Phonological abilitites of children with childhood apraxia of speech

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Sveučilište u Zagrebu

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DOCTORAL DISSERTATION

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Supervisors:

Draženka Blaži, PhD, Full professor

Prof. emer. Ben Maassen

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Doktorski studij "Poremećaji jezika, govora i slušanja"

Dora Knežević

FONOLOŠKE SPOSOBNOSTI DJECE S DJEČJOM GOVORNOM APRAKSIJOM

DOKTORSKI RAD

Mentori:

Prof. dr. sc. Draženka Blaži

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Zagreb, 2022

The supervisors:

Draženka Blaži, PhD, is a full professor with tenure at the Faculty of Education and Rehabilitation Sciences (ERF), University of Zagreb, Department of Speech and Language Pathology. Since the academic year 2000/2001, when she received the title of Assistant Professor, she has been teaching undergraduate and graduate courses in the Speech and Language Pathology programme (ERF), in the postgraduate specialist study programme "Early Intervention in Educational Rehabilitation" and in the PhD programme "Language, Speech and Hearing Impairments". She was Vice Dean for Academic Affairs (2 terms; 2007-2011) and Dean (2 terms) of the Faculty of Education and Rehabilitation Sciences at the University of Zagreb (2 terms; 2007-2011). She is president of the Croatian Logopedics Association (2000-2009; 2014-). She is a member of various associations and continuously collaborates with various ministries and institutions in the field of health, social affairs and education. Her scientific interests are speech and language disorders in children of younger chronological age and speech-language and communication development. She collaborated on 6 scientific projects as an associate and consultant. She was also a member and co-organizer of several international scientific committees of scientific and professional conferences and chairperson of the organizing committee for the 6th Croatian Speech-Language Pathology Congress with international participation. She published over 40 research and professional papers and several book chapters.

Ben Maassen, PhD, is Emeritus Professor of Speech-Language-Literacy Disorders & Clinical Neuropsychologist at the University of Groningen (Center for Language and Cognition), and the University Medical Center Groningen (Research School of Behavioural and Cognitive Neurosciences (BCN), and is affiliated to the Department of Rehabilitation, Radboud University Medical Center, Nijmegen. His main research areas are speech (neurogenic speech disorders, in particular childhood apraxia of speech; perception-production modelling in speech development and speech disorders; speech-related cognitive dysfunctions; Parkinson's disease) and reading (dyslexia and neurocognitive precursors, with focus on neurophysiological assessment and digital game-based learning). He is chair of the International Conference on Speech Motor Control (Nijmegen 2001, 2006; Groningen 2011, 2017, 2022); former member and chair of the Motor Speech Disorders Committee of the International Association for Logopedics and Phoniatrics (IALP); editor-in-chief of the Dutch journal of voice, speech, and language pathology (Tijdschrift voor Stem-, Spraak- en Taalpathologie), and member of the editorial board of several international journals. He has a background in cognitive neuropsychology and speech-language pathology. Previous affiliations: Max Planck Institute for Psycholinguistics, Nijmegen; Donders Institute for Cognition, Radboud University Nijmegen; Department of Child Neurology, Radboud University Medical Center, Nijmegen. He has published several books, over 100 scientific papers (h-index=31) with around 2300 citations by 1600 documents.

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Abstract

Introduction: Childhood apraxia of speech (CAS) is a childhood motor speech disorder characterized by poor planning and/or programming of speech sound sequences in which the precision and consistency of the movements underlying speech are impaired, in the absence of neuromuscular deficits (ASHA, 2007). Although CAS is primarily defined as a motor disorder, recent research suggests that it is a more complex disorder that also includes language impairment (i.e., phonological deficits).

Aim: This study had three general objectives. Since this is the first study of CAS in Croatian, the first goal of this study was to explore speech motor and phonological abilities and also to provide comprehensive data on CAS in another language with a set of speech tasks representing all the processes assumed in speech production (i.e., with reference to the Cascade model; Ozanne, 1995). The second goal was to better understand the cascading effect of motor-speech difficulties on a child's phonological abilities, and the third goal was to distinguish subgroups of children with CAS based on their phonological abilities.

Methods: Children with childhood apraxia of speech (n=30) and typically developing children (n=28) aged 64 to 91 months participated in this study. Exclusion criteria for participants were: neurological or physical cause of the speech sound disorder, motor impairment, cognitive impairment, hearing impairment, and inadequate receptive language skills. For this study, a test battery was created with a series of speech tasks representing different steps in the speech production process.

Results: The present study showed that children with CAS exhibit difficulties at both linguistic and motor levels (i.e., throughout the speech production process), beginning with phonological representations (vowels), phonological awareness, rapid automatized naming, whole-word variability, novel word learning, consonant deletion/substitution, maximum repetition rate, and accurate phoneme productions. In addition, this study showed that the effect of speech motor production on phonological awareness tasks was much more pronounced in children with CAS; that is, they were significantly more successful as long as they did not have to implement motor planning and programming. Third, within the CAS group, three subgroups were identified that showed differences in complex phonological awareness abilities, bi and trisyllabic repetition rates, and correct consonant production.

Key words: childhood apraxia of speech; motor speech disorder; phonological abilities; motor planning; processing level; speech sound disorder

Prošireni sažetak

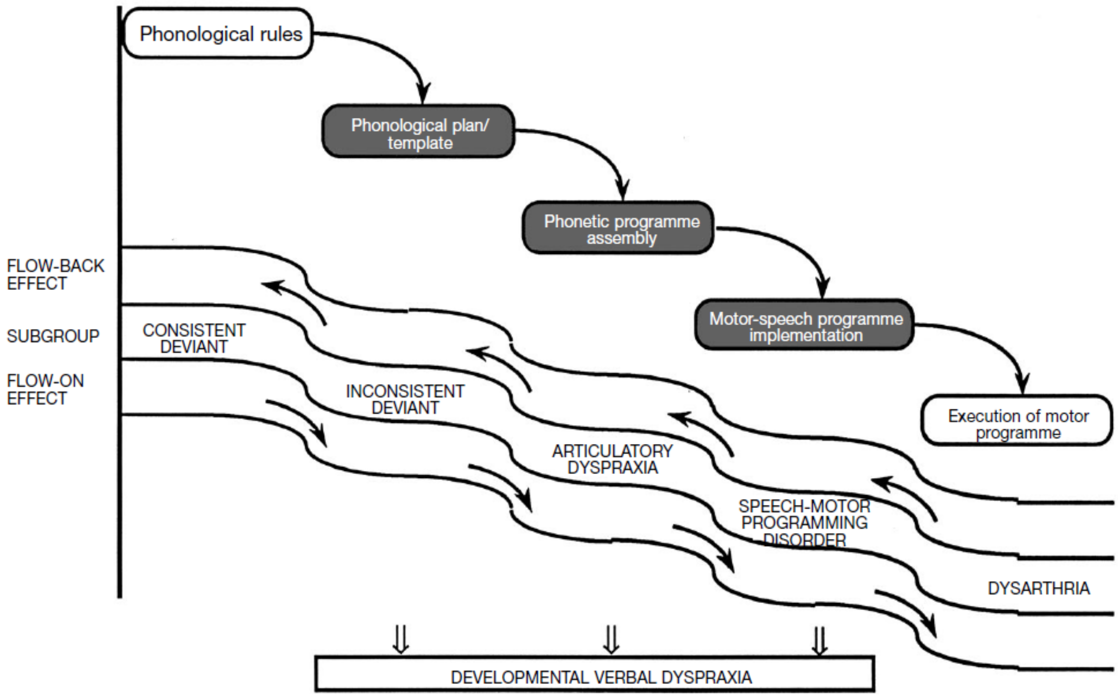
Uvod

Govor je vrlo kompleksan mentalni i fizički process te predstavlja jedan od najučestalijih jezičnih modaliteta. Većini ljudi govor predstavlja jednostavnu i automatiziranu aktivnost, stoga rijetko razmišljaju o njegovoj složenosti. Naime, da bi dijete savladalo govor mora naučiti percipirati i izgovarati suglasnike i vokale, suglasničke skupine, prozodijska svojstva te fonološka pravila jezika kojem je izloženo kako bi u konačnici imalo razumljiv govor (van Haaften et al., 2020). Nažalost, nisu sva djeca jednako brza i uspješna u savladavanju navedenih prepreka te se tada kod djece javljaju teškoće u izgovoru.

Poremećaji izgovora glasova (eng. *speech sound disorders* - SSD) krovni je pojam koji uključuje teškoće s proizvodnjom glasova, njihovom percepcijom i/ili fonološkim reprezentacijama (Limbrick, McCormack & McLeod, 2013). Ova heterogena dijagnostička kategorija uključuje djecu s artikulacijskim poremećajima, fonološkim poremećaja i dječjom govornom apraksijom glasova te čini više od 70% ukupnog broja slučajeva u logopedskoj praksi (Waring i Knight, 2013). Dječja govorna apraksija (DGA) već dugi niz godina predstavlja kontroverzan entitet u znanstveno-istraživačkim krugovima. Općeprihvaćenu definiciju daje Odbor za dječju govornu apraksiju Američkog udruženja logopeda ASHA (2007) koji ovaj poremećaj definira kao neurološki dječji govorni poremećaj u kojem su narušeni preciznost i dosljednost pokreta na kojima se govor temelji, bez prisustva neuromišićnih odstupanja (neprimjereni refleksi i tonus). Osnovni je problem u planiranju i/ili programiranju prostorno-vremenskih parametara slijeda pokreta koji posljedično rezultiraju greškama u govornoj proizvodnji i prozodiji. Iako do danas ne postoji dogovoreni popis značajki specifičnih za DGA, za sljedeće tri značajke postignut je konsenzus među stručnjacima te predstavlja svojevrsni zlatni standard u identificiranju DGA (ASHA, 2007; Iuzzini-Siegeli i sur., 2017): (1) nekonzistentne greške u izgovoru suglasnika i vokala prilikom višestrukog ponavljanja određenog sloga ili riječi, (2) produžena i otežana koartikulacijska tranzicija između glasova i slogova; (3) neprimjerena prozodija.

Napretkom fonoloških istraživanja, istraživači su pokušali identificirati procese u pozadini govora te su u skladu s tim pretpostavkama formirali različite modele čija nam deduktivna priroda omogućava interpretaciju procesa u pozadini govorne proizvodnje. Svi se modeli govorne proizvodnje slažu oko sljedećih procesa: odabir primjerenih riječi (na semantičkoj razini), biranje fonoloških reprezentacija i njihova pretvorba u motoričke reprezentacije te u konačnici izvedba tih pokreta (Maassen & Terband, 2015).

Ovo se istraživanje priklanja hijerarhijskom modelu govorne proizvodnje točnije Kaskadnom modelu planiranja i programiranja govornog izlaza (Ozanne, 1995; 2010) koji detaljnije nadopunjuje model Levelta i suradnika (Levelt, Roelofs, & Meyer, 1999), kod kojih je artikulacijska mrežu završna razina govorne proizvodnje, dok Ozanne (1995) detaljnije razlaže ovu završnu motoričku razinu u dvije: motoričko programiranje i motorička izvedba. Komunikacija među pojedinim razinama uglavnom je jednosmjerna, no ovaj model za razliku od nekih je dvosmjeran te ostavlja mogućnost i obrnutog pravca - utjecaj nižih razina na više. Taj je utjecaj u Modelu predstavljen i grafički; strelice pokazuju protok u oba smjera ostavljajući mogućnost međusobnog utjecaja jedne razine na drugu, pri čemu nije razjašnjeno utječu li govorno-motorička odstupanja na fonološke sposobnosti.



Slika 1. Kaskadni model planiranja i programiranja govornog izlaza (Ozanne, 2010, p.81)

Ovaj model (slika 1) sugerira da djeca s DGA pokazuju odstupanja na tri razine, a to su fonološki plan ili predložak, sklapanje fonetskog plana i implementacija motoričko-govornog programa, tj. da se porijeklo problema nalazi u tranziciji fonološkog koda u artikulacijsko-motorički izlaz. Prva se razina odnosi na fonološko/jezično odstupanje, a preostale dvije na govorno-motoričko odstupanje. Nadalje Ozanne (2005) postavlja tezu da će sva djeca s DGA imati odstupanja na dvije govorno-motoričke razine, dok će samo neka djeca pokazati odstupanja i na razini fonološkog planiranja. Iako ovaj model predlaže odstupanja samo na hijerarhijski nižim razinama modela (motoričke razine), mnoga istraživanja pronašla su dokaze u prilog odstupanjima i na hijerarhijski višim razinama modela tj. na fonološkim razinama (fonološka pravila i sklapanje fonetskog plana). Pa su tako istraživanja pokazala da djeca s DGA pokazuju teškoće s auditivnom diskriminacijom vokala (Maassen, Groenen,& Crul, 2003), zatim da imaju teškoća s proizvodnjom rime (Marion, Sussman, & Marquardt, 1993), fonološkom svjesnošću na razini sloga (Marquardt, Sussman, Snow, & Jacks, 2002) kao i sa sveukupnom fonološkom svjesnošću (McNeill, Gillon, & Dodd (2009). Nadalje, nekonzistentnost u govoru često se navodi kao jedno od obilježja DGA (ASHA, 2007), a dokaz je teškoća s fonološkim planiranjem. Pa tako Iuzzini-Seigel, Hogan, i Green (2017) u svom istraživanju navode da je upravo nekonzistentnost u govoru osnovno obilježje DGA, jer ne može biti objašnjeno jezičnim odstupanjima. S obzirom da 75% spoznaja o DGA dolazi iz engleskog jezika, pitanje je koliko su ta obilježja primjenjiva na hrvatski jezik zbog različitosti u fonološkim sustavima

Ciljevi istraživanja

Ovaj komplicirani i preklapajući odnos između fonološkog i motoričkog razvoja govora otežava potencijalno izoliranje razina na kojoj se javlja teškoća, stoga ne čudi činjenica da trenutno ne postoji slaganje oko pitanja supostojanja teškoća na fonološkim razinama kod djece s DGA. Ovim se istraživanjem ne dovodi u pitanje postojanje teškoća na govorno-motoričkoj razini već se uključivanjem svih razina govorne proizvodnje želi proširiti spoznaja o fonološkim vještinama djece s DGA. Stoga je prvi cilj ovog istraživanja bio sustavno ispitati sva obilježja govorno-motoričkih i fonoloških sposobnosti djece s dječjom govornom apraksijom u hrvatskom jeziku koristeći Kaskadni model kao uporište za identifikaciju razina u procesu govorne proizvodnje. S obzirom na to da je pri ispitivanju fonoloških sposobnosti, posebno na zadacima fonološke svjesnosti gdje je potrebno dati verbalan odgovor (tj. uključuju govorno-motoričku izvedbu), ovim se istraživanjem želio ispitati odnos fonoloških i govorno-motoričkih razina tj. interakcija između zadataka fonološke svjesnosti koji uključuju i ne uključuju davanje verbalnog odgovora (tj. govorno-motoričku izvedbu). S obzirom na heterogenost ove skupine, treći cilj ovog istraživanja bio je ispitati postojanje podskupina djece, posebno hoće li se formirati skupina djece s obzirom na odstupanja na fonološkim razinama govorne proizvodnje. U skladu s tim postavljenje su sljedeće hipoteze:

H1: Djeca s dječjom govornom apraksijom postizat će značajno lošije rezultate na svim razinama govorne proizvodnje (fonološka pravila; fonološko planiranje; fonetsko programiranje; implementacija govorno-motoričkog programa; izvedba) u odnosu na djecu urednog razvoja.

H2: Djeca s dječjom govornom apraksijom postizat će lošije rezultate na zadacima za ispitivanje fonološke svjesnosti kada oni uključuju govorno-motoričku izvedbu (verbalni odgovor) u odnosu na zadatke koji ju ne uključuju (bez verbalnog odgovora).

H3: Identificiranjem obilježja odstupanja na fonološkim razinama formirat će se dodatne podskupine djece unutar skupine djece s dječjom govornom apraksijom.

Metodologija

Ispitanici: U istraživanju je sudjelovalo ukupno 58 ispitanika: djeca s DGA (n=30) i djeca urednog razvoja (n=28). Dob ispitanika varirala je između 5;04 i 7;07, s prosječnom dobi od 6;04. Grupe se nisu razlikovale po dobi, no pregledom raspodjele po spolu uočen je dominanto muški spol u skupini djece s DGA (26 dječaka i 4 djevojčice). U istraživanje nisu uključena djeca s intelektualnim teškoćama, receptivnim jezičnim teškoćama, neurološkim odstupanjima te motoričkim i senzoričkim poremećajima. Svi su ispitanici bili jednojezični govornici hrvatskog jezika, redovito uključeni u odgojno-obrazovnu ustanovu, dok su sva djeca s DGA bila uključena u kontinuirani logopedski tretman.

Materijali: Prije provođena baterije zadataka koja ispituje sve razine govorne proizvodnje, ispitanici su testirani na koloriranim progresivnim matricama (Raven, 1999) za procjenu opće inteligencije te na testu razumijevanja gramatike (Bishop, Kuvač Kraljević et al., 2014) kojim se procjenjivalo jezično razumijevanje. Potom su ispitani na bateriji zadataka koja ispituje sve razine govorne proizvodnje prema Ozanne (1995). Zadaci za ispitivanje fonološkog plana uključivali su AX zadatak diskriminacije vokala te zadatke fonološke svjesnosti preuzete iz Testa za procjenjivanje predvještina čitanja i pisanja (predČiP; Kuvač Kraljević i Lenček, 2011). To su raspoznavanje rime, proizvodnja rime, slogovno stapanje, slogovna raščlamba, fonemsko stapanje i fonemska raščlamba. Zadaci slogovnog i fonemskog stapanja za potrebe ovog istraživanja su konstruirane na način da ne zahtijevaju davanje verbalnog odgovora već je dovoljno pokazati točan odgovor od četiri pokuđena (jedna točan i tri distraktora). Zadaci za ispitivanje fonološkog plana uključivali su zadatak brzog imenovanja (predČip), dva zadatka konzistentnosti: zadatak uzastopnog ponavljanja riječi i zadatak uzastopnog ponavljanja pseudoriječi, konstruirani za potrebe istraživanja po uzoru na CAI tets (Maassen i sur., 2019) te zadatak učenja novih riječi (konstruiran po uzoru na Dodd i sur., 2010). Ispitivanje fonetskog programiranja uključivalo je postotak točno izgovorenih suglasnika u inicijalnoj i finalnoj poziciji u slogu te postotak suglasnika koji nisu ispušteni u ponavljanju 10 riječi i 10 „logatoma“ prema testu artikulacije (Vuletić, 1990). Ispitivanje implementacije govorno-motoričkog plana uključivalo je zadatak govorne dijadohokineze, zadatak preuzet iz CAI testa (Maassen i sur., 2019), a razina govorne izvedbe uključivala je postotak točno izgovorenih suglasnika i vokala u ponavljanju 10 riječi i 10 „logatoma“ prema testu artikulacije (Vuletić, 1990).

Postupak i obrada podataka: Cijeli postupak ispitivanja trajao je između 90 i 100 minuta po ispitaniku. Baterija zadataka procesa govorne proizvodnje trajala je između 45 i 60 minuta s kratkim pauzama (5 min) između zadataka, ovisno o potrebama djeteta. Postupak je proveden u izoliranoj i tihoj sobi jer su se neki odgovori snimali. Nakon prikupljana svih podataka, provedene su sve potrebne predradnje za unošenje i analizu podataka u programu SPSS Statistics, verzija 25.0.

Rezultati i rasprava

Rezultati ovog istraživanja potvrdili su prvu hipotezu kao i pretpostavku Kaskadnog modela govorne proizvodnje a to je da će djeca s DGA pokazati odstupanja na motoričkim razinama modela. Naime, djeca s DGA pokazuju statistički značajno veći broj supstituiranih ili ispuštenih suglasnika u inicijalnoj i finalnoj poziciji u slogu kao i ukupno u broju ispuštenih suglasnika. Djeca s DGA značajno su sporija na zadatku govorne dijadohokineze, i to na svim razinama: jednosložnim (/pa/ta/ka/), dvosložnim (/pata/, /taka/) i trosložnim (/pataka/) sekvencama, s tim da čak 53% djece s DGA nije moglo proizvesti trosložnu sekvencu. Nadalje, djeca s DGA pokazuju značajne teškoće s postotkom uspješno izgovorenih suglasnika (69,9%) i vokala (93.2%) u usporedbi s djecom urednog razvoja (suglasnici = 98.2%; vokali = 99.8%). Ovi rezultati potvrda su teškoća s motoričkim planiranjem i programiranjem djece s DGA, no pokazali su i da djeca s DGA značajno odstupaju i na fonološkim razinama modela, počevši od najviše razine – diskriminacije vokala. Naime, djeca s DGA manje su uspješna razlikovanju vokala od djece urednog razvoja. Nadalje, djeca s DGA postižu značajno niže rezultate na svim ispitanim aspektima fonološke svjesnosti. Iako u usporedbi s djecom urednog razvoja postižu značajno niže rezultate na razini rime, njihovi prosječni rezultati na razini rime (9.1) su unutar graničnog postignuća. Razlikuju se i na zadatku brzog imenovanja gdje u prosjeku imenuju 0.8 slika u sekundi, dok djeca urednog razvoja imenuju u prosjeku 1 sliku u sekundi. Djeca s DGA također pokazuju značajno veću razinu nekonzistentnosti u govoru; na zadatku uzastopnog ponavljanja riječi, zadatku uzastopnog ponavljanja pseudoriječi, kao i na zadatku učenja novih riječi. Na zadatku učenja novih riječi osim nekonzistentnosti pokazuju i značajno veću razinu netočno proizvedenih riječi.

Drugom hipotezom očekivano je da će djeca s DGA postizati lošije rezultate na zadacima fonološke svjesnosti kada oni uključuju govorno-motoričku izvedbu u odnosu na zadatke koji ju ne uključuju. Djeca su testirana uparenim zadacima na istim razinama fonološke svjesnosti: plitka (slogovna raščlamba i slogovno stapanje), plitka-duboka (prepoznavanja i proizvodnja rime) i duboka (fonemska raščlamba i fonemsko stapanje). Zadaci prepoznavanja rime te stapanja ne uključuju govorno-motoričku izvedbu, dok zadaci proizvodnje rime i raščlambe uključuju govornu proizvodnju. Iako je razvojno očekivano i potvrđeno da će i djeca urednog razvoja biti bolja na zadacima koja ne uključuju govorno motoričku izvedbu, taj je efekt govorno-motoričke izvedbe izraženiji kod djece s DGA, što rezultira većim porastom uspješnosti u zadacima koji ne uključuju govorno-motoričku izvedbu u odnosu na one koji ju uključuju. Istraživanje je pokazalo da razlika ne postoji samo na razini sloga, što upućuje na to da djeca s DGA pokazuju odstupanja na dubljim razinama fonološke svjesnosti te da izvedba složenog govornog zadatka može utjecati na višu, jezičnu razinu procesa govorne proizvodnje, no daljina istraživanja fonološke svjesnosti kod djece s DGA su potrebna kako bi potvrdio taj zaključak, no je ovo istraživanje prvi korak u tom smjeru.

Trećom hipotezom pretpostavljeno je da će se identificiranjem obilježja odstupanja na fonološkim varijablama formirati dodatne podskupine djece unutar skupine djece s dječjom govornom apraksijom. Nakon provedene klasterizacije, algoritam k-means identificirao je tri skupine djece s DGA (klaster 1, N=9; klaster 2, N=7; klaster 3, N=13), nakon četvrte iteracije. Varijable koje razlikuju klastere su: proizvodnja rime, fonemska raščlamba, govorna dijadohokineza (dvosložne i trosložne sekvence), postotak točno izgovorenih suglasnika, te postotak točno izgovorenih vokala u inicijalnoj i finalnoj pozivciji u slogu. Klaster 3 predstavlja skupinu djece koja pokazuje nešto lošije rezultate od klastera 1 i slične rezultate kao klaster 2 u proizvodnji rime i raščlambi slogova, međutim pokazuju značajno bolje rezultate u proizvodnji suglasnika i na zadatku govorne dijadohokineze dvosložnih i trosložnih sekvenci. Klaster 2 predstavlja skupinu s najlošijim postignućima na svim spomenutim mjerama. U konačnici možemo reći da je treća hipoteza djelomično potvrđena jer navedene varijable predstavljaju i fonološke i motoričke aspekte procesa govorne proizvodnje.

Zaključak

Ovim su doktorskim istraživanjem po prvi put sustavno ispitana obilježja govorno-motoričkih i fonoloških sposobnosti djece s DGA u hrvatskom jeziku. Uvidom u procese koji prema Kaskadnom modelu (Ozanne, 10995) predstavljaju sve razine govorne proizvodnje možemo zaključiti da djeca s DGA pokazuju odstupanja na svim razinama govorne proizvodnje, i jezičnim i motoričkim tj. hijerarhijski višim i nižim razinama. Nadalje pokazana je interakcija govorno motoričke izvedbe i fonološke svjesnosti koja ukazuje na kompleksan odnos između jezičnih i motoričkih razina koji nije nužno jednosmjeran, te implicira da kod djece s DGA možemo očekivati odstupanja i na višim razinama. Jedno od ograničenja ovog istraživanja je da nisu uključene druge skupine djece kao što su djeca s fonološkim teškoćama stoga nije moguće donositi generalne zaključke koliko su ova odstupanja specifična isključivo za djecu s DGA. Otkrivanjem podskupina djece s DGA još je jednom ukazalo na heterogenost ove skupine, no i potvrdilo da su svim podskupinama zajedničke teškoće u motoričkom planiranju i programiranju. Kao teorijsko polazište u ovom se istraživanju, za kreiranje konstrukta procesa govorne proizvodnje, koristio Kaskadni model (Ozanne, 1995), no moguće je da su pojedina odstupanja odraz višestrukih odstupanja. Primjerice u ovom je modelu postotak točno izgovorenih suglasnika i vokala mjera završne razine govorne izvedbe, no moguće je da je smanjena točnost posljedica i teškoća s fonološkim planiranjem i teškoća s govornom izvedbom. No, ispitivanjem svih razina i njihovim međusobnim korelacijama dobivena je šira slika o procesima u pozadini DGA čime se u konačnici doprinosi kvalitetnijem identificiranju i tretmanu djece s dječjom govornom apraksijom.

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1. INTRODUCTION

It is true that humans, and only humans, have evolved a complex set of voice, hearing, and brain-processing skills enabling very sophisticated vocal communication, also known as speech. Speech usually refers to the ability of humans to produce sounds that are used to convey a message. Speech is only one modality for the expression of language; however, it is not surprising that the broader meaning of speech (freedom of speech) has been recognised as one of the fundamental human rights in the Universal Declaration of Human Rights and in the international human rights laws of the United Nations.

Speech as a modality has a special importance because it is the primary, first-learned modality for hearing language users. It represents a system that consistently relates the meanings of a language with the speech sounds by which the language is communicated (Kent, 2017). Speech sounds may be viewed from two perspectives - as motor production (articulation perspective) and as units that facilitate the expression of meaning (language perspective) - phonemes.

Speaking is a highly complex physical and mental process, "at least as complex as ice dancing" (Maassen & Terband, 2015, p. 331), even though we rarely think of it that way, because for most of us it is like breathing, we do not think about it and barely acknowledge its complexity. The development of speech sounds can be described as the acquisition of individual speech sounds and the organization of those sounds into speech patterns that include both phonetic (i.e., articulatory) and phonological (i.e., phonemic) development (van Haaften et al., 2020). It involves learning a language (its syntax, semantics, and phonology) - a speech code that links meaning to sound, as well as a motor skill by which the speech organs are controlled to produce rapid and overlapping movements (Kent, 2020), but it is because of language that speech has become the most complex motor performance humans perform (Maassen &Terband, 2015).

From the moment children are born, they vocalize. Infants acquire control over the movements of its speech organs by establishing a mapping between articulator movements and their auditory and somatosensory consequences (Terband, 2011). The transition from vocalization to intelligible speech takes many years as children's body structures develop and their perception and production systems become more and more sophisticated and attuned to their language (McLeod, 2017). Therefore, it is not surprising that a developing speech differs from adult speech in misarticulations, slower speech rates, greater variability (errors) in production, and reduced anticipation in articulatory sequencing (Kent, 2020).

1.1. Speech sound disorders

Children's acquisition of speech involves mastery of the perception and production of consonants, vowels, consonant clusters, tones, prosodic features, and phonological rules of the language(s) they are exposed to, with the outcome of intelligible speech (van Haaften et al., 2020). However, not all children acquire spoken language at the same rate; they differ in age of speech onset, rate of development, and types of developmental errors (Dodd, Holm, Crosbie & Hua, 2010). Although most children resolve those differences, unfortunately it is not the case for all of them.

Speech sound disorders (SSD) are not a new area of research; in fact, one of the first papers by Samuel Potter, entitled *Speech and its defects*, appeared in 1882, and since then, a growing concern about speech sound difficulties and other communication disorders led to the founding of the American Academy of Speech Correction in 1925, the forerunner of the American Speech-Language-Hearing Association (ASHA) (Bankson, Bernthal & Flipsen, 2017). It was no longer enough to focus on correcting a lisp; the underlying nature of these problems needed to be understood. In the 1970s, thanks to linguistics, there was a shift in perspective that indicated that SSD should be viewed not only from the perspective of motor production, but also from the perspective that such difficulties may reflect a child's lack of phonological language rules (Bankson et al., 2017).

The foundations of assessment, classification, and intervention for children with SSD have been heavily influenced by psycholinguistic theories (Namasivayam, Coleman, O'Dwyer, & van Lieshout, 2020). With these advances, several systems for classifying SSD subtypes have appeared, such as the Psycholinguistic Framework by Stackhouse and Wells (1997), the Speech Disorders Classification System (Shriberg et al., 2010), and the Model of Differential Diagnosis (Dodd, 2014).

Since then, a growing body of research suggests that SSD is an umbrella term that refers to problems with speech sound production, perception, and/or phonological representation that can impede language comprehension as well (Limbrick, McCormack & McLeod, 2013). SSD can be characterized as a disorder that ranges from something "mild" (such as interdental production of the /s/ sounds) to one that is so severe that speech is completely unintelligible. Typically, SSD are most common in the paediatric population. SSD typically occurs in children younger than 8 years of age, but errors in speech sound production can persist beyond this age and continue into adulthood (Bankson et al., 2017). In addition, research shows that children with SSD account for between 56 percent (Bankson et al., 2017) and 70 percent (Waring & Knight 2013) of the total clinician caseload.

Children with SSD are often divided into two relatively large groups. The first group is organic disorders whose difficulties with speech sound are associated with an obvious aetiology or cause, and the second group is those whose difficulties have no obvious cause (Flipsen, Bernthal & Bankson, 2017a). This second SSD group has been referred to over the years as functional articulation disorders, developmental phonological disorders, idiopathic speech sound disorders, and speech delay of unknown origin. (Flipsen et al., 2017a).

Children with SSD vary in terms of severity, underlying cause, features of the language disorder, and involvement of other aspects of the language system (Dodd, 2011). According to Bowen (2015), children with SSD may exhibit a number of features, including poor stimulability, system and substitution errors, syllable structure errors, consonant distortions, vowel deviations, atypical prosody, unusual tonality, and inappropriate timing. In addition, any or all of these language features may occur singly or in combination, so that children's difficulties may include a mixture of phonetic (articulatory), phonemic (phonological or cognitive-linguistic), structural (craniofacial or syndromal), perceptual, or neuromotor bases (Bowen, 2015).

Even though the term SSD is theory-neutral, this complex relationship between the aetiology (distal), processing deficits (proximal) and the behavioral levels (speech symptoms) is under-specified (Terband, Maassen & Maas, 2019). When we speak of SSD today, there are three main terms that fall under this umbrella term: Phonological Disorder, Articulation Disorder, and Childhood Apraxia of Speech (Bowen, 2015).

1.2. Childhood apraxia of speech (CAS)

Childhood apraxia of speech is a controversial speech sound disorder whose actual existence, nature, and cause continue to be debated. An ever-growing number of studies attempt to illustrate the heterogeneous nature and underlying causes of children with CAS and the changing face of this disorder over time. This chapter reviews current thinking on terminology, definition, causes, epidemiology, and theoretical perspectives on the core problem of CAS.

1.2.1. Terminology and definition of CAS

When one begins to look at the literature on childhood apraxia of speech (CAS), one is likely to feel discouraged after reading the first sentences of the various books and articles:

> „*Childhood Apraxia of Speech (CAS) is a controversial diagnostic label in the literature of communication disorders*…“ (Ozanne, 2010, p. 71).
>
> „*Developmental verbal dyspraxia is a controversial speech disorder with continued debate over its existence, nature, and diagnosis.*“ (McNeill, 2013, p. 49).
>
> „*One of the least understood SSD categories is childhood apraxia of speech (CAS).*” (Flipsen et al., 2017a, p. 132).

This condition was first described in 1954 by Morley et al, who used the term "*developmental dyspraxia*" to describe a group of children whose speech features resembled those of adults after brain injury. Thus, the term "*dyspraxia*" was borrowed from adult literature. Since then, this condition has also been known by various other names.

Originally, the term "*Developmental Apraxia of Speech* (DAS) " was used to indicate that the problem has been evident from speech/ language onset and that the disorder is not caused by hearing impairment, autism spectrum disorder, intellectual disability, or neuromotor disorder such as cerebral palsy, muscle weakness, or incoordination, although it can co-occur with all of these disorders (Ozanne, 2010). Later, the term *developmental* was replaced by the term *childhood* to differentiate it from the adult form and to clarify that it does not resolve with age and without intervention (Ozanne, 2010). Terms such as "*Developmental Articulatory Dyspraxia* (DAD)" and DAS emphasize the articulatory aspect of the disorder, whereas the term "*Developmental Verbal Dyspraxia* (DVD)" used by Stackhouse (1992) refers to the language features often seen in children with CAS. Today, the majority of research uses the term CAS, which was adopted by the American Speech-Language-Hearing Association (ASHA) in 2007, although the term DVD can still be found in the literature, particularly in the United Kingdom (McNeill, 2013). In the present study, the term CAS is used.

Aside from the terminological confusion, there appear to be two other difficulties that have contributed to the controversy with this diagnosis. First, the lack of consensus on the specific distinguishing features of this disorder or, in the past, even whether such features exist at all (Flipsen et al., 2017a).

In 2002, ASHA established an ad hoc committee on this particular topic. This committee was tasked with reviewing the extensive and sometimes conflicting literature in this area. It presented its findings in a technical report (ASHA, 2007). This document is still widely used as a reference point for CAS today. In addition, ASHA states that CAS is a unique diagnostic category as a subgroup of children with SSD of unknown aetiology. ASHA defines CAS as follows:

"*Childhood apraxia of speech (CAS) is a neurological childhood (paediatric) speech sound disorder in which the precision and consistency of movements underlying speech are impaired in the absence of neuromuscular deficits (e.g., abnormal reflexes, abnormal tone). CAS may occur as a result of known neurological impairment, in association with complex neurobehavioral disorders of known or unknown origin, or as an idiopathic neurogenic SSD. The core impairment in planning and/or programming spatiotemporal parameters of movement sequences results in errors in speech sound production and prosody*." (2007, pp. 3–4).

1.2.2. Causes and epidemiology of CAS

Although ASHA (2007) defines CAS as a result of neurological problem, it can also be comorbid with galactosemia, Rett syndrome (Flipsen et al., 2017a), fragile X syndrome (Spinelli et al. 1995), and Down syndrome (Kumin 2006). At one point, autism spectrum disorder (ASD) was included in this list, but a study by Shriberg, Paul, Black, and van Santen (2011) that examined the speech and prosody profile of 46 verbal children with ASD showed that the speech errors observed in this population differed from those observed in CAS, however CAS and ASD are often comorbid, with the frequency of CAS being increased in children with ASD (Tierney et al. 2015). To date, the most common origin of CAS is idiopathic (unknown).

Childhood apraxia of speech (CAS) has been described as a highly heritable condition with strong family aggregation (Lewis et al. 2004). Some evidence of familial aggregation and transmission of CAS has been noted in the K.E. family, where the primary impairment in affected members was the inability to sequence oral movements, a hallmark of CAS (Watkins et al., 2002). However, Lewis et al. (2004) point out that it is not known whether the K.E. family is representative of families with CAS in the general population. Later, it was shown that mutations of FOXP2 were associated only with a specific phenotype of CAS and language impairment without intellectual disability, whereas other genotypes associated with CAS (such as BCL11A, KANSL1, GRIN2A) could lead to a broader phenotype in which CAS may occur as part of the broader spectrum of the condition (Morgan & Webster, 2018).

A study by Liegeois and Morgan (2012) examined the neurological functioning of 45 children with CAS described in 12 studies and found that the majority (96%) of descriptions of CAS with neurological findings were from children with concomitant neurobehavioral diagnoses. Considering that most MRI brain scans did not reveal abnormalities, the authors concluded that neurologic abnormalities in CAS may lie at the sub-macroscopic, metabolic, and/or neurotransmitter levels.

Epidemiological data for CAS is rather limited and inconclusive. An exact prevalence has not been determined, and no systematic population studies have been conducted (ASHA, 2007). One of the most commonly used and cited prevalence studies for this population is a study by Shriberg, Aram, and Kwiatkowski (1997a). They calculated an overall prevalence of 1 to 2 cases per 1000 children, but based solely on clinical referrals. In addition, Broomfield and Dodd (2004) identified only two potential cases of CAS among 936 referrals in the United Kingdom study. Although the exact prevalence may vary depending on the definition of CAS, gender ratio analysis continuously shows that CAS is more prevalent in males than females (McNeill, 2013). Lewis et al. (2004) reported a 2:1 ratio of males to females (sample of 22 children) while Hall, Jordan, and Robin (1993) reported that 74% of 229 cases of CAS reported in the literature were male.

1.2.3. The core problem of CAS

In addition to the controversy over terminology, causes, epidemiology, and symptomatology, there is an ongoing debate about the core problem of CAS. Traditional theoretical viewpoints have been divided into motor and linguistic perspectives (McNeill, 2013).

Traditionally, the underlying nature of CAS was thought to be motoric (Ozanne, 2010). This perspective supports the idea that symptoms are due to a core problem in speech motor planning and/or programming processes (McNeil & Kent, 1990; Shriberg et al., 1997a). Proponents of this perspective do not deny the broader language impairments observed in CAS, but view these difficulties as concomitant or simply a consequence of speech motor control involvement (McNeill, 2013).

However, with advances in phonological research, a debate arose as to whether the nature of the impairment is in fact phonological (Ozanne, 2010), prompting further discussion as to whether CAS should be classified as a "*syndrome*" or solely as a motor-speech disorder (Stein et al., 2020). This representational and speech motor control perspective of CAS was developed in an effort to conceptualise the more linguistic features of CAS as part of the core problem of the disorder, as opposed to the purely motor perspective (McNeill, 2013). This perspective is supported by the research showing co-occurring language impairments among children with CAS (Stackhouse & Wells, 1997; Iuzzini, 2012; Lewis et. al., 2004; Gillon & Moriarty, 2007; Thoonen et. al.,1997).

Therefore, it is not surprising that to date there is no validated list of pathognomonic features. As Flipsen et al. (2017a) noted, identifying CAS features forces us into a circularity problem where we want to know what features make this group distinct, but we need to know what those features are in order to select the right children to look for those features. Still, the ASHA technical report (2007) identified three segmental and suprasegmental features that are consistent with a deficit in the planning and programming of movements for speech that have gained some consensus among researchers in the field of childhood apraxia of speech (Maassen & Terband, 2015):

(1) inconsistent errors on consonants and vowels in repeated productions of syllables or words;
(2) lengthened and disrupted coarticulatory transitions between sounds and syllables;
(3) inappropriate prosody.

Maassen and Terband (2015) noted an important shift in perspective that the fundamental question about CAS should not be which of the speech (and possibly other) symptoms belong to the diagnostic category of CAS, but we should be asking how can we prove that the child with a speech sound disorder (SSD) has a deficit at the level of speech motor planning and programming.

1.3. Speech production in children with CAS

Despite the lack of agreement on the core problem of CAS, research continues in this population. There is a similar ongoing debate in the field of acquired apraxia of speech (AOS) in adults, where McNeil, Pratt, and Fosset (2004) do not question that AOS belongs to motor rather than linguistic levels of processing, but argue that there is a lack of criteria to distinguish between speech motor and linguistic (i.e., phonological) symptoms. The complexity of speech performance suggests that there must be a hierarchy in representation (Maassen & Terband, 2015), which is confirmed by the clinical manifestations of CAS as they occur at a higher level (a higher level than dysarthria) in planning or programming movements for speech (Duffy, 2013). The simultaneous acquisition of well-organised linguistic structures and motor-coordinative commands overlaps in typical development (Nip et al., 2011), which further complicates isolating processing steps in order to identify underlying deficits.

With the development of cognitive neuropsychological and psycholinguistic models of speech production and perception processes, we are beginning to better understand CAS (Maassen, 2002). All models of speech production, like in any motor performance, assume a hierarchy of control (Maassen & Terband, 2015). It is debatable which model best describes all aspects of the speech phenomenon and provides the best insight into the speech motor and linguistic aspects of speech production. However, according to Maassen and Terband (2015) all speech production models agree on "*a preparatory psycholinguistic process of producing a sequence of one or more word forms (a phrase) stored in some short-term memory (buffer), followed by a process that calculates (process of encoding; transcoding; planning; programming) the speech movements that must be made in order to articulate the sequence (phrase). Also, all models agree that the calculations themselves from stored word forms to actual movements are hierarchically structured*" (p. 333).

Kent (2004) considers speech as a cognitive-motor accomplishment and questions the modularity of motor control processes, while Maassen and Terband (2015) note that speech motor processes cannot be clearly separated from cognitive processes due to interaction with higher order psycholinguistic processes, especially in the case of impaired speech systems where this is even more true due to adaptive and compensatory mechanisms.

According to the Information processing model of speech perception and production (Levelt, 1989), speech production begins with the selection of the appropriate word and is followed by phrase planning, which unfolds incrementally with the retrieval of its phonological form and its assembly into a complete phonological phrase, and ends with the transformation of this last level into an articulatory representation that triggers the motor commands for speech production. Phrase planning occurs in parallel with speech motor programming and articulation (Maassen, 2015). The speaker continuously monitors his or her own speech, not only at the level of lexical selection, but also at lower levels of the phonetic plan and during motor programming, including external self-monitoring after the utterance has been produced (Maassen & Terband, 2015). It is therefore justified to ask whether motor deficits influence phonological development or phonological awareness.

Kinematic studies have indeed confirmed that the precision and consistency of movements underlying speech are impaired in children with CAS, but it is less clear which upstream and downstream speech production processes are affected (Maassen, 2015). Upstream processes such as phonological abilities and lexical storage provide the input for phonemic and lexical representations, memory processes that store and retrieve these representations, and transcoding processes for planning and programming motor gestures, while downstream effects lead to speech sound errors that are classified at the perceptual level as substitutions or distortions (Maassen, 2015).

The scheme proposed by Shriberg et al. (1997a) (Figure 1) is based on classical models of speech and speech/language processing (e.g., Dell, 1986; Garrett, 1980; Levelt, 1989), but is intentionally relatively underdeveloped because its research function is useful only for considering speech processing at discrete "stages" or "levels." There are two sets of dichotomies that recur in theoretical approaches to apraxia of speech and are represented by brackets in Figure 1: *Comprehension*/*Decoding* versus *Production*/*Encoding* processes, with *organizational* processes necessarily involved in both activities and Cognitive-*Linguistic/Phonological/Planning* processes versus *Motor-Speech/Phonetic/Programming* processes (Shriberg et al., 1997a). In addition, Shriberg et al. (1997a) mention that newer studies may address one or more of the top five levels of language processing (i.e., auditory-temporal, perceptual-memorial, representational, transformational, selection-retrieval), whereas older studies generally address only the lowest levels of output (i.e., prearticulatory sequencing).

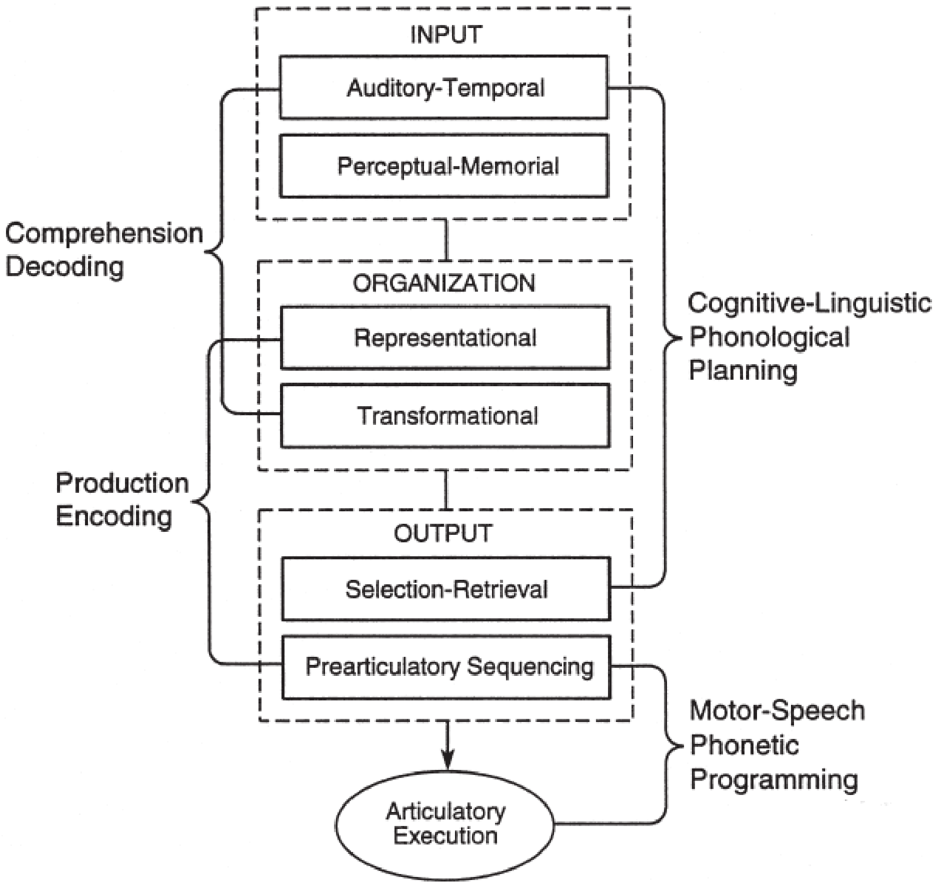


Figure 1. Array of cognitive-linguistic and motor-speech loci implicated in the pathogenesis of suspected CAS. p. 277, Shriberg et al. (1997a, p. 277)

Because there is no obvious physical marker for CAS (Iuzzini-Seigel & Murray, 2017) and speech and language symptoms vary considerably from person to person and over the course of development (Iuzzini-Seigel, Hogan, & Green, 2017), it is difficult to determine exactly which levels of speech production processes are affected. Stackhouse and Wells (1997) note that the deficit in speech motor skills observed in CAS can have implications for a child's language and literacy development ("flow on" effect). One of the most consistent features of the speech of children with CAS is its inconsistency, which may provide inadequate input to the child's developing language system and thus affect auditory processing and later vocabulary knowledge (Stein et al., 2020). Thus, one effect of motor-speech impairment on language and literacy might be that poor speech output impairs accurate encoding of complex words, which in turn affects the child's language system (Stackhouse & Wells, 1997).

This effect of motor-speech difficulties on a child's phonology in CAS could be explained by the Cascade model of speech output processing (Ozanne 1995). This processing model provides a conceptual basis for identifying the underlying deficit in CAS. This model of speech production is conceptually very similar to Level's model, except that Ozanne elaborates further on motor levels of speech production (Figure 2).

The model assumes three levels of deficit in children with CAS: 1. the phonological plan or template; 2. the assembly of the phonetic program/plan; 3. the implementation of the motor-speech program. The first level reflects a phonological deficit, while the other two levels reflect speech motor deficits (Ozanne 1995). Some children with CAS exhibit difficulties at all three levels. However, to be diagnosed with CAS, a child must have a deficit at the motor levels of the model. An important premise of this model is the "flow-on" and "flow-back" effect between levels, which is graphically represented by arrows in Figure 2. This means that this hierarchical model hypothesizes that these processes can influence each other, but to our knowledge there is a gap in understanding whether motor deficits influence phonological development.

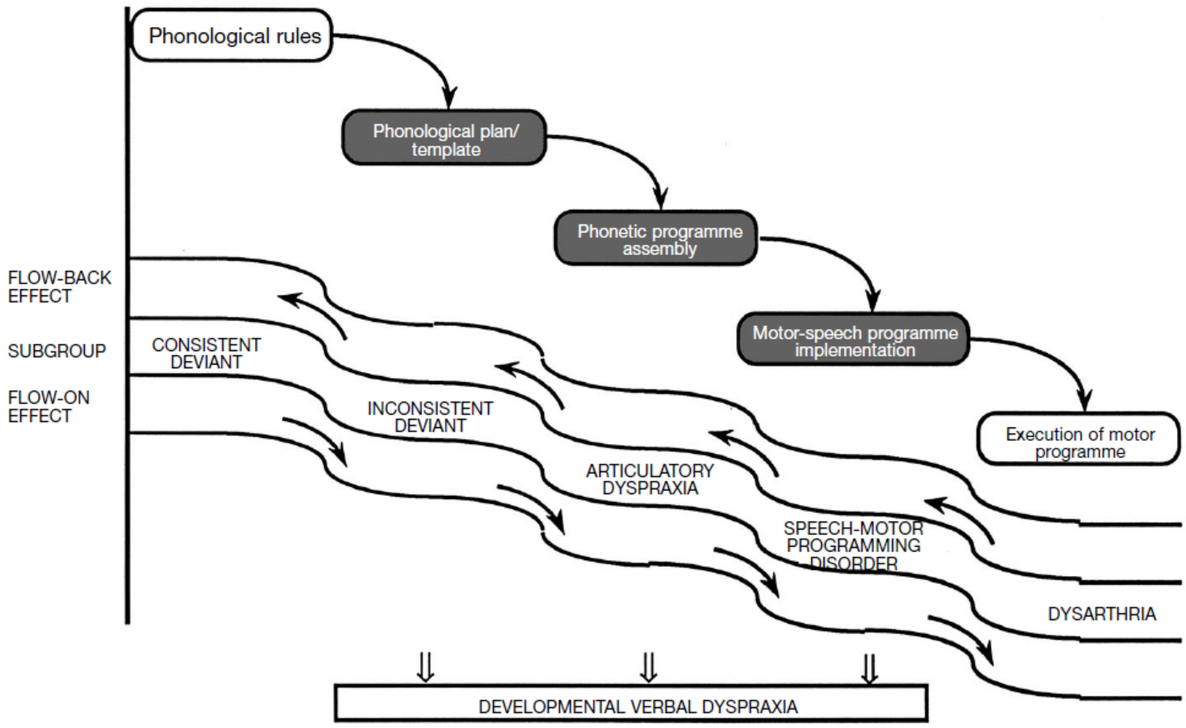


Figure 2. Cascade model of speech output processing (Ozanne, 2010, p.81)

1.4. CAS – more than a motor speech disorder?

Current theories emphasize the complicated relationship between phonological and motor speech development and make the coexistence of features at different levels of speech and language development an expected outcome of an underlying impairment in the planning and programming of speech production (Maassen et al. 2010). Weismer and Green (2015) point out that there is a downside to model-building efforts, which is often confusing with respect to difficult-to-interpret experimental data from individuals with motor speech disorders and the uncertain relationships between these data and the model. In this chapter, the findings from several studies of children with CAS are summarized and explained in terms of the aforementioned speech production models.

1.4.1. Evidence of difficulties with phonological rules

Although the cascade model of speech output processing is viewing CAS as containing deficits at both the linguistic (i.e., phonological plan impairment) and motor levels (i.e., phonetic programming and execution impairments), in order for a child to be diagnosed with CAS, he or she must have a deficit at motor levels of the model (Ozanne, 2010). In addition, this model does not indicate difficulties at the highest level of speech processing (i.e., phonological rules in the Cascade model). However, research has shown that the highest level at which the underlying deficit of CAS was found is the level of lexical representation (i.e., phonological rules).

Shriberg et al. suggested that there is a deficit in phonological representational processes in their 1997 research series. Their results (Shriberg et al., 1997c) showed that children with CAS do not show much self-correction regarding stress, suggesting that representations are not correct and therefore are not perceived as errors. In addition, they found that the production of inappropriate stress does not decrease with speech development, also suggesting a deficit at the representational level. Groenen et al. (1996) performed a categorical classification task on children with CAS and compared it to typically developing children. They found similar results in both groups of children, from which they concluded that children with CAS do not show difficulties in phonetic processing. However, children with CAS showed poorer discrimination results which the authors believed indicated poor auditory processing skills. This was confirmed by the results of a later study by Maassen, Groenen, and Crul (2003).

Maassen et al. (2003, p. 464) asked an important question: "Do children who misarticulate vowels have difficulty forming stable target representations, or is the problem related to inadequate motor control?" Their research (Maassen et al. 2003) showed that auditory perception of vowels is impaired in children with CAS, which in turn affects both production and perception. However, in Lenoci et al.'s (2020) study, high levels of cross-subject variation were found in variability as well as in vowel variability and distinctiveness. Individual variation was found not only in CAS children but also in the speech of typically developing children. Furthermore, speakers with CAS were consistent in showing generalized reduction in F1 distinctions, suggesting that vowels are more centralized and less distinct along the height dimension, indicating the critical nature of height distinctions for speakers with CAS (Lenoci et al., 2020).

Access to a phonologically based representational system is also required to successfully recognize or produce a rhyme, along with: auditory processing of the input target sound and holding it in auditory short-term memory; selection of a unit from the internalized auditory encoded representation that matches the vowel of the input string; and use of this phonological match to drive articulatory output of a rhymed word (Marion, Sussman, & Marquardt, 1993). The Marion et al. (1993) study showed that children with CAS could not produce rhymes, recognize rhymes, or respond to vocalic nuclei in CVCs that represented near-versus-distant rhymes, again confirming difficulties at the phonological level.

Although phonological awareness (i.e., the ability to consciously reflect on and manipulate the subunits of spoken language such as rhymes and syllables) and phonemic awareness (i.e. the ability to discriminate and manipulate individual speech sounds—phonemes) are poorly studied in children with CAS, available data show difficulties with phonological abilities.

Marquardt, Sussman, Snow, and Jacks (2002) showed that children with CAS exhibit problems with phonological awareness at the syllable level: segmenting syllables, judging intra-syllabic sound position and constructing single and consonant cluster arrangements. Furthermore, children with CAS in the McNeill, Gillon, and Dodd (2009) study exhibited poorer phonological awareness than the inconsistent speech disordered group (Dodd, 2005). The performance of the CAS group was inferior to that of the inconsistent group on the measure of receptive phonological awareness, whereas the performance of the two groups on the phonological representation task was comparable. In contrast, there was no difference in the groups' performance on the phonological representation judgement task, which may suggest that the inconsistent group's performance was hindered by inadequate phonological assembly, whereas the CAS group's performance was hindered by their underspecified representational systems. Based on these results, CAS can be viewed as a disorder characterized by an impoverished phonological representation system.

However, all phonological awareness tasks have three main features: (a) the linguistic nature of the stimuli, (b) the phonological complexity of the stimuli, and (c) the response mode (which is almost always a verbal response) (Cunningham et al., 2015). For instance, children who have difficulties in articulating certain phonemes may score poorly on the task despite accurately representing those phonemes. Knowing that variations in performance may result from the effects of each factor, children with CAS are in an unfavourable situation considering the verbal response mode and their problems with speech motor skills. In addition, some studies suggest that phonological tasks requiring a nonverbal response predict reading to the same extent as those requiring a verbal response (Carroll et al., 2003, Gayan & Olson, 2003). The present study aimed to address this methodological problem by including nonverbal responses to phonological tasks.

1.4.2. Evidence of difficulties with phonological planning

Ozanne (1995) conducted a cluster analysis of behaviours that might be indicative of motor programming/planning disorder. The first cluster included behaviours such as inconsistent production of the same word, increased errors with increasing performance load, errors that could not be explained by general articulation or phonological process errors, poor maintenance of phonotactic structure, and vowel errors. The underlying deficit postulated for this group of children is likely to be difficulty assembling the phonological plan for the word or utterance, similar to the children described as having inconsistent phonological disorder in Dodd's model for differential diagnoses.

Children with variable speech (i.e., variable production of phonemes, words, or utterances across multiple repetitions) are likely to be less intelligible because of the unpredictability of their speech production (Holm, Crosbie, & Dodd, 2005). Although speech intelligibility improves with age and intervention, according to Gillon and Moriarty (2007), children with CAS may evidence difficulties when their system is loaded with more complex phonological production tasks. Similarly, Lewis et al. (2004) found that older children with CAS generally have intelligible speech but have difficulty pronouncing new and/or complex multisyllabic words correctly which is then associated with poor learning of speech sound targets in treatment.

Speech inconsistency is indeed the most commonly reported feature across childhood speech disorders by clinicians and researchers (Iuzzini-Seigel, Hogan, & Green, 2017). However, word variability (i.e., variability in repeated production of the same word) is particularly important because it has been associated with CAS (ASHA, 2007) and inconsistent disorders (Dodd, 2005). According to Macrae, Tyler, and Lewis (2014), word variability is shown to peak during developmental change, with a general trend toward decreasing word variability in both typical and disordered speech development.

Iuzzini-Seigel et al. (2017) raise the question of whether speech inconsistency could instead be driven by a higher-order deficit leading to difficulties in sequencing phonological units and acquiring morphological rules. Moreover, reports on speech inconsistency as a defining feature of CAS are inconclusive. Shriberg et al. (1997b) found that error consistency (i.e., the ratio of the most common error class produced per lexical item to the total number of items produced) did not distinguish children with suspected CAS from children with other speech disorders. Similarly, Betz and Stoel-Gammon (2005) found that although children with CAS produced more errors than children with phonological disorders (PD), error consistency did not differ, indicating an equal level of speech inconsistency. Iuzzini (2012) investigated inconsistent performance in preschool-aged children with CAS and phonological disorder and showed that token-to-token inconsistency assessment was less effective than phonemic inconsistency in differentiating between these groups.

King, Jakielski, and Malone (2001) examined variability in repeated productions of connected speech samples in children with CAS and found that of 52 lexical types produced, only two were produced consistently, while the remaining 50 types varied in token production. Iuzzini-Seigel et al. (2017) found that two CAS subgroups (CAS and CAS + language impairment) were equivalent on all speech inconsistency measures (token-to-token inconsistency for mono- and multisyllabic real words and repeated production of the phrase “*buy Bobby a puppy*”), suggesting that speech inconsistency is a core feature of CAS and cannot be attributed to language impairment in this population, whereas children with a language impairment exhibited high levels of sentence inconsistency (i.e., repetition of the sentence "*Buy Bobby a puppy*"). Similarly, Marquardt, Jacks, and Davis (2004) showed that the disorder is characterized by high levels of total token and error token variability including low levels of word target stability and token accuracy.

Children with CAS have shown some other behaviours that may indicate a disorder at the phonological level. For example, research by Lewis et al. (2018) found significant differences, with the CAS group performing more poorly on rapid automatized naming (RAN) than the SSD-only group and the no SSD/LI group. RAN requires naming a small set of randomly presented colours, objects, numbers, or letters as quickly as possible (Lewis et al., 2018). Although most studies have examined the relationship between RAN performance and reading skills, rapid access to and retrieval of phonological information and oral motor skills are also associated with RAN - all skills that may be impaired in children with SSD (Norton & Wolf, 2012; Treiman, 2017) including children with CAS. In the current study, we used the RAN test with pictures of highly frequent one- and two-syllable Croatian words (e.g., clock, dog, heart...) (rather than letters) to eliminate orthography from this measure.

Although the studies that have examined speech variability in CAS are limited in scope and use different methods to capture speech inconsistency, they suggest that variability may be a defining feature of the disorder.

1.4.3. Evidence of difficulties at phonetic programme assembly

There are several models of speech production that suggest that the causal factor is to be found somewhere in the transition from a phonological code to articulomotor output, i.e., phonetic planning, motor programming, or motor execution (Ozanne, 1995; van der Merwe, 1997; Velleman & Strand, 1994). In compliment to the model of Levelt and colleagues (Levelt, Roelofs, & Meyer, 1999), who proposed an articulatory network as the final level of speech production, others (e.g., Ozanne, 1995; Van der Merwe, 1997) further elaborated on this final level into two stages: motor programming and motor execution. Based on Ozanne's (1995) research, two clusters emerged reflecting a deficit in motor programming, which she subdivided into: phonetic programme assembly and motor speech programme implementation.

The underlying deficit of CAS is often thought to be at the level of phonetic programming (Schmidt & Lee, 1999; Van der Merwe, 1997), which can be summarized as the inability to translate an abstract phonological code into motor speech commands (Nijland, 2003). This means that the gesture (defined during phonetic planning) only defines the task of the articulators in an abstract way and does not delineate the exact means to accomplish this task (Nijland, 2003).

Breakdowns at the phonetic programming level include: consonant deletion (i.e., whenever a consonant is omitted in syllable-initial or syllable-final position); spontaneous production of phonemes in words they cannot imitate; use of phonemes in words that do not contain that phoneme; and groping (e.g., trial and error movements on the imitation of single sounds) (Ozanne, 1995).

Research suggests that consonant omissions may be distinctive for children with CAS (Lewis et al., 2004, Shriberg et al., 1997b). Shriberg et al. (1997b) reported that 42% of consonant errors in younger children with CAS were omissions, compared to 25% in children with speech delay. In addition, Lewis et al. (2004) found a high proportion of initial and final consonant deletion, syllable deletion, and cluster reduction in children with CAS compared to children with isolated speech disorders and children who had both speech and language disorders. Overall, 90% of the CAS group deleted syllables, compared with 8% in the isolated speech impairment group and 15% in the speech and language impairment group, respectively. Similar results were confirmed in the study by Jacks, Marquardt, and Davis (2006), in which omission errors were almost exclusively attributed to the deletion of the final consonant in words, regardless of the number of syllables, providing a consistent pattern of syllable errors. Although CAS has been extensively studied in the clinical literature, most of the evidence for impairments in syllable structure production in this population comes from English. A case study by Canault, Thai-van, and Le Normand (2021), based on a two-year longitudinal follow-up of a French boy with CAS, found similarities to previous English studies (Jacks et al., 2006; Lewis et al., 2004) in terms of poorer precision of coda-singleton consonant in coda position and a higher rate of deletions compared to onset-singleton consonant at ages 5 and 6 years.

Another breakdown of the phonetic programming level involves articulatory groping. Although groping is often associated with CAS, not all children with CAS exhibit this feature (Iuzzini-Seigel & Murray, 2017). In Lewis et al.'s (2004) study of children with CAS, speech disorder, and combined speech and language disorders, 5 out of 9 participants with CAS demonstrated groping in a preschool-age conversational sample. There was no data on participant groping at school-age follow-up. In a study by Iuzzini-Seigel et al. (2017), only 1 out of 20 children with CAS showed groping while responding to the GFTA-2, but they hypothesize that it is possible that more children would show this feature on more complex speech tasks (e.g., a polysyllable word test).

In contrast to the two aforementioned studies, Murray et al. (2015) found that articulatory groping accurately identified 54% of preschool-aged participants with CAS, whereas non-speech oral motor groping identified 29% of participants with CAS compared to participants with other speech sound disorders, emphasizing that within-speech groping is associated with CAS, whereas non-speech groping may occur due to oral apraxia (Murray et al., 2015). Iuzzini-Seigel and Murray (2017) suggest that groping should not be considered mandatory to obtain a CAS diagnosis. Furthermore, groping is based on expert judgement and does not easily meet the requirement of being operationalized, so it was not included in the present study.

1.4.4. Evidence of difficulties with motor speech programme implementation

Motor programming is the level of speech production in which phonetic plans characterizing the spatial and temporal targets of articulatory movements are translated into context-dependent motor specifications for the articulators (Nijland, Maassen & van der Meulen, 2003), which are then executed in the motor execution stage (Ozanne, 1995). A breakdown at the level of motor speech programme implementation occurs when the correct motor programme is selected but the wrong timing and force parameters are chosen (Schmidt & Lee, 1999), and it is characterised by slow diadohokinetic (DDK) rates and poor sequencing ability of DDK tasks (Ozanne, 1995).

DDK or maximum repetition rate (MRR) is a task that involves rapid repetition of syllables that are usually a part of a speech mechanism examination that is intended to evaluate oral motor skills independent of phonological abilities (Icht & Ben-David, 2021). This specific task includes successive movements (e.g., syllable to syllable) of connected speech entailing constant approximations of specific articulatory targets because no absolute or static positions are associated with speech sounds (Flipsen, Bernthal, & Bankson, 2017b). That specific overlapping movement involved in producing sequential motor speech elements presents a problem for children with CAS.

Almost 50 years ago, Yoss and Darley (1974) suggested that DDK performance was one of the factors that could distinguish motor speech disorders from other speech sound disorders. Later, Williams and Stackhouse (1998) investigated diadochokinetic abilities in three children with various SSD and paediatric motor speech disorders. One child with developmental verbal dyspraxia showed lower overall scores on two of the measures-accuracy and consistency; one child with specific phonological delay showed a very consistent DDK profile and there were no differences on the rate measures, whereas the third child showed a mixture of dysarthric and dyspraxic features on all three measures-accuracy, rate and consistency. Thoonen et al. (1999) confirmed that CAS can be diagnosed on the basis of maximum fricative prolongation, in combination with difficulty in sequencing speech movements as measured by performance on trisyllabic repetition task. Similarly, Shriberg et al. (2012) confirmed a significantly lower score on the syllable repetition task in CAS children, confirming the idea of a core impairment in motor planning or programming of speech in speakers with CAS.

Meloni et al. (2020) did a study in French-speaking children which showed that there is no significant difference between children with phonological disorders (PD) and typically developing children, but a significant difference between children with CAS and typically developing (TD) children. These results suggest that children with CAS can be distinguished from TD children on the DDK task, whereas children with PD cannot, confirming the premise that children with CAS struggle in specifying timing and force parameters of the motor plan.

In addition to the aforementioned differences, there is also acoustic evidence suggesting deficits in the temporal control of speech. It has been reported that children with CAS produce longer acoustic durations of segments (Nijland et al., 2003) and words (Bahr, 2005) than control subjects. It has been suggested that the degree of variability in the duration of pauses and speech events may distinguish children with CAS from children with other speech disorders (Shriberg et al., 2003).

Even though MRR is a highly language-neutral task there is a big variety in methods that are being used across languages and research groups (see Diepeveen et al., 2019). Therefore, the current MRR computerized protocol (Maassen et al., 2019) in this study aimed at providing an objective and comprehensive insight which can later be used for cross-linguistic comparison.

1.4.5. *Evidence of difficulties with speech execution*

In the final phase of motor execution, motor plans and programs are translated into speech movements. Nevertheless, it can be argued that the motor execution of phonemes depends on all the preceding processes and cannot be studied separately without knowing the cause of the breakdown at this level. For example, a phonemic selection error may be perceived as an execution error (e.g., stopping - production of a stop consonant instead of a fricative or affricate), whereas some phonetic distortions may result in errors that fall into a different phonological category and are perceived as phoneme substitution errors (Ziegler, 2008). Speech execution difficulties have indeed been reported multiple times in children with CAS as articulation difficulties (Ozanne, 1995; Ozanne, 2010) and as severe and persistent speech disorders that prove resistant to speech and language therapy (McNeill, 2013).

Accurate productions are defined as consonant and vowel productions that are free of errors such as distortions, omissions, and substitutions. Speech errors occur when a speaker intends to produce a word but misproduces one or more speech sounds in the word (MacKay & James, 2004). During development, children lack the articulatory control to produce phonemic distinction in the same manner as adults, however, Croatian children are expected to produce all sounds of the Croatian language correctly by the age of 5;06 (Vuletić, 1990). Although consonants are articulatorily more difficult than vowels and they are more susceptible to distortions and substitutions in TD children (Galluzzi, Bureca, Guariglia & Romani, 2015) and the accuracy of vowels reaches adult levels well before that of consonants (Pollock, 2002), children with CAS have been reported to have difficulty producing vowels and vowel errors have been documented in various auditory and acoustic studies (Lenoci et al., 2020). Furthermore, Pollock and Hall (1991) showed that children with CAS have problems with rhoticised vowels and diphthongs. Similar results were obtained in a longitudinal study by Davis, Jacks, and Marquardt (2005), which showed that children with CAS were able to produce almost all vowels but had a high number of vowel errors, resulting in much lower accuracy levels than expected for their age. In addition, speakers with CAS also showed smaller vowel spaces and less pronounced vowel contrasts, both in articulation and acoustics, compared to matched controls (Lenoci et al., 2020).

Regardless of the cause, research has shown that children with SSD (including CAS) have less mature speech motor patterns compared to their typically developing (TD) peers (Case & Grigos, 2018). For example, in his study, Terband (2011) showed an increase in speech sound distortion (as well as coarticulation, searching articulatory behaviour, and variability) with increasing dependence on feedback control in a series of computer simulations, which could result from increased reliance on auditory feedback control due to incorrect and/or imprecise feedforward commands. In addition, a study by Case and Grigos (2018) suggests that, while children with CAS show improvements in accuracy of speech production (percentage of consonants correct - PCC and percentage of vowels correct - PVC), they may require more intensive practice over time to facilitate changes in articulatory movement, which could explain why children with CAS have difficulty retaining newly acquired speech targets and generalizing what they learn in untreated contexts.

This lack of agreement on the cause of production errors in CAS affects our ability to specify the phenomenology of CAS and the theoretical implications of production errors (i.e., where the breakdown occurs in the speech production process).

1.4.6. Conclusions

All the studies discussed in this section confirm the initial statement that CAS is indeed a highly controversial and complex entity. Although various psycholinguistic theories attempt to isolate the processing steps involved in speech production, these results are puzzling and make it difficult to determine the underlying deficit. The processing levels at which deficits have been identified comprise the entire speech production chain, from the highest levels such as lexical storage and retrieval to the lowest level (i.e., speech execution). However, there are only few studies in which psycholinguistic models have been systematically applied to apraxia of speech (Ziegler 2005) or CAS (Maassen et al. 2010; Nijland, 2009; Maassen & Terband, 2015), which weakens the theoretical basis for delineating the core processing deficit as well as the clinical basis for differentiating CAS from other speech sound disorders. In addition, the sample sizes in these studies were predominantly small, usually up to 15 children of varying ages. Furthermore, the classification criteria for a number of these studies do not include a detailed classification protocol, particularly with regard to receptive language measures, making it difficult to generalize conclusions regarding CAS.

2. PRESENT STUDY

The development of the language, cognitive, and neurological systems complicates our understanding of the causes of speech disorders in children. This developmental interaction between different levels of cognitive processing presents a fundamental challenge in isolating the underlying deficit of developmental disorders (Maassen & Terband, 2015).

Although CAS is primarily defined as a motor disorder, recent research suggests that it is a more complex disorder that includes language impairment (i.e., phonological deficits). Despite the dispute over pathognomic features, there is more or less agreement on the core features of CAS (i.e., the "golden standard" proposed by ASHA, 2007). These features are consistent with a deficit at the level of planning and programming speech sounds; however, CAS has been associated with a variety of features and has been shown to involve different difficulties during successive stages of development (Terband, 2011; Maassen, 2002).

Explanations for CAS range from a disturbance localized at the level of phonological representation to those at the level of motor programming and execution. In addition, not all children exhibit the same symptoms, and symptoms change over time, so a checklist approach to CAS is not an ideal solution. This study follows the premise of Maassen and Terband (2015) by constructing a test battery with a set of speech tasks that represent different steps in the speech production process (i.e., with reference to the Cascade model; Ozanne, 1995) using current technological aids. By comparing speech performances obtained with such a test battery, our aim is to determine which processing steps are functioning normally and which are impaired. The current study was limited to comparing the speech production of children with CAS with typically developing children (this study did not include differential diagnoses with other speech sound disorders), while controlling for receptive language impairments.

Considering that most of the data on CAS come from English-speaking children, it is difficult to determine which of these features are relevant in other languages, as there are differences between the various phonological systems, especially in terms of the inventory of consonants and vowels. Therefore, one of the goals of the current study was to provide data on CAS in languages other than English.

The present study does not question the presence of speech motor difficulties in children with CAS, but rather aims to further investigate speech motor abilities as well as phonological abilities, taking into account the involvement of both speech motor production and speech perception processes, and to distinguish subgroups of children with CAS based on these distinct processing levels in order to gain insight into the processes underlying speech production difficulties in children with CAS.

2.1. Questions

Given the complex developmental interactions between processing levels and in order to understand the core of the underlying deficit(s) in CAS, the following questions are posed:

1. Do children with CAS differ compared to TD children at all levels of speech production? In particular, do these two groups differ in terms of:

   1.1. Phonological rules; i.e., is there a significant difference in the AX discrimination task and phonological awareness tasks (rhyme recognition; rhyme production; syllable synthesis; syllable analysis; phonemic synthesis; phonemic analysis)?

   1.2. Assembling the phonological plan for the word or utterance; i.e., is there a significant difference in the RAN test, the word repetition task, the non-word repetition task and in the word learning task?

   1.3. Phonetic programme assembly; i.e., is there a significant difference on errors in syllable-initial consonants (PCCI), in syllable-final consonants (PCCF) and consonant deletion.

   1.4. Implementation of the motor-speech programme; i.e., is there a significant difference in the MRR tasks (monosyllabic, bisyllabic, and trisyllabic repetition)?

   1.5. Speech execution, i.e., is there a significant difference in the accuracy of consonant (PCC) and vowel (PVC) production?

2. Is there an effect of motor speech production on phonological tasks, i.e., do children with CAS perform worse on phonological awareness tasks that require motor speech production (verbal response) than on tasks that do not require motor speech production (nonverbal response)?

3. Is it possible to identify a subgroup of children with CAS considering higher levels of speech production (phonological abilities)?

2.2. Hypotheses

H1: Children with CAS perform worse at all levels of speech production.

H1.1: Children with CAS perform worse at the phonological rules level, i.e., there is a significant difference in the AX discrimination task and the phonological awareness tasks.

H1.2: Children with CAS perform worse at the level of assembling the phonological plan for the word or utterance; i.e., there is a significant difference in the RAN test, the word repetition task, the non-word repetition task, and the novel word learning task.

H1.3: Children with CAS perform worse at the level of phonetic programme assembly, i.e., there is a significant difference in the correctly produced consonants in syllable-final position (PCCF) and syllable-initial positions (PCCI) as well as in the number of omitted consonants (PCnD).

H1.4. Children with CAS perform worse at the level of implementation of the motor-speech programme, i.e., there is a significant difference in the maximum repetition rate (MMR), in monosyllabic, bisyllabic, and trisyllabic repetition.

H1.5: Children with CAS perform worse at the level of speech execution; i.e., there is a significant difference in the accuracy of consonant (PCC) and vowel (PVC) production.

H2: Children with CAS perform worse on phonological tasks requiring motor speech production (verbal response) than on phonological tasks not requiring motor speech production (nonverbal response), i.e., there is an interaction between motor speech production and phonological tasks.

H3: By identifying difficulties at the phonological levels of speech production, subgroup of children with CAS is formed.

3. METHOD

3.1 Participants

This study included a total of 58 participants: children with childhood apraxia of speech (n=30) and typically developing children (n=28). After being informed about the study, parents signed a written informed consent form emphasizing that participation was voluntary and that children could withdraw from the study at any time. In addition, parents could ask for information at any time during data collection by e-mail or telephone. Ethical approval for this study was obtained from the Ethical Review Board of the Faculty of Education and Rehabilitation Sciences.

The age of the participants ranged from 64 to 91 months (5;04 to 7;07 years). The mean age of the participants was 76.17 months (6;04 years). The two groups didn't differ statistically in age, and there were no gender differences in age. However, the gender pattern shows that males predominated in the CAS group (Table 1).

Table 1. Participant's gender structure

		TD group		**CAS group**		**Total**
		f	%	*f*	%	
Gender	Male	15	36.6	26	63.4	**41**
	Female	13	76.5	4	23.6	**17**
	Total	**28**	**100.0**	**30**	**100.0**	**58**

3.1.1. Typically developing participants (TD)

Typically developing children (TD) were recruited from two mainstream kindergartens in Zagreb. In addition, they were recruited by two speech-language pathologists working in these kindergartens. The children had no history of speech and language disorders, motor or cognitive impairments. All children had normal or corrected vision and normal hearing (based on the results of neonatal screening). Only children with the results within the normal range on Raven's Colour Progressive Matrices Test (CPM; Raven, 1999) and on the Test for Reception of Grammar (TROG-2:HR; Bishop, Kuvač Kraljević et al., 2014) participated in the study (SR ≥85). All children were monolingual speakers of Croatian.

3.1.2. Participants with childhood apraxia of speech (CAS)

Participants with CAS were recruited via letters to the speech and language therapist of the Croatian Logopedics Association (kindergarten section and private practice section) or referred directly to the Centre for Rehabilitation, the clinical unit of the Faculty of Education and Rehabilitation Sciences. Participants with CAS were recruited mainly in Zagreb and its surroundings, but there were also children from Zadar, Trogir and Split. The tests were conducted in the Centre for Rehabilitation of the Faculty of Education and Rehabilitation Sciences or in the kindergartens. All tests took place in quiet rooms in one of the above mentioned institutions, with each participant sitting next to the researcher.

Exclusion criteria for participants were: neurological or physical cause of the speech sound disorder (e.g., cleft palate), motor impairment, cognitive impairment, hearing impairment, and inadequate receptive language skills. Motor abilities was tested trough Croatian version of The Developmental Coordination Disorder Questionnaire (DCDQ-HR; Sangster Jokić, Knežević & Wilson, in preparation). The Raven's Colour Progressive Matrices Test (Raven, 1999) was used to assess nonverbal cognitive abilities. Only children whose scores were within the normal range (SR ≥85) participated in the study. To assess receptive language, the Test for Reception of Grammar (TROG -2: HR; Bishop, Kuvač Kraljević et al., 2014) was used. Only children whose scores were in the normal range (receptive language scores above -1.25 SD at TROG - 2) participated in the study. All parents completed a case history form (Appendix A). All children regularly attended kindergarten and one child attended first grade, and they were all monolingual Croatian speakers.

All participants with CAS had a previous CAS diagnosis or were suspected of having CAS. For the purposes of this study, the diagnostic classification for CAS was additionally based on the presence of the three in ASHA (2007) (1) inconsistent consonant and vowel errors, (2) difficulty forming articulatory transitions between sounds and syllables, and (3) prosodic errors. The diagnostic classification for CAS was determined as proposed by Grigos, Moss, and Lu (2015) by the researcher and one experienced SPL. Children diagnosed with CAS showed these features in more than one speech context and within at least three different words (or sounds/syllables in the sequencing task): inconsistent errors were defined as consonant and vowel errors that differed in repeated productions of the same word (e.g., /*svjetiljka*/ Eng. /*lamp*/ produced as /*sletika*/, /*sletilka*/, /*sletitka*/ by the same speaker; difficulties in articulatory transitions were characterized by poor coarticulation, especially when it included phonemes that were present in the child's repertoire (e.g., difficulty in combining the consonant /s/ with different vowels, even though /s/ and the vowels can be produced correctly in other contexts); prosodic errors were identified as incorrect lexical and/or phrasal stress. In addition to the three core features, children with CAS exhibited at least four of the following features throughout different tasks: metathesis (e.g., production of /poklopac/ Eng. /lid/ as /klopopac/), vowel errors (e.g., production of /poklopac/ as /paklopac/), voicing errors (e.g., voiceless sound is replaced by a voiced sound), phoneme distortions, articulatory groping (e.g.., visual struggle accompanying phoneme production), reduced phonetic inventory, and poorer expressive than receptive language skills.

At the time of the study, all children with CAS were undergoing speech and language therapy (at least one year), and none of them had received occupational therapy or physical therapy.

3.2. MATERIALS

For each participant, the procedure lasted between 90 and 100 minutes in total. The first part (tests for exclusion criteria) took between 20 and 30 minutes. A series of tasks representing different steps in the speech production process lasted between 45 and 60 minutes with short breaks (5 minutes) between tasks, depending on the needs of the participants.

3.2.1. Tests for exclusion criteria

Raven's Coloured Progressive Matrices (CPM; Raven, 1999) are suitable for testing general cognitive abilities from the age of 5 to 11 years. This test measures the ability to make perceptive and logical conclusions and offers insight into perceptual, problem solving and learning abilities. The test consists of 12 tasks grouped in 3 sets (36 in total). On each task the participant is presented with a coloured drawing in which one section has been left blank. The participant must choose the image that best fills the white frame in the main drawing. The testing was conducted by a psychologist.

The Test for Reception of Grammar (TROG-2:HR; Bishop, Kuvač Kraljević et al., 2014) was used for assessing receptive language of Croatian grammar with respect to inflexion, functional words, and word order. The TROG-2 consists of 80 items made up of sentences of varying complexity that are ordered from lower to higher complexity. The participant must select one of four images that corresponds to the sentence presented in the item. The testing was conducted by an SLP.

Motor abilities was tested trough Croatian version of The Developmental Coordination Disorder Questionnaire (DCDQ-HR; Sangster Jokić, Knežević & Wilson, in preparation). DCDQ (Wilson, Kaplan, Crawford & Roberts, 2007) is a questionnaire developed to identify subtle motor problems in children aged 5-15 years. Caregivers assess their child’s coordination in comparison with other children of the same age and rate it on a 5-point Likert scale. This questionnaire represents a standardized method for measuring a child's coordination in daily, functional activities. It assembled out of 15 items that examine: control during movement, fine motor and handwriting skills, and general coordination. The Cronbach’s alpha coefficient for the total test is .88. The DCDQ was also significantly correlated with the total impairment score of the Movement ABC ($r = -.59$, $p < .0001$) (Wilson, Kaplan, Crawford, Campbell & Dewey, 2000).

3.2.2. Materials and tasks representing different levels of speech production

The following section explains the tasks and materials used in the mail part of the study.

Table 2. Measured constructs, variables, and materials/tasks used in the present study

Measured constructs	Measured variables	Materials & tasks	Verbal response
Phonological rules	Phonological representations	AX discrimination task	✖
	Phonological awareness	Rhyme recognition (predČiP)	✖
		Rhyme production (predČiP)	✔
		Syllables blending (predČiP)	✖
		Syllables segmentation (predČiP)	✔
		Phonemic blending (predČiP)	✖
		Phonemic segmentation (predČiP)	✔
Phonological planning	Retrieval of phonological information	RAN test (predČiP)	✔
	Whole-word variability	Word repetition task (CAI)	✔
	Whole-word variability	Non-word repetition task (CAI)	✔
	Novel word consistency and correctness	Novel word learning task	✔
Phonetic programme assembly	Consonant deletion/substitution	PCCI PCCF PCnD	✔
Motor speech programme implementation	Maximum repetition rate	Monosyllabic repetition (CAI)	✔
		Bisyllabic repetition (CAI)	✔
		Trisyllabic repetition (CAI)	✔
Speech execution	Accurate phoneme productions	PCC PVC	✔

3.2.3. AX discrimination task

To reduce the load on auditory memory, the AX discrimination task was adopted for the present study. This discrimination task requires phonemic judgment based primarily on phonetic properties and features. It required a response of "same" or "different" on each presented trial. Participants were presented with two consecutive speech tokens (vowels). Vowels were used because they appear to be more sensitive than consonants in terms of auditory-phonetic discrimination (Maassen et al., 2009). Speech tokens were synthesized considering the F1 frequency of vowels in the Croatian language (Table 3; Škarić, 1991). The task was conducted in E-Prime, a stimulus presentation software that recorded both accuracy and total reaction time.

Table 3. F1 frequencies for vowels in Croatian language (Škarić, 1991, p. 186)

	i	e	a	o	u
F1	360	500	700	450	380
F2	2200	1800	1400	1150	750

The participant had to listen to each pair and select the ✔ key on the keyboard if the presented vowels were „ the same“, and ✖ key on the keyboard if the vowels were „different“. There were a total of 16 trials (Table 4). Vowels are usually distinguished by vowel height and vowel backness. Vowel height refers to the vertical position of the tongue (low-1, middle-2, high-3), and vowel backness refers to the position of the tongue relative to the back of the mouth (front-1, middle-2, back-3).

Table 4. Vowel pairs in the AX discrimination task

Trial	Stimuli 1	Stimuli 2	Vowel height	Vowel backness
1	A	A	11	22
2	A	E	12	21
3	E	E	22	11
4	E	I	23	11
5	I	I	33	11
6	I	A	31	12
7	O	O	22	33
8	O	E	22	31

9	U	U	33	33
10	U	A	31	32
11	A	O	13	23
12	I	U	33	13
13	O	I	23	31
14	O	U	23	33
15	E	U	23	13
16	O	A	21	32

All pairs were randomly ordered with a duration of 1000 ms, with the interpair interval of 5000 ms. Prior to the task, there was a preparatory section in which participants had to complete 5 trials to familiarise themselves with the procedure and to ensure that they had fully understood the task. Participants were motivated by the small robot that appeared on the screen, as if it was producing the synthesised vowels. At the end, a winner's trophy appeared on the screen as a reward for helping the robot. Participants never received any differential feedback for their responses, only verbal reinforcement ("*You are doing great*," "*Well done*"). Correctness and reaction time from onset to last response were calculated for each participant for both conditions (vowel height and backness).

3.2.4. Test for the Assessment of Reading and Writing Prerequisites (PredČiP)

Phonological awareness is the ability to consciously think about a spoken word in terms of its basic phonological units - syllable, rhyme, and phoneme (Ouellette & Haley, 2013). In the present study, participants were assessed with the Test for the Assessment of Reading and Writing Prerequisites (PredČiP; Kuvač Kraljević & Lenček, 2012), a standardized test of pre-reading skills and abilities. It consists of language tasks (rapid automatized naming, phonological awareness tasks, letter knowledge, and narration) and visual perception tasks (recognition and copying). The PredČiP tasks used in the study relate to phonological awareness. Syllables and phonemic blending require a verbal response in this test, but for the purposes of this study, verbal response was excluded in these particular tasks and the task was presented differently from its original form. Each item received a binary score, with "1" representing correct responses and "0" representing incorrect responses.

3.2.4.1. Rhyme recognition

In the rhyme recognition task, participants were presented with two words. They had to recognise whether two words rhymed with a yes or a no, verbally or nonverbally (e.g. *seka-teka* - correct answer: yes, or a nod of the head). There were seven pairs, and for each correct answer the participant received one point.

3.2.4.2. Syllable blending

In the syllable blending task, the ability to blend syllables to form words was tested using seven words (as in the predČiP test), but without the answer having to be given verbally. The participant was presented with the syllabically segmented version of the word (*ka-men*, Eng. *stone*) and had to point to the correct answer. The participant was presented with four responses: correct answer (*kamen*), phonological distractor (*kamin*, Eng. *fireplace*); semantic distractor (*drvo*, Eng. *tree*); unrelated distractor (*olovka*, Eng. *pencil*) (Figure 3). The list of words consists of four two-syllabic, two three-syllabic, and one four-syllabic word. All graphics were created by the researcher in Canva, a graphic design platform.

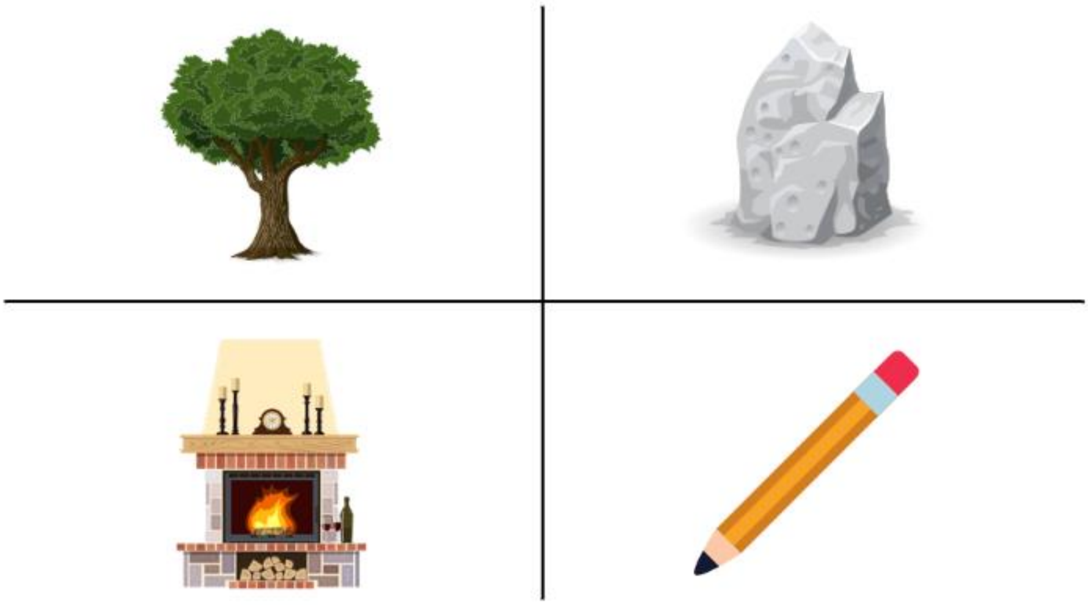


Figure 3. Example of the non-verbal syllable blending task

3.2.4.3. Phonemic blending

In phonemic blending task, the ability to blend phonemes to form words was tested using seven words (as in the predČiP task), but without the answer having to be given verbally. The participant was presented with the phonemically segmented version of the word (*k-o-l-a-č*, Eng. *cake*) and had to point to the correct answer. The participant was presented with four responses: correct answer (*kolač*), phonological distractor (*kotač*, Eng. *wheel*); semantic distractor (*sladoled*, Eng. *ice-cream*); unrelated distractor (*sunce*, Eng. *sun*) (Figure 4). The list of words consists of seven words, ranging from simple CVC words with three phonemes to more complex CCCVCV words with six phonemes. All graphics were created by the researcher in Canva, a graphic design platform.

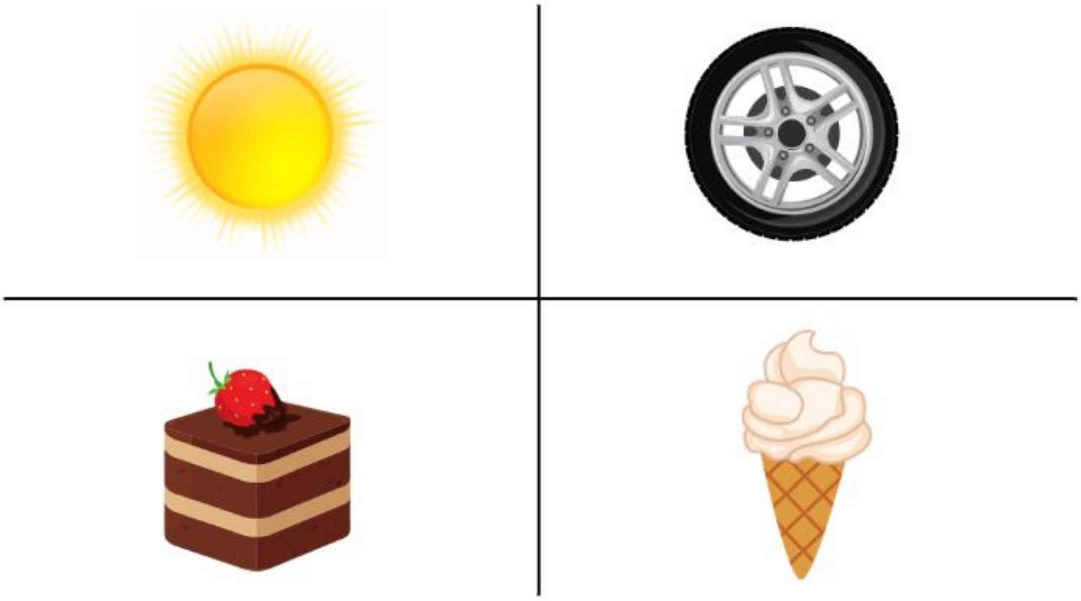


Figure 4. Example of the non-verbal phonemic blending task

3.2.4.4. Rhyme production

In the rhyme production task, participants were asked to produce words that rhyme with the presented word (seven in total) regardless of their meaning. For example, participants were presented with the word *most* (Croatian for *bridge*) and could give several correct answers, such as *kost* (Croatian for *bone*) or *post* (Croatian for *fasting*), all of which rhyme with *most*. However, they could also answer *bost*, which is a correct answer, although it has no meaning in Croatian. If the child consistently omitted, substituted, or distorted certain sounds, he or she was not penalized for doing so. For example, if the participant substitutes /r/ with /j/ and answers *kjv* (instead of *krv*, meaning blood) for the presented word *crv* (Eng. *worm*), it was accepted as a correct answer.

3.2.4.5. Syllable segmentation

In syllable-related tasks representing shallow phonological awareness (Kuvač Kraljević, Lenček & Matešić, 2019), participants were asked to segment seven words into syllables. For example, participants were given the word *kugla* (Eng. *sphere*) and had to segment it into syllables (*ku* - *gla*). The list of words consists of four two-syllabic, two three-syllabic, and one four-syllabic word. Similar to the rhyme production task, the child was not penalized for continuously making omissions, substitutions, or distortions of specific sounds.

3.2.4.6. Phonemic segmentation

In phonemic related tasks, representing deep phonological awareness (Kuvač Kraljević et al., 2020), participants were asked to segment seven words into phonemes. For example, participants were given a word *vuk* (Eng. woolf) and they had to segment it into syllables (*v-u-k*). The answer was accepted as correct if they produced all segments of the word presented. The list of words consists of seven words, ranging from simple CVC words with three phonemes to more complex CCCVCV words with six phonemes. Similar to the rhyme production and syllable segmentation task, the child was not penalized for consistently making omissions, substitutions, or distortions of certain sounds.

3.2.4.7. Rapid automatized naming (RAN)

Wolf et al. (2000) refer to rapid naming as a combination of different subprocesses that begins with attentional, perceptual, and memory processes, then moves into conceptual, phonological, and semantic representations, and ends with motor subprocesses, each of the components having precise timing. In this study, Rapid automatized naming (RAN; Kuvač Kraljević & Lenček, 2012) was conducted to assess lexical retrieval and speed of processing phonological information. The test consists of 20 items (drawings) representing frequent and well known one- and two-syllable Croatian words (e.g., clock, dog, heart...). Before starting the test, the participant was asked to name all items to ensure that all items were known to the participant. After timing each participant, the number of items per second was calculated.

3.2.5. The Computer Articulation Instrument (CAI)

To gain insight into the underlying causes of the child's difficulties, it is necessary to conduct a comprehensive analysis of the child's performance on a range of speech tasks reflecting different levels of processing (van Haaften et al. 2019a). Based on these premises, the Computer Articulation Instrument (CAI) was developed (Maassen et al., 2019). CAI is a computer-based speech-production test developed by Maassen et al. (2019) in the Netherlands. Its psychometric properties indicate that CAI is a reliable and valid instrument for assessing typical and delayed speech development in Dutch children (van Haaften et al., 2019a). It is modular in design and requires interactive administration. Tasks targeting phonological and speech motor skills in children aged 2-7 years include (a) picture naming, (b) non-word imitation, (c) word and non-word repetition, and (d) maximum repetition rate (MRR).

CAI captures the entire chain of speech processes (see Figure 5), from preverbal visual-conceptual processing through lemma access, word form selection, phonological encoding, motor planning, and articulation (motor execution) (van Haaften et al., 2019a). The evaluation of speech production in CAI is based on phonetic transcriptions and acoustic measurements, both computer-implemented. With the author's permission, the word imitation, non-word imitation, and MRR tasks have been translated into Croatian and implemented in CAI.

The assessment was administered using a laptop and a headset with a microphone in a quiet room (or a room with as little background noise as possible) to ensure a good sound level. The acoustic signal was automatically stored on the computer's hard drive in one recording for each of the different tasks. Before each task, the participant was asked to make sure that both the playing and the recording of the sound were working correctly.

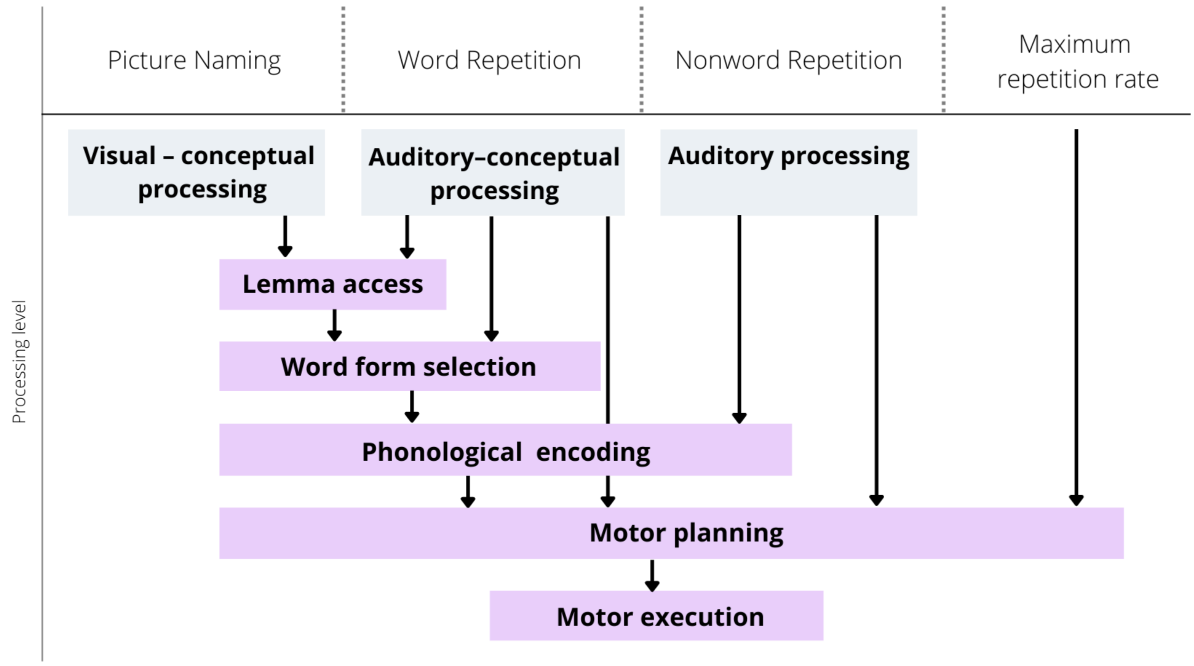


Figure 5. The speech production processes assessed by The Computer Articulation Instrument (based on Maassen & Terband, 2015, Figure 15.2).

3.2.5.1. Word repetition task

The word repetition task aims to assess the variability in speech production that occurs when a child uses multiple productions of the same word (van Haaften et al. 2019b). As explained by Ingram and Ingram (2001), the most efficient way to measure whole-word variability is to elicit a pre-set number of productions for a pre-selected set of words. In this task, a participant is asked to repeat a given word 5 times without visual support. There were 5 blank dots on the screen that turned white as soon as the participant produced the item to help him count the number of repetitions. In the Dutch version, the word and non-word conditions contained three two-syllable and two three-syllable items with equal, complex consonant structures (CVC-CCVC, CCVC-CVC, CVCC-CCVCC, CVC-CV-CCV, CV-CV-CVC). To follow the premise of complex consonant structure for the Croatian version, less frequent syllable structures were chosen (Table 5). The selected consonant structures are based on the research of Kelić (2017), who conducted corpus analyses on the frequency of syllable structure in Croatian.

Table 5. Frequency of Croatian syllabic structures in corpus samples, Kelić (2017)

	Structure:	Frequency:
Frequent	CV	58.34%
	CCV	15.2%
Less frequent	V	10.30%
	CVC	8.75%
	CCVC	2.65%
	VC	1.46%
	CCCV	1.06%
	CVCC	0.33%
	CCCVC	0.18%
	CCVCC	0.14%
	VCC	0.07%
	CVCCC	0.02%

Two-syllable items were formed from two less frequent syllables, while the three-syllable items were formed from two less frequent syllables and one frequent syllable (Table 6). The items were recorded by a trained female native Croatian speaker in the Acoustics laboratory of the Department of Speech and Language Pathology. They were digitized at a sampling rate of 44 kHz and a resolution of 16 bits.

Table 6. The list of words in the Croatian version of the CAI word and non-word task

Word structure (Croatian)	Words	Non-words
CVC-CVC	CIRKUS (Eng. *circus*)	VAJLON
CCVC-CVC	KRUMPIR (Eng. *potato*)	BRUZLJEN
V-CCVC	OBLAK (Eng. *cloud*)	OKRUM
CCCV-CVC-CV	SVJETILJKA (Eng. *lamp*)	STRUMARDA
CCVC-VC-CV	KLIZALJKE (Eng. *ice-skates*)	BRIZONGE

In the word repetition task, the number of different word forms was determined after each session. A production was identified as "different" if at least one phoneme of the target word was replaced, deleted, or added. For example, the words /KRUMPI/ and /KRUMPIN/ are two forms of the target word /KRUMPIR/, neither of which is correct, but this was not measured in this task. Consistency was calculated for each target word by dividing the number of repetitions (5) minus the number of word forms by 5 (representing the proportion of whole-word variability).

3.2.5.2. Non-word repetition

Unlike picture naming and word repetition, a child cannot rely on his or her lexicon in this task and therefore either analyses the phonological structure of the non-word directly or follows the auditory-motor planning pathway (van Haaften et al. 2019b). Similar to the word repetition task, the child is asked to repeat a presented non-word five times, but this time with visual support. Each non-word is accompanied by coloured pictures of "nonsense characters" (Figure 6) displayed on the computer screen to make the task more attractive, especially for younger children.

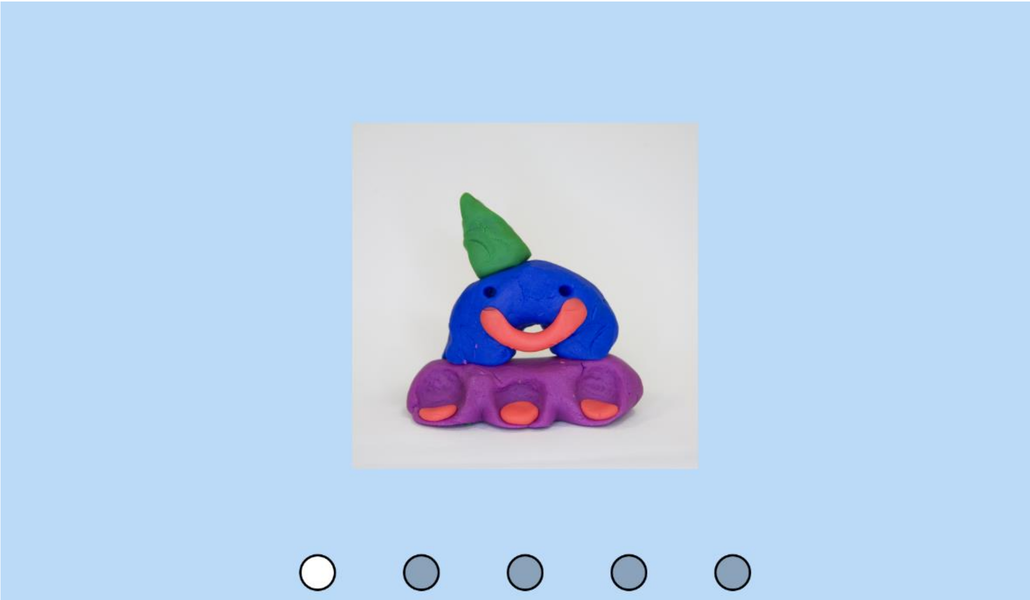


Figure 6. An example of visual support used for the non-word imitation task

The selected non-word items have the same structure as the items in the word repetition task (Table 6). In selecting the non-word items, a small pilot study was conducted to control for wordlikeness (i.e., similarity to actual words in the listener's native language). Wordlikeness has a strong influence on how quickly and accurately non-words are processed (Frisch, Large & Pisoni, 2000). In the preparatory study, first-year students of speech and language pathology at the University of Zagreb (N=55) had to rate the presented non-words on a scale from 1 (this word could never be a Croatian word) to 5 (this word could absolutely be a Croatian word). The words were presented in written form via Google forms. Only items with ratings from 1 to 3 were selected for the non-word task. The items were then recorded by a trained female native Croatian speaker in the acoustics laboratory of the Department of Speech and Language Pathology. They were digitized at a sampling rate of 44 kHz and a resolution of 16 bits. As in the word repetition task, the number of different non-word forms was determined to be "different" if at least one of the phonemes of the target word was produced differently or deleted.

3.2.5.3. Maximum repetition rate (MRR)

Diadochokinesis (DDK) is one of the few objective assessments of motor speech performance. DDK is considered a particularly sensitive index of motor speech impairment because it requires maximum performance (Ziegler, 2002), which is why it is often referred to as maximum repetition rate (MRR). In the CAI instrument it is referred to as MRR.

MRR is a task that includes the repetition of syllables composed of a consonant and a vowel as quickly as possible in a clear manner (Diepeveen et al, 2019). MRR tasks are purely motor tasks, and do not require linguistic knowledge (Maassen & Terband, 2015). It has two components: alternating motion rates (AMR) - repetition of monosyllabic sequences (/pa/, /ta/, /ka/) and sequential motion rates (SMR) - repetition of multisyllabic sequences (/pataka/). These stop consonants /p/, /t/, and /k/ are the consonants that occur in most languages of the world (Schwartz, Boë, Badin & Sawallis 2012) and are among the first acquired consonants (McLeod & Crowe, 2018). These consonants, in combination with a vowel, represent different places of articulation, so MRR protocols typically have multiple components that increase in complexity (Diepeveen et al., 2019).

MRR performances were elicited via a computerized protocol consisting of spoken instructions and examples starting from monosyllabic sequences and ranging from normal to faster speech rates. All instructions were translated from Dutch into Croatian and recorded by a trained female native Croatian speaker in the acoustics laboratory of the Department of Speech and Language Pathology. They were digitized at a sampling rate of 44 kHz and a resolution of 16 bits.

The participant was first given an instruction (“*you are now going to hear a strange word”, “you have to repeat it exactly as you hear it”*) and then an audio sample (Table 7). For the last two instructions, participants were asked to produce the items faster. The first of those two instruction was: "*Good job! Can you do it faster?*" and it included an audio sample (12 syllables at a faster rate). The second instruction asked children to produce the syllable sequences *as fast as possible* without an audio sample.

Table 7. Assessment protocol for the MRR for mono-, bi-, and trisyllabic sequences (based on Diepeveen et al., 2019, p.241, Figure 15.2)

Sequence	Audio target	Speech rate
pa 3x	Yes	Normal speech rate
pa 6×	Yes	Normal speech rate
pa 12x	Yes	Faster speech rate
pa ≥9×	No	As fast as possible
ta 3x	Yes	Normal speech rate
ta 6×	Yes	Normal speech rate
ta 12x	Yes	Faster speech rate
ta ≥9×	No	As fast as possible
ka 3x	Yes	Normal speech rate
ka 6×	Yes	Normal speech rate
ka 12x	Yes	Faster speech rate
ka ≥9×	No	As fast as possible
pataka 3x	Yes	Normal speech rate
pataka 6×	Yes	Normal speech rate
pataka 12x	Yes	Faster speech rate
pataka ≥9×	No	As fast as possible
pata 3x	Yes	Normal speech rate
pata 6×	Yes	Normal speech rate
pata 12x	Yes	Faster speech rate
pata ≥9×	No	As fast as possible
taka 3x	Yes	Normal speech rate
taka 6×	Yes	Normal speech rate
taka 12x	Yes	Faster speech rate
taka ≥9×	No	As fast as possible

Only the last two sequences of the MRR task were analysed (those containing the instructions "*faster*" and "*as fast as possible*"). The first and last syllables were excluded from the analysis, and only sequences with a remaining minimum of 3 syllables were included in the analysis (Diepeveen et al., 2019). As explained by Diepeveen et al. (2019) first and the last syllables were excluded because speakers often produce the first syllable with a longer duration and higher intensity, and the last syllable is often lengthened. The fastest correctly produced syllable sequence is used for further analysis. All productions were analysed in the CAI adapted Praat script (Figure 7). Before extracting the syllable durations and MRR, the marked syllable onsets were depicted in the waveform and inspected visually. MRR was calculated by dividing the total number of syllables by the duration of the trial; it is expressed in syllables per second.

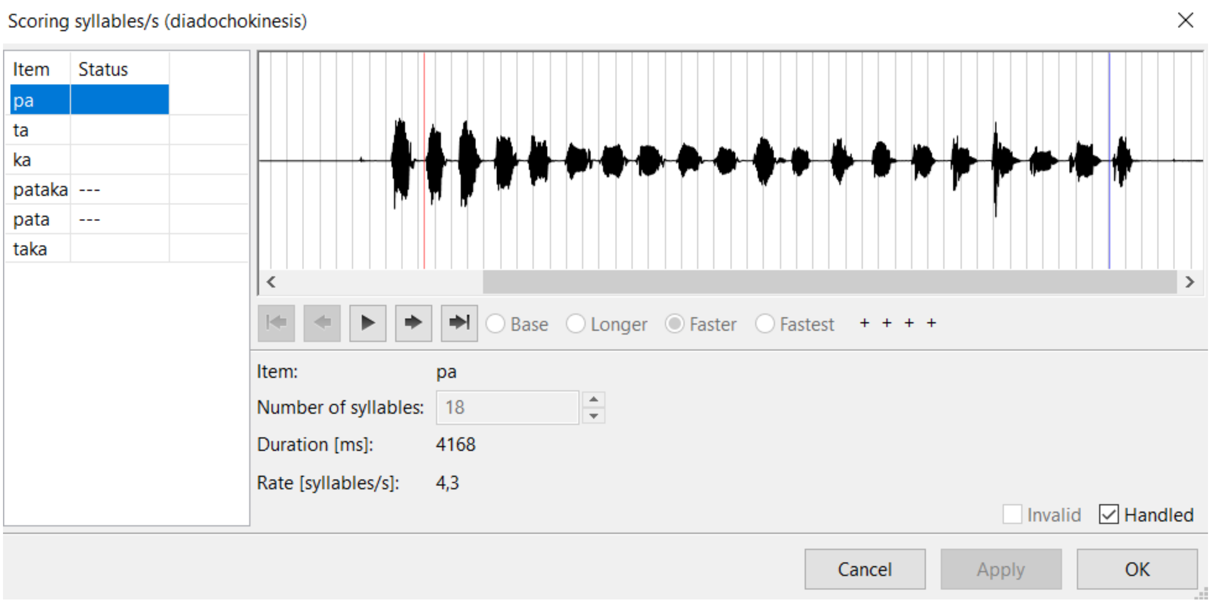


Figure 7. Example of the analysis with the CAI instrument for sequence /pa/, faster attempt

A sequence is marked as fail (0) if the child refused to complete the sequence, if not enough syllables were detected (minimum of 3), if an irregular rhythm (distinct pause) was executed, or if the child made errors (e.g., /pakata/ in- stead of /pataka/). All the further details of the standardized protocol for MRR assessment in young children in Dutch was presented by (Diepeveen et al., 2019).

3.2.6. Novel word learning task

A word learning task assesses children's ability to create and execute motor plans for nonsense words (Dodd et al., 2010). This task is an adaptation of Bradford and Dodd's (1996) and Dodd et al.'s (2010) novel word learning tasks.

Learning a word means having a well-formed phonological representation after repeated auditory exposure and being able to produce the word correctly and not confuse it with a similar sounding word (Alt et. al, 2019). The cognitive load in this task is higher because the child must store information while engaging in other cognitively demanding activities and then create, sort, and select the correct representation and create a phonetic program for a given word (Alt et al., 2019). Therefore, the phonological component of word learning may be the most prone to error.

An appealingly illustrated story of "The Three Little Pigs" was used in this study. Each pig was given a two-syllable nonsense name (CVCV) following the phonotactics of the Croatian language (Table 8), using different consonants (taking into account the manner and place of articulation). In the selected book, there are visually different little pigs depicted three times before the story and twelve times during the story.

Table 8. Two-syllabic nonsense names in the novel word task – Three little pigs

		1st little pig	**2nd little pig**	**3rd little pig**
New name		**Cuni**	**Lugo**	**Žapo**
Syllabic structure		**CVCV**	**CVCV**	**CVCV**
Number of mentions in the instruction		3	3	3
Number of mentions in the story		12	12	12
Manner of articulation:	Stop		✔	✔
	Fricative			✔
	Affricate	✔		
	Nasal	✔		
	Lateral		✔	
	Glide			
Place of articulation:	Bilabial			✔
	Labiodental			
	Dental	✔		
	Alveolar	✔	✔	
	Palatal			✔
	Velar		✔	

The story was recorded in a storytelling manner by a trained actress, a native speaker of Croatian, in the Acoustics Laboratory of the Department of Speech and Language Pathology. These recordings were converted into Microsoft PowerPoint along with the book illustrations (Figure 8). Children's comprehension of the names was tested by asking them to point to the named characters after receiving the instruction:

*"You will hear a story about three little pigs. You have probably heard this story before. This time your task is to remember their names. I will ask you their names after the end of the story. Here they are: Cuni, Lugo and Žapo (the researcher points to them while naming them). Cuni is the oldest and the smartest brother, Lugo is the one with the blue shirt and Žapo is the one with the yellow one. Can you show me Cuni? Lugo? Žapo? (*the researcher makes sure that the participants can distinguish them*). I will ask you their names after the story is finished."*

Figure 8. Microsoft PowerPoint outlook of the Three little pigs

After the story ended, the production of each test word was elicited five times. Each participant was asked to answer the questions (Table 8). If there was no response, participants were prompted with the first syllable (CU); if there was still no response, they were given the whole test word (CUNI). All responses were recorded and later transcribed.

Table 9. Eliciting questions for the novel word learning task

Eliciting question	**Answer**
Can you tell me the names of the three little pigs?	Cuni, Lugo Žapo
What is the name of the oldest brother who is very hardworking?	Cuni
What are the names of the little pigs who were having fun all day long?	Lugo, Žapo
Who built a house out of straw?	Lugo
Who built a house out of sticks?	Žapo
Who built a house out of rocks?	Cuni
Who had to run away from the wolf because he blew away his house? (And who else)	Lugo, Žapo
Who built such a strong house that wolf couldn't blow away?	Cuni
At the end of the story, who was jumping with happiness and laughing to wolf?	Cuni, Lugo, Žapo

Consistency and correctness were calculated for each participant. The production was identified as "different" if at least one phoneme of the target word was substituted or deleted. Consistency was calculated for each target word by dividing the number of repetitions minus the number of word forms by five (which corresponds to the proportion of whole-word variability), as in the word and non-word repetition task. Correctness scores were calculated depending if the production was spontaneous or prompted (Table 10). The maximum score was 30, which means that the participant spontaneously produced both syllables correctly five times.

Table 10. Correctness scoring system for word learning task

Production:	Correct syllables	Score	Example (Lugo)
Spontaneous production	two-syllables	6	Lugo
Syllable prompted	two-syllables	5	Lu → Lugo
Word prompted	two-syllables	4	Lugo → Lugo
Spontaneous production	one-syllable	3	Luno
Syllable prompted	one-syllable	2	Lu → Luko
Word prompted	one-syllable	1	Lugo → Lulo
Spontaneous production	zero	0	Loki
Syllable prompted	zero	0	Lu → Loki
Word prompted	zero	0	Lugo → no answer

3.2.7. Articulation test

Accurate productions are defined as consonant and vowel productions that are free of errors such as distortions, omissions, or substitutions. The idea of articulation tests is to compare the child's utterances to the target word, while percentage of consonants correct (PCC) and percentage of vowels correct (PVC) are measures developed to address the need for a quantitative, relational inventory of mastered phonemes (i.e., measures of phonetic accuracy).

PCC and PVC represent the percentage of consonants and vowels produced correctly, calculated as number of correct consonants/vowels divided by the total number of consonants/vowels and multiplied by 100. Considering the literature review and the impairments in syllable structure production in children with CAS, three additional measures were calculated: the percentage of correct consonants in syllable-initial position - PCCI, the percentage of correct consonants in syllable-final position - PCCF, and the percentage of consonant deletion. Because the child must create a phonetic program while relying on the auditory-motor planning pathway to reproduce less familiar and complex words, and especially non-words, this task can be used to isolate motor programming skills (Vance, Stackhouse & Wells 2005).

Because the focus of our study was on phonological rather than phonetic development, consonant distortions were scored as correct, with only omissions and substitutions considered errors, similar to the PCC-Revised calculation described by Shriberg et. al. (1997c) and van Haaften et al. (2019).

The Vuletić articulation test (1990) is the only standardized articulation test in Croatian. It consists of description of pictures on different topics (children's room, playground, kitchen) and word and non-word imitation tasks. For this study, we chose the word and non-word imitation task because the picture description task is more difficult to administer, inefficient, and unattractive to the child (old-fashioned black and white photographs). The list of words ranges from simple two-syllable words to gradually more complex five-syllable words, from open to closed syllables, and from no consonant cluster to more complex consonant clusters. The words are ranked from familiar to less familiar words (Vuletić, 1990). The task required participants to reproduce pre-recorded words (N=10) and non-words (N=10) (Table x). For the calculation of phonemic speech sound inconsistency, 101 consonant possibilities were included. Of the 25 possible consonant phonemes in Croatian, 14 were represented in three possible positions within the word (initial, middle, and final), representing all possible places and manners of articulation (31 consonants in syllable-initial position and 10 consonants in syllable-final position). All five vowels were also included in different positions within the word, giving the children 64 possibilities to produce them.

Table 11. Word and non-word list, Vuletić articulation test (1990)

Word list	Non-word list
DEVET	TEDEV
LOPTA	TOLPA
KLUPA	PLAKU
TRAKTOR	KROTKAR
POKLOPAC	PLOCOPAK
SLIKARSTVO	PLISTVORKA
KAZALIŠTE	LAKAŠTELIZ
USISAVAČ	ASAČUVIS
KUPAONICA	PAUNOCIKA
ZAKISELJENOST	ISKELJEZANOST

Participants' responses were recorded and transcribed using the International Phonetic Alphabet (IPA) and The Extended Speech Assessment Methods Phonetic Alphabet (X-SAMPA) in SSA (Spontane Spraak Analyse, Eng. Spontaneous Speech Analysis), the Dutch program for analysing transcribed spontaneous speech. It is based on two existing instruments: The Computer Articulation Instrument (CAI; Maassen, 2019) and the Phonological Method for Dutch (FAN; Beers, 1995). After completing the transcriptions, the program calculated PCC, PVC, PCCI, PCCF and PCnD.

3.3 Data analysis

As a prerequisite for data analysis and hypothesis testing, some preliminary analyses and data checks were performed, including checking for missing values, identifying and handling univariate or multivariate outliers, analysing descriptive data, and creating a matrix of bivariate correlations of the composite variables. Data were analysed using SPSS Statistics, software version 25.0.

3.3.1. Missing values

Analysis of the missing values in the database of 58 participants showed that the problem of missing values was not an issue in this study. In addition, the only missing values for the AX discrimination task was for the variable reaction task - RTtime. When analysing the missing values in this variable, it is noticeable that in 10 out of 32 vowel pairs the percentage of missing values exceeds 10%, but the percentage of missing values does not exceed 20%. The highest percentage of missing values (19%) was found for the vowel pair O/U, which could mean that the participants had the greatest problems in discriminating the vowels O and U (more on this in the discussion).

3.3.2. Outliers

For most variables in this study, there are no significant univariate outliers. All univariate outliers identified were participants with CAS. Given the procedure and participant group (CAS group), it is apparent that these observed results represent the abilities of the participants, which is why they remain in the database and were included in further analysis.

Detected univariate outliers with low scores:

- One outlier on variable *Vowel height* and *Correctness* in the AX discrimination task.
- One outlier on variable *Phonemic segmentation*
- Five outliers on variable *Word repetition_OBLAK*
- Four outliers on variable *Two-syllabic_PATA* in the Maximum repetition rate task

Detected univariate outliers with high scores:

- One outlier on variable *Consistency_Lugo* in the Word learning task

Detection of multivariate outliers was performed by regression testing for Mahalanobis distance. The Mahalanobis distance is the distance of a case from the centroid of the remaining cases, where the centroid is the point that lies at the intersection of the means of all variables (Tabachnick & Fidell, 2013). With the value of $p < .01$, df = 31 (number of variables), there were no multivariate outliers. All data were stored in the database and included in further analysis.

3.3.3. Composite variables

In this study, some composite variables were created. For RT in the AX discrimination task there were three composite variables - the average times of each participant for the vowel height condition (AX_RTtime_height), the vowel backness condition (AX_RTtime_ backness), and the average time based on vowel height and backness (AX_RTtime_average). The same was done for the correctness variable (ACC). Average results were also formed for the consistency measures of the word repetition task (WR_Cons) and the non-word repetition task (NWR_Cons). For the MRR, two composite variables were formed based on the average scores for monosyllabic and two-syllable tasks of each participant (MRR_mono; MRR_bi). Although the three-syllable task (MRR_tri) had a single score, it was retained in Table X for clarity. In addition to composite variables, this study also used scalar data representing a single value.

3.3.4. Testing for normality

Formal statistical tests for normal distribution are not the first choice because they lead to rejection of the null hypothesis even when the distribution does not deviate significantly from the normal distribution (Tabachnick & Fidell, 2013). Analysis of skewness and kurtosis indicates that all values that are above ± 1.5 are not normally distributed (Tables 12 and 13). Gravetter and Wallnau (2014) and George and Mallery (2010) indicate that it is acceptable if skewness and kurtosis are both within ±2. In addition, Berman (2022) points out that in order to perform the independent samples *t*-test it is sufficient that the data are only slightly asymmetric, there is no significant number of divergent data, and the group sizes are between 16 and 40. If these criteria are met in this study, we could parametric tests to test the hypothesis.

3.3.5. Correlations

Defining the various constructs used in this study is an important part of the exploratory analysis. In this sense, the relationships (correlations) between the measured variables were tested. Since some variables did not have a normal distribution, Spearman's rank correlation was used. The correlations for all participants are presented in Table 14, while the specific correlations for the two groups studied (CAS and TD) can be found in the Appendix 3 and 4, for which scatter plots were also prepared (available upon request), indicating a linear relationship. These results will be referred to in the following sections.

Table 12. Descriptive statistics for composite variables of all participants (N=58)

Variables:	**Minimum**	**Maximum**	***M***	***SD***	**Skewness**[a]	**Kurtosis**[b]	**Cronbach α**
AX_ACC_ backness	4	16	13.2	2.7	-1.52	1.79	.78
AX_ACC_height	4	16	13.2	2.7	-1.53	1.78	.79
AX_ACC	8	32	26.5	5.4	-1.53	1.790	.90
AX_RTtime _height	127.3	248.4	161.3	22.1	1.720	4.285	.81
AX_RTtime _backness	133.5	248.4	161.8	21.7	1.850	4.625	.808
AX_RT	132.7	248.4	161.5	21.9	1.802	4.479	.907
WR	.32	.80	.7	.1	-0.759	-0.527	.802
NWR	.24	.80	.6	.2	-0.663	-0.539	.835
MRR_Mono	3.53	6.13	4.7	0.5	0.205	0.054	.874
MRR_Bi	.00	6.45	4.2	1.7	-1.312	0.897	.769
MRR_Tri	.00	8.30	3.8	2.4	-0.772	-0.868	-

[a]Standard Error of Skewness is 0.314

[b]Standard Error of Kurtosis is 0.618

Note. AX_ACC_backness = correctness score on the AX task with regard to vowel backness; AX_ACC_height = correctnesscorrectness score on the AX task with regard to vowel height; AX_ACC = average correctness score on the AX task for both conditions; AX_RTtime _height = reaction time from onset to last response on the AX task with regard to vowel height (sec); AX_RTtime _backness = reaction time from onset to last response on the AX task with regard to vowel backness (sec); AX_RTtime = average reaction time from onset to last response for both conditions (sec); WR = word repetition consistency, max. = 0.80 (5 repetitions minus the number of word forms divided by 5); NWR = non-word repetition consistency, max. = 0.80 (5 repetitions minus the number of word forms divided by 5); MRR_Mono = monosyllabic maximum repetition rate (syllable/sec); 20. MRR_Bi = bisyllabic maximum repetition rate (syllable/sec); 21. MRR_Tri = trisyllabic maximum repetition rate (syllable/sec)

Table 13. Descriptive statistics for variables of all participants (N=58)

Variables:	Minimum	Maximum	*M*	*SD*	Skewness[a]	Kurtosis[b]
RAN	.37	1.57	.9	.3	0.174	-0.192
Rhym_Rec	3	7	6.3	1.1	-1.791	2.425
Rhym_Prod	0	7	4.8	2.4	-0.960	-0.492
Syll_Seg	0	7	5.9	1.7	-1.796	2.797
Syll_Blend	1	7	6.3	1.0	-2.917	12.587
Phon_Seg	0	7	3.6	2.9	-0.054	-1.783
Phon_Blend	1	7	5.3	2.1	-0.946	-0.580
Cuni_Corr	0	30	20.3	8.7	-0.481	-0.939
Cuni_NoWf	0	.8	.6	.2	-0.869	0.696
Lugo_Corr	6	30	16.3	6.7	0.752	-0.185
Lugo_NoWf	0	.8	.6	.2	-0.593	-0.477
Žapo_Corr	0	30	17.5	9.8	0.006	-1.202
Žapo_NoWf	0	.8	.6	.2	-1.248	1.232
PCC	39	100	84.9	17.3	-1.046	-0.100
PVC	89	100	98.1	2.9	-1.575	1.576
PCCI	52	100	88.7	14.3	-1.171	0.212
PCCF	10	100	75.3	26.6	-0.918	-0.414
PCnD	77	100	97.6	4.6	-2.622	7.807

[a]Standard Error of Skewness is 0.314

[b]Standard Error of Kurtosis is 0.618

Note. RAN = rapid automatized naming (items per second); Rhym_Rec = rhyme recognition, number correct (max.=7); Rhym_Prod = rhyme production; Syll_Seg = syllable segmentation; Syll_Blend = syllable blending; Phon_Seg = phonemic segmentation; Phon_Blend = phonemic blending; Cuni_NoWf; Lugo_NoWf; Žapo_NoWf= novel word learning consistency (5 repetitions minus the number of word forms divided by 5); Cuni_Corr; Lugo_Corr; Žapo_Corr = novel word learning correctness score; PCC = percentage consonant correct; PVC = percentage vowels correct; PCCI = percentage correct consonants in syllable initial position; PCCF = percentage correct consonants in syllable final position; PCnD = percentage consonant not deleted

Table 14. Spearman's rank correlation for all the measured variables (N=58)

Variables:	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21
1. AX_RT	1.00																				
2. AX_Acc	.13	1.00																			
3. Rhym_Rec	.11	.46**	1.00																		
4. Rhym_Prod	.14	.50**	.62**	1.00																	
5. Syll_Seg	-.05	.23	.29*	.21	1.00																
6. Syll_Blend	.16	.29*	.35**	.23	.14	1.00															
7. Phon_Seg	.20	.50**	.67**	.61**	.29*	.24	1.00														
8. Phon_Blend	.05	.62**	.61**	.63**	.18	.37**	.83**	1.00													
9. RAN	.15	.49**	.37**	.32*	.31*	.43**	.58**	.60**	1.00												
10. WR_Cons	.12	.57**	.72**	.59**	.38**	.38**	.69**	.62**	.49**	1.00											
11. NWR_Cons	.12	.59**	.73**	.60**	.40**	.29*	.74**	.72**	.55**	.89**	1.00										
12. WL_Cons	-.07	.49**	.62**	.49**	.41**	.40**	.49**	.55**	.47**	.67**	.73**	1.00									
13. WL_Corr	-.02	.45**	.55**	.46**	.48**	.27*	.51**	.52**	.54**	.69**	.80**	.77**	1.00								
14. PCC	.01	.45**	.70**	.59**	.35**	.24	.72**	.68**	.40**	.82**	.81**	.64**	.67**	1.00							
15. PVC	.00	.35**	.61**	.52**	.32*	.04	.53**	.41**	.22	.70**	.65**	.55**	.50**	.71**	1.00						
16. PCCI	.15	.47**	.71**	.62**	.29*	.22	.74**	.64**	.38**	.82**	.80**	.56**	.64**	.93**	.74**	1.00					
17. PCCF	.15	.42**	.60**	.61**	.24	.20	.71**	.60**	.45**	.74**	.79**	.57**	.64**	.82**	.68**	.81**	1.00				
18. PCnD	.09	.42**	.63**	.66**	.19	.34**	.64**	.58**	.33*	.64**	.66**	.50**	.51**	.75**	.46**	.77**	.70**	1.00			
19. MRR_Mono	-.06	.36**	.47**	.37**	.43**	.24	.44**	.39**	.37**	.57**	.56**	.60**	.60**	.51**	.52**	.55**	.49**	.38**	1.00		
20. MRR_Bi	-.08	.39**	.48**	.48**	.30*	.20	.51**	.50**	.36**	.59**	.61**	.57**	.59**	.59**	.48**	.59**	.61**	.62**	.68**	1.00	
21. MRR_Tri	.07	.39**	.56**	.49**	.32*	.12	.56**	.49**	.39**	.66**	.66**	.52**	.52**	.69**	.50**	.69**	.58**	.63**	.53**	.65**	1.00

*p<.05, ** p< .01

Note. 1. AX_Acc = Ax discrimination task, correctness score; 2. Ax_RT = Ax discrimination task, total reaction time; 3. Rhym_Rec = rhyme recognition; 4. Rhym_Prod = rhyme production; 5. Syll_Seg = syllable segmentation; 6. Syll_Blend = syllable blending; 7. Phon_Seg = phonemic segmentation, 8. Phon_Blend = phonemic blending; 9. RAN = rapid automatized naming; 10. WR_Cons = word repetition consisntency; 11. NWR_Cons = non-word repetition consistency; 12. WL_Cons = word learning consistency; 13. WL_Corr = word learning correctness; 14. PCC = percentage consonant correct; 15. PVC = percentage vowels correct; 16. PCCI = percentage correct consonants in syllable initial position; 17. PCCF = percentage correct consonants in syllable final position; 18. PCnD = percentage consonant not deleted; 19. MRR_Mono = monosyllabic maximum repetition rate; 20. MRR_Bi = bisyllabic maximum repetition rate; 21. MRR_Tri = trisyllabic maximum repetition rate

4. Results

4.1. Differences in phonological and speech motor abilities

Five constructs were measured to analyse all levels of speech production (see Table 2 in the materials). The Independent Samples *t*-test was conducted to examine whether the two groups of participants differed significantly, while the Mann–Whitney *U* test was used as a nonparametric equivalent to the *t*-test for variables that deviate from the normal distribution.

4.1.1. Phonological rules

For the first level of speech production (i.e., phonological rules), AX discrimination task and phonological awareness tasks (rhyme production; syllable blending; syllable segmentation; phonemic blending; phonemic segmentation) were measured.

As mentioned previously, for the AX task, correctness (ACC) and the reaction time from onset to last response (RTtime) were calculated for each participant under two conditions (vowel height and backness). The results show that the groups differ on the correctness variable (Table 15), on both conditions (AX_ACC_backness; AX_ACC_height), and on average (AX_ACC).

Table 15. Differences between the groups on AX discrimination task (Mann-Whitney U test)

Variable	Group	Mean rank	Sum of ranks	Mann-Whitney *U* test
AX_ACC_backness	TD	38.7	1085	161.5**
	CAS	20.9	627	
AX_ACC_height	TD	38.4	1075	171.5**
	CAS	21.2	637	
AX_ACC	TD	38.7	1083	163.0**
	CAS	20.9	628	
AX_RTtime_height	TD	26.7	748	342.0
	CAS	32.1	963	
AX_RTtime_backness	TD	26.6	746	340.0
	CAS	32.2	965	
AX_RTtime	TD	26.7	748	342.0
	CAS	32.1	963	

p*<.05, **p<.01

Children with CAS were indeed less accurate at discriminating vowels than TD children - they could correctly differentiate 12.2 out of 16 vowel pairs, in contrast to TD children who could correctly differentiate 14.4 out of 16 pairs. In addition, there were some vowel combinations that were more difficult than others. 24/30 children with CAS and 20/28 TD children could not correctly discriminate the vowels /o/ and /u/, while 13/30 children with CAS could not discriminate the vowels /e/ and /i/, but only 1/28 TD children had difficulty discriminating this pair. Furthermore, there were no significant differences between CAS and TD children on correctly recognizing same vowel pairs /a-a/, /e-e/, /i-i/, /o-o/, /u-u/, additionally confirming difficulties with differentiating vowels (Table 16).

Table 16. Differences between the groups on same vowel recognition (Mann-Whitney U test)

Variable	**Group**	**Mean rank**	**Sum of ranks**	**Mann-Whitney *U* test**
AX_AA	TD	29.9	838	408.0
	CAS	29.1	873	
AX_EE	TD	32.4	908	338.0
	CAS	26.8	803	
AX_II	TD	30.9	865	381.0
	CAS	28.2	846	
AX_OO	TD	32.0	895	351.0
	CAS	27.2	816	
AX_UU	TD	30.4	851	395.0
	CAS	28.7	860	

p*<.05, **p<.01

As can be seen in Table 15, the groups also did not differ in total reaction time (i.e., the time they needed to complete the task). However, the total reaction time does not correlate with correctness.

In addition, two way-mixed ANOVAs were performed for accuracy and total reaction time in relation to vowel height and backness. Results from ANOVA show that there is a significant main effect of group ($F(1,56)=11.78$; $p<.01$), with TD children performing better ($M= 14.4$; $SD=0.47$) compared to CAS children ($M=12.1$; $SD=0.45$), but no significant interaction was found between vowel height and backness ($F(1,56)=0.93$; $p>.05$). Height and backness showed no significant interaction with participant group ($F(1,56)=0.93$; $p>.05$), as shown in Figure 9.

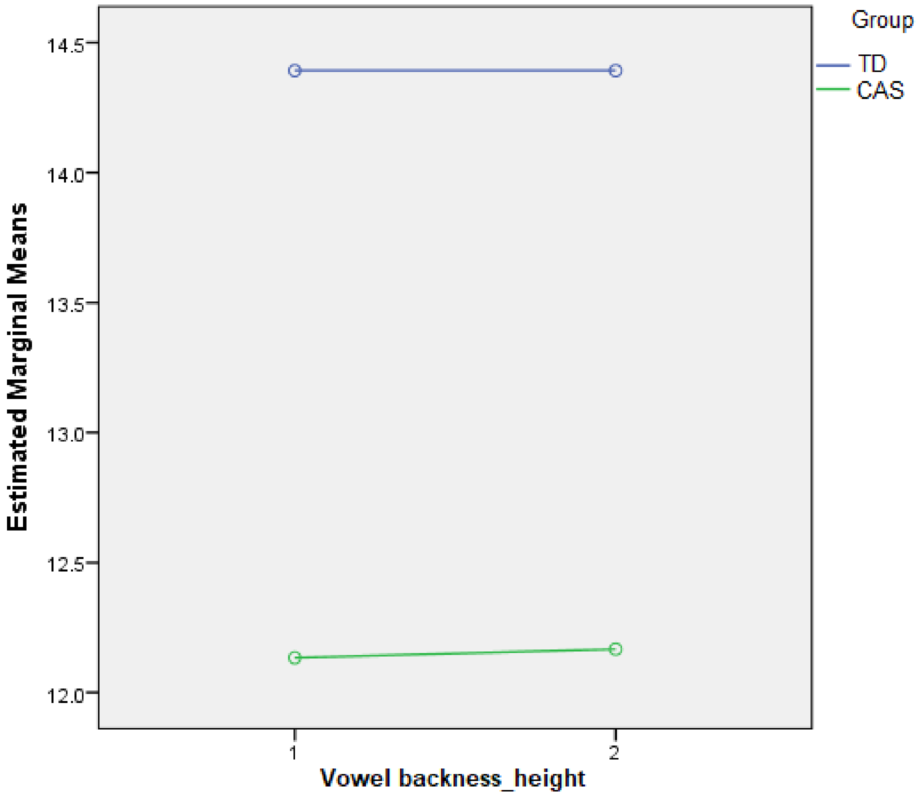


Figure 9. Vowel height and backness interaction with the participant group.

When testing for differences in phonological awareness tasks, the *t*-test was used for rhyme production, phonemic segmentation, and syllable blending, while the Mann-Whitney *U* test was used for rhyme recognition, syllable segmentation, and phonemic blending. The results showed that children with CAS performed significantly worse on all phonological abilities tasks compared to TD children (Table 17).

Table 17. Differences between the groups on rhyme production and phonemic segmentation and blending (t-test)

Variable	Group	*M*	*SD*	*Mdn*	Mann-Whitney *U* test	*t*- test	Effect size
Rhyme recognition	TD	6.7	0.19	7.0	136.5**		
	CAS	5.7	1.29	6.0			
Rhyme production	TD	6.3	0.81	6.0		5.62**	3.53[a]
	CAS	3.4	2.65	3.0			
Syllable blending	TD	6.7	0.48	7.0		2.52*	1.37[a]
	CAS	6.0	1.27	6.0			
Syllable segmentation	TD	6.5	1.04	7.0	247.0**		
	CAS	5.3	2.03	6.0			
Phonemic blending	TD	6.5	1.23	7.0	133.0**		
	CAS	4.2	2.15	5.0			
Phonemic segmentation	TD	5.8	1.71	6.0		7.71**	2.04[b]
	CAS	1.6	2.34	0.5			

**p<.01; *p<.05; *M* – mean; *SD* – standard deviation; *Mdn* - median; [a] – Glass's delta; [b]- Cohen's *d*

Although syllable blending is the simplest level of phonological ability, and children with CAS correctly blended 6 of 7 words presented, they were still statistically different from TD children (6.7 of 7) in this task. In addition, they recognized 5.7 out of 7 presented rhymes, whereas the TD children achieved an average of 6.7 correctly recognized rhymes. In the syllable segmentation task, the CAS children segmented 5.3 and the TD children segmented 6.5 of 7 presented words correctly. Children with CAS had difficulty in rhyme production with only 3.4 out of 7 rhymes produced, while this task was not as difficult for TD children with 6.3 rhymes produced correctly. In the phonemic tasks, children were dealing with smaller units than syllables, which made these tasks even more difficult. As expected, children with CAS performed significantly worse on both phonemic blending and segmentation. Thus, children with CAS successfully blended 4.2 words and segmented only 1.6 out of 7 words, in contrast to TD children who blended 6.5 words and segmented 5.8 words on average (Figure 10).

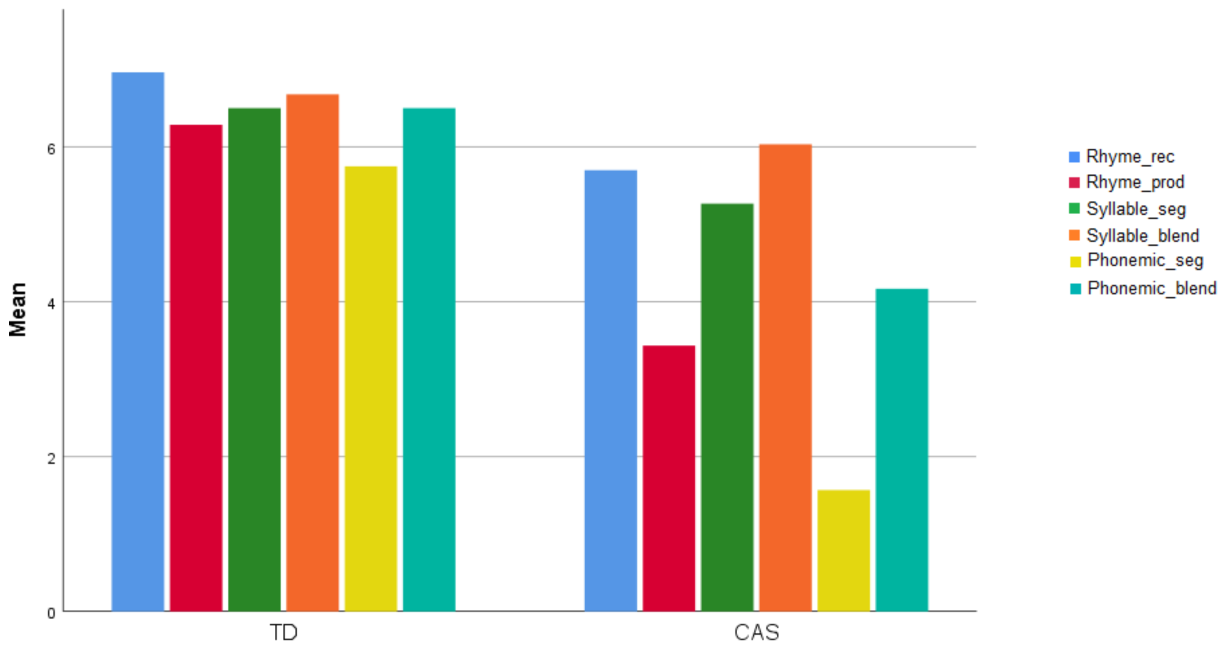


Figure 10. Average scores on phonological awareness tasks for TD and CAS children

For this study, the syllable and phoneme blending tasks were reconstructed so that no verbal response was required, similar to the study by Janssen et al. (2016), and therefore an age-matched control group was used for comparison instead of the available norms. Since the rhyming task was performed in its original form, as suggested by the PredČiP test (Kuvač Kraljević & Lenček, 2012), we can conclude that children with CAS, although performing worse than TD, are within the average range expected for children of this age - based on the available norms. Their average score on both rhyme recognition and rhyme production when added is 9.1, which is within typical range (from 9 to 14) and represents normal performance.

These results suggest that although children with CAS have lower scores on phonological awareness tasks compared to TD children, their phonological abilities related to rhymes are within the range of typical performance for preschool children.

4.1.2. Phonological planning

For the second level of speech production (i.e., phonological planning), RAN test, the word repetition task, the non-word repetition task, and the novel word learning task were measured. Results were normally distributed for both groups, except for *Lugo_NoWf* and *Žapo_NoWf*. Furthermore, all phonological planning tasks show positive correlations (Table X).

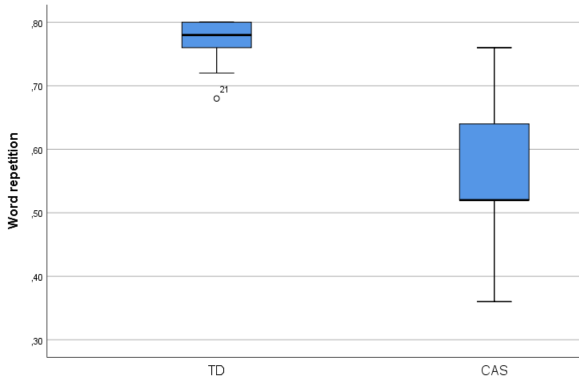
When compared on RAN test, analysis revealed that children with CAS produced significantly fewer items per second than did the TD children, with a large effect (Table 18). CAS children produced less than one item per second on average (*M*=0.77), whereas TD children produced one item per second on average (*M*=1.03).

Table 18. Differences between the groups on RAN test, word repetition task, the non-word repetition task (t-test)

Variable	**Group**	***M***	***SD***	***t*- test**	**Effect size**
WR_total	TD	.77	0.03	10.33**	6.65[a]
	CAS	.57	0.11		
NWR_total	TD	.75	0.05	11.07**	5.63[a]
	CAS	.48	0.12		
RAN	TD	1.0	0.23	4.11**	1.12[b]
	CAS	0.8	0.23		

**p<.01; *p<.05; *M* – mean; *SD* – standard deviation; *t*- statistic t-test; [a] – Glass's delta; [b]- Cohen's *d*

Furthermore, token-to-token inconsistency was calculated for repeated productions of words and non-words. The results show that there is a statistically significant difference between CAS and TD children, with CAS children being more inconsistent (Table 18). In the word repetition task, TD children (*M*=0.77) showed more consistency in repeated productions than CAS children (*M*=0.57) with large effect sizes. The same results were found for non-word repetition, where TD children (*M*=0.75) were also more consistent than CAS children (*M*=0.48), with large effect sizes. This is also evident in Figures 11 and 12, where lower scores represent more inconsistencies. A score of 0 means that the child produced 5 different word forms in 5 repetitions, while a maximum score of 0.8 means that the child produced 1 word form in 5 repetitions. Children with CAS produced an average of 2.15 distinct word forms and 2.55 distinct non-word forms in five consecutive repetitions, whereas TD children produced an average of 1.15 word forms and 1.25 non-word forms, suggesting that TD children have little more than one word from five consecutive repetitions.



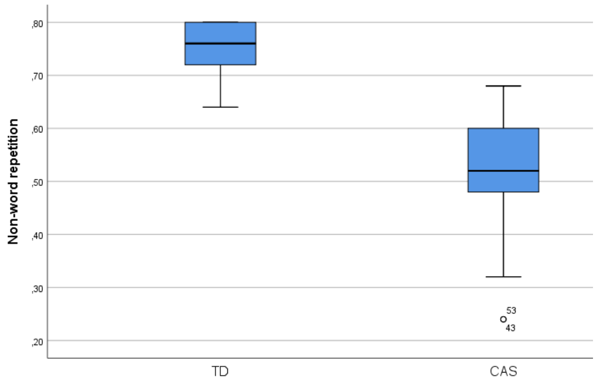


Figure 11. Boxplot of word repetition scores for TD and CAS children

Figure 12. Boxplot of non-word repetition scores for TD and CAS children

In addition, word and non-word consistency showed a high positive correlation with the other variables representing phonological planning, as well as with consonant and vowel production (PCC, PVC, PCCI, PCCF, PCnD) and monosyllabic, bisyllabic, and trisyllabic MRR (Table 14).

In the novel word learning task, consistency (number of word forms - NoWf) and correctness (Corr) were calculated for each word (Cuni, Lugo, Žapo). The results show that the difference between the two groups was statistically significant for both consistency and correctness (Table 19).

Table 19. Difference between the groups on novel word learning task (t-test and Mann-Whitney U test)

Variable	**Group**	***M***	***SD***	***Mdn***	**Mann-Whitney *U* test**	***t*- test**	**Effect size**
Cuni_Corr	TD	25.5	5.51	28.0		5.70**	1.95[a]
	CAS	14.8	8.58	14.0			
Lugo_Corr	TD	19.7	6.37	17.5		4.17**	1.10[b]
	CAS	13.2	5.40	13.5			
Žapo_Corr	TD	23.0	9.29	29.0		4.94**	1.15[a]
	CAS	12.3	7.05	11.0			
Cuni_NoWf	TD	.7	.15	.8		2.90**	0.77[b]
	CAS	.6	.20	.6			
Lugo_NoWf	TD	.7	.15	.7		4.31**	1.06[b]
	CAS	.5	.22	.4			
Žapo_NoWf	TD	.8	.10	.8	178.0**		
	CAS	.6	.21	.6			

**p<.01; *p<.05; *M* – mean; *SD* – standard deviation; *Mdn* - median; [a] – Glass's delta; [b]- Cohen's *d*

The elicited productions of all three target words were statistically less correct for children with CAS, and for all three words, with large effect sizes. In addition, their productions were more inconsistent (Table 19) than the productions of TD children (word Cuni with large effect size). Correctness and consistency for all three words Cuni, Lugo, and Žapo showed a high positive correlation (Table 20). However, correctness and consistency values for the word Lugo did not correlate with correctness and consistency of the words Cuni and Žapo. This could be due to the articulation movement, which is more similar for the words Cuni and Žapo, while Lugo was the most articulatory difficult word due to the transition from the front (alveolar - /l/) to the back (velar - /g/) of the mouth, which distinguishes it from the other two words.

Table 20. Spearman's rank correlation for the novel word learning task

Variables:	**1**	**2**	**3**	**4**	**5**	**6**
1. Cuni_Corr	1.00					
2. Cuni_NoWf	.65**	1.00				
3. Lugo_Corr	.23	-.05	1.00			
4. Lugo_NoWf	.37**	.14	.52**	1.00		
5. Žapo_Corr	.50**	.40**	.21	.33*	1.00	
6. Žapo_NoWf	.38**	.12	.14	.18	.54**	1.00

**p<.01; *p<.05

In summary, children with CAS performed significantly worse on all phonological planning tasks.

4.1.3. Phonetic programme assembly

For the third level of speech production (i.e., phonetic programme assembly), the percentage of correct consonants in syllable-initial position – PCCI, the percentage of correct consonants in syllable-final position – PCCF and PCnD – percentage consonants not deleted were calculated. The results were normally distributed for PCCF, but not for PCCI and PCnD.

Children with CAS evidenced statistically significant difficulties in production of consonants in final position (*M*=54) compared to TD children (*M*=95.4), with a large effect size (Table 21). Mann Whitney *U* test revealed similar results in production of consonants in initial position (*M*=76.3), showing that children with CAS also exhibit more omissions and substitutions (Table 21) then TD children (*M*=98.8) implicating that children with CAS have difficulties with consonants in syllable structure production (i.e. with phonetic programme assembly). Furthermore, the analysis of a specific measure targeting consonant deletion showed that TD children do not delete consonants (*M*=100) while CAS children show statistically more consonant deletions (*M*=95.3) throughout their production (Table 21). PCnD shows a high positive correlation with word and non-word consistency, while PCCF and PCCI show an even higher positive correlation with word and non-word consistency.

Table 21. Percentages correct on PCCF, PCCI and PCnD for both groups

Variable	**Group**	***M***	***SD***	***Mdn***	**Mann-Whitney *U* test**	***t*- test**	**Effect size**
PCCF	TD	95.4	5.08	100.0		8,24*	1.59[a]
	CAS	54.0	26.08	55.0			
PCCI	TD	98.8	2.73	100.0	38.0**		
	CAS	76.3	20.02	81.0			
PCnD	TD	99.9	0.76	100.0	150.5**		
	CAS	95.3	5.67	96.00			

**p<.01; *p<.05; *M* – mean; *SD* – standard deviation; *Mdn* - median; [a] – Glass's delta; [b]- Cohen's *d*

Note. PCCF = percentage consonants correct in syllable-final position; PCCI = percentage consonants correct in syllable-initial position; PCnD: percentage consonants not deleted. Higher values mean better performance, i.e., fewer omissions and deletions.

4.1.4. Motor speech programme implementation

Variables for the fourth level of speech production (i.e. motor speech programme implementation) were formed based on the average scores (syllable per second) for monosyllabic (/pa/, /ta/ and /ka/) and bisyllabic task (/pata/ and /taka/) of each participant. Only the child's best attempt was included in the analysis (regardless of whether the instruction was "*fast*" or "*as fast as possible*"). The results were normally distributed.

According to the data, as expected, the monosyllabic repetition is the least demanding task, while the trisyllabic repetition is the most demanding. In addition, there is an apparent trend toward slower rates in children with CAS as the complexity of the sequence increases, with more children unable to reproduce more complex repetitions; 16 children with CAS were unable to produce the correct three-syllable sequence (Figure 13), and their results were marked as missing values. In contrast, TD children were able to successfully reproduce all sequences regardless of complexity, performing faster as sequence complexity increased.

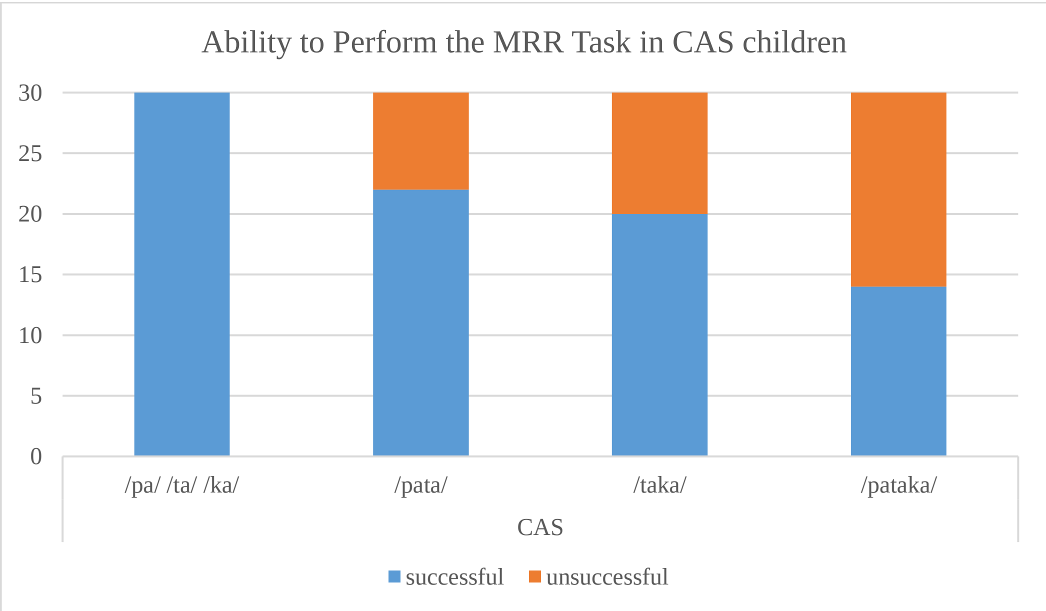


Figure 13. Overview of the ability to perform monosyllabic (/pa/, /ta/, /ka/); two-syllabic (/pata/ and /taka/) and three-syllabic task (/pataka/).

Differences between mean syllable rates for each of the successive sequences revealed that children with CAS had statistically significantly slower maximum repetition rates for both alternating movement rates (i.e., repetition of one-syllable sequences) and sequential movement rates (i.e., repetition of multisyllabic sequences), with large effect sizes for all tasks (Table 22).

Table 22. Difference between the groups on maximum repetition rate tasks (t-test)

Variable	**Group**	***M***	***SD***	***t*- test**	**Effect size**
MRR_Mono	TD	5.0	0.49	4.93**	1.29[b]
	CAS	4.4	0.43		
MRR_Bi	TD	5.3	0.55	5.76**	3.84[a]
	CAS	3.2	1.92		
MRR_Tri	TD	5.5	0.79	7.50**	4.31[a]
	CAS	2.1	2.35		

**p<.01; *p<.05; *M* – mean; *SD* – standard deviation; *t*- statistic t-test; [a] – Glass's delta; [b]- Cohen's *d*

In addition, a high positive correlation was found between performance on monosyllabic, bisyllabic, and trisyllabic repetition tasks (Figure 14). As the monosyllabic rate increases, both the two- and three-syllabic rates increase. The highest correlation was found between mono- and bisyllabic and bi- and trisyllabic rates. Interestingly, this positive correlation was not found in children with CAS between mono- and bisyllabic and mono- and trisyllabic rates, while the correlation between bi- and trisyllabic rates was moderate.

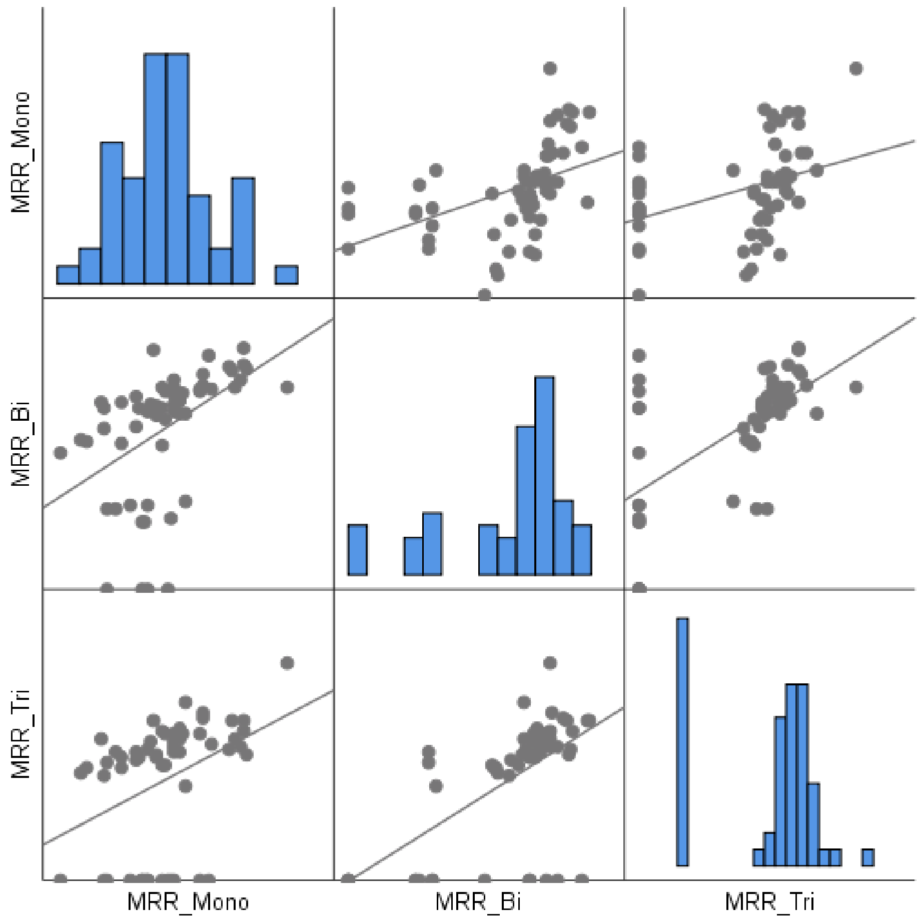


Figure 14. Scatterplot matrix for mono-, bi- and tri- syllabic repetition rates (N=58)

4.1.5. Speech execution

Final level of speech production was measured as percentage of consonants (PCC) and vowels (PVC) productions that are free of omissions or substitutions. Since the two variables are not normally distributed Mann-Whitney *U* test was used to test for differences.

The analysis showed significant differences between children with CAS and TD children on PCC and PVC (Table 23). Children with CAS had more errors in the production of consonants and vowels and showed an overall less consistent phoneme pattern compared to the TD children.

Table 23. Difference between the groups on PCC and PVC (Mann-Whitney U test)

Variable	**Group**	**Mean rank**	**Sum of ranks**	**Mann-Whitney *U* test**
PCC	TD	44.1	1236	10.5**
	CAS	15.9	476	
PVC	TD	40.0	1121	125.0**
	CAS	19.7	590	

**p<.01

Children with CAS produced an average of 69.6% correct consonants, in contrast to TD children who had an average of 98.2% (Figure 15 and 16). They were much better at vowel production, with 93.2%, opposed to TD children, who produced 99.8% vowels correctly. PCC and PVC have a positive moderate to high correlation with all measured variables except total reaction time and syllable blending.

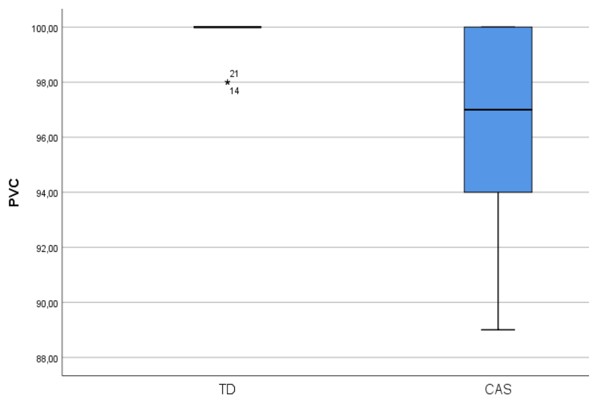


Figure 15. Mean PVC scores for TD and CAS children

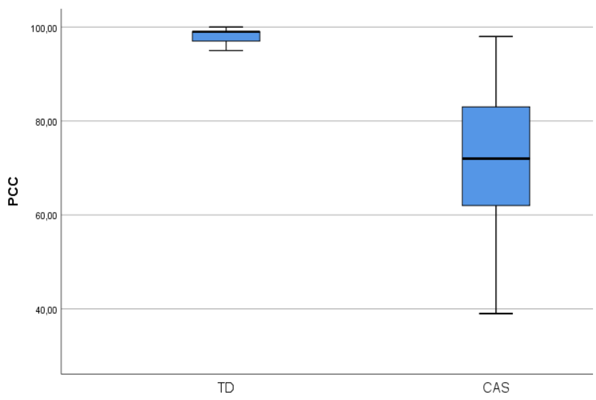


Figure 16. Mean PCC scores for TD and CAS children

4.2. Speech motor production in phonological tasks

Classical tasks on phonological aspects in the speech production process challenge both phonological input and output processes, but the available standardized phonological tasks in Croatian all require a verbal response (PredČiP; Kuvač Kraljević & Lenček, 2012). This study aimed to further investigate phonological processes while controlling for the mode of response, which can be given verbally or nonverbally.

If the child had difficulty articulating certain phonemes (omissions, substitutions, or distortions) but recognized the phonological segment under study (i.e., had an accurate representation of the syllable or phoneme), the response was considered correct.

To examine differences between different tasks with different phonological awareness level with and without verbal response, a dependent-samples *t*-test was performed, except for the syllable blending and segmentation tasks. Due to the non-normal distribution, the Wilcoxon signed-rank test was performed for these variables. The results are presented in Table 24.

Table 24. Differences between the phonological awareness tasks with and without verbal response - CAS children

Variable	**Phonological awareness level**	**Verbal response**	***M***	***SD***	***t***
Rhyme recognition (predČiP)	shallow-deep	✖	5.7	1.29	5.29**
Rhyme production (predČiP)	shallow-deep	✔	3.4	2.65	
Syllable blending (predČiP)	shallow	✖	6.0	1.27	-1.53[a]
Syllable segmentation (predČiP)	shallow	✔	5.3	2.03	
Phonemic blending (predČiP)	deep	✖	4.2	2.15	-7.55**
Phonemic segmentation (predČiP)	deep	✔	1.6	2.34	

**p<.01

M- mean; *SD*- standard deviation; *t*- statistik t-test; [a]- Wilcoxon Signed-Rank Test for Syllables blending

Note. Difficulty level represents the degree of difficulty of the presented task, E = easier, D = more difficult (e.g. blending is easier than segmentation); Verbal response **x** = the task does not require a verbal response; Verbal response ✔ = the task requires a verbal response.

As developmentally expected children with CAS had more success in recognizing rhymes than in producing them (*t*= 5.29; *df*= 29; *p*<.001). Similarly, they were more successful in phonemic blending than in phonemic segmentation, (*t*= -7.55; *df*= 29; *p*<.001). However, there were no differences in the syllables task (*t*= -1.53, *df*= 29; *n.s.*); children with CAS performed equally well on syllable blending and syllable segmentation. In contrast to the rhyme and phonemic awareness tasks, which showed high positive correlations, the syllable awareness tasks showed no correlation at all (Table 25).

Table 25. Spearman's rank correlation for the phonological awareness tasks with and without verbal response (CAS children)

Variables:	**1**	**2**	**3**	**4**	**5**	**6**
1. Rhyme recognition	1.00					
2. Rhyme production	.55**	1.00				
3. Syllable segmentation	-.05	.16	1.00			
4. Syllable blending	.28	.24	.05	1.00		
5. Phonemic segmentation	.37*	.51**	.00	.05	1.00	
6. Phonemic blending	.42*	.64**	-.00	.38*	.71**	1,00

**p<.01; *p<.05

Differences on the same tasks were also analysed for TD children, as a control (Table 26). Similar to the children with CAS, the TD children were statistically more successful (*t*=4.16, *df*= 27; *p*<.001) at rhyme recognition (*M*=6.9; *SD*=0.19) than at rhyme production (*M*=6.3; *SD*=0.08). As expected, TD children also performed better (*t*= -3.21, *df*= 27; *p*= .003) at phonemic blending task (*M*=6.5, *SD*=1.23) then at the phonemic segmentation task (*M*=5.8, *SD*=1.71). There were no differences in the syllable tasks (*t*=-0.56, *df*= 27; *n.s.*).

Table 26. Differences between the phonological awareness tasks with and without verbal response -TD children

Variable	**Phonological awareness level**	**Verbal response**	***M***	***SD***	***t***
Rhyme recognition (predČiP)	shallow-deep	✗	6.9	0.19	4,16**
Rhyme production (predČiP)	shallow-deep	✓	6.3	0.81	
Syllable blending (predČiP)	shallow	✗	6.7	0.48	-0.56[a]
Syllable segmentation (predČiP)	shallow	✓	6.5	1.04	
Phonemic blending (predČiP)	deep	✗	6.5	1.23	-3,21**
Phonemic segmentation (predČiP)	deep	✓	5.8	1.71	

***p*<.01
M- mean; *SD*- standard deviation; *t*- statistik t-test; [a]- Wilcoxon Signed-Rank Test for Syllables blending

In contrast to the CAS children, only phonemic blending and segmentation showed a high positive correlation in the TD children, whereas the results at the other levels of phonological awareness did not correlate (Table 27).

Table 27. Spearman's rank correlation for the phonological awareness tasks with and without verbal response (TD children)

Variables:	1	2	3	4	5	6
1. Rhyme recognition	1.00					
2. Rhyme production	-.20	1.00				
3. Syllable segmentation	.35	-.29	1.00			
4. Syllable blending	.28	.19	.09	1,00		
5. Phonemic segmentation	.19	.08	.27	.15	1.00	
6. Phonemic blending	-.10	.36	-.11	.18	.63**	1.00

**p<.01; *p<.05

In addition, to determine whether there was an effect of verbal response, a total score for three tasks representing equal phonological awareness levels with and without speech motor performance was calculated as a simple linear combination of the scores for these tasks. To determine whether there was an effect of speech motor performance on phonological awareness, a two-way analysis (ANOVA) was conducted.

The results indeed showed a significant main effect of speech motor production (verbal response) (F(1,56)=81.508, p<.001). Moreover, the interaction between the group (TD and CAS) and the condition was also significant (F(1,56)= 25.203, p<.001). Although the effect of speech motor production is seen in both groups, it is more pronounced in children with CAS (Figure 17), with higher success on tasks that do not involve speech motor performance.

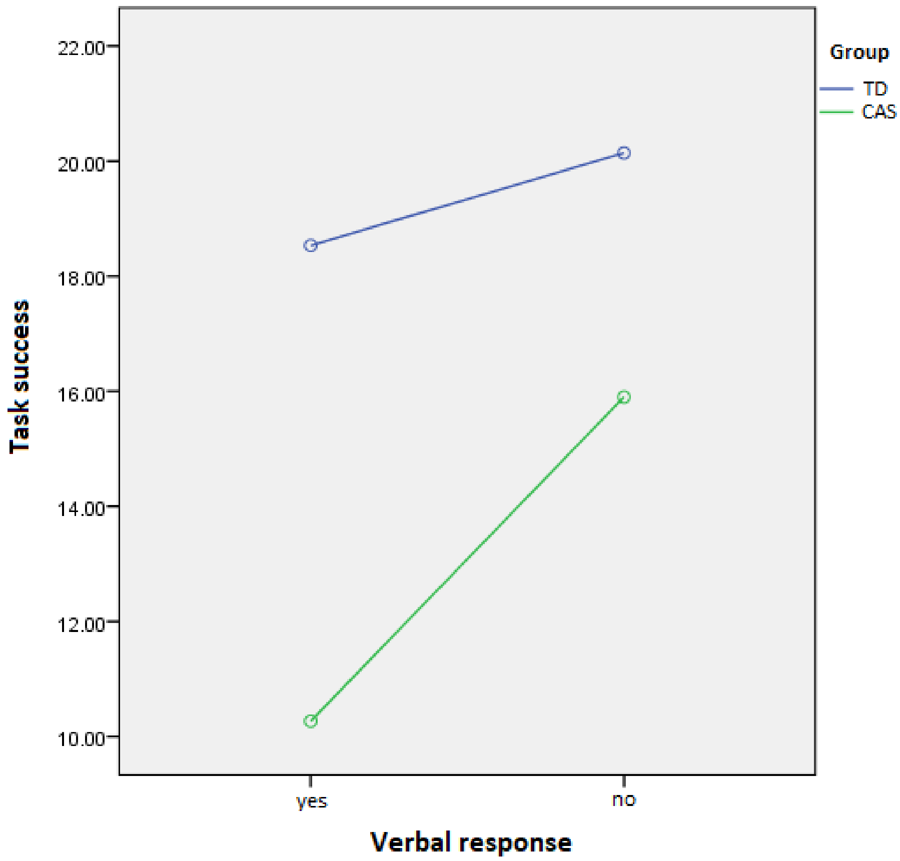


Figure 17. Main effect of the group and speech motor performance (verbal response) and their interaction

Furthermore, performances of TD children between different levels of phonological awareness do not differ. They are equally successful on rhyme (shallow-deep) and syllable (shallow) level ($t(27) = 0.26$, p=n.s.), rhyme (shallow-deep) and phonemic (deep) level ($t(27) = 2.07$, p=n.s.), and syllable (shallow) and phonemic (deep) level ($t(27) = 1.71$, p=n.s). In contrast to TD children, children with CAS differ statistically between all three levels: rhyme (shallow-deep) and syllable (shallow) level ($t(29) = -3.25$, $p<.001$.), rhyme (shallow-deep) and phonemic (deep) level ($t(29) = 5.88$, $p<.001$), and syllable (shallow) and phonemic (deep) level ($t(29) = 6.85$, $p<.001$). However, further analysis showed that there is no difference only between rhyme recognition and syllable blending, both of which are tasks without verbal response but with different levels of phonological awareness (Table 28).

Table 28. Differences between different phonological awareness levels with the same verbal response – CAS children

Variable	Phonological awareness level	Difficulty level	Verbal response	*M*	*SD*	*t*
Rhyme recognition	shallow-deep	E	✖	5.7	1.29	4.03**
Phonemic blending	deep	E	✖	4.2	2.15	
Rhyme production	shallow-deep	D	✔	3.4	2.65	4.26**
Phonemic segmentation	deep	D	✔	1.6	2.34	
Rhyme recognition	shallow-deep	E	✖	5.7	1.29	-1.11
Syllable blending	shallow	E	✖	6.0	1.27	
Phonemic blending	deep	E	✖	4.2	2.15	-4.92**
Syllable blending	shallow	E	✖	6.0	1.27	
Phonemic segmentation	deep	D	✔	1.6	2.34	-6.52**
Syllable segmentation	shallow	D	✔	5.3	2.03	
Rhyme production	shallow-deep	D	✔	3.4	2.65	-3.27**
Syllable segmentation	shallow	D	✔	5.3	2.03	

**$p<.01$

M- mean; *SD*- standard deviation; *t*- statistic t-test

Note. Difficulty level represents the degree of difficulty of the presented task, E = easier, D = more difficult (e.g. blending is easier than segmentation); Verbal response **x** = the task does not require a verbal response; Verbal response ✔ = the task requires a verbal response.

In summary, when compared on the same phonological awareness level with different response models, there is no statistical difference at the syllable level (shallow), while there are significant differences at the rhyme level (shallow-deep) and especially at phonemic level (deep) for both groups of children, although this effect of verbal response at the rhyme and phoneme levels is much larger for the CAS children. Moreover, TD children are equally successful at different levels of phonological awareness. However, they are less successful on tasks that require a verbal response and have a higher level of difficulty. CAS children, on the other hand, differ on tasks with different levels of phonological awareness, with the phonemic level (deep) being the most difficult, but they do not differ on two easier levels (rhyme recognition and syllable blending) that do not require a verbal response.

It is evident that children with CAS have more difficulty at deeper levels of phonological awareness and with more difficult tasks such as segmentation of smaller units that require a verbal response. Thus, one might conclude that having to perform a (difficult) speech task affects the phonological processing in children with CAS, which in turn may affect the linguistic levels of speech production processes. However, further research on phonological awareness is needed to establish this relationship.

4.3. Clusters of children with CAS

The idea of cluster analysis is to measure the distance between each pair of children with CAS in relation to the variables proposed in the study, and then to group children who are similar to each other. One of the aims is to give a better insight into the differences in phonological abilities.

There are a number of algorithms that can be used, however, there is no universal clustering algorithm, so it is important to decide on appropriate clustering techniques. Considering that there are no known subgroups of children with CAS, K-Means clustering was chosen. The K-Means clustering algorithm finds observations in a data set that are similar to each other and assigns them to a group. After calculating K centroids based on Euclidean distance, data points are assigned to the closest centroid that forms the cluster, and this process is repeated until there is no change in centroid position (Kansal, Bahuguna, Singh & Choudhury, 2018).

Of the 30 children with CAS, 29 children were included in the cluster analysis (data were missing for participant 63). The initial analysis involved hierarchical cluster analysis, whereas dissimilarity was determined using Euclidean distance. Before performing the regression analysis, all values were converted to z-scores to make the data comparable. An examination of the resulting agglomeration schedules and dendrograms suggested two cluster solutions (see Appendix 5 and 6).

Further investigation of group membership and sizes and subsequent analysis involved a non-hierarchical K-Means clustering algorithm. Cluster identification was based on the dissimilarity of all previously mentioned variables testing all theorized processes of speech production, except for total reaction time and percentage consonant non deleted, as there were no differences between groups in RT time and the variable PCnD was less informative than PCCI and PCCF. The K-Means cluster algorithm confirmed the three-cluster solution as the most appropriate - cluster 1 (N=9), cluster 2 (N=7), and cluster 3 (N=13). Convergence was reached with the fourth iteration (Table 29).

Table 29. Iteration history (K-means clustering for three-segment solution)

	Change in cluster centers		
Iteration	**1**	**2**	**3**
1	27.305	19.036	27.922
2	5.663	2.995	5.774
3	4.580	.000	2.812
4	.000	.000	.000

Table 30 summarises the descriptive data for each cluster. Cluster 1 appears to represent children with CAS who have better phonological skills and fewer vowel and consonant errors. Cluster 2 appears to represent children with significant consonant errors as well as difficulties with rhyme production, phonemic blending, and segmentation. Children in cluster 2 also show a slower rate of repetition in bisyllabic and trisyllabic sequences. Cluster 3 represents the largest cluster, and children in this cluster are slightly better in consonant production and phonological skills than in cluster 2, but worse than children in cluster 1 (Figure 18).

Table 30. Cluster description

	Cluster 1 (N=9)		Cluster 2 (N=7)		Cluster 3 (N=13)			
Variables:	*M*	*SD*	*M*	*SD*	*M*	*SD*	F	Sig.
RAN	.9	0.17	.8	0.29	.7	0.23	0.68	.516
Rhym_Rec	6.0	1.23	5.9	0.69	5.6	1.45	0.27	.767
Rhym_Prod	5.2	2.28	1.4	1.99	3.5	2.40	5.48	.010*
Syll_Seg	4.7	2.449	5.1	1.95	5.8	1.88	0.76	.477
Syll_Blend	6.0	.866	5.3	2.14	6.5	.66	2.42	.108
Phon_Seg	3.3	2.739	.3	.49	1.2	2.12	4.71	.018*
Phon_Blend	5.3	1.803	3.3	2.21	4.0	2.16	2.09	.144
WR_Pcorr	.6	.11	.5	0.04	.6	0.10	2.01	.155
NWR_Pcorr	.5	0.10	.4	0.14	.5	0.09	3.82	.035
AX_Acc	12.9	2.51	12.1	2.12	12.2	3.00	0.24	.786
MRR_Mono	4.5	0.32	4.3	0.40	4.5	0.45	0.76	.479
MRR_Bi	4.3	1.26	1.6	1.70	3.9	1.71	6.52	.005**
MRR_Tri	2.1	2.45	.6	1.63	3.6	2.09	4.50	.021*
PCC	81.3	11.24	51.6	7.96	76.5	10.93	18.30	.000**
PVC	97.8	2.49	95.1	3.13	96.2	3.72	1.37	.271
PCCI	89.6	8.31	60.86	7.67	81.4	10.46	19.81	.000**
PCCF	84.4	8.82	24.3	9.76	53.1	9.47	82.63	.000**
WL_Cons	.5	0.07	.5	0.15	.6	0.09	1.73	.198
WL_Corr	.5	0.15	.4	0.18	.5	0.14	0.72	.494

$^{**}p<.01$; $^{*}p<.05$

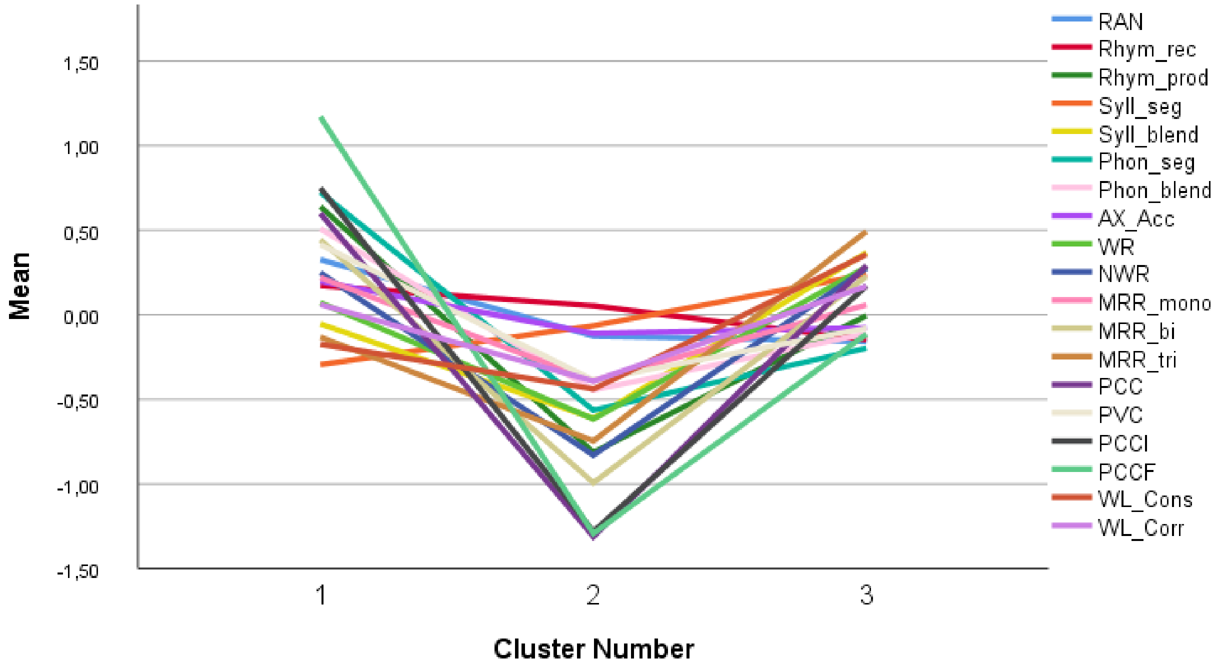


Figure 18. Final cluster centres

To further validate the structure of the cluster solution, a comparison between clusters was performed. Because the cluster sizes were small, the nonparametric Kruskal-Wallis test was used to determine if there were differences, and pairwise comparison (significance values adjusted by Bonferroni correction) was used to further determine differences between clusters. The results are presented in Table 31.

Table 31. Differences between clusters (Kruskal-Wallis test)

Variables:	Cluster	Mean Rank	χ 2	Sig.	Pairwise comparison
RAN	1	18.0	1.621	.445	
	2	13.6			
	3	13.7			
RhymRec	1	16.8	.691	.708	
	2	13.9			
	3	14.4			
RhymProd	1	20.7	9.007	.011*	1-2 (.008)
	2	8.0			2-3 (n.s.)
	3	14.8			3-1 (n.s.)
SyllSeg	1	12.8	2.060	.357	
	2	13.3			
	3	17.4			
SyllBlend	1	13.0	3.532	.171	
	2	11.9			
	3	18.0			

PhonSeg	1 2 3	21.4 10.4 13.0	9.038	.011*	1-2 (.017) 2-3 (n.s.) 3-1 (.045)
PhonBlend	1 2 3	19.2 11.3 14.1	3.723	.155	
WR_Pcorr	1 2 3	15.2 9.1 18.1	5.317	.070	
NWR_Pcorr	1 2 3	16.2 9.1 17.3	4,579	,101	
AX_Acc	1 2 3	17.1 13.0 14.7	.979	.613	
MRR_Mono	1 2 3	16.3 12.6 15.4	.806	.668	
MRR_Bi	1 2 3	19.1 6.4 16.8	9.940	.007**	1-2 (.009) 2-3 (.026) 3-1 (n.s.)
MRR_Tri	1 2 3	14.3 9.0 18.7	6.810	.033*	1-2 (n.s.) 2-3 (.029) 3-1 (n.s.)
PCC	1 2 3	19.6 4.4 17.5	14.547	.001**	1-2 (.001) 2-3 (.003) 3-1 (n.s.)
PVC	1 2 3	18.1 11.2 14.9	2.677	.262	
PCCI	1 2 3	21.4 4.7 16.1	15.776	.000**	1-2 (.000) 2-3 (.013) 3-1 (n.s.)
PCCF	1 2 3	24.9 4.2 13.9	24.072	.000**	1-2 (.000) 2-3(.043) 3-1 (.008)
WL_Cons	1 2 3	13.4 11.3 18.1	3.522	.172	
WL_Corr	1 2 3	15.2 10.5 15.2	2.888	.236	

**p<.01; *p<.05

The results show that there are statistical differences in rhyme production and phonemic segmentation, both representing more complex phonological abilities. Cluster 1 performed statistically better than cluster 2 on both tasks, but compared to cluster 3, they were statistically better only on phonemic segmentation, while there were no differences on rhyme production. There were no differences between clusters 2 and 3 on the aforementioned tasks. Furthermore, there were statistical differences in bi- and trisyllabic maximum repetition rate, with both clusters 1 and 3 performing better than cluster 2, and in bisyllabic maximum repetition rate, cluster 3 performed better than cluster 2. The third aspect in which differences were significant was consonant production. Clusters 1 and 3 performed better than cluster 2 on both the PCC and PCCI, while cluster 1 performed better than clusters 3 and 2, but cluster 3 performed better than cluster 2 (Figure 19). In summary, cluster 3 does not differ significantly from cluster 2 in the phonological awareness tasks, but it does differ from cluster 2 on the (significant) production tasks. Thus the largest group (n=13) with overall intermediate scores, is relatively poor (similar to cluster 2) on phonological awareness.

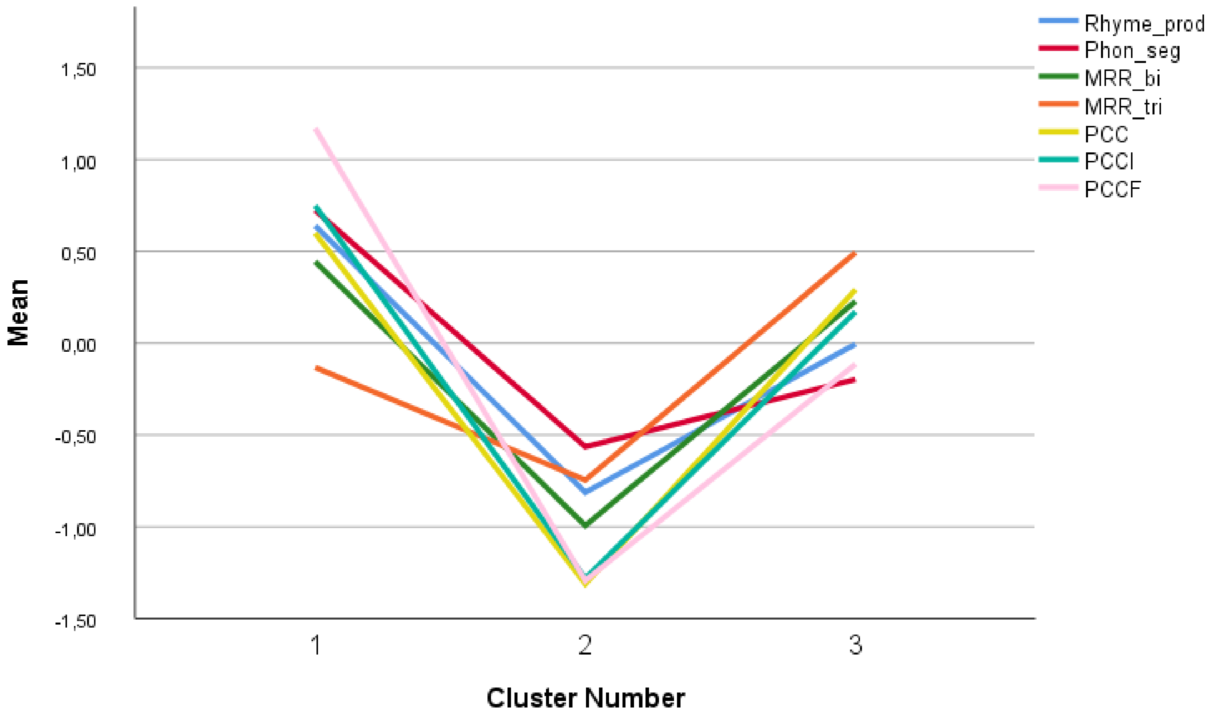


Figure 19. Significant variable differences between the clusters

These three comorbid subgroups could be divided by severity into mild (cluster 1), moderate (cluster 3), and severe (cluster 2) based on performance on the above mentioned measures. This is also supported by the fact that the clusters did not differ with respect to age $H(2)=0.89$, $p=.642$, which means that these differences cannot be attributed to maturation. On all other variables, these three groups showed similar performance.

5. DISCUSSION

The main goal of this study was to investigate phonological abilities as well as speech motor abilities following the premise of the process-oriented approach using a series of speech tasks representing different steps of the speech motor process (i.e., Cascade model; Ozanne, 1995) in order to better understand speech processing deficits in CAS. To describe a speech production process, one could argue about which model best describes the speech phenomena (Levelt 1989; van der Merwe 1997; Ozanne; 1995). All of these symbolic models agree that an abstract linguistic phonological form is planned before a more specified motor plan is encoded (Laganaro, 2012). In compliment to the model of Levelt and colleagues (Levelt et al.,1999), who proposed an articulatory network as the final level of speech production, Ozanne (1995; 2010) further elaborates on this final level: motor programming (which she further subdivides into: phonetic programme assembly and motor speech programme implementation) and motor execution. These models are modular and imply that there is a flow of information between these processes, usually in one direction. However, in the Cascade model (Ozanne, 1995; 2010), the flow is assumed to be bidirectional, which means that these processes can influence each other. The following section discusses the results of this study, as well as other similar studies conducted to determine processing levels underlying CAS.

5.1. Speech production processes in children with CAS

5.1.1. Phonological representations and phonological awareness in children with CAS

Although the Cascade model does not recognize the highest level of speech production (i.e., phonological rules) as the level at which children with CAS have difficulty, the results presented in the previous chapter are inconclusive. This is not surprising given that phonology is not a single construct-according to Ramus, Marshall, Rosen, and van der Lely (2013), it has at least two components: phonological representations and phonological abilities. The problem is that any task tapping phonological abilities necessarily involves phonological representations, so deterioration in phonological representations would degrade performance on all phonological abilities (Ramus et al., 2013).

The relationship between perception and production is well established, and several studies have shown a relationship between consonant perception and speech sound deficits. Vowels appear to be more sensitive to auditory/phonetic discrimination than consonants (Maassen et al., 2003).

The present study shows that children with CAS differ from TD children in vowel discrimination, suggesting that children with CAS do not have a correctly encoded phonological representation of vowels. Although they have difficulty with vowel discrimination, the speed with which they responded on each trial was similar to that of TD children. It would be expected that if they are unsure whether the two vowels are different, they would need more processing time to make a response. However, this is not the case, that is children with CAS did not differ in total reaction time, which could lead us to conclude that they do not have processing problems but access incorrectly encoded phonological representations of vowels.

These results confirm the findings of Maassen et al.'s (2003) study in which children with CAS showed impaired auditory perception of vowels, suggesting that production difficulties in children with apraxic speech are related to target representations of vowels. These results are similar to the study by Groenen et al. (1996), who found no difference in categorical identification between CAS and TD, but poorer discrimination in CAS children. In addition, children with CAS have generally been shown to have difficulty detecting differences in vowel duration (Ingram, Reed & Powell, 2019).

In the present study, children with CAS were found to have difficulty distinguishing two specific vowel pairs. The first pair is /o/-/u/. Both /o/ and /u/ have the back position of the tongue and are relatively close regarding height (/o/ middle; /u/ high), yet this pair was also difficult for TD children. In addition, Lenoci et al. (2020) also found individual vowel production variation not only in CAS children but also in TD children's speech in Italian. However, the /e/-/i/ pair was difficult only for CAS children (13/30 did not differentiate these vowels). Similar to the previous pair, this pair also shares the position of the tongue (front) and height (/e/ middle; /i/ high). In this study, however, no significant interaction was found between vowel height and backness and group, in contrast to the study by Lenoci et al. (2020), which suggests that vowels are more centralized and particularly less distinct along the height dimension, indicating a critical nature of height distinctions for speakers with CAS. However, the difference between these two studies is that Lenoci et al. (2020) investigated the consistency of vowel production, whereas the present study tested only vowel discrimination. Further studies should therefore include both vowel discrimination and vowel production to better understand whether the inconsistencies in vowel production are a consequence of poor vowel discrimination or vice versa.

Furthermore, Zuk et al. (2018) showed no measurable differences between children with CAS and their typically developing peers on speech perception tasks when children with co-occurring language impairment were excluded from the sample (Zuk et al., 2018). On the contrary, Nijland (2009) did control for language comprehension and still found that both children with CAS and children with phonological disorders performed worse on the discrimination task compared to TD children. Similar to Nijland (2009), the present study also found differences between TD and CAS children on the vowel discrimination task when controlling for language comprehension.

These equivocal findings could be explained by differences in the phonological systems of different languages. Lenoci et al.'s (2020) stated that vowel impairments need to be documented in speakers with different vowel systems in order to draw more general conclusions about auditory perception of vowels in CAS. For instance, vowel spaces in non-German languages are overall less crowded, and their diphthongs tend to be more distinct (Lenoci et al., 2020). In contrast to Croatian (non-German language), which consists of five vowels and one diphthong (Figure 20), Dutch has a more complex and crowded vowel structure with monophthongs and diphthongs (Figure 21). In this complex Dutch vowel system, there is less contrast between high and low and front and back vowels, making it more difficult for children to differentiate vowels. For Croatian children discrimination is easier because there are fewer vowels and they are less crowded which makes them easier to differentiate. Regardless, Croatian children with CAS still show difficulty in vowel discrimination.

This study suggests that vowel discrimination may be an important feature in CAS diagnosis and adds to the body of research on speakers of languages with less crowded vowel systems such as Italian (Lenoci et al., 2020).

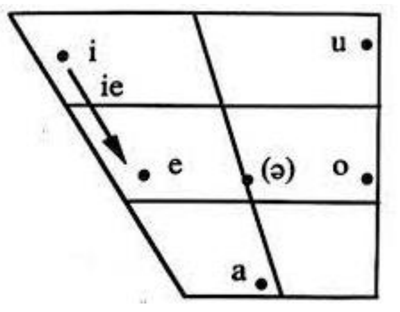


Figure 20. Vowel chart for the Standard Croatian (Landau et al., 1999; p.67)

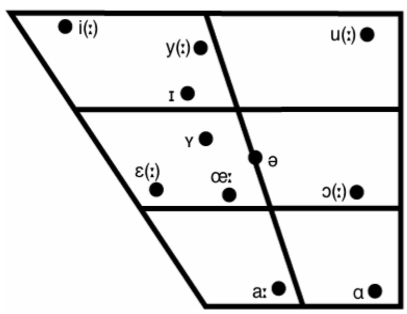


Figure 21. Vowel chart for the Northern Standard Dutch (Gussenhoven, 1992; p. 47)

In addition, phonological and phonemic awareness were also investigated in the present study. Phonological awareness refers to the ability to manipulate units of speech larger than a single phoneme, such as words, syllables, onsets, and rhymes (the vowel and remaining consonants in a word or syllable). Phonemic awareness, on the other hand, is a special case of the more general phonological awareness (at the phoneme level) and refers to the ability to discriminate and manipulate phonemes. These two phonological abilities refer to a continuum that includes attending to, recognizing, discrimination, and manipulating lexical representations (Pufpaff, 2009). Phonological skills are thought to underlie speech, language, reading, and spelling, and poor phonological awareness can lead to inaccurate mappings of phonemes to graphemes (Lewis et al., 2018). Phonological skills require the successful storage, retrieval, and manipulation of phonological codes, and unlike phonological representations, they require additional cognitive processes such as attention, metacognitive processes, and working memory (Ramus et al., 2013), and to develop phonological awareness, one must have good phonological skills.

Phonological difficulties at the phoneme, rhyme, and syllable levels have been reported in children with CAS compared to their typically developing peers on receptive and expressive tasks (Stackhouse & Snowling, 1992; Marion et al., 1993; Marquardt et al. 2002; McNeil et al., 2009; Nijland, 2009). Stackhouse and Snowling (1992) first highlighted the involvement of lexical representation in rhyme studies and indicated that the deficit in phonological representation may interfere with the development of phonological awareness in children with CAS. Their study showed impaired rhyme production and rhyme identification skills in two CAS children aged 10;07 and 11;00 years at baseline and four years later at follow-up. Similar to these findings, Marion et al. (1993) found severe deficits in rhyme production and rhyme identification in four CAS children aged 5 to 7 years compared to TD children, leading them to conclude that deficits in speech motor programming cannot explain such phonological incompetence. Even when controlling for language comprehension, Nijland (2009) found that children with CAS had significantly lower scores on the rhyming task compared to the typically developing children. The results of the present study confirmed the previously mentioned findings. Children with CAS showed poorer performance on rhyme discrimination and rhyme production compared to TD children, but their group average on rhyme tasks was still within the typical developmental range for preschool children based on test norms. This suggests that rhyme difficulties in children with CAS, are not a difficulty but rather a diversity that can be explained by their core problem - assembling the phonological plan and its implementation.

Furthermore, Marquardt et al. (2002) addressed the issue of syllable structure, i.e., whether children with CAS show age-appropriate metalinguistic awareness of the syllable. They studied three children with CAS between the ages of 6 and 8 years and three TD children of the same age. Each child had to detect syllables, judge intrasyllabic position, and judge intrasyllabic structure. A general pattern emerged in all tasks: the CAS children performed poorer. However, they performed better on syllable detection then on the other tasks (i.e., judging intrasyllabic position and judging intrasyllabic structure), with one CAS child achieving the same accuracy as the TD children. This participant had a significantly higher percentile rank on the Peabody Picture Vocabulary Test-Revised, a normal developmental history, and the highest score on the cognitive test of all three children with CAS. The present study examined the ability to blend syllables in words and segment a target word into syllables. The results showed that children with CAS performed worse on both tasks than TD children. Still, CAS children were similar to TD in that both groups were most successful in the syllable synthesis task (averaging 6 correct words out of 7 presented), a task similar to syllable detection task in the study by Marquardt et al. (2002).

As the phonological complexity of the tasks increased, children with CAS had more difficulty completing the tasks, but this was developmentally expected, as children's sensitivity to phonological units follows a hierarchy that begins with syllable awareness (at age 3 to 4 years), followed by rhyme awareness (at age 4 to 5 years), and lastly phoneme awareness (Goswami & Bryant, 1990). In a study by Kuvač Kraljević et al. (2020), a factor analysis examining the rhyme variable (recognition and production) suggested that rhyme was evenly distributed between syllabic and phonemic awareness, representing shallow and deep phonological awareness, and thus could play a role in the transition from syllabic to phonemic awareness during the preschool years. Further analyses confirmed that syllabic phonological awareness reflects shallow phonological awareness and phonemic awareness reflects deep phonological awareness.

In the present study, as expected, children with CAS had the most difficulty with the phonemic task, in which they successfully segmented an average of 1.6 out of 7 words, whereas TD children segmented an average of 5.8 words. Phonemic awareness represents the endpoint of phonological awareness and reflects the child's ability to organize at the finest level of phonology. This may be of concern, as phoneme awareness is considered a better predictor of later reading skills than awareness of larger sound units (Hulme et al., 2002). In addition, McNeil et al. (2009) compared the phonological abilities of children with CAS, children with inconsistent speech disorder (ISD), and TD children. TD children performed better on rhyme awareness, alliteration, and phoneme identity than CAS and ISD children, as expected. Although both groups exhibited comparable receptive vocabulary levels, CAS children performed more poorly than the ISD children on the phonological awareness measures, but no difference was found between the groups on the letter knowledge and reading measures. Furthermore, there was no difference between CAS and ISD children on phonological representation judgement task, with both groups performing poorer then TD children. Researchers indicated that children with CAS are likely to experience more severe phonological awareness deficits than children with other SSD. In contrast to previous studies, children with a standard score of less than 77 on the PPVT receptive vocabulary test were not included in this study, meaning that phonological awareness of participants in this study could not be attributed to significant receptive vocabulary deficits (i.e. receptive language).

Furthermore, children who fail to use phonological information efficiently for decoding are more likely to have persistent reading difficulties (Hogan, Catts & Little, 2005). Using this premise, Lewis et al. (2004) compared the school-age outcomes for children with CAS, SSD only (SSD), and SSD with concomitant language impairment (SSD + LI). Results showed that speech articulation and single word decoding were significantly reduced in children with CAS compared to children with SSD and children with SSD + LI. The severity, persistence, and inconsistency of the speech production difficulties observed with CAS may account for the greater reading difficulties in these children. However, there was no control for language impairment in the CAS group, unlike in the SSD group, in which criteria for language impairment included a scale score of less than 8 on two or more subtests of the Test of Language Development - Primary. In their later study (Lewis et. al, 2018), they confirmed that participants with severe disorders such as CAS + LI in middle childhood and adolescence performed worse on a spelling measure than other participants, including those with other forms of SSD (SSD-only and SSD + LI /no CAS) and those without these disorders (no SSD/LI). Similar to previous studies, there was no CAS-only group (i.e. without language impairment), which is very important because not all children with CAS have concomitant language difficulties and consequent reading problems, although a more recent study by Miller et al. (2019), which examined school-aged children and adolescents (ages 7-18) diagnosed with either CAS or SSD who were not CAS, showed that 65% of participants with CAS compared with 24% of participants with other SSD were classified as low-proficiency readers based on nonsense and single word decoding.

In the present study, children with CAS performed significantly worse than TD children on all phonological awareness tasks. However, their performance on the rhyme task was within the typical developmental range for pre-schoolers, and they were also quite successful on the syllable task, suggesting that CAS children can process larger phonological units. For example, if a child runs more slowly than the other children in the class, it does not automatically mean that the child has a motor impairment, only that he or she runs somewhat more slowly. A similar analogy can be applied to children with CAS - their performance on the rhyme tasks does not automatically indicate an impairment, but rather that they are somewhere within the normal range, as if they were "running slower" i.e. delayed rather than deviant. However, they showed considerable difficulty at the phonemic level, suggesting that CAS children have problems with more precise processing of the smallest unit - the phoneme.

5.1.2. Phonological planning in children with CAS

According to Ozanne (1995), CAS results from an impairment somewhere in the transition from word form retrieval to final articulo-motor output. Most authors agree that a core feature of CAS is inconsistency, suggesting a processing rather than a representational deficit (Nijland, 2003). Difficulty in assembling the phonological plan for a word or utterance is reflected as speech inconsistency. Although the analysis of speech inconsistency might seem straightforward, things are not so simple. As Iuzzini-Seigel et al. (2017) point out, assessing speech inconsistency is a research challenge that first requires choosing the appropriate level of analysis (e.g., token-to-token vs. phonemic) and other stimulus features (e.g., short words vs. longer words or phrases, words vs. non-words). In the present study, token-to-token inconsistency was chosen as the most efficient method for measuring whole-word variability (Ingram & Ingram, 2001) in multisyllabic words and non-words because children with CAS often produce more errors as the number of syllables increases (Davis et al., 1998). Other functions such as verbal short-term memory and attention also contribute to phonological planning, but the contribution of memory processes in responding to non-word repetition tasks is beyond the scope of this study. Nevertheless, the results of these two tasks show a very high positive correlation, suggesting that the same construct is being studied.

Although occasional errors in multisyllabic segmental accuracy are to be expected even in TD children (Benway & Preston, 2020), for example, TD children in the present study showed slightly more than one form in five repetitions (1.15 for words and 1.25 for non-words). However, the present study also confirmed that children with CAS showed statistically greater variability in both word and non-word repetition tasks (2.15 forms in five word repetitions and 2.55 in non-word repetitions), in contrast to TD children who were significantly more consistent in their production.

Similar to a study by Iuzzini-Seigel et al. (2017), the present study examined only multisyllabic words and non-words with complex structure. In their study, token-to-token inconsistency was calculated based on two repetitions, and all impaired groups were more inconsistent on multisyllabic words than the TD group; however, children with CAS and CAS and language impairment (LI) were more inconsistent than children with only LI. Both studies confirm the findings of Marquardt et al. (2004), showing that the overall token and error token variability in children with CAS was high compared with expected levels of functioning in TD children.

It has been argued that tasks such as rapid automatized naming (RAN) address several cognitive abilities, including rapid access to phonological representations and retrieval of phonological information (Alves et al., 2016). There are not many studies examining RAN in CAS children. Lewis et. al (2018) showed that the CAS + LI group had significantly lower scores on RAN than the no SSD/LI and SSD only groups. The present study provided similar results where CAS children (without language impairment) scored lower on the RAN test than the TD group, suggesting difficulties in retrieving phonological information and efficiently assembling a phonological plan.

Another task targeting phonological planning was the novel word learning task, which targets output processing and phonological planning (Dodd, Holm, Crosbie & McIntosh, 2010). As supported by a number of researchers (Ballard, Robin, McCabe & McDonald, 2010; Van der Merwe, 2011), novel word stimuli can be used to directly address underlying planning deficits. Several research studies have shown that children with CAS often have difficulty generalizing newly acquired sounds to new speech contexts. This is evident in studies of CAS treatment when segments that are produced accurately within treatment stimuli do not remain accurate when combined with other segments in different words (Ballard et al., 2010; Davis et al., 1998; Grigos & Kolenda, 2010; Maas, Butalla & Farinella, 2012; Case & Grigos, 2018).

The novel word learning task differs from non-word repetition because the cognitive load is higher - the child must store information while engaging in other cognitively demanding activities, and then create, sort, and select the correct representation and create a phonetic program for a given word (Alt et al., 2019). The Dodd et al. (2010) study used an appealingly illustrated book with the story "The Three Little Pigs" in which each pig was given a two-syllable nonsense name; production of each test word was elicited five times. The same methodology was used in the present study.

The inconsistent speech disordered group in the study of Dodd (2005) performed worse on the expressive naming task than the other three groups. The control, delayed, and consistent groups performed equally well. However, no CAS group was included in this study. They concluded that children who make inconsistent errors have a more general motor planning problem. By this definition, CAS children and the group of children with inconsistent speech disorders might have the same type of deficit, which raises the question of the nature of their deficit and whether they represent different diagnostic categories at all, especially knowing that "pure" cases are very rare and, as Maassen and Terband (2015) noted, differences between developmental speech disorders are more a matter of degree of involvement than diagnostic categories.

In the present study, children with CAS performed worse on the novel word learning task, exhibiting difficulties in both target word correctness and consistency, similar to the inconsistent speech disordered group in the Dodd et al. (2010) study. Children with CAS had difficulty remaining consistent especially in the repetition of the word Lugo, where the movement starts at the alveolar ridge and ends at the posterior part of the tongue against the soft palate, which made motor execution very difficult, and only in this particular task correctness and consistency did not correlate. Similar results were presents in Bradford and Dodd (2009) study where they found that CAS children made significantly more errors than TD children when imitating new words, and that they did not improve their accuracy with successive imitation attempts.

Although this task had a much higher cognitive load, it highly correlated with consistency in the non-word repetition task and weakly to moderately with the word repetition task, leading us to conclude that the phonological component is the most error-prone and that children with CAS have difficulty selecting and sequencing phonemes (i.e., assembling a phonological template or plan for producing an utterance). Presented findings add to the growing body of evidence on linguistic components of the disorder, confirming that children with CAS have difficulties with phonological planning, which represents a linguistic aspect according to the speech processing model, and that speech inconsistency is one of the central features of the CAS profile (e.g., Marquardt et al., 2004; Iuzzini-Seigel et al., 2017).

5.1.3. Consonant deletion and substitution

According to Ozanne (1995), children with CAS may have difficulty selecting and sequencing phonemes (i.e., assembling a phonological template or plan for producing an utterance). Under this premise, consonant deletion and substitution are both phonological processes that represent difficulties at the level of phonetic programming. They indicate that a child has a problem translating an abstract phonological code into motor speech in which he or she either omits or substitutes the target phoneme. Different studies suggest that consonant errors affecting syllable structure (e.g. syllable reduction, initial and final consonant deletion) are more common in CAS than in other speech sound disorders (Shriberg et al., 1997b; Lewis et al., 2004; Jacks et al., 2006).

Shriberg et al. (1997b) reported that 42% of consonant errors were omissions in younger children with CAS (4;10 to 7;00 years) compared to 25% in children with speech delay. Children with CAS in the present study also exhibit more consonant deletions throughout their production compared to TD children. More precisely the present study showed that children with CAS had statistically more errors in consonants in syllable-final position (PCCF=54%) and syllable-initial position (PCCI=76%) than TD children (PCCF=95%, PCCI=99%). In the Jacks et al. (2006) study, simple monosyllables were found to be the only syllable form that was consistently produced correctly, while omission errors were almost exclusively due to the deletion of the final consonant in words, regardless of the number of syllables, reflecting a deficit in syllable construction rather than sound-specific errors, resulting in a consistent pattern of syllable errors. The present study also confirmed that the distribution of phonemic errors was not uniform across syllable positions, with children with CAS omitting or substituting nearly half of the consonants in syllable-final position. Furthermore, 73.5% of syllable structures in the Croatian corpus have CV and CCV structures (both ending in a vowel). Based on this language specificity, it can be concluded that children with CAS have difficulty with phonetic programming of less frequent and more complex syllable structures and clusters of Croatian (CVC, CCVC, CVCC, CCCV), confirming that the error rate increases with increasing utterance complexity (Maassen, 2002). Similar observations have been made in adults with apraxia of speech (AOS); Staiger and Ziegler (2008) found a significant effect of syllable complexity; moreover, AOS patients made 54% more errors on complex (occurrence of at least one cluster) than on simple low-frequency syllables.

Considering those are complex syllables and consonant clusters, we cannot be sure that these errors are exclusively due to a disorder of phonetic programming; they could also be caused by a disruption at higher levels of processing, e.g., an unstable phonological representation or phonological planning of a complex production. Ozanne (1995) reached a similar conclusion, stating that consonant deletions alone are not sufficient evidence for a phonetic programming deficit.

5.1.4. Multiple repetition rates in children with CAS

A breakdown at the level of motor speech programme implementation occurs when the correct motor programme is selected but the wrong timing and force parameters are chosen (Schmidt and Lee, 1999). Diadohokinetic rate (DDK) or maximum repetition rate (MRR) is one of the few objective assessments of motor speech performance used in differential diagnosis to assess breakdowns in underlying motor planning and programming difficulties specifically in CAS differentiation (Thoonen et al., 1996; 1999; Shriberg et al., 2010; 2012).

Although there are various MRR protocols, two task types are most common: alternating motion rates (AMR) - repetition of monosyllabic sequences (/pa/, /ta/, /ka/) and sequential motion rates (SMR) - repetition of multisyllabic sequences (/pata/, /taka/, /pataka/). Both tasks primarily address the transition between sounds and syllables, and aim at motor programming in which spatial and temporal targets for articulatory movements are translated into context-dependent motor specifications for the articulators, which is considered problematic in children with CAS (Nijland et al., 2003).

Present study showed that children with CAS were indeed statistically slower on the mono-, bi-, and trisyllabic sequences). The monosyllabic performances showed a high correlation with the bisyllabic performances and a slightly lower but still high correlation with the trisyllabic performances. The problem with MRR is that there are no universal rates for TD children as a reference point, so it is difficult to determine whether the sequence presented is statistically slower than expected for a given age. However, there have been initial attempts to investigate the universality of speech rates and whether they might be language-independent. For example, research by Diepeveen, Knežević, and Maassen (2022) on differences between TD children in MRR in different languages showed that there were no significant differences between Croatian, German, and Dutch children (5-6 years), except for the sequence /ta/ between Croatian and German. Further research is needed to determine if the norms of one language are transferable to another language. Present study showed that all children with CAS could produce monosyllabic sequences, but more than half of them (16 out of 30) could not produce the trisyllabic sequence, just as in the Thoonen et al. (1996) study, where not all children with CAS could produce a correct trisyllabic sequence, as originally expected by Aram and Glasson (1979). The trisillabic sequence is more difficult to execute because it requires rapid and accurate sequencing and a front-to-back transition, using the lips, tip of the tongue, and back of the tongue to produce the consonants. Although the trisillabic sequence is more difficult to produce, none of the TD children in this study had problems with it, just as in the study by Thoonen et al. (1996). This could explain the fact that performance on monosyllabic tasks in children with CAS does not correlate with bi- and trisyllabic tasks, whereas in TD children, monosyllabic performance highly correlates with bisyllabic performance and moderately with trisyllabic performance. Although children with CAS can successfully produce monosyllabic sequences in the present study, their rates were still significantly slower than the rates of the TD children, which is in contrast to the results of Thoonen et al. (1996).

These results are consistent with the second study by Thoonen et al. (1999), in which they concluded that children can be reliably identified with CAS primarily because they are unable to produce the trisyllabic sequence or because they have a slow rate; sensitivity was reported as 100% and specificity as 91%. More recently, Murray, McCabe, Heard, and Ballard (2015) advised using an oral motor assessment, including the trisyllabic sequence /pataka/, to diagnose CAS. In addition, these findings are consistent with a study by Meloni et al. (2020) of French-speaking children with CAS, showing that they can be discriminated from TD children on the MRR task, whereas children with PD cannot. These studies suggest that MRR rate seems to be a relevant marker for CAS in French speakers, as well as in English, Dutch, and Croatian speakers, regardless of language.

On the other hand, the study by Williams (2015) rather MRR is a marker for speech difficulty in general and not a specific marker for CAS. A very recent study has shown that while both non-word imitation and three-syllable MRR may have utility in identifying CAS, their concurrent validity is not high (Preston, Benway, Leece & Caballero, 2021). However, in this study, a high positive correlation was found between the bi- and tri-syllable MRR and consistency in word and non-word repetition, suggesting an underlying deficit in both phonological planning and motor programming in children with CAS.

Although only rate (syllables per second) was used as a measure in the present study, Yaruss and Logan (2002) reported that very few disfluencies occurred in the transcripts of MRR productions of typically developing children aged 3-7 years, whereas MRR consistency might allow us to distinguish children who use consistent simplification patterns from those who are inconsistent in their responses. Further studies should focus on both rate and fluency and consistency of the sequences produced to gain a more comprehensive insight.

5.1.5. Accurate phoneme production in children with CAS

Children with CAS have difficulties producing sounds and syllables consistently and precisely in order to speak words and sentences in a clear manner (Morgan, Murray & Liégeois, 2018). When speech symptoms are reported in CAS, they usually involve a restricted phonemic repertoire, vowel errors, and unusual or atypical speech errors (ASHA, 2007). Crary, Landess, and Towne (1984) reported a high frequency of consonant omission, cluster reduction, and consonant substitution in the speech of children with CAS. Later, Thoonen et al. (1997) reported a large difference between CAS and TD, finding that children with CAS produced an overall higher rate of consonant errors (substitutions, omissions, distortions) and cluster errors (cluster reductions) than TD children. In a very recent study, Chenausky et al. (2022) found that consonant errors accounted for most of the variance in the severity and intelligibility of single-word speech, which is largely consistent with previous work.

The results presented in this study are consistent with the previously mentioned studies. Children with CAS differed in consonant (PCC = 69.6%) and vowel (PVC = 93.2%) accuracy from TD children, as expected. Grigos and Case (2018) reported very similar decreased consonant (PCC = 69.7%) and vowel accuracy (PVC = 86.1%) in American children with CAS at five to six years of age in connected speech. Moreover, Barrett, McCabe, Masso, and Preston (2020) found that there was a strong correlation between PCC in single words and PCC in connected speech in children with CAS. The average PCC in their study was 80 and PVC was 88 (Barrett et al., 2020).

In the present study, vowel accuracy showed a high positive correlation with non-word repetition and word repetition, while consonant accuracy showed an even higher positive correlation with non-word repetition and word repetition. However, when focusing only on CAS children, vowel accuracy correlated only moderately with word repetition and not at all with non-word repetition, while consonant accuracy correlated highly with both word and non-word repetition. This is in contrast to a study by Chenausky et al. (2022), in which they reported that vowel errors were significantly correlated with single-word severity and intelligibility and may function as a pathognomonic feature of CAS, or to the study by Malmenholt, McAllister, Lohmander, and Östberg (2022), in which a measure of vowel consistency showed promising potential for discriminating between Swedish-speaking children with and without CAS.

Although showing significant differences compared to TD children, Croatian children with CAS seem to have fewer problems with vowels than expected. This is not consistent with Randazzo's (2019) study, which showed that vowel errors are the third most important distinguishing feature for CAS among clinicians, and some authors argue that vowel errors are a key diagnostic feature of CAS (e.g., Jacks et al., 2013). Since more than 75% of CAS studies have been conducted on English-speaking children (Murray et al. 2021), it is quite difficult to determine which markers from English are relevant in other languages, as mentioned by Meloni et al. (2020), because differences between phonological systems may limit the applicability of markers. Although we found that Croatian children with CAS show impairment in auditory perception of vowels, they still show higher vowel accuracy than children in other languages. Further studies are needed to determine whether vowel errors may be the central diagnostic feature for Croatian children, as hypothesised on the basis of English CAS studies.

Comparing the PCC index scores proposed by Shriberg and Kwiatkowski (1982), which include four levels of severity: mild (more than 85% of consonants correct), mild to moderate (between 85% and 65%), moderate (between 50% and 65%), and severe (below 50%), children with CAS would correspond to mild to moderate phonological disorder. In addition, lower consonant accuracy would also be expected in other children with SSD and motor speech disorders, so this feature alone cannot contribute to the differential diagnosis of CAS, but should always be included. A more detailed look would potentially give us a deeper insight into consonant errors, but this is beyond the scope of this paper.

5.2. Speech motor performance in phonological awareness tasks

Franck Ramus (Ramus & Szenkovits, 2008; p. 129) asked himself an important question, „*If at least some individuals with dyslexia had auditory deficits, how could I expect them to perform normally in phonological tasks requiring auditory perception of the stimuli, and how would I be able to unambiguously interpret my data*? ". A similar question could be applied to children with CAS.

It is well known that all phonological awareness tasks have three main features: (a) the linguistic nature of the stimuli, (b) the phonological complexity of the stimuli, and (c) the response mode (Cunningham et al., 2015), and that performance variability can result from the effects of each factor, children with CAS are in a potentially unfavourable situation when considering the response mode-which is almost always a verbal response. As Ramus and Szenkovits (2008) noted, choosing the appropriate task was not trivial when examining different aspects of phonology. Although the previous section provided data demonstrating that children with CAS exhibit difficulties in phonological processing and phonological awareness, as suggested by previous research (Marion et al., 1993; Marquardt et al., 2002; McNeill et al., 2008), one of the the sources of uncertainty is the response mode in phonological tasks, which may influence associations with the outcome.

In Croatian, the available standardized phonological tasks all require a verbal response (PredČiP; Kuvač Kraljević & Lenček, 2012); for the purposes of this study, phonological tasks (syllable and phonemic blending) were reconstructed to not require a verbal response, a methodology similar to the study by Janssen et al. (2016). For this reason, the matched control group was used for comparison rather than the available norms. In order to control for the phonological rather than the motor (articulatory) aspect of this task, errors in speech production (omissions, substitutions, distortions) were not scored as errors in phonological awareness if a child showed them continuously.

Not all phonological awareness tasks are equally easy or difficult; they follow a specific hierarchy that begins with syllable blending, followed by rhyme recognition, syllable segmentation, rhyme production, phonemic blending, and phonemic segmentation (Kuvač Kraljević et al., 2020). These tasks represent three different levels of phonological awareness - the syllable level (shallow), the rhyme level (shallow-deep), and the phonemic level (deep). For each of these levels, there are two tasks: the easier one like blending, which also does not require a verbal response, and the more difficult one like segmentation, which does require a verbal response. TD children are expected to perform better on phonological tasks that require synthesis (or blending) because they developmentally precede segmentation tasks.

Although Stackhouse and Wells (1997) hypothesised long ago that poor speech production may affect accurate encoding of complex words, which in turn affects the child's developing lexicon and language system, the unique contribution of speech motor performance to phonological awareness in children with CAS remains unknown.

The present study confirmed that all children performed better in rhyme recognition and phoneme blending than in rhyme production and phoneme segmentation, with a significant main effect of speech motor production (verbal response) in both groups. However, there was no difference between syllable blending and segmentation in either group. This could mean that children with TD and children with CAS have the same level of phonological awareness at the shallow level. (i.e., that they have mastered the level of syllabic phonemic awareness), which is not the case for the higher levels (i.e., rhyme and phonemic awareness).

Although present in both groups, the effect of speech motor production was much more pronounced in children with CAS. They showed a significant higher success in easier phonological tasks without verbal response (rhyme recognition and phonemic blending); i.e., they were significantly more successful as long as they did not have to implement motor planning and programming (verbal response). This contrasts with participants with dyslexia, who had more difficulty on discrimination tasks than on repetition tasks, highlighting their deficit in input representation (Ramus & Szenkovits, 2008).

These results suggest that children with CAS have more difficulty at deeper levels of phonological awareness and with more difficult tasks such as segmentation of smaller units requiring verbal response. Thus, one might conclude that having to perform a speech task affects the phonological processing in children with CAS, which in turn may affect the linguistic levels of speech production processes indicating a close relationship between phonological awareness and motor aspects of speech production at the deeper levels of phonological awareness. Although further investigation of segmentation and blending is needed to draw conclusions, these results are somewhat consistent with the idea of a "flow-back" effect suggested by the Cascade speech production process, in which motor deficits in children with CAS may affect phonological development (Ozanne, 2010), as well as with the previously mentioned hypothesis of Stackhouse and Wells (1997). Miller et al. (2019) recently showed that significant predictors of low reading proficiency in SSD and CAS groups were oral language, phonological awareness, multisyllabic word repetition, and MRR, indicating that motor speech deficits may also increase risk for reading difficulties, although to a lesser degree. These results are not consistent with the theory that poor phonological awareness in children with CAS cannot be accounted for by motor speech planning difficulties (Marion et al. 1993, Marquardt et al. 2002) and that indistinct phonological representations underlie the speech, language, and reading difficulties associated with CAS proposed by McNeill et al. (2009).

In summary, indistinct phonological representations may indeed be the result of CAS and then play a role in further speech and language development and symptomatology, whereas phonological awareness and motor aspects of speech production may have a more complex relationship than the classical one direction hierarchical relationship.

5.3. Subgroups of CAS children

As mentioned earlier, the present study did not question the presence of motor aspects of this disorder, but rather aimed to investigate the entire speech production process, taking a more process-oriented approach. There are many previously mentioned studies that have characterized the speech deficits associated with CAS, but there are fewer studies that have explicitly and systematically examined these other clinical features, particularly the phonological aspects that might help us understand the core problems in CAS and potentially play a role in the further development of speech and the symptomatology.

Previous attempts to divide children with SSD into subgroups relied on various classifications of features of the child's speech, and there was one factor that seemed to distinguish subgroups of SSD-the presence or absence of comorbid LI (Lewis et al., 2011). Similarly, the classification of CAS into subgroups has long been of interest because it can lead to better differential diagnosis and more successful treatment (Stein et al., 2020), and it also suggests that children with similar types of errors share a common aetiology.

In their study, Stein et al. (2020) investigated whether there are comorbid subgroups within CAS that can be defined by language and reading ability and the presence of certain clinical symptoms. Their analysis revealed three subgroups with deficits of varying severity in language and reading skills. The results suggest that the severity of comorbid subtypes within CAS is manifested in deficits in language and phonological processing skills associated with reading impairment.

Nijland (2003) also tested the homogeneity of the groups by clustering individuals within the research groups based on motor functioning, memory, sensory functioning, and attention. Although cluster analyses showed that the CAS group was more heterogeneous than the TD group, they were more heterogeneous on most cognitive functions, except for sequential tasks, where they were homogeneously impaired.

In the present study, we investigated whether there was a subgroup within the CAS group that could be identified by the presence of difficulties in phonological abilities. Similarly, to Stein et al. (2020), this study also revealed 3 clusters of children with CAS. Cluster 1 represents mild group that exhibits better results on deep phonology (rhyme production and syllable segmentation) and correct consonant productions. Cluster 3 represents moderate group that exhibits somewhat poorer results than cluster 1 and similar results to cluster 2 on rhyme production and syllable segmentation. However, they exhibit better results than cluster 2 on correct consonant productions and bi- and trisyllabic repetition rates. Cluster 2 represents severe group that exhibits the poorest results on mentioned measures. There was no significant difference across clusters with respect to age. On other variables, these three groups showed similar performance, suggesting that the difficulties in phonological planning and motor programming are the same for all three subgroups. Although there were no significant differences on other variables, consonant productions are one of the core features of CAS and highly correlate with several other parameters such as word and non-word consistency and MRR, suggesting that the groups may differ in terms of the severity of phonetic planning and motor programming. The group that showed significantly better results in the phonological awareness tasks (rhyme production and phoneme segmentation) also performed best in consonant production, indicating once again the complex relationship between phonological and motor aspects of speech production.

Although our findings support the notion of CAS as a heterogeneous disorder, for which symptoms have been reported to vary considerably across individuals and over the course of development (Iuzzini-Siegel et al., 2017), this study found that these three CAS subgroups are still a fairly homogeneous group, exhibiting similar difficulties, especially in aspects of phonological planning and motor programming, while differing in their phonological abilities and consonant production, which cannot be attributed to difficulties with receptive language. In the present study, the idea of comorbidity of CAS and language impairment is not discarded, but the main aim was to investigate CAS as a primary speech motor disorder with phonological features that are not due to a higher level language impairment in order to better understand the relationship between higher and lower levels of speech process in CAS, which is why we controlled for receptive language.

In the absence of accepted pathognomic diagnostic criteria, it is not surprising that there is a high degree of clinical disagreement among practicing SLPs regarding their criteria for diagnosing CAS (Forrest, 2003), leading to the variability/heterogeneity of diagnoses across different studies. As suggested in previous studies (Malmenholt, Lohmander, & McAllister, 2017), in addition to an invitation to participate in this study, SLPs in this study were sent a detailed description of the core diagnostic criteria and accompanying difficulties (with examples) recommended in the CAS literature (including some phonological aspects) before participants were recruited. Three core features suggested by ASHA had to be met, whereas additional phonological aspects were not crucial for participant inclusion (i.e., they were not mandatory). This inclusion of the phonological aspects may have helped to create a more homogeneous group of children with CAS. This was recognized as an important step because a previous study on CAS features in Croatia showed that out of 70 SPLs, only 25.7% recognized phonological difficulties and 35.5% recognized cluster reduction as a CAS feature (Blaži, Knežević, Šarić & Blaži, 2019). The inclusion of phonological difficulties in CAS description allowed us to see a bigger picture and make them a common feature of CAS in Croatian - not an exclusion criterion. Considering the course of speech development, and the fact that a particular deficit can have effects on other levels of processing (Maassen & Terband, 2015), the use of the checklist approach that do not include phonological deficits limits participants with CAS to difficulties at the motor level, which makes it difficult to obtain a more comprehensive picture and to understand the possible effects of these difficulties on higher processes. If we focus only on the three features proposed by ASHA (2007), we run the risk of limiting our knowledge of CAS. As Terband (p. 150, 2011) has nicely explained – "*if a speech-motor disorder can lead to deficits at different levels of speech production, such as poor phonological processing and lexical representation, one could argue that CAS and phonological disorder more often than not occur in combination. This would imply that a phonological disorder should not be an exclusion criterion for the diagnosis of CAS, but rather be accepted as comorbidity.*".

5.4. Cascade model of speech output processing and CAS

Cascade model of speech output processing Ozanne (1995) proposed that children with CAS exhibit difficulties at three levels of motor speech control: phonological planning, phonetic programming, and speech motor control, but to be diagnosed with CAS, a child only needs to exhibit difficulties at the motor levels of the model (phonetic programming and motor programming). The presence of motor speech difficulties is indeed a necessary prerequisite for the diagnosis of CAS; however, several studies have shown that CAS is not limited to the presence of motor speech difficulties.

By comparing speech performances obtained from a battery targeting all processing stages, we were able to determine which processing stages were functioning normally and which were impaired (Maassen & Terband, 2015). Based on the data presented, we can conclude that the Cascade model of speech output processing is a good theoretical model for understanding CAS. By systematically applying this model, we conclude that, in contrast to Ozanne (2010), the children with CAS in the present study show difficulties at all levels of speech processing, as shown in Table 32. Beginning with difficulties in discriminating vowels and extending to difficulties in auditory processing and phoneme selection and manipulation (i.e., phonemic awareness). In addition, they show significant whole-word variability in word and non-word repetition tasks, as well as in the novel word task, where they also provide fewer correct productions. Finally, they produce fewer items per second on the RAN task. The transcription analysis showed that they have difficulty sequencing and producing consonants in syllable-initial and syllable-final positions. Also, as expected, they have significantly lower maximum repetition rates at all three levels (monosyllabic, bisyllabic, and trisyllabic repetition) and show less accurate phoneme production for both consonants and vowels compared to TD children whose speech patterns resemble those of adults. Thus, in the present study, we found evidence of deficits in CAS at both linguistic and motor levels (i.e., throughout the speech production process).

Table 32. Difficulties exhibited by children with CAS at different levels of the Cascade model of speech output processing.

Cascade model (Ozanne, 1995)	**Measured variables**	**Exhibiting difficulties**
Phonological rules	Phonological representations (vowels)	**yes**
	Phonological awareness	**yes**
Phonological planning	Rapid automatized naming	**yes**
	Whole-word variability	**yes**
	Novel word learning	**yes**
Phonetic programme assembly	Consonant deletion/substitution	**yes**
Motor programme implementation	Maximum repetition rate	**yes**
Speech execution	Accurate phoneme productions	**yes**

An important assumption of other models of speech production is that information travels between levels in one direction (e.g., Levelt, 1989), but this model assumes the possibility of bidirectionality. Moreover, further controlling for the receptive language and response mode in phonological tasks, as shown above, could provide a good empirical argument for a "flow back" effect of speech production (i.e., lower levels) on phonological processes (i.e., higher levels), indicating that motor deficits can affect phonological development and not just vice versa. These findings justify Maassen et al. (2010) to question the traditional notion of a separation between phonological and motor issues. Moreover, these findings are not only theoretically important, but could also lead to treatment that targets the specific underlying impairment, which is ultimately most important.

6. VERIFICATION OF THE HYPOTHESES

H1: Children with CAS perform worse at all levels of speech production.

This hypothesis is accepted (see below).

H1.1: Children with CAS perform worse at the phonological rules level, i.e., there is a significant difference in the AX discrimination task and the phonological awareness tasks.

This hypothesis is accepted. Significant differences were found in discriminating vowels, and on all phonological awareness tasks children with CAS children performing worse.

H1.2: Children with CAS perform worse at the level of assembling the phonological plan for the word or utterance; that is, there is a significant difference in the RAN test, the word repetition task, the non-word repetition task, and the novel word learning task.

This hypothesis is accepted. The results showed that children with CAS performed worse on the RAN test, on word and non-word repetition task, and novel word learning task.

H1.3: Children with CAS perform worse at the level of phonetic programme assembly, i.e., there is a significant difference in the correctly produced consonants in syllable-final and syllable-initial positions as well as in the number of omitted consonants.

This hypothesis is accepted. The present study showed that children with CAS produced statistically more consonant omissions, errors in consonants in syllable-final position and syllable-initial position than TD children.

H1.4: Children with CAS perform worse at the level of implementation of the motor-speech programme, i.e., there is a significant difference in the MRR tasks, in monosyllabic, bisyllabic, and trisyllabic repetition.

This hypothesis is accepted. Children with CAS performed worse on all MRR tasks, i.e., they had statistically lower repetition rates on monosyllabic, bisyllabic, and trisyllabic repetition.

H1.5: Children with CAS perform worse at the level of speech execution; i.e., there is a significant difference in the accuracy of consonant (PCC) and vowel (PVC) production.

This hypothesis is accepted. Children with CAS had lower accuracy in the production of consonants and vowels.

H2: Children with CAS perform worse on phonological tasks requiring motor speech production (verbal response) than on phonological tasks not requiring motor speech production (nonverbal response), i.e., there is an interaction between motor speech production and phonological tasks.

This hypothesis is partially accepted. Children with CAS and TD children performed better on two tasks without motor speech production-rhyme recognition and phonemic blending-than on tasks with motor speech production-rhyme production and phonemic segmentation. However, there was no difference in the syllable task with respect to motor speech production. In addition, there was a significant interaction between motor speech production and phonological task, which was more pronounced in children with CAS.

H3: By identifying difficulties at the phonological levels of speech production, subgroup of children with CAS is formed.

This hypothesis is partially accepted. Three clusters were identified based on differences at the phonological level and the level of phonetic and motor programming.

7. LIMITATIONS AND IMPLICATIONS FOR FURTHER RESEARCH AND CLINICAL WORK

The present study once again underlined the complexity and multidimensionality of the speech production process in children with developmental speech disorders. And although it provided for the first time some valuable answers regarding CAS in Croatian, it opened new questions that need to be further explored.

The results of the present study show that children with CAS have difficulties at all levels of speech production, because we did not limit children with CAS to the speech motor aspect of the disorder, but also included the phonological aspect, as suggested in the literature. However, we did not include other developmental domains such as fine and gross motor skills, short-term memory, and procedural learning skills. Future research should go beyond speech motor abilities and examine other perceptuo-motor and cognitive functions, as speech motor and phonological abilities are not the only closely related functions (Nijland et. al, 2015).

In the present study, we investigated children with CAS without additional language impairment and compared their results with those of typically developing children. Because we did not include children with other speech sound disorders such as phonological disorders or with specific language disorders, we cannot draw conclusions about the specificity of the results for CAS. Further research should include more groups with developmental speech disorders.

Another limitation is that there are no standardized tests targeting different aspects of speech production in Croatian. For this study, several tasks and procedures were constructed and translated for Croatian, but they need further testing and validation to fully generalize the results and disseminate their use. However, this study was a response to a limitation mentioned in the study by van Haaften et al. (2019) regarding the Computer Articulation Instrument, which is currently only available in Dutch, and represents a first step towards the translation of CAI into other languages. However, further research is needed to evaluate this adaptation.

Future research should broaden our knowledge on phonological and reading abilities of school children with CAS, to better understand if phonological abilities are just delayed rather than deviant i.e. do they persist into school age and affect reading?

As noted earlier, over 75% of CAS studies were retrospective studies, case-control studies, and/or studies with English-speaking children (Murray et al. 2021). The present study contributes to a better understanding of CAS in languages other than English, while further studies should make a more detailed comparison of the structural commonalities and differences between English and Croatian in order to better understand which features from English are relevant in Croatian (e.g., vowel discrimination in Croatian children with CAS). Future research should implement similar measurements so that results are comparable across languages and continue to optimize assessment stimuli.

Clinicians have reported difficulties in identifying and differentiating CAS from other developmental disorders in several studies (Forest, 2003; Malmenholt et al., 2017); in the study by Blaži et al. (2019), 61.5% of Croatian SLPs reported difficulties in differentiating CAS from phonological disorders. This study confirms that children with CAS - in addition to the expected difficulties in the motor domain - also show difficulties with phonological abilities. Spencer, Davison, Boucher, and Zuk (2022) note that in addition to evidence-based intervention targeting accurate and consistent speech production for children with CAS, SLPs should always do a comprehensive assessment of speech perception, language, and emergent literacy skills. These findings will hopefully focus clinicians' attention on the inclusion of phonological skills in the assessment and treatment of CAS. In addition, we hope to shed light on the process-oriented approach as opposed to the checklist approach, as this approach gives a complete characterization of the speech profile so that underlying processing deficits can be identified and effectively treated (Maassen & Terband, 2015).

8. CONCLUSIONS

This study had three general objectives. Since this is the first study of CAS in Croatian, the first goal of this study was to explore speech motor and phonological skills and also to provide comprehensive data on CAS in another language with a set of speech tasks representing all the processes assumed in speech production. The second goal was to better understand the cascading effect of motor-speech difficulties on a child's phonological abilities, and the third goal was to distinguish subgroup of children with CAS based on their phonological abilities.

First, by comparing performance from a battery that targeted all levels of speech processing, the results showed that children with CAS performed worse compared to typically developing children at all levels of speech production. Consistent with the predictions of the Cascade model (Ozanne, 1995), this study confirmed that children with CAS exhibit difficulties at the proposed levels (i.e., phonological planning, phonetic programming, and speech motor skills) as well as in phonological representation and phonemic awareness (i.e., phonological rules) which represents the highest level of the model. As expected, children with CAS consistently showed difficulty with token-to-token inconsistency and retrieval of phonological information (i.e., phonological planning); they exhibit more consonant errors affecting syllable structure (i.e., phonetic programming); their monosyllabic, bisyllabic, and trisyllabic maximum repetition rates were significantly slower when they could produce the sequence (i.e., motor programming); they exhibit less accurate consonant and vowel production (i.e., speech execution). It is important to emphasize that these specific tasks are not unidimensional and that, for example, less accurate consonant and vowel productions can be attributed not only to difficulties in speech execution but also to phonological planning. However, for the purpose of testing the Cascade model, they were arranged as the model suggests (i.e., testing specific levels).

Second, the present study, along with the previously mentioned studies (Stackhouse & Snowling, 1992; Marion et al., 1993; Marquardt et al., 2002; McNeil et al., 2009), provided convincing evidence that children with CAS have difficulty with phonological abilities, although it was difficult to determine whether their performance was merely delayed or deviant. Moreover, by eliminating the effect of verbal response in common phonological abilities tasks, this study showed that the effect of speech motor production in phonological tasks was much more pronounced in children with CAS; that is, they were significantly more successful as long as they did not have to implement motor planning and programming (verbal response)-

providing an empirical argument for a "flow back" effect of speech production (i.e., at lower levels) on phonological processes (i.e., at higher levels).

Third, within the CAS group three subgroups were identified, evidencing differences on complex phonological awareness abilities (rhyme production and phonemic segmentation), bi and trisyllabic repetition rate, and correct consonant production (PCC, PCCI and PCCF). These three comorbid subgroups could be classified by severity as mild (cluster 1), moderate (cluster 3), and severe (cluster 2) based on their phonological awareness abilities and phonetic planning and motor programming

In summary, the results of the present study support the framework for speech processing proposed by Ozanne (1995; 2010) in that it allows for a more detailed view of the motor aspect of speech processing, but this study has revealed a broader range of difficulties in CAS than originally proposed by the Cascade model. Focusing on the speech features and viewing CAS only as a paediatric speech motor disorder one may miss the bigger picture and all the processes underlying these speech features. There is a specific, complex relationship between speech motor control and phonology and thus a strong association between deficits in both domains (Maassen et al., 2010), which means that one does not exclude the other and that phonological difficulties should not be considered an exclusion criterion for the diagnosis of CAS.

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9. REFERENCES

Alt, M., Arizmendi, G. D., Gray, S., Hogan, T. P., Green, S., & Cowan, N. (2019). Novel Word Learning in Children Who Are Bilingual: Comparison to Monolingual Peers. *Journal of speech, language, and hearing research: JSLHR*, *62*(7), 2332–2360. https://doi.org/10.1044/2019_JSLHR-L-18-0009

Alves, L. M., Siqueira, C. M., Ferreira, M., Alves, J. F., Lodi, D. F., Bicalho, L., & Celeste, L. C. (2016). Rapid Naming in Brazilian Students with Dyslexia and Attention Deficit Hyperactivity Disorder. *Frontiers in psychology*, *7*, 21. https://doi.org/10.3389/fpsyg.2016.00021

American Speech-Language-Hearing Association. (2007). *Childhood apraxia of speech* [Technical report]. http://www.asha.org/policy

Aram, D. M., & Glasson, C. (1979). *Developmental apraxia of speech*. Paper presented at the Annual Conventionof the American Speech-Language-Hearing Association,Los Angeles, CA.

Bahr, R. H. (2005). Differential diagnosis of severe speech disorders using speech gestures. *Topics in Language Disorders*, *25*(3), 254-265.

Ballard, K. J., Robin, D. A., McCabe, P., & McDonald, J. (2010). A treatment for dysprosody in childhood apraxia of speech. *Journal of speech, language, and hearing research: JSLHR*, *53*(5), 1227–1245. https://doi.org/10.1044/1092-4388(2010/09-0130)

Bankson, N. W., Bernthal, J. E., & Flipsen, P. (2017). Introduction to the Study of Speech Sound Disorders. In J. E. Bernthal, N. W. Bankson, & P. Flipsen JR. (Eds.), *Articulation and phonological disorders: Speech sound disorders in children* (8th ed., pp. 1-6). Pearson.

Barrett, C., McCabe, P., Masso, S., & Preston, J. (2020). Protocol for the Connected Speech Transcription of Children with Speech Disorders: An Example from Childhood Apraxia of Speech. *Folia phoniatrica et logopaedica: official organ of the International Association of Logopedics and Phoniatrics (IALP)*, *72*(2), 152–166. https://doi.org/10.1159/000500664

Beers, M. (1995). The phonology of normally developing and language impaired children. (Unpublished doctoral dissertation). University of Amsterdam, The Netherlands.

Benway, N. R., & Preston, J. L. (2020). Differences Between School-Aged Children with Apraxia of Speech and Other Speech Sound Disorders on Multisyllable Repetition. Perspectives of the ASHA special interest groups, 5(4), 794–808. https://doi.org/10.1044/2020_persp-19-00086

Berman H. B. (2022, August 08). *Hypothesis Test: Difference Between Means*. Stat Trek. https://stattrek.com/hypothesis-test/difference-in-means

Betz, S. K., & Stoel-Gammon, C. (2005). Measuring articulatory error consistency in children with developmental apraxia of speech. *Clinical linguistics & phonetics*, *19*(1), 53–66. https://doi.org/10.1080/02699200512331325791

Blaži, A., Knežević, D., Blaži, D. & Šarić, L. (2019) Typical characteristic for identification and assessment of childhood apraxia of speech in Croatia. In *The Abstract Book of Poster Presentations IALP 2019: Innovations in Supporting Communication Participation*.

Bowen, C. (2015). *Children's Speech Sound Disorders: Second Edition*. John Wiley & Sons

Bradford, A. & Dodd, B. (2009). Do all speech-disordered children have motor deficits? *Clinical Linguistics & Phonetics*, *10*(2), 77–101. https://doi.org/10.3109/02699209608985164

Broomfield, J., & Dodd, B. (2004). The nature of referred subtypes of primary speech disability. *Child Language Teaching and Therapy*, *20*(2), 135–151. https://doi.org/10.1191/0265659004ct267oa

Canault, M., Thai-van, H., & Le Normand, M. T. (2021). Simplification of syllable structure in childhood apraxia of speech: a 2-year follow-up French case study. *Clinical linguistics & phonetics*, *35*(10), 945–963. https://doi.org/10.1080/02699206.2020.1839971

Carroll, J. M., Snowling, M. J., Stevenson, J., & Hulme, C. (2003). The development of phonological awareness in preschool children. *Developmental psychology*, *39*(5), 913. https://doi.org/10.1037/0012-1649.39.5.913

Chenausky, K. V., Gagné, D., Stipancic, K. L., Shield, A., & Green, J. R. (2022). The Relationship Between Single-Word Speech Severity and Intelligibility in Childhood Apraxia of Speech. *Journal of speech, language, and hearing research: JSLHR*, *65*(3), 843–857. https://doi.org/10.1044/2021_JSLHR-21-00213

Crary, M. A., Landess, S., & Towne, R. (1984). Phonological error patterns in developmental verbal dyspraxia. *Journal of clinical neuropsychology*, *6*(2), 157–170. https://doi.org/10.1080/01688638408401206

Cunningham, A. J., Witton, C., Talcott, J. B., Burgess, A. P., & Shapiro, L. R. (2015). Deconstructing phonological tasks: The contribution of stimulus and response type to the prediction of early decoding skills. *Cognition*, *143*, 178-186. https://doi.org/10.1016/j.cognition.2015.06.013

Davis, B. L., Jakielski, K. J., & Marquardt, T. P. (1998). Developmental apraxia of speech: Determiners of differential diagnosis. *Clinical linguistics & phonetics*, *12*(1), 25-45. https://doi.org/10.3109/02699209808985211

Dell, G. S. (1986). A Spreading-Activation Theory of Retrieval in Sentence Production. *Psychological review*, *93*(3), 283-321. https://doi.org/10.1037/0033-295X.93.3.283

Diepeveen, S., Haaften, L. van, Terband, H., Swart, B. de & Maassen, B. (2019). A Standardized Protocol for Maximum Repetition Rate Assessment in Children. *Folia Phoniatrica et Logopaedica*, *71*(5–6), 238–250. https://doi.org/10.1159/000500305

Diepeveen, S., Knežević, D., & Maassen, B. (2022, August 26). *Differences between diadochokinesis rates in children across three European languages* [Poster]. 8th International Conference on Speech Motor Control, Groningen, Netherlands

Dodd, B. (2005). Differential diagnosis and treatment of children with speech disorder (2nd ed.). Whurr Publishers.

Dodd, B. (2014). Differential diagnosis of pediatric speech sound disorder. *Curr. Dev. Disord. Rep. 1*, 189–196. https://doi.org/10.1007/s40474-014-0017-3

Dodd, B., Holm, A., Crosbie, S., & McIntosh, B. (2010). Core vocabulary intervention for inconsistent speech disorder. In Williams, A. L., McLeod, S., & McCauley, R. J. (Eds.), *Interventions for speech sound disorders in children* (pp. 117-136). Brookes Publishing.

Flipsen, P., Bernthal, J. E., & Bankson, N. W. (2017a). Classification and Comorbidity in Speech Sound Disorders. In J. E. Bernthal, N. W. Bankson, & P. Flipsen JR. (Eds.), *Articulation and phonological disorders: Speech sound disorders in children* (8th ed., pp. 132-149). Pearson.

Flipsen, P., Bernthal, J. E., & Bankson, N. W. (2017b). Motor-Based Treatment Approaches. In J. E. Bernthal, N. W. Bankson, & P. Flipsen JR. (Eds.), *Articulation and phonological disorders: Speech sound disorders in children* (8th ed., pp. 132-149). Pearson.

Forrest, K. (2003). Diagnostic criteria of developmental apraxia of speech used by clinical speech-language pathologists. *American journal of speech-language pathology*, *12*(3), 376–380. https://doi.org/10.1044/1058-0360(2003/083)

Frisch, S. A., Large, N. R., & Pisoni, D. B. (2000). Perception of Wordlikeness: Effects of Segment Probability and Length on the Processing of Nonwords. *Journal of Memory and Language*, *42*(4), 481–496. https://doi.org/10.1006/jmla.1999.2692

Galluzzi, C., Bureca, I., Guariglia, C., & Romani, C. (2015). Phonological simplifications, apraxia of speech and the interaction between phonological and phonetic processing. *Neuropsychologia*, *71*, 64–83. https://doi.org/10.1016/j.neuropsychologia.2015.03.007

Ganger, J., & Brent, M. R. (2004). Reexamining the vocabulary spurt. *Developmental psychology*, *40*(4), 621–632. https://doi.org/10.1037/0012-1649.40.4.621

Garrett, M. F. (1980). Levels of processing in sentence production. In Butterworth B. (Ed.) *Language production*. Vol. 1: *Speech and talk* (pp. 176–220). Academic Press.

Gayán, J., & Olson, R. K. (2003). Genetic and environmental influences on individual differences in printed word recognition. *Journal of experimental child psychology*, *84*(2), 97-123. https://doi.org/10.1016/S0022-0965(02)00181-9

George, D., & Mallery, M. (2010). *SPSS for Windows Step by Step: A Simple Guide and Reference, 17.0 update (10a ed.).* Pearson.

Gillon, G. T., & Moriarty, B. C. (2007). Childhood Apraxia of Speech: Children at Risk for Persistent Reading and Spelling Disorder. *Seminars in speech and language*, *28*(1), 48–57. https://doi.org/10.1055/s-2007-967929

Goswami, U., & Bryant, P. (1990). *Phonological Skills and Learning to Read*. Hove: Lawrence Earlbaum Associates.

Gravetter, F., & Wallnau, L. (2014). *Essentials of statistics for the behavioral sciences (8th ed.)*. Wadsworth.

Grigos, M. I., & Case, J. (2018). Changes in movement transitions across a practice period in childhood apraxia of speech. *Clinical linguistics & phonetics*, *32*(7), 661–687. https://doi.org/10.1080/02699206.2017.1419378

Grigos, M. I., & Kolenda, N. (2010). The relationship between articulatory control and improved phonemic accuracy in childhood apraxia of speech: a longitudinal case study. *Clinical linguistics & phonetics*, *24*(1), 17–40. https://doi.org/10.3109/02699200903329793

Grigos, M. I., Moss, A., & Lu, Y. (2015). Oral Articulatory Control in Childhood Apraxia of Speech. *Journal of speech, language, and hearing research : JSLHR*, *58*(4), 1103–1118. https://doi.org/10.1044/2015_JSLHR-S-13-0221

Groenen, P., Maassen, B., Crul, T., & Thoonen, G. (1996). The specific relation between perception and production errors for place of articulation in developmental apraxia of speech. *Journal of speech and hearing research*, *39*(3), 468–482. https://doi.org/10.1044/jshr.3903.468

Gussenhoven, C. (1992). Dutch. *Journal of the International Phonetic Association, 22*(1-2), 45-47. https://doi.org/10.1017/S002510030000459X

Hall, P. K., Jordan, L. S., & Robin, D. A. (1993). *Developmental apraxia of speech: theory and clinical practice*. Pro-Ed.

Hogan, T. P., Catts, H. W., & Little, T. D. (2005). The relationship between phonological awareness and reading: implications for the assessment of phonological awareness. *Language, speech, and hearing services in schools*, *36*(4), 285–293. https://doi.org/10.1044/0161-1461(2005/029)

Holm, A., Crosbie, S., & Dodd, B. (2005). Treating inconsistent speech disorders. In B. Dodd (Ed.), *Differential diagnosis and treatment of children with speech disorder* (2nd ed., pp.182–201). Whurr Publishers.

Horga, D., & Liker, M. (2016). Artikulacijska fonetika: anatomija i fiziologija izgovora. Ibis grafika d.o.o.

Hulme, C., Hatcher, P. J., Nation, K., Brown, A., Adams, J., & Stuart, G. (2002). Phoneme awareness is a better predictor of early reading skill than onset-rime awareness. *Journal of experimental child psychology*, *82*(1), 2–28. https://doi.org/10.1006/jecp.2002.2670

Icht, M., & Ben-David, B. M. (2021). Evaluating rate and accuracy of real word vs. non-word diadochokinetic productions from childhood to early adulthood in Hebrew speakers. *Journal of communication disorders*, *92*, 106112. https://doi.org/10.1016/j.jcomdis.2021.106112

Ingram, D., & Ingram, K. D. (2001). A Whole-Word Approach to Phonological Analysis and Intervention. *Language, speech, and hearing services in schools*, *32*(4), 271–283. https://doi.org/10.1044/0161-1461(2001/024)

Ingram, S. B., Reed, V. A., & Powell, T. W. (2019). Vowel Duration Discrimination of Children with Childhood Apraxia of Speech: A Preliminary Study. *American journal of speech-language pathology*, *28*(2S), 857–874. https://doi.org/10.1044/2019_AJSLP-MSC18-18-0113

Iuzzini, J. (2012). *Inconsistency of speech in children with childhood apraxia of speech, phonological disorders, and typical speech* (Doctoral dissertation).

Iuzzini-Seigel J. (2022). Prologue to the Forum: Care of the Whole Child: Key Considerations When Working with Children with Childhood Apraxia of Speech. *Language, speech, and hearing services in schools*, 1–5. Advance online publication. https://doi.org/10.1044/2022_LSHSS-22-00119

Iuzzini-Seigel, J. & Murray, E. (2017). Speech Assessment in Children with Childhood Apraxia of Speech. *Perspectives of the ASHA Special Interest Groups*, *2*(2), 47–60. https://doi.org/10.1044/persp2.sig2.47

Iuzzini-Seigel, J., Allison, K. M., & Stoeckel, R. (2022). A Tool for Differential Diagnosis of Childhood Apraxia of Speech and Dysarthria in Children: A Tutorial. *Language, speech, and hearing services in schools*, 1–21. Advance online publication. https://doi.org/10.1044/2022_LSHSS-21-00164

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

Jacks, A., Marquardt, T. P., & Davis, B. L. (2006). Consonant and syllable structure patterns in childhood apraxia of speech: developmental change in three children. *Journal of communication disorders*, *39*(6), 424–441. https://doi.org/10.1016/j.jcomdis.2005.12.005

Jacks, A., Marquardt, T. P., & Davis, B. L. (2013). Vowel production in childhood and acquired apraxia of speech. In *Handbook of vowels and vowel disorders* (pp. 344-364). Psychology Press.

Janssen, C., Segers, E., Mcqueen, J., M., & Verhoeven, L. (2016). Transfer from implicit to explicit phonological abilities in first and second language learners. *Bilingualism: Language and Cognition*, *20*(04), 795–812. https://doi.org/10.1017/s1366728916000523

Kansal, T., Bahuguna, S., Singh, V., & Choudhury, T. (2018). Customer Segmentation using K-means Clustering. 2018 *International Conference on Computational Techniques, Electronics and Mechanical Systems (CTEMS)*. https://doi.org/10.1109/ctems.2018.8769171

Kelić, M. (2017). *Phonological representations and phonological skills in children with dyslexia in Croatian* (Doctoral dissertation).

Kent, R. (2004). Models of speech motor control: Implications from recent developments in neurophysiological and neurobehavioral science. In B. Maassen, R. Kent, H.F.M. Peters, P.H.H.M. vanLieshout, and W. Hulstijn (Eds.), *Speech Motor Control in Normal and Disordered Speech* (pp. 1–28). Oxford University Press.

Kent, R. (2017). Normal Aspects of Articulation. In J. E. Bernthal, N. W. Bankson, & P. Flipsen JR. (Eds.), *Articulation and phonological disorders: Speech sound disorders in children* (8th ed., pp. 7-48). Pearson.

King, J. L., Jakielski, K. J., & Malone, K. A. (2001). Quantifying variability in developmental apraxia of speech: case study results. American Speech-Language-Hearing Association Convention, New Orleans, LA.

Kuvač Kraljević, J. & Lenček, M. (2012). *Test za procjenjivanje predvještina čitanja i pisanja (PredČiP)*. Jastrebarsko/Zagreb: Naklada Slap.

Kuvač Kraljević, J., Lenček, M. & Matešić, K. (2020). Phonological Awareness and Letter Knowledge: Indicators of Early Literacy in Croatian. *Croatian Journal of Education, 21* (4), 1263-1293. https://doi.org/10.15516/cje.v21i4.3130

Laganaro, M. (2012). Patterns of impairments in AOS and mechanisms of interaction between phonological and phonetic encoding. *Journal of speech, language, and hearing research: JSLHR*, *55*(5), S1535–S1543. https://doi.org/10.1044/1092-4388(2012/11-0316)

Landau et al. 1999. „What is the International Phonetic Alphabet? “. In *Handbook of the International Phonetic Association: A guide to the Use of the International Phonetic Alphabet*. Cambridge University Press.

Lenoci, G., Celata, C., Ricci, I., Chilosi, A. & Barone, V. (2020). Vowel variability and contrast in Childhood Apraxia of Speech: acoustics and articulation. *Clinical Linguistics & Phonetics*, *35*(11), 1–25. https://doi.org/10.1080/02699206.2020.1853811

Levelt, W. J., Roelofs, A., & Meyer, A. S. (1999). A theory of lexical access in speech production. *The Behavioral and brain sciences*, *22*(1), 1–75. https://doi.org/10.1017/s0140525x99001776

Levelt, W., J. (1993). *Speaking: From intention to articulation*. MIT press.

Lewis, B. A., Avrich, A. A., Freebairn, L. A., Taylor, H. G., Iyengar, S. K., & Stein, C. M. (2011). Subtyping Children with Speech Sound Disorders by Endophenotypes. Topics in language disorders, 31(2), 112–127. https://doi.org/10.1097/TLD.0b013e318217b5dd

Lewis, B. A., Freebairn, L. A., Hansen, A., Taylor, H. G., Iyengar, S., & Shriberg, L. D. (2004). Family pedigrees of children with suspected childhood apraxia of speech. *Journal of Communication Disorders*, *37*(2), 157–175. https://doi.org/10.1016/j.jcomdis.2003.08.003

Lewis, B. A., Freebairn, L., Tag, J., Benchek, P., Morris, N. J., Iyengar, S. K., Taylor, H. G., & Stein, C. M. (2018). Heritability and longitudinal outcomes of spelling skills in individuals with histories of early speech and language disorders. *Learning and individual differences*, *65*, 1–11. https://doi.org/10.1016/j.lindif.2018.05.001

Liégeois, F. J., & Morgan, A. T. (2012). Neural bases of childhood speech disorders: lateralization and plasticity for speech functions during development. *Neuroscience and biobehavioral reviews*, *36*(1), 439–458. https://doi.org/10.1016/j.neubiorev.2011.07.011

Limbrick, N., McCormack, J., & McLeod, S. (2013). Designs and decisions: the creation of informal measures for assessing speech production in children. *International journal of speech-language pathology*, *15*(3), 296–311. https://doi.org/10.3109/17549507.2013.770552

Lin, Z. (2004). IPA English vowel chart. Retrieved October 10, 2022, from https://commons.wikimedia.org/wiki/File:English_vowel_chart.png

Maas, E., Butalla, C. E., & Farinella, K. A. (2012). Feedback frequency in treatment for childhood apraxia of speech. *American journal of speech-language pathology*, *21*(3), 239–257. https://doi.org/10.1044/1058-0360(2012/11-0119)

Maassen, B. (2002). Issues Contrasting Adult Acquired Versus Developmental Apraxia of Speech. *SEMINARS IN SPEECH AND LANGUAGE*, *23*(4), 257–266. https://doi.org/10.1055/s-2002-35804

Maassen, B. (2015). Developmental Models of Childhood Apraxia of Speech. In R. H. Bahr & E. R. Silliman, (Eds.), *Routledge Handbook of Communication Disorders* (1st ed., pp. 236-252). Routledge.

Maassen, B., & Terband, H. (2015). Process-oriented diagnosis of childhood and adult apraxia of speech (CAS and AOS). In M. A., Redford (Ed.), *The handbook of speech production* (1st ed., pp. 331–350). Wiley.

Maassen, B., Groenen, P., & Crul, T. (2003). Auditory and phonetic perception of vowels in children with apraxic speech disorders. *Clinical linguistics & phonetics*, *17*(6), 447–467. https://doi.org/10.1080/0269920031000070821

Maassen, B., Nijland, L., & Terband, H. (2010). Developmental models of childhood apraxia of speech. In B. Maassen, & P. van Lieshout (Eds.) *Speech Motor Control. New Developments in Basic and Applied Research* (pp. 243–258). Oxford University Press),

Maassen, B., van Haaften, L., Diepeveen, S., Terband, H., van den Engel-Hoek, L., Veenker, T., & De Swart, B. (2019). Computer Articulation Instrument. Amsterdam, the Netherlands: Boom Uitgevers

MacKay, D. G., & James, L. E. (2004). Sequencing, speech production, and selective effects of aging on phonological and morphological speech errors. *Psychology and aging*, *19*(1), 93–107. https://doi.org/10.1037/0882-7974.19.1.93

Macrae, T., Tyler, A. A., & Lewis, K. E. (2014). *Lexical and Phonological Variability in Preschool Children With Speech Sound Disorder. American Journal of Speech-Language Pathology*, *23*(1), 27. https://doi.org/10.1044/1058-0360(2013/12-0037)

Malmenholt, A., Lohmander, A., & McAllister, A. (2017). Childhood apraxia of speech: A survey of praxis and typical speech characteristics. *Logopedics, phoniatrics, vocology*, *42*(2), 84–92. https://doi.org/10.1080/14015439.2016.1185147

Malmenholt, A., McAllister, A., Lohmander, A., & Östberg, P. (2022). Speech feature profiles in Swedish 5-year-olds with speech sound disorder related to suspected childhood apraxia of speech or cleft palate. *International journal of speech-language pathology*, *24*(2), 156–167. https://doi.org/10.1080/17549507.2021.1968951

Marion, M. J., Sussman, H. M., & Marquardt, T. P. (1993). The perception and production of rhyme in normal and developmentally apraxic children. *Journal of communication disorders*, *26*(3), 129–160. https://doi.org/10.1016/0021-9924(93)90005-u

Marquardt, T. P., Jacks, A., & Davis, B. L. (2004). Token-to-token variability in developmental apraxia of speech: three longitudinal case studies. *Clinical linguistics & phonetics*, *18*(2), 127–144. https://doi.org/10.1080/02699200310001615050

Marquardt, T. P., Sussman, H. M., Snow, T., & Jacks, A. (2002). The integrity of the syllable in developmental apraxia of speech. *Journal of communication disorders*, *35*(1), 31–49. https://doi.org/10.1016/s0021-9924(01)00068-5

McLeod, S. (2017). Speech sound acquisition. In J. E. Bernthal, N. W. Bankson, & P. Flipsen JR. (Eds.), *Articulation and phonological disorders: Speech sound disorders in children* (8th ed., pp. 49-92). Pearson.

McLeod, S., & Crowe, K. (2018). Children's consonant acquisition in 27 languages: A cross-linguistic review. *American Journal of Speech-Language Pathology*, *27*(4), 1546–1571. https://doi.org/10.1044/2018_AJSLP-17-0100

McNeil, M. R., & Kent, R. D. (1990). Motoric Characteristics of Adult Aphasic and Apraxic Speakers. Cerebral Control of Speech and Limb Movements. In: G. E. Hammond (Ed.), *Cerebral Control of Speech and Limb Movements* (pp.349–386). Elsevier Science Publishers. https://doi.org/10.1016/s0166-4115(08)60655-x

McNeil, M. R., Pratt, S.R., & Fosset, T.R.D. (2004). The differential diagnosis of apraxia of speech. In B. Maassen, R. Kent, H. F. M. Peters, P. H. H. M. van Lieshout, & W. Hulstijn (Eds.), *Speech Motor Control in Normal and Disordered Speech* (pp. 389-414). Oxford University Press.

McNeill, B. (2013). Developmental verbal dyspraxia. In L. Cummings (Ed.), *The Cambridge Handbook of Communication Disorders* (Cambridge Handbooks in Language and Linguistics). Cambridge: Cambridge University Press. https://doi.org/10.1017/CBO9781139108683.005

McNeill, B. C., Gillon, G. T., & Dodd, B. (2009). Phonological awareness and early reading development in childhood apraxia of speech (CAS). *International journal of language & communication disorders*, *44*(2), 175–192. https://doi.org/10.1080/13682820801997353

Meloni, G., Schott-Brua, V., Vilain, A., Loevenbruck, H., Consortium, E., & MacLeod, A. (2020). Application of childhood apraxia of speech clinical markers to French-speaking children: A preliminary study. *International journal of speech-language pathology*, *22*(6), 683–695. https://doi.org/10.1080/17549507.2020.1844799

Miller, G. J., Lewis, B., Benchek, P., Freebairn, L., Tag, J., Budge, K., Iyengar, S. K., Voss-Hoynes, H., Taylor, H. G., & Stein, C. (2019). Reading Outcomes for Individuals with Histories of Suspected Childhood Apraxia of Speech. *American journal of speech-language pathology*, *28*(4), 1432–1447. https://doi.org/10.1044/2019_AJSLP-18-0132

Morgan, A. T., & Webster, R. (2018). Aetiology of childhood apraxia of speech: A clinical practice update for paediatricians. *Journal of paediatrics and child health*, *54*(10), 1090–1095. https://doi.org/10.1111/jpc.14150

Morgan, A. T., Murray, E., & Liégeois, F. J. (2018). Interventions for childhood apraxia of speech. *The Cochrane database of systematic reviews*, *5*(5), CD006278. https://doi.org/10.1002/14651858.CD006278.pub3

Morley D. E. (1952). A ten-year survey of speech disorders among university students. *The Journal of speech disorders*, *17*(1), 25–31. https://doi.org/10.1044/jshd.1701.25

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

Murray, E., McCabe, P., Heard, R., & Ballard, K. J. (2015). Differential diagnosis of children with suspected childhood apraxia of speech. *Journal of speech, language, and hearing research: JSLHR*, *58*(1), 43–60. https://doi.org/10.1044/2014_JSLHR-S-12-0358

Namasivayam, A. K., Coleman, D., O'Dwyer, A., & van Lieshout, P. (2020). Speech Sound Disorders in Children: An Articulatory Phonology Perspective. *Frontiers in psychology*, *10*, 2998. https://doi.org/10.3389/fpsyg.2019.02998

Nijland L. (2003). *Developmental apraxia of speech: deficits in phonetic planning and motor programming* (doctoral dissertation).

Nijland L. (2009). Speech perception in children with speech output disorders. *Clinical linguistics & phonetics*, *23*(3), 222–239. https://doi.org/10.1080/02699200802399947

Nijland, L., Maassen, B., & van der Meulen, S. (2003). Evidence of motor programming deficits in children diagnosed with DAS. *Journal of speech, language, and hearing research: JSLHR*, *46*(2), 437–450. https://doi.org/10.1044/1092-4388(2003/036)

Nijland, L., Maassen, B., Van Der Meulen, S., Gabreëls, F., Kraaimaat, F. W., & Schreuder, R. (2003). Planning of syllables in children with developmental apraxia of speech. *Clinical linguistics & phonetics*, *17*(1), 1–24. https://doi.org/10.1080/0269920021000050662

Nijland, L., Terband, H., & Maassen, B. (2015). Cognitive Functions in Childhood Apraxia of Speech. *Journal of speech, language, and hearing research: JSLHR*, *58*(3), 550–565. https://doi.org/10.1044/2015_JSLHR-S-14-0084

Nip, I. S., Green, J. R., & Marx, D. B. (2011). The co-emergence of cognition, language, and speech motor control in early development: A longitudinal correlation study. *Journal of Communication Disorders*, *44*(2), 149–160. https://doi.org/10.1016/j.jcomdis.2010.08.002

Norton, E. S., & Wolf, M. (2012). Rapid automatized naming (RAN) and reading fluency: implications for understanding and treatment of reading disabilities. *Annual review of psychology*, *63*, 427–452. https://doi.org/10.1146/annurev-psych-120710-100431

Ouellette, G. P., & Haley, A. (2013). One complicated extended family: The influence of alphabetic knowledge and vocabulary on phonemic awareness. *Journal of Research in Reading*, *26*, 29-41. https://doi.org/10.1111/j.1467-9817.2010.01486.x

Overby, M. S., Caspari, S. S., & Schreiber, J. (2019). Volubility, Consonant Emergence, and Syllabic Structure in Infants and Toddlers Later Diagnosed With Childhood Apraxia of Speech, Speech Sound Disorder, and Typical Development: A Retrospective Video Analysis. *Journal of speech, language, and hearing research : JSLHR*, *62*(6), 1657–1675. https://doi.org/10.1044/2019_JSLHR-S-18-0046

Ozanne A. (1995). The search for developmental verbal dyspraxia. In B. Dodd (Ed.), *Differential diagnosis and treatment of children with speech disorder* (1st ed.). Whurr Publishers.

Ozanne, A. (2005). Childhood apraxia of speech. In B. Dodd (Ed.), *Differential diagnosis and treatment of children with speech disorder* (2nd ed., pp. 71–82). Whurr Publishers.

Pollack, K. & Hall, P. (1991) An analysis of vowel misarticulations of five children with developmental apraxia of speech. *Clinical Linguistics and Phonetics 5*: 207–24. https://doi.org/10.3109/02699209108986112

Preston, J. L., Benway, N. R., Leece, M. C., & Caballero, N. F. (2021). Concurrent Validity Between Two Sound Sequencing Tasks Used to Identify Childhood Apraxia of Speech in School-Age Children. *American journal of speech-language pathology*, *30*(3S), 1580–1588. https://doi.org/10.1044/2020_AJSLP-20-00108

Pufpaff, L. A. (2009). A developmental continuum of phonological sensitivity skills. *Psychology in the Schools*, *46*(7), 679-691. https://doi.org/10.1002/pits.20407

Ramus, F., & Szenkovits, G. (2008). What phonological deficit? *Quarterly journal of experimental psychology*, *61*(1), 129-141. https://doi.org/10.1080/17470210701508822

Ramus, F., Marshall, C. R., Rosen, S., & van der Lely, H. K. (2013). Phonological deficits in specific language impairment and developmental dyslexia: towards a multidimensional model. *Brain : a journal of neurology*, *136*(Pt 2), 630–645. https://doi.org/10.1093/brain/aws356

Randazzo, M. (2019). A Survey of Clinicians with Specialization in Childhood Apraxia of Speech. *American journal of speech-language pathology*, *28*(4), 1659–1672. https://doi.org/10.1044/2019_AJSLP-19-0034

Raven, J. C. (1999). Kolorirane progresivne matrice - CPM. Jastrebarsko: Naklada Slap.

Sangster Jokić, C., Knežević D., & Wilson, B. N. (2020). Validacija mjernog instrumenta za rano prepoznavanje razvojnog poremećaja koordinacije: Ispitivanje psihometrijskih svojstava hrvatske inačice Upitnika za procjenu koordinacije (DCDQ-HR). [Unpublished manuscript] Katedra za radnu terapiju; Zdravstveno veleučilište Zagreb.

Schmidt, R., & Lee, T. (1999). *Motor Control and Learning: A Behavioral Emphasis*. Champaign, Human Kinetics.

Schwartz, J. L., Boë, L. J., Badin, P., & Sawallis, T. R. (2012). Grounding stop place systems in the perceptuo-motor substance of speech: On the universality of the labial–coronal–velar stop series. *Journal of Phonetics*, *40*(1), 20–36. https://doi.org/10.1016/j.wocn.2011.10.004

Shriberg, L. D., & Kwiatkowski, J. (1982). Phonological disorders III: a procedure for assessing severity of involvement. *The Journal of speech and hearing disorders*, *47*(3), 256–270. https://doi.org/10.1044/jshd.4703.256

Shriberg, L. D., Aram, D. M., & Kwiatkowski, J. (1997a). Developmental apraxia of speech: I. Descriptive and theoretical perspectives. *Journal of speech, language, and hearing research: JSLHR*, *40*(2), 273–285. https://doi.org/10.1044/jslhr.4002.273

Shriberg, L. D., Aram, D. M., & Kwiatkowski, J. (1997b). Developmental apraxia of speech: II. Toward a diagnostic marker. *Journal of speech, language, and hearing research: JSLHR*, *40*(2), 286–312. https://doi.org/10.1044/jslhr.4002.286

Shriberg, L. D., Austin, D., Lewis, B. A., McSweeny, J. L., & Wilson, D. L. (1997c). The percentage of consonants correct (PCC) metric: extensions and reliability data. *Journal of speech, language, and hearing research: JSLHR*, *40*(4), 708–722. https://doi.org/10.1044/jslhr.4004.708

Shriberg, L. D., Fourakis, M., Karlsson, H. K., Lohmeier, H. L., McSweeney, J., Potter, N. L., et al. (2010). Extensions to the speech disorders classification system (SDCS). *Clin. Linguist. Phonet. 24*, 795–824. https://doi.org/10.3109/02699206.2010.503006

Shriberg, L. D., Green, J. R., Campbell, T. F., McSweeny, J. L., & Scheer, A. R. (2003). A diagnostic marker for childhood apraxia of speech: the coefficient of variation ratio. *Clinical linguistics & phonetics*, *17*(7), 575–595. https://doi.org/10.1080/0269920031000138141

Shriberg, L. D., Lohmeier, H. L., Strand, E. A., & Jakielski, K. J. (2012). Encoding, memory, and transcoding deficits in Childhood Apraxia of Speech. *Clinical linguistics & phonetics*, *26*(5), 445–482. https://doi.org/10.3109/02699206.2012.655841

Škarić, I. (1991). Fonetika hrvatskoga književnog jezika. In S. Babić, D. Brozović, M. Moguš, S. Pavešić, I. Škarić, S. Težak (Eds.), *Povijesni pregled, glasovi i oblici hrvatskoga književnog jezika: Nacrt za gramatiku* (pp. 61-379). HAZU - Globus

Spencer, C., Davison, K. E., Boucher, A. R., & Zuk, J. (2022). Speech Perception Variability in Childhood Apraxia of Speech: Implications for Assessment and Intervention. *Language, speech, and hearing services in schools*, 1–16. Advance online publication. https://doi.org/10.1044/2022_LSHSS-21-00170

Stackhouse, J., & Snowling, M. (1992). Barriers to literacy development in two cases of developmental verbal dyspraxia. *Cognitive Neuropsychology*, *9*(4), 273-299. https://doi.org/10.1080/02643299208252062

Stackhouse, J., & Wells B. (1997). *Children's speech and literacy difficulties: A psycholinguistic framework*. Wiley

Stackhouse, J., & Wells, B. (1997). *Children's speech and literacy difficulties: A psycholinguistic framework*, vol. 1. Wiley Incorporated.

Staiger, A., & Ziegler, W. (2008). Syllable frequency and syllable structure in the spontaneous speech production of patients with apraxia of speech. *Aphasiology*, *22*(11), 1201-1215. https://doi.org/10.1080/02687030701820584

Stanford University (2010). Retrieved October 10, 2022, from https://web.stanford.edu/~kfpotts/teaching/F10_L1/F10_L1_English-IPA.doc

Stein, C. M., Benchek, P., Miller, G., Hall, N. B., Menon, D., Freebairn, L., Tag, J., Vick, J., Taylor, H. G., Lewis, B. A. & Iyengar, S. K. (2020). Feature-driven classification reveals potential comorbid subtypes within childhood apraxia of speech. *BMC Pediatrics*, *20*(1), 519. https://doi.org/10.1186/s12887-020-02421-1

Tabachnick, B. G., & Fidell, L. S. (2013). *Using multivariate statistics (6th edition).* Pearson Education Inc.

Terband, H. (2011). *Speech Motor Control in Relation to Phonology: Neurocomputational Modeling of Disordered Development* (Doctoral dissertation).

Terband, H., Maassen, B., and Maas, E. (2019). A psycholinguistic framework for diagnosis and treatment planning of developmental speech disorders. *Folia Phoniatr. Logop. 3*, 1–12. https://doi.org/10.1159/000499426

Thoonen, G., Maassen, B., Gabreëls, F., & Schreuder, R. (1999). Validity of maximum performance tasks to diagnose motor speech disorders in children. *Clinical Linguistics and Phonetics, 13*(1), 1-23.

Thoonen, G., Maassen, B., Gabreëls, F., Schreuder, R., & de Swart, B. (1997). Towards a standardised assessment procedure for developmental apraxia of speech. *International Journal of Language & Communication Disorders*, *32*(1), 37-60. https://doi.org/10.3109/13682829709021455

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

Tierney, C., Mayes, S., Lohs, S. R., Black, A., Gisin, E., & Veglia, M. (2015). How Valid Is the Checklist for Autism Spectrum Disorder When a Child Has Apraxia of Speech? *Journal of developmental and behavioral pediatrics: JDBP*, *36*(8), 569–574. https://doi.org/10.1097/DBP.0000000000000189

Treiman, R. (2017). Learning to spell words: Findings, theories, and issues. *Scientific Studies of Reading*, *21*(4), 265–276. https://doi.org/10.1080/10888438.2017.1296449

Turner, S. J., Vogel, A. P., Parry-Fielder, B., Campbell, R., Scheffer, I. E., & Morgan, A. T. (2019). Looking to the Future: Speech, Language, and Academic Outcomes in an Adolescent with Childhood Apraxia of Speech. *Folia phoniatrica et logopaedica : official organ of the International Association of Logopedics and Phoniatrics (IALP)*, *71*(5-6), 203–215. https://doi.org/10.1159/000500554

van der Merwe, A. (1997). A theoretical framework for the characterization of pathological speech sensorimotor control. In M.R. McNeil (Ed.), *Clinical Management of Sensorimotor Speech Disorders* (pp. 1–25). Thieme Medical Publishers.

van der Merwe, A. (2011). A speech motor learning approach to treating apraxia of speech: Rationale and effects of intervention with an adult with acquired apraxia of speech. *Aphasiology*, *25*(10), 1174–1206. https://doi.org/10.1080/02687038.2011.582246 van Haaften, L. van, Diepeveen, S., Engel-Hoek, L. van den, Jonker, M., Swart, B. de & Maassen, B. (2019a). 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. https://doi.org/10.1044/2018_jslhr-s-18-0274

van Haaften, L. van, Diepeveen, S., Terband, H., Vermeij, B., Engel-Hoek, L. van den, Swart, B. de & Maassen, B. (2019b). Profiling Speech Sound Disorders for Clinical Validation of the Computer Articulation Instrument. *American Journal of Speech-Language Pathology*, *28*(2S), 844–856. https://doi.org/10.1044/2018_ajslp-msc18-18-0112

van Haaften, L., Diepeveen, S., Engel-Hoek, L. den, 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. https://doi.org/10.1111/1460-6984.12575

Vance, M., Stackhouse, J., & Wells, B. (2005). Speech-production skills in children aged 3-7 years. *International journal of language & communication disorders*, *40*(1), 29–48. https://doi.org/10.1080/13682820410001716172

Velleman, S., & Strand, K. (1994). Developmental verbal dyspraxia. In J. E. Bernthal & N. W. Bankson (Eds.), *Child Phonology: Characteristics, Assessment, and Intervention with Special Populations* (pp. 110-139). Thieme Medical Publishers, Inc.

Vuletić, D. (1990). Test artikulacije. Zagreb: Fakultet za defektologiju Sveučilišta u Zagrebu.

Watkins, K. E., Vargha-Khadem, F., Ashburner, J., Passingham, R. E., Connelly, A., Friston, K. J., Frackowiak, R. S., Mishkin, M., & Gadian, D. G. (2002). MRI analysis of an inherited speech and language disorder: structural brain abnormalities. *Brain : a journal of neurology*, *125*(Pt 3), 465–478. https://doi.org/10.1093/brain/awf057

Weismer, G., & Green, J. R. (2015). Speech production in motor speech disorders: Lesions, models, and a research agenda. In M. A. Redford (Ed.), *The handbook of speech production*, (1st ed., pp. 298-330). John Wiley & Sons, Inc

Williams, P. (2015) *The Diadochokinetic Skills of Children with Speech Difficulties* (Doctoral dissertation).

Williams, P., & Stackhouse, J. (1998). Diadochokinetic Skills: Normal and Atypical Performance in Children Aged 3–5 Years. *International Journal of Language & Communication Disorders*, *33*(S1), 481-486. https://doi.org/doi:10.3109/13682829809179472

Wilson, B. N., Kaplan, B. J., Crawford, S. G., Campbell, A., & Dewey, D. (2000). Reliability and validity of a parent questionnaire on childhood motor skills. *The American journal of occupational therapy: official publication of the American Occupational Therapy Association*, *54*(5), 484–493. https://doi.org/10.5014/ajot.54.5.484

Wolf, M., Bowers, P. G., & Biddle, K. (2000). Naming-speed processes, timing, and reading: a conceptual review. *Journal of learning disabilities*, *33*(4), 387–407. https://doi.org/10.1177/002221940003300409

Yaruss, J. S., & Logan, K. J. (2002). Evaluating rate, accuracy, and fluency of young children's diadochokinetic productions: a preliminary investigation. *Journal of fluency disorders*, *27*(1), 65–86. https://doi.org/10.1016/s0094-730x(02)00112-2

Yoss, K. A., & Darley, F. L. (1974). Developmental apraxia of speech in children with defective articulation. *Journal of speech and hearing research*, *17*(3), 399–416. https://doi.org/10.1044/jshr.1703.399

Zaretsky, E., Velleman, S. L., & Curro, K. (2009). Through the magnifying glass: Underlying literacy deficits and remediation potential in Childhood Apraxia of Speech. *International Journal of Speech-Language Pathology*, *12*(1), 58–68. https://doi.org/10.3109/17549500903216720

Ziegler, W. (2002). Task-Related Factors in Oral Motor Control: Speech and Oral Diadochokinesis in Dysarthria and Apraxia of Speech. *Brain and Language*, *80*(3), 556–575. https://doi.org/10.1006/brln.2001.2614

Ziegler, W. (2005). A nonlinear model of word length effects in apraxia of speech. *Cognitive neuropsychology*, *22*(5), 603–623. https://doi.org/10.1080/02643290442000211

Ziegler, W. (2008). Apraxia of speech. In G. Goldenberg, B.L. Miller, (Eds.) *Handbook of clinical neurology*, *88*, (3rd series, pp. 269-285). Elsevier B.V.

Zuk, J., Iuzzini-Seigel, J., Cabbage, K., Green, J. R., & Hogan, T. P. (2018). Poor Speech Perception Is Not a Core Deficit of Childhood Apraxia of Speech: Preliminary Findings. *Journal of speech, language, and hearing research: JSLHR*, *61*(3), 583–592. https://doi.org/10.1044/2017_JaSLHR-S-16-0106

Appendix 1. Case history form

Povjerljivost informacija o identitetu je zajamčena, Neposredno nakon prikupljanja, podatci će biti kodirani te će na taj način biti zaštićena sva prava i anonimnost sudionika istraživanja.

Anamnestički upitnik

Ime i prezime djeteta:		
Spol:	**M**	**Ž**
Datum rođenja:		
Prvi jezik djeteta (dominantni jezik kojim se dijete služi):		
Je li dijete izloženo još nekom jeziku?		
Dijete je uključeno u predškolsku ustanovu:	Da	Ne
Dijete ide samo u malu školu:	Da	Ne
Dijete ide u školu:	Da	Ne
Je li dijete uključeno u neki oblik terapije?	Da	Ne
Ako da, koju i koliko dugo?		

TRUDNOĆA / POROD I PERINATALNI PODACI:

Tijek trudnoće: Ako je trudnoća bila rizična, koje su se teškoće javile?	uredan rizičan
Tjedan gestacije (označiti) :	☐ Prijevremeni porođaj (prije 28. tjedna) ☐ Prijevremeni porođaj (između 28. i 32. tjedna) ☐ Prijevremeni porođaj (između 32. to 37. tjedna) ☐ Porođaj u terminu (iza 38. tjedna)
Je li porod bio uredan? Ako nije, koja vrsta komplikacije se javila?	
APGAR:	
Ostalo moguće teškoće ili napomene:	

RANI PSIHOMOTORIČKI RAZVOJ:

U kojoj je dobi dijete prohodalo?	

RANI JEZIČNO - GOVORNI RAZVOJ:

Kada je dijete počelo brbljati:	
Prva riječ sa značenjem:	

Povjerljivost informacija o identitetu je zajamčena, Neposredno nakon prikupljanja, podatci će biti kodirani te će na taj način biti zaštićena sva prava i anonimnost sudionika istraživanja.

Spajanje riječi:	
Je li bilo dužih zastoja/nazadovanja u jezično-govornom razvoju? *Ako da, što je primiječeno?	
Je li dijete zainteresirano za slikovnice?	
Je li dijete zainteresirano za crtanje/pisanje?	

TEŠKOĆE/KOMPLIKACIJE:

Komplikacije s hranjenjem:	
Komplikacije sa sisanjem/gutanjem/žvakanjem:	
Hospitalizacije (kada, gdje, zašto?):	
Česte upale uha, grla, nosa?	
Ima li vaše dijete dodatnih teškoća ili nekih medicinskih dijagnoza (epilepsija, neurološke bolesti, kognitivne teškoće, oštećenja sluha, vida, motoričke teškoće…) *Ako da, koje?	Da Ne
Ostalo:	

Appendix 2. General characteristics of children with CAS

CASE ID	GROUP	GENDER	AGE (months)	TROG-2: HR score	SLP threapy	Additional difficulties
1	CAS	F	69	106	yes	no
2	CAS	M	69	92	yes	no
4	CAS	M	73	108	yes	no
5	CAS	M	84	97	yes	no
7	CAS	F	86	103	yes	no
8	CAS	M	77	84	yes	no
9	CAS	M	71	86	yes	no
19	CAS	M	79	91	yes	no
26	CAS	F	64	90	yes	no
29	CAS	M	75	89	yes	no
31	CAS	M	76	99	yes	no
32	CAS	F	80	88	yes	no
42	CAS	M	80	97	yes	no
44	CAS	M	87	85	yes	no
48	CAS	M	67	90	yes	no
51	CAS	M	65	99	yes	no
52	CAS	M	78	109	yes	no
53	CAS	M	87	93	yes	no
54	CAS	M	71	95	yes	no
56	CAS	M	71	89	yes	no
57	CAS	M	75	103	yes	no
59	CAS	M	67	95	yes	no
60	CAS	M	84	85	yes	no
61	CAS	M	68	84	yes	no
62	CAS	M	68	92	yes	no
63	CAS	M	90	85	yes	no
64	CAS	M	91	98	yes	no
68	CAS	M	85	98	yes	no
69	CAS	M	82	85	yes	no
71	CAS	M	86	88	yes	no

Appendix 3. Spearman's rank correlation for all the measured variables for CAS children

Variables:	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21
1. AX_RT	1.00																				
2. AX_Acc	.26	1.00																			
3. Rhym_Rec	.14	.26	1,00																		
4. Rhym_Prod	.21	.35	.55**	1,00																	
5. Syll_Seg	-.15	.09	-.05	.16	1,00																
6. Syll_Blend	.14	.41*	.28	.24	.05	1,00															
7. Phon_Seg	.25	.33	.37*	.51**	.00	0,05	1,00														
8. Phon_Blend	.17	.53**	.42*	.64**	.00	,38*	,71**	1,00													
9. RAN	.22	.51**	.09	.33	.14	,38*	,47**	,52**	1,00												
10. WR_Cons	.07	.48**	.51**	.51**	.23	,50**	.34	,48**	.37*	1,00											
11. NWR_Cons	.18	.40*	.54**	.61**	.10	,45*	,52**	,75**	.37*	,80**	1,00										
12. WL_Cons	-.14	.12	.28	.27	.34	,39*	-.04	.20	-.03	,51**	.51**	1,00									
13. WL_Corr	-.05	.31	.20	.42*	.21	,45*	.11	.40*	.33	,45*	,57**	.48**	1,00								
14. PCC	.09	.35	.42*	.57**	.03	.21	.57**	,57**	.14	,66**	,60**	.27	.31	1,00							
15. PVC	-.03	.17	.36	.47**	.18	-.09	.04	.06	-.09	,45*	.22	.25	-.01	,49**	1,00						
16. PCCI	.26	.36*	.43*	.61**	-.05	.18	.66**	,55**	.20	,59**	.59**	.18	.36	,93**	,44*	1,00					
17. PCCF	.38*	.26	.28	.65**	-.05	.10	.54**	,43*	.19	.25	.35	-.01	.25	,66**	,38*	,73**	1,00				
18. PCD	.15	.20	.37	.58**	-.06	.29	,51**	,47*	.12	,40*	,47**	.26	.20	,73**	.19	,74**	,65**	1,00			
19. MRR_Mono	.02	.17	.16	.30	.32	.20	.05	.07	.20	,48**	.31	.32	.50**	.24	.31	.29	.13	-.03	1,00		
20. MRR_Bi	-.03	.34	.21	.41*	.21	.23	.41*	,42*	.25	,56**	,58**	.38*	.44*	.59**	.28	,53**	.60**	,58**	.27	1,00	
21.MRR_Tri	.06	.08	.19	.34	.15	-.01	.35	.17	.01	.35	,38*	.13	.12	,40*	.15	,42*	.23	.38*	-.10	.38*	1,00

*p<.05, ** p< .01

Note. 1. AX_Acc = Ax discrimination task, correctness score; 2. Ax_RT = Ax discrimination task, reaction time; 3. Rhym_Rec = rhyme recognition; 4. Rhym_Prod = rhyme production; 5. Syll_Seg = syllable segmentation; 6. Syll_Blend = syllable blending; 7. Phon_Seg = phonemic segmentation, 8. Phon_Blend = phonemic blending; 9. RAN = rapid automatized naming; 10. WR_Cons = word repetition consisntency; 11. NWR_Cons = non-word repetition consistency; 12. WL_Cons = word learning consistency; 13. WL_Corr = word learning correctness; 14. PCC = percentage consonant correct; 15. PVC = percentage vowels correct; 16. PCCI = percentage correct consonants in syllable initial position; 17. PCCF = percentage correct consonants in syllable final position; 18. PCD = percentage consonant deletion; 19. MRR_Mono = monosyllabic maximum repetition rate; 20. MRR_Bi = bisyllabic maximum repetition rate; 21. MRR_Tri = trisyllabic maximum repetition rate

Appendix 4. Spearman's rank correlation for all the measured variables for TD children

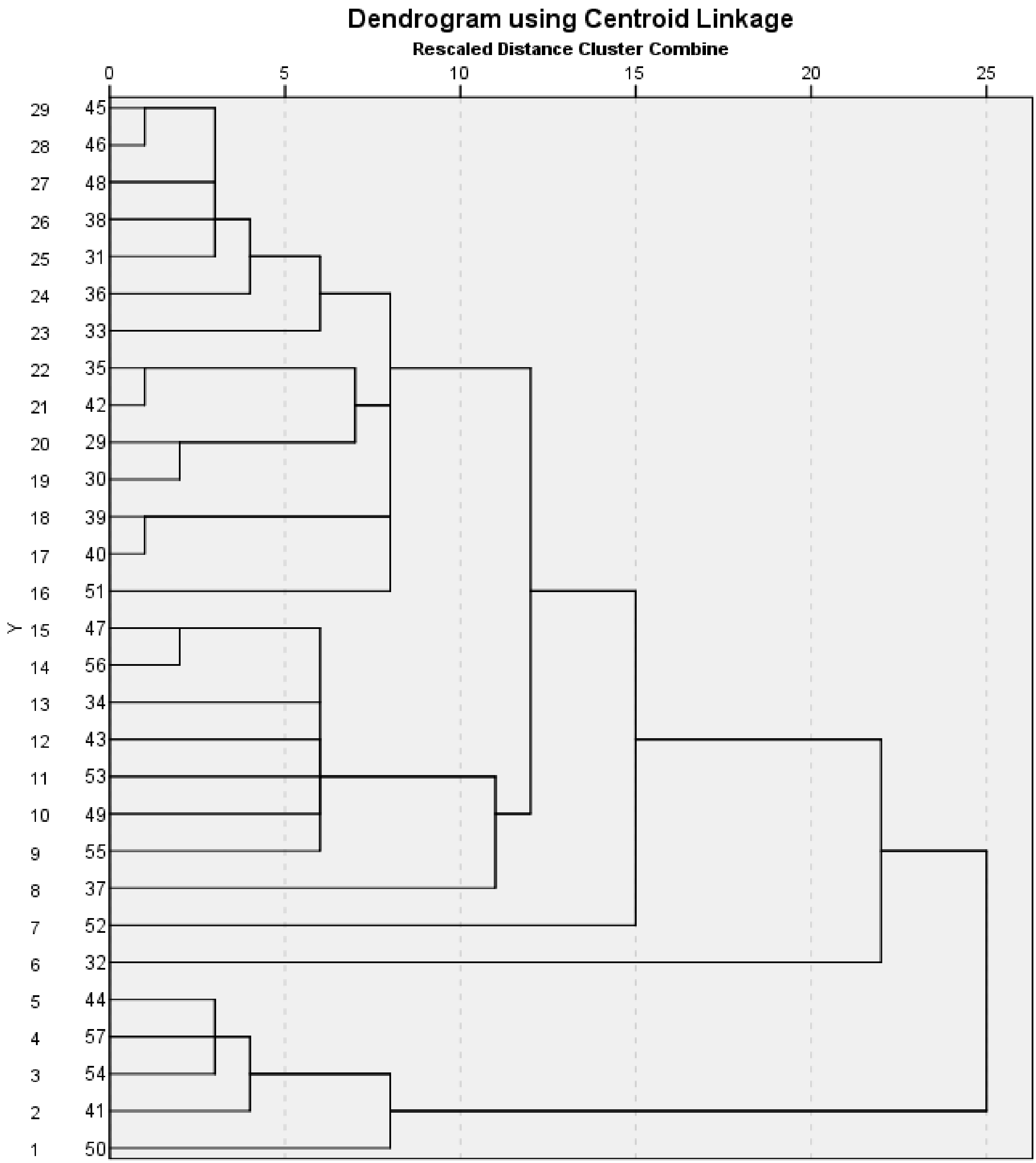
Variables:	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21
1. AX_RT	1,00																				
2. AX_Acc	.00	1,00																			
3. Rhym_Rec	.01	-.04	1,00																		
4. Rhym_Prod	.02	.46*	-.20	1,00																	
5. Syll_Seg	.11	.02	.34	-.29	1,00																
6. Syll_Blend	.18	.01	.28	.19	.09	1,00															
7. Phon_Seg	.14	.11	.19	.08	.27	.15	1,00														
8. Phon_Blend	-.12	.38*	-.10	.36	-.11	.18	.63**	1,00													
9. RAN	.09	.08	.23	-.09	.18	.33	.41*	.36	1,00												
10. WR_Cons	.27	.09	-.18	.12	.02	-.01	-.02	-.07	-.02	1,00											
11. NWR_Cons	.09	.19	-.10	-.08	.26	-.41*	.10	-.03	.18	.34	1,00										
12. WL_Cons	-.29	.21	.34	.24	.01	.14	.09	.11	.37	-.19	-.01	1,00									
13. WL_Corr	-.06	-.06	-.06	-.09	.35	-.24	.05	-.15	.29	.01	.40*	.38*	1,00								
14. PCC	-.25	-.23	-.09	-.07	.13	-.24	-.08	.09	-.15	-.07	-.06	-.05	-.02	1,00							
15. PVC	.14	-.06	-.05	-.06	.11	-.19	.11	-.14	.07	.12	.23	.00	.24	.12	1,00						
16. PCCI	.12	-.12	-.11	-.05	.03	-.23	-.16	-.13	-.27	.12	-.04	-.34	-.03	.58**	.51**	1,00					
17. PCCF	-.13	-.28	-.18	-.06	-.20	-.18	.00	-.12	.19	-.01	.33	.13	.18	.15	.30	-.04	1,00				
18. PCD	-.20	-.04	-.04	.12	-.11	-.13	-.19	-.10	-.32	-.18	-.24	-.27	-.16	.20	-.05	.30	-.18	1,00			
19. MRR_Mono	-.22	.07	.23	-.05	.16	.05	.01	.04	.15	-.26	-.12	.23	.18	-.31	.10	-.10	-.08	.13	1,00		
20. MRR_Bi	-.14	.07	.18	.12	-.12	-.13	.20	.23	.00	-.14	.02	.21	.11	-.16	.07	-.03	.05	.32	.64**	1,00	
21.MRR_Tri	.04	-.10	.27	.03	-.05	-.23	.02	.04	.12	-.03	-.06	-.04	-.06	.12	-.09	.26	.02	.32	.39*	.45*	1,00

*p<.05, ** p< .01

Appendix 5. Agglomeration Schedule

Stage	Cluster Combined		Coefficients	Stage Cluster First Appears		Next Stage
	Cluster 1	Cluster 2		Cluster 1	Cluster 2	
1	45	46	5,201	0	0	8
2	39	40	6,121	0	0	20
3	35	42	6,543	0	0	19
4	47	56	6,832	0	0	17
5	29	30	7,552	0	0	19
6	44	57	8,401	0	0	7
7	44	54	7,319	6	0	11
8	45	48	8,930	1	0	9
9	38	45	8,129	0	8	10
10	31	38	8,713	0	9	12
11	41	44	9,611	0	7	23
12	31	36	9,939	10	0	13
13	31	33	12,193	12	0	22
14	34	43	12,441	0	0	15
15	34	53	12,653	14	0	16
16	34	49	12,289	15	0	17
17	34	47	10,576	16	4	18
18	34	55	10,248	17	0	24
19	29	35	14,024	5	3	21
20	39	51	14,807	2	0	21
21	29	39	15,525	19	20	22
22	29	31	15,294	21	13	25
23	41	50	15,651	11	0	28
24	34	37	18,829	18	0	25
25	29	34	20,214	22	24	26
26	29	52	25,124	25	0	27
27	29	32	34,818	26	0	28
28	29	41	39,229	27	23	0

Appendix 6. Dendrogram for cluster analysis



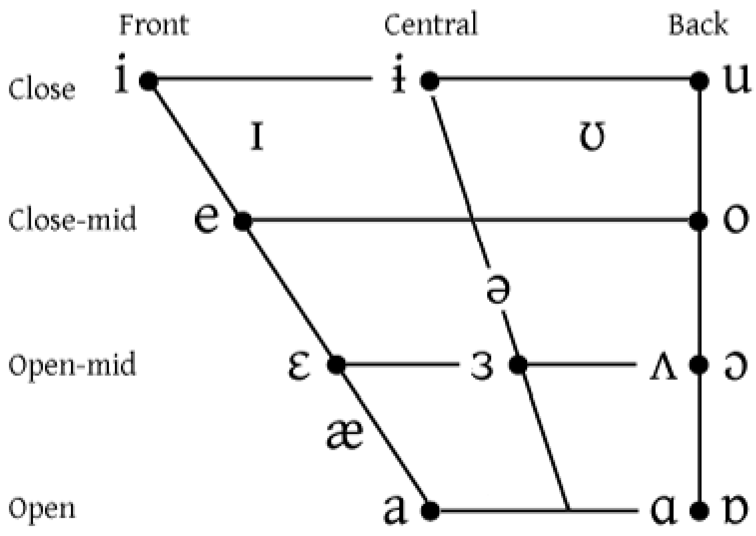
Appendix 7. International Phonetic Alphabet (IPA) symbols for Croatian consonants (Horga & Liker 2016: p266).

MJESTO / NAČIN		BILABIJALNI	LABIODEN-TALNI	DENTALNI	ALVEOLARNI	POSTALVEO-LARNI	ALVEOLO-PA-LATALNI	PALATALNI	VELARNI
STUPANJ SUŽ.	OKL	p b		t d					k g
	AFR			ts		tʃ . dʒ	tɕ dʑ		
	FRI		f	s z		ʃ ʒ			x
	APR		ʋ					j	
OBLIK SUŽ.	NAZ	m		n			ɲ		
	LAT			l			ʎ		
DINAMIKA	VIB				r				

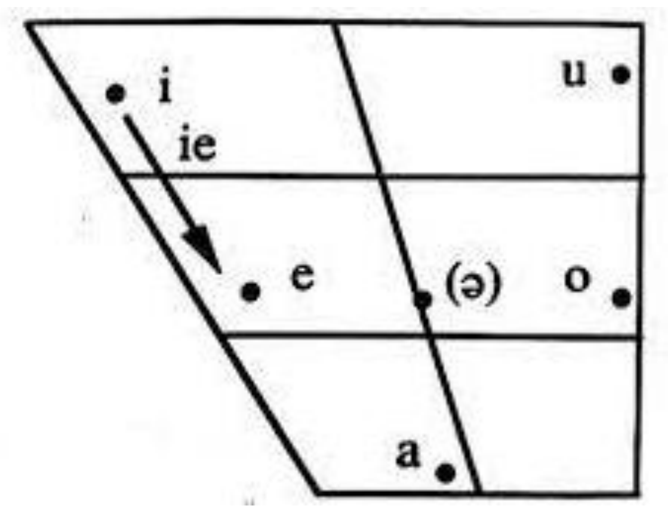
Appendix 8. International Phonetic Alphabet (IPA) symbols for English consonants (Stanford, 2010)

	Bilabial		Labio-dental		(Inter-) dental		Alveolar		Palato-alveolar		Velar		Glottal	
Stop	p	b					t	d			k	g		
Fricative			f	v	θ	ð	s	z	ʃ	ʒ			h	
Affricate									ʧ	ʤ				
Nasal		m						n				ŋ		
Lateral								l						
Approximant		w						r		j				

Appendix 9. IPA English vowel chart (Lin, 2004)



Appendix 10. IPA Croatian vowel chart (Landau et al., 1999; p.67)



10. CURRICULUM VITAE

Dora Knežević was born in Split, Croatia, in 1992. After completing her Master's degree in Speech and Language Pathology (SLP) at the Faculty of Education and Rehabilitation Sciences, University of Zagreb in 2015, she began working as an SLP at the Potočnica Kindergarten in Zagreb. Since 2016, she has been working as a research and teaching assistant at the Department of Speech and Language Pathology at the Faculty of Education and Rehabilitation Sciences, University of Zagreb, under the supervision of Prof. Draženka Blaži, PhD. Since 2017, she has been pursuing a PhD at the Postgraduate Programme of Speech, Language and Hearing Disorders at the same Faculty. Her main scientific interests are speech sound disorders and motor speech disorders in children. She actively collaborates with colleagues from different Croatian and foreign institutions. She is part of the SLP Unit at The Center for Rehabilitation, a teaching base and clinical unit of the Faculty of Education and Rehabilitation Sciences, where she does her clinical work. She is also a member of the Croatian Logopedics Association, where she works for the promotion, modernization and digitization of SLP in Croatia. She also became a member of the Motor Speech Committee of the International Association for Logopedics and Phoniatrics (IALP) since 2021, but she is most proud of the Association of Students of Speech and Language Pathology Logomotiva, which she founded with students in 2019 and still actively mentors and supports. She has several professional and personal interests in which she continues her education through various seminars and workshops. She has participated in several national and international conferences and has published four scientific and one professional paper.

Knežević, D. (in press). Motor abilities of children with Childhood apraxia of speech. *The Croatian Review of Rehabilitation Research*

Olujić Tomazin, M., Matić Škorić, A., & Knežević, D (in press). Students' experiences and characteristics of online teaching at the beginning of the COVID pandemic in Croatia. In A. Zovko, N. Vukelić, I. Miočić, (Eds). Prema postpandemijskom obrazovanju: kako osnažiti sustav odgoja i obrazovanja.

Knežević, D.,Blaži, D., & Hostonski, J. (2020). Govorno-motoričke sposobnosti djece s kašnjenjem u jezično-govornom razvoju. Klinička psihologija, 13, 1-2, 35-46. https://doi.org/10.21465/2020-KP-1-2-0003

Blaži, D., Knežević, D. i Zglavnik, I. (2020). Načini hranjenja i roditeljska zabrinutost kod djece s različitim vrstama orofacijalnih rascjepa. *Logopedija, 10* (1), 1-6. https://doi.org/10.31299/log.10.1.1

Knežević, D. (2019). Are children with childhood apraxia of speech a subgroup of children with developmental coordination disorders?. *Logopedija*, *9* (1), 9-13. https://doi.org/10.31299/log.9.1.2