Apraxia of Speech: Preliminary Findings. *J Speech Lang Hear Res, 61*(3), 583-592. doi:10.1044/2017_JSLHR-S-16-0106