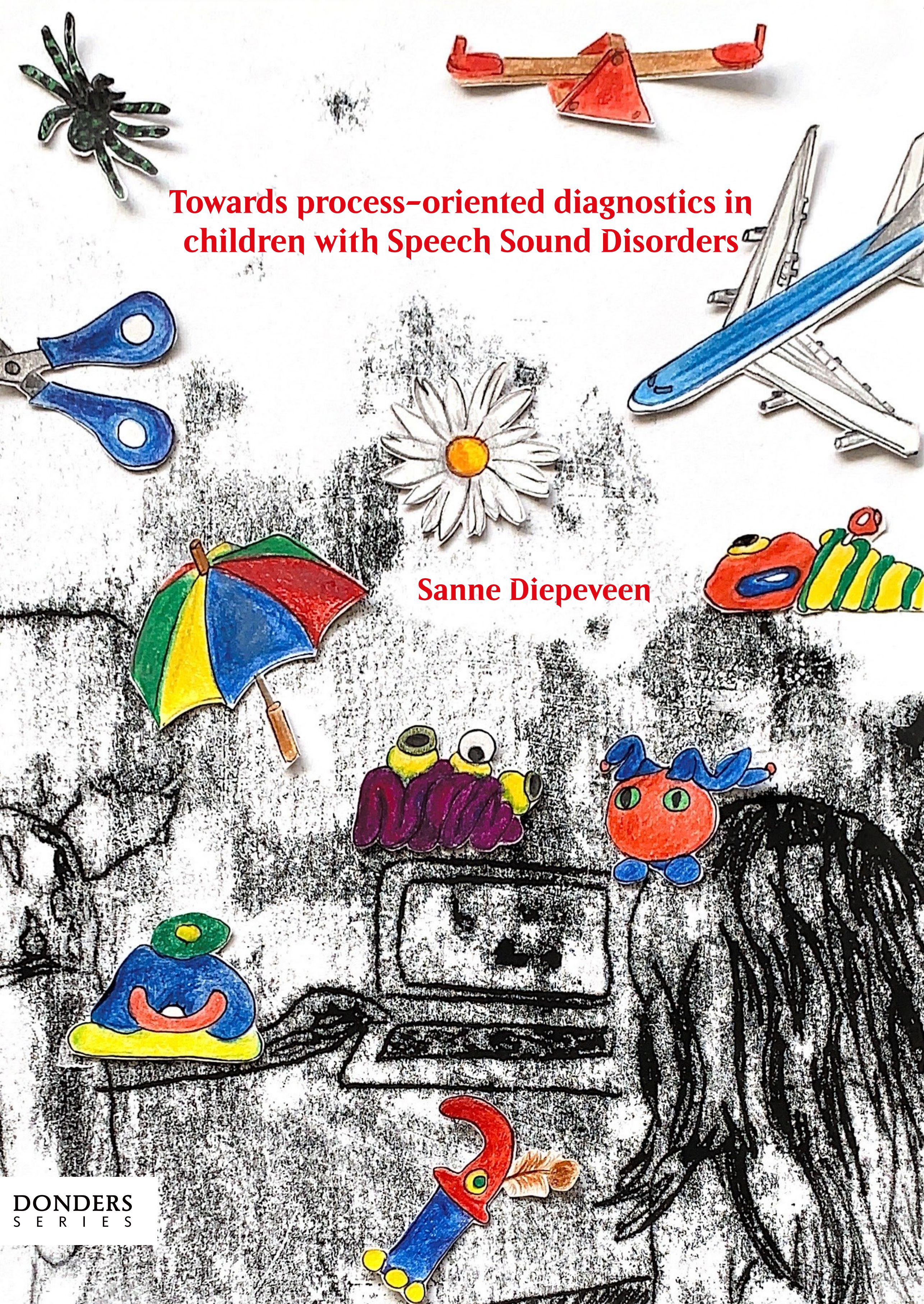
Towards process-oriented diagnostics in children with Speech Sound Disorders

Sanne Diepeveen



DONDERS SERIES

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ISBN: 9789464690682
Layout and Cover: ProefschriftMaken | proefschriftmaken.nl
Printed: ProefschriftMaken | proefschriftmaken.nl
Cover illustration: Elly van Santen

Towards process-oriented diagnostics in children with Speech Sound Disorders

Proefschrift

ter verkrijging van de graad van doctor
aan de Radboud Universiteit Nijmegen
op gezag van de rector magnificus prof. dr. J.H.J.M. van Krieken,
volgens besluit van het college voor promoties
in het openbaar te verdedigen op

maandag 28 november 2022

om 16.30 uur precies

door

Susanna Johanna Helena Diepeveen

geboren op 13 maart 1978

te Boxmeer

Promotor: Prof. dr. B.A.M. Maassen (Rijksuniversiteit Groningen)

Copromotoren: Dr. H.R. Terband (University of Iowa, Verenigde Staten)

Dr. B.J.M. de Swart

Manuscriptcommissie: Prof. dr. P.J. van der Wees

Prof. dr. R. Jonkers (Rijksuniversiteit Groningen)

Dr. E. Janse

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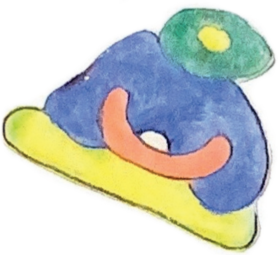

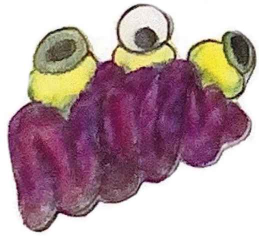

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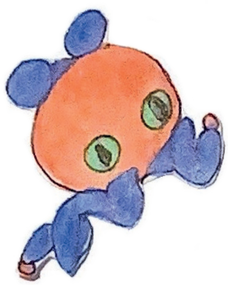
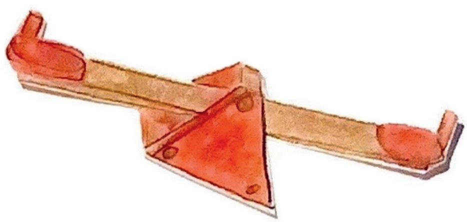
CHAPTER 1



General Introduction



Children learn to speak to express their needs and wishes and to share their thoughts with others. Learning to speak begins at an early age: the auditory system functions even before birth to learn which sounds, melodies, and rhythms of the ambient language are pronounced in each word, and children typically produce their first word about 12 months after birth (Hoff, 2014). However, learning to speak is not easy for all children. Children with a speech sound disorder (SSD) struggle to produce speech sounds and do not reach the developmental milestones. SSD is an umbrella term that applies to all or combinations of difficulties with speech perception, phonological representation, or motor production of speech sounds and speech segments (American Speech-Language-Hearing Association, 2021).

Speech disorders are among the most common communication disorders in the caseloads of speech and language pathologists (SLPs) who work with children (Joffe & Pring, 2008; Priester et al., 2009; Waring & Knight, 2013). Between the ages of 4 to 8 years, an estimated 3.4–3.8% of all children have an SSD (Eadie et al., 2015; Shriberg et al., 1999; Wren et al., 2016). Meijer et al. (2020) reported that 46% of all referrals for speech and language therapy in private SLP practices in the Netherlands in 2019 involved children aged 4 to 7 years. In 21% of those cases, the child had been diagnosed with an SSD (Meijer et al., 2020). A prospective community study by Morgan et al. (2017) recruited 1,494 children before the age of 12 months and reassessed them repeatedly until the age of 13 years. Data were analysed until the age of seven years. At age four, 164 (11%) of the children produced non-age-appropriate speech errors. 93 of these children were reassessed at age seven; the other 71 children were not included for various reasons (e.g., parents declined further follow-up). Of the remaining 93 children, more than 40% were still classified as having persistent speech errors. Such speech problems can complicate a child’s acquisition of academic and psychosocial skills (Cabbage et al., 2018; McCormack et al., 2009, 2010; Preston et al., 2013). For example, children who have a speech problem at the beginning of the reading process are at increased risk of developing reading difficulties. Therefore, early detection combined with treatment of an SSD might prevent problems later in life (Hayiou-Thomas et al., 2017; Raitano et al., 2004; Tambyraja et al., 2020).

An SLP must assess the cause of a child’s speech problem before they can choose the right intervention. Since there are many types of SSDs, it can be difficult for SLPs to detect the underlying causes of speech problems and correctly diagnose children (American Speech-Language-Hearing Association, 2021; Bernthal et al., 2017). The causes of speech problems lie in the processes underlying speech production, such as articulatory or motor skills and cognitive-linguistic skills (Stoel-Gammon & Vogel-Sosa, 2014). Children with an SSD may have difficulties in one or both of these domains (Stoeckel & Caspari, 2020). It is complicated to identify the processes that underlie a child's speech difficulties. For example, a speech error such as [tat] instead of 'kat' ('cat') may be the result of an immature phonological system or a motoric simplification. In the former case, it may be that the /k/ has not yet been incorporated into the child's phonological system in word-initial position. In the latter case, the speech error could be due to a motoric simplification; in this context, producing a /t/ instead of a /k/ involves less variation in articulation. Therefore, it is important to gather information about the different speech processes so they can be disentangled in assessment (Bates & Titterington, 2017). Diagnosing the underlying processing deficits forms the foundation for determining a suitable intervention (Shulga et al., 2020). Therefore, it is important for SLPs to have adequate assessment tools to gain a good understanding of underlying speech processes.

This chapter continues with a description of typical speech development. Subsequently, it will describe the classification models for differential diagnosis of SSDs and present a novel speech assessment. The chapter ends with the outlines and aims of this thesis.

Typical speech sound development

The Dutch language contains 19 consonants (shown in Table 1) plus four additional consonants that occur only in loan words (in parentheses) (Mennen et al., 2007). Most consonants occur in syllable-initial positions (except /ŋ/); every consonant occurs in word-final position except voiced plosives, voiced fricatives, and /h/. The consonants /c, ʃ, ʒ, ɲ/ only occur in loan words and/or as allophones. The vowels can be divided into a set of long vowels /i, y, u, e, ø, o, a/, a set of short vowels /ɪ, ɛ, ɔ, ʉ, ɑ/, a reduced vowel /ə/, and three diphthongs /ɑu, ɛi, ʉy/ (Jonkers et al., 2014; Mennen et al., 2007; Van Haaften, Diepeveen, van den Engel-Hoek et al., 2020). Syllables consist of a vowel, zero or up to three consonants in the initial positions, and zero or up to four consonants in the final position.

Table 1. Dutch consonants

	Manner of articulation				
Place of articulation	Plosives	Fricatives	Nasals	Liquids	Glides
Bilabial	p, b		m		
Labiodental		f, v			w
Alveolar	t, d	s, z	n	l, r	
Postalveolar	(c)	(ʃ), (ʒ)	(ɲ)		
Palatal					j
Velar	k, (g)	x	ŋ		
Glottal		h			

Many researchers have examined the typical development of speech sounds in Dutch (Beers, 1995; Fikkert, 1994; Levelt et al., 2000; Levelt, 1994; Priester et al., 2013; Stes, 1977; van den Berg et al., 2017). Our research group used the Picture Naming task from the Computer Articulation Instrument (CAI; van Haaften et al., 2020; see also the Speech Assessment section in this chapter for more information) to examine the speech sound development of Dutch children. 1,503 children aged 2;0 to 6;11 years completed the Picture Naming task; 14 age groups were formed with a range of 4 months for children aged 2;0 to 5;11 years and a range of 6 months for those aged 6;0 to 6;11 years. Over 100 children were assessed for each age group. The sample was representative of the general Dutch population in terms of gender, geographic region, degree of urbanisation, and parental socio-economic status (Maassen et al., 2019). SLP students and SLPs administrated the CAI. The children named the pictures that appeared on the computer screen, and their utterances were automatically recorded in the CAI programme and later transcribed. The results showed:

1) The number of correctly produced speech sounds (Percentage Consonant Correct-Revised, PCC-R) and vowels (Percentage Vowel Correct, PVC) in the words increased with age. PVC was systematically higher than PCC-R. Differences between these scores were larger for the youngest age groups.
2) Vowels were acquired at 3;4 years. Children produced all syllable-initial consonants for ≥ 75% correctly, except for the voiced fricatives /v/ and /z/ and the liquid /r/ at 3;7 years. All final consonants were acquired at 4;4 years.
3) Regarding the degrees of phonological complexity described by Beers (1995), we found that degree 1 consonants (syllable-initial /p/, /t/, /m/, /j/ and /n/) were produced correctly at 2;0 years. The dorsal consonant /k/ (degree 2) was correctly produced at 2;8 years. The continuants /s/, /x/ and /h/ (degree 3) were acquired at 2;4 years. Degree 4 with the consonants /b/, /f/ and /w/ was complete at 2;8 years. And degree 5 (consonants /l/ and /r/) was acquired at 3;8 years.
4) Simple syllable structures (CV, CVC, and V) emerged first in speech development. This was followed by structures with a cluster of two consonants (CCV, CCVC, CVCC) and then syllables with clusters of three (CCCVC). At 4;7 years, the children could produce all syllable structures except for CCVCC (which was acquired after the age of 6;11 years).
5) Younger children used more phonological simplification processes than older children. At 4;4, children did not use phonological simplification processes in their speech, except for the final cluster reduction from two to one consonant (44.5% of the children) and occasionally the initial cluster reduction from three to two consonants (14.3% of the children). We did

not observe the unusual processes that Beers (1995) described in her data (i.e., backing, nasalisation, H-sation, and lateralisation).

The results of this study form reference norms that enable Dutch SLPs to distinguish children with speech problems from typically developing children. This was previously impossible because there were no norm-based speech assessments available in the Netherlands (van Haaften et al., 2020).

The psycholinguistic model

Children with SSDs form a heterogeneous group in terms of severity, symptoms, and underlying causes. Differential diagnosis of subtypes of SSDs is a complex process; SLPs must distinguish between underlying speech processes that can be articulatory, motor speech, and/or cognitive-linguistic based (Namasivayam et al., 2020; Terband et al., 2019). The psycholinguistic model presented in Figure 1 can aid in describing/differentiating the underlying speech processing levels involved (Levelt, 1989; Stackhouse & Wells, 1997; Terband et al., 2019). A speaker must first conceptualise what (s)he wants to say and formulate a preverbal message. Grammatical encoding is then used in the process of formulating utterances which entails the selection of a lemma (containing meaning and grammatical information) and the lexeme or word form. This process results in a grammatical code. Next, the grammatical code must be given a phonological form code. This process is called phonological encoding. During this process, speech sounds and syllables are selected and sequenced in a phonological phrase of linguistic/symbolic units. After the phonological encoding, the speaker constructs a motor movement plan that comprises the selection and sequencing of the articulatory movement goals and adapts these goals to the phonetic environment. This plan defines and organises all

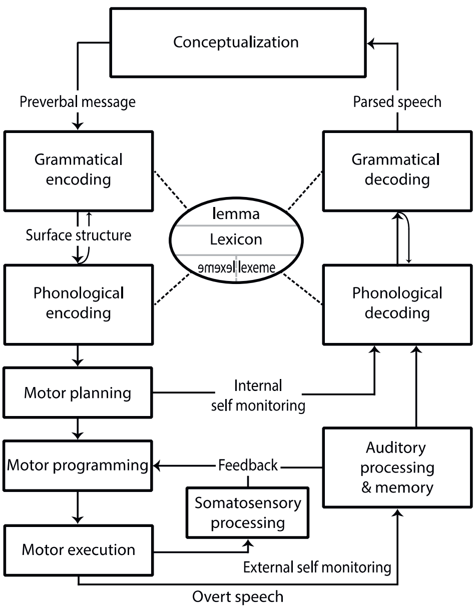


Figure 1. Adult model of speech processing (Terband et al., 2016b, 2016a) (adapted from Levelt, 1989; Levelt et al., 1999; Van der Merwe, 1997, 2009; Guenther, 1994; Guenther & Perkell, 2004) displaying the sensorimotor and memory functions involved in speech production and perception (Terband et al., 2019).

movement patterns of the sounds, the sound transitions, the syllables, and the word boundaries. The motor plan is then implemented in a motor programme with muscle-specific information, i.e., the speaker constructs the neural signals that lead the articulators to achieve their movement goals considering articulatory context, sensory information, and (meta)linguistic requirements (e.g., prosody, pitch, lexical stress). In the motor execution phase, the speaker executes the speech motor programme and the constructed neural signals set the articulators in motion. These stages form a constant process during speaking that is constantly monitored at various levels: a person receives feedback from an internal self-monitoring system based on somatosensory and auditory feedback. During each processing stage, difficulties may occur that affect the speech output. Thus, identifying the origin of the overt speech symptoms requires a speech assessment that provides information about each stage of the speech production process.

Speech assessment and diagnosis

Most SLPs use a naming task to assess children’s speech (McLeod & Baker, 2014). Priester et. al (2009) concluded that 70% of SLPs in the Netherlands use the naming task Logo-art and do not use the same assessment for every child. Logo-art is a speech assessment in which children name different pictures. The words in this assessment contain each consonant of the Dutch language once in a particular position. Recently, Yeh and Liu (2021) concluded that in terms of intelligibility, speech accuracy, and phonemic inventory, a Picture Naming task provides more information to differentiate children with SSD from typically developing children than a coherent or spontaneous speech sample. Macrae (2017) questioned whether a naming task with each consonant only once in a particular postion would gather enough information about a child’s speech. He suggested that a speech assessment should include a naming task with at least two opportunities to express each consonant in different word positions and an analysis of the child’s spontaneous speech. Others also proposed obtaining a spontaneous or connected speech sample to gain a more comprehensive view of a child’s speech (Barrett et al., 2020; Fabiano-Smith, 2019; Masterson et al., 2005). However, speech cannot be only assessed with a Picture Naming task or a connected speech sample because these kinds of assessments only provide the SLP an inventory of speech errors (e.g., how many omissions or which substitutions occur). They do not help the SLP analyse why the omissions or substitutions occurred, and this information is crucial to selecting an appropriate intervention (Terband et al., 2019). As mentioned above, the production of speech involves a chain/variety of processes, and a problem can exist in any of these processes. Therefore, a speech assessment should contain multiple tasks to assess the different underlying processes and the association between them (Terband et al., 2019).

There was no such integrated assessment in the past, which is why our research group developed the Computer Articulation Instrument. The CAI is a novel, norm-referenced speech assessment that combines different tasks in one instrument and can provide an overview of different speech processes based on the model explained in the previous paragraph (see also Chapters 5 and 6 of this thesis). The CAI consists of four tasks which are administered through a computer. The SLP and the child sit side by side facing a computer screen. The SLP operates the computer while the computer program gives instructions. During the assessment, the child's speech is automatically recorded and saved on the computer's hard drive. The SLP analyses the child's speech in the CAI after the assessment. The first task in the CAI is Picture Naming, in which the child names 60 pictures presented one by one on the computer screen. In the other three tasks, the stimuli are presented auditorily. In the second task (NonWord Imitation), the child repeats monosyllabic, bisyllabic, and/or trisyllabic words (depending on their age). The third task (Word and NonWord Repetition) is a consistency task in which the child repeats five words and non-words five times. The final task is a diadochokinesis task named Maximum Repetition Rate (MRR): the child repeats three monosyllabic sequences (/pa/, /ta/, /ka/), two bisyllabic sequences (/pata/, /taka/), and one multisyllabic sequence (/pataka/) as quickly and accurately as possible (Maassen et al., 2019). The parameters of the CAI are presented in Table 2.

Table 2. Parameters of the Computer Articulation Instrument

Task	Parameter	
PN	PCCI	Percentage of Consonants Correct in syllable-initial position
	PVC	Percentage of Vowels Correct
	Level 5	Percentage of correct consonants /l/ and /R/
	RedClus	Percentage of Reduction of Initial consonant Clusters from 2 consonants to 1
	CCVC	Percentage of correct syllable structure CCVC (C=consonant, V=vowel)
NWI	PCCI	Percentage of Consonants Correct in syllable-initial position
	PVC	Percentage of Vowels Correct
	Level 4	Percentage of correct consonants /b/, /f/ and / ʋ /
	Level 5	Percentage of correct consonants /l/ and /R/
	RedClus	Percentage of Reduction of Initial consonant Clusters from 2 consonants to 1
	CVC	Percentage of correct syllable structure CVC
	CCVC	Percentage of correct syllable structure CCVC
WR	PWV Word	Proportion of Whole-Word Variability – Word Repetition
NWR	PWV Nonword	Proportion of Whole-Word Variability – NonWord Repetition
MRR	MRR pa	Number of syllables per second of sequence /pa/
	MRR ta	Number of syllables per second of sequence /ta/
	MRR ka	Number of syllables per second of sequence /ka/
	MRR pataka	Number of syllables per second of sequence /pataka/
	MRR pata	Number of syllables per second of sequence /pata/
	MRR taka	Number of syllables per second of sequence /taka/

Note. PN = Picture Naming; NWI = NonWord Imitation; WR = Word Repetition; NWR = NonWord Repetition; MRR = Maximum Repetition Rate.

In a previous study on the CAI, van Haaften et al. (2019) collected normative data from 1,524 typically developing Dutch-speaking children (aged 2;0 to 7;0 years). The children were tested at school by SLP students or an SLP. After the session, the recorded speech was analysed according to a purpose-fitted protocol. Continuous norms were calculated which showed that children improved with age on all the tasks of the CAI. Therefore, SLPs can use this normative data to distinguish between typically developing children and those who are delayed in speech or have a potential speech disorder. In addition, five factors emerged from this data: 1) all Picture Naming parameters; 2) the segmental parameters of NonWord Imitation (PCCI, PVC, level 4, and level 5); 3) the syllabic structure parameters of NonWord Imitation (RedClus, CVC, and CCVC); 4) (Non)Word Repetition consistency, and 5) all MRR parameters. This confirms that the CAI consists of several separate tasks.

Inter-rater reliability and test-retest reliability were also calculated with the Intraclass Correlation Coefficient (ICC) for a subgroup of the total sample (van Haaften et al., 2019). The ICC for interrater reliability showed a range from sufficient to good, except for percentage of vowels correct on Picture Naming and NonWord Imitation and for the MRRs for bisyllabic and trisyllabic items. The variation between the ICCs might be due to the use of different raters. However, the point-to-point interrater agreement of the phonetic transcriptions of the Picture Naming and NonWord Imitation responses (total number of consonants, vowels, and word and syllable boundaries) was high for both tasks. The ICCs for test-retest reliability were sufficient for most parameters of the Picture Naming task except for the PCCI and the PVC. The test-retest reliability for the other tasks of the CAI (NonWord Imitation, Word and NonWord Repetition, and MRR) were insufficient, likely due to the children's development between the two measurements and the effect of learning. However, these parameters are still part of the analysis of the CAI because they could play an important role in diagnosing children with SSDs and obtaining a complete speech profile (van Haaften et al., 2019). These results indicate that the CAI is a reliable and valid instrument for mapping children's speech and can be used to differentiate between typical and atypical speech development.

Aims and outline of the thesis

As described above, it is important that an SLP uses process-oriented diagnostics to thoroughly analyse the child's whole speech chain. In a study by Priester et al. (2009), SLPs reported using a picture naming task to diagnose children with an SSD, but that task alone is not enough to analyse the whole speech chain. So, what do SLPs do for clinical reasoning when assessing children with an SSD and, subsequently, choosing an adequate intervention? The CAI can play a role in process-oriented diagnostics since its tasks facilitate the analysis of underlying speech processes. In this respect, it is important to have a clear picture of the different CAI tasks, such as the MRR. It is also important to investigate the role of the CAI in distinguishing typically developing children from children with an SSD and investigate whether the CAI can be used to distinguish between different types of SSDs.

The following three aims were formulated to investigate these topics further. The first aim of this thesis was to gain insight into the clinical reasoning of SLPs during the process of diagnosing children with an SSD and determining the treatment trajectory. The second aim was to determine the role the recently developed CAI might play in the diagnostic process, and determine the instrument's reliability, validity, and responsiveness for assessing underlying speech production processes and deficits. This thesis especially focused on the contribution of the MRR task (the other tasks are discussed in two recently published studies: see van Haaften et al., 2019, 2020). The third aim was to investigate the role the CAI could play in differentiating subtypes of SSDs and thus its clinical value for assessing speech profiles.

Outline of the thesis

Chapter 2 addresses the clinical reasoning of the diagnostic and intervention process of children with SSDs (first aim). The aim of this study was to investigate the steps an SLP takes in diagnosing and guiding children with an SSD and their parents. 137 SLPs filled in a questionnaire and 33 SLPs participated in a semi-structured interview. The questionnaire and interview contained questions about the clinical reasoning SLPs use in the diagnostic and intervention process for SSDs.

Our group developed the CAI to help SLPs make differential diagnoses of children with SSDs. In previous publications about the CAI, we looked at the psychometric evaluation of the instrument (van Haaften et al., 2019) and the phonological development of children with the Picture Naming task (van Haaften et al., 2020).

This thesis adds the MRR task, which is presented in Chapters 3 and 4 (second aim). **Chapter 3** contains a study that included 1,524 children aged 2;0 to 6;11 years who performed the MRR task. The study describes the development and implementation of the administration procedure. We tested several variants of this administration protocol to develop a valid and reliable administration protocol for the CAI.

Chapter 4 describes the norm values of the MRR for children (*n* = 1041) between 3;0 and 6;11 years and the profiles of performance to describe developmental trends. This study used the proposed protocol from Chapter 3 and norm values were collected and described per age group.

The second part of the thesis describes how the CAI can be used to assess and diagnose children and classify their speech problems (third aim). The clinical application and validation of the CAI was examined in three experiments involving children with SSDs from different age groups.

Chapter 5 describes two groups of children with SSDs aged between 3;0 and 6;4 years. This study assessed possible performance profiles and validated those against clinical judgements. The first study compares the scores on the Picture Naming (PN) task of the CAI with a judgement by the SLP on intelligibility. The second study validates the speech profiles for the four to six factor scores extracted from all four tasks of the CAI: PN; NonWord Imitation (NWI); percentage whole-word variability (PWV); and MRR with the clinical judgements of severity of the speech sound disorder by their SLP.

Chapter 6 reports the results of a study in which the CAI was used to examine a group of children aged 4;0 to 6;11 years with SSDs. A factor analysis was conducted on all the tasks of the CAI followed by a cluster analysis to see if profiles of groups of children could be detected.

Chapter 7 contains the general discussion of the earlier chapters. It looks at clinical decision making, diagnostic needs, the speech profiles of children with speech problems, and the role of the CAI. Furthermore, it describes clinical decision making based on the results of a clinical assessment of children with SSDs using the CAI and recommendations for SLPs' clinical practice.

This thesis is based on published journal articles, so some overlap between the chapters is inevitable.

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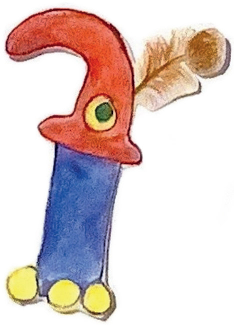
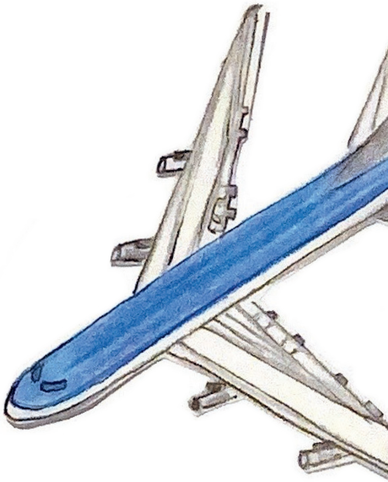
CHAPTER 2





Clinical reasoning for SSDs: Diagnosis and intervention in SLPs' daily practice

Sanne Diepeveen, Leenke van Haaften, Hayo Terband,
Bert de Swart, Ben Maassen

Published in American Journal of Speech-Language Pathology 2020; 29(8): 1529–1549

Abstract

Purpose: This study aims to give an insight in clinical reasoning (diagnosis and intervention) of SLPs in the Netherlands for children with SSD.

Method: The study featured a mixed-method (qualitative and quantitative) design. Semi-structured interviews containing nondirective, open-ended questions were conducted with 33 SLPs, which were analysed using a constant comparative analysis. Other SLPs (137) filled out a questionnaire on the same topics. Multiple-choice questions were analysed by descriptive frequencies, while open-ended questions were analysed thematically.

Results: The results indicate that SLPs use a variety of assessments to diagnose SSD, complemented by observation and often case history. In total 85 different diagnostic labels were reported. The choice of intervention is based on what is appealing to the child, matches his/her age as well as the specific diagnosis and severity. Interventions are used for multiple speech disorders and, according to SLPs, parents play a large role in diagnostics and intervention.

Conclusions: These results reveal the need for a (1) clear and consistent terminology of diagnoses in the field of paediatric SSD; (2) fast and easy-to-administer comprehensive differential diagnostic instrument in combination with an instrument to assess participation in everyday life; (3) tool to conduct a case history online.

Introduction

Clinical reasoning - or practice decision making - refers to thinking about and making decisions as a health care provider in a context-dependent situation. It is a contextualised interactive phenomenon (Higgs & Jones, 2008) that comprises two elements: (1) diagnostic reasoning -collecting and analysing information- and (2) therapeutic reasoning -making sure the patient's circumstances and needs are included (Ajjawi & Higgs, 2008).

Classification systems for speech sound disorders

Clinical reasoning is difficult to capture for speech sound disorders (SSD). Clinicians rarely talk about clinical reasoning itself (Hoben, Varley, & Cox, 2007) and the field of SSD underwent large changes in the last decades, which causes clinicians to change their way of thinking. Several diagnostic classification systems and models were presented with either an etiological, a descriptive-linguistic or a processing approach (Tyler, 2010). Two systems are predominant at the moment (Waring & Knight, 2013; Terband, Maassen & Maas, 2019): Dodd's Model of Differential Diagnosis (MDD; Dodd, 2014) and Shriberg's Speech Disorders Classification System (SDCS; Shriberg et al., 2017; Shriberg et al., 2010). The MDD is a psycholinguistic model of speech production an development containing the categories: phonological disorder, phonological delay, consistent atypical phonological disorder, inconsistent phonological disorder, phonetic articulation disorder and developmental apraxia of speech. The SDCS is based on the presumed aetiological background of the speech problem and contains the categories: speech delay, genetic, otitis media, psychosocial, motor speech disorders, apraxia, dysarthria, not otherwise specified, residual speech errors, /s/ and /r/. However, not all categories have specific and sensitive diagnostic markers to differentiate them (Terband, Maassen & Maas, 2019). Also, a consensus on a classification system fails, due to the heterogeneity among children with SSD: they differ in severity, aetiology, speech characteristics, involvement of other aspects of the linguistics system, treatment response, and maintenance factors (Dodd, 2011). As Waring and Knight (2013) concluded, there is a need for "an inclusive, universally agreed-upon classification system that meets the needs of clinicians and researchers" (p. 25). Furthermore, such a classification system should contain directions for speech-language pathologists (SLPs) to choose treatment and be universally applied and implemented (McLeod & Baker, 2014). A framework that could help during the diagnostic process is the *International Classification of Functioning, Disability and Health* (ICF; World Health Organization, 2001). The ICF comprises a holistic approach that covers all three possible angles (etiological, descriptive-linguistics and processing) in its different components. McLeod and Bleile (2004) described which kind of examination and intervention could be used for each of the levels of the ICF (body structures, body functions, activity, and participation).

First examinations are described, followed by examples of interventions. For body structures, SLPs should conduct an oromuscular and an audiological assessment. At the level of body functions SLPs should analyse a speech sample (phonological processes). McLeod and Bleile (2004) combined the levels of activity and participation; an SLP can conduct an intelligibility assessment and gather information about successes and difficulties in participating in everyday live. For the intervention two sets of goals could be applicable: impairment-based goals and socially-based goals. Interventions at the level of body structure are an operation and acceptance of the different facial structure. Goals for body functions entails accurate productions, reduce unusual speech patterns, and increase intelligibility. For activity and participation, they suggest using the following goals: correctly pronounce names and words the child likes to utter; promote communication successes by collaborating with a teacher and peers.

Diagnostic process

To diagnose children with SSD, clinicians have the choice of several assessments. In a survey of SLPs in the United States, more than 50% of the clinicians reported to estimate intelligibility. They also conduct a standardised single-word test and a hearing screening, and they test stimulability and oral motor skills (Skahan, Watson and Lof, 2007). This should be supplemented with additional single-word testing to get an overview of all the consonants and vowels in all positions multiple times, connected speech sample, phonological analyses and it may include inconsistency testing (Macrae, 2016). Fabiano-Smith (2019) provided an evidence-based protocol of ten steps, which starts with a detailed case history (step 1). Step 2 (Routine Assessments) includes an oral peripheral assessment and a full hearing evaluation. In step 3, the SLP obtains a connected speech sample. This sample is then used to get a phonetic inventory (step 4). Consonant accuracy is subsequently established with the help of a wordlist of a standardized single-word test (step 5). In step 6, the SLP analyses the speech errors in the phonetic transcription of the speech sample. Step 7 entails a phonological error pattern analysis and is followed by a measure of whole-word proximity in step 8. Step 9 comprises testing the stimulability with the Scaffolding Scale of Stimulability (Glaspey & Stoel-Gammon, 2005) and in the last step (step 10), the SLP estimates the intelligibility of the child (Fabiano-Smith, 2019). These ten steps can help the SLP to diagnose a child properly.

However, what fails is a single assessment that can differentiate between the various diagnostic categories (Terband, Maassen, & Maas, 2019; Terband, Maassen, & Maas, 2016). Current diagnostic instruments tend to focus on speech sound disorders separately. For example, Hodson Assessment of Phonological Patterns (Hodson, 2004); HAPP-3) and Metaphonbox (Leijdekker-Brinkman, 2005) for a phonological disorder; PROMPT (Hayden, 2008), The Nuffield Dyspraxia

Programme (Williams & Stephens, 2004) or the pause marker (PM) (Shriberg et al., 2017) for Childhood Apraxia of Speech (CAS). SLPs depend on their own clinical reasoning to try to establish a differential diagnosis of the speech disorder.

All mentioned diagnostic instruments measure the problems on the function level of the ICF, but this is not enough to get a broad view on the problems parents and child experience during everyday activities (McLeod, 2004). SLPs can use a case history to ask parents about the problems they experience on the activity and participation levels. In addition, an SLP can perform an observation to see how the child functions during, for example, a game. In order to examine the speech skills at the level of participation, the SLP can ask parents to fill out the Intelligibility in Context Scale (ICS) (McLeod, Harrison, & McCormack, 2013).

Intervention process

If an SLP established a diagnosis, she/he has to choose from multiple interventions (Baker & McLeod, 2011a). For example, in the case of a phonological disorder an SLP can choose between cycles approach (Hodson & Paden, 1983), minimal pair intervention (Leijdekker-Brinkman, 2005) or combine them. In a survey of 2084 SLPs in the United States SLPs reported using a traditional intervention more often than other inventions. They use certain parts of phonological interventions, and they provide phonological awareness training (Brumbaugh & Smit, 2013). The choice for an intervention requires the SLP to consider the differential diagnosis as well as the therapeutic, the scientific perspectives and elements of the child's background, such as age, family circumstances, parents' collaboration (Baker & McLeod, 2011b; Dodd & Bradford, 2000). Clinical reasoning thus plays a critical role in the work of an SLP, both during the diagnostic process as during the invention. However, little is known about how this process takes shape in daily practice.

Some authors concluded that SLPs use their experience rather than scientific knowledge (Brumbaugh & Smit, 2013; Mcleod & Baker, 2004; Joffe & Pring, 2008). This is due to a lack of time to read scientific articles and to the lack of sufficient scientific evidence for certain (combinations of) interventions or methods. Many SLPs were found to combine interventions or methods, without any scientific evidence such as a randomized controlled trial (Baker & McLeod, 2011b; Joffe & Pring, 2008). However, they should be able to apply these combinations of interventions based on their clinical experience (Sizer et al., 2016). Although expert opinions are not the strongest evidence, this should play a bigger role in evidence-based practise (e.g. Hofmeijer, 2014). An expert can detect differences and patterns in treatment outcomes and eventually a randomised control trial can be started. Therefore, it is imperative to understand how clinicians diagnose children with SSD and how they plan the intervention. Patterns in the decision-making process can serve as starting points for further research, for example treatment efficacy.

Aims of the study

The aim of the present study was to investigate clinical reasoning of SLPs in daily practice using a mixed-method (combining qualitative and quantitative) design. SLPs active in several settings in the Netherlands participated in either a semi-structured interview containing nondirective, open-ended questions or a questionnaire containing both closed (multiple-choice or rating) and open-ended questions. Both interview and questionnaire focused on what choices the SLPs make during the processes of diagnosing children with SSD and planning and administering the intervention. The research questions were as follows:

(1) Which steps does an SLP take to eventually diagnose a child with a suspicion of SSD and are there differences in method between SLPs?
(2) What are the labels (diagnosis) and which labels are more common?
(3) Which methods and didactics are used by the SLPs?
(4) What is the role of the parents in the whole process?

Method

Medical-ethical Permission from an ethical review board

Given the fact that all the participating SLPs were adults and the questions in the interview and questionnaire did not solicit any personal information about their clients, permission from an ethical review board was not required for the present study according to the Dutch administrative authority (CCMO, 2018).

Mixed-method design

A concurrent triangulation (use of multiple methods) design mixed-method study was used in which quantitative and qualitative data are gathered. An important advantage of such a research design is that two complementary datasets are analysed and cross-verified with the results merged into a single overall interpretation, thereby increasing the validity of the findings (Bekhet & Zauszniewski, 2012).

The interview was used as the qualitative method design to get a more in-depth view on the decisions an SLP makes during the diagnostic and intervention process for children with SSD, while the questionnaire was used as the quantitative method design to collect data from a large group of participants. The data from the interviews was used to supplement the questions in the questionnaire.

Interview

Participants

Thirty-three SLPs working in a private practice or school in the Netherlands were interviewed, 23 from the province Gelderland (eastern part of the Netherlands) and 10 from Brabant (southern part of the Netherlands), between September 2013 and December 2014. Table 1 presents an overview of the background of the participants. The SLPs were recruited via email and/or telephone (obtained via an internet search). The trained research assistants conducted the interview in pairs at the participating SLPs workplace. The participants gave their written informed consent. They could withdraw from the study at any time. One research assistant executed the interview, while the other observed and intervened, if necessary, by asking to clarify an answer or to get additional information. The interviews were audio recorded. The data was processed anonymously.

Data collection

Semi-structured interviews were conducted comprising nondirective, open-ended questions organised in three sections: background of the SLP, decision making in diagnosis of SSD and intervention of SSD (see Appendix 1). Ten research assistants from HAN University of Applied Sciences (final year SLP students) were trained to conduct the interviews according to a predefined protocol (interview guide, see Appendix 1) developed by the first author in consultation with the co-authors and three independent SLP lecturers. The training comprised both theoretical knowledge about collecting qualitative data (reading and attending lectures), and practical skills (role-playing and conducting several pilot interviews with feedback from the first author).

Data analysis

The interviews were transcribed verbatim by the research assistant who was present at the interview and was checked by a research assistant who was not present at the interview. All SLPs participated in a member-checking process of the verbatim; no changes were necessary according to the SLPs. During the analysis process, the objective was to identify key issues in the data. A constant comparative method was used (Corbin & Strauss, 2008); this is the reason why validity could not be determined. First, one interview was segmented and coded independently by the first author and three research assistants. Comments of the SLP were labelled with a word or short phrase that expressed the key issue (code). The resulting codes were then evaluated between the first author and the research assistants, and a consensus list of codes was composed. Second, this list was used to code the remaining interviews with assistance of the computer software Atlas-TI© version 7 (www.atlasti.com, GmbH, Berlin, 1993-2013) to organise the data. Each interview was coded individually; the code list was adapted when new codes appeared in the interviews. The new codes were discussed between the first author and the research assistants to increase the rigour and transparency of the process. No new codes appeared after the 29th participant, meaning saturation was reached. All interviews were then checked again with the completed list. After coding all interviews, the codes were organised in coherent themes or sub-themes (axial coding; Corbin & Strauss, 2008) if possible, by the first author and the research assistants independently. The themes or sub-themes were compared and discussed if these did not match.

Questionnaire survey

Participants

SLPs with experience in the field of children with SSD were recruited via newsletters from professional associations and a Facebook-group for SLPs in the Netherlands. After three weeks, the call was repeated. The questionnaire was available on a website between May and September 2014, during which it was filled out by 137 SLP that did not participate in the interview. The SLPs filled out the questionnaire anonymously and gave their consent when they submitted the filled-out questionnaire. Most participants (n = 130) completed the questions about both diagnosis and intervention while seven SLPs indicated that their work does not comprise therapy and only answered the questions about diagnosis. The distribution of gender (see Table 1), region (North n = 10 (7.3%); West n = 64 (46.7%); East n = 36 (25.5%); South n = 27 (20.5%)) and workplace (see Table 1) was representative for SLPs in the Netherlands (Kwaliteitsregister, 2017).

Data collection

The questionnaire survey was administered online to make it easily accessible for all SLPs in the Netherlands and to enable the participants to respond at a self-chosen moment. The questionnaire was developed by the first two authors, taking into account the comments of six professionals in the field of SSD on a pilot version. The questionnaire (see Appendix 2) consisted of 27 questions distributed over three sections: background (8 questions), assessment process (10), and intervention approaches and additional aspects of the intervention (9). Information was elicited via either closed multiple choice questions (12), closed rating questions 5-point Likert scales ranging from strongly agree to strongly disagree) (11) or open-ended questions (13). The questionnaire was administered using Limesurvey© software (Schmitz, 2012) and took approximately 20 minutes to complete.

Data analysis

The open-ended questions were analysed thematically by the first author and checked by the second author, while the closed questions and multiple-choice questions were analysed by

Table 1. Demographic background of the SLPs that participated in the study (questionnaire *n* = 137, interview *n* = 33).

Demographic variable	No. of questionnaire Participants	% of questionnaire Participants	No. of interview Participants	% of interview Participants
Gender				
Female	135	98.5	32	97.0
Male	2	1.5	1	3.0
Region				
North	10	7.3	0	0.0
West	64	46.7	0	0.0
East	36	25.5	23	69.7
South	27	20.5	10	30.3
Years of experience				
0-5	20	14.6	5	15.2
6-10	23	16.8	7	21.2
11-15	19	13.9	4	12.1
16-25	42	30.7	11	33.3
25+	33	24.1	6	18.2
Work setting				
Private practice	97	70.8	29	87.9
Primary school	5	3.6	0	0.0
Primary school (Special Education)	10	7.3	4	12.1
Secondary school (Special Education)	7	5.1	0	0.0
Hospital	3	2.2	0	0.0
Specialist day care centre: young children	15	10.9	0	0.0
Work hours per week				
0-8	1	0.7	0	0.0
9-16	10	7.3	0	0.0
17-24	42	30.7	13	39.4
25-32	42	30.7	7	21.2
33-40	33	24.1	11	33.3
>40	9	6.6	0	0.0
				Missing 2
Degree				
MA	35	25.5	1	3.0
BA	102	74.5	32	97.0
Education course in SSD after graduation				
Yes	100	73.0	31	90.9
No	37	27.0	2	9.1
Number of courses in SSD after graduation*				
1	46	33.6	8	24.2
2	30	21.9	12	36.4
3	14	10.2	5	15.2
>4	9	9.1	5	15.2

*Most followed courses: Metaphon, Cycles approach and PROMPT means of descriptive frequencies. There were some missing data towards the end of the questionnaire from some of the participants, particularly the open-ended questions were not filled out by everyone (55.5% completed every question). Reported percentages are for valid data only, excluding the missing responses for the questions with missing values.

Results

First of all, the results from the questionnaire indicated that the age of the children in the caseload of the participating SLPs is dependent on the work setting. The children attending a private practice are generally between two and ten years old, with exceptional situations of older children up to 19 years of age. School based SLPs report seeing children between four and seven years of age. Furthermore, the responses indicated that most of the children below four years of age have a diagnosis of phonological disorder while most of the older children have the diagnosis phonetic articulation disorder. The results regarding the processes of diagnostics and planning and administering intervention are described in the following sections. To provide a complete overview of the current practice of the SLPs in these respects, the results from the questionnaire and the interviews are combined in the text. Note that throughout the paper, *n* is used to denote sample size (the total number SLPs that responded to the questionnaire/interview or to a specific question), whereas n denotes the number of response types (the number of specific answers to a question).

Diagnostic process

The participants reported that they take between two and four sessions (one session is 30 minutes in the Netherlands) to do a complete assessment, including a case history, before they diagnose a child. The number of sessions depended on the severity of the speech problems and the child's cooperation during the assessment. In the following sections, we will present the results on the different steps of the diagnostic process separately.

Case history: Manner of conducting and themes

Not all SLPs conduct a case history; this is the case for SLPs in schools, special day care centers or hospitals. Parents are often not present at the SLP sessions. "…*it is difficult to get all the information; it depends on the collaboration of the parents. I do not have the time to conduct a case history; my caseload is large."* (P5). SLPs who conduct a case history differ in four aspects of the case history: which questions they ask the parent or guardian, the presence or absence of the child during the intake, whether they combine an interview with an assessment during the first session or not. "…*I hear from some colleagues, that they do not conduct an interview, …and they start with an assessment. I never do an assessment the first time.*" (P30).

All responses to questions about the case history from the interviews were labeled and then clustered in nineteen items. Subsequently, these items were included in the questionnaire where the respondents were asked to select the five items that they considered the most important for a case history. The results indicated that most of the SLPs considered it important to ask the parent whether the child has hearing problems (n = 103), how the speech and language of the child developed (n = 94) and about the course of the speech disorder (n = 71; see Table 2).

Table 2. Most important questions in SLPs' case history as reported in the questionnaire (*n* = 137; SLPs had to choose five items).

Theme	n	Item	n
Hearing	103	Speech- and language disorders in the family	30
Speech-and language development	94	Reactions of the environment on intelligibility	28
Course of the disorder	71	Child's awareness of intelligibility	25
Reactions of the child when misunderstood	53	Multilingualism	25
Oral habits	44	Compensatory behaviour	24
Feeding development	35	Pregnancy and childbirth	13
Sensorimotor development	34	Psychosocial development	11
Babbling	33	Sensory perception	8
First word	33	Diseases	1
Cognitive development	31		

Observation: Manner of conducting and themes

To get an impression of what SLPs attend to during the case history and assessment, an open-ended question was included in the questionnaire inquiring what SLPs consider important to observe in a child with SSD, in general and in the speech-language domain specifically. The responses were clustered into 21 topics (Table 3). The SLPs mentioned that they observe the communication strategy of the child the most (n = 62), for example pointing and making gestures, followed by the oral skills of the child (n = 56). SLPs mentioned observation of the speech characteristics (e.g., phonological processes, speech sound distortions) 45 times.

Table 3. What SLPs observe in a child with SSD as reported in the questionnaire (*n* = 132).

Observation topics	n	Observation topics	n
Communication strategy of the child	62	Feeding development	12
Oral skills	56	Child awareness of the SSD	12
Speech characteristics	45	Groping	9
Interaction with the parent/caretakers/SLP	40	Cognitive skills	8
Coping with the speech problems	33	Auditory processing	8
Intelligibility	28	Pragmatic skills	5
Language development	25	Imitation	3
Hearing	22	Reading/writing development	3
Sensory and motor development	19	Motivation	2
Attention/focus	19	Rate of speech of the parents/caretakers	1
Reaction of the parents when they do not understand the child	14		

Assessments used by SLPs

In the interviews, the SLPs were asked which assessments they use for a suspected diagnosis and why they prefer those assessments for that specific SSD. An overview of the responses for the three most common speech problems in children, phonological disorder, phonetic articulation disorder and CAS, is presented below. The responses of the SLPs who participated in the questionnaire are presented in Table 4.

1. *Phonological disorder:* Nine SLPs reported to use the Hodson Assessment of Phonological Patterns (HAPP) (Hodson & Paden, 1983) because of the ability to calculate the severity of the disorder. This instrument is mostly used with children of four years of age or younger because it contains objects instead of pictures. Another reason to assess a child with HAPP is if the child has a severe speech disorder. One SLP (P1) stated to regret not being able to use HAPP with older children and to use the Metaphon screening (Leijdekker-Brinkman, 2005) instead for older children. Metaphon is also chosen for children with a severe disorder (n = 5). When the disorder is less severe, seven SLPs reported to prefer the Nederlands Articulatie Onderzoek (NAO, Baarda, de Boer-Jongsma, & Haasjes-Jongsma, 2014) (picture naming), which is a two-way scoring assessment (correct or false analysis). One of the SLPs indicated to also assess the non-speech movements of children with a phonological disorder.
2. *Phonetic articulation disorder*: Seven SLPs indicated to rather observe a child than conduct an assessment. One (P15) argued that she can see a lisp during a conversation with a child. When SLPs do an assessment, they use the NAO (n = 8) or HAPP (n = 3). Four SLPs stated that they assess the non-speech movements of children with a phonetic articulation disorder.
3. *Childhood Apraxia of Speech*: 17 SLPs mentioned how they assess children with a suspicion of CAS. Most of them reported to use the Dyspraxieprogramma (Erlings-van Deurse, Freriks,

Goudt-Bakker, & Meulen, 1993) based on The Nuffield Dyspraxia Programme, but not the whole assessment. As one SLP indicated, "*The whole program is too long and boring….*" (P7). The items from the Dyspraxieprogramma that are used the most are: repeating speech movements, diadochokinetic sequences, repeating long words, non-speech oral movement assessment, repeat a word five times and an observation of groping behavior.

Overall, three reasons for why an SLP uses an assessment were given frequently: (1) the assessment is in possession of the SLP, (2) the assessment is quick, easy to use and clear or (3) the SLP recently followed a course on the assessment. One SLP (P9) indicated, "*I have got all the other assessments in my cupboard, but I do not use them. It is time consuming, and an observation combined with a simple picture naming task, namely the NAO, and some good thinking gives me plenty of information*." Furthermore, the results on the questionnaire indicated that 37.6% of the SLPs (*n* = 132) always use the same assessment when assessing a child with SSD and that 40.8% choose an assessment that fits the child's speech problem based on information gained from the case history and/or observation.

Table 4 presents quantitative data of which assessment the SLPs use. The SLPs mentioned on average three assessments, with a range of one to six. Most of the selected assessments are on the level of body functions (ICF). SLPs named the NAO (Baarda et al., 2014) the most followed by Metaphon (Leijdekker-Brinkman, 2005) and the HAPP (Hodson & Paden, 1983). However, when asked which assessment they use the most, the most frequent answer was the NAO, but HAPP was answered more often than Metaphon. Six other speech production assessment were preferred more than six times. When an assessment was named less than six times, we gathered them in a single category (Other speech assessment). Forty SLPs reported to use a speech sample but only six SLPs stated that this assessment is their first choice. Furthermore, some of the SLPs (questionnaire, *n* = 30; interview *n* = 3) mentioned the use of a language assessment or the use of a language sample to investigate the speech problems (e.g. the Frog story). Two SLPs gave a reason for the use of a language assessment; P14 stated "*I want to get a clear understanding of the language development.*' While the other (P7) reported '*I use a language assessment to observe the speech of the child*."

Table 4. Speech assessments used by SLPs (*n* = 132) to examine SSD. Questionnaire outcomes are presented in the 2nd and 3rd column, comprising all the mentioned assessments (*n* = 133) and only the most used assessment (*n* = 126) respectively. Outcomes of the interviews (*n* = 33) are presented in columns 4 and 5. In the interviews, the SLPs were only asked to mention which assessment they use and for which suspicions of SSD. The last column presents the addressed level of the ICF (Body structure, Body functions, Activity or Participation).

Speech assessment	**Mentioned (questionnaire)**		**Most used (questionnaire)**		**Mentioned (interview)**		**Suspicion of a specific diagnosis (interview)**	**ICF Category addressed by the assessment**
	n	%	n	%	n	%		
Nederlands Articulatie Onderzoek (NAO) (Picture naming, two-way scoring)	100	75.8	61	46.2	27	81.8	Phonetic articulation disorder (mostly) Phonological disorder	Body functions
Metaphon	66	50.0	17	12.9	17	51.5	Phonetic articulation disorder Phonological disorder (mostly)	Body functions
Hodson Assessment of Phonological Patterns	41	31.1	29	22.0	18	54.5	Phonetic articulation disorder Phonological disorder (mostly)	Body functions
Speech sample	40	30.3	6	4.5	2	6.1	Phonetic articulation disorder (mostly) Phonological disorder Childhood Apraxia of Speech	Body functions; Activity
Dyspraxieprogramma	30	22.7	1	0.8	12	36.4	Childhood Apraxia of Speech	Body functions
Schlichtingtest (Language assessment)	19	14.4	0	0.0	3	9.1	Phonetic articulation disorder Phonological disorder Childhood Apraxia of Speech	Body functions

Oral motor assessments	18	13.6	0	0.0	4	12.1	Phonetic articulation disorder (mostly) Phonological disorder	Body functions
PROMPT	13	9.8	1	0.8	2	6.1	Phonetic articulation disorder (mostly) Phonological disorder Childhood Apraxia of Speech	Body functions
Taaltest Alle Kinderen®: klankarticulatie (Picture naming, two-way scoring)	11	8.3	1	0.8	4	12.1	Phonetic articulation disorder (mostly) Phonological disorder	Body functions
Self-made speech assessment	6	4.5	7	5.3	7	21.1	Phonetic articulation disorder (mostly) Phonological disorder	Body functions
Other speech assessments	13	9.8	2	1.5	3	9.1		Mostly Body Function, one assessment Participation (Intellibility in Context Scale-ICS-NL)
Other language assessments	11	8.3	1	0.8	0	0.0		Body functions
Hearing	3	2.3	0	0.0	1	3.0		Body functions
Sensory Profile	1	0.8	0	0.0	0	0.0		Body functions; Activity

2

During the interviews, the SLPs were also asked to give their view on the most optimal way to conduct an assessment. Sixteen SLPs responded that they would like to have an assessment that can be used for differential diagnosis. An additional eight SLPs reported that they want to have a tool that is fully computer-based to save time and make the process easier.

Differential diagnosis

In an open-ended question (questionnaire) the SLPs were asked to mention the diagnosis they formulate for children with SSD in order of frequency. SLPs mentioned a total number of 85 different diagnoses with an average of three per SLP. Many similarities were observed among the different diagnoses, for example “dyspraxia”, “verbal dyspraxia”, “CAS”. Therefore, the 85-mentioned labels were combined into seven categories (see Table 5) and these were used in the further analysis. In some cases, the SLP mentioned two different labels that were eventually combined in the same category, for example within the category *Phonological disorder/delay*, one SLP gave two different diagnoses: “*Phonological delay*” and “*Phonological disorder”,* this was counted only once. The category *Combination of diagnoses* contains diagnoses in combination with each other (for example phonological disorder with CAS). The most mentioned diagnosis is a phonological disorder/delay, followed by a phonetic articulation disorder (see Table 5).

The SLPs in the interviews reported similar terms to what was reported in the questionnaires (Table 5). All (100%) interviewed SLPs see children with a phonetic articulation disorder. Most (97%) of the SLPs who participated in the interview have children with phonological disorders in their caseload. The majority of the SLPs (69.7%) stated to diagnose children with CAS. However, most SLPs expressed that the children in their caseload do not have a pure CAS, but rather some characteristics of CAS in combination with a severe phonological disorder. As one participant said, “*Yes, a pure dyspraxia almost never occurs. It is frequently combined with a phonological problem*” (P24). Only one of the five SLPs with less than 5 years of experience diagnosed a child with CAS. Furthermore, only a small number of SLPs see children with dysarthria (9.1%), reportedly because “*The children with dysarthria are already in a special day-care centre*” (P16). In the private practices, only SLPs with more than 16 years of experience reported to have had children with dysarthria in their caseload. Finally, eight SLPs reported that their caseload also comprises children that stutter or clutter or children with a cleft palate.

Table 5. Diagnoses as reported by SLPs in the questionnaires (*n* = 132) categorized into groups, with distinctive characters of each diagnostic category based on the interviews (*n* = 33).

Speech sound disorder	No. of times reported (questionnaire)		Distinctive characters of the speech problem according to SLPs (interview)	ICF Category of the distinctive character
	n	%		
Phonological disorder/delay (33 different labels)	127	96.2	Simplification processes	Body functions
			Deletion of speech sounds	Body functions
			Child can pronounce the speech sound, but not in a syllable or word	Body functions
			Language problem	Other
			Structural and consistency in speech pattern	Body functions
			Often unintelligible speech	Activity
			Frustration	Participation
			No awareness of speech problem	Participation
			Hearing problems	Body structure Body functions
			Sensory processing problem	Body functions
Phonetic articulation disorder (23 different labels)	113	85.6	Lisp	Body functions
			Child can pronounce the sound, but does this in a wrong manner	Body functions
			Wrong tongue placement	Body functions
			Problems in muscle tone of lips and tongue	Body functions
			Abnormal dental position	Body structure
			Short frenulum	Body structure
			Substitution of just one speech sound	Body functions
			High gag reflex	Body functions
			Jaw instability	Body structure
Childhood Apraxia of Speech (CAS) (13 different labels)	59	44.7	Inconsistent speech pattern	Body functions
			Problems with repeatedly pronouncing a word	Body functions
			Groping	Body functions
			Frustration	Participation
			Unintelligible speech	Activity
			Difficulties with imitating a speech sound	Body functions
			Late talker	Other
			Feeding problems	Other
			Problems with coordinating of the lips, tongue, and jaw	Body functions
			More errors when pronouncing longer words or sentences	Body functions
Dysarthria (1 label)	6	4.5	Difficulties in forming a sound	Body functions
			Difficulties in intelligible pronunciation of sounds	Activity
			Fluctuating muscle tone	Body functions
			Hyper nasality	Body functions
Fluency disorders	11	8.3		
Combination of diagnosis	10	7.6		
Other diagnoses (not speech related)	8	6.1		

During the interviews, the SLPs were also asked how they recognise the different speech disorders. The responses are presented in Table 5. Following McLeod (2004), the reported distinctive characters were matched with an ICF level. Some of the distinctive characters could not be specified because the characters were too broadly formulated to fit into a single ICF level and were therefore labelled as *Other*. The characters of a phonetic articulation disorder as mentioned in this survey only regarded the level of body functions, whereas the reported characters of phonological disorder, CAS and dysarthria regarded all levels of the ICF.

Intervention

Factors that influence choice of the intervention

The results from the questionnaires indicated that predominantly child-related factors are important for an SLP when they choose an intervention method or material. Especially the extent to which an intervention is appealing to the child was reported to play a role in the choice of a specific intervention (49.3%). Furthermore, they also take into account as one of the main reasons age (37.3%) and specific diagnosis and severity of the disorder (31.3%).

Similar reasons were also mentioned during the interviews. In contrast to the questionnaire, however, the experience of the SLP was frequently expressed in the interviews (n = 17): "*What is your own experience, what works and what does not work…"* (P31). In addition, the behaviour of the child was only stated in the interviews (n = 19), probably because the SLPs were asked to provide a more comprehensive answer. "*…and shy children…you just keep in mind the behaviour*" (P23).

In both the interview and the questionnaire, the SLPs were also asked which intervention method or material they would choose given a specific SSD. Table 6 presents the outcomes of the questionnaire for the three diagnoses that were mentioned the most often, i.e., phonological disorder, phonetic articulation disorder, and CAS. With respect to the diagnosis dysarthria (that was mentioned only four times), SLPs reported to use PROMPT (Hayden, 2008; 75.0%) or Neuro Developmental Treatment (Howle, 2002; 25.0%). SLPs report to use almost every intervention for all three diagnoses; the differences lie in the frequencies. For example, the Cycles approach (Hodson & Paden, 1983) is commonly used with phonological disorder (n = 69), but not often with phonetic articulation disorder (n = 4) or CAS (n = 3). In the interviews, the SLPs indicated to use the same methods/materials per disorder. As was the case with respect to the diagnostic assessment, most of the used interventions address the level of body functions (Table 6).

In reply to follow-up questions about the chosen intervention, 62.9% of the SLPs (questionnaire) reported to follow the instructions of the method/material as described in the manual or handbook. Furthermore, the SLPs (*n* = 89) reported to be uncertain about their choice of intervention for children with CAS (34.8%), followed by children with a phonological disorder (28.2%). Again, the interviews sketched the same picture.

Table 6. Method/materials used per diagnosis as reported in the questionnaire (*n* = 91).

Method/Material	**Phonological disorder**		**Phonetic disorder**		**CAS**		**ICF Category addressed by the assessment**
	n	%	n	%	n	%	
Cycles approach	69	75.8	4	4.4	3	3.3	Body functions Activity
Metaphon(box)	70	76.9	7	7.7	4	4.4	Body functions
Dyspraxieprogramma	22	24.2	15	16.5	41	45.1	Body functions
Logo-art/ Widget Inprint *(picture database)*	42	46.2	53	58.2	12	13.2	Body functions
Oral myofunctional therapy/Garliner	2	2.2	31	34.1	1	1.1	Body functions Body structure
Van Riper	4	4.4	12	13.2			Body functions
PROMPT	12	13.2	14	15.4	7	7.7	Body functions Body structure
Self-made material	8	8.8	14	15.4			
Minimale parenspel *(minimal pairs game)*	10	11.0					Body functions
Transparant/TenT *(program to practice sentences with 3Dmaterials or pictures)*	2	2.2	3	3.3	1	1.1	Body functions

Story card/ Card sequence	1	1.1	1	1.1			Body functions
3Dmaterial/games	18	19.8	13	14.3	4	4.4	Body functions Activity
Phonological awareness	1	1.1					Body functions
Core vocabulary	1	1.1					Body functions
Sound gestures	3	3.3	3	3.3			Body functions
ToP Taalprogramma *(program with themes, for example, farm animals (pictures/story cards))*	1	1.1	1	1.1	1	1.1	Body Function
Psycholinguistic Orientated Phonological Therapy (P.O.P.T.)	1	1.1					Body functions
Mirror	2	2.2	4	4.4	2	2.2	Body functions
Tongbrekers *(cards with tongue twisters)*			1	1.1	1	1.1	Body functions
Therapy according to Golding-Kushner	1	1.1	3	3.3			Body functions Body structure
Oral motor exercises	1	1.1	12	13.2	2	2.2	Body functions
Drost			1	1.1			Body functions
Sensory integration			1	1.1	1	1.1	Body functions

The SLPs were also asked to explain which didactic strategies they use when administering the intervention. The responses from the questionnaire (n = 73) and the interviews (n = 33) combined for the three most common diagnoses (i.e., phonological disorder, phonetic articulation disorder, and CAS) are presented in Table 7. The results showed many similarities between the three disorders, except for the structure of the intervention. The SLPs reported that for children with a phonological disorder they start with practicing target words, whereas they start intervention with exercises on the sound level for children with phonetic articulation disorders or CAS. While children with CAS practice with two alternating sounds, children with a phonetic articulation disorder already practice sounds in words. The responses further indicated that SLPs use awareness and imitation as didactic strategies for children with phonological and phonetic articulation disorders, but not for children with CAS. For these children an SLP uses drilling and motor learning; these techniques are not used for other speech disorders.

When children are able to pronounce sounds in words and sentences, the SLPs reported to change the didactic strategy and allocate some time of the intervention session to stimulate the intelligibility in real life (Activity and Participation level). According to some SLPs, this is not included in the chosen intervention method. For example, P14 illustrated this by saying: "…*I use longer words, memory, and story picture cards. These exercises are not included in a method….I use these materials to stimulate natural speech*." Another SLP (P31) stated *"...exercises at home, for example, a conversation with dad or mum. A child can learn to execute what he has learned*". One SLP (P33) shared her uncertainty about the transition phase. "*In my practice the child performs well, then the child goes home, and we are back at square one.*"

Table 7**.** Didactic strategies for the three diagnoses as reported in the questionnaires (*n* = 73) and the interviews (*n* = 33) combined.

	Speech sound disorder	
Phonological disorder	**Phonetic articulation disorder**	**CAS**
Awareness of speech problem	Awareness of speech problem	
Tactile	Tactile	Tactile
Auditory exercises and bombardment	Auditory exercises	Auditory exercises
Visual	Visual	Visual
Imitation	Imitation	
Repetition	Repetition	Repetition
Playful	Playful	Playful
		Drill
		Motor learning
Structure: production of target words	Structure: (Auditory Discrimination), sound, word, sentence level and spontaneous speech	Structure: start with one sound, alternate two sounds, etc. Finally pronounce words and sentences

Factors influencing the effect of the therapy

During the interview, the SLPs were asked how they keep their intervention period short and effective. The most important factor according to the SLPs was whether parents stimulate their child at home. As some SLPs reported, therefore they try to actively involve parents in the intervention and give parents a clear description of the homework. SLPs in the questionnaire were further asked for their perspective on the role of parents during the intervention in an open-ended question. The responses were combined into six categories. Most SLPs consider parents as a co-therapist (89.5%). As one SLP (P14) wrote: "*Parents are co-therapist. It is not beneficial to only practice during a therapy session*." Furthermore, 88.7% of the SLPs (data from statements, Likert-scales) reported that they provide homework; specific exercises for parents and child to work on together. The responses further indicated that the provision of homework is mainly dependent on the work setting. Homework was reported to be more or less standard in private practices, but less common in specialist day care centres and schools. SLPs find it is also important to have a collaboration with the school or nursery of the child.

Another important effective intervention requirement was that the children should have fun during the intervention and practice by playing. Furthermore, the SLPs considered it important to pause the intervention when there is no visible progress. The intervention period was reported to be shorter and more effective when SMART goals (Specific, Measurable, Attainable, Realistic and Timely) are formulated at the start of the sessions and targets are adjusted if necessary. Goals are important to be formulated together with the parents, for example, P6 said: "*I always ask parents; What would you like to achieve in six months? Well, nine out of ten times they want to understand their child better*.". Other factors in choosing the goals for the therapy sessions are the capability of the child to pronounce sounds in isolation or even in a word and the normal speech development.

Additionally, the questionnaire contained an open-ended question about the steps the SLPs take when the intervention does not have any or just a little effect. This question was answered by 68 SLPs and the responses were combined into seven categories. The results showed that when the intervention has no or little effect most SLPs (86.7%) consult parents, teachers or a colleague and/or change the intervention method (58.8). Other practices reported were to refer the child to another SLP or another discipline (48.5%), to conduct an additional assessment in order to find a cause (36.8%), and to temporarily pause the intervention (35.5%). Nine SLPs (13.2%) stated that they would stop the intervention sessions altogether.

The last question of the interview was what SLPs would like to change or add to their options regarding intervention (*n*=21). The majority of the SLPs (*n*=14) responded that they would like to have one complete/combined method that they could use for all children. As one SLP pointed out: "*I would like to talk with influencers on SSD to create an ideal articulation intervention which represents all existing methods*" (P9). In addition, four SLPs reported the need for variable and challenging materials to motivate the children to practice and three SLPs expressed the wish for a computer-based intervention. For example, "*There are probably so many apps, but a homework app focused on speech might be a good idea?"* (P11).

Discussion

The purpose of the present study was to get a complete overview of the entire process SLPs go through in their practice as a professional, from the registration of a child with (suspected) SSD until dismissal. In a mixed-method design combining interviews and questionnaires, we surveyed the practices and opinions of a total number of 170 SLPs in several professional settings in the Netherlands. The results showed a strong congruence between the quantitative and the qualitative data. Before attending the outcomes regarding the intervention process, we will first discuss the process of how SLPs come to establish a diagnosis.

Diagnostic process

First of all, the results revealed a number of differences in the diagnostic process dependent on work setting. SLPs in private practices always conduct a case history to determine which topics are important for the specific child, whereas SLPs working in education, special day care centres or hospitals do not always conduct a case history. The absence of parents during school hours is a problem for SLPs in education or special day care centres and an extensive case history is often considered too time-consuming in hospitals. A recent study by Harrison et al. (2017) found that the judgement of parents is a valid and important tool in diagnosing speech problems. This stresses the need for all SLPs to conduct a case history, because it helps in establishing a diagnosis. In addition, we believe that conducting a (detailed) case history with a parent can save time on the longer run, because a better fitting assessment plan can be conducted if parents provide a detailed description of the child's (speech) problems. It also could provide important information to set the goals later in the intervention sessions.

With respect to the speech assessments that are used for diagnosis, the results first and foremost showed a large variety between individual SLPs, independent of work setting. SLPs in the Netherlands are known to use a variety of assessment tools Priester, Post and Goorhuis-Brouwer (2009), similar to assessment tool usage in other countries and languages (Joffe & Pring, 2008; McLeod & Baker, 2004, 2014; Skahan, Watson, & Lof, 2007). By far the most popular assessment amongst the SLPs was a naming task with pictures or 3D-materials to help evaluate the children's speech sound inventory. A spontaneous speech sample was often mentioned as well but was not often selected as the most frequent assessment option. With respect to the use of spontaneous speech samples, it is often questioned whether these contain enough information about all the different speech sounds (Eisenberg & Hitchcock, 2010). For a complete phonological analysis, a speech assessment should contain at least two opportunities for a child to utter each consonant in each word position including two-element and three element consonant clusters and SLPs may need to consider additional spontaneous speech samples (Macrae, 2017).

Several reasons were reported as to why SLPs choose a specific assessment. A frequently mentioned criterion was that the tool should be quick and easy to use. Joffe and Pring (2008) argued that SLPs have little interest in a more detailed assessment, possibly because of the time it involves. A naming task, which is used often, is fast and easy. However, Terband, Maassen and Maas (2019) state that an SLP needs to assess a child with several assessment instruments to differentiate between phonological and speech motor deficits, since this is not possible based on one single assessment. Malmenholt, Lomander and McAllister (2016) found that SLPs usually administer self-assembled assessment batteries for diagnosing a child with a suspicion of CAS. Our data shows that most SLPs use parts of the *Dyspraxieprogramma* (Erlings-van Deurse et al., 1993) to diagnose speech motor problems such as CAS, but in the interviews several SLPs reported to find differentiating CAS from other speech problems particularly difficult. Like Terband, Maassen and Maas (2019) argue, this is probably the reason why some SLPs use more than one assessment to differentiate between the different SSDs. Another reason why it is important to administer more than just a naming task is the need of additional information about the child's functioning in social situations (McLeod, Harrison, & McCormack, 2012). Most SLPs use observation to get information about the activity and participation level of the ICF (*Communication strategy of the child; Interaction with the parent/caretakers/SLP; Coping with the speech problems; Intelligibility)* and only a few SLPs use an assessment (ICS-NL;

McLeod, Harrison, & McCormack, 2013) to assess the impact of the intelligibility in everyday life.

In summary, three problems occur in diagnosing children with SSD in practice, (1) not every SLP conducts a case history, (2) SLPs rarely collect information at all the levels of the ICF, especially on participation in everyday life determined by parents/caregivers, and (3) SLPs often only use a picture-naming task. The SLPs in the present study reported that their main motives are time and ease of use. The relevant research literature clearly shows that the assessment process contains a diversity of tasks containing all levels of the ICF (including a questionnaire for the parents/caregivers of a child for example the ICS) and a comprehensive analysis of the speech sample. Therefore, we suggest the use of a fast and easy-to-administer comprehensive differential diagnostic instrument in combination with the ICS (McLeod, Harrison and McCormack, 2013) and a thorough case history and observation. The ICS is easy-to-administer; it adds the view of a parent/caregiver and it is an instrument to get information about the participation in everyday life of the child. In the questionnaire are questions about the comprehension of the speech by strangers, but also by a teacher. Parents reported children to be less intelligible for less familiar communication partners (Van Doornik, Gerrits, McLeod & Terband, 2018). Additionally, we suggest replacing the live case history with the possibility to use an online case history tool, to enable SLPs to further save time and collect the necessary information even if direct contact with parents/caregivers is not possible (e.g. school setting).

Labelling a diagnosis

In the questionnaire, the 137 SLPs reported a total number of 85 different labels in response to the question which diagnoses they formulate. This number is a factor 10-20 larger than the number of different disorders in current classification systems such as the MDD (Dodd, 2014) and the SDCS (Shriberg et al., 2017; Shriberg et al., 2010). However, only an average of three different diagnostic labels were mentioned per SLP. The 85 different labels mainly reflected naming idiosyncrasies of individual SLPs, and we could combine them into seven categories. There are similarities and differences between the labels used by the SLPs in this study and the two classification systems. If we compare our data with the MDD, all the diagnoses in this model appear in our data, although some disorders under different names. For example, the MDD label phonetic articulation disorder was named by the SLPs, but the term articulation was also named frequently. A second example is the use of one label for the group phonological disorder where the MDD uses the three labels phonological delay, consistent atypical phonological disorder and inconsistent phonological disorder. Although all these were also mentioned by the SLPs, in the Dutch version of the International Classification of Impairments, Disabilities, and Handicaps (ICIDH; Raaijmakers & Dekker, 1993) these labels are not common to use, only a phonological disorder is mentioned.

One diagnosis that appeared in our study, dysarthria, is not mentioned in the MDD (Dodd, 2014) and this is also where the MDD and the SDCS (Shriberg et al., 2017; Shriberg et al., 2010) differ. The SDCS also differentiates between typology and aetiology, unlike most classification systems (Shriberg et al., 2017). In this survey, we did not make distinctions based on typology in the sense that the SDCS does, for example by comparing the persistence of the speech problems resulting in different age groups (three to nine or older than nine). The typology used in our study was more of a characterization of the disorder. The SLPs in our survey also did not mention the aetiology in the descriptions of the different diagnostic labels although aetiology is covered in the case history (for example *hearing*).

Although the diagnostic labels of the MDD and SDCS, as well as the five groups that we chose in the present study, are widely used in the literature, the ICIDH and in education of SLPs, this apparently does not mean SLPs use these exact labels or groupings in daily practice. Our findings show that there is no consensus on terminology of the different SSDs and that there are a lot of idiosyncrasies in the diagnostic labels that are used. This makes it difficult to communicate among SLPs, let alone communicate well with other disciplines and parents. Although we did find some agreement in the descriptions of the different SSD labels, some of the features named by the SLPs were not specific for just one diagnostic label. For example, simplification processes was mentioned as a distinctive character of phonological disorder but can also indicate CAS (Terband, Maassen and Maas, 2019). How such specific errors are interpreted is highly dependent on the individual SLP and his/her personal background.

Intervention process

Our results indicate that most SLPs choose the intervention based on availability and own experience, rather than scientific evidence regarding treatment of the diagnosed deficit. Almost all the interventions mentioned by the SLPs are used for all three disorders (phonological, phonetic articulation disorders and CAS), the differences reside in how many SLPs use a specific intervention for the different disorders. There is no common ground in choosing an intervention.

Also, SLPs often combine interventions or adapt the intervention themselves while there is little scientific ground for this. For example: for children with CAS the *Dyspraxieprogramma* is often paired with the Cycles approach or Metaphon. There is some evidence of efficacy for an intervention based on speech motor skills and linguistics skills. Unfortunately, there is not much evidence about establishing maintenance and generalization of this combination in intervention (Murray, McCabe, & Ballard, 2014).

These results suggest there is a no consensus on which intervention to use for a particular child with a specific SSD among SLPs. The fact that SLPs combine interventions does not in itself have to be a problem. The SLPs have their reasons and they see children progress during the sessions. We recommend gathering information about these combinations and to investigate the effectiveness of these combination further.

Non-speech oral motor movements

About 10% of the SLPs reported non-speech oral motor movements (NSOM) as part of the diagnostic process (observation, case history and assessment) as well as the intervention process. The reason to discuss NSOM here separately is their controversial status in the field of speech disorders. For example, Ziegler (2003) claimed that impaired speech and non-speech movements should be kept separate and recently, also Maas (2016) argued that speech is only speech when all components involved in speech are present. NSOM thus might not be relevant to observe or examine and to address as part of an SSD intervention. Our present findings indicate that SLPs use non-speech oral motor exercises (NSOMEs) in the intervention process for all diagnostic groups, similar what has been observed in other countries (Brumbaugh & Smit, 2013; Joffe & Pring, 2008; Rumbach, Rose, & Bomford, 2016; Ruscello, 2008; Watson & Lof, 2009). In their recent review, Lee and Gibbon (2015) concluded that there is no strong evidence that NSOMEs are an effective treatment or addition to the intervention of children with phonological speech disorder. Similarly, Pennington, et al. (2016) found only three studies in which nonspeech exercises were examined for children with dysarthria and none of these showed any improvement. The lack of strong evidence for the effectiveness of NSOMEs should have implications for professionals in making decisions in relation to the intervention plan.

Parent involvement

Every SLP in our survey involves parents at a given moment during the guidance of children with SSD. How and how often, however, is dependent on the work setting. SLPs in schools or day-care facilities have less opportunity to work with parents closely. Similarly, an Australian survey found that SLPs working in a school setting were less likely to have a parent present in their intervention sessions (Pappas, McLeod, McAllister, & McKinnon, 2008). Working with parents is a necessity for all SLPs at some point. For example, it is not possible to conduct a case history without the help of a parent (or caregiver) and every SLP in our study conducts a case history somewhere in the process.

After the case history, the amount of involvement of the parent is variable, for example during the assessments the parents are more in the background. However, SLPs involve parents in choosing the goals and the type of intervention, which is also reported in other surveys regarding clinical reasoning in the UK, Portugal, Australia and China (Joffe & Pring, 2008; Oliveira, Lousada, & Jesus, 2015; Sugden, Baker, Munro, Williams, & Trivette, 2017; To, Law, & Cheung, 2012). Similar to what Sugden et al. (2017) concluded in their survey among Australian SLPs, the SLPs in our survey further reported that parents were also involved in stimulating the child during the intervention period. More than half of the SLPs indicated to involve the parents in the home practice exercises as part of the intervention. Parents are expected and invited to be co-therapist, and to aid in improving their child's speech by correcting and modelling. The reasons the SLPs gave for involving parents during the intervention was to ensure an effective and short intervention period.

Even though the scope of our survey was broader than most of the previous studies (Joffe & Pring, 2008; Oliveira et al., 2015; Sugden et al., 2017; To et al., 2012), which describe only involvement of parents for children with phonological disorders, the results are comparable. Nevertheless, it would be interesting to ask SLPs more specifically about the involvement of parents in intervention for different diagnostic groups. Like Sugden et al. (2017) argue, an insight into the view of parents would also be interesting, so that SLPs could better match the wishes and possibilities of parents to the intervention involvement.

Limitations

In the present study, the participating SLPs were recruited through an advert on social media and in a newsletter from the professional association of the Netherlands. The sample of SLPs included in the questionnaire survey that responded to the advert included a somewhat high percentage of SLPs with a master's degree (25.5 %), which might not be representative for the whole profession. However, we found a great deal of similarities between the data of the questionnaire with the data of the interviews. The latter had a smaller percentage of SLPs with a master's degree (3.0%). During the interview process, we reached a point of saturation. It is possible that there is a bias, and that the SLPs who participated all happened to have a particular style in diagnosing and treating children with SSD, but the variance in the answers showed this cannot be the case. Additionally, the results of the questionnaire and the interviews were similar. This means the results are likely to be reliable for this sample of SLPs.

Future research

In contrast to most recent surveys, the present study featured not only a questionnaire, but also an interview as suggested by McLeod and Baker (2014). This combination enabled us to get a clear idea of the clinical reasoning of SLPs, why they choose a particular assessment, diagnosis and an intervention for an individual child. However, this type of study does not provide insight into the quality of those decisions. Possibilities for future studies would be to observe SLPs in their clinical decision making as proposed by McLeod and Baker (2014) or to survey the dossiers of children with SSD.

The data in this study showed a variety of diagnostic labels (n = 85) that are used by SLPs in daily practice, indicating a lack of clear common ground for differential diagnosis and a lack of lack of agreement about identifying and the terminology. A terminological debate about SSD in the Netherlands or maybe even worldwide thus seems to be warranted. The CATALISE study of Bishop et al. (2016), designed to address similar problems in the field of developmental language disorders, could serve as both inspiration and example for such an exercise.

Conclusions

Overall, the results of this study indicate that there is no consensus on terminology in the field of SSD clinical practice. This can influence access to services and hinder practice as well as research (Bishop, Snowling, Thompson, & Greenhalgh, 2016). It also seems that most of the SLPs choose an assessment and intervention based on the availability of materials or their own experience. Unfortunately, this might not be the best option for that particular child. At the present, however, this might be difficult because a universally agreed-upon classification system with diagnostic markers for every SSD is lacking (Waring & Knight, 2013). In addition, our findings indicate that there is a need among SLPs for a fast and easy-to-administer comprehensive differential diagnostic instrument, to enable SLPs to further save time and collect the necessary information even if direct contact with parents/caregivers is not possible (e.g. school setting).

Our findings further revealed that there are no clear directions for adapting the intervention based on the characteristics of individual children. SLPs rely on their own clinical experience, and at the present simply have no other option. There is a pressing need for research to establish which intervention (or combinations of interventions) is most effective for particular diagnoses, as well as for specific groups of children.

It will take a while before we have a solution for these problems. In the short term, the authors advise to improve education for SLP students and SLPs, so that SLPs are well equipped to consider all different aspects of choosing a diagnostic instrument and intervention.

Acknowledgements

We gratefully thank all the participating SLPs and also, we want to thank the SLP students who participated in the data collection.

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Appendix 1:

Interview guide which was used in the interviews of the SLPs. 'I' in this context is the interviewer, you is the interviewed SLP.

Background questions	
Questions about background, work, career	I would like to get an impression of your work and experience as a speech and language therapist. When did you graduate as a speech and language therapist? Can you tell us something about your career? Can you briefly tell us about this practice / school / institution? Do you have multiple workplaces? Have you ever worked in a different setting or settings?
Work experience and courses for children with speech sound disorders	How long have you been working with children with a speech sound disorder? How many children with a speech sound disorder do you treat on average per week? What kind of speech problems do the children have? Did you follow a course on speech sound disorders? Which?
Age of the children	How old are the children? (age minimum to maximum)
Diagnostic process	
Introduction	I am curious about the way of making choices when diagnosing a child with a speech sound disorder
Process	Suppose you are being called by a mother who says her child does not speak well. OR Imagine, a new child is registered who does not speak well. What are the steps you take until the child leaves your practice again? Is this process the same for every speech sound disorder?
Differentiation (try to elicit that the speech and language therapist tells something about the several speech disorders, but rather not name the disorders yourself)	Which speech sound disorders do you diagnose? How do you differentiate between the different speech sound disorders? Does it ever happen that you have doubts between certain speech sound disorders? How do you handle this? Does it happen that children have multiple speech problems? How do you determine the diagnosis? What information parents give you, gives you an idea of a certain speech problem?

	Do you use assessments during the diagnostic process? If yes which one and why? You have already told about .... and .... disorders. Do you see children with other speech sound disorders? Do you really see no other children than those with .... and ...... disorders?
Factors	Which factors play a role in diagnosing a speech sound disorder? - For example: personal factors, external factors, participation level, activities level etc.
Dream question	What should be the ideal situation when diagnosing a child with a speech sound disorder? What is your dream?
Intervention process	
Introduction	Now I am curious about which choices you make when a child with a speech sound disorder receives an intervention.
Process	Suppose you have a diagnosis. What do you do next? - What do you think about before you start the intervention? - If you set goals, what do you take into account? If children have multiple speech sound disorders, which disorder will you first set goals for? Which choices do you make? - Why? Can you give an example? How do you decide which intervention or approach is best for a child?
Methodology (try to elicit that the speech and language therapist tells something about the intervention, but rather not name the intervention yourself)	Could you indicate for each speech sound disorder what kind of intervention / approach you use? (name here which speech sound problems the speech and language therapist has already mentioned) What plays a role in the choice of the intervention? Are there certain interventions that you use the most and why? Do you ever use multiple methods or different ways? - How do you use these different methods during the intervention?
Didactics (try to elicit that the speech and language therapist tells something about the approaches, but rather not name the approaches yourself)	How do you use an intervention? (ask about every intervention that is mentioned) Which didactic approach do you use with the intervention? (ask about every method that is mentioned) Do you give homework? Which homework do you give to children with speech sound disorders? What instructions do you give the parents / guardians about the homework?
Factors	Which factors play a role in the intervention of children with speech sound disorders?

Dream question	What should be the ideal situation for the intervention process? What is your dream?
Closing	
Closing the interview	Would you like to add something to this conversation?

Appendix 2:

The questions were published via Limesurvey©. Some questions arose from earlier answers given by the SLP. This is indicated in the questions below.

Background (8 questions)

1. What is your gender?
    a. Female
    b. Male
2. In which province do you work?
    a. Drenthe
    b. Gelderland
    c. Groningen
    d. Flevoland
    e. Friesland
    f. Limburg
    g. Noord-Brabant
    h. Noord-Holland
    i. Overijssel
    j. Utrecht
    k. Zeeland
    l. Zuid-Holland
3. When did you graduate as an SLT?
    a. 0 -5 years ago
    b. 5 -10 years ago
    c. 10-15 years ago
    d. 15-25 years ago
    e. More than 25 years ago
4. Did you complete another education course as well?
    a. No
    b. Yes, namely:
5. Did you take a post-graduate course on speech sound disorders for children?
    a. No
    b. Yes, namely:
6. In which setting do you work with children with speech problems? Multiple answers possible.
    a. Private practice
    b. Primary school
    c. Primary school (Special Education)
    d. Secundary school (Special Education)
    e. Hospital
    f. Specialist day care centre: young children
7. How many hours per week do you work as an SLT?
    a. 0 - 8
    b. 9 - 16
    c. 17 - 24
    d. 25 - 32
    e. 33 - 40
    f. More than 40
8. What is your role in guiding children with speech problems?
    a. Only diagnostics
    b. Only therapy
    c. Diagnostics and therapy

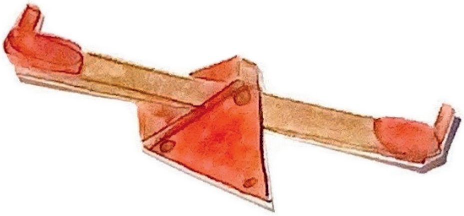
Assessment process (10)

9. What is usually the course of the diagnostic process? (example first a case history, then ...) *(open ended question)*
10. Which components do you find important to be discussed in the case history in children with speech problems?
    a. Pregnancy and child birth
    b. Babbling
    c. Speech- and language development
    d. Multilingualism
    e. Course of the speech disorder
    f. Child's awareness of intelligibility
    g. Reactions of the environment on intelligibility
    h. Reactions of the child when misunderstood
    i. Compensatory behaviour
    j. Speech- and language disorders in the family
    k. Feeding development
    l. Oral habits
    m. Sensory perception
    n. Cognitive development
    o. Psychosocial development
    p. Hearing
    q. Diseases
    r. Medication
11. What do you find important to observe in a child with a speech problem, both in general functioning as in speech-language? *(open ended question)*
12. Which assessment(s) do you use to diagnose a speech sound disorder in children? (multiple answers possible) *(open ended question)*
13. Which assessment do you use most often? *(open ended question)*
14. Why do you use this assessment most often? *(open ended question)*
15. Which speech problems in children do you diagnose? Name them in the order in which you most often state them. (multiple answers possible) *(open ended question, the answers of this question will be used in question 16, 18, 19, 22, 23)*
16. What characteristics or combination of characteristics (from assessment data) make that you choose the following diagnosis (s)? (more than one characteristic per diagnosis is possible) *(open ended question)*
17. Here are a number of statements. Do you want to indicate to what extent you agree with the statement? (scale: strongly agree, agree, disagree / disagree, disagree, strongly disagree)
    a. I always use the same assessment with every child with a suspicion of a speech sound disorder.
    b. I have sufficient possibilities (eg assessment material) to diagnose a child with a speech sound disorder.
    c. I use the ICF during the diagnostic process.
    d. I have sufficient knowledge to diagnose a child with a speech sound disorder.
    e. When I have diagnosed a speech sound disorder the diagnoses does not change during the intervention process.
    f. A child with a speech sound disorder can have multiple speech diagnoses.
    g. I sometimes delay the diagnosis until I have given a number of intervention sessions. This is mainly in children with ...
18. Would you like to complete the following statement?
    a. I feel confident when diagnosing children with ... (multiple answers possible)
    b. I am insecure when diagnosing children with ...... (multiple answers possible)

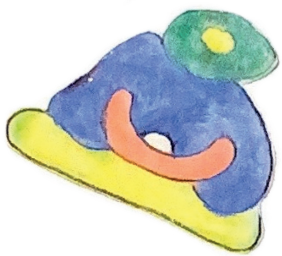
Intervention approaches and additional aspects of the intervention (9)

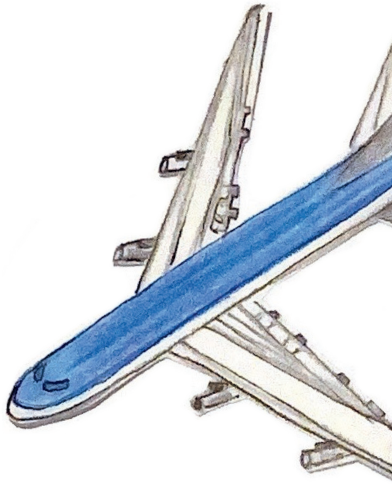
19. Which interventions or methods do you use with children with the following speech sound disorder? *(open ended question)*

20. What are important reasons for your choice of a specific intervention for a certain child? *(open ended question)*
21. Here are a number of statements. Do you want to indicate to what extent you agree with the statement? (scale: strongly agree, agree, disagree / disagree, disagree, strongly disagree)
    a. I exactly follow the description / instruction of the intervention.
    b. The environmental factors of the child play a part in the choice of the intervention.
    c. The personal factors of the child play a part in the choice of the intervention.
    d. I always give homework.
22. Would you like to complete the following statement?
    a. I feel confident when choosing an intervention for children with ... (multiple answers possible)
    b. I am insecure when choosing an intervention for children with ...... (multiple answers possible)
23. Can you briefly characterize the structure of the intervention for children with a .....? *(open ended question)*
24. What is the role of parents during the intervention? *(open ended question)*
25. How do you ensure that the child also applies what he has learned during the intervention in daily life? *(open ended question)*
26. What do you do if no progress is visible? (multiple answers possible) *(open ended question)*
27. Do you think there is enough material on the market for the intervention of children with speech sound disorders?
    a. Yes
    b. No, what do you wish?



CHAPTER 3





A standardized protocol for Maximum Repetition Rate assessment in children

Sanne Diepeveen, Leenke van Haaften, Hayo Terband,
Bert de Swart, Ben Maassen

Published in Folia Phoniatrica Logopaedica 2019;71(5-6):238-50

Abstract

Background/Aims: Maximum repetition rate (MRR) is often-used in the assessment of speech-motor performance in older children and adults. The present study aimed to evaluate a standardized protocol for MRR-assessment in young children for Dutch.

Methods: The sample included 1,524 children of 2-7 years-old with no hearing difficulties and Dutch spoken in their nursery or primary school and was representative for children in the Netherlands. The MRR protocol featured mono-, tri- and bisyllabic sequences and was computer-implemented to maximize standardization.

Results: <50% of the two-year-olds could produce >1 monosyllabic sequence correctly. Children who could not correctly produce ≥2 monosyllabic sequences could not produce any of the multisyllabic sequences. The effect of instruction ("faster" and "as fast as possible") was small and multiple attempts yielded a faster MRR in only 20% of the cases. MRR's did not show clinically relevant differences when calculated over different numbers of repeated syllables.

Conclusions: The MRR-protocol is suitable for children of three years and older. If children cannot produce at least two of the monosyllabic sequences, the multi-syllabic tasks should be omitted. Furthermore, all fast attempts of each sequence should be analysed to determine the fastest MRR.

Introduction

Diagnosing a child with motor speech disorders (MSD, for example childhood apraxia of speech (CAS), developmental dysartria) is not always an easy task for a speech-language pathologist (SLP). Children with MSD form a heterogeneous group due to etiological factors, but also due to individual differences in the pattern of development of the speech disorder leading to individual differences in speech errors (Shriberg et al., 2017b). Because of these differences, it is difficult to assess the underlying deficits (issues in speech-motor planning, programming, or execution) in children with MSD. In clinical practice, underlying deficits are often examined separately, but multiple factors may be involved (Dodd & McIntosh, 2008). This is why the precise identification of MSD in many children is difficult, especially for those who do not have an evident primary problem (Rvachew et al., 2005). Nevertheless, the precise identification of MSD is important for a correct diagnosis and treatment planning. If the intervention is better adapted to the individual diagnosis, this will ensure better progress (Rvachew et al., 2005), which can lead to great benefits for further speech and language development of the child.

A misdiagnosis or under-diagnosis can occur because SLPs simply rely on a diagnostic checklist for identifying MSD. These lists are often not specific enough to distinguish between the possible different underlying deficits. At the moment there is no specific test protocol for diagnosing children with MSD (Shriberg, 2003). Recently, Shriberg, et al., (2017a) proposed to use a Pause marker to identify children with childhood apraxia of speech (CAS). This could be helpful, but there is a need for a gold standard for all children with MSD. Furthermore, there are only a few objective instruments for mapping children's motor speech skills and there is no norm referenced assessment based on a large dataset. This is a major problem, as a good understanding of normal speech is necessary for the interpretation of MSDs (Wong et al., 2011). Finally, the existing assessments may be hard for children to complete, and it also demands a lot of the SLPs judgement ability. The outcomes therefore may not represent the children’s true abilities (McCauley & Strand, 2008).

However, an often-used objective assessment for the clinical judgment of the motor speech performance of older children and adults is the maximum repetition rate (MRR) (Icht & Ben-David, 2014; Rvachew & Brosseau-Lapré, 2012; Wang et al., 2008; Yang et al., 2011). The MRR frequently contains two types of stimuli: repetition of monosyllables (papapa) and of multiple syllable sequences (pataka) (Rvachew & Brosseau-Lapré, 2012). MRR is also called diadochokinetic (DDK) and both terms are used in the literature. We choose the term MRR instead of diadochokinetic (DDK), There is much debate about using meaningful (e.g. “*patticake”* or “*pat-a-cake*”) or non-meaningful stimuli (e.g. “*pataka*”), however, Williams and Stackhouse (2000) concluded that it is desirable to use non-meaningful stimuli to assure that the children’s performance is not influenced by their linguistic abilities. Furthermore, the MRR contains often the consonants /p/, /t/ and /k/ in a sequence (Kent et al., 1987), such that the three major articular organs are examined, namely the lips, the jaw and the tongue (Thoonen et al., 1996). Thus, the different consonants represent multiple levels of physiological complexity since each consonant has a different place of articulation and age of acquisition. These consonants cannot be produced in isolation in succession, which is why the consonants are combined with a vowel (Kent et al., 1987). Thus, the syllables /pa/, /ta / and /ka/ were used in several studies (Icht & Ben-David, 2014; Rvachew et al., 2005; Thoonen et al., 1996). MRR-protocols typically consist of multiple components, which increase in complexity. First, the child should repeat the monosyllabic sequences /papa../, /tata../ and /kaka../. Second, bisyllabic sequences such as /pata../ and /taka../ are administered. The MRR ends with the repetition of the trisyllabic sequences /pataka../ (Thoonen et al., 1996). During the assessment of MRR children are asked to repeat the different sequences in one breath at the highest possible pace. The sequences are meant to be repeated without errors and interruptions (Thoonen et al., 1996). Many children struggle with the unnatural situation of the MRR, which requires a specific approach with regard to instruction and practice opportunities (Wit et al., 1993). The MRR appears to be difficult for younger children, who make relatively more articulation errors during MRR-tasks as compared to conversational speech (Yaruss & Logan, 2002). Williams and Stackhouse (2000) found in their study of 30 typically-developing children aged three to five that the MRR was more sensitive when the score was based on accuracy and consistency instead of rate of the productions. However, Yaruss and Logan (2002) found no age-related increase in such MRR-accuracy and consistency scores. Overall, young children show much more variability in their performance than older children, such that the timing, speed and fluency of speech movements become less variable when children get older (Juste et al., 2012). Several studies showed that children make fewer mistakes during the performance of monosyllabic sequences compared to the multisyllabic sequences (Vance et al., 2005) and the rate of the sequences decreases as the task becomes more complex (Padovani et al., 2009; Shriberg et al., 2009).

Measuring MRR

The MRR used to be measured perceptually without any support of instrumental methods that can visualize the acoustic waveform. However, Gadesmann and Miller (2008) noted that the use of only perceptual evaluation is not acceptable for clinical diagnosis because perceptual measurement is not accurate enough. Nowadays, there are several programs that semi-automatically interpret the various MRR results. Some examples of these types of programs are the Diadochokinetic Rate Analysis, which is part of the Motor Speech Profile Model (Milenkovic, 2001), TOCS+ MPT Recorder™ (Hodge & Gotzke, 2011) and Praat (Boersma & Weenink, 2016). Although in these programs the task of the examiner is reduced to simply counting syllables, difficulties still occur when the speaker repeats the syllables quickly and irregularly. In this case, the individual syllables are too close together, which makes it difficult to detect the syllable boundaries which will influence the reliability of the value.

There is no uniform method of measuring the MRR, which makes it difficult to compare the results of different children worldwide. There are three methods being used: (1) counting syllables repeated in a certain amount of time (count by time), (2) measuring the time needed to repeat a given number of syllables (time by count), or (3) assess how many syllables can be produced in one breath (Gadesmann & Miller, 2008). As a consequence, there is large variability with respect to the collected norm data, which in its turn leads to difficulties with the interpretation of the MRR results (McCauley & Strand, 2008; Williams & Stackhouse, 2000).

Clinical use of the MRR

The MRR-performance of children with MSD differs compared to typically-developing children. Authors (Lewis et al., 2004; Thoonen et al., 1996, 1999; Wit et al., 1993) of four separate studies concluded that children with MSD (spastic dysarthria and CAS), differ in their performance on the MRR. More recently, Murray and coworkers (2015) advised to use an oral motor assessment to diagnose CAS, which includes the trisyllabic sequence: /pataka/, and polysyllabic word accuracy to diagnose CAS. The mentioned authors concluded that the MRR is a valuable tool in the differential diagnosis of underlying speech motor deficits, which is supported by the differences in MRR-performances between children diagnosed with dysarthria (Wit et al., 1993) and apraxia of speech (Thoonen et al., 1996) as compared to controls. Others dispute this because they did not find such differences between children with a typical development and children with MSD (Bradford & Dodd, 1996; Ozanne, 2005). Our opinion is that, although MRR does not necessarily reflect the primary speech disorder in all cases with SSD, MRR can play a role in the differential diagnosis to assess disorders in underlying articulo-motor planning and programming (Maassen & Terband, 2015; Rvachew et al., 2005). Interpreting only performance on the

MRR task is insufficient to assess the underlying speech problem; this requires multiple tasks and the assessment of a comprehensive speech profile (Maassen & Terband, 2015). Thus, in a large validation study, we assessed performances on the MRR as well as other speech tasks (picture naming, nonword imitation, word and nonword repetition) with the recently developed diagnostic instrument, Computer Articulation Instrument (CAI) (Maassen et al., 2019) - in a group of 1,524 typically-developing children. Factor analyses on the task performances showed separate factors for each of the four tasks (Van Haaften et al., 2017). The diagnostic value of these norm data resides in now being able to compare MRR performance of children with MSD to typical development.

As discussed above, SLPs use the MRR as an assessment tool for children with MSD. To date, available norm data are based on small samples of children, especially in the younger age groups (2-5 years old), and there is still much debate on the manner to conduct the MRR and the method to calculate the MRR. The aim of this study was to optimize a standardized protocol - which was based on previous studies in the Dutch language (Thoonen et al., 1996, 1999; Wit et al., 1993). Other research questions of the present study were: are children aged two to seven years able to perform the MRR-task, and what kind of instruction and how many attempts do children need to produce their fastest sequence; during clinical work we noticed some children were slower after the instruction to go as fast as possible.

Materials and Method

Participants

This study was part of a large normative study of a new Dutch instrument, the Computer Articulation Instrument (CAI), to assess children's speech problems (for more details see Maassen et al. (2019)). 1,524 Dutch-speaking children in the Netherlands were recruited between January 2008 and April 2015. The children were recruited via nurseries (47) and mainstream primary schools (71) in the Netherlands. The sample was representative for gender, urbanization, and geographic region. Inclusion criteria were as follows: (1) no hearing difficulties; (2) the Dutch language was spoken in the nursery or primary school. Table 1 shows the number of subjects per MRR sequence per age group (14 age groups were selected) and gender of the children. Not all children executed all MRR–tasks. Furthermore, in some cases the audio files were damaged due to technical problems, or the individual syllables were not recognisable because of background noise and the recordings were excluded from the sample.

Table 1. Age and gender for the 14 age groups of the normative sample.

Age group (years;months)	Mean age (years;months)	Total number of subjects	Gender		Number of subjects per sequence					
			Boys	Girls	/pa/	/ta/	/ka/	/pataka/	/pata/	/taka/
2;0-2;3	2;1	72	30	42	59	59	58	55	57	52
2;4-2;7	2;5	102	55	47	79	81	80	70	77	66
2;8-2;11	2;9	101	46	55	83	82	81	71	81	69
3;0-3;3	3;1	104	52	52	90	90	88	83	89	78
3;4-3;7	3;5	110	61	49	90	92	94	89	94	83
3;8-3;11	3;9	102	57	45	95	95	94	86	94	90
4;0-4;3	4;1	100	55	45	85	84	81	80	83	83
4;4-4;7	4;5	115	60	55	93	94	94	89	95	91
4;8-4;11	4;9	116	56	60	94	94	94	91	92	90
5;0-5;3	5;1	121	66	55	104	106	106	103	104	103
5;4-5;7	5;5	128	71	57	113	111	114	109	112	114
5;8-5;11	5;9	117	64	53	103	105	104	101	102	104
6;0-6;5	6;2	117	69	48	107	108	107	104	108	106
6;6-6;11	6;8	119	57	62	108	108	109	108	109	109
Total		1524	799	725	1303	1309	1304	1239	1297	1238
% sample		100%	52.4%	47.6%	84.5%	84.9%	84.6%	80.4%	84.1%	80.3%

The children were randomly selected from a list based on age group and gender. All parents/caregivers of the randomly selected children were asked for permission via an informed-consent letter to include their child anonymously in this large study. If parents gave permission, they filled out a short questionnaire containing questions about the speech and language development, multilingualism and health condition (e.g. loss of hearing) of the child. The protocol has been assessed by an ethics committee (Radboud University Nijmegen Medical Centre) and the study could be carried out.

Data collection

The children were seen individually by two SLP students or one SLP. In total, 14 SLPs assessed the younger children (2 to 4 years of age) and 110 SLP students administered the CAI for the older children (4 to 7 years of age). All these research-assistants were trained to assess children with the CAI by the first two authors and a precise instruction in the form of a guideline was given. To assure a flawless administration of the CAI, students worked in pairs.

An assessment session with a child contained the four tasks of the Computer Articulation Instrument (CAI): Picture naming (PN), Nonword imitation (NWI), Word- and Nonword repetition (WR, NWR) and Maximum Repetition Rate (MRR) (Maassen et al., 2019; Van Haaften et al., 2017). The assessment was administered at the child's nursery or primary school in a quiet room (or the room with the least possible background noise). The CAI was administered using a laptop and the acoustic signal was automatically stored on the computer's hard disk in one recording for each of the different tasks. The child and research-assistant were seated in front of the computer next to each other with a microphone and both wore a headset, or speakers were present, to provide a good sound level of the automated instruction of the CAI. The whole CAI would take approximately 30 minutes with the MRR being the last section of the CAI. The administration of the MRR took about five to ten minutes per child.

MRR Administration

A protocol (Table 2) for the assessment of the MRR-task was developed based on previous studies in Dutch and other languages (Modolo et al., 2010; Thoonen et al., 1996; Wit et al., 1993). The instruction was given by the CAI computer program to maximize standardization and the children were asked to imitate the following sequences: first three monosyllabic sequences (/papa../, /tata../ and /kaka../), followed by one trisyllabic sequence (/pataka.../) and finally two bisyllabic sequences (/pata../ and /taka../). First, the children were asked to repeat a short sequence of three syllables (e.g. /papapa/) in a normal speaking rate, followed by a longer sequence of six syllables in a normal rate (e.g. /papapapapapa/). The next instruction included imitation of a series of several syllables at a faster rate (the audio example contained 12 syllables at a faster rate). Finally, the children were asked to produce the syllable sequences as fast as possible. If necessary, the child got three attempts for every sequence (the CAI was programmed a maximum of three attempts per sequence) to collect an accurate or faster repetition of the sequence; the third attempt was given if the first two were both incorrect or the research-assistant had the impression that the child could produce a faster rate.

If a child refused to utter a sequence, the research-assistant tried to motivate the child and the sequence would be repeated or the research-assistant presented the next sequence. If the child kept on refusing during the next sequences, the session was ended.

Table 2. Assessment protocol for the MRR.

	Sequence	Trial	Instruction
1	pa	papapa	Sequence of three syllables, normal speech rate.
		pa…6x	Sequence of six syllables, normal speech rate.
		pa…12x	After an audio example, a faster speech rate (five syllables per second), sequence of twelve syllables
		pa…≥ 9x	As fast as possible (without an example) a sequence of minimal nine syllables.
2	ta	tatata	Sequence of three syllables, normal speech rate.
		ta…6x	Sequence of six syllables, normal speech rate.
		ta…12x	After an audio example, a faster speech rate (five syllables per second), sequence of twelve syllables
		ta…≥ 9x	As fast as possible (without an example) a sequence of minimal nine syllables.
3	ka	kakaka	Sequence of three syllables, normal speech rate.
		ka…6x	Sequence of six syllables, normal speech rate.
		ka…12x	After an audio example, a faster speech rate (five syllables per second), sequence of twelve syllables
		ka… ≥ 9x	As fast as possible (without an example) a sequence of minimal nine syllables.
4	pataka	pataka	Sequence of three syllables, normal speech rate.
		pataka…4x	Sequence of twelve syllables, normal speech rate.
		pataka…4x	After an audio example, a faster speech rate (five syllables per second), sequence of twelve syllables
		pataka…≥ 3x	As fast as possible (without an example) a sequence of minimal nine syllables.
5	pata	pata	Sequence of two syllables, normal speech rate.
		pata…3x	Sequence of six syllables, normal speech rate.
		pata…6x	After an audio example, a faster speech rate (five syllables per second), sequence of twelve syllables.
		pata…≥ 4x	As fast as possible (without an example) a sequence of minimal eight syllables.
6	taka	taka	Sequence of two syllables, normal speech rate.
		taka…3x	Sequence of six syllables, normal speech rate.
		taka…6x	After an audio example, a faster speech rate (five syllables per second), sequence of twelve syllables.
		taka…≥ 4x	As fast as possible (without an example) a sequence of minimal eight syllables.

MRR Analysis

After all the data with the basic protocol were collected, the process of analysing the samples started with the goal to maybe alter the protocol procedure of assessing and analysing the MRR-task. Since the program stored one whole recording of all trials per child the recordings were cut in smaller sequences by hand with Praat software, version 6.0.21 (Boersma & Weenink, 2016).

Six students at HAN University of Applied Sciences (HAN-UAS) and three SLPs analysed the mono-, tri- and bi-syllable sequences according to a protocol, which is shown in Table 3. They were all trained by the first author and started with analysing one practice sample of one audio-recording, which contained all the six sequences. The students received instructions on how to use and interpret the protocol (for example, which syllable sequence is suitable for further analyses if the child took a breath or pause?) only the last two items of the MRR-task were analysed (those elicited by the instructions "faster" and "as fast as possible"). Any occurring speech errors were registered per sequence in an excel file (for example /papadada/).

Table 3. Analysis protocol for calculating the MRR.

The sequence is considered correct if: - the syllables are pronounced fluently in succession; dialect variances are accepted
The sequence is considered partially correct if: - the sequence contains a single error (for example /papatapapapa/); then the sequences before and after the error are considered, and the longest and best sequence (at least 3 syllables) is selected. - the sequence contains noise or other interfering elements, but a good sequence can be analysed before or after the noise or interfering element; then the longest and best sequence is selected (at least 3 syllables). - the sequence contains pauses or interruptions; then the series are evaluated before and after the pause, and the longest and best sequence (at least 3 syllables) is selected. Pauses can arise from: o Inhalation: The child inhales during the execution of the sequence and then continues with the sequence. This also applies to syllables that are pronounced on an inhalation. o Rhythm: The child deviates from the rhythm of the sequence and a pause occurs. This is seen in waveform representation with a striking distance between two syllables.
The sequence is considered incorrect if: - the sequence in total consists of four syllables or less - the sequence is influenced by phonological processes (eg substitution, reduction, assimilation, metathesis, addition, etc.); these sequences were marked in an excel file for error analysis. - the sequence is influenced by one of the following issues: o Noise or other interfering elements Noise due to an interruption on the part of the examiner or other audible sounds, that makes the individual syllables unrecognisable. o Sound volume Low volume that makes the individual

syllables unrecognisable.

o Syllables cannot be distinguished

Syllables in the waveform cannot clearly be distinguished from each other.

The audio-recordings, containing just one sequence and attempt, were analysed by the first author and one of the SLPs with the help of a customized Praat-script (developed by one of the authors; HT). The script detected and marked syllable onsets by localising the noise burst of the voiceless plosives. The first and the last syllable were excluded because speakers often produce the first syllable with a longer duration and higher intensity (Thoonen et al., 1996) and the last syllable is also often lengthened (Ackermann et al., 1995). Before extracting the syllable durations and MRR, the marked syllable onsets were depicted in the waveform and inspected visually and any errors in the number of syllables indicated by the script were corrected manually. If corrected manually, the script could not give the separate durations of all the individual syllables and only the MRR value (total number of syllables divided by total duration of the sequence) was given. Figure 1 gives an example of one of the sequences with the markers. Only sequences with a remaining minimum of three syllables were included in the analysis. In some cases, the script could not detect syllable onsets correctly. These samples were analysed by hand to determine the number of syllables and the duration of the sequence. Eventually, all data of the MRR were merged in SPSS, version 24 for Windows (SPSS Inc., Chicago, IL, USA).

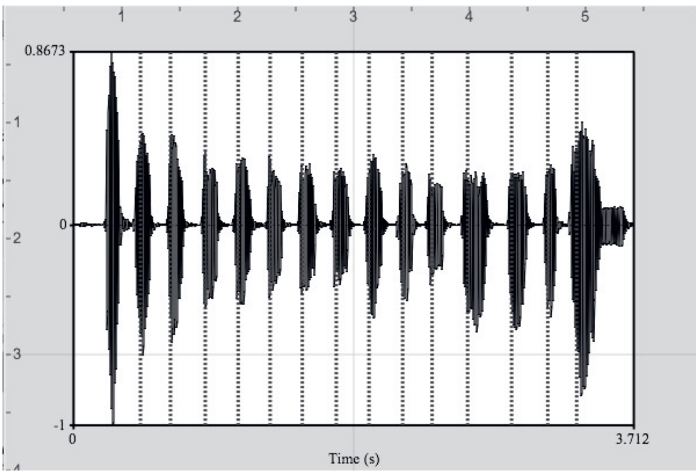


Figure 1. Example of the analysis with the Praat-script of one sequence /tatata/, fastest first attempt.

3

Reliability

Interrater and test-retest reliability were examined and described. We will only describe the result of this study; all details can be found in the publication by van Haaften et al. (2019). Interrater reliability was good for the monosyllabic sequences /pa/ (ICC 0.81) and /ka/ (ICC 0.83) and sufficient for /ta/ (ICC 0.77). For /pataka/, /pata/and /taka/, we found insufficient interrater reliability with ICCs ranging from 0.41 to 0.62. Especially the younger children had difficulty performing the /pataka/, /pata/ and /taka/; had we included whether the attempts were successful or not, the ICC might have been much higher. Test-retest reliability showed a sufficient reliability measure (ICC 0.70) for /pa/, and for the other sequences the test-retest reliability was insufficient with ICCs ranging from 0.18 to 0.60. Reasons for these low scores could be the rapid development of the younger children during the interval between test and retest, or a test-retest training effect because children were significantly more competent on /pataka/ on the second test than on the first test (t (53) = -3.02, p = .004).

Statistical Analysis

First, frequency tables were constructed to determine how many children produced the different sequences of the MRR. Then, a comparison was made between the completion of the different sequences, for example monosyllabic sequences versus multisyllabic sequences. Frequency tables were also constructed for the MRR value of the different sequences and attempts. Means and SDs of all parameters were calculated per age group and repeated measures ANOVA's were conducted to compare the best performance on the fast (with example) and the fastest (without example) attempt per sequence. Furthermore, to determine if the gold standard of ten syllables should be maintained, means and SDs over all the first 3-10 syllables each were calculated per sequence and differences were investigated. Furthermore, intraclass correlation coefficients (ICCs) with two-way mixed-effects models featuring no fixed effects were calculated between the MRRs over each of the numbers of syllables compared with the gold standard of ten syllables.

Results

First the results of all children are described to answer the questions if children of all ages can perform the MRR-tasks and if all children can perform all the different MRR-tasks (mono-, bi- and trisyllabic sequences). Subsequently, we investigate whether one of the instructions (fast or faster) elicits faster MRRs and whether it matters to ask multiple attempts per sequence. The last part of the results addresses the question if there is a difference in MRR when calculated over less than 10 syllables per sequence.

Ability to perform the MRR task

Tables 4 and 5 show the number of children executing and failing the different sequences of the MRR. For the 2-4-year-olds, not all audio-recordings included all MRR sequences because sometimes the child refused to utter one or multiple sequences, and sometimes the SLP foresaw that the child would not execute the bi- and trisyllabic sequences after finishing the monosyllabic sequences. These cases are marked *No sequence* in Tables 4 and 5. A sequence is marked *Fail* if the child refused to complete the sequence, if not enough syllables were detected (minimum of three), if an irregular rhythm (distinct pause) was executed, or if the child made errors (for example /pada/ instead of /pata/). For /pa/ 62 children refused to utter any syllables, for /ta/ 91 children, /ka/ 77 children, /pataka/ 156 children, /pata/ 100 children and for /taka/ 129 children. For each of the monosyllabic sequences, the results show that about 80% of children could produce the sequence correctly. For the multisyllabic sequences, the percentage of children that could produce the sequence correctly is lower, i.e., 65.1% for /pataka/and slightly higher percentages for /pata/ (75.9) and /taka/ (77.7%).

Table 4. Fail and pass of all monosyllabic sequences.

Age group	/pa/					/ta/					/ka/				
(years;months)	n	% Pass	% Fail		% No sequence	n	% Pass	% Fail		% No sequence	n	% Pass	% Fail		% No sequence
			Errors	<3 syllable				Errors	<3 syllable				Errors	<3 syllable	
2;0-2;4	59	30.5%	1.7%	39.0%	28.8%	59	32.2%	1.7%	32.2%	33.9%	58	34.5%	3.4%	37.9%	24.1%
2;4-2;8	79	35.4%	2.5%	44.3%	17.7%	81	34.6%	3.7%	37.0%	24.7%	80	35.0%	6.3%	32.5%	26.3%
2;8-3;0	83	66.3%	3.6%	21.7%	8.4%	82	64.6%	1.2%	20.7%	13.4%	81	63.0%	3.7%	23.5%	9.9%
3;0-3;4	90	71.1%	1.1%	18.9%	8.9%	90	75.6%	0.0%	13.3%	11.1%	88	75.0%	2.3%	14.8%	8.0%
3;4-3;8	90	66.7%	5.6%	20.0%	7.8%	92	71.7%	1.1%	14.1%	13.0%	94	69.1%	5.3%	18.1%	7.4%
3;8-4;0	95	90.5%	1.1%	5.3%	3.2%	95	87.4%	2.1%	5.3%	5.3%	94	88.3%	3.2%	3.2%	5.3%
4;0-4;4	85	85.9%	7.1%	3.5%	3.5%	84	90.5%	2.4%	3.6%	3.6%	81	91.4%	1.2%	2.5%	4.9%
4;4-4;8	93	96.8%	0.0%	1.1%	2.2%	94	93.6%	2.1%	2.1%	2.1%	94	92.6%	3.2%	1.1%	3.2%
4;8-5;0	94	97.9%	0.0%	1.1%	1.1%	94	97.9%	1.1%	0.0%	1.1%	94	96.8%	0.0%	0.0%	3.2%
5;0-5;4	104	99.0%	1.0%	0.0%	0.0%	106	94.3%	0.9%	1.9%	2.8%	106	95.3%	1.9%	0.0%	2.8%
5;4-5;8	113	99.1%	0.0%	0.9%	0.0%	111	94.6%	2.7%	0.0%	2.7%	114	95.6%	2.6%	0.0%	1.8%
5;8-6;0	103	99.0%	1.0%	0.0%	0.0%	105	100.0%	0.0%	0.0%	0.0%	104	99.0%	1.0%	0.0%	0.0%
6;0-6;6	107	98.1%	1.9%	0.0%	0.0%	108	99.1%	0.9%	0.0%	0.0%	107	98.1%	1.9%	0.0%	0.0%
6;6-7;0	108	99.1%	0.9%	0.0%	0.0%	108	97.2%	1.9%	0.0%	0.9%	109	99.1%	0.9%	0.0%	0.0%
Total	1303	1095	24	122	62	1309	1095	20	103	91	1304	1091	33	103	77
% sample	100.0%	84.0%	1.84%	9.4%	4.8%	100.0%	83.7%	1.5%	7.9%	6.9%	100.0%	83.7%	2.5%	7.9%	5.9%

Table 5. Fail and pass of all tri- and bisyllabic sequences.

Age group	/pataka/					/pata/					/taka/				
(years;months)	n	% Pass	% Fail		% No	n	% Pass	% Fail		% No	n	% Pass	% Fail		% No
			Errors	<3 syllable	sequence			Errors	<3 syllable	sequence			Errors	<3 syllable	sequence
2;0-2;4	55	16.4	23.6	18.2	41.8	57	22.8	15.8	35.1	26.3	52	23.1	13.5	11.5	51.9
2;4-2;8	70	17.1	24.3	20.0	38.6	77	29.9	11.7	32.5	26.0	66	34.8	15.2	13.6	36.4
2;8-3;0	71	23.9	32.4	11.3	32.4	81	45.7	11.1	24.7	18.5	69	52.2	10.1	10.1	27.5
3;0-3;4	83	47.0	21.7	13.3	18.1	89	57.3	14.6	13.5	14.6	78	66.7	10.3	7.7	15.4
3;4-3;8	89	42.7	24.7	9.0	23.6	94	60.6	10.6	18.1	10.6	83	59.0	14.5	6.0	20.5
3;8-4;0	86	62.8	17.4	7.0	12.8	94	75.5	9.6	9.6	5.3	90	86.7	4.4	1.1	7.8
4;0-4;4	80	68.8	17.5	0.0	13.8	83	80.7	9.6	2.4	7.2	83	80.7	9.6	1.2	8.4
4;4-4;8	89	75.3	18.0	1.1	5.6	95	89.5	4.2	1.1	5.3	91	89.0	6.6	0.0	4.4
4;8-5;0	91	68.1	24.2	0.0	7.7	92	84.8	10.9	0.0	4.3	90	81.1	13.3	0.0	5.6
5;0-5;4	103	79.6	14.6	0.0	5.8	104	91.3	4.8	1.0	2.9	103	84.5	12.6	1.0	1.9
5;4-5;8	109	83.5	12.8	0.0	3.7	112	89.3	7.1	0.0	3.6	114	91.2	5.3	0.0	3.5
5;8-6;0	101	91.1	8.9	0.0	0.0	102	94.1	5.9	0.0	0.0	104	91.3	7.7	0.0	1.0
6;0-6;6	104	86.5	11.5	0.0	1.9	108	95.4	4.6	0.0	0.0	106	94.3	5.7	0.0	0.0
6;6-7;0	108	91.7	7.4	0.0	0.9	109	100.0	0.0	0.0	0.0	109	96.3	3.7	0.0	0.0
Total	1239	807	218	58	156	1297	985	105	107	100	1238	962	111	36	129
% sample	100.0	65.1	17.6	4.7	12.6	100.0	75.9	8.1	8.3	7.7	100.0	77.7	9.0	2.9	10.4

Next, we investigated the number of correctly produced sequences per individual. Table 6 provides an overview of the number of monosyllabic sequences that children in the different age groups have performed, showing that only 21% of the children under the age of three can perform all three monosyllabic sequences.

Table 6. Numbers of children producing zero to three of the three monosyllabic sequences correctly per age group.

Age group (years;months)	**Monosyllabic sequence**			
	0	**1**	**2**	**3**
2;0-2;4	42	12	9	9
2;4-2;8	58	18	12	14
2;8-3;0	32	14	20	35
3;0-3;4	23	13	19	49
3;4-3;8	31	14	18	47
3;8-4;0	8	8	14	72
4;0-4;4	18	5	13	64
4;4-4;8	23	2	8	82
4;8-5;0	23	0	4	89
5;0-5;4	16	2	7	96
5;4-5;8	14	3	10	101
5;8-6;0	12	1	3	101
6;0-6;6	9	0	7	101
6;6-7;0	10	0	7	102
Total	319	92	151	962
% sample	20.9	6.0	9.9	63.1

In order to determine the capability of carrying out the bi- and trisyllabic sequences in relation to the children’s abilities to produce the monosyllabic sequences, we cross-tabulated the number of correctly produced monosyllabic sequences with the correct production of the bi- and trisyllabic sequences (Table 7). The results indicate that children who can produce at least two monosyllabic sequences are more likely to subsequently also correctly produce a bi- or trisyllabic sequence. The children who can only produce less than two monosyllabic sequences have a much lower chance of performing a tri- and bisyllabic sequences, showing a weak positive relation (rs = 0.278, n = 1,524, p < .001).

Table 7. Cross-tabulation of numbers of children correctly producing zero to three bi- or trisyllabic sequences in relation to their number of correctly produced monosyllabic sequences.

Monosyllabic sequences	Tri- and bisyllabic sequences									
	None		One		Two		Three		Total	
	n	%	n	%	n	%	n	%	n	%
None	300	19.7	15	1.0	3	0.2	1	0.1	319	20.9
One	50	5.3	17	1.1	17	1.1	8	0.7	92	6.0
Two	29	1.9	24	1.6	56	3.7	42	3.8	151	9.9
Three	33	2.2	84	5.5	226	14.8	619	56.6	962	63.1
Total	412	27.0	140	9.2	302	19.8	670	44.0	1524	100.0

Choosing the best sequence and the number of syllables

Children under 35 months of age have more difficulty with executing the different sequences for that reason these children were excluded in further analyses which resulted in the inclusion of 1041 children.

During data collection, the question raised which attempt or sequence (the one after the instruction ‘faster’ or ‘as fast as possible’) would actually be the fastest MRR, as we observed that children do not always go faster if they have been instructed to go as fast as possible. In addition, some children got up to three attempts to produce a sequence fast or as fast as possible because the administrator estimated that the child could go faster. As there is no evidence in literature, to our knowledge, regarding which attempt is the fastest overall, we compared the performance of the children after the instructions ‘fast’ and ‘as fast as possible’, as well as the performances on the extra attempts. Of the total group, 742 children got more than one attempt for at least one of the sequences. To determine whether instruction has an effect on the realized rate, we compared the best attempt of the children on the ‘fast’ instruction with the best attempts on the ‘as fast as possible’ instruction (Table 8). A repeated measures ANOVA yielded significant effects of instruction for all sequences except for /ka/ (Table 8), indicating that for all sequences except /ka/ children achieved a higher rate on average upon the instruction ‘as fast as possible’ as compared to the preceding ‘faster’ instruction. However, the data must be interpreted with caution, since the effect sizes are rather small, in particular for the monosyllabic sequences (Table 8).

Table 8. Comparison of the best performance on the two instructions "faster" (with example) and "as fast as possible" (without example).

Sequence	Instruction	N	MRR Mean	SD	df	F	p	ω²
/pa/	faster	790	4.4	.6	1, 550	26.601	<.001	.009
	fastest	752	4.5	.7				
/ta/	faster	821	4.2	.5	1, 615	56.115	<.001	.0019
	fastest	719	4.4	.7				
/ka/	faster	829	4.2	.5	1, 613	0.314	.575	.000
	fastest	775	4.2	.6				
/pataka/	faster	687	3.7	.7	1, 411	84.558	<.001	.049
	fastest	627	4.1	1.0				
/pata/	faster	735	4.1	.6	1, 477	88.687	<.001	.049
	fastest	707	4.5	.9				
/taka/	faster	743	4.1	.6	1, 476	75.459	<.001	.041
	fastest	677	4.4	.9				

Note. N gives the number of children who produced the sequence; the statistical test and the calculation of mean and SD were conducted with repeated measures ANOVA on less (df+1) pairwise comparisons.

Table 9. Kruskal-Wallis statistics for the comparison of MRR values for the different numbers of syllables in the produced sequences.

Sequence	N	df	H	p
/pa/	983	8	20.29	.009
/ta/	987	8	12.99	.112
/ka/	981	8	14.21	.076
/pataka/	893	8	7.472	.487
/pata/	953	8	13.82	.087
/taka/	934	8	15.51	.050

Number of syllables

In recent MRR protocols (Rvachew & Brosseau-Lapré, 2012; Yaruss & Logan, 2002; Thoonen et al., 1996; wit, et al., 1993) the number of syllables that are required/prescribed for analysis are 10 or 12 syllables per sequence. Our clinical experience, however, is that not many children can produce 10- or 12-syllable sequences, especially children with MSD. Because the aim of our research project is to develop an assessment for children with MSD it is important to evaluate if the protocol can also be administered with less than 10 syllables.

Table 10. Mean (and SD) syllable rate for each of the different sequence lengths per number of syllables.

Number of syllables	/pa/ n=992			/ta/ n=996			/ka/ n=990			/pataka/ n=902			/pata/ n=962			/taka/ n=943		
	N	M	SD	N	M	SD	N	M	SD	N	M	SD	N	M	SD	N	M	SD
3	183	4.5	0.8	165	4.4	0.8	140	4.1	0.7	120	4.2	1.2	115	4.2	0.8	111	4.2	0.9
4	158	4.6	0.7	161	4.4	0.7	143	4.2	0.7	142	4.1	0.9	156	4.5	1.0	119	4.5	0.9
5	155	4.6	0.6	129	4.6	0.7	124	4.2	0.6	113	4.1	1.0	128	4.4	0.9	100	4.4	0.9
6	112	4.5	0.6	104	4.4	0.7	135	4.2	0.6	101	4.1	0.9	127	4.6	2.0	128	4.6	0.9
7	107	4.7	0.6	107	4.4	0.7	131	4.3	0.5	113	4.0	0.9	96	4.5	0.7	119	4.4	0.8
8	75	4.6	0.5	76	4.4	0.6	90	4.2	0.6	63	3.9	0.8	92	4.5	0.9	83	4.3	0.7
9	68	4.7	0.7	82	4.5	0.5	74	4.3	0.5	68	4.1	0.8	60	4.6	0.7	70	4.4	0.7
10	43	4.6	0.6	61	4.5	0.5	45	4.2	0.5	62	3.9	0.8	53	4.6	0.7	65	4.5	0.7
>10	91	4.7	0.5	111	4.6	0.6	108	4.3	0.6	120	3.9	0.8	135	4.5	0.7	148	4.5	0.7

Table 11. Intraclass correlation coefficients for the comparison of syllable rate for each of the sequence lengths 3 to 9 with 10 or more.

Sequence	Comparison syllables	*N*	ICC	CI lb	CI ub
/pa/	3 with 10	92	.904	.858	.936
	4 with 10	92	.945	.918	.963
	5 with 10	92	.966	.949	.977
	6 with 10	92	.975	.962	.983
	7 with 10	92	.985	.978	.990
	8 with 10	92	.994	.991	.996
	9 with 10	92	.997	.996	.998
/ta/	3 with 10	105	.882	.831	.918
	4 with 10	105	.910	.871	.938
	5 with 10	105	.936	.907	.956
	6 with 10	105	.965	.949	.976
	7 with 10	105	.978	.968	.985
	8 with 10	105	.991	.987	.994
	9 with 10	105	.995	.993	.997
/ka/	3 with 10	102	.843	.776	.891
	4 with 10	102	.904	.886	.934
	5 with 10	102	.929	.897	.952
	6 with 10	102	.957	.936	.970
	7 with 10	102	.977	.966	.984
	8 with 10	102	.986	.980	.991
	9 with 10	102	.995	.993	.997
/pataka/	3 with 10	58	.881	.807	.928
	4 with 10	58	.899	.835	.939
	5 with 10	58	.913	.857	.947
	6 with 10	58	.962	.936	.977
	7 with 10	58	.982	.969	.989
	8 with 10	58	.983	.972	.990
	9 with 10	58	.989	.981	.993
/pata/	3 with 10	86	.735	.620	.819
	4 with 10	86	.806	.718	.869
	5 with 10	86	.869	.806	.913
	6 with 10	86	.918	.877	.946
	7 with 10	86	.953	.929	.969
	8 with 10	86	.978	.966	.985
	9 with 10	86	.992	.988	.995
/taka/	3 with 10	105	.824	.752	.877
	4 with 10	105	.863	.805	.905
	5 with 10	105	.905	.864	.935
	6 with 10	105	.945	.921	.963
	7 with 10	105	.974	.962	.982
	8 with 10	105	.987	.980	.991
	9 with 10	105	.995	.992	.996

Note. ICC = intraclass correlation coefficient; CI lb= confidence interval lower bound; CI ub= confidence interval upper bound

Therefore, Kruskal-Wallis test (none of the sequences met the test for equality of variance) per sequence was executed to see if there are differences between the MRR values for each of the sequence lengths with a minimum of three syllables and combining the sequences longer than 10 syllables. Sequence /pa/ showed a significant result and no significant difference in syllable rate between the different sequence lengths was observed for the other sequences (see Table 9). In Table 10 the descriptive values of the mono-, tri- and bisyllabic sequences are presented.

To compare MRR when calculated over different numbers of syllables per child and not between the children as described in Table 10, the MRRs of each successive number of syllables were calculated for children who produced 10 or more syllables in a sequence. Differences between the mean syllable rate for each of the successive sequence lengths from three to nine were studied by estimating intraclass correlation coefficients (ICCs). Table 11 shows good to excellent ICCs for every sequence length (except for the mean syllable rate of sequence length three in comparison with the mean rate sequence length ten of /pata/, which has a moderate ICC).

Discussion

In the current study, we adapted an existing MRR protocol and evaluated this protocol in a sample of 1,524 typically developing Dutch children from two to seven years old; the largest group of children of which MRR assessment is described thus far. The results showed first that children under 30 months of age have severe difficulty with executing the tasks properly and even for children up to three years of age it is still difficult. Most of the previous studies (Henry, 1990; Yaruss & Logan, 2002) described groups of children from three years and older, simply because this is the youngest age at which children tend be referred to an SLP (Tiffany, 1980). Although there still is much debate about administering the MRR at this young age, these studies concluded that children from three years of age can perform the MRR-task. The present results corroborate and extend these findings in a large sample, showing that administering MRR-tasks in younger children is indeed problematic. For that reason, we conclude that MRR should not be assessed in children under the age of three and we adjusted the MRR protocol for future use accordingly (which is part of the CAI test battery).

Second, the results showed that children who have difficulty performing the monosyllabic series cannot perform the bi- and trisyllabic sequences. In itself this seems obvious since the bi- and trisyllabic sequences are articulatorily much more difficult to pronounce than the monosyllabic sequences (Terband et al., 2011; Wong et al., 2011). The importance, however, is that this establishes that MRR for the monosyllabic sequences and MRR for the bi- and trisyllabic sequences should be separate outcome measures that should both be included in the MRR task report. Furthermore, we included in the protocol that the bi- and trisyllabic sequences should not be administered if children could not produce the monosyllabic sequences to reduce the burden of the test battery.

According to the assessment protocol, the test administrator is instructed to ask children to redo the sequence up to two times again if he/she suspects it was not performed at the child's maximum capacity. In 14% of the cases, the child was asked to repeat a sequence, and in 2% of the cases, the child got a third attempt of one or more of the sequences. Our results showed that most children were actually the fastest at the first attempt compared to the other attempts; only about 20% of the children were faster on the second or third attempt. However, it seems important to give children a second or even a third attempt if the administrator expect children to be even faster, because for about 12% of the attempts the child was faster at the second or third attempt. In most protocols there is a gradual build-up of number of syllables and pace of the sequences to be produced. After several trials the children can be asked to produce the sequence as quickly as possible without an example. The expectation was that children show the fastest rate with the instruction to go as fast as possible, but this has not been explored in any published data. On the surface, a substantial number of children performed the fastest MRR with the instruction to go faster. The results showed a distinct pattern underneath. For the two monosyllabic sequences /pa/ and /ta/ and for the bi- and trisyllabic sequences the instruction to perform the sequence "as fast as possible" yielded the fastest MRR while for /ka/ the performance was the same between the two instructions. However, the effect sizes are very small and therefore it is debatable if the difference between the performance for the two instructions is clinically relevant. The difference could be an effect of learning how to conduct the task. Within the protocol the child first practices the sequence (build up) and when the child is familiar with the sequence, the child is asked to produce it as fast as possible, thereby requiring maximal performance. However, we noticed children going louder and not that much faster and the effect size of the difference between the two instructions is very small. The advice is to choose the fastest attempt which can be either performed with the last or the second last instruction and/or attempt.

Recent studies report the use of 10 to 12 syllables (Rvachew & Brosseau-Lapré, 2012; Thoonen et al., 1996; Wit et al., 1993; Yaruss & Logan, 2002). However, this study showed that a large number of especially the youngest children do not reach the criterion of sequence length 10. Instead, they produce sequence lengths in the range from three to about 10 syllables after exclusion of the first and last syllable. Gadesmann and Miller (2008) compared the following methods of the same sequence children pronounced: number of syllables for the first 5 s, the time of pronouncing a number of repetitions (five times) and the total duration of the maximum sequence length uttered in one breath, and thereby showed that the results are identical irrespective of the method of assessment. Based on this study and our own data, we conclude that a sequence of at least five syllables, such that the mean rate is based on measuring the duration of at least 3 syllables, is sufficient.

MRR is the most common measure, but in the literature, there are also indications that other measures of the MRR-task can provide valuable information on the development of speech motor skills and therefore a better understanding of the underlying problems in children with MSD. In children with MSD measuring speech variability can yield important information about the speech motor control system and to support the identification, assessment, and treatment of the underlying speech process (McCabe et al., 1998; Murdoch et al., 1995; Preston & Koenig, 2011; Terband et al., 2011; Thoonen et al., 1999; Williams & Stackhouse, 2000; Wit et al., 1993). The coefficient of variation of the syllable durations could be added to investigate the variability of the sequences, as well as the Normalized Pairwise Variability Index (NPVI), which in previous studies has been used to investigate stress-timing and syllable-timing (Grabe & Low, 2002; van Brenk, 2015). However, some reservation is required in this respect as the current speech-to-result set-up for most variability measures is not yet sufficiently automated to serve as an easily applicable analysis tool in the daily practice of speech therapists (van Brenk, 2015). The goal for us is to see if and how the assessment of variability as an outcome measure of the MRR-task could be implemented in the Computer Articulation Instrument.

In the present study, we asked parents or caregivers whether their child had a history of hearing problems and if they had any doubts about his/her hearing. It is possible that the child could have a mild hearing problem because parents and caregivers can overlook a mild hearing problem (Lo et al., 2006). In the Netherlands, the hearing of all children is recorded during the regular governmental hearing screening after two weeks after birth (neonatal screening) and at the age of four (Lanting et al., 2017). Furthermore, the research-assistants were asked to pay particular attention to signs of hearing problems. This is why we did not include a whole hearing screening, but it is possible that a few children had a mild hearing problem.

In the field of adult MSD, there has been debate about the potential utility of nonspeech oral motor tasks (Kent, 2015; Maas, 2016; Weismer, 2006; Ziegler & Ackermann, 2013) and recently, Staiger et al. (2017) suggested that MRR is not a speech-like skill and therefore MRR is unusable in clinical assessment of MSD in adults. We would like to stress here, however, that results that hold for adults with acquired disorders do not necessarily hold for children with developmental disorders. As pointed out elsewhere in this special issue following Bishop (1997) and Karmiloff-Smith et al. (2006; 2003), developmental disorders are characterized by association rather than dissociation of functions (Terband et al., 2016, 2019). Whereas the adult speech production system is highly redundant, and the different processes and representations are highly overlearned, children have an incomplete system that is still in development. At the age of four to six years, children still make speech errors in conversational speech or in naming pictures that can be based on an incomplete phonological system or an immature motoric speech system (Maassen & Terband, 2015). The dissociation between MRR and other speech tasks found for adults thus cannot be extended to children. In fact, correlations between performance on speech tasks and different nonspeech motor tasks have been found in several groups of children with speech disorder, among which children with Childhood Apraxia of Speech (Nijland et al., 2015) and children with Fetal Alcohol Spectrum Disorders (Terband et al., 2018).

In addition, and even more importantly, the MRR serves an important function in differential diagnosis of developmental speech disorders, as for example also expressed in the 2011 Speech-language pathology medical review guidelines from the American Speech-Language-Hearing Association (2011). Several studies have reported differences between children with and without MSD on the MRR (Thoonen et al., 1996; Wit et al., 1993) and the MRR has been shown to be discriminative between CAS and developmental dysarthria (Thoonen et al., 1996, 1999). We therefore propose that for a comprehensive speech assessment the following tasks should be administered: picture naming, nonword imitation, word and nonword repetition and MRR (Van Haaften et al., 2017). The present study yields directions for administering the MRR-tasks and norm values to interpret the performances relative to typically developing children. Research with diverse groups of children with SSD with the comprehensive test battery is required to validate the MRR and evaluate its contribution to the speech diagnosis. Such studies are currently conducted by our research group.

In summary, the new MRR-protocol describes how to assess children from three years of age; if a child cannot perform more than two monosyllabic sequences the session can be ended. In the clinical report of the MRR the score for the monosyllabic and for the bi- and trisyllabic sequences must be given separately. Children do not have to be encouraged to perform a sequence of at least ten syllables. For each MRR-sequence, the test administrator should analyze the attempts the child has produced upon the last two instructions and then determine which attempt was the fastest.

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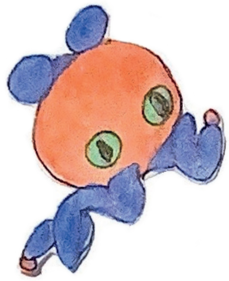

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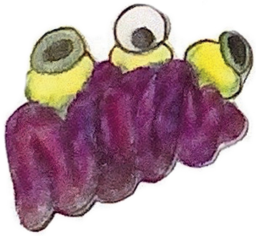

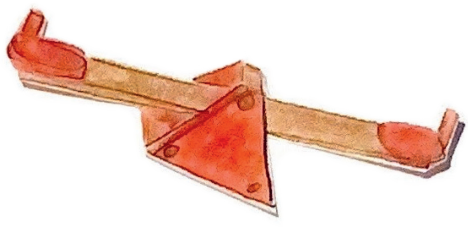

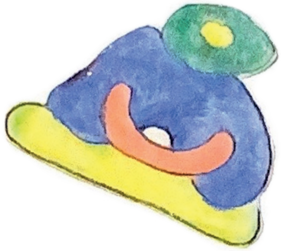

Maximum Repetition Rate in a large cross-sectional sample of typically developing Dutch-speaking children

Sanne Diepeveen and Leenke van Haaften, Hayo Terband,
Bert de Swart, Lenie van den Engel-Hoek, Ben Maassen

Published in International Journal of Speech-Language Pathology 2021, 23(5), 508-518

Abstract

Purpose: The current study aims to provide normative data for the maximum repetition rate (MRR) development of Dutch-speaking children based on a large cross-sectional study using a standardised protocol.

Method: A group of 1,014 typically developing children aged 3;0 to 6;11 years performed the MRR task of the Computer Articulation Instrument (CAI). The number of syllables per second was calculated for mono-, bi-, and trisyllabic sequences (MRR-pa, MRR-ta, MRR-ka, MRR-pata, MRR-taka, MRR-pataka). A three-way mixed ANOVA was conducted to compare the effects of age and gender on MRR scores in different MRR sequences.

Result: The data analysis showed that overall MRR scores were affected by age group, gender and MRR sequence. For all MRR sequences the MRR increased significantly with age. MRR-pa was the fastest sequence, followed by respectively MRR-ta, MRR-pata, MRR-taka, MRR-ka and MRR-pataka. Overall MRR scores were higher for boys than for girls, for all MRR sequences.

Conclusion: This study presents normative data of MRR of Dutch-speaking children aged 3;0 to 6;11 years. These norms might be useful in clinical practice to differentiate children with speech sound disorders from typically developing children. More research on this topic is necessary. It is also suggested to collect normative data for other individual languages, using the same protocol.

Introduction

Maximum repetition rate (MRR), or *diadochokinesis*, involves alternating motion rate tasks comprising speech like syllables (Kent, 2015). MRR is one of the most commonly used oral-motor assessments in clinical practice (Icht & Ben-David, 2014; Williams & Stackhouse, 2000). It is suggested as an important part of a test battery to differentiate between various speech disorders (Diepeveen, Van Haaften, Terband, De Swart, & Maassen, 2019; Maassen & Terband, 2015; Terband, Maassen, & Maas, 2019). However, there is also still a debate about the clinical value of the MRR. A higher-faster-farther approach might not be a good assessment because in speech speed is not a necessary skill (Ziegler et al., 2019). Although this is the case, MRR can play a role in diagnosing underlying articulomotor planning and programming problems (Maassen & Terband, 2015; Rvachew et al., 2005; Van Haaften, Diepeveen, Terband, et al.. 2019). MRR is therefore often used in the assessment of children with a suspicion of a motor speech disorder (MSD) and/or childhood apraxia of speech (CAS) (Murray, McCabe, Heard, & Ballard, 2015; Thoonen, Maassen, Gabreels, & Schreuder, 1999), and it has been used in the characterization of speech language phenotypes (e.g., Peter et al., 2017; Peter, Matsushita, & Raskind, 2012; Turner et al., 2015). To be able to interpret the results of the MRR adequately, it must be part of a set of speech tasks. By comparing the results of the MRR task with the results of other tasks (i.e. picture naming, nonword repetition) a complete speech profile can be obtained. The results of the MRR should not be used solely to diagnose children with speech sound disorders, because many children with SSD show similar behavioural symptoms in speech. The traditional way of diagnosing children with SSD might not be sufficient, because the different levels involved in speech influence each other (Namasivayam et al., 2020). The underlying processes involved in speech production are lemma access, word form selection, phonological encoding, speech motor planning and programming, and speech motor execution (Terband, Maassen, & Maas, 2019). Insight into the deficits that might be the underlying causes of an SSD, requires an extensive analysis of a child's performance on a range of speech tasks that reflect different underlying processes. A study of our research group (Van Haaften, Diepeveen, Van den Engel-Hoek et al., 2019) showed the distinctive function of four different speech tasks of a new speech production test battery for children: the Computer Articulation Instrument (CAI). The CAI contains the tasks picture naming, nonword imitation, word and nonword repetition and MRR. Factor analyses were conducted based on the assumption that clusters of selected parameters would reflect different aspects of speech production, either within or across tasks. Factor analyses revealed five meaningful factors: all picture-naming parameters (PN), the segmental parameters of nonword imitation (NWI-Seg), the syllabic structure parameters of nonword imitation (NWI-Syll), (non)word repetition consistency (PWV), and all MRR parameters. Each task reflects different aspects of speech production.

Furthermore, the construct validity was underlined by the weak correlations between CAI factor scores, indicating the independent contribution of each factor to the speech profile. In another study with 41 children (age 3;0 to 6;4; 26 boys and 15 girls) with SSD data were collected from the four tasks of the CAI. The children were categorised in two groups, moderate or a severe SSD indicated by their speech language pathologist (SLP). Results indicated a significant difference between the two groups for picture naming, nonword imitation (segmental and syllable structure) and the bisyllabic and trisyllabic MRR factor (Van Haaften, Diepeveen, Terband et al., 2019). The findings of these two studies suggest that the MRR should be part of the diagnostic process. Normative data of MRR is essential to differentiate children with delayed or disordered speech development from typically developing children. The availability of these data is important for SLPs to make clinical decisions.

Several studies have investigated MRR in typically developing children. The overall conclusion, across languages, is that MRR increases with age. Contrasting results were found in studies investigating gender differences and differences between specific MRR sequences. Some studies found differences between boys and girls (Modolo, Berretin-Felix, Genaro, & Brasolotto, 2011) or between MRR sequences (Blech, 2010; Prathanee, Thanaviratananich, & Pongjanyakul, 2003), while other studies found no differences between gender (Fletcher, 1972; Icht & Ben-David, 2015; Wong, Allegro, Tirado, Chadha, & Campisi, 2011; Zamani, Rezai, & Garmatani, 2017) or MRR sequence (Rvachew, Ohberg, & Savage, 2006; Thoonen, Maassen, Wit, Gabreels, & Schreuder, 1996). However, considerable methodological differences exist between the studies, with different methods of data collection and different scoring methods of MRR. Several studies used a time-by-count procedure (the time needed to repeat a certain number of syllables) (Blech, 2010; Fletcher, 1972; Prathanee et al., 2003; Rvachew et al., 2006; Thoonen et al., 1999; Thoonen et al., 1996; Yaruss & Logan, 2002; Zamani et al., 2017), while in other studies a procedure of count-by-time was used (the number of syllables repeated in a certain amount of time) (Henry, 1990; Icht & Ben-David, 2015; Juste et al., 2012; Modolo et al., 2011; Robbins & Klee, 1987). Because of these methodological differences, the normative data is difficult to compare. To reduce these differences, a standardised protocol is proposed in a study by Diepeveen et al. (2019). In this protocol, it is suggested that MRR should not be assessed in children under the age of 3 years. The maximum age up to seven years has been chosen, because previous research has shown that speech sound development continues up to seven years (Priester and Goorhuis-Brouwer, 2013). Monosyllabic sequences and bi- and trisyllabic sequences should be described as separate outcome measures and if children cannot produce the monosyllabic sequences, the bi- and trisyllabic sequences should not be administered. Nonsense syllabic sequences are used instead of real words as MRR is supposed to measure motor speech abilities rather than linguistic skills (Williams & Stackhouse, 2000). The measurement procedure follows the time-by-count principle. The data indicates that children do not have to be encouraged to perform series of at least ten syllables, but that series of five syllables is sufficient for a reliable and valid calculation of the MRR (Diepeveen et al., 2019). After exclusion of the first and last syllable, the mean rate is then based on the duration of at least three syllables.

Most of the MRR studies in typically developing children are based on a small number of children and relatively limited age ranges (Blech, 2010; Prathanee et al., 2003; Rvachew et al., 2006; Thoonen et al., 1999; Thoonen et al., 1996; Wong et al., 2011; Yaruss & Logan, 2002). As typically developing children show progress in speech motor skills as they grow older, normative data is required for consecutive age groups. Therefore, the aim of the present study is to provide normative data for the MRR development of Dutch-speaking children aged 3;0 to 6;11 years based on a large cross-sectional study using the standardised protocol by Diepeveen et al. (2019). Differences between age groups, gender and MRR sequences are described.

Method

Participants

The 1,014 participants of this study participated in a large normative study in the context of the development of a new speech production test battery in Dutch: the Computer Articulation Instrument (CAI; Maassen et al., 2019; Van Haaften, Diepeveen, Van den Engel-Hoek et al., 2019). The CAI consists of four tasks: (1) picture naming, (2) nonword imitation, (3) word and nonword repetition, and (4) maximum repetition rate (MRR) task. The data of the MRR task was used for the current study. Between January 2008 and April 2015, typically developing Dutch-speaking children aged between 2;0 and 7;0 were recruited via nurseries (n = 47) and mainstream primary schools (n = 71) in the Netherlands. Inclusion criteria were no hearing loss and Dutch being the spoken language at the nursery or primary school. The sample was representative for gender, geographic region and degree of urbanisation (Van Haaften et al., 2019). The parents or caregivers were asked to fill out a questionnaire containing questions about hearing problems, speech and language development, developmental problems and whether the child is seen by an SLP. Children were excluded if they had developmental problems that could influence the speech performance. See Maassen et al. (2019) and Van Haaften et al. (2019) for detailed information on sample characteristics and data collection. As Diepeveen et al. (2019) concluded that the MRR protocol of the CAI is applicable for children of 3 years and older, this study only used the data of children aged between 3;0 and 7;0, divided in 11 age groups. Table 1 shows the number of subjects per MRR sequence per age group and gender.

Table 1. Sample composition: numbers of children per age group, broken down by gender.

Age group (years;months)	Total number of children	M_{age}	Gender (*n*)	
			Boys	Girls
3;0-3;3	68	3;01	32	36
3;4-3;7	65	3;05	34	31
3;8-3;11	86	3;08	46	40
4;0-4;3	77	4;01	42	35
4;4-4;7	90	4;05	48	42
4;8-4;11	93	4;08	43	50
5;0-5;3	103	5;01	54	49
5;4-5;7	111	5;05	61	50
5;8-5;11	104	5;08	55	49
6;0-6;5	108	6;02	63	45
6;6-6;11	109	6;07	53	56
Grand total	1,014		531	483
% sample	100		52.4	47.6

Ethical considerations

The research ethics committee of the Radboud University Nijmegen Medical Centre stated that this study does not fall within the remit of the Medical Research Involving Human Subjects Act (WMO). Therefore, this study can be carried out (in the Netherlands) without an approval by an accredited research ethics committee. The study was conducted according to the ethical principles and guidelines in the Netherlands. For example, informed consent was obtained from all parents or caregivers.

Procedure

In the CAI project 14 SLPs administrated the test for the younger children (2 to 4 years of age) and 110 SLP students (working in pairs) assessed the older children (4 to 7 years of age). All assessors were trained in the administration of the MRR task by the first two authors. The assessment took place at the child's nursery or primary school in a quiet room. The CAI was administered using a computer laptop and the acoustic signal (minimum of 44.1 Hz; 16 bits) was automatically stored on the computer's hard disk. The child and SLP or SLP student were seated side by side in front of the computer. Both wore a headset, or a speaker and microphone were used. Testing took approximately 30 minutes for all the tasks of the CAI. The administration of the MRR task took about five to ten minutes per child.

MRR administration

For the administration of the MRR task the CAI uses the protocol described by Diepeveen et al. (2019). This protocol was developed based on previous studies in the Dutch language (Thoonen et al., 1999; Thoonen et al., 1996; Wit, Maassen, Gabreels, & Thoonen, 1993). Instructions were given by the CAI computer program to maximise standardisation. During the task children are required to reproduce pre-recorded sequences on one single breath: first three monosyllabic sequences (/papa../, /tata../ and /kaka../), followed by one trisyllabic sequence (/pataka.../) and finally two bisyllabic sequences (/pata../ and /taka../). It was not possible to change the order of sequences; the computer program was fixed.

First, the children were asked to repeat a short sequence of three syllables (e.g. /papapa/) in a normal speaking rate after an audio model. Second, children were asked to repeat a longer sequence of six syllables in a normal rate (e.g. /papapapapapa/). The third instruction included imitation of a sequence of 12 syllables at a faster speech rate after an audio example. Finally, the children were asked to produce the syllable sequences as fast as possible, without an audio model. The CAI allows a maximum of three attempts per sequence.

MRR analysis

Six SLP students of HAN University of Applied Sciences and three SLPs analysed the mono-, tri- and bi-syllabic sequences according to the analysis protocol for calculating the MRR proposed by Diepeveen et al. (2019). They were trained by one of the first authors (SD) and practiced with one sample before analysing the other samples. Since the program stores all tasks and all trials of a child in one recording, the recordings were spliced into fragments per trial manually with Praat software, version 6.0.21 (Boersma & Weenink, 2016). First the administrator determined if the sequence was pronounced correctly. The sequence was correct when the syllables were pronounced fluently in succession and had no articulation errors, allowing for dialect variances. The test administrator analysed the attempts the child has produced upon the last two instructions, calculated the syllables per second and recorded this in the database. The audio-recordings, each containing just one attempt of one sequence, were analysed with the help of a customised Praat-script (developed by one of the authors; HT). The script detected and marked syllable onsets by localising the noise burst of the voiceless plosives. The first and the last syllable were excluded because speakers often produce the first syllable with a longer duration and higher intensity (Thoonen et al., 1996) and the last syllable is also often lengthened (Ackermann, Hertrich, & Hehr, 1995). Before extracting the number of syllables, syllable durations and MRR score, the marked syllable onsets were depicted in the waveform and inspected visually and any errors in the number of syllables indicated by the script were corrected manually. Figure 1 gives an example of one of the sequences with the markers. Only sequences with a remaining minimum of three syllables, after exclusion of the first and last syllable, were included in the analysis. In 30% of the cases, the script could not detect syllable onsets correctly. These samples were analysed manually to determine the number of syllables and the duration of the sequence; administrators used both visual examination of the waveform and playback of the audio recording. In pilot study for our MRR-protocol, we studied the reliability (n = 126) between the computer script and the manually analysed recordings. The intraclass correlation coefficients (ICCs) were sufficient to good: /pa/ = .79; /ta/ = .90; /ka/ = .85; /pataka/ = .74; /pata/ = .79; /taka/ = .76. MRR score was calculated by dividing the number of syllables of the sequence by the duration of the sequence (syll/s). Eventually, number of syllables, duration time, and MRR score were merged in SPSS, version 24 for Windows (SPSS Inc., Chicago, IL, USA). The fastest correctly produced series of syllables, based on the number of syllables, is used for analysis.

Not all children completed all MRR sequences for reasons of shyness or inattentiveness. Furthermore, in some cases the audio files were damaged due to technical problems or background noise that prevented recognising the individual syllables. In this case, the recordings were excluded from the sample. Table 2 shows the number of children from whom an analysable MRR sequence was collected.

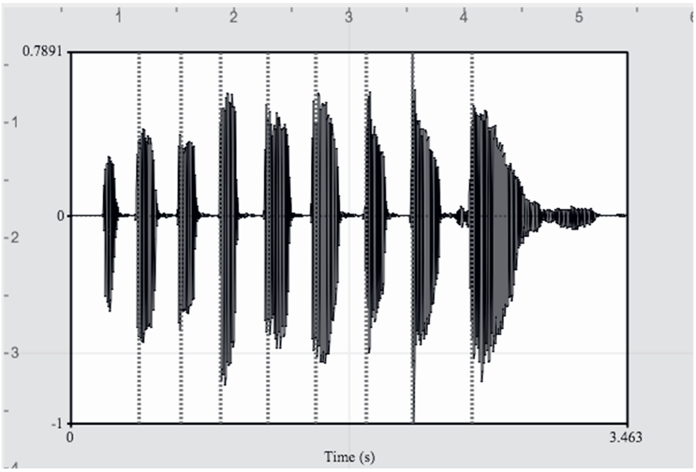

Figure 1. Example of the analysis with the Praat-script of one of the maximum repetition rate sequences.

Table 2. Descriptive statistics (means and standard deviations) of the MRR score (syll/s) per age group and gender, broken down by MRR sequence.

Gender	Age group		MRR sequence					
			MRR-pa	MRR-ta	MRR-ka	MRR-pataka	MRR-pata	MRR-taka
Total	3;0 – 3;3	*n*	37	37	37	37	37	37
		M	3.95	3.91	3.66	3.40	4.01	3.81
		SD	0.59	0.56	0.46	0.55	0.88	0.78
	3;4 – 3;7	*n*	38	38	38	38	38	38
		M	4.06	4.06	3.76	3.54	3.99	4.08
		SD	0.50	0.51	0.57	0.83	0.60	0.82
	3;8 – 3;11	*n*	51	51	51	51	51	51
		M	4.15	4.11	3.84	3.74	4.03	4.07
		SD	0.52	0.67	0.53	0.87	0.79	0.83
	4;0 – 4;3	*n*	60	60	60	60	60	60
		M	4.27	4.17	4.00	3.82	4.35	4.25
		SD	0.57	0.61	0.54	0.73	0.90	0.78
	4;4 – 4;7	*n*	77	77	77	77	77	77
		M	4.59	4.40	4.14	3.88	4.41	4.38
		SD	0.51	0.57	0.54	0.82	0.76	0.74
	4;8 – 4;11	*n*	77	77	77	77	77	77
		M	4.55	4.42	4.20	3.93	4.49	4.47
		SD	0.67	0.62	0.56	0.90	0.97	0.83
	5;0 – 5;3	*n*	87	87	87	87	87	87
		M	4.64	4.40	4.33	4.04	4.49	4.36
		SD	0.54	0.59	0.48	0.79	0.70	0.84
	5;4 – 5;7	*n*	97	97	97	97	97	97
		M	4.82	4.69	4.37	4.14	4.68	4.53
		SD	0.55	0.54	0.46	0.83	0.72	0.57
	5;8 – 5;11	*n*	94	94	94	94	94	94
		M	4.83	4.70	4.45	4.35	4.55	4.70
		SD	0.62	0.62	0.47	0.89	0.84	0.80
	6;0 – 6;5	*n*	99	99	99	99	99	99
		M	4.96	4.87	4.48	4.37	4.86	4.64
		SD	0.51	0.66	0.49	0.96	0.91	0.72
	6;6 – 6;11	*n*	103	103	103	103	103	103
		M	5.03	4.92	4.63	4.51	4.80	4.96
		SD	0.56	0.59	0.56	0.86	0.83	0.78
	Total	*n*	820	820	820	820	820	820
		M	4.64	4.52	4.26	4.07	4.51	4.48
		SD	0.64	0.67	0.58	0.90	0.86	0.81
Boys	3;0 – 3;3	*n*	18	18	18	18	18	18
		M	3.95	3.86	3.63	3.28	4.14	3.57
		SD	0.56	0.62	0.52	0.68	1.06	0.78
	3;4 – 3;7	*n*	21	21	21	21	21	21
		M	4.24	4.18	3.87	3.58	4.23	4.24
		SD	0.48	0.47	0.66	0.64	0.53	0.84
	3;8 – 3;11	*n*	28	28	28	28	28	28
		M	4.27	4.22	3.90	3.90	4.14	4.21

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		SD	0.45	0.76	0.53	1.00	0.82	0.93
	4;0 – 4;3	*n*	33	33	33	33	33	33
		M	4.36	4.31	4.03	4.00	4.52	4.19
		SD	0.51	0.66	0.60	0.73	0.96	0.89
	4;4 – 4;7	*n*	38	38	38	38	38	38
		M	4.64	4.39	4.29	3.83	4.45	4.35
		SD	0.49	0.59	0.57	0.77	0.92	0.73
	4;8 – 4;11	*n*	37	37	37	37	37	37
		M	4.51	4.51	4.18	3.94	4.50	4.46
		SD	0.75	0.58	0.64	1.03	1.03	0.95
	5;0 – 5;3	*n*	44	44	44	44	44	44
		M	4.68	4.49	4.34	4.04	4.65	4.44
		SD	0.59	0.71	0.47	0.77	0.71	0.97
	5;4 – 5;7	*n*	56	56	56	56	56	56
		M	4.80	4.68	4.30	4.26	4.66	4.48
		SD	0.55	0.57	0.47	0.94	0.74	0.57
	5;8 – 5;11	*n*	52	52	52	52	52	52
		M	4.90	4.76	4.46	4.39	4.55	4.69
		SD	0.72	0.62	0.53	0.90	0.80	0.84
	6;0 – 6;5	*n*	57	57	57	57	57	57
		M	4.94	4.96	4.55	4.43	4.92	4.71
		SD	0.50	0.72	0.5	1.11	0.95	0.80
	6;6 – 6;11	*n*	51	51	51	51	51	51
		M	5.21	4.98	4.62	4.53	4.98	5.02
		SD	0.63	0.59	0.59	0.86	0.92	0.83
	Total	*n*	435	435	435	435	435	435
		M	4.70	4.59	4.29	4.13	4.60	4.49
		SD	0.66	0.70	0.60	0.94	0.89	0.87
Girls	3;0 – 3;3	*n*	19	19	19	19	19	19
		M	3.95	3.97	3.69	3.51	3.89	4.03
		SD	0.63	0.49	0.40	0.38	0.69	0.72
	3;4 – 3;7	*n*	17	17	17	17	17	17
		M	3.84	3.91	3.61	3.49	3.69	3.88
		SD	0.44	0.54	0.41	1.04	0.55	0.77
	3;8 – 3;11	*n*	23	23	23	23	23	23
		M	4.02	3.98	3.75	3.54	3.90	3.89
		SD	0.57	0.54	0.53	0.65	0.75	0.67
	4;0 – 4;3	*n*	27	27	27	27	27	27
		M	4.17	3.97	3.97	3.61	4.15	4.32
		SD	0.63	0.51	0.46	0.68	0.81	0.62
	4;4 – 4;7	*n*	39	39	39	39	39	39
		M	4.54	4.41	4.00	3.92	4.36	4.42
		SD	0.52	0.56	0.47	0.88	0.57	0.76
	4;8 – 4;11	*n*	40	40	40	40	40	40
		M	4.59	4.34	4.22	3.92	4.48	4.48
		SD	0.60	0.65	0.49	0.79	0.92	0.71
	5;0 – 5;3	*n*	43	43	43	43	43	43
		M	4.60	4.30	4.31	4.04	4.33	4.28
		SD	0.48	0.44	0.49	0.83	0.65	0.68

5;4 – 5;7	*n*	41	41	41	41	41	41
	M	4.85	4.69	4.47	3.98	4.72	4.59
	SD	0.54	0.51	0.43	0.63	0.70	0.56
5;8 – 5;11	*n*	42	42	42	42	42	42
	M	4.74	4.61	4.45	4.29	4.54	4.71
	SD	0.46	0.61	0.39	0.88	0.89	0.76
6;0 – 6;5	*n*	42	42	42	42	42	42
	M	4.99	4.74	4.38	4.30	4.79	4.54
	SD	0.52	0.57	0.43	0.71	0.86	0.61
6;6 – 6;11	*n*	52	52	52	52	52	52
	M	4.86	4.86	4.64	4.50	4.63	4.91
	SD	0.43	0.60	0.54	0.87	0.69	0.72
Total	*n*	385	385	385	385	385	385
	M	4.58	4.44	4.23	4.02	4.42	4.46
	SD	0.62	0.63	0.55	0.84	0.80	0.74

Note. n = number of children from whom an MRR sequence was analysed; *M* = mean of the MRR score (syll/s); *SD* = standard deviation of the mean MRR score (syll/s); MRR-pa = number of syllables per second of sequence /pa/; MRR-ta = number of syllables per second of sequence /ta/; MRR-ka = number of syllables per second of sequence /ka/; MRR-pataka = number of syllables per second of sequence /pataka/; MRR-pata = number of syllables per second of sequence /pata/; MRR-taka = number of syllables per second of sequence /taka/

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Reliability

Interrater and test-retest reliability of the MRR scores (syll/s) were examined and described by Van Haaften et al. (2019). In this study, typically developing children aged between 2;0 and 7;0 were included. To measure interrater reliability the audio recordings of 103 children were randomly selected and scored by 33 raters. Their MRR scores were compared with those of one independent rater. A total of 107 children were randomly selected for the test-retest reliability study; these children were examined twice within three months by the same administrator. Two raters scored the audio recording of the initial test and retest, with the same rater scoring the tests of the same child. Interrater reliability, calculated with interclass correlation coefficient (ICC), was good for the monosyllabic sequences /pa/ (ICC 0.81) and /ka/ (ICC 0.83) and sufficient for /ta/ (ICC 0.77). The interrater reliability for the bisyllabic and trisyllabic items was insufficient, with ICCs ranging from 0.41 to 0.62. Especially the younger children (i.e., the 2- to 3-year-olds) had difficulties performing the bisyllabic and trisyllabic items, whereas a large number of children were not able to perform the task at all. The data of children who failed to perform the task were not included in the reliability study; had we included whether the attempts were successful or not, the ICC might have been higher. Test-retest reliability was sufficient for /pa/ (ICC 0.70) and insufficient for the other sequences, with ICCs ranging from 0.18 to 0.60. Reasons for these low scores could be the rapid development of the younger children during the interval between test and retest or a test-retest training effect. Based on these results, and the results of the study of Diepeveen et al. (2019), the younger children aged between 2;0 and 3;0 were not included in the current study. Further details and interpretations of the reliability study are discussed in van Haaften, Diepeveen, van den Engel-Hoek et al. (2019).

Statistical Analysis

To compare the effects of age and gender on MRR scores in different MRR sequences, and to test the hypotheses that there is a difference between the six MRR sequences and between boys and girls for the 11 age groups, a two-way mixed ANOVA was conducted. MRR score (syll/s) was the dependent variable, MRR sequence was the within-subject factor with six levels (MRR-pa, MRR-ta, MRR-ka, MRR-pataka, MRR-pata, MRR-taka), and there were two between-subject factors: age group (11 age groups) and gender (2 levels: boys and girls). Mauchly's test of Sphericity was conducted to test the hypothesis that the variances of differences between conditions are equal. Bonferroni correction was applied for post hoc comparisons. Statistical analyses were performed using SPSS version 20 for Windows (SPSS Inc., Chicago, IL, USA).

Results

The results of the mean number of syllables / second per age group and per sequence are presented in Figure 2. The percentage of children (in relation to the total number of children of the respective age group) who could perform the sequence correctly (fluently in succession; no articulation errors, allowing for dialect variances) is shown at the beginning of the bars.

The mean and standard deviations of each MRR sequence are depicted by age group and gender in Table 2, showing data of children who could perform all the six sequences correctly. Mauchly's test indicated that the assumption of sphericity was violated (χ^2 (14) = 521.6, $p< .001$), therefore degrees of freedom were corrected using Huynh-Feldt estimates of sphericity ($\varepsilon = .85$).

The two way mixed ANOVA revealed a significant effect of the within-subject factor 'MRR sequence' ($F(4.24, 3382.89) = 100.16$, $p< .001$, effect size or partial $\eta^2 = .112$), which means that the MRR scores were significantly different for the MRR sequences. Post-hoc analyses showed that the difference between mean MRR scores was significant for most of the pairwise comparisons but was not significant between MRR-ta and the bi-sylabic sequences MRR-pata ($p = 1.000$) and MRR-taka ($p = 1.000$), nor between MRR-pata and MRR-taka ($p = 1.000$). The fastest sequence is MRR-pa ($M = 4.64$, $SD = 0.64$) and the slowest sequence is MRR-pataka ($M = 4.07$, $SD = 0.90$), see Table 2.

The effect of between-subject factor 'age group' was also significant ($F(10, 798) = 29.96$, $p< .001$, effect size or partial $\eta^2 = .273$). The number of syllables per second increased with age for all MRR sequences. As shown in Table 2, MRR sequences increased on average with 1.02 syllables per second from the youngest to the oldest age group.

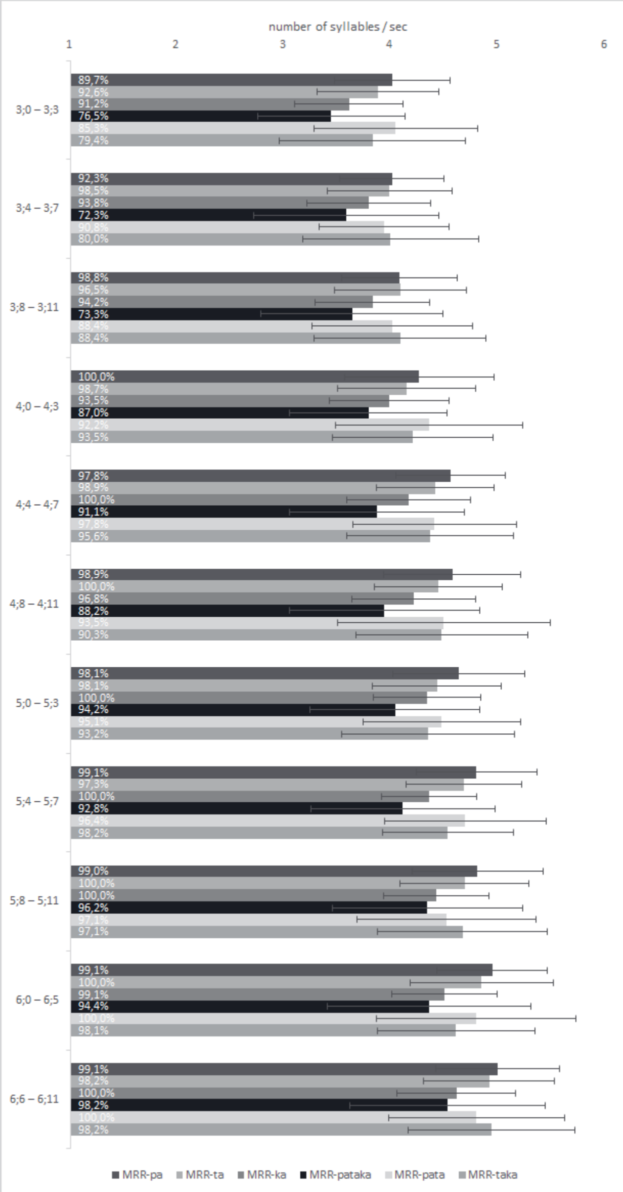

Figure 2. Mean number of syllables / second per age group and per sequence. The percentage of children able to perform the task (in relation to the total number of children of the respective age group) are shown at the beginning of the bars.

The statistical analysis also yielded a significant effect of the between-subject factor 'gender' on overall MRR scores ($F(1, 798) = 9.49$, $p= .002$, effect size or partial $\eta^2 = .012$). As shown in Table 2, MRR scores were higher for boys than for girls for all MRR sequences.

No significant interaction was found between ‘MRR sequences’ and ’age group’ (F(42.39, 3382.89) = 1.181, p = .196, effect size or partial η^2 = .015), ‘MRR sequences’ and ‘gender’ (F(4.24, 3382.89) = 2.172, p = .066, effect size or partial η^2 = .003), ’age group’ and ‘gender’ (F(10, 798) = .876, p = .555, effect size or partial η^2 = .011), or ‘MRR sequences’ and ‘age group’ and ‘gender’(F(42.39, 3382.89) = 1.069, p = .351, effect size or partial η^2 = .013).

Discussion

This study presents normative data of MRR from a large population of Dutch-speaking children aged 3;0 to 6;11 years. Tight ranges of age groups were used to be able to examine the relationship between age and MRR score. A cross-sectional study was performed, using a standardised protocol (Diepeveen et al., 2019). This protocol was used for both the administration of the MRR task and the analysis of the MRR scores. Effects of age, MRR sequence and gender were investigated.

Effect of age on MRR scores

For all MRR sequences the number of syllables per second increased significantly and monotonously with age. No interaction was found between MRR sequence and age group. The MRR score of all sequences was about 1 syllable per second faster for the oldest age group when compared with the youngest age groups. These results are in accordance with the findings in previous studies (Henry, 1990; Icht & Ben-David, 2015; Juste et al., 2012; Modolo et al., 2011; Prathanee et al., 2003; Robbins & Klee, 1987; Zamani et al., 2017). Thus, MRR score increases with age, which is likely to be caused by maturation of the speech motor system (Kent, Kent, & Rosenbek, 1987). Our study included children from 3;0 to 6;11 years of age. Fletcher (1972) found an increase of MRR score in a study with 48 children between the ages of 6;0 and 13;0 years. Wong et al. (2011) demonstrated that MRR score still increases up to the age of 18 years. Between 18 and 60 years of age, Knuijt, Kalf, Van Engelen, Geurts, and de Swart (2019) found stable MRR scores, with a decrease in maximum number of syllables per second from 60 years of age. To conclude, the increase in MRR score seen in the current study in children aged 3 to 7 years is in line with the results of other studies in older children and with studies in adults.

Effect of MRR sequences on MRR scores

The present results show that at the group level typically developing children produce the monosyllabic sequence MRR-ta slower than MRR-pa, and MRR-ka was slower than MRR-pa and MRR-ta. This is in agreement to similar studies with children (Kent et al., 1987; Prathanee et al., 2003; Robbins & Klee, 1987; Rvachew et al., 2006; Thoonen et al., 1996) and adults (Knuijt et al., 2019; Padovani, Gielow, & Behlau, 2009). The production of velar sounds takes longer than the production of alveolar and lip sounds. This might be due to the involvement of physiological factors. The production of /ka/ requires movement of the tongue dorsum, which has a larger mass than the tongue tip, required for pronouncing /ta/; larger inertia of the larger mass, might be (part of) the explanation. The difference in speed between MRR-pa and MRR-ta, with MRR-ta being slower, could be explained by an earlier neurological maturation of jaw and lip movements as compared to tongue tip movements. Lip and jaw movements stabilise earlier in speech motor control development as compared to tongue movement (Terband, Maassen, Van Lieshout, & Nijland, 2011; Terband, Van Brenk, Van Lieshout, Nijland, & Maassen, 2009).

Taken all MRR sequences into account, our results show that MRR-pataka is the slowest sequence, which is probably due to the fact that the motor program of trisyllabic sequences is more complex than mono- or bisyllabic sequences (Wright et al., 2009). Furthermore, it can also be due to physiological aspects as described above. However, contradictory results are described in previous studies. In the studies of Rvachew et al. (2006) and Thoonen et al. (1996) the monosyllabic sequences were slower than the trisyllabic sequences, whereas several other studies found that in their population the MRR-pataka was slower than the monosyllabic sequences (Blech, 2010; Modolo et al., 2011; Wong et al., 2011). Differences in these outcomes are probably due to the use of different protocols. In addition to other studies, our study also investigated the MRR rate of bisyllabic sequences. The mean MRR rate of both bisyllabic sequences was similar to MRR-ta, and thus faster than the production of the monosyllabic sequence MRR-ka. Also, no previous studies have described normative data of MRR scores based on such a large representative sample as in our study. To summarise, the data of our study shows influences from *physiological factors*; larger movement inertia of the tongue body as compared to the tongue tip (i.e. MRR-ta > MRR-ka); from *neurological maturation*; jaw and lips movements stabilise earlier than tongue tip and tongue body movements (i.e. MRR-pa > MRR-ta and MRR-ka):, and *sequence complexity*; sequencing is more complex when more different units must be produced (i.e. MRR monosyllabic sequences > MRR bisyllabic sequences > MRR trisyllabic sequences). How these three factors (physiological factors, neurological maturation and sequence complexity) interact will have to be investigated further.

Gender differences

For all MRR sequences, overall rates were higher for boys than for girls. Prathanee et al. (2003) also found significant higher MRR scores for boys than for girls for /pə/, /tə/, /kə/, and /pə-tə/. Modolo et al. (2011) described older children and found for the 8-year-old children that boys performed faster on /pa/ and girls performed faster on /ta/ and /ka/. For the 9-year-old children these results were different; girls were overall faster than boys. At the age of 10 years girls were still faster than boys, except for the sequences /pataka/. However, other studies (Fletcher, 1972; Henry, 1990; Icht & Ben-David, 2015; Robbins & Klee, 1987; Wong et al., 2011; Zamani et al., 2017) found no differences

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between the performance of boys and girls in similar age ranges as our study. Our findings suggest that at the level of motor speech tasks, less taxing on linguistic skills, boys outperform girls. This is in contrast with studies that found boys showing a slower maturation of the speech motor development (Smith & Zelaznik, 2004), and in contrast with studies concluding that phonological accuracy measures of girls are better than that of boys (Dodd, Holm, Hua, & Crosbie, 2003). However, the results of this study should be interpreted with care; the sample is large, yet the effect size is small (Pek & Flora, 2018). Further research is needed.

Clinical implications and future perspectives

Despite of the ongoing debate on the clinical value of MRR, it has been suggested to have an important function in the assessment of children with MSD, and especially in children with CAS (Murray et al., 2015). Children with MSD show difficulties on MRR tasks when compared to typically developing children, more specifically with the speed(ing up) (Henry, 1990; Thoonen et al., 1996; Wit et al., 1993) and with the sequencing of different speech sounds (Henry, 1990; Thoonen et al., 1996). The studies of Thoonen (1999; 1996) indicate that monosyllabic MRR sequences differentiate children with spastic dysarthria from children with CAS and typically developing children. In addition, MRR can contribute to a first step in differential diagnosis between different types of speech sound disorders (SSD), and especially between different types of MSD. MRR offers insight into possible underlying motor execution impairments (Terband et al., 2019), and is thereby a potential added value in describing a complete speech profile. With only tasks like picture naming and nonword imitation it is not possible to distuingish a speech motor execution impairment from problems in lemma acces, word form selection, and phonological encoding (Van Haaften, Diepeveen, Terband, et al.. 2019).

In this protocol, articulation errors were not included in the analysis. As a result, there are missing values in the norm dataset. However, we consider the remaining data as sufficient to draw conclusions. Studies are currently being conducted to collect MRR data from children with SSD. With the normative data presented in this study and MRR data from children with SSD, clinicians will be able to distinguish typically developing children from children with SSD.

The present study is the largest available study using a standardised administration procedure for the age range 3;0 to 6;11 years. However, the test-retest of the norm group shows a low score for the bi- and tri-syllabic sequences. This is related to a test-retest effect; children were significantly faster on the second test moment because they know what they are expected (Diepeveen, et al. 2019). The normative data of our study is based on a large and representative sample of only Dutch-speaking children. Therefore, the clinical usability of our data in other languages must be discussed. Icht and Ben-David (2014) demonstrated that MRR score is influenced by language differences. They found significant differences in adults in MRR scores between English, Portuguese, Farsi and Greek-speaking persons, with the mean MRR in the Portuguese and Greek sample being faster than the mean MRR in the English sample and the mean MRR in Farsi being slower than in English. Prathanee et al. (2003) found differences in speech rate on an MRR task between English-speaking and Thai-speaking children. They therefore suggest using the norm data of English with English-speaking children and the Thai norms for children who speak Thai. They suggest that the shorter height, and coinciding smaller lung volume, of Thai children when compared to Western children, influences the slower MRR score of Thai children. However, we hypothesise that this explanation is not plausible, since lung volume is related mainly to length of sequence (Pennington et al., 2006) and not to speed of the articulation. Furthermore, Diepeveen et al. (2019) showed that length of sequence is independent of rate. The described language differences can be a possible explanation for the differences found between the results of the present study and other studies, besides differences in sample size and sample representativeness. For example, in the English language the voiceless stops (/p, t, k/) are aspirated in syllable initial position, whereas in Dutch these stops are not aspirated. These findings suggest that reference norms cannot be generalised across languages. In addition, in the past different protocols were used for measuring MRR score (time-by-count or count-by-time measures), making it even more difficult to compare normative data between languages (Diepeveen et al., 2019). We suggest to use this protocol for MRR studies in children for further studies in other languages.

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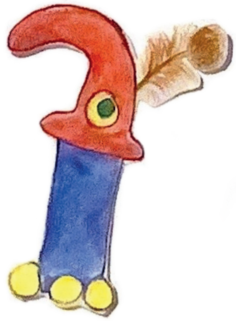
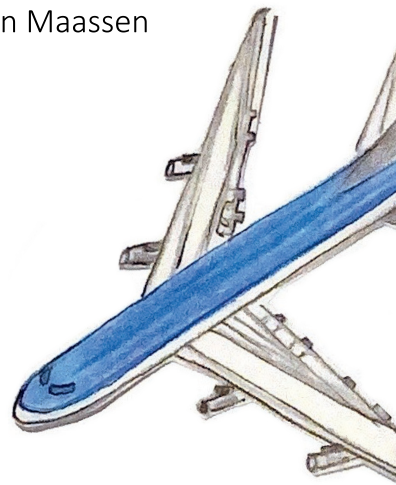


CHAPTER 5

5

Profiling speech sound disorders for clinical validation of the Computer Articulation Instrument

Leenke van Haaften, Sanne Diepeveen, Hayo Terband, Bernadette Vermeij, Lenie van den Engel-Hoek, Bert de Swart, Ben Maassen



Published in American Journal of Speech-Language Pathology 2019 Jul 28(7): 844–856

Abstract

Purpose: The current paper presents data from two studies on clinical groups of children referred for speech assessment. Aim of these studies is to validate the Computer Articulation Instrument (CAI) with the known-group validation method, and to determine the differential diagnostic power of the resulting speech profiles.

Method: Study 1 examined known-group validity by comparing the scores of 93 children diagnosed with speech-language difficulties on the task picture naming (PN) of the CAI with intelligibility judgments given by speech-language pathologists (SLPs). In study 2 the speech profiles of 41 children diagnosed with speech sound disorders (SSD), consisting of four to six factor scores extracted from the four tasks of the CAI: PN; nonword imitation (NWI); word and nonword repetition ((N)WR); and maximum repetition rate (MRR), were validated against clinical judgments of severity of the SSD given by SLPs.

Results: In study 1, a repeated-measures ANOVA revealed a significant effect of intelligibility level on the PN-performance of the CAI and there were highly significant correlations between intelligibility and PN-performance in the expected direction. Neither intelligibility level nor PN-performance were related to nonverbal intelligence and language scores. The ANOVA and a series of t-tests in study 2 revealed significant differences between the moderate and severe group for the CAI-factors based on PN and NWI and the bi- and tri-syllabic sequences of MRR (MRR-BiTri), but not for the factor word and nonword proportion of whole-word variability (PWV) based on WR and NWR, and the mono-syllabic sequences of MRR (MRR-Mono). These results suggest that especially the tasks PN, NWI and MRR-BiTri are most sensitive for diagnosing SSD.

Conclusions: The findings of these two studies support the known-group validity of the CAI. Together with the results of a previous study of our group on reliability and validity (van Haaften et al., submitted), we can conclude that the CAI is a reliable and valid tool for assessment of children with SSD.

Introduction

Children with speech production problems are one of the four subtypes that can be distinguished in children with a specific language impairment (Van Weerdenburg, Verhoeven, & Van Balkom, 2006). They show a specific profile as compared to the other subtypes of children with language impairments: difficulties with lexical-semantic abilities, with auditory conceptualization, or with verbal sequential memory (Van Weerdenburg et al., 2006). Recently, Bishop et al. (2017) proposed to use the term Developmental Language Disorder (DLD) when a language disorder was not associated with a known biomedical etiology. They state that DLD is a heterogeneous category that encompasses a wide range of problems, including expressive phonological problems. Phonological problems in preschoolers that are not accompanied by other language problems do not meet the criteria for DLD. Therefore, Bishop et al. (2017) propose to use the more general term Speech Sound Disorder (SSD) for such cases. SSD is an umbrella term that includes expressive phonological problems and problems with speech production that have motor or physical origins, or involve misarticulations such as a lisp, where a sound is produced in a distorted way without losing the contrast with other sounds. Children with SSD are one of the most common clinical populations for speech-language pathologists (SLPs; Mullen & Schooling, 2010); the reported prevalence is highly variable, ranging from 2.3 to 24.6% (Eadie et al., 2015; Law et al., 2000). They form a heterogeneous group, showing variability in severity, etiology, proximal causes, speech error characteristics and response to treatment (Dodd, 2011).

There are several widely recognized classification systems for SSD featuring a variety of approaches, i.e., etiology, descriptive-linguistics, and psycholinguistic and psychomotor processing (Waring & Knight, 2013). In current practice, symptom patterns form the basis of diagnostic classification (Dodd, 1995b, 2014). The Speech Disorders Classification System (SDCS) described by Shriberg et al. (2017), divides SSD into three classes, based on etiology: Speech Delay (SD), Speech Errors (SE) and Motor Speech Disorder (MSD; including dysarthria, childhood apraxia of speech (CAS) and motor speech disorder – not otherwise specified). Examples of symptoms of MSD include slow speech rate, distorted substitutions of speech sounds, increased difficulty with multisyllabic words and prosodic errors. Yet, there is no validated list of diagnostic patterns for differential diagnosis of SSDs. For example, one of the speech symptoms that is described for different types of SSD is inconsistency of speech errors. From a phonological point of view, high inconsistency of speech errors could indicate an unstable phonological system, also called a phonological planning deficit (Dodd, 1995a; Macrae, Tyler, & Lewis, 2014) or unstable lexical representations (Sosa & Stoel-Gammon, 2012). However, inconsistency is also a characteristic of CAS (Davis, Jakielski, & Marquardt, 1998; Forrest, 2003; Iuzzini-Seigel, Hogan, & Green, 2017). In the latter case, inconsistency is explained by an unstable motor system (articulo-motor planning and programming). Thus, the same symptom can refer to different underlying deficits, and the same deficit can result in different symptoms, leading to a wide variety of symptoms within subtypes and much symptomatic overlap between subtypes of SSDs. Therefore, in clinical practice a re-orientation from behavioral diagnostics to process-oriented diagnostics is required in order to reveal the proximal causes of SSD (Terband & Maassen, 2012).

Psycholinguistic and psychomotor models give a conceptual basis to analyze speech disorders and form the basis for a process-oriented diagnostic classification system based on the identification of the breakdown in the chain of sequential and parallel speech processes (Baker et al., 2001). Rather than categorization of SSDs based on single symptoms or sets of symptoms, process-oriented diagnostics primarily focus on speech profiles comprising clustered symptoms that can be interpreted in terms of the underlying speech production processes. An example of a psycholinguistic processing model is the model described by Levelt (1989), in which *conceptualizing a preverbal message*, either from memory or from perception, is the first process in speaking. The next process is formulating a word or sentence, driven by two steps of lexicalization: selecting a lemma, containing meaning and grammatical information, and the corresponding lexeme or word form, which forms the input for the next stage of *phonological encoding*. Phonological encoding entails specifying the sequence of speech sounds together with their syllabic and prosodic structure. These syllables are the basic units of articulo-motor planning and programming. The final process of actually performing the articulatory movements is *execution*, resulting in an acoustic speech signal (Maassen & Terband, 2015). Levelt, Roelofs, and Meyer (1999) validated this processing model with normal speech production data, and Nijland (2003) further elaborated on the planning, execution, and monitoring stages of the model, and applied it to analyses of SSD. By conducting different speech experiments in children with CAS, Nijland (2003) could conclude that both phonetic planning and motor programming are deviant in children with CAS. Levelt's model is relevant for analyzing SSD, because of the stage's lexeme retrieval, phonological encoding, and self-monitoring, which are the processes underlying consistent and inconsistent phonological disorder. MSDs, of which CAS and dysarthria are the main diagnostic categories, can be described by means of the motor planning, programming, and execution processes. However, the main objective of a process-oriented approach is not to categorize, but to give a complete characterization of the speech profile, such that underlying processing deficits can be identified. Insight into the deficits that might be the underlying causes of the child's difficulty requires an extensive analysis of a child's performance on a range of speech tasks that reflect different levels of processing. Based on these premises, the Computer Articulation Instrument (CAI) was developed (Maassen et al., 2019). The CAI consists of a battery of speech production tasks and is based on a series of studies of Dutch children with developmental and acquired speech sound disorders (Nijland, Maassen, & van der Meulen, 2003; Nijland, Maassen, van der Meulen, et al., 2003; Nijland, Terband, & Maassen, 2015; Thoonen et al.,1999; 1994). The CAI has a modular structure, and it provides an interactive administration and scoring of four speech tasks. The tasks comprise (1) picture naming (PN), (2) nonword imitation (NWI), (3) word and nonword repetition (WR and NWR) and (4) maximum repetition rate (MRR), thereby covering phonological and speech motor skills.

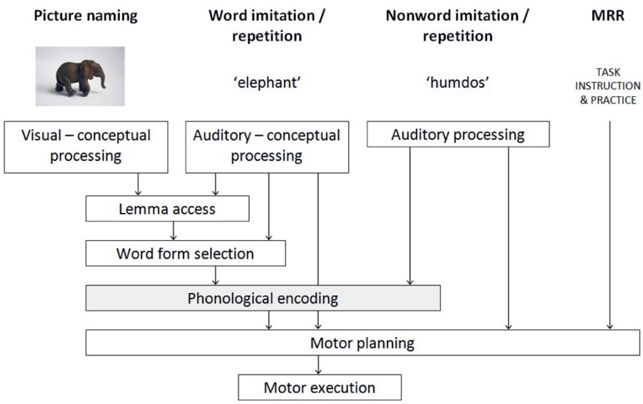


Figure 1. The speech production processes assessed in the four tasks of the Computer Articulation Instrument (Maassen & Terband, 2015; Figure 15.2). MRR = maximum repetition rate.

As demonstrated in Figure 1, picture naming taps into the whole chain of speech processes, from preverbal visual-conceptual processing to lemma access, word-form selection, phonological encoding, motor planning, and articulation (motor execution) (Maassen & Terband, 2015). During nonword imitation a child is asked to reproduce nonwords (or nonsense words). In contrast to picture naming, a child cannot revert to its lexicon during this task and thus the child either needs to analyze the phonological structure of the nonword directly, addressing the phonological decoding and encoding system, or follows the auditory-to-motor-planning pathway. In word and nonword repetition a child is asked to repeat a word or nonword 5 times. This task aims to assess variability in speech production, which occurs when a child uses multiple productions of the same word or nonword. *Maximum repetition rate* is a pure motor task (articulo-motor planning and programming) and does not require any knowledge of words, syllables, or phonemes. The evaluation of speech production in the CAI is based on phonetic transcriptions and acoustic measurements. Both the tasks and speech analyses are computer-implemented (Van Haaften et al., 2019). Rather than focusing on single diagnostic markers, two types of analyses are conducted within the CAI: (1) objective and quantitative assessment of symptoms, and (2) contrasting severity of symptoms across tasks. The outcome of this assessment battery is a speech performance profile that can be interpreted as characteristics of breakdown in underlying processes. Normative data from 1,524 children in the age range of 2;0 to 6;11 years have been collected, such that performance on the CAI as a whole, as well as the profile of performances on the different tasks, can be quantified in percentile scores which allows for interpretation in terms of strengths and weaknesses (Maassen et al., 2019).

In a previous study of our research group, we assessed the psychometric properties of the CAI, including reliability and construct validity (Van Haaften et al., 2019). Overall, sufficient to good values were found for interrater reliability, but intraclass correlation coefficients (ICCs) on test-retest reliability were low, probably due to better performance at retest reflecting a test-retest learning effect in addition to normal development. The study also described two aspects of construct validity. The first aspect, criterion validity, was confirmed by clear and significant age trends in CAI-parameters in a large sample of typically developing children aged between 2 and 7 years. The second aspect of construct validity, structural validity, was assessed by factor analysis and correlations. Factor analyses on a total number of 20 parameters revealed five meaningful factors: picture naming (PN); segmental quality of nonword imitation (NWI-Seg); quality of syllabic structure of nonword imitation (NWI-Syll); word and nonword proportion of whole-word variability (PWV), based on WR and NWR; and maximum repetition rate (MRR). Weak correlations were found between CAI factor scores, indicating the independent contribution of each factor to the speech profile.

Further steps are needed in the validation process of the CAI. The ultimate goal is to assess the strengths of the five CAI factors in identifying breakdown of speech processes in children with SSD (process-oriented diagnostics), which will be described in future papers. The more immediate step, determining known-group validity, is presented in the current study. Known-group validity is a third aspect of construct validity and refers to the degree to which a measure is sensitive to differentiate between subgroups that are hypothesized to have different scores (Portney & Watkins, 2009). To assess this aspect of construct validity of the CAI, this paper presents data from two studies on clinical groups of children with speech language impairments and SSD. The aim of *study 1* is to determine known-group validity by comparing the scores of children with speech language impairments, as diagnosed on the basis of language and intelligence tests, on one task of the CAI (picture naming) with intelligibility judgments given by SLPs. *Study 2* aims to determine the diagnostic power of all four tasks of the CAI by comparing the five CAI-factors: PN, NWI-Seg, NWI-Syll, PWV, and MRR (see also Table 4) with a severity judgment of the speech difficulties (mild, moderate, severe) of children with SSD.

Study 1

The first study was designed to validate the scores on the task picture naming of the CAI with intelligibility judgments (good, moderate, poor) in children diagnosed with speech language impairments. For this study the parameter 'percentage of consonants correct' of the task picture naming is used (PN-PCC), and nonverbal intelligence and language tests for the speech language impairment-diagnosis.

Method

Ethics, Consent and Permissions

The research ethics committee of the Radboud University Nijmegen Medical Centre stated that this study does not fall within the remit of the Medical Research Involving Human Subjects Act (WMO) (file number CMO 2016-2985). Therefore, this study can be carried out (in the Netherlands) without an approval by an accredited research ethics committee. Informed consent was obtained from all parents or guardians.

Participants

A total of 93 children aged between 3;0 and 4;0 years participated in this study (see Table 1). The sample consisted of 73 boys and 20 girls, representative for the gender distribution in children with speech language impairments. All children attended one of the intervention centers for preschoolers with speech language impairments at the NSDSK, a specialized diagnostic and intervention center for children with hearing loss or speech language impairments. Before admission to the center, these children had been referred to an Audiology Center (AC) by their family doctor or health care physician on the basis of suspected speech language impairment. At the AC, nonverbal intelligence is assessed by a psychologist, receptive and expressive language tests are administered by an SLP, and hearing status is evaluated by audiometry. Children meet the criteria for referral to a speech language impairment intervention center when they have difficulties in language production and / or language comprehension and / or when their speech is highly unintelligible. Admission takes place if they have a score of at least 1.5 standard deviation (SD) below the mean on at least one standardized, norm-referenced language test. Children with 25 dB or more hearing loss were excluded for this study.

Nonverbal intelligence and language skills were assessed within a time period ranging from three months before until three months after the start of the intervention. If language scores were missing or were older than three months at the start of the intervention, language performance was assessed by the SLP of the intervention center within three months after the intervention started.

Materials and procedures

Nonverbal intelligence was assessed with the Snijders-Oomen Nonverbal Intelligence Test 2½-7-Revised (SON-R 2 ½ - 7) (Snijders et al., 2003), yielding a nonverbal intelligence quotient (NVIQ). Vocabulary was tested with the Dutch version of the Peabody Picture Vocabulary Test-III (PPVT-III-NL) (Schlichting, 2005), yielding a vocabulary quotient (QPPVT). The Schlichting Test for Language Comprehension and Language Production (Schlichting & Spelberg, 2010a; 2010b) was used to measure receptive (receptive language quotient: RLQ) and expressive language skills (sentence and word production quotient: SWQ). These norm-based standard scores or Q-scores (M = 100, SD = 15) of each test were used for the analyses.

In addition to the measures for nonverbal intelligence and language, the CAI was administered to all the children (Maassen et al., 2019). For this study the task picture naming of the CAI was used. The task was administered by SLPs of the speech language impairment early intervention group, specifically trained in the administration of the CAI. Picture naming contains 60 words, covering the full inventory of vowels, consonants, clusters, and syllable structures of the Dutch language. For this study, the parameter picture naming-percentage consonants correct (PN-PCC) was used for analyses. Individual's PN-PCC scores were transformed into z-scores by subtracting the mean of the normative group and dividing by the standard deviation of the study group; this was done for three age-groups (36-39, 40-43, 44-47 months) separately. The reason for dividing by the standard deviation of the study group rather than the standard deviation of the norm group was, that the former was approximately three times as large as the latter (18.9 compared to 6.3). Applying the broader confidence intervals of the study group yields the more conservative estimates. Z-scores were transformed into Q-scores (formula: $Q = 100 + 15*z$) to make them comparable to the cognitive and language scores NVIQ (nonverbal intelligence quotient), QPPVT (vocabulary quotient), RLQ (receptive language quotient), and SWQ (sentence and word production quotient).

For each child, the SLP rated the intelligibility on a three-level scale: good, moderate or poor. The same method is used in the study of Lohmander, Lundeborg, and Persson (2016). Twenty-two children were rated with a 'good' intelligibility, 46 were rated as 'moderate', and 25 children were rated with a 'poor' intelligibility.

Table 1. Number of children per age category and completed tests.

Age category	N	Boys	Girls	NVIQ	QPPVT	RLQ	SWQ	PN-PCC-Q
36-39 months	29	23	6	26	25	23	22	29
40-43 months	35	28	7	32	33	21	22	35
44-47 months	29	22	7	26	28	19	17	29
total	93	73	20	84	86	63	61	93
% missing values				9.7%	7.5%	32.3%	34.4%	0%

Note. NVIQ = nonverbal intelligence quotient; QPPVT = Peabody Picture Vocabulary Test, vocabulary quotient; RLQ = receptive language quotient; SWQ = rentence and word production quotient; PN-PCC-Q = CAI picture naming percentage consonant correct quotient.

Statistical analysis

To test the hypothesis that there is a difference in mean Q-scores of the nonverbal intelligence test, language tests and CAI for the three intelligibility levels, a one-way repeated-measures ANOVA was conducted with Q-score as dependent variable, test-instrument as within-subject factor (five levels: NVIQ, QPPVT, RLQ, SWQ and PN-PCC-Q) and intelligibility level as between-subject factor (three levels: good, moderate, poor). Mauchly's test of Sphericity was conducted to test the hypothesis that the variances of differences between conditions are equal. Bonferroni correction was applied for post hoc comparisons. A series of ANOVAs was performed to evaluate differences between Q-scores for the three levels of intelligibility. Levene's test of Equality of Error Variances was conducted to test the homogeneity of variance assumption. Bonferroni correction was applied for post hoc comparisons. Correlations between Q-scores and intelligibility levels were calculated with Spearman's rank correlation coefficients, and correlations between the Q-scores of the different tests were calculated with Pearson's' rank correlation coefficients. Missing values were replaced by the mean per age group (i.e., mean imputation method). All statistical analyses were performed using SPSS version 20 for Windows (SPSS Inc., Chicago, IL, USA).

Results

Mean Q-scores and standard deviations of all tests for the three intelligibility levels are shown in Table 2. Comparing the profiles of Q-scores across tests, it was found that in the levels moderate and poor intelligibility, on average, children achieved the highest scores on the nonverbal intelligence test, followed by the vocabulary test, the receptive language test, and the expressive language tests. Lowest Q-scores were obtained for PN-PCC-Q. In contrast, children with a 'good' intelligibility also showed the highest scores for the nonverbal intelligence, but in this group PN-PCC-Q was higher than the language Q-scores, which were approximately equal. Thus, of all Q-scores, PN-PCC-Q shows the largest decrease between groups from good to poor intelligibility.

Table 2. Mean Q-scores for the nonverbal intelligence, language and speech tests.

Intelligibility score	N	NVIQ		QPPVT		RLQ		SWQ		PN-PCC-Q	
		M	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*
Good	22	102.8	11.6	84.7	18.8	78.6	11.3	78.2	10.4	92.5	5.99
Moderate	46	99.9	11.2	89.9	16.6	80.9	13.7	74.0	9.50	73.4	11.3
Poor	25	100.9	11.4	90.9	18.2	82.2	14.0	71.3	9.33	62.5	14.1

Note. NVIQ = nonverbal intelligence quotient; QPPVT = Peabody Picture Vocabulary Test, vocabulary quotient; RLQ = receptive language quotient; SWQ = sentence and word production quotient; PN-PCC-Q = CAI picture naming percentage consonant correct quotient.

A one-way repeated-measures ANOVA was conducted with the Q-scores of the five test instruments as repeated measures, and intelligibility level as between subject variable. Mauchly's test indicated that the assumption of sphericity had been violated, χ^2 (9) = 58.9, $p < .001$, therefore degrees of freedom were corrected using Huynh-Feldt estimates of sphericity ($\varepsilon = .78$). The results show that the within-subject factor 'test instrument' was significant, $F(3.10, 278.96) = 79.78$, $p < .001$, effect size or partial $\eta^2 = .47$, which means that the scores on the test instruments were significantly affected by intelligibility level. The between-subject factor 'intelligibility level' was marginally significant, $F(2, 90) = 3.09$, $p = .051$, effect size or partial $\eta^2 = .064$. Post-hoc analyses showed that the difference of mean Q-scores was not significant between level 'good' and 'moderate' ($p = .217$), nor between level 'moderate' and 'poor' ($p = .556$) but was significant between level 'good' and 'poor' ($M = 6.78$, $SE = 2.47$, $p = .022$). In addition, there was a significant interaction between intelligibility levels and 'test instrument', $F(6.20, 278.96) = 10.00$, $p < .001$, effect size or partial $\eta^2 = .18$. To further examine this interaction, a series of ANOVAs was conducted to test the differences between the three intelligibility levels for the Q-scores of each test instrument separately. There was no significant difference between intelligibility levels for NVIQ ($F(2, 90) = 0.47$, $p = .626$), QPPVT ($F(2, 90) = 0.87$, $p = .421$), RLQ ($F(2, 90) = 0.43$, $p = .650$) or SWQ ($F(2, 90) = 3.07$, $p = .051$). For the latter, marginally significant factor SWQ, post-hoc analyses revealed a significant mean difference between level 'good' and 'poor' ($p = .047$), and no significant mean differences between level 'good' and 'moderate' ($p = .276$), or 'moderate' and 'poor' ($p = .795$). For PN-PCC-Q, the Levene's test for equality of variances was significant, indicating that the requirement of homogeneity of variance was violated. Therefore, the Welch F-ratio was calculated, showing that the difference in mean PN-PCC-Q between intelligibility levels was significant ($F(2, 51.28) = 69.48$, $p = <.001$).

Table 3 shows correlations between intelligibility and Q-scores. A strong, significant correlation was found between PN-PCC-Q and intelligibility (Spearman's r (93) = .69, $p < .001$), which is in the expected direction: PN-PCC-Q decreases when the intelligibility level decreases. No other Q-scores, not even the expressive language score SWQ, correlated significantly with intelligibility nor with PN-PCC-Q. There were weak, significant correlations between the outcome of the nonverbal intelligence test and language tests, and moderate correlations among the language tests, with correlations between RLQ and SWQ, and between QPPVT and RLQ being moderate, and between QQPVT and SWQ being weak. No significant correlations were found between PN-PCC-Q and the Q-scores of the nonverbal intelligence test and language tests. Inspection of the scatter plots did not reveal any outliers.

Table 3. Spearman's rank correlations and Pearson's rank correlations between intelligibility levels and Q-scores and between Q-scores (*N* = 93).

		Intelligibility level	NVIQ	QPPVT	RLQ	SWQ	PN-PCC-Q
Intelligibility level	Spearman's *r*	1	.027	-.14	-.11	.20	.69**
NVIQ	Pearson's *r*		1	.36**	.31**	.35**	.10
QPPVT	Pearson's *r*			1	.52**	.36**	-.22
RLQ	Pearson's *r*				1	.48**	-.15
SWQ	Pearson's *r*					1	.21
PN-PCC-Q	Pearson's *r*						1

Note. NVIQ = nonverbal intelligence quotient; QPPVT = Peabody Picture Vocabulary Test, vocabulary quotient; RLQ = receptive language quotient; SWQ = sentence and word production quotient; PN-PCC-Q = CAI picture naming percentage consonant correct quotient.
**. Correlation of factor scores is significant at the .01 level (2-tailed).
*. Correlation of factor scores is significant at the .05 level (2-tailed).

5

Study 2

The second study aims to determine the diagnostic power of all four tasks of the CAI. For this, the relation between the five CAI-factors (picture naming (PN), nonword imitation segmental (NWI-Seg), nonword imitation syllable structure (NWI-Syll), word and nonword proportion of whole-word variability (PWV), maximum repetition rate (MRR)) and clinical judgments of severity of the speech disorder by the SLPs is investigated.

Methods

Ethics, Consent and Permissions

The ethics approval for study 1 also applied to study 2.

Participants

The participants in study 2 were 41 children with an age range from 3;0 to 6;4 years, 26 boys and 15 girls. For this study children with SSD were recruited from several institutions: nineteen children from primary healthcare services, one child from an AC and 21 children from a special school for language- and hearing-impaired children. All parents or caregivers were given an information letter. After obtaining the signed parental consent form, the child was included in the study.

The parents or caregivers of all 41 children were asked to provide information about the children's hearing status. They were asked whether the child had a history of hearing problems, if hearing problems had been recorded during the regular governmental (neonatal) hearing screening, and if available, if they could provide us with hearing acuity data (pure-tone thresholds). Thirty children passed a bilateral hearing screening at 20 dB. Parents or caregivers of the other eleven children reported no history of hearing problems and no hearing problems recorded during the regular governmental (neonatal) hearing screening.

Prior to the procedures of this study, a speech diagnosis was reported by the SLP of the child, based on clinical observation and a standard speech-language protocol, including standardized language tests. Speech was observed with different instruments. Until now, for the Dutch language no standardized and normalized speech assessment is available. All children were diagnosed with SSD, most of them (*N* = 36) with a phonological disorder (PD), two children with CAS, and three children with an unknown diagnosis because no details were available about the children's speech apart from the fact that their SSD was severe. Differential diagnosis was part of the clinical reasoning process of the SLP and was done based on diagnostic criteria described in studies like Forrest (2003) and Shriberg and Kwiatkowski (1994).

Materials and procedures

For the present study, all participants were tested on their speech skills with the CAI. All four tasks (PN, NWI, (N)WR and MRR) were administered. Both the administration of the tests and the analyses of the speech are computer implemented. Table 4 shows the parameters used to assess task performance; a detailed description of the CAI and these parameters, as well as a description of the normative dataset, is presented in Maassen et al. (2019) and Van Haaften et al. (2019); for all parameters percentile scores can be determined. A factor analysis on all 20 parameters of the normative data, obtained from a total number of 1,524 children, yielded five factors; 1) picture naming (PN), 2) nonword imitation segmental (NWI-Seg), 3) nonword imitation syllable structure (NWI-Syll), 4) proportion word variability (PWV) of words and nonwords, and 5) maximum repetition rate (MRR) (Van Haaften et al., submitted). For the present study, factor scores were calculated based on the factor weights obtained from this factor analysis. Because there were many missing values in the MRR-task (see below), separate factor scores were calculated on only the monosyllabic MRR-sequences (/papa../, /tata../, /kaka../, yielding factor MRR-Mono), and the bi- (/pata../, /taka../) and tri-syllabic (/pataka../) sequences, yielding factor MRR-BiTri.

Table 4. CAI parameters per speech task and extracted factors.

Task	Factor	Parameter	
PN	PN	PCCI	Percentage of consonants correct in syllable-initial position
		PVC	Percentage of vowels correct
		Level 5	Percentage of correct consonants /l/ and /R/
		RedClus	Percentage of reduction of initial consonant clusters from 2 consonants to 1
		CCVC	Percentage of correct syllable structure CCVC (C=consonant, V=vowel)
NWI	NWI-Seg	PCCI	Percentage of consonants correct in syllable-initial position
		PVC	Percentage of vowels correct
		Level 4	Percentage of correct consonants /b/, /f/ and / ʋ /
		Level 5	Percentage of correct consonants /l/ and /R/
		CVC	Percentage of correct syllable structure CVC
	NWI-Syll	RedClus	Percentage of reduction of initial consonant clusters from 2 consonants to 1
		CCVC	Percentage of correct syllable structure CCVC
WR	PWV	PWV Word	Proportion of whole-word variability – word repetition
NWR		PWV Nonword	Proportion of whole-word variability – nonword repetition
MRR	MRR-Mono	MRR-pa	Number of syllables per second of sequence /pa/
		MRR-ta	Number of syllables per second of sequence /ta/
		MRR-ka	Number of syllables per second of sequence /ka/
	MRR-BiTri	MRR-pataka	Number of syllables per second of sequence /pataka/
		MRR-pata	Number of syllables per second of sequence /pata/
		MRR-taka	Number of syllables per second of sequence /taka/

Prior to the administration of the CAI, severity of the SSD was judged by the child's SLP (*N* = 11) on a severity scale with three categories: mild, moderate and severe, following the categories proposed by Dodd (1995c). An SLP rated the severity of a speech sound disorder as *mild* when a child is mostly intelligible in spontaneous speech, but errors are obvious and distracting from content. The severity was rated *moderate* when single words are often intelligible in context, but connected speech is often difficult to understand, particularly out of context. The category *severe* was rated when most utterances are unintelligible on the first meeting. Also, the persistence of the speech disorder and the consequences on communication abilities were taken into account when rating severity. The category 'moderate' was scored for 14 children and 27 children were scaled as 'severe'. None of the children was scaled as having a 'mild' speech disorder. Therefore, the statistical analyses of this study are based on two severity categories: moderate and severe. Table 5 shows the distribution of the participants in the three severity categories by speech diagnosis.

The tasks of the CAI were administered by (student) SLPs specifically trained in the administration of the CAI.

Table 5. Speech diagnosis by severity categories.

	Speech disorder			
Severity category	PD	CAS	Unknown	Total
Mild	0	0	0	0
Moderate	13	1	0	14
Severe	23	1	3	27
Total	36	2	3	41

Note. PD = phonological disorder; CAS = childhood apraxia of speech

Statistical analyses

The factor PWV had two missing values, and these were replaced by the overall PWV mean (M = -1.20) (i.e., mean imputation method). Much more missing data were observed for the MRR-tasks, due to speech-motor difficulties and/or shyness or inattentiveness of the child; also, a few recordings could not be analyzed due to the low acoustic quality. Out of the total number of 41 children, only 23 produced at least two mono-syllabic sequences correctly (44% missing), and only 9 out of these 23 (amounting to a total of 78% missing data) at least two of the bi- or tri-syllabic sequences. Because of this large number of missing values, no imputation was applied, but a separate analysis was conducted instead on the group of 23 children. The 14 children who were not able to produce the bi- or tri-syllabic sequences were assigned the lowest z-score, such that failure to produce these sequences was marked as poor performance. One-way repeated measures ANOVAs were conducted to test the hypothesis that there is a difference in CAI-factors for the two severity categories, comprising two levels: 'moderate' and 'severe'. Because of the missing data in factors MRR-Mono and MRR-BiTri, the first analysis was conducted on the four remaining factors: PN, NWI-Seg, NWI-Syll, PWV. Subsequently, a one-way repeated-measures ANOVAs was conducted with six CAI-factors, including MRR-Mono and MRR-BiTri. Mauchly's tests of Sphericity were conducted to test the hypothesis that the variances of differences between conditions are equal. Next, if in the ANOVA either severity level or the interaction between severity level and CAI-factor was significant, a series of independent t-tests were conducted to evaluate the difference in factor-scores between the moderate and severe group for each of the four or six CAI-factors separately. Levene's test of Equality of Error Variances was conducted to test the homogeneity of variance assumption. Correlations between CAI-factors and severity categories were calculated by Spearman's rank correlation coefficients (r), and correlations between the CAI-factors were assessed by calculating Pearson's' rank correlation coefficients (r). All statistical analyses were performed using SPSS version 20 for Windows (SPSS Inc., Chicago, IL, USA).

Results

Table 6 shows that, on average, children with a speech disorder of moderate severity have higher factor-scores on PN, NWI-Seg, NWI-Syll and PWV, than children with a severe speech disorder. For the children with a severe speech disorder mean factor scores ranged from -1.13 to -1.72, and for the children with moderate severity between -0.18 and -1.07. Thus, all mean scores were below the population average.

Table 6. Means and standard deviations of the factor-scores of four CAI-factors per severity category.

Severity category		**PN**	**NWI-Seg**	**NWI-Syll**	**PWV**
Moderate	N	14	14	14	14
	Mean	-1.07	-0.88	-0.18	-1.03
	SD	0.52	0.83	0.75	0.81
Severe	N	27	27	27	27
	Mean	-1.72	-1.69	-1.13	-1.29
	SD	0.56	0.73	0.80	0.67
Total	N	41	41	41	41
	Mean	-1.45	-1.42	-0.81	-1.20
	SD	0.62	0.85	0.89	0.72

Note. PN = factor score of all parameters of picture naming; NWI-Seg = factor score of the segmental parameters of nonword imitation; NWI-Syll = factor score of the syllable structure parameters of nonword imitation; PWV = factor score of the two PWV parameters of word and nonword repetition.

First, a one-way repeated-measures ANOVA with the four CAI-factors PN, NWI-Seg, NWI-Syll and PWV was conducted. Mauchly's test indicated that the assumption of sphericity had been violated, χ^2 (5) = 15.13, p = .010, therefore degrees of freedom were corrected using Huynh-Feldt estimates of sphericity (ε = .91). The results show that the within-subject factor 'CAI-factors' was significant, F(2.74, 106.96) = 18.29, p < .001, effect size or partial η^2 = .32, indicating that the factor-scores of the CAI were significantly affected by the severity of the speech disorder. The between-subject factor 'severity category' was also significant, F(1, 39) = 11.98, p = .001, effect size or partial η^2 = .24; there was a significant difference in factor-scores between the children with a moderate and severe speech disorder. There was also a significant interaction between CAI-factors and severity categories, F(2.74, 106.96) = 3.70, p = .017, effect size or partial η^2 = .087. To further examine this interaction, a series of independent t-tests was conducted to test the differences between the two severity categories for each CAI-factor separately. Significantly lower factor-scores for the severe versus moderate group were found for PN (t(39) = 3.62, p = .001), NWI-Seg (t(39) = 3.21, p = .003), and NWI-Syll, t(39) = 3.67, p = .001. No significant difference was found between the mean factor-scores of the moderate and severe group for the CAI-factor PWV (t(39) = 1.11, p = .27).

Table 7. Means and standard deviations of the factor-scores of six CAI-factors per severity category

Severity category		PN	NWI-Seg	NWI-Syll	PWV	MRR-Mono	MRR-BiTri
Moderate	N	10	10	10	10	10	10
	Mean	-1.05	-0.80	-0.29	-0.97	-1.12	-1.15
	SD	0.46	0.69	0.80	0.85	0.89	1.56
Severe	N	13	13	13	12	13	13
	Mean	-1.52	-1.45	-1.14	-1.28	-0.60	-2.31
	SD	0.67	0.84	0.75	0.75	0.85	0.06
Total	N	23	23	23	22	23	23
	Mean	-1.31	-1.17	-0.77	-1.14	-0.82	-1.81
	SD	0.62	0.83	0.87	0.80	0.89	1.16

Note. PN = factor score of all parameters of picture naming; NWI-Seg = factor score of the segmental parameters of nonword imitation; NWI-Syll = factor score of the syllable structure parameters of nonword imitation; PWV = factor score of the two PWV parameters of word and nonword repetition; MRR-Mono = factor score of the monosyllabic items of maximum repetition rate parameters; MRR-BiTri = factor score of the bi- and trisyllabic items of maximum repetition rate parameters.

The second one-way repeated-measures ANOVA was conducted with all six CAI-factors, including MRR-Mono and MRR-BiTri, on 23 children with complete data on these factors (Table 7). A one-way repeated-measures ANOVA was conducted with these CAI-factors (PN, NWI-Seg, NWI-Syll, PWV, MRR-Mono and MRR-BiTri). Mauchly's test indicated that the assumption of sphericity had been violated, χ^2 (14) = 32.99, p = .003, therefore degrees of freedom were corrected using Huynh-Feldt estimates of sphericity (ε = .87). Like the analysis with four factors, the results show that the six factor-scores of the CAI were significantly affected by the severity level of the speech disorder; the within-subject factor 'CAI-factors' was significant, $F(4.3, 90.9) = 6.40$, $p < .001$, effect size or partial η^2 = .23. The between-subject factor 'severity category' was also significant, $F(1, 21) = 4.60$, $p = .04$, effect size or partial η^2 = .18, as well as the interaction between CAI-factors and severity categories, $F(4.3, 90.9) = 4.17$, $p = .003$, effect size or partial η^2 = .17. To further examine this interaction, independent t-tests were conducted to test the differences between the two severity categories for all six factors. For NWI-Syll ($t(21) = 2.61$, $p = .016$) and MRR-BiTri ($t(0.0) = 2.35$, $p = .043$) the differences between the mean factor-scores of the moderate and severe groups reached significance. No significance difference was found between the severity groups for PWV. For PN and NWI-Seg the differences were only marginally significant in this second analysis, most likely due to less power as compared to the first analysis. It is remarkable that there is no difference between moderate and severe groups for MRR-Mono, but a large, significant difference for MRR-BiTri. We will come back to this issue in the general discussion.

Table 8 shows correlations between severity category and CAI-factors. Moderate, significant correlations were found between severity category and PN, NWI-Seg and NWI-Syll. Children with a severe disorder had lower CAI-factor scores. The factor scores of PN, NWI-Seg, and NWI-Syll showed strong correlations; the correlations with PWV and MRR-BiTri were weak to moderate. No significant correlations were found between MRR-Mono and any other CAI-factor.

Table 8. Spearman's rank correlations and Pearson's correlations between severity category and CAI-factors and between CAI-factors.

		Severity category	PN	NWI-Seg	NWI-Syll	PWV	MRR-Mono	MRR-BiTri
	N	41	41	41	41	41	23	23
Severity category	Spearman's *r*	1	-.53**	-.50**	-.50**	-.19	.28	-.32
PN	Pearson's *r*		1	.80**	.81**	.39*	-.09	.41*
NWI-Seg	Pearson's *r*			1	.68**	.60**	.12	.53*
NWI-Syll	Pearson's *r*				1	.51**	-.03	.44*
PWV	Pearson's *r*					1	-.07	.21
MRR-Mono	Pearson's *r*						1	-.02
MRR-BiTri	Pearson's *r*							1

Note. PN = factor score of all parameters of picture naming; NWI-Seg = factor score of the segmental parameters of nonword imitation; NWI-Syll = factor score of the syllable structure parameters of nonword imitation; PWV = factor score of the two PWV parameters of word and nonword repetition; MRR-Mono = factor score of the monosyllabic items of maximum repetition rate parameters; MRR-BiTri = factor score of the bi- and trisyllabic items of maximum repetition rate parameters.
**. Correlation of factor scores is significant at the .01 level (2-tailed).
*. Correlation of factor scores is significant at the .05 level (2-tailed).

Discussion

The CAI is a computer-based assessment for speech production with a range of speech tasks that reflect different levels of processing (phonological and speech motor skills), and it provides normative data based on a sample of 1,524 children in the age range of 2;0 to 6;11 years. A previous study on psychometric characteristics of the CAI revealed sufficient interrater reliability, test-retest reliability and construct validity (Van Haaften et al., 2019). In this current paper we report known-group validity, based on the outcome of two studies in children with speech language impairment and SSD.

The known-group validity of the CAI was supported by the results of study 1. These results confirm the hypothesis that PN-PCC-Q is significantly affected by intelligibility level. There was a significant difference between the intelligibility levels with respect to the PCC parameter of the task picture naming of the CAI and there was a highly significant correlation between the intelligibility levels and PN-PCC-Q in the expected direction. Correlations between PCC and intelligibility measures were also found in previous studies (Lagerberg et al., 2015; McLeod, Harrison, & McCormack, 2012; Neumann, Rietz, & Stenneken, 2017). In the study of McLeod et al. (2012), significant correlations were found between PCC (measured with the Phonology subtest of the Diagnostic Evaluation of Articulation and Phonology; DEAP) and the outcome of the Intelligibility in Context Scale (ICS). Unfortunately, the ICS could not be administered in our study, because the children in study 1 fell out of its age range (too young). Therefore, the intelligibility was scored by the SLPs on a scale with three levels: good, moderate, poor. In study 1 and study 2 subjective judgments of SLPs with ordinal scales were used. Due to this subjectivity no optimal objective measurements were collected, which is a limitation of this study. No reliability measures are reported for these scales. However, it is a common way to judge children's speech and they are used in several other studies (Gordon-Brannan & Hodson, 2000; Lohmander et al., 2016). Further validation studies are needed to corroborate the diagnostic value of the CAI. The present study with 'expert judgment' is the first step in this validation process. Different studies describe those experienced listeners tend to give higher intelligibility ratings than inexperienced listeners (Doyle, Swift, & Haaf, 1989; Landa et al., 2014). In the current study, the ratings were assigned by SLPs who are experienced listeners. As a consequence, the rating 'poor intelligibility' must be considered as indication of a serious speech difficulty. It emphasizes the validity of the strongly related parameter PN-PCC-Q. The results of our study showed a quite stable pattern of nonverbal intelligence and language scores in the children with a speech language impairment across intelligibility levels. Intelligibility level shows no or only a very weak, non-significant correlation with the outcomes on the nonverbal intelligence and language tests; similarly, no or a very weak, non-significant correlation was found between PN-PCC-Q and the outcomes on the nonverbal intelligence and language tests. The results of these correlations show that the PCC of picture naming of the CAI measures a distinct aspect of the language domain. This corresponds to the subtypes described by Van Weerdenburg et al. (2006), in which children with a speech sound disorder are one of the four distinct subtypes.

Study 2 supports the diagnostic power of the CAI-factors in a group of children with SSD. All children, with either a moderate or severe SSD, showed scores below average on the CAI-factors picture naming (PN), nonword imitation segmental (NWI-Seg) and syllable structure (NWI-Syll), proportion of whole-word variability (PWV), MRR-Mono and MRR-BiTri, with mean factor-scores being between -0.77 and -1.81.

Comparison of four CAI-factors (without MRR) revealed significant differences among these factors as well as between the two severity categories. The severity of the speech disorder is mainly expressed in the parameters of picture naming and nonword imitation, as shown by the significant difference between the moderate and severe group for the CAI-factors PN, NWI-Seg and NWI-Syll, whereas PWV is stable across the two groups. These results suggest that especially PN and NWI are the most sensitive tasks to diagnose SSD. This is in line with other authors who stated that nonword imitation, in which the articulatory competence is tested separately from lexical knowledge, is an important part of an assessment battery for children with SSD (Vance, Stackhouse, & Wells, 2005). Other authors have also suggested to not only use picture naming in a speech assessment but include a nonword imitation task to gain better insight in the speech production of a child

(Geronikou & Rees, 2016; Hodges et al., 2017). Nonword imitation is also associated with phonological short-term memory (Gathercole, 2006). Poor performance on NWI can be influenced by difficulties with phonological short-term memory, and not just speech production difficulties. Krishnan et al. (2017) suggest that nonword imitation skills have a unique role in the process of remembering and reproducing novel words. They found that nonword imitation abilities were associated with oromotor praxis, reading fluency, and audiovisual sequence reproduction accuracy. The finding that PWV is relatively stable across severity groups might be related to the multiple origins of inconsistency. As elaborated in the introduction section, inconsistency could indicate unstable lexical representations, an unstable phonological system, or unstable motor planning as is typical for CAS.

When all six CAI-factors were compared (including MRR), significant differences were found among the six factors and the two severity categories. Differences between the moderate and severe group were found for PN, NWI-Seg, NWI-Syll and MRR-BiTri. Remarkably, no difference between moderate and severe groups were found for MRR-Mono, whereas there was a significant difference between the moderate and severe group for MRR-BiTri. The severe group showed the lowest z-score for MRR-BiTri (-2.31) when compared with the other CAI-factors. These results implies that MRR-BiTri is an important factor in diagnosing SSD, like PN and NWI. MRR-BiTri is especially useful in differential diagnosis of SSD with a motor origin (CAS and dysartria), as mentioned in other studies (Rvachew, Hodge, & Ohberg, 2005; Thoonen et al., 1996). The fact that PN, NWI and MRR-BiTri of the CAI were the most affected in the severe speech disorder group underlines the importance of these tasks in diagnosing SSD. No differences between the two severity groups were found for the factors PWV and MRR-Mono. They correlate less with the SLPs' judgments of severity than the other factors. Nevertheless, the mean factor-scores are below average in the SSD-groups as compared to typically developing children with the same age. This indicates that these tasks do contribute to the diagnostic differentiation between typical and atypical development. In studies on speech development, speech variability, as assessed with the tasks word and nonword repetition, has been found to be relatively high in young typically developing children (2- and 3-year-olds; (Sosa, 2015), and such variability decreases with age (Holm, Crosble, & Dodd, 2007). In a previous study (Van Haaften et al., 2019) we also found minor decreases of the PWV with age. Increased variability has also been associated with certain types of speech disorders, such as CAS (Davis et al., 1998; Dodd, 1995b; Forrest, 2003; Holm et al., 2007; Iuzzini-Seigel et al., 2017) and inconsistent phonological disorders (Dodd, 1995b). In the present study, PWV shows a mean below average factor score, and a moderate to strong correlation (.39 to .60) to the PN and NWI factors, although the PWV scores for moderate and severe disorders do not differ. To get a better understanding of these complex relations, a scatterplot of PWV and NWI-Seg factor-scores was made, see Figure 2. Regression lines show a small difference in PWV between moderate and severe disorders; interestingly, for both severity-groups the correlation with NWI-Seg is equally strong. This suggests that PWV can serve as a diagnostic marker for SSD; validation studies with other speech and language diagnoses need to be conducted.

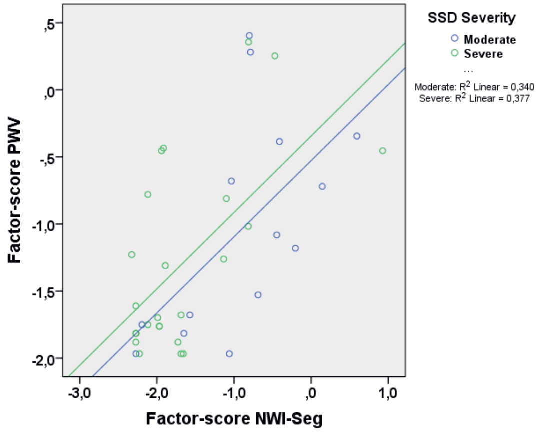


Figure 2. Scatterplot of segmental quality of nonword imitation (NWI-Seg) and word and nonword proportion of whole-word variability (PWV factor-scores), showing the correlations for both groups of children with moderate and severe SSD. Although the difference in PWV between the two groups is small, the correlations with NWI-Seg are moderate to strong.

MRR-performance of mono-syllabic sequences shows no relation with the other task-parameters, suggesting that MRR-Mono assesses an independent aspect of speech production. This is in accordance with such studies as the one by Staiger et al., (2017), who concluded from factor analyses of speech data from patients with neurological movement disorders as compared to control subjects, that speech tasks and oral motor tasks such as rapid syllable repetition measure separate traits. Krishnan et al. (2017) studied the correlation between nonword imitation and other tasks. They also found no correlation between MRR-Mono and nonword imitation, whereas an alternate MRR task (like MRR-BiTri) correlated significantly with nonword imitation. From the perspective of process-oriented approach, Maassen and Terband (2015) argued that MRR, being **a** pure motor task that does not require any knowledge of words, syllables, or phonemes, can be used to assess speech motor skills. Still, like PWV, mean MRR-Mono factor scores are below the population average, thus, like PWV, might serve as a diagnostic marker for SSD. However, in contrast to PWV, MRR-Mono does not correlate with severity. Further studies are needed to delineate the role of the purely repetitive (MRR-Mono) and sequential (MRR-BiTri) variant in SSD.

The present study yields strong indications that comparison of the performance on the different speech tasks of the CAI provides information on the underlying speech processing difficulties of children with SSD. Interestingly, the children with SSD show a distinct factor-structure, that differs from that of the normative study. As mentioned in the introduction, in the normative study on 1,524 typically developing children, weak and very weak correlations between factor-scores were found, from which it can be concluded that the CAI-factors represent independent components of the speech production process. Align with psycholinguistic models, like Levelt's model, the current study describes the speech profile of a group of children with SSD by conducting different speech tasks covering all different speech processes (phonological and speech motor skills).

A limitation of the present study is the use of a heterogeneous group of children with SSD, without analyzing the results of different subgroups. This is an important next step in process-oriented diagnostics. The crucial statistical remark to be made here is that factor analysis is based not on average skills, but on variability in skills and especially co-variance. It can be argued that in a typical population variability in skills is not caused by specific underlying factors, but rather reflects random noise. In contrast, in an atypical population like children with SSD, underlying deficits can cause large covariance if task requirements show overlap; analyzing this structure of overlapping and non-overlapping task performances is the first step in process-oriented diagnostics. Future investigations are needed to compare subgroups of children with different types of SSD, such that more profiles of CAI-factors can be determined to further reveal the proximal causes of SSD. Following the results of the study, the most important implication for clinical practice is to distinguish typical speech development from atypical speech development by the administration of different speech tasks, such as incorporated in the CAI. This allows for process-oriented diagnostics, which is important for targeted intervention in children with SSD.

Acknowledgements

The authors wish to thank all the children who participated, their parents, and all the SLPs for valuable help with the administration and scoring of the tests. This research was partially supported by Het Revalidatiefonds (The fund for Rehabilitation).

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CHAPTER 6

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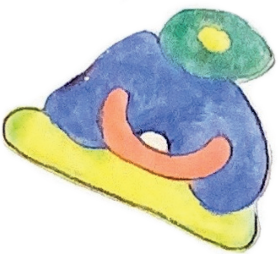

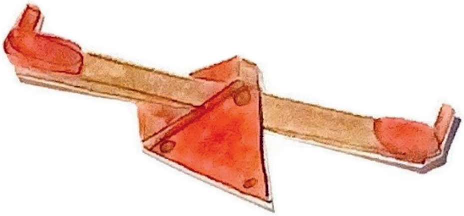

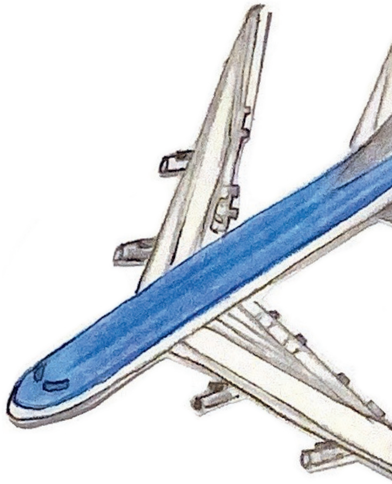

Process-oriented profiling of speech sound disorders

Sanne Diepeveen, Hayo Terband, Leenke van Haaften,
Anne Marie van de Zande, Charlotte Megens-Huigh,
Bert de Swart & Ben Maassen

Published in Children 2022; 9, 1502: 1-23

Abstract

The differentiation between subtypes of speech sound disorder (SSD) and the involvement of possible underlying deficits is part of ongoing research and debate. The present study adopted a data-driven approach and aimed to identify and describe deficits and subgroups within a sample of 150 four to seven-year-old Dutch children with SSD. Data collection comprised a broad test battery including the Computer Articulation Instrument (CAI). Its tasks Picture Naming (PN), NonWord Imitation (NWI), Word and NonWord Repetition (WR; NWR) and Maximum Repetition Rate (MRR) each renders a variety of parameters (e.g., percentage of consonants correct) that together provide a profile of strengths and weaknesses of different processes involved in speech production. Principal Component Analysis on the CAI parameters revealed three speech domains: 1) all PN parameters plus three parameters of NWI; 2) the remaining parameters of NWI plus WR and NWR; 3) MRR. A subsequent cluster analysis revealed three subgroups, which differed significantly on intelligibility, receptive vocabulary, and auditory discrimination but not on age, gender and SLPs diagnosis. The clusters could be typified as three specific profiles: 1) phonological deficit; 2) phonological deficit with motoric deficit; 3) severe phonological and motoric deficit. These results indicate that there are different profiles of SSD, which cover a spectrum of degrees of involvement of different underlying problems.

Introduction

A substantial part of the caseload of speech and language pathologists (SLPs) consists of children with a speech sound disorder (SSD). Prevalence estimates vary, ranging from approximately 3.4% to 24.6% of children in the age of 4 to 8 years being diagnosed with an SSD (Eadie et al., 2015; Shriberg et al., 1999; Wren et al., 2016). Children with SSD are a heterogenous group in terms of symptoms and severity as well as regarding (suspected) underlying deficits (and comorbidities), which makes diagnosing children with SSD a complicated affair (American Speech-Language-Hearing Association, 2021; Bernthal et al., 2017).

Speech development

Speech is the product of a variety of linguistic and speech motor processes working together (Levelt, 2001; Maassen, 2015; Namasivayam et al., 2020; Terband, Maassen, et al., 2019). During speech production, the first process is the conceptualization of a preverbal message from memory or from perception, for example seeing a picture of a cat in a naming task. Next is the formation of an utterance (word or sentence), which is executed by two lexicalization steps: the selection of a lemma, which contains meaning and grammatical word information, and the related lexeme or word form. This lexeme is the input for the next phase, phonological encoding, which consists of generating the sequence of speech sounds and the syllabic and prosodic structures. The selected syllables are the basic elements of the next phase: articulomotor planning and programming. Here, the motor plans and programs for the different speech movements are formed. Motor planning involves the selection and sequencing of articulatory movement goals which are then implemented in muscle specific motor programs (motor programming). Finally, the articulatory movements are executed (motor execution). The neural signals are sent to peripheral systems and transformed into coordinated muscle activity, resulting in an acoustic speech signal (Levelt, 1989; Maassen & Terband, 2015; Stackhouse & Wells, 1997; van Haaften, Diepeveen, van den Engel-Hoek, et al., 2019).

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Children develop adult-like speech both through the development of motor skills and through the expansion of the language system, especially the storage of words with their associated phonemes (lexeme) and the sound system (phonology). Around the age of 24 months, an expressive vocabulary spurt is observed in typically developing (TD) children. During this spurt, a temporary increase in the variability of jaw movements is found, which is believed to be due to the speech motor system rearranging itself to match the rapid cognitive and linguistic development (Green et al., 2002; Nip et al., 2011; Vuolo & Goffman, 2018). Saletta et al. (2018) found that a task with a higher linguistic load was associated with increased speech motor variability in TD children's speech. Thus, linguistic/phonological development influences the speech motor system and vice versa. Both developmental systems can present problems in children with SSD and in intervention an SLP should use different therapy methods for the two systems. An SLP has to investigate both systems in the diagnostic phase. Problems of interpretation arise when an SLP uses only a naming task in the assessment process. Naming the picture of, for example, a cat in a speech assessment does not provide enough information to differentiate a linguistic deficit from a speech motor deficit based on speech errors alone (overt symptoms). If, in the example of the target word 'cat', the /k/ is substituted into [t] this may be interpreted as the phonological process of fronting; the child substitutes a sound produced with the tongue further back in the mouth for one made with the tongue tip just behind the teeth, at the front of the mouth. However, this substitution can also be seen as a simplification of the word 'cat'; the child uses not two different articulatory movement goals, /k/ and /t/, but only one which is easier to produce. The present study set out to investigate the results of a process-oriented speech assessment in a large sample of children with SSD. Using a data-driven approach we investigated if subgroups can be distinguished and how they compare.

Current practice in speech assessments and interpretation

As mentioned above, diagnosing children with SSD is a hard task due to the ambiguity of the diagnostic markers for SSD subtypes and the overlap of speech symptoms between the different diagnostic labels. According to SLPs' reports, a wide variety of different speech assessments are used to diagnose children with SSD and often more than one assessment is used for a single child/per case (Diepeveen et al., 2020; Joffe & Pring, 2008; Malmenholt et al., 2017; McLeod, 2004; Mcleod & Baker, 2014; Priester et al., 2009; Skahan et al., 2007). The obtained assessment data are interpreted based on the SLP's own clinical experience and not on the basis of a clearly formulated set of objectified criteria, as evidenced from data from the Netherlands (Diepeveen et al., 2020) and the United Kingdom (Joffe & Pring, 2008). From questionnaires and interviews of a total of 170 SLPs in the Netherlands, Diepeveen et al. (2020) found that there is no consensus on the terminology and there are many idiosyncrasies in diagnosis and treatment planning of SSDs. A reported 85 different diagnostic labels were used for children with SSD and the speech symptoms associated with these labels showed large overlap. Furthermore, the reports indicated that intervention methods were used for a variety of different diagnostic labels and methods incongruent with their described purpose. The Nuffield Dyspraxia Programme, for example, was also used with children who had been diagnosed with a phonological problem (Diepeveen et al., 2020). Overall, the study concluded that there is no consensus among SLPs in the Netherlands on the terminology and there are many idiosyncrasies in diagnosis and treatment planning of SSDs.

SLPs have different classification systems at their disposal that differentiate subtypes of speech disorders in children (see Waring and Knight (2013) for an overview). Two of the systems that are commonly used are Shriberg's Speech Disorders Classification System (SDCS) (Shriberg et al., 2010) and Dodd's Model of Differential Diagnosis (MDD) (Dodd, 2014; Ttofari Eecen et al., 2019). These two systems have a different approach on classifying SSD. The SDCS is based on the behavioral phenotype of the child's speech and etiological background, whereas the MDD is based on a descriptive-linguistic approach. SDCS and MDD have been subject of prevalence studies, which are shortly summarized below.

The SDCS is an organized framework to distinguish between several subtypes of SSD. It has four levels: etiological processes (distal causes), speech processes (proximal causes), clinical typology (behavioral phenotypes) and diagnostic markers (critical signs of phenotype). At the clinical typology level, three different types are described; each characterized by a specific set of disorders. The three main groups are: speech delay (SD), speech errors (SE) and motor speech disorder (MSD) (Shriberg et al., 2019). In a study of 97 children with SSD, Vick et al. (2014) discovered two groups of children based on five speech tasks and also non-speech tasks. One group (76%) met the criteria of SD and a smaller group (10.3%) met the criteria of motor speech disorder – not otherwise specified (MSD-NOS). Differences between the groups were on atypical speech movements such as a higher variability in measures of articulatory kinematics and a poor performance on iambic lexical stress word imitation in the MSD-NOS group. To further examine the use of the SDCS for the motor speech disorder group and to estimate the prevalence of the types of motor speech disorders, Shriberg et al. (2019) used a sample of 415 children with idiopathic speech delay. A conversational speech sample of each child was used to complete a narrow phonetic transcription, a prosody-voice coding, and an acoustic analysis. These were then entered into the SDCS analysis program and based on the outcomes of the three measures a child was classified in a group. The classification of MSD applied, was Speech Motor Delay, Childhood Dysarthria, Childhood Apraxia of Speech (CAS), and concurrent Childhood Dysarthria and CAS. The following results emerged: 82.2% of the children that met the SDCS criterion for SD at assessment had no MSD; 17.8% with SD met criteria of one of the subgroups of MSD. Of the latter group, 12% was classified has having a Speech Motor Delay; 3.4% met criteria for Childhood Dysarthria and 2.4% children were classified with CAS. None of the children were classified has having the combination Childhood Dysarthria and CAS.

Another model that is often used by SLPs is Dodd's (2014) Model for Differential Diagnosis (MDD). The MDD model contains the following diagnostic labels: 1) articulation disorder: substitutions or distortions of sound (e.g. lateral lisp); 2) phonological delay: speech error patterns typical of younger children; 3) consistent atypical phonological disorder: consistent error patterns of unusual non-developmental errors; 4) inconsistent phonological disorder: inconsistent error pattern of the same lexical item and no oromotor difficulties; and 5) CAS: inconsistency in speech, oromotor signs, slow speech rate, disturbed articulation, short utterance length, poorer performance in imitation. For each of these labels a description is given of the speech problems that can be seen during assessment (Dodd, 2014). Ttofari-Eecen et al. (2019) conducted a validation study for the MDD model and assessed a group of children who speak standard Australian English with the Goldman-Fristoe Test of Articulation 2 (GFTA-2) (Sounds-in-Words and Stimulability sections; Goldman et al., 2000), the Diagnostic Evaluation of Articulation and Phonology (DEAP Inconsistency Assessment; Dodd et al., 2006) and the Verbal Motor Production Assessment for Children (VMPAC; Hayden, 2008). A total of 126 children were eventually divided into the five groups: suspected atypical speech motor control (10%); inconsistent phonological disorder (15%); consistent atypical phonological disorder (20%); phonological delay (55%); and articulation disorder (0%). Ttofari-Eecen et al. (2019) concluded that although the model was designed for the use of children with an articulation or phonological delay or disorder only, the model can be used by SLPs in clinical practice to differentiate children with suspected SSD including children with motor speech disorder such as dysarthria or CAS.

In the MDD and the SDCS, classification is done through the description of the error patterns of the speech output and these errors are compared with typically developing children. Within the SDCS, the extensive use of etiological criteria is also included (Waring & Knight, 2013). The question is whether an SLP can differentiate between the different diagnostic labels based on the error pattern and/or etiology. Both the MDD and the SDCS models leave little room for selecting multiple diagnoses per child, as shown in the two studies described above; all 415 children in Shriberg et al. (2019) and all 126 children in Ttofari-Eecen et al. (2019) received only one diagnosis. Speech errors and/or the etiological background are matched to a specific diagnostic label in both models, and thus these classification systems seem to leave no room for diagnosing the gradual involvement of multiple underlying deficits belonging to one or more different diagnostic labels (Terband, Maassen, et al., 2019).

Diagnostic profiling within the psycholinguistic framework

As mentioned above, some children with SSD present problems in multiple processes, both linguistic and speech motor (Stoeckel & Caspari, 2020). An SLP should therefore assess these multiple processes in a child with SSD to find out which one or more of these underlying processes show deficient functioning. The Psycholinguistic Framework aids SLPs to examine at a cognitive or psycholinguistic level where in the speech and language process the impairment is situated (Baker et al., 2001). This framework is a psycholinguistic speech-processing model and comprises a ‘box and arrow’ model of speech processing skills and representations that serves as a guide for compiling individual profiles of strengths and weaknesses (Stackhouse & Wells, 1997). By comparing speech symptoms under different elicitation conditions within this framework, the proximal causes of SSD can be studied since involvement of underlying processes is different in different speech conditions. In a nonword imitation setting, for example, an alternative speech production route starts from auditory input. Since the child has no lexical representation of the target nonword available, the child must use either the phonological decoding and encoding system (analyze and select combinations of familiar consonants and vowels, possibly syllables) or the auditory-to-motor-planning pathway (repeating the sounds without phonological interpretation, such as in repeating click-sounds).

The problems experienced by children with SSD can be at the level of word-form retrieval, phonological encoding, motor planning and programming, and/or articulation (motor execution). Systematic comparison of speech symptoms under varying conditions allows for assessing a profile of intact and deficient processes. This calls for a shift in the clinical reasoning skills of SLPs from a more diagnostic classification system such as the MDD or the SDCS model (diagnostic categories based on error patterns within a naming task or spontaneous speech) to a process-orientated view (Terband, Maassen, et al., 2019). In other words, an SLP should identify the possible deficiencies of the underlying speech processes (Maassen, 2015; Namasivayam et al., 2020; Terband, Maassen, et al., 2019). Unfortunately, current diagnostic instruments are not designed to provide fine-grained information about the involvement of the different underlying speech production processes (Terband, Maassen, et al., 2019). For example, Geronikou and Rees (2016) conducted a small study to profile four Greek speaking children with SSD based on nonword auditory discrimination, mispronunciation detection, naming, real word repetition and nonword repetition. The children could be profiled as having issues with either phonological or motor representations and the authors concluded that there is a need for a study with a wider range of consonants and clusters in different positions in words in the diagnostic instrument and they also advised to use a larger group of children. Such a study is possible with a new diagnostic instrument developed and released in the Netherlands, the Computer Articulation Instrument (CAI) (Maassen et al., 2019). The basic idea of the CAI is that speech is elicited in different contexts, which each tap into different levels of the production process such that functioning of production processes can be assessed by comparing performances. In addition, the sample of elicited words and nonwords contain all consonants and clusters in different positions, in most cases in at least two different words/nonwords, depending on the frequency of occurrence of the consonants and clusters in the Dutch language. Thus, the instrument yields comprehensive speech profiles from several speech tasks that reflect the functioning of different speech production processes - including phonological skills and speech motor skills; a comparison of those speech profiles gives an indication of possible underlying deficits.

The first aim of the present study was to determine which components emerge in a sample of 150 children with SSD with Principal Component Analysis (PCA) of the speech measures (20 parameters) of the CAI. This analysis was previously conducted for a norm group of 1524 typically developing Dutch-speaking children aged between 2;0 and 7;0 indicated five meaningful components: [1] picture naming (PN); [2] segmental quality of nonword imitation (NWI); [3] quality of syllabic structure of NWI; [4] word and nonword proportion of whole-word variability (PWV), based on word- and nonword repetition (WR and NWR); and [5] mono- and multi-syllabic sequences of maximum repetition rate (MRR) (van Haaften, Diepeveen, van den Engel-Hoek, et al., 2019). PCA is not premised on average skills, but on the variation of skills and particularly on covariance. In a typical population, variation of skills may not be expressed in specific underlying components, due to a ceiling effect. In contrast, in an SSD population, underlying deficits may cause large covariance. If the components are similar, this could mean that children with SSD go through similar developmental milestones as typically developing children, which could be interpreted as an overall speech delay. In contrast, a different component structure could imply a deviant speech profile, which would indicate specific speech deficits. The components can also provide information about the tasks in which specific speech symptoms appear, which helps interpretation regarding the psycholinguistic processes involved.

The second aim was to test whether profiles can be differentiated and identified with the CAI test battery (Maassen et al., 2019) in the same sample of children with SSD. To this end, we conducted k-means cluster analysis, an unsupervised machine learning method to partition data into a k number of groups (clusters) by minimizing variances within clusters, maximizing group similarity. This analysis was exploratory with no preconceived hypotheses about how children would group.

Method

Participants

Participants were recruited in collaboration with the SPEECH study (Van Doornik et al., 2018). 150 children aged 4;0 to 6;6 years (M = 5;2 years) participated in this study. The sample consisted of 94 boys and 56 girls; this ratio between boys and girls is consistent with other international studies (Shriberg et al., 2019; Wren et al., 2016). The children were recruited through private practices (n = 60), special schools for language- and hearing-impaired children (n = 60), a rehabilitation center (n = 16), regular schools (n = 12) and an audiological center (n = 2) in the Netherlands. The children lived in different regions of the Netherlands (North, n = 13; East, n = 44; South, n = 20; West, n = 73). Three children also spoke a language other than Dutch: German, English, and Spanish.
Inclusion criteria were as follows:

- Aged 4 to 6;11 years;
- Dutch as the primary language as indicated by parental report;
- No history of hearing problems based on parents' or caregivers' information (further indicated by care givers) about the child's hearing status;
- A speech sound disorder (SSD) diagnosed by the referring SLP.

At the time of the study 138 children received speech and language therapy. One of these children had scores on the CAI above percentile 16 (see below) and was excluded for this study. Twelve children were recruited through regular schools and had no history of speech or language therapy; they were recruited for the control group of another study (Van Doornik et al., 2018) and were found to have an SSD. These cases were referred to an SLP and were added to the SSD group.

Diagnoses were based on clinical observation and/or a Dutch speech assessment (note that no normalized and standardized assessments were available at the time) and determined by the child's SLP. The majority of the children were diagnosed with a phonological disorder (n = 105), seventeen children with CAS, nine children with a phonetic articulation disorder, five children with dysarthria and the diagnosis of two children was not further specified by the SLP. Eleven children (those recruited through regular schools and not receiving speech therapy at the time of the study) were not previously diagnosed and were referred to an SLP after the diagnostic session; these children did not receive a diagnostic label. Of all children, thirty-two children received more than one diagnosis; sixteen children were diagnosed with a phonological disorder in combination with a phonetic disorder; ten children with CAS and a phonological disorder; two children with dysarthria and a phonological disorder; one child with CAS and a phonetic disorder. Three children received three diagnoses (CAS, phonological disorder, and a phonetic disorder).

Receptive vocabulary of 123 children was determined with the Peabody Picture Vocabulary Test-III-NL (PPVT-III-NL; Schlichting, 2005) (n = 79) or another comprehension test (n = 44) was available in the child's file. Ninety-one children had a quotient score above 85 (range 85-129; 32 children had a score below the 85 (range 66-84). The other children (n = 26) were judged to have a normal comprehension level of the Dutch language, as determined by a professional (teacher, daycare employee and/or SLP), caregivers and the examiner. Comprehension language scores within normal range were not an inclusion criterion, since a comorbidity of a language impairment is common for children with SSD (Eadie et al., 2015).

Data collection

Caregivers were first asked to complete a questionnaire containing questions about their child's speech and language development, and health condition. They also completed the Intelligibility in Context Scale (ICS; McLeod et al., 2013). If the child already received speech therapy, the SLP was also asked to fill out a questionnaire about the child's speech and language abilities. The children were subsequently seen during one or two sessions by 12 student-SLPs or SLPs specifically trained in the administration of the different assessments. The assessment took place at school, private practice, rehabilitation center, or audiological center facilities, in a quiet room.

Materials

During the one or two sessions a receptive vocabulary task, the Peabody Picture Vocabulary Test-III-NL (PPVT-III-NL; Schlichting, 2005); an auditory discrimination test (phonemic judgement) part of the Testinstrumentarium Taalontwikkelingsstoornissen (TTOS-ADT; Verhoeven et al., 2013) and the Computer Articulation Instrument (CAI) were conducted. The framework of the CAI is an integrated model of the cognitive and sensorimotor functions involved in speech production and perception (see Figure 1; Terband, Maassen, et al., 2019).

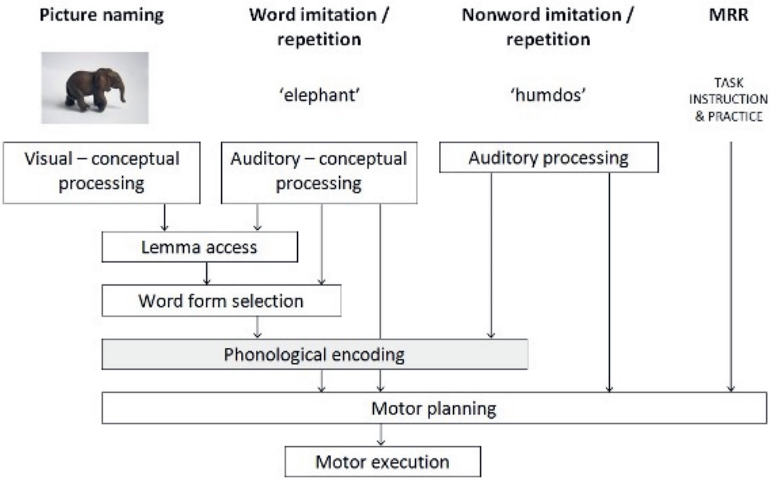

Figure 1. The speech production processes assessed in the four tasks of the Computer Articulation Instrument (Maassen & Terband, 2015; Figure 15.2). MRR = maximum repetition rate. Printed with permission.

The first task of the CAI, picture naming (PN), examines the child's ability to retrieve the stored information about a real word and contains the whole chain of the speech production process, from preverbal visual-conceptual processing to lemma access, word-form retrieval, phonological encoding, motor planning, and articulation (motor execution; see Figure 1; Maassen &

Terband, 2015). In the second task, the child is asked to imitate nonwords (NWI). Due to the nature of the task, the child has no lexical representation of the target utterance available, which means the child must use either the phonological decoding and encoding system or the auditory-to-motor-planning pathway. For the word (WR) and nonword repetition (NWR) tasks, the child is asked to repeat five words or nonwords five times to assess the variability of the speech of the child, which taps into the stages of motor planning and motor programming and stability of the phonological representation of the word form. The final task, maximum repetition rate (MRR), provides a window into the child's motor execution by examining the child's ability to repeat six different sequences as fast as possible (e.g., patakapataka…). For more information on the reliability, validation, and collection of the norms of the CAI, see van Haaften et al. (2019).

Data analysis

A computer or laptop with the CAI, which automatically stored the acoustic signal on the hard disk, was used. The children were seated in front of a microphone and wore open-back headphones to provide a good sound level of the automated instructions. The recordings were transcribed (broad phonetic transcription) and analyzed according to the CAI examiner's manual (Maassen et al., 2019) on the computer by the student-SLPs or SLPs. The student-SLPs worked in pairs and the SLPs worked alone. Following the psychometric evaluation guidelines (van Haaften, Diepeveen, van den Engel-Hoek, et al., 2019), all student SLPs and SLPs were required to practice the transcription and other analyses of the CAI with two practice-examples of children with SSD. After the training session, the results of the transcription and the analysis corresponded between the student-SLPs or SLPs. The transcriptions of the CAI of all children in this study were checked and differences were discussed between the student-SLPs or SLPs. The transcriptions were also checked by the first author (SD) or Anniek van Doornik (collaboration partner in collecting the data). After the transcription and analysis, an automated report was generated of several outcome measures of all CAI-tasks. The outcome measures (percentiles) were based on the data of the norm group (Maassen et al., 2019). Table 1 contains the outcome measures per speech task (parameters) used in the statistical analysis and the number of completed tasks per age group.

Table 1. Parameters/outcome measures per speech task

Task	Parameter		Completed tasks
PN	PCCI	Percentage of consonants correct in syllable-initial position	149
	PVC	Percentage of vowels correct	149
	Level 4	Percentage of correct consonants /b/, /f/ and /ʋ/	149
	Level 5	Percentage of correct consonants /l/ and /R/	149
	RedClus	Percentage of reduction of initial consonant clusters from 2 consonants to 1	149
	CV	Percentage of correct syllable structure CV	149
	CVC	Percentage of correct syllable structure CVC	149
	CCVC	Percentage of correct syllable structure CCVC (C=consonant, V=vowel)	149
	SP	Simplification processes, total score of the processes: fronting, stopping of fricatives, voicing, devoicing and gliding	149
	UP	Unusual processes, total score of the processes: backing, unusual stopping, Hsation, nasalisation and denasalisation	149
NWI	PCCI	Percentage of consonants correct in syllable-initial position	146
	PVC	Percentage of vowels correct	146
	Level 4	Percentage of correct consonants /b/, /f/ and /ʋ/	146
	Level 5	Percentage of correct consonants /l/ and /R/	146
	RedClus	Percentage of reduction of initial consonant clusters from 2 consonants to 1	146
	CV	Percentage of correct syllable structure CV	146
	CVC	Percentage of correct syllable structure CVC	146
	CCVC	Percentage of correct syllable structure CCVC	146
	SP	Simplification processes, total score of the processes: fronting, stopping of fricatives, voicing, devoicing and gliding	146
	UP	Unusual processes, total score of the processes: backing, unusual stopping, Hsation, nasalisation and denasalisation	146
WR	PWV	Proportion of whole-word variability – Word repetition	149
NWR	PWV	Proportion of whole-word variability – Nonword repetition	147
MRR	pa	Number of syllables per second of sequence /pa/	133
	ta	Number of syllables per second of sequence /ta/	133
	ka	Number of syllables per second of sequence /ka/	131
	pata	Number of syllables per second of sequence /pata/	120
	taka	Number of syllables per second of sequence /taka/	115
	pataka	Number of syllables per second of sequence /pataka/	111

Note. PN = Picture Naming; NWI = NonWord Imitation; WR = Word Repetition; NWR = NonWord Repetition; MRR = Maximum Repetition Rate.
Note. Level 4 and 5 are part of the five degrees of complexity of phonological contrasts of Dutch syllable-initial consonants described by Beers (1995)

Statistical Analysis

All statistical analyses were performed using SPSS Version 26. All raw scores were transformed per age group (four/five/six years old) into z-scores to control for speech development to be able to compare the different variables with each other in a single analysis; the z-scores were calculated only with the raw scores of the 150 children with an SSD. To also control for outliers, z-scores lower than -2.33 or higher than 2.33 were replaced by -2.33 or 2.33, respectively; these were the lowest/highest z-scores observed in the CAI norm group. This was the case for eight z-scores in the entire database. Not all children could perform a correct sequence for the MRR task, due to speech–motor difficulties and/or due to shyness or inattentiveness of the child. Additionally, some recordings could not be analyzed due to the low acoustic quality. In cases where children made speech errors, for example replacing a sound with another sound, the missing score was replaced by the lowest z-score (-2.33) of the norm group. This was the case for ten children for the sequence pa; 15 for ta; 19 for ka; 59 for pataka; 29 for pata and 35 for taka.

A principal component analysis (PCA) with varimax rotation (listwise exclusion) was conducted to determine which components are present and to identify clusters of items.
The Kaiser-Meyer-Olkin (KMO) measure was calculated prior to the PCA to determine whether the sample size was adequate; a value larger than 0.5 is deemed acceptable (Field, 2017). The number of principals components (PC) was determined on the criterion for eigenvalues greater than 1. Components were retained if they featured at least three parameters. The CAI-parameters were considered for a PC if they had an absolute factor loading value of more than .4. The parameters with the highest factor loading on a PC were included in that PC (Field, 2017).

Using the same procedure and criteria, a series of additional PCAs was performed subsequently on each of the subsets of variables loading significantly on one PC in the first analysis (see Table 2). There were several reasons to conduct this additional series. Because PCA necessarily applies listwise exclusion, the relatively large number of missing values in the MRR task also limited the number of datapoints for the other components. In the complementary PCAs per subset, all available data for that PC could be included. Factor loadings could thus be verified on all available data and composite performance scores could be obtained for the maximum number of children, including those with missing values on other PCs. These additional PCAs per subset also functioned as a check if the PCs should not be broken down into sub-components on the larger sample. Next, Pearson product-moment correlations were calculated to determine relationships between PCs. A split-half reliability of the PCs (comparing the outcomes when using half of the dataset, randomly selected, with the outcomes using the full dataset) was conducted to check whether the results were stable. If the results of the split-half procedure are similar to the results of the whole group, this confirms the outcomes of the results of the conducted analysis.

Subsequently, we conducted an exploratory k-means cluster analysis with the z-scores of all CAI parameters to test whether distinctive profiles could be identified in our sample of children with SSD. K-means clustering is an unsupervised machine learning method to partition data into a predetermined k number of clusters. In an iterative manner, the observations are divided into groups in a way that minimizes the within-cluster variance and maximizes the variance between clusters. To determine which number of clusters provided the best fit, a comparison was made between analyses with two to four clusters. First, the Iteration History of every number of clusters was compared to determine the best solution. After this procedure the graphs of the clusters were observed to see how the outcomes of the parameters were combined in the different clusters. For example, a two cluster-composition could mean the outcomes of the parameters are clustered in a group with children that score reasonably well and a group with children that score very low. Finally, the number of children in the different clusters were observed to see if there were clusters with a very small number of children in it.

In order to check for possible bias due to age or gender, the distributions of age and gender were compared across clusters. The construct validity was examined by comparing the clusters with respect to parameters of the CAI. The external validity (criterion) was also examined by comparing the clusters with the outcomes of the ICS (objective measure of severity), receptive vocabulary (PPVT-III-NL), auditory discrimination test (T-TOS-ADT), indication of the severity of the speech problem judged by the SLP and care givers (subjective measure of severity), the diagnosis given by the SLP and setting of the child (for example a private practice). This was analyzed with an ANOVA or a Chi-squared test, depending on the level of measurement of the variable; significance was defined as $p < 0.05$ for all tests.

Results

The results of the PCAs are presented first, along with the analysis of correlations between the PCs. Next, we describe the results of the cluster analysis, followed by a comparison between clusters of the PCs identified in the PCA as well as all the non-CAI variables. Note that all children in our sample have atypical speech development, which was verified with the percentage of consonants correct in syllable-initial position (PCCI) scores of the tasks Picture Naming and NonWord Imitation. The scores on these tasks were transformed into z-scores compared to the norm group. Note that these are different z-scores than used in the analysis of this study and were calculated with the average and the standard deviation of the norm group of the CAI (Maassen et al., 2019). All children scored below a z-score of -1.5 on at least one of the two parameters (PN-PCCI z-score M = -4.79, SD = 4.77; NWI-PCCI z-score M = -2.91, SD = 2.68), and no z-score higher than 1 occurred thus confirming the diagnosis of SSD for all children.

Principal Component Analysis

A PCA with orthogonal rotation (varimax) was conducted on all speech parameters of the CAI. The KMO measure confirmed adequacy of the sample for the analysis (KMO = .870). The analysis yielded a solution in which three components had an eigenvalue higher than 1, (12.7, 2.64 and 1.94 respectively). This three-component solution explained 61.7% of the variance. All principal components had a Cronbach's alpha's higher than .74, which indicates the internal consistency of the components were acceptable. The results of the PCA are presented in Table 2. Parameters loading high on the first PC were all the parameters of the PN task plus the following parameters of the NWI task: Level5, Simplification processes and the Unusual processes (PN+) (an explanation of the parameters can be found in Table 1). The second PC included WR, NWR and almost all the parameters of the NWI task except for Level5, Simplification processes and the Unusual processes (NWI/PWV). The last PC contained all the parameters of the MRR. It should be noted that the parameters NWI-PCCI, NWI-level4, NWI-SP and NWI-UP also had high loadings (above .4) on one of the other two components; these parameters were included with the PC on which the highest loading was calculated. The grouping was confirmed by repeating the analysis with half of the SSD group.

Table 2. Principal Component Analysis results for Picture Naming (PN), NonWord Imitation (NWI), Word (WR) and NonWord (NWR) Repetition, and Maximum Repetition Rate (MRR). The highest component loading of each parameter is displayed in boldface.

Task	Parameter	Component		
		1	2	3
PN	PCCI	**0.896**	0.262	0.224
	PVC	**0.655**	0.430	0.177
	RedClus	**0.817**	0.089	0.163
	Level 4	**0.728**	0.170	0.180
	Level 5	**0.631**	0.237	-0.053
	CV	**0.432**	0.249	0.400
	CVC	**0.563**	0.364	0.080
	CCVC	**0.797**	0.262	0.176
	SP	**0.801**	0.197	0.185
	UP	**0.768**	0.199	0.150
NWI	PCCI	0.601	**0.730**	0.153
	PVC	0.367	**0.823**	0.155
	RedClus	0.561	**0.648**	0.072
	Level 4	0.508	**0.680**	0.053
	Level 5	**0.469**	0.319	0.124
	CV	0.104	**0.731**	0.283
	CVC	0.349	**0.715**	0.257
	CCVC	0.477	**0.481**	0.050
	SP	**0.632**	0.532	0.133
	UP	**0.637**	0.542	0.002
WR	PWV	0.085	**0.730**	0.111
NWR	PWV	0.284	**0.566**	0.175
MRR	pa	-0.202	0.271	**0.726**
	ta	-0.004	0.240	**0.786**

	ka	0.202	0.094	**0.667**
	pata	0.230	0.130	**0.708**
	taka	0.198	-0.148	**0.720**
	pataka	0.255	0.226	**0.445**
Eigenvalues		12.70	2.64	1.94
% of variance		45.37	9.42	6.93
Cronbach's α		.945	.909	.796

Note. PN = Picture Naming; NWI = NonWord Imitation; WR = Word Repetition; NWR = NonWord Repetition; MRR = Maximum Repetition Rate; PCCI = Percentage of consonants correct in syllable-initial position; PVC = Percentage of vowels correct; Level 4 = percentage of correct consonants /b/, /f/ and /ʋ/; Level 5 = percentage of correct consonants /l/ and /R/; RedClus = percentage of reduction of initial consonant clusters from 2 consonants to 1; CV = percentage of correct syllable structure CV; CVC = percentage of correct syllable structure CVC; CCVC = percentage of correct syllable structure CCVC; SP = Simplification processes, total score of the processes: fronting, stopping, voicing, devoicing and gliding; UP = Unusual processes, total score of the processes: backing, atypical stopping, Hsation, nasalisation and denasalisation ; WR-PWV = Proportion of whole-word variability – Word Repetition; NWR-PWV = Proportion of whole-word variability – NonWord Repetition; MRR-pa = number of syllables per second of sequence /pa/; MRR-ta = number of syllables per second of sequence /ta/; MRR-ka = number of syllables per second of sequence /ka/; MRR-pataka = number of syllables per second of sequence /pataka/; MRR-pata = number of syllables per second of sequence /pata/; MRR-taka = number of syllables per second of sequence /taka/.

A complementary series of PCAs was performed to obtain composite performance scores for all children, including those with missing values on other components, and to verify factor loadings and check if the PCs should not be broken down into sub-components on the larger sample. All three PCAs yielded a one-component solution. Within this additional PCA, the first component (PN+), comprising the PN parameters and the phonological processes of the NWI task explained 63.2% of the variance (KMO = .884); the second PC (NWI/PWV) comprising the remaining NWI parameters and the two repetition tasks (WR and NWR) explained 61.8% of the variance (KMO = .889), and the third PC (MRR) containing all MRR parameters explained 50.2% of the variance (KMO = .788).

Pearson product-moment correlations between the components of the second PCA were calculated. Moderate and significant correlations were found between PN+ (PC 1) and MRR (PC 3), and between NWI/PWV (PC 2) and MRR (PC 3). The correlation between PN+ and NWI/PWV was high. The results are shown in Table 3.

Table 3. Pearson correlations between Principal Components, *n* = 100

Components	PN+	NWI/PWV	MRR
PN+	-	.793**	.420**
NWI-/PWV	.793**	-	.375**
MRR	.375**	.420**	-

Note. PN = Picture Naming; NWI = NonWord Imitation; PWV = proportion of whole-word variability, Word and NonWord Repetition; MRR = Maximum Repetition Rate.

**Correlation of Principal Components is significant at the .01 level (two-tailed).

Cluster analysis

A k-means cluster analysis was conducted with the same CAI parameters as used in the PCA (see Table 2). Forty-nine children out of a total 149 children were not included due to listwise exclusion (exclusion because of missing data); some children did not complete all the tasks due to failure or refusal. To check which number of clusters would fit best, the remaining 100 children were each allocated to one of either two, three or four clusters. The three-cluster analysis yielded the clearest results, which are shown in Table 4 and Figure 2. The two-cluster analysis yielded one group of children who performed poorly on all parameters and one group who performed slightly better on all parameters. The four-cluster analysis yielded one group that scored significantly worse and one group that performed significantly better, each compared to the other three clusters. However, no clear interpretation could be made of the profiles of the other two, intermediate clusters. Therefore, the analysis of three clusters was chosen to be described here. To validate this choice the same procedure was applied on a random selection of half of the 100 cases. The clusters yielded approximately the same mean scores for the three clusters as for the one based on all 100 children for the k-means cluster analysis, and the same components emerged for the PCA.

The three clusters that emerged differed significantly from each other with respect to the parameters PN-level 5, PN-CCVC, NWI-Level 4, NWI-Level 5, and all MRR parameters with all differences showing large effect sizes ($\eta^2 > .14$). The children in cluster I outperformed children in cluster II and III, and children in cluster II scored better than children in cluster III. However, most of the CAI-parameters were not normally distributed, therefore, if a difference between the three groups was found to be significant at the 5% level, the comparison was reanalyzed using the Bonferroni corrected listwise comparisons for the non-normally distributed parameters. When this was applied, clusters I and II were not significantly different from each other on these parameters (Picture naming: PCCI, PVC, Level 4, RedClus, CV, CVC, SP, UP; NonWord Imitation: PCCI, PVC, RedClus, CV, CVC, SP, UP and the Word/NonWord Repetition), whereas cluster III was significantly different from clusters I and II. Children in cluster III scored lower than children in cluster I and II on all parameters. In Figure 2 the performance of the children on the tasks for the three clusters is shown.

Table 4. Measures age, gender, parameters of the CAI in three subgroups of children with SSD identified by cluster analysis (*n* = 100).

Variable		Norm group	Cluster			ANOVA		
		(*n* = 121)	I (*n* = 46)	II (*n* = 28)	III (*n* = 26)	F	p	η²
Age (age in months (SD))		61.5 (1.10)	62.1 (8.40)	60.2 (9.09)	61.3 (8.69)	.404	.669	.008
% boys		66 (54.5%)	28 (49.1%)	18 (31.6%)	11 (19.3%)	3.177	.204	.178
PN	PCCI	96.8 (3.7)	90.6 (6.99)	85.0 (11.25)	56.2 (12.31)	106.197	<.001$^{*I=II,\ I/II>III}$	.686
	PVC	97.7 (3.1)	97.7 (2.97)	97.0 (2.59)	87.0 (7.81)	48.267	<.001$^{*I=II,\ I/II>III}$	.499
	Level 4	~	90.5 (13.53)	82.1 (20.89)	46.6 (28.06)	40.478	<.001$^{*I=II,\ I/II>III}$	.455
	Level 5	93.6 (11.0)	73.1 (20.87)	69.8 (20.96)	34.7 (24.69)	27.78	<.001*	.364
	RedClus	97.0 (6.2)	89.5 (16.05)	89.0 (14.63)	69.3 (23.25)	12.080	<.001$^{*I=II,\ I/II>III}$	.199
	CV	~	94.9 (6.09)	91.5 (10.12)	78.5 (17.22)	18.864	<.001$^{*I=II,\ I/II>III}$	.280
	CVC	~	93.9 (5.12)	92.3 (6.89)	82.6 (10.30)	21.553	<.001$^{*I=II,\ I/II>III}$	.308
	CCVC	94.4 (9.9)	82.1 (20.14)	70.7 (25.20)	30.4 (22.81)	45.436	<.001*	.484
	SP	2.8 (5.2)	13.4 (13.26)	27.9 (27.44)	82.8 (38.47)	61.197	<.001$^{*I=II,\ I/II>III}$	.558
	UP	0.2 (0.5)	2.8 (4.16)	5.5 (7.28)	19.6 (11.68)	42.634	<.001$^{*I=II,\ I/II>III}$	.468
NWI	PCCI	87.7 (6.9)	78.3 (13.30)	71.9 (15.61)	39.4 (9.99)	74.758	<.001$^{*I=II,\ I/II>III}$	.607
	PVC	93.5 (4.8)	91.9 (8.41)	88.6 (11.29)	70.5 (14.96)	31.844	<.001$^{*I=II,\ I/II>III}$	.396
	Level 4	87.8 (11.8)	76.3 (18.63)	72.5 (20.23)	30.3 (18.26)	53.376	<.001*	.524
	Level 5	87.2 (12.8)	72.0 (20.70)	68.8 (25.10)	31.1 (17.74)	33.562	<.001*	.409
	RedClus	92.3 (11.8)	86.6 (14.94)	85.2 (18.81)	68.4 (23.37)	8.843	<.001$^{*I=II,\ I/II>III}$	.154
	CV	96.8 (7.8)	94.8 (9.04)	92.9 (13.15)	74.4 (21.14)	18.770	<.001$^{*I=II,\ I/II>III}$	.279
	CVC	93.3 (5.7)	90.96 (8.18)	87.32 (9.15)	71.5 (18.12)	23.551	<.001$^{*I=II,\ I/II>III}$	.327
	CCVC	83.0 (25.2)	75.1 (30.40)	67.9 (36.88)	32.1 (35.98)	14.043	<.001*	.225
	SP	7.2 (7.4)	27.5 (23.86)	39.8 (30.29)	105.2 (43.27)	52.502	<.001$^{*I=II,\ I/II>III}$	.520

	UP	2.1 (2.2)	9.7 (7.20)	11.9 (8.65)	34.1 (14.24)	55.043	<.001$^{*I=II, I/II>III}$	.532
WR	PWV	.23 (.04)	.30 (.07)	.32 (.11)	.47 (.16)	21.483	<.001$^{*I=II, I/II>III}$	.307
NWR	PWV	.28 (.08)	.35 (.12)	.40 (.15)	.51 (.21)	9.242	<.001$^{*I=II, I/II>III}$	.160
MRR	pa	4.64 (0.61)	4.44 (0.51)	3.82 (0.58)	3.97 (0.78)	7.092	.001*	.143
	ta	4.44 (0.60)	4.45 (0.44)	3.65 (0.68)	3.53 (0.83)	17.092	<.001*	.284
	ka	4.34 (0.51)	4.01 (0.59)	3.14 (0.80)	2.91 (0.91)	11.255	<.001*	.220
	pata	4.49 (0.73)	4.59 (0.92)	2.81 (0.88)	2.78 (1.01)	23.710	<.001*	.404
	taka	4.37 (0.75)	4.32 (0.79)	2.64 (0.56)	3.15 (0.99)	23.332	<.001*	.418
	pataka	4.09 (0.82)	3.47 (1.18)	2.33 (0.57)	2.81 (1.07)	12.745	<.001*	.372

Note. PN = Picture naming; NWI = Nonword imitation; WR = Word repetition; NWR = Nonword repetition; MRR = Maximum repetition rate; PCCI = Percentage of consonants correct in syllable-initial position; PVC = Percentage of vowels correct; Level 4 = percentage of correct consonants /b/, /f/ and /ʋ/; Level 5 = percentage of correct consonants /l/ and /R/; RedClus = percentage of reduction of initial consonant clusters from 2 consonants to 1; CV = percentage of correct syllable structure CV; CVC = percentage of correct syllable structure CVC; CCVC = percentage of correct syllable structure CCVC; SP = Simplification processes, total score of the processes: fronting, stopping, voicing, devoicing and gliding; UP = Unusual processes, total score of the processes: backing, atypical stopping, Hsation, nasalisation and denasalisation ; WR-PWV = Proportion of whole-word variability – Word repetition; NWR-PWV = Proportion of whole-word variability – Nonword repetition; MRR-pa = number of syllables per second of sequence /pa/; MRR-ta = number of syllables per second of sequence /ta/; MRR-ka = number of syllables per second of sequence /ka/; MRR-pataka = number of syllables per second of sequence /pataka/; MRR-pata = number of syllables per second of sequence /pata/; MRR-taka = number of syllables per second of sequence /taka/, ~ no score because of a ceiling effect in the norm group. Note. Redclus for the norm group is inverted.

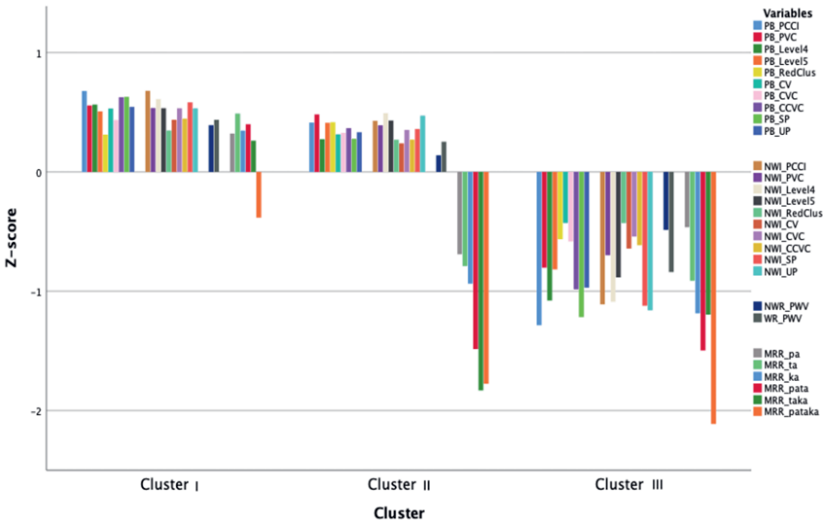

Figure 2. Overview of the distribution of the parameters across the clusters in z-scores.

Cluster comparison with non-CAI variables

Two Chi-squared tests indicated that age and gender did not differ between the three clusters (see Table 4). A series of ANOVAs with post-hoc pairwise comparisons indicated that the clusters did differ in the performance of the children on some of the additional assessments. With respect to the receptive vocabulary assessment (PPVT-III-NL), the auditory discrimination task (TTOS-ADT) and the speech intelligibility (ICS), the children in cluster I outperformed the children in clusters II and III while the cluster II children in turn also outperformed the children in cluster III (I > II > III; see Table 5).

The SLPs and caregivers were asked to rate the child's speech problem in the questionnaire. SLPs and caregivers were asked 'How would you estimate the severity of the speech problem?' and they could answer with Mild, Moderate or Severe. To see if the SLP's judgement correlated with the distribution in the clusters, a comparison (Chi-squared test) was made between the clusters with respect to the judgement of the severity of the SSD. There was a significant difference between the three clusters on the three severity levels; these differences showed a moderate effect size (V between .3 and .5) (see Table 5). Most of the children in cluster III were judged to have a severe speech problem, 19 (59.4%) children were considered to have a severe speech problem judged by SLPs and 13 (52.0%) children judged by their caregivers. The label moderate was mostly given to the children in cluster I and II. The label mild was given by the SLPs to 12 (80.0%) and by their caregivers to 16 (76.2%) children in cluster I; three children in cluster I were labeled by their caregivers as having no speech problem.

For 88 children, the diagnosis that they had received from their SLP was known (the diagnosis by the SLP based on the SLP's assessment of the child); for 12 children, the SLP's diagnosis was not known. The label phonological disorder was most often given by the SLPs followed by the diagnosis of CAS; phonetic disorder and dysarthria were the least frequent diagnoses. The result of the Chi-squared test showed no interaction between the diagnostic labels and the clusters.
The analysis also included the number of children attending a particular setting. The clusters differed significantly regarding setting. In clusters I and II, the largest category consisted of children who received speech therapy in a private practice. In cluster III, however, the largest category was special education for children with speech and language disorders. The settings audiologic center and rehabilitation center were divided roughly equally across the three clusters. The children who were initially recruited for the control group, but who turned out to have a speech problem, were mainly placed in Cluster I.

Table 5. Measures PPVT-III-NL, TTOS-ADT, ICS, intelligibility level SLP and parents, diagnosis and setting in three subgroups of children with SSD identified by cluster analysis (*n* = 100).

Variable		**Cluster** I (*n* = 46)	II (*n* = 28)	III (*n* = 26)	**ANOVA for continuous and $\chi^2$for categorical variables**		**η^2 for continuous and V for categorical variables**
PPVT-III-NL		102.8 (14.00)[$]	101.5 (10.94)[$$]	90.8 (11.86)[$$$]	5.201	.008[*]	.152
T-TOS (ADT)		63.4 (27.67)[+]	52.9 (34.58)[++]	34.4 (25.17)[+++]	5.959	.004[*]	.153
ICS		4.0 (.40)[^]	3.8 (.44)[^^]	3.5 (.51)[^^^]	9.801	<.001[*]	.201
Intelligibility affected (SLPs) (*n* = 73)					28.027	<.001[*]	.438
	mild	12 (80.0%)	3 (20,0%)	0 (0.0%)			
	moderate	11 (42.3%)	11 (42.3%)	4 (15.4%)			
	severe	5 (15.6%)	8 (25.0%)	19 (59.4%)			
Intelligibility affected (parents) (*n* = 78)					22.478	.001[*]	.380
	no speech problem	3 (100.0%)	0 (0.0%)	0 (0.0%)			
	mild	16 (76.2%)	3 (14.3%)	2 (9.5%)			
	moderate	10 (34.5%)	11 (37.9%)	8 (27.6%)			
	severe	5 (20.0%)	7 (28.0%)	13 (52.0%)			
Diagnosis (*n* = 88)					7.266	.297	.058
	Phonetic disorder	5 (62.5%)	2 (25.0%)	1 (12.5%)			

	Phonological disorder	27 (42.2%)	17 (26.6%)	20 (31.3%)			
	Childhood Apraxia of Speech	3 (25.0%)	6 (50.0%)	3 (25.0%)			
	Dysarthria	0 (0.0%)	2 (50.0%)	2 (50.0%)			
Setting					32.744	<.001*	.405
	Private practice	22 (53.7%)	15 (36.6%)	4 (9.8%)			
	Special education	9 (26.5%)	6 (17.6%)	19 (55.9%)			
	Rehabilitation centre	4 (33.3%)	5 (41.7%)	3 (25.0%)			
	Audiologic centre	1 (50.0%)	1 (50.0%)	0 (0.0%)			
	Recruited as control group	10 (90.9%)	1 (9.1%)	0 (0.0%)			

Note. PPVT-III-NL = quotient of word comprehension; TTOS-ADT = percentile of auditory discrimination test. ICS = average score on the Intelligibility of Context Scale.
Note. Data missing in this group: $^{\$}$ 16 children, $^{\$\$}$ 15 children, $^{\$\$\$}$ 8 children; $^{+}$ 13 children; $^{++}$ 11 children, $^{+++}$ 7 children; ^ 13 children, ^^ 5 children, ^^^ 1 child

Comparison of clusters and factors

To see if the clusters differed from each other on the scores on the three principal components (PC) identified in the PCA, a single multivariate ANOVA was conducted. The clusters differed significantly on each PC: PN+ (PC 1; $F = 144.15$, $p = .000$, $\eta^2 = .748$); NWI/PWV (PC 2; $F = 57.15$, $p = .000$, $\eta^2 = .541$) and MRR (PC 3; $F = 88.66$, $p = .000$, $\eta^2 = .646$). Table 6 presents the mean PC scores for the three clusters. Children in the largest cluster (I, n = 46) scored best on all components. Children in cluster II (n = 28) showed a different pattern: they scored similar to children in cluster I on the PN+ PC and on the NWI/PWV, but they scored weak on the MRR. The children in cluster III (n = 26) scored very low on all the PCs.

Table 6. Mean components and standard deviation of the three clusters per factor

	Components					
	PN+		NWI-/PWV		MRR	
Cluster	*M*	*SD*	*M*	*SD*	*M*	*SD*
I	.72	.46	.64	.61	.87	.58
II	.49	.50	.41	.69	-.76	.46
III	-1.25	.52	-.96	.58	-.72	.77

Discussion

The overall aim of this study was to determine the possibility of profiling children with SSD based on underlying deficits. For this, the CAI was administered, and a two-step analysis procedure was conducted, comprising a Principal Component Analysis (PCA) to find components, followed by a cluster analysis (k-means clustering) to find distinct profiles.

Which components emerged and how do these relate to norm group outcomes?

The PCA yielded three stable and meaningful components. The first component (labeled PN+) consisted of all picture naming (PN) parameters plus three parameters of the nonword imitation (NWI) task: NWI-level5 and the phonological processes (see Table 1 for explanation of the parameters). The second component (labeled NWI/PWV) consisted of the remaining NWI parameters and the two-proportion whole-word variability (PWV) parameters, based on word repetition (WR) and nonword repetition (NWR). The third component (labeled MRR) contained all maximum repetition rate (MRR) parameters. The results of the PCA in the current group of children with SSD differed from the results of the PCA in the CAI norm group, which consisted of Dutch children with typical development (n = 1.524) aged two to seven years (van Haaften, Diepeveen, van den Engel-Hoek, et al., 2019). In the norm group, five components were discovered: PN; segmental quality of NWI; quality of syllabic structure of NWI; word and nonword proportion of whole-word variability (PWV) and MRR. Note that the phonological processes were not included in the norm group, probably because their frequency of occurrence was too low among the 4–7-year-olds. The component MRR emerged as one component in both samples.

6

The five components from the norm group (van Haaften, Diepeveen, van den Engel-Hoek, et al., 2019) were used in a previous study to compare the scores of 41 children with SSD (van Haaften, Diepeveen, Terband, et al., 2019). That is, the components' weights obtained from the PCA of the norm group were used to calculate component scores for the children with SSD. In this study, the child's SLP had scored the severity of the speech disorder as moderate or severe (mild did not occur). Children in the moderate group obtained better scores than children in the severe group on parameters of the Picture Naming and NonWord Imitation tasks, whereas word and nonword repetition consistency were equal for these two groups. Furthermore, the moderate and severe groups differed with respect to the MRR-bi-and trisyllabic parameters, but not with respect to the MRR-monosyllabic sequences. Thus, this study provided evidence that comparison of performance on the different speech tasks of the CAI can provide distinct profiles which are different from the norm group and related to severity of SSD.

In the present study, not only the number of components in the clinical sample was smaller than that of the norm group, but also the composition of the first two components was different. In the norm group, all the parameters of the PN task loaded onto one component, and segmental quality of NWI and quality of syllabic structure of NWI on two separate components. In contrast, in the SSD group the specific phonological parameters of the NWI task, namely the NWI-level5, and the two phonological processes, loaded onto the PN component rather than on the NWI. Thus, the first component in the SSD group comprised both segmental and syllabic aspects of picture naming as well as specific phonological aspects of nonword imitation. Therefore, phonological encoding is a stronger component in the SSD group. The second component in the SSD group contained the remaining parameters of NWI, reflecting overall segmental quality (PCC, PVC) and quality of syllable structure (CV, CVC, CCVC), plus the percentage whole-word variability (WR-PWV and NWR-PWV). Interpretation? Related to the chain: auditory – memory – encoding/assembly. The difference between the norm group and the children with SSD regarding the parameters WR-PWV and NWR-PWV could be due to the fact that the typical children are consistent in this task already at an early developmental stage, resulting in a ceiling effect, whereas large differences were found between the SSD subgroups. Overall, the two components in the children with SSD as compared to four components in the norm group, seem to indicate a much clearer dissociation in the SSD group as compared to the norm group between phonological processes of speech production (word form retrieval and phonological encoding) and the processes that follow (motor planning, programming, and the stability of those processes). For naming pictures, children use the whole chain of the speech production process, and thereby rely on their vocabulary and –for the speech production process– specifically on the stored word forms (lexemes). In contrast, for repeating nonwords speakers use either the phonological decoding and encoding systems, or the auditory-to-motor-planning pathway (or both). The statistical result that PN and NWI-parameters load largely on different components, indicates that this distinction in underlying processing has significant impact on the quality of production. This implies that it is important to assess both tasks to get a broad view on the whole speech production process and on parts of the chain. Children who make relatively few errors in speech production when imitating nonwords may have relatively little difficulty in pronouncing new words they are learning, which could be a starting point for a method of intervention.

Which clusters emerged?

After the PCA analysis, a cluster analysis (k-means clustering) was conducted to see if subgroups would emerge from the data. Three clusters were found. The children in cluster I (n = 46) outperformed the children in the other two clusters on all parameters, while the children in cluster

III (n = 26) scored lowest on all parameters. However, compared to the norm group, the children in cluster I scored lower on all parameters of PN and NWI. Although the cluster I group shows little or no vowel replacement in their speech as well as few errors in the simple syllable structures (CV and CVC), these children do make cluster reduction errors and phonological processes do still occur in more complex syllables. Therefore, this cluster can be labelled as phonological deficit. The children in cluster II (n = 28) showed a different pattern: they scored similar to the cluster I children on the PN+ and NWI/PWV principal components, but they scored weak on the MRR. As such, this cluster could be labeled as a phonological deficit with motoric deficit. The children in cluster III (n = 26) scored very low on all components, and this cluster could thus best be labeled as severe phonological and motoric deficit.

How do the different clusters compare to each other and to norm data?

McLeod (2020) concluded in her review that 11 studies found a weak to moderately significant correlation between ICS and PCC. In our study this correlation calculation was not part of the research question, but we found a severity trend as well. As discussed above for each task of the CAI a difference can be observed between the clusters. This can be further supported by data on the intelligibility of the children as assessed by the caregivers and the SLPs. The intelligibility on the ICS is significantly different between the three clusters; the intelligibility of children in cluster I is better than that of cluster II and the children in cluster III show the lowest intelligibility. This was also confirmed by the responses of the speech therapists to the question of how severe they thought the speech problem was. Here too, the clusters differed significantly from each other; the severity of the SSD is rated as least severe for the children in cluster I and more severe for the children in cluster II, and the children in 670 cluster III are the most severe cases according to the SLPs (severity: III>II>I).

With respect to error patterns, a first difference between the three clusters that can be observed is in vowel production. PN-PVC and NWI-PVC in cluster I and II showed a fairly high score and do not differ much from the children with a typical speech development. In typical development, five-year-old children achieve a mean PVC of 97.0 (SD = 3.9) in naming pictures and 90.5 (SD = 7.5) in repeating nonwords (see Table 3) (Maassen et al., 2019). The cluster I and II children in the present study showed similar averages and did not differ significantly from each other. However, the children in cluster III obtained significantly lower PVC-scores compared to the norm data. Roepke and Brosseau-Lapré (2021) also observed differences in vowel production for 39 typically developing children compared to 45 children with SSD. They concluded that no conclusion could be drawn from their study as to whether these speech errors are systematic and reflect speech severity because the children were not matched on language ability but on age; another pattern might have been obtained if children were matched on language ability. However, a clear pattern was visible in our study: the children with the most severe speech disorder (cluster III; severe phonological and motoric deficit) showed lower PVCs than the other two less severe speech disorder groups.

Regarding consonant production, the results showed a similar profile among the clusters in the SSD group, cluster I and II children had similar averages of PCCI on both PN and NWI while the children in cluster III scored lower on both tasks. In the case of PCCI, however, all children with SSD scored lower compared to the norm group data (percentage for the five-year-old: PCCI-PN = 95.2, SD = 5.2; PCCI-NWI = 82.5, SD = 10.1). These findings indicate once more that measures such as the percentage consonants correct can serve as a severity index (Dale et al., 2020; Shriberg et al., 1997). Consistency of errors was also measured in the present study, by means of the proportion whole-word variability when repeating five words and five nonwords five times (PWV-WR and PWV-NWR respectively). The children in cluster I and II scored the same and the children in cluster III were significantly less consistent in repeating the five words and nonwords. Compared to children in the norm group the mean inconsistency scores of the two tasks were slightly higher for the children in cluster I and II, and the children in cluster III showed the largest variability.

The last task of the CAI is the Maximum Repetition Rate (MRR). The results showed that children in cluster I outperformed children in cluster II and III on all MRR parameters and that the cluster II children outperformed the children in cluster III, all with a large effect size. In comparison to the norm group (mean of the five years old ranges from 3.74 syll/sec to 4.29 syll/sec for the different sequences in the norm group (Maassen et al., 2019; Diepeveen et al., 2021), the children in cluster I scored similar on all MRR parameters. The children in cluster II produced the monosyllabic sequences slightly slower than the children of the norm group, and the bi- and trisyllabic sequences were produced at least one syllable per second slower than the norm group. The cluster III children produced the /pa/ sequences somewhat slower than the norm group as well and produced all other sequences with at least one syllable per second slower (Maassen et al., 2019; Diepeveen et al., 2021). Children in cluster II and III were slightly better on the mono syllabic sequences compared to the bi-tri syllabic sequences. This difference may be a predictor of motor planning and programming problems. Ozanne (2005) performed a cluster analysis of 18 behaviors that could reveal an underlying speech motor planning and programming problem on a dataset in a study of 100 children (ages 3;0-5;6) with SSD of unknown origin. The most common problems of the children were incorrect DDK sequences (38%), slow DDK rate (35%) and an increase in errors with increased linguistic load (27%), which corroborates our findings.

In the past, several debates have taken place about the potential value of nonspeech oral motor tasks such as the MRR (Ziegler et al., 2019). Criticism has mainly come from the field of adult acquired disorders, but most studies with children conclude that MRR should be part of the assessment of SSD (Chenausky et al., 2020; Diepeveen et al., 2019; Murray et al., 2015). The current study confirms that MRR performance has a distinctive contribution to the diagnosis of SSD. The distinction between Cluster I and II is primarily based on MRR, and the distinction between Cluster I and III on both MRR and the phonological components. Across clusters, the correlations between the phonological components are high, and the correlations between these clusters and MRR are moderate. This shows that MRR contributes to diagnostic classification as an indicator of speech motor involvement (Cluster 1 versus Cluster 2) and can be considered an indicator of severity (Clusters 2 and 3).

In summary, three conclusions can be drawn from the analysis of the clusters: 1) there are different profiles of SSD; 2) in which severity plays a role and 3) that cover a spectrum of degrees of involvement of different underlying problems.

In this study, the group of children with missing values in the MRR, because children could not or refused to perform a sequence, was not included in the cluster analysis. Why the children refused is not known; children were not asked to give an explanation. They might have refused out of boredom of the session. In addition, not all typically developing children in the MRR norm group performed a sequence either (Diepeveen et al., 2021). In the future, qualitative analysis (e.g., 0=no MRR; 1=could not perform a long sequence; 2=could not perform a sequence correctly due to a speech error, etc.) could be used to assess the number of children who performed a sequence.

How do these relate to diagnostic classification systems?

We cannot make a direct, quantitative comparison, between our results and the results of the previously mentioned two studies in the introduction classifying children with the SCDC (Shriberg et al., 2019) and with Dodd's model (Ttofari Eecen et al., 2019), due to the large differences in tasks and data analysis method. This study applied a data-driven cluster analysis, while the other two studies aimed to classify the children according to pre-determined profiles that (are assumed to) correspond to certain subtypes of speech disorders. Furthermore, in our data, severity of the speech disorder also plays a role in clustering the outcomes of the CAI, while speech severity is not included in the validity studies of SCDC and in Dodd's model.

Dodd's Model for Differential Diagnosis The children in the consistent atypical phonological disorder group and the children in the phonological delay group in the study of Ttofari-Eecen et al. (2019) had at least one (a)typical phonological error pattern and had no difficulty repeating the 25 words of the DEAP Inconsistency Assessment (Dodd et al., 2006) multiple times. This group could be compared to the children in cluster I, who also had at least one typical and/or atypical phonological error pattern. However, children in cluster I had a higher mean score on the Word and NonWord repetition tasks of the CAI compared to the norm group of the CAI; children in cluster I scored les consistent. Therefore, they might not be similar to the consistent atypical phonological disorder group and the phonological delay group of the MDD model; these children do not have a lower score on an inconsistency assessment compared to a norm group. The children in cluster II performed similar to the children in the inconsistent phonological disorder group of the Ttofari-Eecen-study; they had a typical and/or atypical phonological error pattern and were inconsistent in their speech. The children in cluster III can be compared with the suspected atypical speech motor control group based on the overall low scores on the CAI-parameters, including the MRR-task. Ttofari-Eecen et. al. (2019) also found oromotor problems in their population; unfortunately, the results of the Dutch oromotor task was not known for all children in our study.

Speech Disorders Classification System (SDCS) Comparison with the SDCS is even more complicated as the different categories of the SDCS are defined at different levels: etiological processes (distal causes), speech processes (proximal causes), and clinical typology (behavioral phenotype) (Shriberg et al., 2017). Focusing on the categories based on clinical typology, the children in cluster I (phonological deficit cluster) can probably best be compared to children in the Speech Delay group (SD), as they showed no evidence of motor involvement (scores on PWV and MRR that are only slightly below the norm). The children in the other two clusters, with poor MRR would probably fall within the Motor Speech Disorder group (MSD). Further differentiation between subgroups of MSD requires additional speech motor tasks, which is beyond the scope of this study.

Clinical implications and future research

In the future, the tasks of the CAI will be supplemented with components that can provide a more detailed view of problems with motor planning and programming. Examples of these components: are systematic manipulation of conditions during speech such as speeding up; blocking auditory feedback and exercises to determine a short-term learning effect (Terband, Maassen, et al., 2019); as well as acoustic measurements of coarticulation and variability (Terband, Namasivayam, et al., 2019). The aim of the CAI is to provide SLPs with sufficient information to plan a well-fitting intervention that is specifically tailored to the individual child. In 2010, Williams et al. reported 23 different interventions for children with SSD. There are currently even more interventions available that were not included in that article, for example the Dynamic Temporal and Tactile Cueing (DTTC) (Strand et al., 2006) and since 2010 a few new interventions entered the market, for example Rapid Syllable Transition Treatment (ReST) (McCabe et al., 2017). More fine-grained analyses of underlying processing deficits could give a large contribution to the design of tailor-made therapy plans.

A classification of the different interventions and mapping these onto the outcomes of the process-oriented assessment might be a solution, as already described by several authors (Rvachew

& Brosseau-Lapré, 2016; Stackhouse & Wells, 1997; Wren et al., 2018). In their review, Wren and colleagues (2018) proposed a framework of five different categories of interventions: 1) environmental, 2) auditory–perceptual, 3) cognitive–linguistic, 4) production, and 5) integrated. For the children in the present study, it would perhaps be best to offer the children in cluster I (phonological deficit cluster) an intervention in the auditory-perceptual category or the cognitive-linguistics category because as the results show these children showed problems mainly with the tasks PN and NWI. This suggests that these children experience problems primarily in lemma access, word form retrieval, and phonological encoding. To treat these problems, the SLP can choose an intervention that falls under the auditory perceptual interventions or the cognitive-linguistic interventions. The auditory perceptual interventions target the perceptual skills of the child to change the speech output. The aim is to immerse the child in an auditory stimulation of word targets as well as auditory discrimination exercises that stimulate the child's phonemic awareness, for example cycles approach. The cognitive-linguistics interventions stimulate the higher-level processing to promote change in the speech through confronting a child with their reduced set of contrasts or increasing awareness of sounds in speech, for example Metaphon (Wren et al., 2018).

To help SLPs make a choice between these two interventions, Bron et. al. (2013) developed a flowchart for Dutch SLPs in which, for example, age is one of the factors. Younger children could have more difficulty with a cognitive-linguistic intervention (such as Metaphon), because this form of intervention relies more on the child's cognitive abilities; the children must learn to hear the differences between their pronunciation of the word and the correct pronunciation and they must also understand that their pronunciation refers to a different concept than the word they mean to pronounce, for example the difference between 'hat' and 'rat'. Younger children or children with lower cognitive abilities and/or children with an inconsistent error pattern tend to benefit more from a phonological cycles approach then from Metaphon (Bron et al., 2013).

The second group of children (cluster II, phonological and motoric deficit) scored worse on the speech motor tasks of the CAI (sequences of the MRR with more problems with the bi-tri syllabic sequences) than the children in cluster I; they also have more problems with the pronunciation of /l/ and /r/ (level 5) and the CCVC structures. These children have problems with the following underlying speech processes: lemma access, word form selection, phonological encoding and speech motor planning and programming (see Figure 1). The interventions in the category production could be a good choice; they can benefit from the guidance on phonetic placement or manner and imitation in combination with one of the interventions in the auditory perceptual or the cognitive-linguistic group.

The last group of children (cluster III, severe phonological and motoric deficit) score low on all the tasks and especially on the speech motor tasks. What also distinguishes this group from the other two clusters is the additional lower score on the auditory discrimination task. Integrating an auditory perceptual intervention with one that is more focused on the motor speech system (production) could help to fill the child's phonological system and reduce the speech motor difficulties. Currently, SLPs combine interventions and usually choose the intervention based on availability and own experience (Diepeveen et al., 2020; Joffe & Pring, 2008). Hopefully this will change in the future and SLPs will make their decisions during clinical reasoning on a process-oriented assessment and the framework described by Wren et al. (2018). Further development of treatment planning frameworks, flow charts and decision trees on additional assessments leading to specific treatment recommendation/prescription are warranted.

Conclusion

In summary, the results of this study demonstrated three underlying principal components of the CAI-parameters for a group of children with SSD. The components showed a different pattern compared to a study with typically developing children with the same CAI-parameters. Three different clusters of children could be identified. The largest group showed problems compared to the norm group only at the phonological level and could be characterized as having a phonological deficit. The second, much smaller group had the same problems but also experienced some difficulties at the speech motor level. This group was termed as having phonological and motor deficits. The third group, equal in number to the second, showed extensive problems at both the phonological and speech motor level and could be characterized as having severe phonological and motor deficits. This data-driven clustering shows that there seems to be a difference in severity of the speech disorders amongst the three clusters, and different profiles of speech processing problems could be detected in our sample. The profiles are informative with respect to treatment planning in that each profile implies a specific intervention approach. More comparative research is needed to test the diagnostic accuracy of process-orientated diagnosis methods including more and different children, for example children with dysarthria, and controlling for possible additional factors such as behavioral characteristics and language impairment (Allison et al., 2020).

Supplementary Materials: Not applicable.

Author Contributions: Conceptualization, S.D., B.M., H.T., and B.S.; methodology, S.D., B.M., and H.T.; data collection, S.D., L.H., A.Z., C.M-H.; data analyses, S.D., B.M., and H.T.; writing—original draft preparation, S.D.; writing—review and editing, S.D., B.M. and H.T.; visualization, S.D., B.M., and H.T.; supervision, B.M., H.T., and B.S.; project administration, S.D. All authors have read and agreed to the published version of the manuscript.
Funding: This research received no external funding.

Institutional Review Board Statement: The study was conducted in accordance with the Declaration of Helsinki,and approved by the research ethics committee of the Radboud University Nijmegen Medical Centre (file number: CMO 2016-2985), and the internal review board ETCL of de faculty of Humanities, Utrecht University (file number: doorn026-01-2017).

Informed Consent Statement: Informed consent was obtained from all subjects involved in the study.
Data Availability Statement: The data presented in this study are available on request from the corresponding author. The data are not publicly available due to articles in preparation.
Acknowledgments: The authors would like to thank all children that participated in this study and their parents or caregivers for their time and effort, Anniek van Doornik, student-SLPs and SLPs for their help with participant recruitment and test administration, and two anonymous reviewers for their constructive comments on an earlier version of this manuscript.
Conflicts of Interest: The authors declare no conflict of interest.

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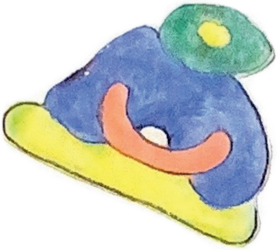
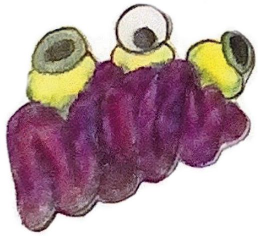
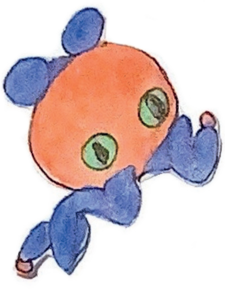
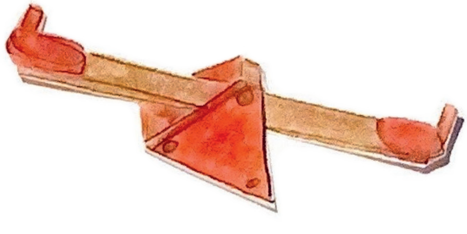




CHAPTER 7



Summary and General Discussion



Summary

Speech sound disorders (SSDs) are among the most common disorders that speech and language pathologists (SLPs) encounter when working with children (Joffe & Pring, 2008; Priester et al., 2009; Waring & Knight, 2013). Speech problems can affect children's (academic) performance (Cabbage et al., 2018; McCormack et al., 2009; McCormack et al., 2010; Preston et al., 2013), so it is important that they be diagnosed at an early age. After a child is registered for speech-language therapy, the SLP conducts a case history, an observation, and a speech assessment during the first appointments with the child and their parent(s)/caregiver(s). The SLP then takes further steps to shape clinical reasoning and decide on a suitable intervention. In several countries, it is common to use a picture naming task to assess the speech of children with an SSD (Broome et al., 2017; Joffe & Pring, 2008; McLeod & Baker, 2017; Priester et al., 2009; Skahan et al., 2007). We wanted to know how SLPs in the Netherlands generally diagnose children with SSDs. We also wanted to identify how SLPs make decisions in their clinical reasoning about children with SSDs and choose the most suitable intervention.

In order to obtain a correct diagnosis, an SLP assess the different speech processes and concludes where in the speech process the child has difficulties. Terband, Maassen and Maas (2019) suggest that an SLP includes tasks in the speech assessment that assess the following skills: lemma access, word form selection, phonological encoding, speech motor planning and programming, and speech motor execution. Since there is no single speech assessment instrument that can summarise the different underlying speech processes, we developed the Computer Articulation Instrument (CAI). It comprises four tasks that together provide a comprehensive overview of the speech production processes. In a recent study, our research group concluded that the CAI is a reliable and valid instrument for mapping speech (van Haaften et al., 2019). We used the CAI to study phonological development in typically developing children and set norms (van Haaften et al., 2020; see information in the introduction of this thesis). In the present study, we wanted to further examine the reliability and validation of the speech-motor tasks in the CAI and evaluate the use of the CAI in clinical practice.

This thesis focuses on three aims: 1) to gain insight into the clinical reasoning of SLPs during the diagnostic process and the treatment trajectory of children with SSD; 2) to determine the role the recently developed CAI might play in the diagnostic process, and determine the instrument's reliability, validity, and responsiveness for assessing underlying speech production processes and deficits (especially for the MRR); and 3) to investigate the role the CAI could play in differentiating subtypes of SSDs and thus its clinical value for assessing speech profiles.

Chapter 1 sketched the theoretical framework of the project. It provided background information on the development of speech and outlined a model that can guide process-oriented diagnostics. Furthermore, it elucidated how this model led us to develop the CAI and detailed the aims and outline of this thesis in this context.

Clinical reasoning

Chapter 2 explored the clinical reasoning Dutch SLPs use when treating children with SSDs. There is broad agreement that the clinical reasoning of experienced clinicians is based on an interaction between domain-specific knowledge, experience, and intuition (Ginsberg et al., 2016; Higgs & Jones, 2008). Clinical reasoning can be complex when it relates to children with SSDs. Several classification systems have been developed with either an aetiological, a descriptive-linguistic, or a processing approach (Tyler, 2010), such as the Model of Differential Diagnosis (MDD, Dodd, 2014) and the Speech Disorders Classification System (SDCS, Shriberg et al., 2010, 2017). Children with SSDs form a heterogeneous group. The differences between children are related to aetiology and speech characteristics, as well as to underlying processing deficits, severity, involvement of other aspects of the linguistics system, treatment response, environmental factors, and maintenance factors (see also Dodd, 2011). This makes it difficult to get indications for purposeful speech intervention for children with SSD based on the existing classification systems, which are mostly based on aetiology or speech characteristics alone.

Another approach that could help in the diagnostic process is the International Classification of Functioning, Disability and Health (ICF) (Cunningham et al., 2017; McCormack et al., 2010; McLeod, 2004, 2006). The holistic approach of the ICF encompasses three components (aetiological, descriptive-linguistic, and processing) that are described in three perspectives: description of the physiological functions and anatomical parts of the body (body structures and body functions), description of the execution of a task or action (activity), and description of the involvement in a real-life situation (participation). An SLP can gain insight from these three perspectives of the ICF in the diagnostics process. Most SLPs use a standardised single-word test to obtain information about the physiological functions and anatomical parts of the child's body (Skahan et al., 2007). According to Macrae (2017), a single-word test should include a survey of all the consonants and vowels in all positions multiple times, a connected speech sample, and a phonological analysis, and it may include inconsistency testing. An SLP using the ICF approach also gathers information on contextual factors (environmental and personal factors) for example during the case history (McLeod & Bleile, 2004). After the diagnostic phase, the SLP chooses an intervention that fits the differential diagnosis, the therapeutic and scientific perspectives, and elements of the child's background, such as age, family circumstances, and parental collaboration (Baker & McLeod, 2011; Dodd & Bradford, 2000). SLPs are forced to make choices throughout the whole process of guiding children with SSDs and their parents/caregivers, but little was known about these choices. Thus, we conducted a study to gain insight into this process in the Netherlands.

That study featured a concurrent triangulation (use of multiple methods) design. 137 SLPs completed an online survey for the quantitative study, and 33 SLPs were interviewed by SLP students for the qualitative study. The SLPs were recruited via social media or approached directly by the students.

The results from the questionnaire and interviews indicated that SLPs take two to four sessions to complete the diagnostic process. Most SLPs specified that they conduct a case history with the child's parents/caregivers in which they ask questions that cover 19 topics. Most SLPs ask about the child's hearing and speech language development, the course of the disorder, and behavioural aspects (such as the child's reaction when misunderstood).

Next, SLPs conduct an assessment that also contains an observation of several skills such as the child's communication strategy, oral skills, speech characteristics, and/or interactions with the parent/caregiver. Dutch SLPs often use the Nederlands Articulatie Onderzoek (NAO) (Baarda et al., 2014), the Metaphon assessment (Leijdekker-Brinkman, 2005), and the Hodson Assessment of Phonological Patterns (HAPP, Hodson, 2004). The NAO is mainly used when an SLP suspects a phonetic disorder, while the Metaphon assessment and HAPP are mostly used for a suspicion of a phonological disorder. A small group of SLPs also use the Dyspraxieprogramma (Erlings-van Deurse et al., 1993), which is based on The Nuffield Dyspraxia Programme, but they do not use the entire assessment due to its length and they do not use it often.

After the assessment, the SLP formulates a diagnosis. Our study found that SLPs in the Netherlands use 85 labels. We could identify several clusters of labels, which yielded five unique categories. The most common of the five was a phonological disorder, followed by a phonetic disorder, Childhood Apraxia of Speech (CAS), dysarthria, and fluency disorders.

After the diagnosis, the SLP plans the intervention based on the diagnostic label combined with the child's age and behaviour and the SLPs own experience. We found that most of the interventions mentioned are being used for all diagnostic labels. In the case of a phonological disorder, SLPs often use the cycle approach (Hodson & Paden, 1983) and Metaphon (Leijdekker-Brinkman, 2005). For children diagnosed with a phonetic disorder, Logo-art and oral myofunctional therapy are often chosen. SLPs often select the Dyspraxieprogramma when planning an intervention for children diagnosed with CAS.

The structure of the invention also differs for these three diagnoses (we did not include dysarthria and fluency disorders because few SLPs mentioned them). Differences include using target words or specific speech sounds or practising alternating sounds before practising real words. SLPs also use different didactic strategies, such as imitation and awareness versus drilling and motor learning. SLPs reported that they focus on helping the child apply the learned strategies in everyday life, which, they say, is not always included in the intervention programme. The SLPs mentioned that the involvement of parents/caregivers during the intervention period and the extent to which the child practises at home are factors that can reduce the duration of the intervention period.

Furthermore, the intervention period is reportedly shorter and more efficient if SMART goals (specific, measurable, achievable, realistic, and timely) are defined at the beginning of the sessions and the goals are revised if needed. According to the SLPs, it is important to formulate the goals with the parents/caregivers. If progress stagnates, the SLP will provide a break for the child during the intervention period. SLPs hope that a method will be developed that can be used with all children and that will motivate and stimulate them. In addition, they expressed that it would be ideal if the material could be presented/administered via a computer or tablet.

Overall, we concluded that there is no consensus among SLPs in the Netherlands on the terminology of the different SSDs and there are many idiosyncrasies in the diagnostic labels used. Our results also indicated that most SLPs choose an intervention based on availability and their own experience, rather than scientific evidence about the treatment of the diagnosed deficiency.

Maximum Repetition Rate

To help SLPs differentiate between underlying speech processes, our research group developed a speech assessment: the Computer Articulation Instrument. Previous studies have examined the use of the CAI with typically developing children and determined that it is a valid and reliable instrument (van Haaften et al., 2020; Maassen et al., 2019; van Haaften et al., 2019). This thesis focuses on the development and validation of the Maximum Repetition Rate (MRR) task and its contribution to profile assessment; this was described in Chapters 3 and 4.

Chapter 3 described the establishment of the protocol for the diadochokinetic task of the CAI. This task is often used to differentiate children with an underlying speech motor deficit from children with problems based on a not (yet) properly developed phonological system (Icht & Ben-David, 2014; Rvachew & Brosseau-Lapré, 2012; Wang et al., 2008; Yang et al., 2011). However, there is no uniform method of administering and analysing the MRR task, which makes it difficult to compare the results of different studies worldwide (Gadesmann & Miller, 2008). For this study, we developed a protocol based on existing research (Rvachew & Brosseau-Lapré, 2012; Thoonen et al., 1996; Wit et al., 1993). In this protocol, the diadochokinetic task consisted of six sequences: /papa../, / tata../, and /kaka../, pata../ and /taka../, /pataka../ (Maassen et al., 2019). We used the following method to obtain the MRR: total number of syllables divided by total duration of the sequence (count per time).

We recruited 1,524 Dutch-speaking children aged 2;0 to 6;11 years and the sample was representative for gender, urbanisation, and geographic region. In total, 14 SLPs assessed the younger children (2 to 4 years of age) and 110 SLP students administered the protocol for the older children (4 to 7 years of age). All the research assistants were trained to assess children using the protocol. The computer gave the instruction which maximised the standardisation of the task. The children were asked to imitate the following sequences: three monosyllabic sequences (/papa../, /tata../ and /kaka../), one trisyllabic sequence (/pataka.../), and two bisyllabic sequences (/pata../ and /taka../). For each sequence, the children were given some practice trails in the following order: 1) a short sequence of three syllables at a normal speech rate; 2) a longer sequence of six syllables at a normal rate; and 3) a series of several syllables at a faster rate (the audio example contained 12 syllables at a faster rate). After these practice trails, the children were asked to produce the syllable sequences as quickly as possible without an example, and they were given up to three attempts for each sequence.

The acoustic signal (child's speech) was automatically stored on the computer's hard disk in one recording. These samples were analysed according to the protocol. For example, a sequence could not contain a pause or a speech error. For each of the six correctly uttered sequences, the MRR was calculated for the trail with fast rate and the fastest rate. We determined how many children produced the different sequences of the MRR and we drew a comparison between completing the different sequences (e.g., monosyllabic sequences versus multisyllabic sequences). Repeated measures ANOVAs were conducted to compare the best performance on the fast (with example) and fastest (without example) attempt per sequence. Differences were calculated between the first three to ten syllables of each sequence. Finally, we calculated intraclass correlation coefficients with two-way mixed-effects models featuring no fixed effects between the MRRs over each number of syllables compared with the gold standard of ten syllables.

Four conclusions could be drawn from this study and were included in the final CAI protocol. First, children under the age of three have great difficulty in performing this task; many children could not produce a sequence with three or more syllables. Children made more errors in the trisyllabic sequence, /pataka../, than in the other sequences. Furthermore, only 21% of the younger children (age <3;0) produced all three monosyllabic sequences. Therefore, we decided to exclude this younger group of children from the norm data for the CAI. The remaining children (age 3;0 to 6;11) were included in the further analysis to determine whether they could perform all the sequences, and which attempt per sequence was the fastest.

Second, we noted that not all children could perform all the sequences, and the bi- and trisyllabic sequences were especially difficult. Therefore, the new CAI protocol prescribes that the bi- and trisyllabic sequences should not be administered if children cannot produce the monosyllabic sequences, to ease the burden of the whole test battery.

Third, when determining which attempt was the fastest, we found that children were faster when they received the instruction “as fast as possible” than when they were asked to imitate the example with a faster speech rate. However, this result had only a small effect size, so it must be interpreted with caution. The CAI protocol for the MRR task therefore stipulates that the SLP should determine which attempt of the last two instructions is fastest and select that attempt for comparison with the norm data.

The final (fourth) conclusion from this study was that it is unnecessary to obtain a 10- to 12-syllable sequence as previous research suggested (Rvachew & Brosseau-Lapré, 2012; Thoonen et al., 1996; Wit et al., 1993; Yaruss & Logan, 2002). We found that many children generated sequence lengths of three to about ten syllables (excluding the first and last syllable of the sequence). We also found no differences in repetition rate between shorter and longer sequences (except for the sequence /pa../ with a small effect size). Consequently, we decided to specify that a child must produce at least five syllables per sequence.

Chapter 4 described a large cross-sectional study about the normative data for the MRR of a group of typically developing Dutch children aged 3;0 to 6;11. The protocol from Chapter 3 was used to analyse the data about the 1,041 children. All MRR sequences increased significantly with age. The sequence /pa../ was the fastest of all the sequences, followed by /ta../, /pata../, /taka../, /ka../, and /pataka../. This result was in line with previous studies conducted in Dutch and in other languages. Another conclusion of this study was that boys pronounce the different sequences more quickly than girls. The outcomes of this study could help an SLP determine whether a child has a typical development or not. It remains to be seen whether these norm data can also distinguish a child with a suspected motor speech disorder from a child with speech problems based on an incomplete phonological system. The next section describes two studies to clinically validate the CAI.

Clinical validation

Chapter 5 described two studies used to determine the clinical value of the CAI in the diagnostic phase. Two groups of children with speech language impairments and SSD participated in the two studies. The design of the two studies was the same. The children were assessed with the CAI by an SLP. The first study included 93 children aged 3;0 and 4;0 years. These children had been diagnosed with an SSD and attended an intervention class for young children with a developmental language disorder. We compared the child’s scores on the Picture Naming task of the CAI to the intelligibility judgements (good, moderate, poor) given by the child's SLP. Children with a poor intelligibility judgement had lower CAI scores on the Picture Naming task than children with a moderate intelligibility judgement. Children with a good intelligibility judgement scored highest on the Picture Naming task. We found no correlation between nonverbal intelligence and language scores and the performance on picture naming and intelligibility.

The second study described in **Chapter 5** involved a group of 41 children with SSDs aged 3;0 to 6;4. The children had been diagnosed with a phonological disorder (*n* = 36), CAS (*n* = 2), or an unknown disorder (*n* = 3). The aim of this study was to determine the relationship between the severity of speech difficulties as judged by SLPs and the five CAI factors (van Haaften et al., 2019). For the CAI factors based on Picture Naming, NonWord Imitation, and the bisyllabic and trisyllabic sequences of MRR, we discovered a significant difference in performance between the severity groups. The group labelled 'severe SSD' scored worse on these factors than children labelled 'moderate SSD'. For the factor word and nonword proportion of whole-word variability (PWV) and the monosyllabic sequences of the MRR, we found no significant differences between the moderate and severe groups. This implies that the results on Picture Naming, NonWord Imitation, and the bisyllabic and trisyllabic sequences of MRR could help an SLP diagnose a child with SSD.

The last study (**Chapter 6**) described a larger group of children with SSDs (*n* = 150). Some of the data from Chapter 5 were also used in this study. The sample consisted of 94 boys and 56 girls aged 4;0 to 6;6 years. Most of them had been diagnosed with a phonological disorder (*n* = 105); the others had been diagnosed with CAS (*n* = 17), a phonetic articulation disorder (*n* = 9), dysarthria (*n* = 5), or a disorder not further specified by the SLP (*n* = 2). Some children received multiple diagnoses. 11 children were recruited through regular schools and did not receive speech therapy at the time of the study, but they scored below percentile 16 on the CAI and thus were included in this study. The goal of this study was to examine whether the CAI can be used to differentiate and identify profiles (Maassen et al., 2019).

The results found slightly different factors for this SSD group than found in the factor analysis of the norm group (van Haaften et al., 2019). Only three factors occurred: 1) all Picture Naming (PN) parameters plus the following parameters of the NonWord Imitation task (NWI): Level5, Simplification processes, and the Unusual processes; 2) the remaining parameters of the NonWord Imitation parameters including Word Repetition and NonWord Repetition; 3) the parameters of the MRR. Thus, the CAI can provide distinct factor profiles that are different from the norm group. The fact that PN and NWI parameters load largely on different factors implies that it is important to assess both tasks to get a broad view of the whole speech production process and parts of the chain. Children who make relatively few speech production errors when imitating nonwords may have relatively little difficulty in pronouncing new words they are learning, which could be a starting point for an intervention method.

A subsequent cluster analysis with a K-means analysis showed that the children could be divided into three groups. Cluster I was the largest group and had the highest scores on every CAI parameter. Children scored lower than the norm group on the Picture Naming task (which uses the entire chain of speech processes) and the NonWord Imitation task. There was no difference in scores compared to the norm group on the MRR task, which assesses motor planning and programming. Thus, this group of children mainly has problems with the phonological system and this group was labelled as having a phonological deficit. The children in Cluster II had similar scores as those in Cluster I and scored significantly lower on a few CAI parameters (syllabic structure parameters and the MRR task). These children seem to have more difficulty with motor planning and programming, so Cluster II was named mild phonological deficit with motoric deficit. Cluster III was named severe phonological and motoric deficit because of the low scores on all the CAI parameters.

When the three clusters were compared to other variables, we found that the clusters differ on intelligibility (Intelligibility in Context Scale (ICS); McLeod et al., 2013), word comprehension (Peabody Picture Vocabulary Test-III-NL (PPVT-III-NL); Schlichting, 2005), auditory discrimination (Testinstrumentarium Taalontwikkelingsstoornissen (TTOS-ADT); Verhoeven, et al., 2013), and the judgements of the SLPs and the parents/caregivers about the severity of the SSD. The children from Cluster I scored highest on these variables, followed by the children in Cluster II and then the children in Cluster III. We found no significant differences between the clusters for age, gender, or the diagnosis given by the SLP. Three conclusions were described: 1) there are different profiles of SSD; 2) severity plays a role; and 3) they cover a spectrum of degrees of involvement of different underlying problems.

General Discussion

This thesis had three aims: 1) to gain insight into the clinical reasoning of SLPs during the diagnostic process and the treatment trajectory of children with SSD; 2) to determine the role the recently developed CAI might play in the diagnostic process, and determine the instrument's reliability, validity, and responsiveness for assessing underlying speech production processes and deficits (especially for the MRR); and 3) to investigate the role the CAI could play in differentiating subtypes of SSDs and thus its clinical value for assessing speech profiles. To gain insight into these goals, six studies were conducted and described in five papers (see Chapters 2-6). The results of these studies are important to the clinical work of SLPs and could influence their clinical reasoning for children with SSDs.

This general discussion will address the steps SLPs take during clinical reasoning and present the results of the chapters described in this thesis. It will also discuss the clinical implications of the results of this thesis and make recommendations for further research.

When guiding a child with an SSD and their parents/caregivers, the SLP takes several steps: 1) case history, 2) observation, 3) process-oriented assessment, 4) conclusion of the diagnostic process, 5) intervention decision (method, materials, and goals), 6) intervention process, 7) continuous evaluation during the intervention, and 8) final evaluation. These steps are not sequential; for example, the observation starts when the child enters the examination room and continues during all the following steps. It may also be necessary to return to the assessment process when, for example, a child makes little progress during the intervention and a re-evaluation is needed. The following paragraphs explain the steps in more detail and outline what we have learned from the studies described in this thesis. Results from Chapter 2 relate to all eight steps of the clinical reasoning process and Chapters 3 to 6 focus mainly on step 3 (process-oriented assessment). The final chapter (Chapter 6) also examines step 4 (conclusion of the diagnostic process).

Case history

After the parents have enrolled their child in speech-language therapy, an SLP invites them to an initial session in which the SLP takes a case history (1). The information obtained in a case history has valuable information the SLP can use to diagnose the child and plan the intervention. Parents provide information about their child's speech and language development, which can provide initial insight into speech motor skills or phonological development. Parents can also talk about their child's motivation and character. Taking a case history in the 'office' is feasible for SLPs in private practice, but that is not the case for SLPs in schools (see Chapter 2).

Based on the findings in Chapter 2, we recommend that each SLP conducts a case history at the beginning of the intervention process. A form that parents can fill in at home could be a tool to obtain information in all settings (school and private practice), and we recommend that a digital form be developed. Recent years have taught us that conducting a case history over video call is another option. In Chapter 2, we mentioned that the following categories are important to include in a case history and can be included on an online case history form: demographic information, hearing, speech- and language development (e.g., babbling, first words), course of the disorder, the child's communication skills, the child's reactions when misunderstood/compensatory behaviour, the child's awareness of intelligibility, reactions of the environment on intelligibility, oral habits, cultural environment, multilingualism, developmental history (sensorimotor-, psychosocial-, cognitive development), birth/health/medical history, the child's preference/likes/dislikes, and the parents'/child's motivation and ability to practice. However, it remains important that SLPs continue to ask for clarification of written information received from parents/caregivers.

It would be advisable to carry out a study on the validity of the information obtained by a case history with a form compared to a face-to-face interview or a combination of both. Another recommendation is to use the Speech Participation and Activity – Children (SPAA-C, McLeod, 2004), a tool that can help to generate a picture of the child's interests and strengths from other people in the child's life, such as grandparents or a teacher. It is designed to consider a child's ability to participate fully in society. Unfortunately, it has not yet been translated into Dutch.

Observation

As mentioned above, observation is an ongoing process during all phases. The topics an SLP observes can differ by phase. Each child and their parents/caregivers are different, and the SLP observes certain items depending on the situation. As stated in Chapter 2, SLPs mentioned that they observe the following topics: the child's communication strategy, oral skills, speech characteristics, interaction with other individuals, coping with/awareness of the speech problems, intelligibility, language development, hearing/auditory processing, sensory/motor/cognitive development, attention/focus, groping, and rate of the child's and parents' speech (Diepeveen et al., 2020). SLPs observe these topics during the assessment and intervention phases. In the intervention phase, the SLP also monitors the child's progress towards defined goals. This could involve the generalisation of the exercises in daily practice observed during small talk (Chapter 2). During a session, the SLP also keeps track of how often the child achieves a speech target correctly and the child's failures, in order to provide the appropriate feedback (Waelkens, 2017). To our knowledge, no observation tool exists for children with SSDs; the question is whether such a tool would be desirable to use.

Process-oriented assessment

In the next step (3), the SLP uses speech assessments to gather information. Speech disorders do not develop in a vacuum, but are influenced by and affect language and more general cognitive strengths and weaknesses from the onset (Maassen & Terband, 2015; Terband, Maassen, & Maas, 2019). Macrae (2016) suggested that an assessment battery is needed to determine whether a child has an SSD, which type of SSD, and which evidence-based intervention might be most suitable.

Differential diagnosis of SSDs remains a clinical challenge. It is difficult to choose the right assessment because children with SSDs form a heterogeneous group; the children differ in severity, aetiology, proximal causes, and speech characteristics (Dodd, 2014; Ttofari Eecen et al., 2018). One study described in Chapter 2 (Diepeveen et al., 2020) used a wide variety of assessments to assess children, depending on the information from the case history and/or the observations. The most popular assessment was a naming task which was used for all children regardless of the suspicion of a certain diagnosis. To get an overview of all aspects, a comprehensive assessment battery should include a hearing test, an oral-musculature assessment, single-word production (including polysyllabic words and nonwords), and connected speech sampling (Murray et al., 2020; Terband, Maassen, et al., 2019). Others also recommend administering a language comprehension test (Bleile, 2002; McLeod & Baker, 2017; Miccio, 2002; Tyler & Tolbert, 2002).

Existing speech assessments do not assess the underlying processes involved: lemma access, word form selection, phonological encoding, speech motor planning and programming, and speech motor execution (Terband, Maassen, & Maas, 2019). To fill this gap, we created the CAI (Maassen et al., 2019) which has been proven to be a valid and reliable instrument. The CAI includes norm data of children aged 2 to 7 years for four tasks (Diepeveen et al., 2019; 2021; Haaften et al., 2020; van Haaften et al., 2019). Those four tasks are: 1) Picture Naming; 2) NonWord Imitation; 3) Word and NonWord Repetition; and 4) Maximum Repetition Rate (Maassen et al., 2019).

The Picture Naming task is included because it involves the entire chain of speech processes, from visual conceptual processing to lemma access, word form selection, phonological encoding, speech motor planning and programming, and speech motor execution (Terband, Maassen, et al., 2019). In Chapter 5, we show that this task can differentiate between severity of the speech problem: a poor intelligibility judgement of the child’s speech was related to a CAI score that was lower than that of children with a moderate intelligibility score. A good intelligibility judgement was related to the highest scores on the Picture Naming task (van Haaften et al., 2019).

This was also confirmed in the last study of this thesis (Chapter 6). The percentage consonant correct initial (PCCI) of the norm group (one outcome of the Picture Naming (PN) task) was higher than the PCCI of the SSD group. A measure such as the PCCI is a quantitative measure of the severity of the speech problem (Dale et al., 2020; Shriberg et al., 1997). The second task in the CAI is NonWord Imitation (NWI). During this task, children cannot use the lexicon because the lexicon contains no nonwords. It therefore directly uses the phonological encoding system. Children analyse the phonological structure of the nonword or choose the route of the auditory to the speech motor planning system. An SLP could compare the results of the PN task and the NWI task; this allows the SLP to determine to what extent the lexicon plays a role in the speech production process. Krishnan et al. (2013) concluded that imitation of non-words depends not only on phonological storage or phonological working memory but also on non-linguistic motor skills and planning. In the studies in this thesis, the nonword task was a separate factor; this means the nonword task measures something else than the other tasks. Overall, the children performed worse on the parameters of the NWI task than the PN task, but no cluster was found with children who had a relatively large discrepancy between the NWI task and the PN task.

The next task, the repetition of (non)words, was added because it involves the production process of word form selection more than PN does. When a child repeats a word, they not only select a word form based on the lemma, but also directly based on auditory input (Richard Hanley et al., 2004). This task assesses the consistency of speech production. Dodd (2014) added the inconsistent phonological disorder category to the Model of Differential Diagnosis (MDD); it refers to children who are inconsistent in their speech because they produce inconsistent errors during multiple repetition of the same word. This was confirmed by Ttofari-Eecen et al. (2018), who found a group of children (19%) who met the diagnostic criteria of the inconsistent phonological disorder category. The repetition task of the CAI might also differentiate between children with consistent errors and those with inconsistent errors; we did not examine this in the current thesis. We do know that consistency plays a role in differentiating between groups of children with SSDs. In the study described in Chapter 6, we found that one group of children performed significantly worse on this task than the other two groups (Diepeveen et al., submitted).

The last task of the CAI, Maximum Repetition Rate (MRR), was described in several studies in this thesis. First, a protocol was proposed to score children's performance on the six parameters of the CAI. This led to the exclusion of children younger than three years of age. Second, norm scores were presented; the MRR sequences improved significantly with age. Third, the MRR was used in two other studies described in this thesis that included children with SSD.

This task has been credited with playing a role in detecting speech motor planning and programming in several studies (Thoonen et al., 1996; Wit et al., 1993) in which the diadochokinetic task did distinguish between children with dysarthria and CAS and other children. Children with dysarthria and CAS are known to have problems with speech motor planning and programming

(Morgan & Liegeois, 2010; Terband et al., 2019). In this thesis, we found that two groups of children with SSD performed worse on the MRR sequences than one other group, which means the MRR could help to distinguish groups of children (Diepeveen et al., submitted). We found no differences between the severity of the speech problem for the monosyllabic sequences of the MRR. However, an SLP could use the bi- and trisyllabic sequences of MRR to help diagnose a child with SSD (van Haaften et al., 2019).

Conclusion of the diagnostic process

The study described in Chapter 5 found no correlation between the diagnoses given by the SLPs and the clusters that emerged based on the children's CAI results. The question is whether this was due to using a new diagnostic tool or due to the SLPs' clinical reasoning processes. In Chapter 2, we described how SLPs in the Netherlands use 85 labels to diagnose children with SSD. There is also an overlap in the symptoms that SLPs assign to different diagnostic labels. This causes problems in step 4 (conclusion of the diagnostic process).

Based on the studies described in this thesis, it might be more interesting to avoid assigning a specific diagnostic label to a child, but instead to determine which underlying process causes the speech problems and change to a diagnostic taxonomy or dimensional diagnosis. After all, if we know which underlying process causes problems, an SLP can connect it to a targeted intervention (Terband, Maassen, et al., 2019). This shift to a dimensional diagnostic process means that SLPs need to learn to think differently and use different assessments in their diagnostic process, such as the CAI.

Lecturers at Dutch universities of applied sciences (SLP deparments) could introduce this method into the diagnostics of children with SSD. If students learn to work this way during their education, they could take this method with them during internships and into their future careers. In addition, post-graduate courses about children with SSDs could teach SLPs to use an instrument that maps out the underlying speech processes to diagnose children with SSDs, such as the CAI.

A problem is that SLPs in the Netherlands are required to submit a diagnostic code to the patient's health insurer. The possible codes are phonetic articulation disorder, phonological articulation disorder, general articulation disorder, other articulation disorder, dysarthria, and apraxia/dyspraxia (Raaijmakers & Dekker, 1993). Clearly, this system must change.

During step 4 (conclusion of the diagnostic process), SLPs also create an overview of other elements that could influence the child's functioning in communication. This might include information gathered during steps 1 (case history) and 2 (observation), such as character, motivation, and opportunities to practise at home. These factors could influence the choices SLPs make in the next step (intervention decision (method, materials, and goals).

In conclusion, an SLP can use the CAI to determine whether a child has an SSD. It has been shown that the CAI can distinguish between groups of children: in one study (Chapter 6) three distinct groups could be detected in a group of 150 children diagnosed with an SSD. The groups were labelled based on their CAI profile (results on the different tasks). The first group was labelled as the phonological deficit group, the second group as the mild phonological and motoric deficit group, and the third group as the severe phonological and motoric deficit group.

Intervention decision, process, and evaluation

Once an SLP has identified the underlying processes or diagnosis and concluded the diagnostic process, they should reflect on the intervention decision (5), the process (6), and the evaluation of the intervention (7 and 8). These steps are further examined in Chapter 2 of this thesis. The results show that SLPs choose an intervention based on availability and their experience with it. They may use the same intervention for children with different underlying speech process deficits (Diepeveen et al., 2020). This also could be due to a lack of studies with higher levels of evidence, as Baker and McLeod (2011) described in a review of 134 published studies. The most frequently described studies (74.1%) involved lower levels of evidence (quasi-experimental studies, 41.5%; nonexperimental case studies, 32.6%). SLPs also combine interventions that probably do not achieve maintenance and generalisation of the learned speech sounds or words (Murray et al., 2014).

A child's performance on the CAI can help an SLP make a well-founded decision about which intervention to choose for that child. A child with a similar speech profile as the children in the phonological deficit group might be better off with an intervention that taps into the lemma access, word form selection, and phonological encoding. Such as, interventions that can be included in the auditory perceptual interventions or the cognitive-linguistic interventions (Wren et al., 2018). For the children in the mild phonological deficit and motoric deficit group, an intervention with a focus on planning and programming speech sounds could be a good starting point. The last group (severe phonological deficit and motoric deficit) (Diepeveen et al., submitted) would be better off with a programme that combines an auditory perceptual intervention with an intervention more focused on the motor speech system.

SLPs in the Netherlands believe it is important to set goals with the parents/caregivers because this helps them make progress during the intervention. The SLPs in our survey (Chapter 2) also indicated that they involve parents/caregivers in the selection of an intervention. SLPs also try to encourage them to get involved during the intervention period. Australian SLPs do that as well (Sugden et al., 2018). The Dutch SLPs indicated that they would like parents/caregivers to act as a co-therapist in correcting and modelling the child's speech to improve their speech skills. The SLPs believed this would ensure a short and effective intervention period (Diepeveen et al., 2020).

However, SLPs should be aware of challenges and obstacles that parents experience while completing home exercises, such as finding time to practise and knowing how to complete the tasks (Sugden et al., 2019; Thomas et al., 2017; Watts Pappas et al., 2016).

Future perspectives

This thesis describes the CAI as a tool for diagnosing children with SSDs and determining the severity of the speech disorder. In addition, the CAI has been shown to be helpful in identifying difficulties with underlying speech processes. To further investigate the underlying speech process deficits, a direct manipulation of the speech processes could complement the CAI. An example of such a tool is auditory perturbation: auditory feedback plays a role in sensory goals during speech sound production and in the acquisition and adaptation of the speech motor programme.

Furthermore, it plays a role in the control and correction of speech movements (Guenther & Perkell, 2004; Perkell, 2013). For example, children with CAS have an impairment in the coordinative structures underlying speech motor coordination, which causes poor feedforward control. This was examined in a series of computer simulations using the DIVA model. Four key speech-motor symptoms were detected: stronger coarticulation, distorted productions of speech sounds, searching articulatory behaviour, and high variability across productions (Terband et al., 2009).

Another study investigated the effect of auditory feedback masking on vowel space and voice onset time (VOT) for voiceless plosives in speech in school-aged children with CAS versus a control group (Iuzzini-Seigel et al., 2015). The children with CAS had a smaller vowel space area in a condition with masking of auditory feedback, and they produced shorter VOTs.

Terband et al. (2014) compared 17 typically developing children to 11 children with SSD (aged 3;9 – 8;7 years) to investigate the ability to compensate for and adapt to perturbed auditory feedback. The typically developing children were able to compensate and adapt their formant frequencies better than the SSD group. The degree of compensation and performance correlated with the PCCI of the NonWord Repetition task of the CAI. The authors suggested that impaired auditory-motor integration could play a role in SSDs.

All these findings have implications for SLPs during the assessment phase. Iuzzini-Seigel et al. (2015) recommend that SLPs vary in valid communication contexts instead of only assessing children in a quiet clinical surrounding. SLPs should also incorporate feedback manipulations into their intervention.

Another tool that can be implemented in the CAI is the (Normalised) Pairwise Variability Index. This has been used to quantify stress production in children with and without CAS (see Terband, Namasivayam, et al., 2019). Shriberg et al. (2008) investigated timing and stress characteristics of three siblings with CAS. One of the three had a significantly poorer score compared to the age-matched control. Other studies also have found that measuring speech variability can make an important contribution to the identification, assessment, and treatment of adults and children with speech disorders (Barbier et al., 2013; Marquardt et al., 2004; Terband et al., 2011). Although investigating speech variability may be a useful tool in assessing children with SSDs, some hesitation is necessary. Software automation and hardware integration are essential to make this method an easy-to-apply clinical tool and the technics for distorted speech are not that advanced yet.

A second aim for the future is to develop new guidelines for process-oriented diagnosis and treatment planning. Baker and McLeod (2011) published a review of intervention studies involving children with SSD published between 1979–2009. They identified 134 studies, 46 of which included distinct intervention approaches and 24 of which mentioned no specific procedures. These studies also included varying levels of evidence. That may be why SLPs choose an eclectic approach and combine perceptual, linguistic, and oral motor activities (Diepeveen et al., 2020; Lundeborg Hammarström et al., 2019).

Also, many children have speech problems that include a mix of phonological and motor deficits (Diepeveen et al., submitted; Namasivayam et al., 2020). SLPs are generally trained to focus on speech output skills, to think and work according to a diagnostic classification model based on behavioural symptoms, and to plan interventions based on workplace availability and personal experience (Diepeveen et al., 2020; Hegarty et al., 2020; Joffe & Pring, 2008; Sugden et al., 2018; Wren et al., 2018).

The new guidelines should also contain new interventions that Dutch SLPs did not mention in the interviews and questionnaire (Chapter 2). One new intervention is Rapid Syllable Transition Therapy (ReST; McCabe et al., 2017). It uses pseudo-word targets with varying lexical stress to simultaneously target articulation, prosodic accuracy, and coarticulatory transitions. A study by Murray et al. (2015) examined 26 children with CAS (aged 4–12 years) who were treated with ReST or the Nuffield Dyspraxia Programme–Third Edition (Williams & Stephens, 2004). Both groups progressed during the intervention sessions and the effects were still visible four months after the last intervention session. In a review study of interventions for children with CAS, ReST was one of three interventions with sufficient evidence for Phase III trials and interim clinical practice (Murray et al., 2014); this was confirmed by a quality appraisal of the published systematic reviews (Springle et al., 2020). Another of the three interventions in the review was the Nuffield Dyspraxia Programme (Williams & Stephens, 2004), which is also widely used in the Netherlands (Diepeveen et al., 2020). The third intervention with sufficient evidence is Dynamic Temporal and Tactile Cueing (DTTC), which aims to improve the efficiency of neural processing for better sensorimotor planning and programming of speech (Strand, 2020). DTTC was not mentioned in our interviews with Dutch SLPs.

The mentioned interventions mainly are aimed at improving motor speech skills. One of the new interventions aimed at improving phonological skills is Core Vocabulary Therapy (CTV). It is intended for children with problems related to intelligibility (Williams et al., 2021). Their phonological system is unstable, leading to inconsistent speech errors. The aim of CTV is to help the child produce as much speech as possible and practise this production consciously every day, so the transfer to spontaneous speech improves. These children become more aware of their own pronunciation and learn to control and correct themselves. Several studies have shown that children with inconsistent phonological speech made progress when treated with CTV (Crosbie et al., 2005; Dodd & Bradford, 2000; Flanagan & Ttofari Eecen, 2018). One of these studies compared two interventions (CTV and phonological contrast therapy); the data analysis showed that using CTV had a greater effect on children with inconsistent phonological speech disorder than the other intervention and a greater effect on those children than on children with a consistent phonological disorder (Crosbie et al., 2005).

The new guideline should not only help SLPs select the most suitable (eclectic) intervention for a specific child, but it should include advice on how to involve parents/caregivers in the intervention and, most important, how to encourage practise at home. The use of a tablet could be an option here (Dural & Ünal-Logacev, 2018; McKechnie et al., 2020; Wren & Roulstone, 2008). Saving time is crucial here because SLPs report having limited time and resources to help children and their parents/caregivers (Joffe & Pring, 2008; Lee, 2018). An online tool or computer app for SLPs could help them implement this new guideline in daily practice and assist them with clinical reasoning, diagnosis, and treatment planning.

In summary, the aim for the future is to improve primary care for children with SSDs by developing an evidence-based, process-oriented digital clinical route map for diagnosis and intervention planning and monitoring, in the form of an easy-to-use app.

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Nederlandse Samenvatting

Spraakstoornissen behoren tot de meest voorkomende stoornissen die logopedisten tegenkomen in het logopedisch veld (Joffe & Pring, 2008; Priester et al., 2009; Waring & Knight, 2013). Spraakstoornissen kunnen van invloed zijn op de (academische) prestaties van kinderen (Cabbage et al., 2018; McCormack et al., 2009; McCormack et al., 2010; Preston et al., 2013). Daarom is het belangrijk dat de kinderen op jonge leeftijd worden gediagnosticeerd en er een interventie wordt gestart. Nadat een kind is aangemeld voor logopedie, voert de logopedist een anamnese, een observatie en een spraak-taalonderzoek uit tijdens de eerste afspraken met het kind en zijn ouder(s)/verzorger(s). De logopedist neemt dan verdere stappen om het klinisch redeneren vorm te geven en te beslissen over een geschikte interventie. In verschillende landen is het gebruikelijk om een benoemtaak van plaatjes te gebruiken om de spraak van kinderen te beoordelen (Broome et al., 2017; Joffe & Pring, 2008; McLeod & Baker, 2017; Priester et al., 2009; Skahan et al., 2007). Onze onderzoeksgroep wilde weten hoe logopedisten in Nederland over het algemeen kinderen met spraakstoornissen diagnosticeren. Ook wilden we achterhalen hoe logopedisten in hun klinisch redeneren beslissingen nemen over kinderen met spraakstoornissen en de meest geschikte interventie kiezen.

Om tot een juiste diagnose te komen, beoordeelt een logopedist de verschillende spraakprocessen en concludeert waar in het spraakproces het kind problemen heeft. Terband, Maassen en Maas (2019) stellen voor dat een logopedist taken opneemt in het spraakonderzoek die de volgende vaardigheden beoordelen: het activeren van een lemma (semantische en grammaticale informatie van een woord), selecteren van een woordvorm (lexeem), fonologische encoderen, spraakmotorische planning en programmering, en de spraakmotorische uitvoering. Aangezien er geen enkel spraakonderzoek bestond dat de verschillende onderliggende spraakprocessen onderzoekt, hebben wij het Computer Articulatie Instrument (CAI) ontwikkeld. Het bestaat uit vier taken die samen een uitgebreid overzicht geven van de spraakproductieprocessen. In een recente studie concludeerde onze onderzoeksgroep dat het CAI een betrouwbaar en valide instrument is om spraak in kaart te brengen (van Haaften et al., 2019). We hebben het CAI gebruikt om de fonologische ontwikkeling bij normaalontwikkelende kinderen te bestuderen en normen te stellen (van Haaften et al., 2020; zie informatie in de inleiding van dit proefschrift). In de huidige studie wilden we de betrouwbaarheid en validatie van de spraak-motorische taken in het CAI verder onderzoeken en het gebruik van het CAI in de klinische praktijk evalueren.

Dit doelstellingen van dit proefschrift zijn: 1) het verkrijgen van inzicht in het klinisch redeneren van logopedisten tijdens het diagnostisch proces en het behandeltraject van kinderen met een spraakstoornis; 2) het bepalen van de rol die het recent ontwikkelde CAI speelt in het diagnostisch proces, en het bepalen van de betrouwbaarheid, validiteit en responsiviteit van het instrument voor het beoordelen van onderliggende spraakproductieprocessen en tekorten (met name voor de Diadochokinesetaak); en 3) het onderzoeken van de rol die het CAI speelt in het differentiëren van subtypen van spraakstoornissen en daarmee de klinische waarde ervan voor het beoordelen van spraakprofielen te bepalen.

Hoofdstuk 1 schetst het theoretisch kader van het project. Er wordt achtergrondinformatie over de ontwikkeling van spraak gegeven en er wordt een model dat richting kan geven aan procesgerichte diagnostiek gepresenteerd. Verder wordt er toegelicht hoe dit model leidde tot de ontwikkeling van het CAI en worden de doelen en de opzet van dit proefschrift in deze context nader toegelicht.

Klinisch redeneren

In **hoofdstuk 2** wordt het klinisch redeneren onderzocht van Nederlandse logopedisten bij het onderzoeken en behandelen van kinderen met spraakstoornissen. Er is een brede overeenstemming dat het klinisch redeneren van ervaren therapeuten gebaseerd is op een interactie tussen domeinspecifieke kennis, ervaring, en intuïtie (Ginsberg et al., 2016; Higgs & Jones, 2008). Klinisch redeneren kan complex zijn bij kinderen met een spraakstoornis. Er zijn verschillende classificatiesystemen ontwikkeld met ofwel een etiologische, een beschrijvend-linguïstische, ofwel een linguïstische verwerkingsbenadering (Tyler, 2010), zoals het Model of Differential Diagnosis (MDD, Dodd, 2014) en het Speech Disorders Classification System (SDCS, Shriberg et al., 2010, 2017). Kinderen met spraakstoornissen vormen een heterogene groep. De verschillen tussen kinderen hangen samen met etiologie en spraakkenmerken, maar ook met onderliggende verwerkingsdeficiënties, ernst, betrokkenheid van andere aspecten van het taalsysteem, behandeleffect, omgevingsfactoren, en transfer van het geleerde in de therapie (zie ook Dodd, 2011). Dit maakt het moeilijk om een doelgerichte spraakinterventie voor kinderen met spraakstoornissen op basis van de bestaande classificatiesystemen te bepalen, aangezien deze meestal alleen gebaseerd zijn op etiologie van de spraakstoornis of op de spraakkenmerken.

Een andere benadering die kan helpen bij het diagnostische proces is de International Classification of Functioning, Disability and Health (ICF) (Cunningham et al., 2017; McCormack et al., 2010; McLeod, 2004, 2006). De holistische benadering van de ICF omvat drie perspectieven: beschrijving van de fysiologische functies en anatomische delen van het lichaam (lichaamsstructuren en lichaamsfuncties), beschrijving van de uitvoering van een taak of handeling (activiteit), en beschrijving van de betrokkenheid bij een real-life situatie (participatie). Een logopedist verkrijgt inzicht door gegevens te verzamelen op drie perspectieven van de ICF tijdens het diagnostisch proces. De meeste logopedisten gebruiken een plaatjesbenoemtaak waarbij elke klank een keer wordt aangeboden in een bepaalde positie om informatie te verkrijgen over de fysiologische functies en anatomische delen van de spraak van het kind (Skahan et al., 2007). Volgens Macrae (2017) moet een spraakonderzoek een overzicht bevatten van alle medeklinkers en klinkers in alle posities en deze moeten meerdere keren worden bekeken. Daarnaast is het belangrijk om de spontane spraak te bekijken waarmee een fonologische analyse gedaan kan worden, en de inconsistentie tussen woorden moet bekeken worden. Een logopedist die de ICF benadering gebruikt, verzamelt ook informatie over contextuele factoren (omgevings- en persoonlijke factoren), bijvoorbeeld in de anamnese (McLeod & Bleile, 2004). Na de diagnostische fase kiest de logopedist een interventie die past bij de differentiaaldiagnose, de therapeutische en wetenschappelijke perspectieven, en elementen van de achtergrond van het kind, zoals leeftijd, gezinsomstandigheden, en medewerking van de ouders (Baker & McLeod, 2011; Dodd & Bradford, 2000). Kortom, logopedisten worden gedwongen om keuzes te maken gedurende het hele proces van het begeleiden van kinderen met spraakstoornissen en hun ouders/verzorgers, maar er is weinig bekend over deze keuzes. Daarom hebben we een onderzoek uitgevoerd om inzicht te krijgen in dit proces in Nederland.

In deze studie wordt gebruik gemaakt van een gelijktijdig triangulatie-ontwerp (gebruik van meerdere methoden). 137 logopedisten vulden een online enquête in voor de kwantitatieve studie, en 33 logopedisten werden geïnterviewd door logopediestudenten voor de kwalitatieve studie. De logopedisten werden gevonden via sociale media of rechtstreeks benaderd door de studenten.

Uit de resultaten van de vragenlijst en de interviews komt naar voren dat logopedisten twee tot vier sessies nodig hebben om het diagnostisch proces te voltooien. De meeste logopedisten gaven aan dat zij een anamnese afnemen met de ouders/verzorgers van het kind waarin zij vragen stellen die 19 onderwerpen bevatten. De meeste logopedisten vragen naar het gehoor, de spraaktaalontwikkeling van het kind, het verloop van de stoornis en gedragsaspecten (zoals de reactie van het kind wanneer het verkeerd begrepen wordt).

Vervolgens voeren logopedisten een onderzoek uit dat ook een observatie van verschillende vaardigheden bevat, zoals de communicatiestrategie van het kind, mondelinge vaardigheden, spraakkenmerken, en/of interacties met de ouder/verzorger. De logopedisten in dit onderzoek gebruiken vaak het Nederlands Articulatie Onderzoek (NAO) (Baarda et al., 2014), het Metaphon onderzoek (Leijdekker-Brinkman, 2005), en de Hodson Assessment of Phonological Patterns (HAPP, Hodson, 2004). Het NAO wordt vooral gebruikt wanneer een logopedist een fonetische stoornis vermoedt, terwijl Metaphon en HAPP vooral worden gebruikt bij een vermoeden van een fonologische stoornis. Een kleine groep logopedisten gebruikt ook het Dyspraxieprogramma (Erlings-van Deurse et al., 1993), dat gebaseerd is op The Nuffield Dyspraxia Programme, maar zij gebruiken niet het gehele onderzoek vanwege de lengte en ook gebruiken zij het onderzoek niet vaak.

Na het verwerken van de onderzoeksresultaten in combinatie met de gegevens uit de anamnese stelt de logopedist een diagnose. Uit onze studie blijkt dat logopedisten 85 verschillende labels gebruiken. Deze labels konden in vijf categorieën worden ondergebracht. De meest genoemde van de vijf labels was een fonologische stoornis, gevolgd door een fonetische stoornis, spraakontwikkelingsdyspraxie (SOD), dysartrie, en vloeiendheidsstoornissen (zoals stotteren).

Na de diagnose bepaalt de logopedist welke interventie op basis van het diagnostische label in combinatie met de leeftijd en het gedrag van het kind en de eigen ervaring van de logopedist passend zal zijn. De meeste genoemde interventies worden gebruikt bij alle diagnostische labels. In het geval van een fonologische stoornis gebruiken logopedisten vaak de cyclusbenadering (HAPP, Hodson & Paden, 1983) en Metaphon (Leijdekker-Brinkman, 2005). Bij kinderen met de diagnose fonetische stoornis wordt vaker gekozen voor Logo-art en een orale-myofunctionele therapie. Logopedisten kiezen voor het Dyspraxieprogramma bij kinderen met de diagnose SOD.

De structuur of aanpak van de interventies verschilt ook voor deze drie diagnoses (dysartrie en vloeiendheidsstoornissen zijn niet opgenomen omdat weinig logopedisten deze noemden). Verschillen zijn onder andere het gebruik van doelwoorden of specifieke spraakklanken of het oefenen van wisselende klanken alvorens echte woorden te oefenen. Logopedisten gebruiken ook verschillende didactische strategieën, zoals imitatie en bewustwording van de spraakfouten versus drilling en motorisch leren. Logopedisten meldden dat zij zich richten op het ondersteunen van het kind bij het toepassen van de geleerde strategieën in het dagelijks leven, hetgeen volgens hen niet altijd in het interventieprogramma is opgenomen. De logopedisten vermeldden dat de betrokkenheid van ouders/verzorgers tijdens de interventieperiode en de mate waarin het kind thuis oefent factoren zijn die de duur van de interventieperiode kunnen verkorten.

Bovendien is de interventieperiode naar verluidt korter en efficiënter als aan het begin van de sessies SMART-doelen (specifiek, meetbaar, haalbaar, realistisch en tijd) worden vastgesteld en de doelen zo nodig worden bijgesteld. Volgens de logopedisten is het belangrijk om de doelen samen met de ouders/verzorgers te formuleren. Als de vooruitgang stagneert, zorgt de logopedist voor een therapiepauze voor het kind. Voor de toekomst hopen de logopedisten dat er een methode wordt ontwikkeld die bij alle kinderen bruikbaar is en die de kinderen motiveert en stimuleert. Daarnaast zou het ideaal zijn als het materiaal via een computer of tablet wordt gepresenteerd/gegeven, aldus de geïnterviewde logopedisten.

In het algemeen kan er geconcludeerd worden dat er geen consensus bestaat onder logopedisten in Nederland over de terminologie van de verschillende spraakstoornissen. Uit de resultaten blijkt ook aan dat de meeste logopedisten een interventie kiezen op basis van beschikbaarheid en hun eigen ervaring, in plaats van op wetenschappelijk bewijs voor de interventie.

Diadochokinesetaak

Om logopedisten te helpen onderscheid te maken tussen onderliggende spraakprocessen, is er een spraakonderzoek ontwikkeld: het Computer Articulatie Instrument (CAI). Eerdere studies hebben het gebruik van het CAI bij kinderen (normgroep) onderzocht en vastgesteld dat het een valide en betrouwbaar instrument is (van Haaften et al., 2020; Maassen et al., 2019; van Haaften et al., 2019). Deze thesis richt zich op de ontwikkeling en validatie van de Diadochokinesetaak en de bijdrage daarvan aan een profielbeschrijving voor kinderen met een spraakprobleem; dit is beschreven in hoofdstuk 3 en 4.

Hoofdstuk 3 beschrijft de totstandkoming van het protocol voor de Diadochokinesetaak van het CAI. Deze taak wordt vaak gebruikt om kinderen met een onderliggende spraakmotorische achterstand te onderscheiden van kinderen met problemen op basis van een (nog) niet goed ontwikkeld fonologisch systeem (Icht & Ben-David, 2014; Rvachew & Brosseau-Lapré, 2012; Wang et al., 2008; Yang et al., 2011). Er is echter geen uniforme methode voor het toedienen en analyseren van de Diadochokinesetaak, wat het moeilijk maakt om de resultaten van verschillende studies wereldwijd te vergelijken (Gadesmann & Miller, 2008). Voor deze studie hebben we een protocol ontwikkeld op basis van bestaand onderzoek (Rvachew & Brosseau-Lapré, 2012; Thoonen et al., 1996; Wit et al., 1993). In dit protocol bestond de Diadochokinesetaak uit zes reeksen: /papa../, /tata../, en /kaka../, pata../ en /taka../, /pataka../ (Maassen et al., 2019). We gebruikten de volgende methode om de maximale prestatie (Maximum Repetition Rate, MRR) te verkrijgen: totaal aantal lettergrepen gedeeld door de totale duur van de reeks (telling per tijd).

1,524 Nederlandstalige kinderen in de leeftijd van 2;0 tot 6;11 jaar deden mee na toestemming van ouders. De steekproef was representatief voor geslacht, urbanisatie, en geografische regio. 14 Logopedisten hebben de jongere kinderen (2 tot 4 jaar) onderzocht en 110 studenten van verschillende logopedieopleidingen in Nederland hebben het CAI afgenomen bij de oudere kinderen (4 tot 7 jaar). Al deze onderzoeksassistenten werden getraind in het afnemen van het CAI. Doordat de computer de instructie geeft, is de afname gestandaardiseerd. De kinderen werd gevraagd de volgende reeksen te imiteren: drie monosyllabische reeksen (/papa../, /tata../ en /kaka../), één trisyllabische reeks (/pataka../), en twee bisyllabische reeksen (/pata../ en /taka../). Voor elke reeks kregen de kinderen enkele oefenreeksen in de volgende volgorde: 1) een korte reeks van drie lettergrepen in een normaal spreektempo; 2) een langere reeks van zes lettergrepen in een normaal spreektempo; en 3) een reeks van meerdere lettergrepen in een sneller spreektempo (het audio-voorbeeld bevatte 12 lettergrepen in een sneller spreektempo). Na deze oefeningen werd er aan het kind gevraagd (instructie via de computer) om de reeks zo snel mogelijk te produceren; er werd geen voorbeeld van de reeks gegeven. De kinderen kregen maximaal drie pogingen voor elke reeks.

Het akoestische signaal (de spraak van het kind) werd automatisch in één opname opgeslagen op de harde schijf van de computer. Deze opnames werden geanalyseerd volgens het protocol. Een reeks mocht bijvoorbeeld geen pauze of een spraakfout bevatten. Voor elk van de zes correct uitgesproken reeksen werd de MRR berekend voor de reeks met de snelle snelheid en de reeks met de snelste snelheid. In het onderzoek is eerst gekeken naar hoeveel kinderen de verschillende reeksen van de MRR produceerden en is er een vergelijking gemaakt tussen het voltooien van de verschillende reeksen (bv. monosyllabische sequenties versus multisyllabische sequenties). Repeated measures ANOVAs werden uitgevoerd om de beste prestaties op de snelle (met voorbeeld) en snelste (zonder voorbeeld) poging per reeks te vergelijken. Daarnaast is gekeken naar de verschillen tussen de snelheid van de reeksen als er tussen de drie of tien lettergrepen waren meegenomen in de verschillende reeksen.

Uit deze studie konden vier conclusies worden getrokken, die in het uiteindelijke CAI-protocol werden opgenomen. Ten eerste, kinderen onder de drie jaar hebben grote moeite met het uitvoeren van deze taak; veel kinderen konden geen reeks met drie of meer lettergrepen produceren. Kinderen maakten meer fouten in de trisyllabische reeksen, /pataka../, dan in de andere reeksen. Bovendien produceerden slechts 21% van de jongere kinderen (leeftijd <3;0) alle drie de monosyllabische reeksen. Daarom werd er besloten deze jongere groep kinderen uit te sluiten van de normgegevens voor het CAI. Deze taak kan een logopedist nu niet meer afnemen in het CAI. De kinderen (leeftijd 3;0 tot 6;11) werden opgenomen in de verdere analyse in deze studie om te bepalen of zij alle reeksen konden uitvoeren, en welke poging per reeks het snelst was.

Ten tweede stelden we vast dat niet alle kinderen alle reeksen konden uitvoeren, en dat de bi- en trisyllabische reeksen moeilijk waren. Daarom is in het nieuwe CAI-protocol opgenomen dat de bi- en trisyllabische reeksen niet worden afgenomen als kinderen de monosyllabische reeksen niet kunnen produceren, om de last van een afname van de hele testbatterij te verlichten.

Ten derde, bij het bepalen welke poging het snelst was, vonden we dat kinderen sneller waren wanneer ze de instructie "zo snel mogelijk" kregen dan wanneer hen gevraagd werd het voorbeeld met een snellere spreeksnelheid te imiteren. Dit resultaat had echter slechts een kleine effectgrootte, zodat het met de nodige voorzichtigheid moet worden geïnterpreteerd. In het CAI-protocol voor deze taak is nu opgenomen dat de logopedist moet bepalen welke poging van de laatste twee instructies het snelst is en die poging moet selecteren voor vergelijking met de normgegevens.

De laatste (vierde) conclusie uit deze studie is dat het niet nodig is om een reeks van 10 tot 12 lettergrepen door de kinderen te laten uiten, zoals in eerdere onderzoeken is uitgevoerd als procedure (Rvachew & Brosseau-Lapré, 2012; Thoonen et al., 1996; Wit et al., 1993; Yaruss & Logan, 2002). Veel kinderen in de normgroep produceerde reeksen van drie tot ongeveer tien lettergrepen (exclusief de eerste en laatste lettergreep van de reeks). In het onderzoek zijn geen verschillen in snelheid (MRR) tussen kortere en langere reeksen gevonden (behalve voor de reeks /pa../ met een kleine effectgrootte). Daarom is in het protocol van het CAI nu opgenomen dat kinderen ten minste vijf lettergrepen per reeks moeten produceren om de snelheid van de reeks te laten berekenen in het CAI programma.

Hoofdstuk 4 beschrijft een groot cross-sectioneel onderzoek naar normgegevens voor de Diadochokinesetaak van een grote groep normaal ontwikkelende Nederlandse kinderen in de leeftijd van 3;0 tot 6;11. Het protocol uit hoofdstuk 3 werd gebruikt om de gegevens van de 1.041 kinderen te analyseren. De spreeksnelheid van de kinderen bij de verschillende reeksen nam significant toe met de leeftijd. De reeks /pa../ was de snelste van alle reeksen, gevolgd door /ta../, /pata../, /taka../, /ka../, en /pataka../. Dit resultaat is in overeenstemming met eerdere studies in het Nederlands en in andere talen. Een andere conclusie van deze studie was dat jongens de verschillende reeksen sneller uitspreken dan meisjes. De uitkomsten van deze studie helpen een logopedist bij het bepalen of een kind een normale ontwikkeling heeft op deze taak of niet. De vraag is of met deze normgegevens ook een kind met een vermoeden van een motorische spraakstoornis onderscheiden kan worden van een kind met spraakproblemen op basis van een onrijp fonologisch systeem of een verkeerde opslag van woorden in het fonologisch systeem.

Klinische validatie

Hoofdstuk 5 beschrijft twee studies die gebruikt zijn om de klinische waarde van het CAI in de diagnostische fase vast te stellen. Twee groepen kinderen met spraak-taal stoornissen namen deel aan de twee studies. De opzet van de twee studies was hetzelfde. De kinderen werden onderzocht met het CAI door een logopedist. De eerste studie omvatte 93 kinderen in de leeftijd van 3;0 en 4;0 jaar. Deze kinderen waren gediagnosticeerd met een spraakstoornis en namen deel aan een vroegbehandelingsgroep voor jonge kinderen met een spraak-taalontwikkelingsstoornis. Het percentage correcte consonanten initiaal (PCCI) op de Plaatjesbenoemtaak van het CAI werd vergeleken met het oordeel van de logopedist over de verstaanbaarheid (goed, matig, slechte) van het kind. Kinderen met een slecht verstaanbaarheidsoordeel hadden lagere PCCI-scores op de Plaatjesbenoemtaak dan kinderen met een matige verstaanbaarheid. Kinderen met een goede verstaanbaarheid scoorden het hoogst op de PCCI. Er werd geen correlatie gevonden tussen de non-verbale intelligentie en de taalscores met de prestatie op de Plaatjesbenoemtaak en het oordeel van de verstaanbaarheid.

De tweede studie beschreven in hoofdstuk 5 betrof een groep van 41 kinderen met een spraakstoornis in de leeftijd van 3;0 tot 6;4. De kinderen waren gediagnosticeerd met een fonologische stoornis (n = 36), spraakontwikkelingsdyspraxie (n = 2), of een onbekende stoornis (n = 3). Het doel van deze studie was om de relatie te bepalen tussen de ernst van de spraakstoornis beoordeelt door de logopedist en de vijf CAI-componenten (van Haaften et al., 2019). Voor de CAI-componenten gebaseerd op de Plaatjesbenoemtaak, de Nonwoorden imitatietaak, en de bisyllabische en trisyllabische reeksen van de Diadochokinesetaak, ontdekten we een significant verschil in prestaties tussen de ernstscore die de kinderen hadden gekregen. De groep met het label 'ernstige spraakstoornis' scoorde slechter op deze componenten dan kinderen met het label 'matige spraakstoornis'. Voor de component Woordrepetitietaak en Nonwoordrepetitietaak en de monosyllabische reeksen van de Diadochokinesetaak, werden er geen significante verschillen gevonden tussen de kinderen met een matige of een ernstige spraakstoornis. Dit impliceert dat de resultaten op de Plaatjesbenoemtaak, de Nonwoorden imitatietaak, en de bisyllabische en trisyllabische reeksen van de Diadochokinesetaak een logopedist kunnen helpen bij het diagnosticeren van een kind met een spraakstoornis.

De laatste studie (hoofdstuk 6) beschrijft een grotere groep kinderen met spraakstoornissen (n = 150). Een deel van de onderzoekspopulatie uit hoofdstuk 5 is ook in deze studie gebruikt. De steekproef bestond uit 94 jongens en 56 meisjes in de leeftijd van 4;0 tot 6;6 jaar. De meesten van hen waren gediagnosticeerd met een fonologische stoornis (n = 105); de anderen kinderen waren gediagnosticeerd met een spraakontwikkelingsstoornis (n = 17), een fonetische articulatiestoornis (n = 9), dysartrie (n = 5), of een stoornis niet nader gespecificeerd door de logopedist (n = 2). Sommige kinderen kregen meerdere diagnoses. 11 kinderen werden gevonden via reguliere scholen en kregen geen logopedie op het moment van de studie, maar ze scoorden onder percentiel 16 op het CAI en werden dus opgenomen in deze studie. Het doel van deze studie was om te onderzoeken of het CAI gebruikt kan worden om profielen te differentiëren en te identificeren.

In deze groep kinderen met een spraakstoornis werden er iets andere componenten gevonden dan in de eerdere analyse van de normgroep (van Haaften et al., 2019). Er kwamen slechts drie componenten naar voren: 1) alle parameters van de Plaatjesbenoemtaak plus de volgende parameters van de Nonwoordimitatietaak: Level5, normale spraakprocessen, en de afwijkende spraakprocessen; 2) de overige parameters van de Nonwoordimitatietaak inclusief de Woord repetitietaak en de Nonwoordrepetitietaak; 3) de parameters van de Diadochokinesetaak. Terwijl er in de normgroep vijf componenten naar voren kwamen. Het feit dat Plaatjebenoemtaak en

Nonwoordimitatietaak parameters grotendeels op verschillende componenten laden bij de kinderen met een spraakstoornis, impliceert dat het belangrijk is om beide taken af te nemen en te beoordelen om een breed beeld te krijgen van het gehele spraakproductieproces. Bijvoorbeeld: kinderen die relatief weinig spraakfouten maken bij het imiteren van nonsenswoorden kunnen relatief weinig moeite hebben met het uitspreken van nieuwe woorden die ze aan het leren zijn, wat een aanknopingspunt kan zijn voor de keuze van de behandelmethode.

Na de principiële componenten analyse is er een clusteranalyse met een K-means analyse gedaan. Hieruit blijkt dat de kinderen in drie groepen konden worden verdeeld. Cluster I was de grootste groep en had de hoogste scores op alle CAI parameters (taken). Kinderen scoorden lager dan de normgroep op de Plaatjes benoemtaak (die de hele keten van spraakprocessen gebruikt) en de Nonwoordimitatietaak. Er was geen verschil in scores ten opzichte van de normgroep op de Diadochokinesetaak; deze taak kan een oordeel geven over de motorische planning en programmering van de spraak. De kinderen in cluster I heeft dus vooral problemen met het fonologische systeem en deze groep werd bestempeld als een groep met een 'fonologische achterstand'. De kinderen in cluster II hadden vergelijkbare scores als de kinderen in cluster I, maar scoorden significant lager op een paar CAI parameters (syllabische structuur parameters van de Plaatjesbenoemtaak en de Nonwoordimitatietaak en de parameters van de Diadochokinesetaak). Deze kinderen lijken meer moeite te hebben met de motorische planning en programmering van de spraak, dus werd het label 'milde fonologische achterstand met een motorische achterstand' gegeven aan de kinderen in cluster II. De kinderen in cluster III scoorden laag op alle CAI-parameters en werd daarom gelabeld met 'ernstige fonologische en motorische achterstand'.

Wanneer de drie clusters werden vergeleken met andere variabelen, vonden we dat de clusters verschillen op verstaanbaarheid (Intelligibility in Context Scale (ICS); McLeod et al, 2013), het woordbegrip (Peabody Picture Vocabulary Test-III-NL (PPVT-III-NL); Schlichting, 2005), de auditieve discriminatie (Testinstrumentarium Taalontwikkelingsstoornissen (TTOS-ADT); Verhoeven, et al., 2013), en het oordeel op de ernst van de spraakstoornis door de logopedist en de ouders/verzorgers. De kinderen uit cluster I scoorden het hoogst op al deze variabelen, gevolgd door de kinderen uit cluster II en vervolgens de kinderen uit cluster III. We vonden geen significante verschillen tussen de clusters voor leeftijd, geslacht, of de diagnose gegeven door de logopedist. Drie conclusies konden worden getrokken uit dit onderzoek: 1) er zijn verschillende profielen te ontdekken in een groep kinderen met een spraakstoornis; 2) de ernst van de spraakstoornis speelt een rol; en 3) er is een spectrum van mate van betrokkenheid van verschillende onderliggende problemen zichtbaar bij kinderen met een spraakstoornis.

Dit proefschrift beschrijft het CAI als een instrument voor het diagnosticeren van kinderen met spraakstoornissen en het bepalen van de ernst van de spraakstoornis. Daarnaast is aangetoond dat het CAI behulpzaam is bij het identificeren van kinderen die problemen hebben met onderliggende spraakprocessen. Om de onderliggende problemen met de spraakprocessen verder te onderzoeken, zou een directe manipulatie van de spraakprocessen een aanvulling kunnen zijn op het CAI, zoals het aanbieden van achtergrond ruis tijdens bepaalde taken uit het CAI. Een ander instrument dat in het CAI kan worden geïmplementeerd is de (Genormaliseerde) Pairwise Variability Index.

Een tweede doel voor de toekomst is het ontwikkelen van nieuwe richtlijnen voor procesgerichte diagnostiek en het bepalen van de inhoud van het behandelplan. Baker en McLeod (2011) publiceerden een review van interventiestudies bij kinderen met spraakstoornissen gepubliceerd tussen 1979-2009. Zij identificeerden 134 studies, waarvan 46 studies verschillende interventiebenaderingen beschreven en 24 geen specifieke procedures vermeldden. Deze studies bevatten ook verschillende niveaus van wetenschappelijk bewijs. Dat kan de reden zijn waarom logopedisten kiezen voor een eclectische aanpak en perceptuele, linguïstische en mondmotorische activiteiten combineren in de behandelsessies (Diepeveen et al., 2020; Lundeborg Hammarström et al., 2019).

Het uiteindelijke doel voor de toekomst is dan ook om de logopedische zorg voor kinderen met spraakstoornissen te verbeteren door het ontwikkelen van een evidence-based, procesgeoriënteerde digitale klinische routekaart voor diagnose en het plannen en monitoren van de behandeling, in de vorm van een eenvoudig te gebruiken app.

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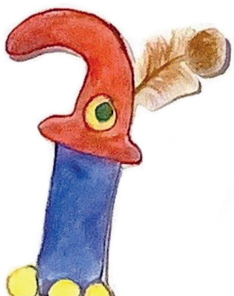
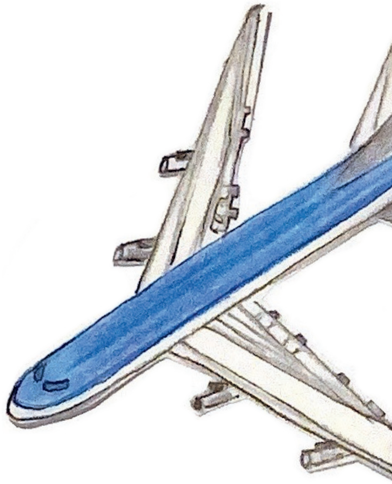
APPENDICES





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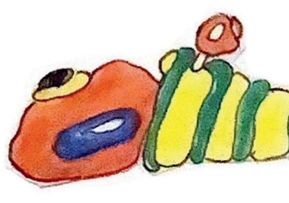

Dankwoord

En dan zit je promotietraject erop. Een aantal keer heb ik al de vraag gekregen: 'Wat ga je nu doen?' Het antwoord daarop is steevast, dat ik geen vooropgesteld plan heb. Zo ben ik ook in dit promotietraject gerold. Van wat een kortdurend project zou zijn om data te verzamelen voor de normering van het Computer Articulatie Instrument, ontwikkelde zich naar een boekje met artikelen waarvan ik hoop dat ze een bijdragen leveren aan het logopedische werkveld. Maar ik heb dit niet alleen gedaan en ik wil een aantal mensen bedanken voor hun suggesties, ondersteuning, inspiratie en ook vooral ontspanning en gezelligheid!

Allereerst ben ik dankbaar voor alle kinderen en hun ouders die meededen aan dit onderzoek. Maar ook de kinderen en hun ouders die ik in mijn loopbaan mocht begeleiden en die mij inspireerde. En ook de studenten inspireerde mij. De vragen die zij stelden over een specifieke cliënt, brachten mij aan het denken en beïnvloedde de manier waarop dit promotietraject gevormd is. Een speciale dank voor de studenten en ook logopedisten die hebben bijdrage aan de dataverzameling.

Speciale dank gaat uit naar mijn promotieteam:
Ben, ooit volgde ik een college van jou en inspireerde je mij. Dank voor je ondersteuning de afgelopen jaren. Ik heb veel van je geleerd! Je liet mij vrij in het uitvoeren van dit onderzoek. Je liet mij zien hoe je goed onderzoek uitvoert en hoe je dit op een wetenschappelijke wijze op papier zet.
Bert, zonder jou was ik niet gestart met dit promotietraject. Wat startte met een grapje in een vergadering, werd door jou serieus genomen. Ik kon altijd bij je aankloppen voor advies. Ook dank dat je mij de mogelijkheid gaf om in de koppelstructuur deel te nemen.
Hayo, soms op afstand, maar altijd goed bereikbaar! Dank voor al jouw adviezen, niet alleen tekstueel, maar ook rondom aanpak van stappen in dit traject. Dank voor de discussies over het diagnosticeren en behandelen van deze kinderen.

Bij het promotieteam hoor jij, Leenke, eigenlijk ook nog. Wat hebben we samen een hoop werk verricht. Een echt team, zoals jij al schreef; zo voelt het voor mij ook. Dank voor de gesprekken die we voerden over het project, ons werk en over van alles en nog wat.

Ook dank aan de medeauteurs van mijn artikelen: Bernadette, Charlotte, Anne Marie, dank voor het verzamelen van data, maar ook bedankt voor alle suggesties. Dan, Lenie, ook al was je co-promotor van Leenke en niet van mij, toch hoor je zeker ook een beetje bij mijn promotieteam. Je gaf waardevolle suggesties. Dank voor alles!

Lieve collega's van de HAN, heel erg bedankt voor jullie steun en bijdrage. Allereerst Theo, Menno en Bert voor de steun in de aanvraag voor een promotietraject. Menno, dank dat je meeleefde in alle stappen die ik heb genomen. Dank ook aan het College van Bestuur van de HAN voor het mogelijk maken van een HAN beurs.
En natuurlijk ook mijn collega's van de opleiding logopedie, bedankt! Dank voor de inhoudelijke gesprekken, gesprekken over proces, gesprekken over andere onderwerpen en vooral ook dank voor jullie vragen tussendoor om te checken hoe het met mij ging. Daarnaast ook dank voor alle collega's die niet meer werken bij de afdeling logopedie. Zonder jullie ondersteuning was ik niet zover gekomen.
Anita, wat fijn dat we elkaar nog steeds zien, ook al wonen we wat verder van elkaar af. Dank voor al jouw gesprekken en ontspannende momenten. Volgend jaar weer naar London?
Pamela, ook jou ben ik dankbaar voor alle gesprekken en je interesse in mijn bevindingen.
Emmy, nog niet zo lang geen collega's meer van elkaar, maar dank voor de spellenavondjes.
Mariëlle, we hebben elkaar gevonden in het doorlopen van een promotietraject. Dank voor de lunchgesprekken. Veel succes met jouw laatste loodjes.

Dan, collega's van lectoraat Neurorevalidatie, heel erg bedankt voor jullie hulp, adviezen, jullie vragen en interesse in wat ik allemaal aan het doen was.
Collega's van iXperium Health, ik geniet en leer van jullie passie voor (zorg)technologie, onze interprofessionele samenwerking, het samen ontdekken en het inspireren van collega's en studenten om meer (zorg)technologie te gebruiken. We hebben nog genoeg te doen!

Beste oud-collega's van het RadboudUMC, ook jullie bedankt voor het bijdragen aan mijn ontwikkeling binnen dit promotietraject. Ik hoop dat we elkaar in de toekomst blijven treffen.

Lieve (oud)collega's van Krebbers Logopedie en Dyslexie: Marjo, Ted, Vera, Marloes, Maaike, Karolijn, Daphne, Rosanne, Tanja en Susan. Dank voor jullie steun de afgelopen jaren en interesse in mijn project. Ook dank voor de fijne gesprekken over ons vakgebied en de hulp bij een 'ingewikkelde' casus. Ik leer veel van jullie.

En dan, lieve familie en vrienden, ook jullie waren steeds geïnteresseerd in mijn vorderingen. Dank voor alle ontspannen middagen en avonden met spelletjes, BBQ/kampvuur, roadtrips en andere activiteiten.

Lieve Petra, dank voor jouw vriendschap! Al sinds de start van de opleiding logopedie trokken we met elkaar op en ik ben blij dat we elkaar nog steeds zien met of zonder Ronnie, Fenna en Niek.

Lieve (al heel lang, ga niet zeggen hoelang) vriendinnen, Helen, Aline, Elles en Elly. Wat hebben we veel meegemaakt. Ieder heeft op haar eigen wijze een bijdrage gehad. Helen, dank dat we al zo lang elkaar steunen en samen plezier hebben. Ik ben blij dat je mijn paranimf wilde zijn. Aline, dank voor de ontspannen logeerpartijen bij jou en het soms meedenken. Elles, dank voor je gesprekken, maar vooral ook bedankt voor je nuchtere blik op momenten dat ik even vastliep. Elly, dank voor je gezelligheid en je kijk op het werken met mensen. En niet te vergeten je tekentalent waardoor ik nu een hele mooie persoonlijke omslag heb voor dit proefschrift.
Dank ook aan jullie gezinnen voor onze gezellige wandel-, zwem-, speel-, spel-, feest-, eet/drinkmomenten. Dank, Wietse, Silke, Femke, Stephan, Jan, Bart, Dirk, Tim, Rick, Jeannôt, Niek en Eef. En Silke, bedankt dat ik een foto en een video-opname van ons samen mocht gebruiken.

Lieve Wil en Rinie, ik wil jullie ook graag bedanken voor de steun en interesse die jullie hebben getoond. Ook dank voor al het lekkere eten en de gezellige spelmomenten.

Lieve pap en mam, dank voor alle ondersteuning en interesse in wat ik doe. Ik kan altijd bij jullie aankloppen voor een gesprek, hulp of wat dan ook. Dank voor alles wat jullie doen.

Lieve Mayke en Leon, lief zusje, dank dat je mijn paranimf wilde zijn. Ook dank voor jouw ondersteuning de afgelopen jaren en de interesse in wat ik nu allemaal aan het doen was. Leon, bedankt voor de gezellige momenten. Dat we nog vele fijne momenten met elkaar mogen hebben.
Lieve Bob, Ruth en Anoek, lief broertje, dank dat je zo nu dan een stuk tekst wilde corrigeren en meedacht over van alles en nog wat. Ruth, also thanks for all your support and the nice moments we have together. Anoek, heerlijk om met jou te spelen on- en offline. Ik hoop dat ik nog vaak bij jullie op zoek kan komen in Engeland of jullie in Nederland.

Lieve Martijn, mijn dank is groot voor alles wat je voor me doet. Je haalt mij uit mijn piekerstand met een of andere nuchtere opmerking. Je hebt mij ook veel tijd bespaard door delen van de dataverwerking te automatiseren. Dank ook voor onze gezellige autoritjes. Ik kijk al uit naar de volgende.

Curriculum Vitae

Sanne Diepeveen was born in Boxmeer, the Netherlands, on March 13th, 1978. She grew up in Nieuw Bergen. After her graduation from secondary school (Scholengemeenschap Stevensbeek in Stevensbeek) in 1996, she started her bachelor's degree in Speech and Language Therapy (SLT) at the HAN University of Applied Sciences in Nijmegen. She began working as a speech and language therapist (SLT) in a private practice in Tiel after her graduation. After eighteen months, she decided to start a second study, Child psychology (Pedagogische Wetenschappen, orthopedagogiek) at Radboud University in Nijmegen. She then combined her study with a position as an SLT at a school for special education (SBO De Vlinder in Bemmel), and various other temporary SLT positions at Stichting De Waarden and Maashorst.

After graduation, she started working as a lecturer at the speech and language therapy department at the HAN University of Applied Sciences (HAN) in Nijmegen combined with a position at the Wijchen municipality (speech-language prevention at pre-school and primary schools) and a temporary position at Kentalis, special education in Oss. She then transferred to the Department of Pediatric Neurology at the Radboud University Medical Center (RadboudUMC). After this position, she applied to a private practice, Krebbers Logopedie en Dyslexie, where she still works as an SLT and child psychologist.

The start of her PhD project began with a position in the HAN's Neurorehabilitation research group on the development of the Computer Articulation Instrument (CAI). As the project progressed, it was possible to apply for a grant to start a PhD project. This was combined with Leenke van Haaften's PhD project; Leenke and Sanne worked on this together with Ben Maassen, Bert de Swart, Hayo Terband and Lenie van den Engel-Hoek. The CAI was launched by BOOM Publishing in 2019.

During this period, Bert de Swart also initiated a project that allowed lecturers from HAN's Academy of Paramedical Studies to gain work experience at the RadboudUMC. Sanne was then placed at the department of Reabilitation of the RadboudUMC, Amalia's Children's Hospital in Nijmegen, where she had previously worked. In addition, SLTs colleagues from that department were assigned to teach at the HAN's SLT department. After a several years, this collaboration stopped and Sanne became a member of HAN's iXperium Health. This consortium aims to introduce students to (healthcare) technology. This suits Sanne well because she herself tries out the latest technologies in her clinical work. Moreover, the CAI is also a new technological tool that can be used in SLT practice.

Data management form

General information about the data collection

This research project involves human subject data. Participants volunteered to participate, and anonymity and confidentiality were assured. Written informed consent for collecting these data was obtained from all parents or legal representatives of the participants. The research ethics committee of the Radboud university medical center, Nijmegen stated that this research project (Chapter 2-6) does not fall within the remit of the Medical Research Involving Human Subjects Act (WMO). Therefore, the studies could be carried out (in the Netherlands) without an approval by an accredited research ethics committee. Data were collected and stored at the Radboud university medical center and the HAN University of Applied Sciences.

FAIR principles

Findable

The raw and processed data and accompanying files (descriptive files, syntax files, recordings of interviews etc.) of this research project are stored in a folder on the server of the department of Rehabilitation at Radboud university medical center (Q:\Research\041 CAI) and the HAN secure network drive ('R disk'). These folders are only accessible by the main researchers of this project. Documentation to describe the datasets is provided on the department server. The privacy of the participants is warranted by use of encrypted and unique individual subject codes. The paper data of the studies are stored in the research group's archive (HAN Lectoraat Neurorevalidatie en Innovatie). All data will be stored for at least 10 years after publication.

Accessible

Only members of the research group have access to the databases. It is not yet possible to make the data available in a public repository because participants only gave informed consent to use their data for purposes as explained on the signed informed consent form. However, requests for data can be made by contacting the staff secretary of the Department of Rehabilitation of the Radboud university medical center (secretariaatstaf.reval@radboudumc.nl). A suitable way to share the data will then be sought. In the future it will be explored how our data can be published in a public repository.

Interpretable

Documentation was added to the data sets to make them interpretable. The documentation contains links to publications, references to the location of the data sets and descriptions of the data sets. The data was stored in SPSS format. No existing data standards were used such as vocabularies, ontologies, or thesauri.

The individual interviews described in **Chapter 2** were recorded. The recordings were transcribed in MSWord and analysed using Atlas-ti. The anonymized data in MSWord and Atlas-ti format are stored on the HAN secure network drive ('R disk'). Documentation was added to the Atlas-ti data set to make it interpretable.

Reusable

The data can be reused within the period of 10 years by the principal researchers for purposes described in the signed informed consent form.

PhD Portfolio

Name PhD student	S.J.H. Diepeveen
Department	Radboud university medical center, Department of Rehabilitation Donders Graduate School for Cognitive Neuroscience
Promotor	Prof. Dr. B.A.M. Maassen
Co-promotors	Dr. B.J.M. de Swart Dr. H.R. Terband

Activities	**Year**	**ECTS**
a) Courses & Workshops		
Schrijven van wetenschappelijke teksten, Radboud University Medical Center	2013	3.0
Academic writing, Radboud University Medical Center	2014	3.0
Graduate School Introduction day, Donders Institute	2016	0.3
Basiscursus regelgeving & organisatie voor klinisch onderzoekers (BROK), Nederlandse Federatie van UMC's	2016	1.5
Basiskwalificatie Examinering, CITO/HANacademy	2016	2.0
Graduate School day, Donders Institute	2018	0.3
Scientific Integrity course, Donders Institute	2018	0.3
How to prepare your poster presentation, Donders Institute	2018	0.3
Improving your statistical inferences, Coursera, Technische Universiteit Eindhoven	2018	1.0
Writing in the sciences, Coursera, Stanford University	2018	2.0
Graduate School day 2, Donders Institute	2019	0.4
Het onderzoekend handelen van studenten effectief begeleiden, HAN University of Applied Sciences	2019	0.3
Improving your statistical question, Coursera, Technische Universiteit Eindhoven	2019	1.0
b) Symposia, Congressess and Conferences		
Ehealth voor mensen met communicatieve beperkingen: wetenschap en praktijk, Nijmegen, oral presentation and organisation	2013	1.25
NVLF congres, oral presentation	2017	1.25
Symposium Diagnostiek van spraakstoornissen bij kinderen, RadboudUMC & HAN University of Applied Sciences, oral presentation and organisation	2017	1.25
7th Speech Motor Control congress, Groningen, poster presentation (2x)	2017	1.25
Symposium on Research Integrity	2020	0.1
Madonna Motor Speech Conference, poster presentation	2022	1.0
HAN Promovendicongres 2022	2022	0.1
Symposium HAN logopedie ism Koninklijke Kentalis, presentatie en organisatie	2019-2022	3.0
c) Other		
Member of the Neurorehabilitation Research Group, HAN University of Applied Sciences	2013-2022	3.0
Reviewing scientific publications	2021-2022	0.1
Teaching activities		
d) Lecturing		
Bachelor Speech- and Language Therapy, HAN University of Applied Sciences	2013-2022	6.0
Post HBO cursus Computer Articulatie Instrument, HAN University of Applied Sciences	2018-2022	2.0
e) Supervising bachelor and master thesis		
Bachelor Speech Language Therapy, HAN University of Applied Sciences	2013-2022	5.0
Bachelor and master Taal- en spraakpathologie, Radboud University	2016-2022	3.0
Total		**43,7**

Publication list

2022	Diepeveen, S., van Haaften, L., van der Zande, A., Megens-Huigh, C., Terband, H., de Swart, B., & Maassen, B., (2022). Process-oriented profiling of speech sound disorders. *Children*, *9*, 1502: 1-23
	Steultjens, E., Lindenschot, M., Diepeveen, S., Zajec, J., de Groot, I., Nijhuis-van der Sanden, R., Koene, S., Graff, M., (2022). Tailored interviewing to uncover the perspectives of children with multiple disabilities on daily activities: a qualitative analyses of interview methods and interviewer skills. *Australian Occupational Therapy Journal, 9*: 1-15
2021	Diepeveen, S.,/van Haaften, L., Terband, H., *van den Engel-Hoek, L.,* de Swart, B., & Maassen, B., (2021). Maximum Repetition Rate in a large cross-sectional sample of typically developing Dutch speaking children*. International Journal of Speech-Language Pathology, 23(5),* 508-518
2020	Diepeveen, S., van Haaften, L., Terband, H., de Swart, B., & Maassen, B., (2020). Clinical reasoning for Speech Sound Disorders: Diagnosis and intervention in Speech-Language Pathologists' daily practice. *American Journal of Speech-Language Pathology*, *28(8)*, 1529-1549.
	van Haaften, L., Diepeveen, S., van den Engel-Hoek, E., de Swart, B., & Maassen, B. (2020). Speech sound development in typically developing 2–7-year-old Dutch- speaking children: A normative cross-sectional study. *International Journal of Language & Communication Disorders, 55(6)*, 971-987.
2019	Diepeveen S, van Haaften L, Terband H, de Swart B, Maassen B. (2019). A Standardized Protocol for Maximum Repetition Rate Assessment in Children. *Folia Phoniatrica Et Logopaedica*, *71*(5-6):238-50.
	van Haaften L, Diepeveen S, van den Engel-Hoek L, Jonker M, de Swart B, Maassen B. (2019). The Psychometric Evaluation of a Speech Production Test Battery for Children: The Reliability and Validity of the Computer Articulation Instrument. *Journal of Speech, Language, and Hearing Research*, *62*(7):2141-2170.
	van Haaften L, Diepeveen S, Terband H, Vermeij B, van den Engel-Hoek L, de Swart B, Maassen B. (2019). Profiling Speech Sound Disorders for Clinical Validation of the Computer Articulation Instrument. *American Journal of Speech-Language Pathology, 28*(2S):844-856.
	Maassen, B., van Haaften, L., Diepeveen, S., Terband, H., van den Engel-Hoek, L., Veenker, T., & De Swart, B. (2019). *Computer Articulation Instrument*. Boom Uitgevers.
	Terband, H., Namasivayam, A., van Brenk, F., Diepeveen, S., Mailend, M.-L., Maas, E., van Lieshout, P., & 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.
2015	Daemen, L., Langens, L., de Beer, J. & Diepeveen, S. (2015). Verhalen oefenboek. Het prentenboek voor logopedie. *Nederlands tijdschrift voor Logopedie, 87,* 6 – 12.
	Orgassa, A. & Diepeveen, S. (2015). Special Issue: eHealth, gastredactie *Stem-, Spraak- en Taalpathologie*.
2012	Diepeveen, S. (2012). Samenvatting rapport: Autismespectrumstoornissen, een leven lang anders, *Logopedie & Foniatrie, 83,* 88 – 90
2010	Diepeveen, S. (2010). Autisme als contextblindheid, interview met Peter Vermeulen. *Logopedie & Foniatrie, 82,* 58 – 61.
2008	Diepeveen, S. (2008). Casus Max: een cyclische aanpak. *Logopedie & Foniatrie*, *80*, 324-328.
2007	Rodenburg D., Diepeveen, S. & de Moor, J. (2007). Begeleiding van kinderen en jongeren met autisme, van onderzoek naar praktijk. In Didden, R. & Huskens, B. *De weg naar passende arbeid voor jongeren met autismespectrumstoornissen.* Bohn Stafleu van Loghem

Donders Graduate School for Cognitive Neuroscience

For a successful research Institute, it is vital to train the next generation of young scientists. To achieve this goal, the Donders Institute for Brain, Cognition and Behaviour established the Donders Graduate School for Cognitive Neuroscience (DGCN), which was officially recognised as a national graduate school in 2009. The Graduate School covers training at both Master's and PhD level and provides an excellent educational context fully aligned with the research programme of the Donders Institute.

The school successfully attracts highly talented national and international students in biology, physics, psycholinguistics, psychology, behavioral science, medicine, and related disciplines. Selective admission and assessment centers guarantee the enrolment of the best and most motivated students.

The DGCN tracks the career of PhD graduates carefully. More than 50% of PhD alumni show a continuation in academia with postdoc positions at top institutes worldwide, e.g. Stanford University, University of Oxford, University of Cambridge, UCL London, MPI Leipzig, Hanyang University in South Korea, NTNU Norway, University of Illinois, North Western University, Northeastern University in Boston, ETH Zürich, University of Vienna etc. Positions outside academia spread among the following sectors: specialists in a medical environment, mainly in genetics, geriatrics, psychiatry, and neurology. Specialists in a psychological environment, e.g. as specialist in neuropsychology, psychological diagnostics or therapy. Positions in higher education as coordinators or lecturers. A smaller percentage enters business as research consultants, analysts or head of research and development. Fewer graduates stay in a research environment as lab coordinators, technical support, or policy advisors. Upcoming possibilities are positions in the IT sector and management position in pharmaceutical industry. In general, the PhDs graduates almost invariably continue with high-quality positions that play an important role in our knowledge economy.

For more information on the DGCN as well as past and upcoming defenses please visit: http://www.ru.nl/donders/graduate-school/phd/

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