Describing speech abilities and trajectories of speech development in a heterogeneous group of children with autism

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Declaration of Authorship

This thesis has not been submitted for any other degree at The University of Sydney or any other university. The work contained in this thesis is my own. To the best of my knowledge and belief, no work has been published elsewhere or written by any other authors, except when appropriately referenced within the thesis. Approval for these studies was given by The University of Sydney Human Ethics Committee (Reference numbers: 2012/712, 2012/1305).

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Declaration of Completion

This is to certify that the thesis entitled *Describing speech abilities and trajectories of speech development in a heterogeneous group of children with autism,* submitted by Kate Broome in fulfilment of the requirements for the degree Doctor of Philosophy, is in a form ready for examination.

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Publications and Presentations Arising from this Thesis

Peer reviewed publications

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Peer reviewed presentations

Broome, K., McCabe, P., & Docking, K. (2014). *Speech development in children with autism spectrum disorder.* Presentation at the Speech Pathology Australia National Conference, Melbourne, Victoria, Australia, 2014.

Broome, K., McCabe, P., Doble, M., & Docking, K. (2017). *Speech is not language: A longitudinal study reporting the speech development of children with autism spectrum disorder.* Presentation at the Speech Pathology Australia National Conference, Sydney, New South Wales, Australia, 2017.

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Notes on Style

This is a thesis with publications, and as such three papers in the style of journal articles are embedded in the thesis. These articles represent Chapters 2, 3, and 4. As this thesis is comprised of individually published studies, there is some inherent repetition, particularly in the required background information for each paper. Attempts were made to minimise repetition of information as much as possible.

Spelling

- The three included journal articles (Chapters 2, 3, and 4) are written in American English in accordance with the instruction for authors from the respective journal in which the articles were published or submitted for publication.
- The remainder of the thesis (Chapters 1, 5, 6, and 7) is written in Australian/British English.

Style

- The three journal articles (Chapters 2, 3, and 4) are written in styles requested by the relevant journal in which the article was published or submitted for publication.
- The remainder of the thesis is written in accordance with APA 7th style, except for tables, which are presented single-spaced for ease of reading.
- The updated systematic review (Chapter 6) includes only a short introduction to reduce repetition between chapters. To assist comparison between the update (Chapter 6) and the original systematic review (Chapter 2), a similar style was adopted for the two chapters.
- Phonetic symbols and diacritics are from the International Phonetic Alphabet (IPA).
- Semicolons are used to refer to a child’s age in years;months.
- References are embedded in the text of the thesis with reference lists found at the end of each chapter.

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Glossary of Acronyms

Acronym	Meaning
AC	Auditory Comprehension
AD	Autistic Disorder
ADOS	Autism Diagnostic Observation Schedule
ALSPAC	Avon Longitudinal Study of Parents and Children
AMMT	Auditory Motor-Mapping Training
AMR	Alternating Motion Rate
APA	American Psychiatric Association
ASD	Autism Spectrum Disorder
ASHA	American Speech-Language-Hearing Association
ASPECT	Autism Spectrum Australia
BPVS-II	British Picture Vocabulary Scale – Second Edition
CAS	Childhood Apraxia of Speech
CBR	Canonical Babbling Ratio
CDI	MacArthur-Bates Communicative Development Inventory
CELF-3	Clinical Evaluation of Language Fundamentals – Third Edition
CELF-4	Clinical Evaluation of Language Fundamentals – Fourth Edition
CELF-P	Clinical Evaluation of Language Fundamentals – Preschool Edition
CINAHL	Cumulative Index to Nursing and Allied Health Literature
CSBS	Communication and Symbolic Behavior Scales
C	Consonant
CA	Chronological Age
DAS	Differential Abilities Scales
DEMSS	Dynamic Evaluation of Motor Speech Skill
DQ	Developmental Quotient
DSM-IV	Diagnostic Statistical Manual of Mental Disorders – Fourth Edition
DSM-5	Diagnostic Statistical Manual of Mental Disorders – Fifth Edition
EC	Expressive Communication

ELO	Evaluation du Language Oral
ERIC	Education Resources Information Centre
ESCS	Early Social and Communicative Scales
EVT	Expressive Vocabulary Test
FWFST	First Words, First Sentences Test
GFTA-2	Goldman-Fristoe Test of Articulation – Second Edition
GMDS-ER	Griffiths Mental Development Scale – Extended Revised
HAPP-3	Hodson Assessment of Phonological Patterns – Third Edition
HCA	Hierarchical Cluster Analysis
IQ	Intelligence Quotient
KSPT	Kaufman Speech Praxis Test
LENA	Language Environment Analysis
LLBA	Linguistic and Language Behavior Abstracts
MBL	Mean Babbling Level
MLU	Mean Length of Utterance
MSD-NOS	Motor Speech Disorder – Not Otherwise Specified
MSEL	Mullen Scales of Early Learning
NDIS	National Disability Insurance Scheme
NVDQ	Nonverbal Developmental Quotient
NVIQ	Nonverbal Intelligence Quotient
PCC	Percentage of Consonants Correct
PCC-R	Percentage of Consonants Correct – Revised
PDD-NOS	Pervasive Developmental Disorder – Not Otherwise Specified
PEPS-C	Profiling Elements of Prosodic Speech in Children
PLS-4	Preschool Language Scale – Fourth Edition
POP	Polysyllable Preschool Test
PPC	Percentage of Phonemes Correct
PPVT-R	Peabody Picture Vocabulary Test – Revised
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-Analyses

PVC	Percentage of Vowels Correct
RPM	Raven's Progressive Matrices
SAEVD-R	Stark Assessment of Early Vocal Development - Revised
SDCS	Speech Disorders Classification System
SLP	Speech-Language Pathologist
SMR	Sequential Motion Rate
SMRS	Scientific Merit Rating Scale
SSD	Speech Sound Disorder
SSL	Syllable Structure Level
SWPT	Single Word Polysyllable Test
TLC	Test of Language Competence
TLS	Total Language Score
TROG-2	Test for Reception of Grammar – Second Edition
V	Vowel
VDQ	Verbal Developmental Quotient
VIQ	Verbal Intelligence Quotient
VMPAC	Verbal Motor Production Assessment for Children
WAIS	Wechsler Adult Intelligence Scale
WISC-3	Wechsler Intelligence Scales – Third Edition
WISC-IV	Wechsler Intelligence Scales – Fourth Edition
WISC-V	Wechsler Intelligence Scales – Fifth Edition
WPPSI – III	Wechsler Preschool and Primary Scale of Intelligence – Third Edition

The research presented in this thesis spans many years. Over this time, there have been considerable developments in the terminology used in autism research. For example, there has been an important shift from disorder to difference, and towards identify-first language. The language used throughout this thesis reflects this changing terminology.

Preface

The initial inspiration for this research came from children on the autism spectrum on my own clinical caseload. While most children on the spectrum presented with impairments in the areas of language and social communication, a small group of children appeared to have significant speech deficits. Over time a pattern began to emerge. For some children, their speech ability was more impaired than other aspects of their communication profile, such as their receptive language and nonverbal communication, and seemed to be a core barrier to their development of verbal communication. Many of these children made amazing gains in their social communication, increasing their desire to communicate, and developing functional alternative and augmentative communication methods. Some children even learnt to read and write simple words. However, sometimes despite years of early intervention, their speech developed very little.

The parents of these children wanted to understand why their children were not progressing verbally. As a clinician faced with an unexpected presentation, I turned to the literature looking for evidence and information about these children. Low verbal children were under-represented in the literature, and it was impossible to provide parents with information regarding possible aetiology and prognosis of their child's communication deficit. Most communication research focused on the social communication and language deficits experienced by many children on the autism spectrum, providing little guidance on best practice for the assessment and management of speech impairments in this population. My colleagues and I had to extrapolate from existing speech sound disorder literature, trialing assessment and intervention approaches not yet researched with children with autism. Frustrated with the lack of research and determined to find the evidence for these children and their families, I began this doctoral research.

Thesis Abstract

Children diagnosed with autism spectrum disorder (ASD) form a heterogeneous population. While the variation in language and social communication abilities are well documented, little is known about the speech of children on the spectrum. The small body of research to date reports three main findings: 1) differences in the prelinguistic speech of young children with ASD relative to typically developing children, 2) the presence of a small subgroup of minimally verbal children with a significant speech sound disorder (SSD), and 3) high rates of mild speech difficulties in older highly verbal children with ASD. The speech capacity and development of children across the entire autism spectrum remains largely undescribed. This thesis aimed to provide detailed descriptive speech data for a heterogeneous group of children with ASD, to explore the possibility of subgroups based on this speech data, and to examine the trajectories of speech development in these children. To address these aims, four main projects are reported in this thesis.

Firstly, a systematic review was completed to provide a summary of speech assessment practices used in research with children with ASD. A systematic search of eight databases was used to find peer-reviewed research articles published between 1990 and 2014. The systematic review identified 21 articles that met the inclusion criteria. Assessment methods included connected speech samples, single-word naming tasks, speech imitation tasks, and analysis of the production of words and sentences. Given the large variation in participant characteristics and reporting standards in the studies included in the systematic review, it was difficult to draw comparisons. As part of the systematic review, a narrative review was completed to ascertain the core components of an evidence-based paediatric speech assessment which, together with the results of the systematic review, provide clinical and research guidelines for best practice. The results of this systematic review were used to guide assessment selection in the subsequent longitudinal study.

The second project is a cross-sectional study reporting the results from the first data point of the longitudinal study and is included as a published paper. This study aimed to provide detailed descriptive baseline speech data and then use this data to explore whether subgroups exist within a heterogeneous cohort based on speech ability. Despite growing interest in the area of speech and autism, large gaps remain in the literature. There is limited information regarding the speech ability of young children with ASD across a range of functional levels, and few studies have reported detailed description of the speech skills of children with ASD. This study included 23 children aged 2;0-6;11 years with a diagnosis of ASD. Independent and relational speech analyses are reported from single-word naming tasks and spontaneous speech samples. Hierarchical cluster analysis identified three descriptive speech subgroups: A) children with high receptive and expressive vocabularies, high nonverbal communication, and high speech ability (n = 10), B) children with very low expressive vocabularies and low speech ability, but higher receptive vocabularies and nonverbal communication (n=3), and C) children with low vocabularies, low nonverbal communication, and low speech development (n=10). This is the first study to provide detailed descriptive speech data of a heterogeneous cohort of children with ASD and to use these data to explore the possibility of subgroups. Clustering suggests a small number of children may present with a unique communication profile which warrants further exploration.

The third project presents the data from the longitudinal study for 22 of the same children described in the second project. This is the first longitudinal study detailing the speech development of children with ASD. The aims of this study were: 1) to describe changes in participant’s speech capacity over 12 months, 2) examine the stability of cluster membership over 12 months, and 3) describe what variables may explain changes in speech capacity over time. Four clusters emerged from clustering. Cluster membership remained stable for Cluster A and Cluster B children from Time 1 to Time 2. The Cluster C children from Time 1 had varied trajectories of speech development. One child made significant gains and joined Cluster A at Time 2 (n=11). Three children remained at the prelinguistic stage of language development and made very little speech gains over 12 months. These three children formed Time 2 Cluster C (n=3). Five children made gains across all areas of communication and formed Time 2 Cluster D (n=5). Findings of this study suggest that a child's consonant repertoire and receptive vocabulary at Time 1 may be important variables to predict cluster membership at Time 2.

Chapter 5 provides further detail regarding the three Cluster B children. These children presented with a consistent and unique communication profile of high receptive vocabularies and use of nonverbal communication, in the presence of low speech and low expressive vocabularies. This profile suggests a co-occurring speech sound disorder (SSD). The challenge of differentially diagnosing an SSD in minimally verbal children is discussed.

Finally, an update of the original systematic review is presented in Chapter 6 to summarise the current state of evidence for the speech assessment of children with ASD. This update includes the results of a systematic search of the same eight databases using the same search terms, to find peer-reviewed research articles published between January 2015 and August 2021. Twenty-seven articles met inclusion criteria. There has been a significant increase in studies, particularly those investigating the speech of minimally verbal children with ASD. This research has seen a rise in studies using speech imitation tasks to assess the speech of less verbal children. Further, a number of studies adopted multiple assessment measures to describe the speech of children with ASD, in keeping with best practice speech guidelines. Together with the results from the longitudinal study, future research and clinical speech assessment guidelines are discussed.

Collectively, the results from the four studies in this thesis highlight the value of describing the speech capacity of children with ASD across the heterogeneous spectrum. Children at all linguistic levels can complete a speech assessment, although some specific modifications for children with ASD should be considered. For example, we recommend including echolalia in a child's speech sample, collecting speech samples whenever the child is most vocal - which may not be when interacting with others in play, and considering the whole communication profile of the child when interpreting their speech ability. Regarding the last point, many children with ASD have co-occurring language and social communication difficulties, and therefore, a score below normal limits on formal standardised assessments or poor speech ability during sampling, does not necessarily indicate a speech sound disorder. Some children may have low levels of speech, language and nonverbal communication. The results from the systematic reviews and longitudinal study suggest that a strengths-based speech assessment, focused on what the child can do, provides important descriptive information for differential diagnosis, baseline data collection, and intervention planning.

Results from the longitudinal study suggest descriptive speech subgroups exist within the heterogeneous population of children with ASD. These subgroups emerged from the data even when the number or type of subgroups were not selected a priori. Children with low language, nonverbal communication, and speech at Time 1 had varied communication trajectories. Some children who initially presented as low verbal made significant gains and were verbal by Time 2. Further, a small subgroup of children with ASD present with a unique communication profile, with high levels of receptive vocabulary and nonverbal communication in the presence of very low expressive vocabulary and speech ability. These children do not develop speech along the same trajectory as children with comparable receptive vocabularies. Over 12 months, the speech capacity of children in this subgroup did not increase, despite improvements in receptive vocabulary and nonverbal communication. This profile suggests a co-occurring SSD, although more data is required to differentially diagnose a motor speech impairment from a phonological disorder. The combination of a child's receptive vocabulary and consonant repertoire may predict the trajectory of speech development. Further research is required to explore these findings.

Chapter 1: Introduction

"The example of autism shows particularly well how even abnormal personalities can be capable of development and adjustment. Possibilities of social integration which one would never have dreamt of may arise in the course of development. This knowledge determines our attitude towards complicated individuals of this and other types. It also gives us the right and the duty to speak out for these children with the whole force of our personality." — Hans Asperger, 1944

The evolution of autism

In 1944, paediatrician Hans Asperger published some of the earliest descriptions of children with autism, descriptions strikingly similar to current accounts nearly 80 years later. To him, autism was not a rare condition of infancy, but instead observed in all ages, across all sectors of society. In his seminal work, Asperger described the diversity of presentations of autism, from children who were nonverbal, to those meticulous and precocious in their language use (Silberman, 2015). He emphasised the importance of focusing on children's strengths rather than their impairments (Baron-Cohen, 2015). Asperger's name became synonymous with the intelligent little 'professors' he described in his 1944 publication (Wing, 2005), somewhat clouding his previous and ongoing descriptions of heterogeneity. Understanding children's neurological differences and focusing on their strengths, speaks to the heart of the modern neurodiversity movement (Baron-Cohen, 2015). Unfortunately, the path for children with autism since the 1940s has been far from smooth.

Asperger's work was overshadowed by child psychiatrist Leo Kanner's descriptions published the year earlier. Infantile autism was described as a rare condition of early childhood. Parents of children with autism were described as cold and unemotional and were blamed for causing their child's autism (Silberman, 2015). In the years that followed, children with autism were placed in institutions and parents were discouraged from visiting.

It would take many decades for the world to return to the concept of autism as a spectrum of behaviours, ranging in severity and presentation.

Lorna Wing, a British psychologist and the mother of a child with autism, reclaimed the notion of a broad spectrum in the 1970s. Along with her colleague, Judith Gould, she reported much higher prevalence rates of autism than the previously reported 4 in 10,000 (Wing & Gould, 1979). Prevalence rates for autism spectrum disorder (ASD) have increased exponentially since the 80s, possibly reflecting broader diagnostic criteria, increased autism awareness, and changes in diagnostic preferences (May et al., 2017). In Australia, it is estimated that one in 50 children live with ASD (May et al., 2017; Randell et al., 2015), a rate similar to estimates from the United States (Maenner et al., 2020) and the United Kingdom (Roman-Urrestarazu et al., 2021). With the rise in diagnoses, there has thankfully been a shift away from the harsh historical treatment of children with autism, toward an improved understanding of the differences in the brain development of these children.

The term neurodiversity was first used by Australian sociologist Judy Singer in her honours thesis in 1998 and published in a book chapter in the following year (Singer, 1999). Neurodiversity recognises the natural strengths and gifts of cognitive varied individuals, and the many societal barriers that may contribute to their disability (DeThorne & Searsmith, 2021; Singer, 2017). Neurodiversity is more of a movement than a word. Under this paradigm, children with autism and their families are empowered to find supports, to foster their strengths, and lead their life on their own terms. In Australia, the current National Disability Insurance Scheme (NDIS) aims to provide neurodiverse individuals and their families these exact freedoms of choice and support. It is time to look beyond the autism diagnosis and see the individual, with their unique strengths and specific needs.

Heterogeneity

Autism is now widely recognised as a heterogeneous, developmental, lifelong, neurodiverse diagnosis. According to the current Diagnostic Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association, 2013), a diagnosis of ASD is made when a child presents with both restricted and repetitive interests and behaviours, and social communication impairments. Language and cognitive impairments are not core features of ASD, and therefore, children with this diagnosis can vary widely in these domains. While some children present with superior language skills, it is estimated that around 60% of children with ASD have co-occurring language impairments (Levy et al., 2010) and up to 30% of children remain nonverbal or minimally verbal through the preschool years (Norrelgen et al., 2015; Tager-Flusberg & Kasari, 2013).

Interestingly, this modern view is somewhat of a homecoming to the original descriptions outlined by Hans Asperger nearly 80 years ago. Throughout history, people with autism have been excluded, initially from being diagnosed under Kanner's narrow criteria, and then from families and society, and from research. Neurodiversity offers a new chapter of understanding and inclusion. In the studies presented in this thesis, we purposefully set out to capture the diversity of children on the autism spectrum. We did not want to exclude the less verbal children, nor did we desire to omit highly verbal children. Instead, we recognised the value of detailing the variation that exists within the spectrum. Describing individual presentations and subgroups within autism may provide important information regarding aetiology and possible trajectories of development within this population (Georgiades et al., 2013; Loth et al., 2017). Clinicians and parents want to be able to recognise their children in the research. If we continue to exclude and only study homogenous groups, it is possible we are missing something and, more importantly, someone.

Speech of children with ASD

Given the diagnostic criteria for autism, communication research with children with ASD has logically focused on their universal social communication difficulties and common language impairments. Unfortunately, children unable to complete standardised assessments, due to innate challenges such as difficulty following spoken instructions and social cues, and limited attention, have been excluded from previous research (Plesa Skwerer et al., 2016). This has resulted in a paucity of communication data regarding children at early stages of development. It is only in the past decade that research attention has turned to exploring the many barriers to functional expressive language for the less verbal children with ASD (Kasari et al., 2013; Paul et al., 2013; Patten et al., 2014; Thurm et al., 2015). Speech deficits have been hypothesised by clinicians to be one such barrier for a small group of children with ASD (Dawson, 2010; Prizant, 1996), although empirical evidence is limited.

What is speech?

As the speech capacity of children with ASD begins to be researched and reported, it is important that clear terminology is adopted. In the ASD literature, the term *speech* has historically been used to refer to any verbal output, including everything from spoken words, and nonspeech vocalizations (i.e. squeals), to babble and sounds. In this thesis, speech refers to spoken sounds and pronunciation and encompasses articulation, phonology, and motor speech. Articulation refers to the physiological movement involved in producing speech sounds. An impairment at this level represents motor mislearning and usually results in the distorted production of an individual sound (Dodd, 2005). Phonology refers to the sound patterns used in a language (Dodd, 2005). A phonological impairment includes patterns of errors, such as sound or syllable substitutions, omissions, and additions (Vihman, 1996). Children with phonological impairments do not have the same system of sounds and syllables as adult members of their community. Motor speech refers to planning, programming, sequencing, and executing motor movements for speech. Motor speech impairments include Childhood Apraxia of Speech (CAS), dysarthria, and according to the Speech Disorders Classification System (SDCS; Shriberg et al., 2010), motor speech disorders – not otherwise specified. Children were excluded from the studies in this thesis if they had a history of structural (i.e. cleft palate) or sensory/perceptual (i.e. hearing) impairments, and so these speech difficulties are not expected in the studies in this thesis.

An overview of speech research with children with ASD

The body of literature examining the speech of children with ASD is small and inconsistent. While some propose significant speech sound disorders (SSD) which impact functional expressive language development (Chenausky et al., 2019; Rapin et al., 2009), others describe only mild impairments (Shriberg et al., 2011), and some conclude speech to be unimpaired and even advanced in this population (Kjelgaard & Tager-Flusberg, 2001). Given the known variation within the spectrum of autism, heterogeneity in the results is not unexpected. Some of this variation likely reflects differences in participant characteristics, terminology, and assessment procedures. To advance this body of work, consistent reporting standards are needed. This will enable results to be compared and collated and help paint a clearer picture of the speech capacity of children with ASD for future researchers, clinicians, and families. What follows is a brief overview of the literature, both at the inception of the studies in this thesis as well as the current state of evidence. This information is expanded in the systematic review in Chapter 2 and the updated review in Chapter 6.

Prior to commencing the studies in this thesis, most research examining the speech of children with ASD focused on either infants at risk of ASD (e.g., Paul et al., 2011; Plumb & Wetherby, 2013) or highly verbal older children (e.g., Shriberg et al., 2011). Studies of verbal children with ASD identified higher rates of mild articulation and phonological speech difficulties than in the neurotypical population (Cleland et al., 2010; Rapin et al., 2009;

Shriberg et al., 2011). Re-analysis of the Shriberg et al (2011) data suggested a small subgroup of children with ASD previously identified with a speech disorder may present with Speech Motor Delay (SMD; Shriberg et al, 2019). SMD is a newly proposed classification and defined as 'a delay in the development of precise and stable articulation that does not meet criteria for dysarthria or apraxia of speech' (Shriberg, et al., 2019, p. 710). These studies largely focused on school-aged highly verbal children, and it remained unknown if these results could be generalised to younger or less verbal children with ASD. There was a large gap in knowledge regarding the speech capacity of children across all levels of linguistic development, particularly those aged between 2 and 6 years.

A study by Schoen and colleagues (2011) stood out as one of the few to provide detailed accounts of the speech capacity of children with ASD. These authors compared the speech of 30 toddlers with ASD, aged 18-36 months, to language and age-matched controls. All toddlers in the Schoen study could be described as prelinguistic or low verbal, producing less than 30 spoken words. Descriptive data regarding consonant accuracy, single consonant and consonant blend inventories, and mean syllable structure level were reported. Differences between the speech of children with ASD and age-matched controls were found, although the differences were not significant when compared to language-matched controls. Further research was needed to apply this level of detail to older and more verbal children.

Clinicians have long suspected a subgroup of low verbal children with ASD to have co-occurring CAS (Dawson, 2010; Prizant, 1996). Childhood Apraxia of Speech (CAS) is an organic speech disorder of motor planning and programming (ASHA, 2007). At the time the studies in this thesis were designed, there was little evidence to support these clinical hypotheses. Only two research teams had studied this hypothesis directly. Velleman and colleagues (2010) assessed the speech of 10 children with ASD (aged 4;0-6;5) using the Verbal Motor Production Assessment for Children (VMPAC; Hayden & Square, 1999), Hodson Assessment of Phonological Patterns – Third Edition (HAPP-3; Hodson, 2004), and acoustic analysis. All participants were reported to use at least phrase-level expressive language and had a verbal, nonverbal, or combined intelligence quotient (IQ) above 70. Using standard scores from the VMPAC, six of the ten children scored in the 'severe deficit' range and one child scored in the 'moderate deficit' range. Only two participants scored in the average range on the HAPP-3. These results provide preliminary support for the hypothesis of a speech impairment, but not specifically CAS, in some children with ASD. However, caution needs to be taken when interpreting standard scores with this group of children. It is important to interpret speech capacity in light of overall communication ability. Some children in the Velleman et al (2010) study presented with low language and low speech, possibly representing impaired global communication development rather than a specific SSD. Shriberg and colleagues (2011) reported no evidence for CAS in 46 children with ASD (aged 4;0-7;0 years), although their participants were all highly verbal children with high speech intelligibility. It remained unclear if similar findings would emerge when studying the speech of less verbal children.

Since the commencement of the studies in this thesis, there has been a shift in the research towards studying less verbal children with ASD. This move has introduced changes in terminology, further support for a small subgroup of children with ASD and suspected SSDs, and the emergence of speech intervention studies. The term 'minimally verbal' is now used more widely, typically referring to children who produce less than 20 verbal words (Kasari et al., 2013). A number of researchers have been interested in identifying the predictors of expressive language growth for minimally verbal children with ASD (Saul & Norbury, 2020; Woynaroski et al., 2016; Yoder et al., 2015). Results of these studies report early consonant inventory as one such predictor, highlighting the importance of describing the speech of children with ASD.

The series of studies completed by Chenausky and colleagues have added a new level of depth to this area (Chenausky et al., 2016; 2017a; 2017b; 2017c; 2018; 2019; 2021). In their 2019 paper, Chenausky and colleagues studied the speech of 54 minimally verbal and low verbal children with ASD, aged 4;4 – 18;10. Their responses during the Kaufman Speech Praxis Test (Kaufman, 1995) were coded for signs of CAS. Children were categorized as within normal limits (n=12), non-CAS speech difficulties (n=16), suspected CAS (n=13), or insufficient speech (n=13). The results of this study add support for the possibility of both functional (i.e. articulation and phonology) and organic (i.e. CAS) speech sound disorders in less verbal children with ASD. Further research is needed to examine if similar subgroups emerge when latent grouping methods are employed with a more heterogeneous cohort of children.

Identifying speech differences in children with ASD is of clinical and theoretical importance. Clinically, children with ASD and a suspected speech deficit will require additional tailored speech assessment and may need targeted speech interventions that are not typically included in early intervention with this population. Theoretically, whether speech differences in children with ASD represent comorbidity or a specific phenotype is out of the scope of this thesis. Instead, it is hoped that the information presented here provides preliminary evidence to base future population studies in this area.

Chapters in this thesis

The primary study in this thesis is a longitudinal study describing the speech capacity and development of a heterogeneous cohort of children with ASD. To inform study design and assessment selection, we first needed to review the literature for best assessment practices with children with ASD.

Chapter 2: Systematic review

Chapter 2 is a published systematic review (Broome et al., 2017) reporting speech assessment practices in peer-reviewed papers published between 1990 and 2014. Children with ASD have known difficulties completing standardised assessments (Charman et al., 2003). Some examples of these challenges include difficulties following the lead of the examiner, maintaining attention, understanding the instructions, and social anxiety in novel situations (Plesa Skwerer et al., 2016; Tager-Flusberg, 1999). This has led to children with ASD not being well described and comments that children could not be assessed rather than descriptions of abilities obtained through other means, such as using informal assessments, or swapping assessments for other tools. A primary aim of our longitudinal study was to capture the speech capacity in a heterogeneous cohort of children with ASD. To achieve this goal, we assumed a different approach to assessment than what is used with neurotypical children would be needed, and turned to the literature for guidance. Chapter 2 outlines the results of this review and provides clinical and research guidelines that were considered when designing the following studies.

Chapter 3: Cross-sectional study

Chapter 3 presents a published paper outlining the cross-sectional results from Time 1 in the longitudinal study (Broome et al., 2021). Cross-sectional studies have dominated the speech literature with children with ASD to date. Studies of this type can provide valuable detailed information of abilities at one point in time. Unfortunately, few studies have provided the level of detail needed to draw comparisons between studies or to describe individual abilities and variation. The cross-sectional study presented in Chapter 3 aimed to provide this level of detail regarding the speech capacity of children with ASD and to explore whether descriptive speech subgroups exist within the heterogeneous cohort. The Time 1 data was then used as important baseline data in which to explore speech growth in the following longitudinal study.

Chapter 4: Longitudinal study

Chapter 4 presents the results from the 12-month longitudinal study (Broome et al., submitted). This study is the first to report descriptive speech trajectories for a cohort of children with ASD. The paucity of longitudinal studies in this area limits our profession's capacity to provide information regarding possible SSD prognoses. Longitudinal studies, encompassing different abilities and age ranges are of particular importance as we endeavour to understand the inherent heterogeneity in the autism spectrum (Loth et al., 2017). The results presented in Chapter 4 provide important longitudinal data to expand our knowledge of the speech of children with ASD and the possible barriers to their verbal development.

Chapter 5: A closer look at the children in Cluster B

Three children from the cross-sectional and longitudinal studies in Chapters 3 and 4 presented with a unique communication profile that remained stable over 12 months. These children had strengths in their receptive vocabulary and nonverbal communication, in the presence of very low expressive vocabularies and speech capacity. Further detailed descriptive data for these three children are presented in Chapter 5, along with a discussion regarding the complexities of differentially diagnosing an SSD in low verbal children.

Chapter 6: Updated systematic review

Chapter 6 reports results from an update to the original systematic review in Chapter 2. This update was completed to examine the current research evidence and provide updated context in which to interpret the findings of the studies in this thesis and explore possible directions for future research in this area. Published peer-reviewed articles from 2015 to August 2021 were included in this update. Results indicate that a number of well-designed studies focused on examining the speech of minimally verbal children with ASD have been published since the original review. Speech treatment studies with children with ASD are also beginning to emerge in the literature.

Chapter 7: Discussion

Chapter 7 brings together the learnings of the systematic reviews, cross-sectional study, and longitudinal study in this thesis. Collectively, the studies in this thesis highlight the value of describing the speech capacity of children across the heterogeneous autism spectrum. The implications of this body of work are discussed in relation to future research and clinical practice.

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Chapter 2: A Systematic Review of Speech Assessments for Children with Autism Spectrum Disorder: Recommendations for Best Practice

Paper 1: A Systematic Review of Speech Assessments for Children with Autism Spectrum Disorder: Recommendations for Best Practice

The paper presented in this chapter has been published as:

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Author Contribution Statement

As co-author of the above paper and primary supervisor, I confirm that Kate Broome made the following contributions:

- Conception of the research questions and search terms in collaboration with co-authors
- Database searches, data entry and data analysis
- Interpretation of data in collaboration with co-authors
- Writing the first draft of the paper with subsequent drafts developed in collaboration with co-authors
- Journal submission
- Journal revisions in collaboration with co-authors and resubmission

Name: Patricia McCabe

Date: 28 November 2021

Note. Table 1 incorrectly has crosses not ticks next to Cleland et al (2010), DeMouy et al (2011) and Grossman et al (2010). These articles were included in the systematic review.

AJSLP

Review Article

A Systematic Review of Speech Assessments for Children With Autism Spectrum Disorder: Recommendations for Best Practice

Kate Broome,[a,b] Patricia McCabe,[a] Kimberley Docking,[a] and Maree Doble[a]

Purpose: The purpose of this systematic review was to provide a summary and evaluation of speech assessments used with children with autism spectrum disorders (ASD). A subsequent narrative review was completed to ascertain the core components of an evidence-based pediatric speech assessment, which, together with the results of the systematic review, provide clinical and research guidelines for best practice.
Method: A systematic search of eight databases was used to find peer-reviewed research articles published between 1990 and 2014 assessing the speech of children with ASD. Eligible articles were categorized according to the assessment methods used and the speech characteristics described.
Results: The review identified 21 articles that met the inclusion criteria, search criteria, and confidence in ASD diagnosis. The speech of prelinguistic participants was assessed in seven articles. Speech assessments with verbal participants were completed in 15 articles with segmental and suprasegmental aspects of speech analyzed. Assessment methods included connected speech samples, single-word naming tasks, speech imitation tasks, and analysis of the production of words and sentences.
Conclusions: Clinical and research guidelines for speech assessment of children with ASD are outlined. Future comparisons will be facilitated by the use of consistent reporting methods in research focusing on children with ASD.

Autism spectrum disorders (ASD) are a group of complex neurodevelopmental disorders characterized by impairments in social communication and interaction, in addition to the presence of restricted and repetitive behaviors (American Psychiatric Association, 2013). A comprehensive communication assessment is a core component of the diagnostic evaluation for ASD (Ozonoff, Goodlin-Jones, & Solomon, 2005) and guides intervention planning. The challenge when completing this assessment is to utilize measures that capture each individual's communication ability. As many children with ASD use self-directed or context-specific communication, they may have particular difficulty demonstrating their ability during a formal assessment (Charman, Drew, Baird, & Baird, 2003; Paul, Chawarska, Cicchetti, & Volkmar, 2008). There is extensive literature available detailing the language deficits of children with ASD, and guidelines for language assessment have been provided (e.g., Charman et al., 2003; Volden et al., 2011). Yet very little is known about the speech development of these children. If speech deficits are present in the ASD population, speech assessments should be included in the routine communication assessment. Speech-language pathologists (SLPs) would benefit from consistent research evidence indicating best practice when completing a speech assessment with children with ASD.

Why Should We Assess the Speech of Children With ASD?

We should assess the speech of children with ASD because: (a) the results can be used as an early positive behavioral marker for ASD, and (b) there are anecdotal reports of a higher incidence of speech sound disorders in children with ASD. These points are explained further below.

Positive Behavioral Marker for ASD

Children diagnosed with ASD at a young age presently have better access to early intervention services, with identification of positive behavioral markers present prior to 3 years of age vital (Chawarska, Klin, Paul, & Volkmar, 2007). Expressive language delay is often the first indicator

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to prompt investigation of children with ASD (De Giacomo & Fombonne, 1998). However, because many aspects of language difficulty overlap with other developmental disorders, a differential diagnosis of ASD at a young age can be challenging (Tager-Flusberg, Paul, & Lord, 2005).

A difference in the prelinguistic vocalizations of young children later diagnosed with ASD has also been suggested as an important positive behavioral marker characterizing this disorder (Plumb & Wetherby, 2013; Schoen, Paul, & Chawarska, 2011). Prelinguistic vocalizations are defined as those sounds that are produced prior to the use of first words. They are typically categorized into speech-like and non-speech-like vocalizations. Early speech-like sounds provide the foundation for the emergence of first words. Models of early vocal development typically describe four to five stages, from early reflexive sounds, to the use of cooing, simple babble, reduplicated babble and finally complex variegated vocalizations (Mitchell et al., 2006; Nathani, Ertmer, & Stark, 2006; Stark, 1980). This development typically occurs in the first 12 months of life, after which time a dramatic increase in the use of complex vocalizations occurs and first words emerge. The development of prelinguistic vocalizations in children with ASD may differ from typically developing children. Children with ASD have been reported to produce fewer speech-like sounds, fewer consonants and fewer complex syllable forms during the prelinguistic stage of development (Paul, Augustyn, Klin, & Volkmar, 2005; Paul, Fuerst, Ramsay, Chawarska, & Klin, 2011; Werner & Dawson, 2005).

Speech Sound Disorders in Children With ASD

The speech development of children with ASD in the linguistic stage of development has not been widely studied. The few studies that have been completed vary greatly in research methodology and in participant numbers, age and level of functioning. Not surprisingly, the results of these studies also vary, making it difficult to make comparisons and draw clear conclusions. Delayed speech development (Cleland, Gibbon, Peppé, O'Hare, & Rutherford, 2010; Rapin, Dunn, Allen, Stevens, & Fein, 2009; Shriberg, Paul, Black, & Santen, 2011; Shriberg et al., 2001; Wolk & Edwards, 1993; Wolk & Giesen, 2000), disordered speech development (Cleland et al., 2010; Rapin et al., 2009; Wolk & Edwards, 1993; Wolk & Giesen, 2000) and advanced speech development (Kjelgaard & Tager-Flusberg, 2001) have all been reported. Such variability in the speech development of children with autism typifies the heterogeneity of the population.

Pediatric speech sound disorders (SSD) overall are a group of disorders affecting a child's ability to produce intelligible speech. Impairments can occur at the linguistic, motor planning, or motor execution stages of speech production. A variety of definitions and classification systems have been used to describe the subgroups of pediatric SSDs (Campbell et al., 2003; Dodd, 2005; Ingram, 1997; Shriberg, Austin, Lewis, McSweeny, & Wilson, 1997b) with the terms *articulation disorder, phonological disorder,* and *motor speech disorder* commonly used. The difference in the labels given to children with SSDs may explain, in part, the wide variation in reported prevalence rates within the neurotypical population (McKinnon, McLeod, & Reilly, 2007). There is a general trend of higher prevalence rates of SSDs in children across the population at younger ages compared to older children (Campbell et al., 2003; Law, Boyle, Harris, Harkness, & Nye, 2000; McKinnon et al., 2007; Shriberg, Tomblin, & McSweeny, 1999). For example, 5%–15% of preschool-aged children were reported to have an SSD (Campbell et al., 2003; Law et al., 2000) compared with 1%–6% of primary school-aged children (McKinnon et al., 2007; Shriberg et al., 1999).

More recent research suggests that children with ASD are at significantly increased risk for concomitant articulation and phonological speech disorders (Cleland et al., 2010; Rapin et al., 2009; Shriberg et al., 2011). This research stems from a diverse representation of professional backgrounds, such as allied health, psychology, and education. As a result, differences in terminology exist, with selected terms often not well defined within any given article. Without clear definitions, it is difficult to draw comparisons between studies. To improve clarity, the following terms are detailed below: articulation, phonology, motor speech disorders, and prosody.

Articulation refers to the motor movements needed to produce speech sounds (Dodd, 2005; Stoel-Gammon & Bernhardt, 2013). Articulation errors are often based in inaccurate motor learning, an impaired ability to execute the correct motor movements for the phonetic production of the sound. Articulation errors have been reported in approximately one third of high functioning children with ASD, with particular difficulties producing sibilants (e.g., /s/) and rhotics (e.g., /r/) (Shriberg et al., 2011; Shriberg et al., 2001). These errors appear to be persistent, continuing into adolescence and adulthood (Shriberg et al., 2001).

Phonology is a linguistic term referring to the sound patterns and contrasts of a language (Dodd, 2005). Phonological error patterns result in the systematic restructuring or simplification of the adult target of a word, including substitutions, omissions and additions (Vihman, 1996). Phonological error patterns can be categorized into whole-word processes and segment change processes (Ingram, 1989; Vihman, 1996). Cleland et al. (2010) reported a phonological speech delay in 12% of the high functioning children with ASD in their study, with a total of 41% of participants producing some speech errors. Rapin et al. (2009) also reported phonological speech impairments in 24% of their participants. Unfortunately, no description of the types of errors produced by these children was provided and therefore it is impossible to determine if they were actually articulation errors or were indeed phonological in nature.

Motor speech disorders result in difficulties planning, programming, sequencing, and/or executing motor movements for speech. Motor speech disorders are caused by impairments in the central and/or peripheral nervous system. Motor speech disorders include dysarthria, childhood apraxia of speech (CAS) and, according to the Shriberg et al. (2010) Speech Disorders Classification System, motor speech disorders—not otherwise specified (MSD-NOS). Dysarthria is defined as impaired speech production due to difficulties in the muscular control of the speech mechanism (Duffy, 2000). Dysarthria is an umbrella term encompassing a number of subtypes. The prevalence of dysarthria in the general pediatric population or in the ASD population is not known. CAS is a disorder of speech motor planning and programming. Practicing SLPs are reported to believe that one out of every six children with ASD attending speech-language pathology has CAS (Dawson, 2010) although this is not supported by the few studies completed to date (Shriberg et al., 2011; Velleman et al., 2010).

Prosody refers to the suprasegmental features of speech, which includes the use of stress, pitch, rate, intonation, and loudness (O'Connor, 2012; Peppé, McCann, Gibbon, O'Hare, & Rutherford, 2007). These features enhance communication by adding grammatical, pragmatic, and affective meaning to linguistic information (Diehl, Bennetto, Watson, Gunlogson, & McDonough, 2008). Prosodic deficits have been reported in high functioning children with ASD (McCann & Peppé, 2003), although the nature and etiology of these deficits remain unclear. More specifically, deficits with phrasing, stress, inappropriate loudness and inappropriate pitch have been reported (Paul et al., 2005; Peppé, Cleland, Gibbon, O'Hare, & Castilla, 2011; Peppé et al., 2007; Shriberg et al., 2011; Shriberg et al., 2001).

The limited research documenting the speech of children with ASD has focused on the speech of high functioning, verbal school-aged children. To date, little is known about the prevalence and nature of SSDs in younger and/or less verbal children with ASD. From the scant research reporting SSD in verbal children with ASD, higher rates of articulation, phonology, and prosody deficits have been noted. It is currently unclear whether there are higher rates of SSDs in the less verbal population of children with ASD.

The presence of concomitant SSDs in children with ASD is of important theoretical and clinical interest. Three explanations for the presence of SSDs in children with ASD are postulated, which include: (a) co-occurrence with no causal relationship, (b) a common cause, or (c) one is the causal pathway to the other. If there is no relationship between SSD and ASD, the same proportion of children with ASD may be expected to have an SSD as would be expected in the general population. However, if the incidence of SSD associated with ASD is higher than the occurrence in the general population, as has been reported in a number of studies (Cleland et al., 2010; Rapin et al., 2009; Shriberg et al., 2011), such an association is worth exploring further.

Summary

Children with ASD represent a heterogeneous population and can present with a number of complex communication disorders. The challenge for SLPs is to achieve the best possible outcomes for all children with ASD. Although traditional language and behavioral approaches have been reported to assist a number of children with ASD (Tager-Flusberg et al., 2005), some children identified with SSDs may require speech treatment approaches to be incorporated into their comprehensive management plan. SLPs need to be informed coherently and consistently as to how best to assess the speech of children with ASD so that any co-occurring SSDs can be diagnosed and the appropriate evidence-based intervention selected.

This article presents a systematic review designed to provide clinical and research guidelines for the speech assessment of children with ASD. This review follows the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Moher, Liberati, Tetzlaff, Altman, & Group, 2009). These guidelines, and in particular the PRISMA flow diagram, were used to improve the transparency and quality of reporting. This systematic review aims to:

1. provide a summary and evaluation of speech assessments described in the literature that have been used with children with ASD and
2. recommend core components that SLPs should use for an evidence-based speech assessment of children with ASD.

Part A: Systematic Review

Method

Systematic Search

Identification

A systematic search was conducted to determine the research evidence dedicated to the assessment of speech development in children with autism. Peer-reviewed journal articles were identified using the following electronic databases: Cumulative Index to Nursing and Allied Health Literature (CINAHL), Education Resources Information Center (ERIC), Linguistic and Language Behavior Abstracts (LLBA), MEDLINE, PsychINFO, Scopus, speechBITE and Web of Science. Key words used were: "autis*" or "pervasive developmental disorder" AND "child*" or "child develop*" or "infant" or "toddler" or "adolescen*" AND "speech" or "speech disorder" or "articulat*" or "articulation disorder" or "phonology" or "phonetics" or "prosody" or "apraxia" or "dyspraxia" or "childhood apraxia of speech" or "dysarthria" or "vocaliz*." Specific search terms varied depending on the search engine's dictionary of associated terms. For reliability purposes, a second independent speech pathology researcher replicated the MEDLINE search with 100% of search results matching.

Screening

All references were exported to Endnote X5 (Thomson Reuters, 2011) where duplicates were removed. Of the 2,687 imported articles, 1,203 duplicates were removed, leaving 1,484 for further analysis. Due to the broad nature of the initial search, references were further filtered according to title, abstract, and keywords. Articles were excluded from this review if they: (a) did not include children with ASD, (b) did not focus, at least in part, on the speech of children with ASD, or (c) focused on augmentative or alternative communication, such as the use of speech generating devices. Of the 1,484 articles screened, 1,378 articles were excluded based on these criteria. A randomly selected 20% sample of the 1,484 articles was screened by the independent rater. There was 94.6% agreement on inclusion and exclusion of these articles. Following this initial search, the reference lists of the retrieved articles were hand-searched for additional articles that met the inclusion criteria. Additionally, authors and key words were searched again in Google Scholar to ensure all relevant articles were uncovered. An additional 10 articles were assessed for eligibility as a result of these secondary searches.

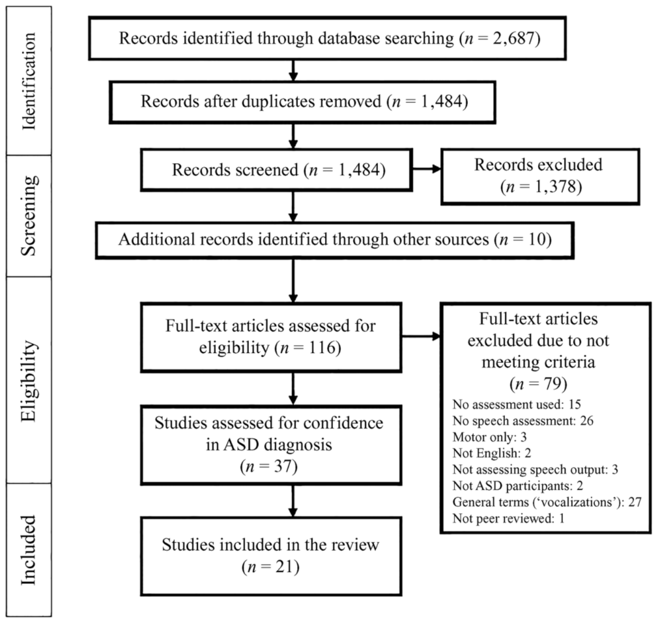
Eligibility

Inclusion criteria. The full articles for the selected 116 references were obtained and assessed according to the following inclusion criteria: (a) peer-reviewed articles published between 1990 and December 2014; (b) written in English; (c) at least one child with ASD assessed; (d) either oral motor skills or at least one aspect of speech development including articulation, phonology, or prosody assessed and reported; (e) assessed the speech output of children with ASD and not just speech perception; and (f) provided some descriptive detail regarding the speech characteristic(s) that were assessed (e.g., not referring to speech in general terms only, such as “vocalizations,” “verbalizations,” “babble,” or “”vocal stereotypy”). Articles were included if they reported using informal or unpublished speech assessment measures if they otherwise met the inclusionary criteria. Thirty-seven articles met the inclusion criteria.

Confidence in ASD diagnosis. Two 5-point scales were applied to each article to rate confidence in the ASD diagnosis of the participants and were adapted from the Core Assessment Battery, as outlined by Ozonoff et al. (2005), and the Scientific Merit Rating Scale (SMRS), an initiative of the National Autism Association National Standards Project (National Autism Center, 2009). The adapted scale from the Core Assessment Battery outlines five key components required in a diagnostic assessment of ASD.

Although the SMRS was developed to evaluate the experimental rigor of treatment studies, the Participant Ascertainment dimension of this scale is particularly relevant to the current review. It refers to the diagnostic methods used to determine eligibility for participant inclusion in the studies. A rating of 5 on each scale indicated confidence in the reported ASD diagnosis. These scores were then added to form one accumulated confidence score out of 10. All articles with an accumulated score of 4 or less were excluded from further review, resulting in 16 articles being excluded (Figure 1). One study was difficult to rate. Shriberg et al. (2011) recruited 46 children previously diagnosed with ASD. Following a thorough diagnostic assessment battery, including all five key assessment components outlined by Ozonoff et al. (2005), 17 participants (37%) were recategorized as

Figure 1. Flow diagram of study selection (adapted from Preferred Reporting Items for Systematic Reviews and Meta-Analyses; Moher et al., 2009).



borderline ASD. These participants met DSM-IV (American Psychiatric Association, 2000) clinical criteria for ASD based on history and presentation but did not meet the threshold on one or two standardized assessment tools. As these participants met clinical criteria for ASD, this article was included in the systematic review and the appropriate score on the SMRS was given. Intrareliability was conducted by the first author. Interrater reliability was completed by an independent postgraduate SLP rater who was trained in both rating scales prior to scoring all articles that met inclusion criteria. Intra- and interrater agreement was 100% for excluded articles. Intrareliability for the remaining 21 articles was 100% on the Core Assessment Battery and 95.24% using the SMRS. Interrater reliability was 90.48% using the Core Assessment Battery and 95.24% using the SMRS. The results are reported in Table 1.

Analysis

The review identified 21 articles that met the inclusion criteria, search criteria and confidence in ASD diagnosis. All articles were reviewed by the first author and an independent rater and were coded according to: (a) the language developmental stage of the participants, (b) the speech domain that was assessed, (c) the speech assessment method used, and (d) the speech characteristics that were described.

Developmental Language Level

Articles were categorized according to whether participants were determined to be at the prelinguistic communication stage or using words. Participants were defined as

Table 1. Rating of confidence in diagnosis of autism spectrum disorder using two 5-point scales.

Reference	Core Assessment Battery	SMRS	Total score	Included
Bellon-Harn, Harn, & Watson (2007)	1	3	4	✗
Beltrame et al. (2011)	0	0	0	✗
Bonneh et al. (2008)	0	3	3	✗
Cleland et al. (2010)	2	3	5	✗
Demouy et al. (2011)	4	5	9	✗
El Mogharbel, Sommer, Deutsch, Wenglorz, & Laufs (2005)	0	0	0	✗
Fine, Bartolucci, Ginsberg, & Szatmari (1991)	1	2	3	✗
Gernsbacher, Sauer, Geye, Schweigert, & Goldsmith (2008)	0	1	1	✗
Grossman et al. (2010)	4	3	7	✗
Hailpern, Harris, La Botz, Birman, & Karahalios (2012)	0	0	0	✗
Iverson & Wozniak (2007)	0	0	0	✗
Kjelgaard & Tager-Flusberg (2001)	4	3	7	✓
Local & Wootton (1995)	1	2	3	✗
McCann et al. (2007)	1	3	4	✗
McCleery et al. (2006)	2	3	5	✓
Nadig & Shaw (2012)	4	3	7	✓
Oller et al. (2010)	0	1	1	✗
Osterling et al. (2002)	3	3	6	✓
Page & Boucher (1998)	1	3	4	✗
Paul et al. (2005)	5	3	8	✓
Paul et al. (2011)	0	0	0	✗
Peppé et al. (2006)	3	3	6	✓
Peppé et al. (2007)	3	3	6	✓
Peppé et al. (2011)	3	3	6	✓
Plumb & Wetherby (2013)	4	5	9	✓
Rapin et al. (2009)	4	3	7	✓
Riches et al. (2011)	4	3	7	✓
Schoen et al. (2011)	4	3	7	✓
Sharda et al. (2010)	1	3	4	✗
Sheinkopf et al. (2000)	3	3	6	✓
Shriberg et al. (2001)	5	3	8	✓
Shriberg et al. (2011)	5	3	8	✓
Van Santen et al. (2010)	3	3	6	✓
Wan et al. (2011)	2	3	5	✓
Werner & Dawson (2005)	3	3	6	✓
Wolk & Edwards (1993)	0	0	0	✗
Wolk & Giesen (2000)	0	2	2	✗

Note. Core Assessment Battery (Ozonoff et al., 2005); Scientific Merit Rating Scale (SMRS; National Autism Center, 2009).

being first word users when they used at least ten intelligible words spontaneously and symbolically (Stoel-Gammon, 1989).

Speech Domain

Articles were separated according to whether segmental or suprasegmental aspects of speech output were assessed. Segmental speech includes phonetics and phonology and typically refers to the use of vowels and consonants. Suprasegmental speech refers to the aspects that are above the level of the single phoneme and includes the prosodic elements of stress, pitch, rate, intonation, rhythm, and loudness.

Speech Assessment Methods

Categories included using conversational speech samples, object or picture naming tasks, and speech imitation tasks. A number of the studies used multiple assessment procedures and were therefore included in a number of categories.

Speech Characteristics

All articles were coded according to the aspects of speech that were described. These characteristics included phonemic repertoires, syllable shapes, intelligibility of speech, phonological processes, and prosodic features.

Interrater reliability, conducted by the first author, was 100% for categorizing all 21 articles according to the participants' language level. Intrarater reliability, conducted by an independent postgraduate SLP rater, was 95.23%. Intrarater reliability was 100% for categorizing the 15 articles with verbal participants according to the speech domain assessed. Interrater reliability was 93.33%.

Results

The review identified 21 articles that met the inclusion criteria. A four-phase flow diagram adapted from PRISMA guidelines (Moher et al., 2009) outlines the study selection process (Figure 1). Of the 21 articles that met the inclusion criteria, seven included participants at a prelinguistic level of language development and 15 assessed the speech of verbal children. Schoen et al. (2011) included both prelinguistic and verbal participants, and therefore this article is included in both categories. As a result, the total number of articles included in Tables 2 and 3 is 22 rather than 21. The participants included in the McCleery et al. (2006) article produced 0–26 single words, which would also place these participants into both the prelinguistic and verbal groups. After correspondence with the first author, it became clear that these participants produced fewer words during direct observation than on parent report and therefore can all be included in the prelinguistic group for the purposes of this review (J. McCleery, email communication, October 4, 2015).

Overall, 747 participants were included in the 21 articles. All of these participants were diagnosed with ASD prior to 2013 and therefore often received labels of autistic disorder (AD) or pervasive developmental disorder—not otherwise specified (PDD-NOS). Various researchers have questioned the validity of this categorical approach and have argued that AD, PDD-NOS., and Asperger's disorder are not independent groups. Instead, these diagnoses occur along a continuum and providing levels of functioning is more informative than categorical information (Volkmar & McPartland, 2014). In keeping with the current diagnostic standards in the DSM-V (American Psychiatric Association, 2013), the subcategory labels are not reported in this review. In order to compare participants across studies we have provided intelligence and language function information where it was reported by the authors (Table 2). Unfortunately, due to the heterogeneity of assessment tools used and the lack of reporting of standard deviations, score ranges, or individual data points, it is difficult to compare the intellectual function or language ability of participants across articles.

Prelinguistic Level

Prelinguistic language was defined as producing fewer than 10 recognizable words (Stoel-Gammon, 1989; Thal, 1995). Of the seven articles that assessed the speech

Table 2. Participant information and speech assessment details from studies with prelinguistic participants with autism spectrum disorders (ASD).

Reference	Research design	*n*	Mean age (mo.)	Cognitive level	Language level	Speech data collection method	Speech analyses
McCleery et al. (2006)	Prospective case control study	14	40.00	Mean mental age[a]: 18.2 mo.	Receptive vocabulary[b]: age equivalent 12 mo. Expressive vocabulary[b]: mean no. words, 7 (0–26)	Speech imitation task	Consonant repertoire
Osterling et al. (2002)	Retrospective case control study	20	12.00	Full-scale IQ*: 64.00 (26.44)	Not reported	Connected speech sample	Frequency of vocalizations
Plumb & Wetherby (2013)	Retrospective case control study	50	21.29	Nonverbal DQ[c]: 76.00 (25.81); Verbal DQ[c]: 68.00 (31.11)	Core language[d]: 73.88 (12.62)	Connected speech sample	Categorized vocalizations
Schoen et al. (2011)	Prospective case control study	21	28.33**	Did not report standard scores[c]	Expressive vocabulary[d]: mean no. words, 3.52 (0–8)	Connected speech sample	Consonant repertoire and syllable shapes
Sheinkopf et al. (2000)	Prospective case control study	15	44.67	Mean mental age[e]: 22.13 (5.07)	Expressive language age[f]: 14.18 (2.70)	Connected speech sample	Categorized vocalizations
Wan et al. (2011)	Single subject multiple-baseline design	6	79.00	Did not report standard scores[c]	Receptive language age[c]: > 22 months; Expressive vocabulary[g]: no words	Single-word naming task	Transcription of treatment probe
Werner & Dawson (2005)	Retrospective case control study	36	42.96	Full-scale IQ[c]: 57.33 (20.60); Nonverbal IQ[c]: 64.01 (19.83); Verbal IQ[c]: 50.49 (24.07)	Did not complete standardized language assessment	Connected speech sample	Frequency of simple and complex babble

Note. Standard scores (standard deviations in brackets); standard scores and standard deviations combined for studies that reported separate diagnostic groups. IQ = intelligence quotient; DQ = developmental quotient

*Reports from previous assessments. Data not for all participants
**Mean age for all participants with ASD, including verbal participants
[a]Bayley Scales for Infant Development (Bayley, 1969). [b]MacArthur-Bates Communicative Development Inventory (CDI; Fenson et al., 1993). [c]Mullen scales of early learning (MSEL; Mullen, 1995). [d]Communication and Symbolic Behavior Scales (CSBS; Wetherby & Prizant, 2002). [e]Merrill-Palmer Scales of Mental Tests (Stutsmart, 1948). [f]Reynell Developmental Language Scales (Reynell & Gruber, 1990). [g]Expressive Vocabulary Test (EVT; Williams, 2007).

Table 3. Participant information and speech assessment details from studies with verbal participants with autism spectrum disorders (ASD).

Reference	Research design	*N*	Mean age (mo.)	Cognitive level	Language level	Speech data collection method	Speech analyses	Speech domain
Cleland et al. (2010)	Prospective cohort study	69	114.52	Nonverbal IQ[a]: 102.96 (14.39)	Receptive vocabulary[b]: 92.89 (16.49); Receptive language[c]: 93.89 (17.17); Expressive language[d]: 84.12 (16.30)	Single-word naming task	Standard scores	Segmental
Demouy et al. (2011)	Prospective case control study	22	117.44	Nonverbal IQ[e]: 76.91 (13.99)	Did not report standard scores[f]	Speech imitation task	Acoustic analysis	Suprasegmental
Grossman et al. (2010)	Prospective case control study	16	148.00	Full-scale IQ[g]: 106.70 (10.60); Nonverbal IQ[g]: 109.60 (19.10)	Receptive vocabulary[h]: 107.00 (15.40)	Closed-sentence task	Acoustic analysis	Suprasegmental
Kjelgaard & Tager-Flusberg (2001)	Prospective cohort study	89	88.07	Full-scale IQ[i]: 68.49*(24.38)	Core language[d]: 72.32* (17.71); Expressive vocabulary[j]: 68.99* (23.62); Receptive vocabulary[h]: 70.37*(22.68)	Single-word naming task; speech imitation task	Standard Scores	Segmental
Nadig & Shaw (2012)	Prospective case control study	15	132.00	Nonverbal IQ[e]: 105.00 (15.00)	Core language[d]: 109.00 (13.00)	Connected speech sample	Acoustic analysis	Suprasegmental
Paul et al. (2005)	Prospective case control study	27	201.60	Nonverbal IQ[e]: 95.20 (25.60); Verbal IQ[e]: 103.90 (23.80)	Receptive language[d]: 98.60 (21.40); Expressive language[d]: 94.50 (18.10)	Production of words and sentences	Percentage correct	Suprasegmental
Peppé et al. (2006, 2007)	Prospective case control study	31	118.00	Reports of normal nonverbal intelligence	Receptive vocabulary[b]: 81.40 (16.20); Receptive language[c]: 79.60 (17.90); Expressive language[c]: 69.80* (8.50)	Production of words and sentences	Raw scores	Suprasegmental
Peppé et al. (2011)	Prospective case control study	71	114.90	Reports of normal nonverbal intelligence	Expressive language[d]: 84.53	Production of words and sentences	Raw scores	Suprasegmental
Rapin et al. (2009)	Prospective cohort study	62	103.20	Did not report group mean	Did not report group mean	Single-word naming task	Standard scores	Segmental

(*table continues*)

Table 3. *(Continued).*

Reference	Research design	*N*	Mean age (mo.)	Cognitive level	Language level	Speech data collection method	Speech analyses	Speech domain
Riches et al. (2011)	Prospective case control study	16	176.00	Nonverbal IQ[e]: > 80	Some CELF-3 subtests only	Speech imitation task	Coding of phonemes and syllable shapes	Segmental
Schoen et al. (2011)	Prospective case control study	9	28.33**	Did not report standard scores[k]	Expressive vocabulary[l]: mean no. words, 15.89 (6.47)	Connected speech sample	Consonant repertoire and syllable shapes	Segmental
Shriberg et al. (2001)	Historical case control study	30	253.80	Nonverbal IQ[e]: 89.05 (26.77)	Core language[m]: 89.55 (21.51)	Connected speech sample	Narrow phonetic transcription, prosody-voice coding, acoustic analysis	Segmental; Suprasegmental
Shriberg et al. (2011)	Historical case control study	46	69.90	Nonverbal IQ[e]: 102.80 (16.10)	Core language[d]: 97.80 (17.80)	Connected speech sample	Narrow phonetic transcription, prosody-voice coding, acoustic analysis	Segmental; Suprasegmental
Van Santen et al. (2010)	Case control study	26	78.84	Nonverbal IQ[e]: 117.63 (11.48)	MLU: < 3	Production of words and sentences	Raw scores, acoustic analysis	Suprasegmental

Note. Standard scores (standard deviations in brackets); standard scores and standard deviations combined for studies that reported separate diagnostic groups; IQ = intelligence quotient; CELF-3 = Clinical Evaluation of Language Fundamentals–Third Edition; MLU = mean length of utterance.

*not all participants completed this assessment
**mean age for all participants with ASD, including prelinguistic participants
[a]Raven's Progressive Matrices (RPM; Raven, Court, & Raven, 1986). [b]British Picture Vocabulary Scale–Second Edition (BPVS-II; Dunn, Dunn, Whetton, & Burley, 1998). [c]Test for Reception of Grammar–Second Edition (TROG-2; Bishop, 2003). [d]Clinical Evaluation of Language Fundamentals: CELF-3 (Semel, Wiig, & Secord, 2000), CELF-4 (Semel, Wiig, & Secord, 2003), CELF-P (Wiig, Secord, & Semel, 1992). [e]Weschler Intelligence Scales: WISC-3 (Wechsler, 1992), WISC-IV (Wechsler, 2003), WPPSI (Wechsler, 2002), WAIS (Wechsler, 1997). [f]Evaluation du Language Oral (ELO; Khomsi, 2001). [g]Kaufman Brief Intelligence Test (Kaufman & Kaufman, 2004). [h]Peabody Picture Vocabulary Test—Revised (PPVT-R; Dunn & Dunn, 1981). [i]Differential Abilities Scales (DAS; Elliott, 1990). [j]Expressive Vocabulary Test (EVT; Williams, 2007). [k]Mullen Scales of Early Learning (MSEL; Mullen, 1995). [l]Communication and Symbolic Behavior Scales (CSBS; Wetherby & Prizant, 2002). [m]Test of Language Competence (TLC; Wiig & Secord, 1989).

of children with ASD at a prelinguistic language level, three were retrospective case-control studies (Osterling, Dawson, & Munson, 2002; Plumb & Wetherby, 2013; Werner & Dawson, 2005), three prospective case-control studies (McCleery, Tully, Slevc, & Schreibman, 2006; Schoen et al., 2011; Sheinkopf, Mundy, Oller, & Steffens, 2000) and a single treatment article that adopted a single-subject multiple baseline design (Wan et al., 2011). Overall, there were 162 participants across the seven articles, with ages ranging from 12 months to 79 months at the time of data collection (Table 2).

Assessment Methods

Connected speech samples. Although connected speech samples were used by the majority of authors, the methods and speech analyses reported vary greatly (Osterling et al., 2002; Plumb & Wetherby, 2013; Schoen et al., 2011; Sheinkopf et al., 2000; Werner & Dawson, 2005). Number of utterances or the length of the sample was reported in only three articles. Schoen et al. (2011) analyzed a 15-minute connected speech sample from the Communication and Symbolic Behavior Scales (CSBS) Behavior Sample. The first 50 utterances were transcribed using broad phonemic transcription. These authors reported that 56% of the participants with ASD did not produce 50 utterances, and therefore all speech-like utterances were transcribed for these participants. Although it could be assumed that the prelinguistic participants did not produce 50 utterances, this was not clearly stated. Sheinkopf et al. (2000) collected a connected speech sample taken during the Early Social and Communicative Scales (ESCS) and reported a mean of 117.73 utterances, although a standard deviation of 85.91 represents wide variability. Plumb and Wetherby (2013) analyzed all vocalizations produced during the CSBS Behavior Sample and reported an average length of 18 min and 10 s. These authors did not report the number of utterances; however, they did analyze the data according to the proportion of transcribable vocalizations.

The speech characteristics reported included phonetic repertoires and syllable shapes. Two studies reported the level of syllable shapes produced (Plumb & Wetherby, 2013; Schoen et al., 2011) and a further two studies reported the frequency of simple or complex babbling (Sheinkopf et al., 2000; Werner & Dawson, 2005), although these terms were not well defined. One article collected data on "babbling," although due to difficulties with reliability the authors collapsed this information with vocalizations and did not report further on babbling (Osterling et al., 2002). Only Schoen et al. (2011) reported the children's consonant inventories from the collected connected speech samples.

Speech imitation. McCleery et al. (2006) used a syllable imitation task to ascertain participants' consonant repertoires. These participants were required to imitate a consonant-vowel (CV) syllable, with the vowel being a neutral schwa. Motivating toys were used as reinforcers. All imitated speech and spontaneous speech were transcribed and consonant repertoires were reported.

Single-word naming task. Wan et al. (2011) transcribed the probe assessment data collected during a treatment study using Auditory Motor-Mapping Training (AMMT). During probe tasks, participants named 15 trained two-syllable words and 15 untrained two-syllable words. Each two-syllable target was broken down into two CV syllables. Each CV syllable was scored separately. Close approximations were considered correct if two of the three consonants' dimension of voicing, place or manner were correct. The percentage of correct CV syllable productions was reported. Consonant repertoires or percent consonants correct were not reported.

Verbal Participants

Participants were categorized as being verbal if they produced 10 or more recognizable words (Stoel-Gammon, 1989; Thal, 1995). It is important to make a clear distinction between children who were using words and were termed *verbal* and those who were prelinguistic but vocalizing. In the ASD literature, the terms verbalizations and vocalizations are used interchangeably and often refer to the sounds made by children prior to the use of first words. Fifteen articles were included in this category (Table 3). These articles were further categorized into those that assessed segmental aspects of speech and those that assessed suprasegmental speech. Two articles assessed both segmental and surprasegmental speech and were included in both categories (Shriberg et al., 2011; Shriberg et al., 2001).

Segmental Speech

A total of seven articles assessed segmental aspects of speech. Of these articles, three were prospective cohort studies (Cleland et al., 2010; Kjelgaard & Tager-Flusberg, 2001; Rapin et al., 2009) and four were prospective case-control studies (Riches, Loucas, Baird, Charman, & Simonoff, 2011; Schoen et al., 2011; Shriberg et al., 2011; Shriberg et al., 2001; Table 3). There were 321 participants across the seven articles, with ages ranging from 18 months to 184 months.

Assessment Methods

Connected speech samples. Connected speech samples were used to assess segmental components of speech in three studies (Schoen et al., 2011; Shriberg et al., 2011; Shriberg et al., 2001). The assessment method used by Shriberg et al. (2011) mirrored that used in their earlier study (Shriberg et al., 2001). Both studies used a connected speech sample taken during the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DeLavore, & Risi, 2000) standardized interview. An average of 48.9 utterances were taken for the participants with Asperger's disorder, and an average of 35 utterances were collected for the participants with high functioning autism in the 2001 study (Shriberg et al., 2001). The average number of utterances sampled was not reported in the 2011 study. The researchers completed narrow phonetic transcription in both studies. Shriberg et al. (2001) reported the frequency and types of consonant and vowel errors. In the later 2011 study, acoustic analyses were completed to classify subtypes of speech sound disorders. Schoen et al. (2011) collected 15-minute connected speech samples from the CSBS. The first 50 utterances were to be transcribed; however, more than half of the participants did not produce 50 utterances. Prelinguistic and verbal participants were included in this study, and the average number of utterances produced by the verbal participants is unclear. Broad phonemic transcription was completed, and the consonant inventories and levels of syllable shapes were reported.

Single-word naming task. Standardized single-word naming tasks were used in three studies. Kjelgaard and Tager-Flusberg (2001) used the Goldman-Fristoe Test of Articulation (Goldman & Fristoe, 1986) and Cleland and colleagues (2010) used the second edition of the same test (Goldman & Fristoe, 2000). Rapin et al. (2009) used the Photo Articulation Test (Pendergast, Dickey, Selmar, & Soder, 1984). All authors reported standard scores; however, only Cleland et al. (2010) included additional phonetic and phonological analyses of all speech error types.

Speech imitation. Two authors used speech imitation tasks to assess segmental aspects of speech. Nonword repetition tasks were used in both studies (Kjelgaard & Tager-Flusberg, 2001; Riches et al., 2011). The speech characteristics reported varied. Riches et al. (2011) used broad phonemic transcription and syllable coding schemes and reported the number of consonant substitutions, changes to syllable structure and phonemic errors produced. Kjelgaard and Tager-Flusberg (2001) reported standard scores only.

Suprasegmental Speech

All 10 studies that assessed suprasegmental aspects of speech were prospective case-control studies (Demouy et al., 2011; Grossman, Bemis, Plesa Skwerer, & Tager-Flusberg, 2010; Nadig & Shaw, 2012; Paul et al., 2005; Peppé et al., 2011; Peppé, McCann, Gibbon, O'Hare, & Rutherford, 2006; Peppé et al., 2007; Shriberg et al., 2011; Shriberg et al., 2001; Van Santen, Prud'Hommeaux, Black, & Mitchell, 2010; Table 3). A total of 284 participants with ASD, with ages ranging from 48–588 months, were included in these studies. Two articles (Peppé et al., 2006, 2007) reported on the same 31 participants so these participants were only counted once.

Assessment Methods

Connected speech samples. Shriberg et al. (2001) and Shriberg et al. (2011) collected connected speech samples from the ADOS and completed perceptual prosody voice coding using the Prosody-Voice Screening Profile (Shriberg, Kwiatkowski, & Rasmussen, 1990). This tool provides perceptual coding information on phrasing, rate, stress, loudness, pitch, voice, quality, and resonance. Acoustic analyses of loudness, pitch and stress were also completed in the later study (Shriberg et al., 2011). Nadig and Shaw (2012) also assessed the suprasegmental aspects of speech from connected speech samples. Acoustic analyses of pitch range, mean pitch and speech rate were taken from 10–13 s of uninterrupted speech segments. Perceptual ratings of pitch and speech rate were also reported.

Production of words and sentences. A number of articles have been published from the same team of researchers using the Profiling Elements of Prosodic Speech in Children (PEPS-C; Peppé & McCann, 2003) to assess the suprasegmental speech of children with ASD (Peppé et al., 2011; Peppé et al., 2006; Peppé et al., 2007). This is a standardized assessment tool that assesses a child's ability to perceive and produce pitch variation, contrastive stress and pausing. Auditory-perceptual scores from the PEPS-C were reported in each study. Van Santen et al. (2010) used a modified contrastive stress task from the PEPS-C, in addition to a lexical stress task (Paul et al., 2005) and an emphatic stress task (Shriberg et al., 2001). Van Santen et al. reported auditory-perceptual scores in addition to acoustic analyses of pitch, amplitude and duration. The ability to produce lexical stress was also assessed by Grossman et al. (2010). These authors used a cloze-sentence task to elicit different lexical stress patterns and reported acoustic analyses of pitch, intensity and duration. Paul et al. (2005) used a nonstandardized prosody protocol. Lexical and pragmatic functions of stress, intonation, and phrasing were assessed and the percentage of correct productions was reported. Demouy et al. (2011) used a sentence imitation task to assess the production of intonation patterns. Acoustic analyses of duration and pitch intonation patterns were reported.

Discussion

The purpose of this systematic review was to determine the core components of an evidence-based speech assessment for children with ASD to guide clinical and research practices. A total of 21 studies met inclusion criteria. The heterogeneity of the participants included in these studies, in addition to variability in terminology and reporting standards, made it challenging to identify trends across the 21 studies.

Prelinguistic Level

Seven studies reported the assessment of speech of children with ASD at the prelinguistic level of development. The majority of these studies used a connected speech sample to analyze the speech of children with ASD. Research assessing prelinguistic speech in typically developing children and children with delayed expressive language has routinely used connected speech samples and standard data collection methods (Oller, 1986; Paul, Norbury, & Ebrary, 2012; Rice et al., 2010; Stark, 1980; Stoel-Gammon, 1989). Stoel-Gammon (1987) argued that the use of a connected speech sample at this stage of language development provides a more valid sample of the child's phonological system when compared with a single-word naming task. Further, it is recognized that a sample of at least 50 speech-like utterances during a parent–child interaction provides adequate data for speech analysis (Mitchell, 1997; Morris, 2010; Nathani et al., 2006; Paul & Jennings, 1992; Stoel-Gammon, 1989).

Of the five studies that used a connected speech sample to obtain speech data, only two reported number of utterances (Schoen et al., 2011; Sheinkopf et al., 2000), and neither study specified that 50 utterances were sampled for each participant. Although most studies used an appropriate data collection method with prelinguistic participants, no study collected a large enough sample (Morris, 2010; Stoel-Gammon, 1989) to be representative of the child's speech.

A standard protocol for reporting the prelinguistic speech development of children with ASD is needed. Only two studies in the current review analyzed prelinguistic speech according to phonetic and syllabic level of complexity (Plumb & Wetherby, 2013; Schoen et al., 2011). Both studies used the Syllable Structure Level model (SSL; Olswang, Stoel-Gammon, Coggins, & Carpenter, 1987). The SSL model provides general information on the level of phonetic complexity, such as vowel and consonant production and use of syllables. It does not provide any detailed description of the child's phonetic repertoire or use of stress.

A disparity exists between the body of research assessing prelinguistic speech in typically developing children and children with delayed expressive language and the current standards used to report the prelinguistic speech of children with ASD. It cannot be explained by the compliance challenges associated with assessing children with ASD, as the use of connected speech samples provides more opportunity to follow the child's lead and capture any self-directed or context-specific language than formal standardized assessments (Charman et al., 2003; Paul et al., 2008). Differences in speech analysis and reporting protocols are more likely a result of the varied professional backgrounds of researchers interested in this area and the contrasting definitions for terms such as speech, vocalizations, verbalizations, and babbling that this brings. To move forward, it would be beneficial for researchers to use a common vocabulary for speech sampling and analysis (e.g., Mitchell, 1997).

Verbal Participants

Fifteen studies assessed the speech of children who produced 10 or more words and were classified as being verbal. Although a range of assessment methods were used across the studies, including single-word naming tasks, connected speech samples, and speech imitation, only one study employed more than one speech sampling method (Kjelgaard & Tager-Flusberg, 2001). Most published speech assessment guidelines recommend using multiple speech sampling methods to gain a complete picture of a child's speech production (e.g., Bernhardt & Holdgrafer, 2001; McLeod & Baker, 2014; Morrison & Shriberg, 1992; Stoel-Gammon, 1987; Stoel-Gammon & Williams, 2013). Morrison and Shriberg (1992), for example, reported differences in sound production during single-word naming tasks versus connected speech and emphasized the importance of using both methods to obtain a more accurate sample of a child's speech. No study in the current review used both single-word naming tasks and connected speech sampling when assessing the speech of children with ASD. There are significant limitations in using only one assessment method, and questions arise regarding the validity of the speech sample collected.

Standard reporting protocols are also needed in the body of research assessing the speech of verbal children with ASD. Standard scores were reported in all studies that used published single-word naming tasks; however, only one study provided any further speech analysis on the sampled data (Cleland et al., 2010). Although standard scores provide some information on the severity of a child's speech difficulty, they do not provide enough descriptive information for the differential diagnosis of an SSD. The American Speech-Language-Hearing Association (2004b) recommends reporting phoneme repertoires, syllable shapes, error analyses, intelligibility, and severity ratings. No study in this review included all of this information. Only six studies completed any phonetic transcription (Cleland et al., 2010; McCleery et al., 2006; Riches et al., 2011; Schoen et al., 2011; Shriberg et al., 2011; Shriberg et al., 2001), and only consonants were transcribed in two of these studies (Cleland et al., 2010; McCleery et al., 2006). Complete phoneme repertoires were not reported in any study, although one study did provide information regarding consonant repertoires (Schoen et al., 2011). Information regarding syllable shapes was also limited to a few studies (Riches et al., 2011; Schoen et al., 2011), as were error analyses (Cleland et al., 2010; Shriberg et al., 2011; Shriberg et al., 2001) and intelligibility (Shriberg et al., 2001).

Differences in the reporting standards in the ASD literature compared with the SSD literature may be the result of a number of factors, including researchers' frame of reference, the research question, and publication restrictions. Researchers in the area of ASD are from diverse professional backgrounds, including psychology, education, and allied health. The professional backgrounds of the researchers will shape their understanding of speech development as distinct from language development, which will in turn shape the assessment methods and reporting protocols they adopt. Additionally, a number of the studies reviewed here did not complete their research with the purpose of assessing speech. The research did not aim to differentially diagnose or treat SSDs and therefore did not include the required speech analyses to do so.

Confidence in ASD Diagnosis

Practice parameters for the assessment of ASD have been outlined by a number of authors (e.g., Filipek et al., 2000; National Autism Center, 2009; Ozonoff et al., 2005). These parameters emphasize the importance of a comprehensive developmental assessment for children identified at risk of ASD, which takes a developmental perspective, includes information from multiple sources, and is multidisciplinary (Filipek et al., 2000; Ozonoff et al., 2005). Children with ASD form a heterogeneous population, and in order to draw comparisons between studies, it is imperative that results from standardized cognitive and language assessment tools be reported for each participant. It is not enough to report diagnostic categories. Seventeen articles were excluded from this systematic review due to lack of confidence in the ASD diagnosis of the participants. One article that was excluded (McCann, Peppé, Gibbon, O'Hare, & Rutherford, 2007) was identified as reporting on the same participants as two included studies (Peppé et al., 2006; Peppé et al., 2007). This highlights the importance of reporting enough information in every article to make the diagnostic process clear to the reader.

What Can We Conclude From the Systematic Review?

The participants, assessment tasks and reporting standards in the studies included in the systematic review varied widely. Although a number of well-designed studies with clear diagnostic criteria have been completed, without standard reporting protocols for speech sampling and analysis, it remains difficult to draw comparisons between studies. Given this heterogeneity, the results do not provide clear clinical or research guidelines for best practice in the speech assessment of children with ASD. This highlights an important challenge.

In the absence of recommendations from the systematic review, it is essential to return to first principles and those of published guidelines for the speech assessment of a more general pediatric population. A narrative review of the pediatric speech literature follows.

Part B: Narrative Review

This narrative review provides a broad overview of recent published recommendations for best practice in the assessment of pediatric SSDs. The purpose of the review is to provide a best-evidence synthesis of the literature, according to the American Speech-Language-Hearing Association (2004a) system for ranking levels of evidence. The highest level of evidence is a meta-analysis (Level 1a) followed by a systematic review (Level 1b) of published random controlled trials. With no published meta-analysis or systematic review, this review encompasses all other levels of evidence inclusive of the most recent large pediatric population studies, differential diagnosis studies and published expert opinions. Two large surveys of practicing SLPs completed by experts in the field have also been included in the review. The focus here is on speech production assessments. It is assumed that appropriate cognitive, language, hearing, and speech perception assessments are completed. For consistency of reporting we have utilized the same framework as the systematic review above by categorizing findings according to the child's linguistic level of development. Children are referred to as prelinguistic if they produce less than 10 recognizable words. Children are defined as verbal if they produce more than 10 recognizable words.

It should be noted that at no stage of speech development does nonspeech oral motor movement correlate to speech accuracy (McCauley, Strand, Lof, Schooling, & Frymark, 2009). Therefore, an oral motor assessment will be the same regardless of a child's speech or linguistic level. The results of the narrative review are provided under the following headings: oral motor assessment, prelinguistic level, and verbal children.

Oral Motor Assessment

An evidence-based speech assessment must include an examination of the oral mechanism (Kent, 2015). A comprehensive assessment of the structures and function of the oral mechanism is crucial to identify underlying structural abnormalities (e.g., cleft palate) impacting speech development and in the differential diagnosis of childhood motor speech disorders. There are a number of published assessment tools (Dodd, Hua, Crosbie, Holm, & Ozanne, 2002; Hayden & Square, 1999; Robbins & Klee, 1987; St. Louis & Ruscello, 2000; Thoonen, Maassen, Wit, Gabreëls, & Schreuder, 1996), although to date there are very few standardized assessments with good evidence of reliability and validity (McCauley & Strand, 2008). Clinicians and researchers may instead use informal methods to complete the oral motor assessment. Regardless of the assessment tool used, there are a number of core components to include; the strength, symmetry, accuracy, speed, and range of movement of the lips, tongue, face, and palate are assessed at rest, during static postures and during nonverbal oral movements (Duffy, 2000; Kent, 2015; Strand, McCauley, Weigand, Stoeckel, & Baas, 2013). Nonverbal tasks can also include sequences of postures (e.g., smile, blow). A number of assessment tools also assess motor speech function, such as the imitation of single phonemes, syllables and sequential/alternating motion rates (SMR, AMR; Thoonen et al., 1996). For the purposes of this review, a clear distinction between an oral motor assessment that assesses the oral mechanism at rest and during nonverbal movement versus speech assessment tasks will be made.

Prelinguistic Level

The understanding several decades ago that the sounds produced in later babbling were similar to those in a child's first words shifted the focus of research onto prelinguistic speech assessments (Stoel-Gammon, 1985; Vihman, Macken, Miller, Simmons, & Miller, 1985). Since this time, the methods for collecting and analyzing speech data have evolved.

Assessment Methods

It is widely recognized that a connected speech sample provides the most representative sample of a child's prelinguistic speech (Oller, 1986; Paul et al., 2012; Rice et al., 2010; Stark, 1980; Stoel-Gammon, 1989). Connected speech samples are typically collected during natural interactions between the child and a familiar caregiver. Prelinguistic vocalizations can be categorized into speech-like sounds, including consonants and vowels, and nonspeech-like sounds, such as squeals, cries and laughing. Prelinguistic speech-like sounds are typically referred to as babbling and are the focus of a speech assessment at this stage of development. There appears to be agreement that a sample of at least 50 speech-like vocalizations taken during a parent–child interaction provides adequate data for speech analysis (Mitchell, 1997; Morris, 2010; Nathani et al., 2006; Paul & Jennings, 1992; Stoel-Gammon, 1989).

Speech analyses taken from connected speech samples alone have limitations. Differences in the number of utterances and phonological complexity will make it difficult to draw comparisons between samples (Stoel-Gammon & Williams, 2013). This also makes it hard to compare samples from the same child over time, which is often used to identify improvements in an individual's speech system. Further, little information will be obtained regarding a child's ability to imitate sounds or words.

To address these limitations, there has been a recent shift toward developing standardized speech assessments appropriate for very young children and/or children at an early stage of language development (Dodd & McIntosh, 2010; Shriberg et al., 2009; Stoel-Gammon & Williams, 2013; Strand & McCauley, in press). Unlike other published speech assessment tasks that aim to assess all phonemes across all positions in words, assessment tasks appropriate for children at this stage of development should focus on early developing consonants and vowels, syllable shapes, and stress patterns. Specifically, this would mean mainly single- or two-syllable words, containing mainly stops and nasals, with stress on the first syllable (Stoel-Gammon, 1998). The development of these tools highlights the need for more information regarding a child's phonetic stimulability and word imitation, even at very early stages of development. Imitation deficits of varying forms have been identified in the autism spectrum population. Motor imitation significantly dominates this body of research and deficits have been reported across a variety of tasks including actions on objects and imitation of body movements (Charman et al., 1997; DeMyer et al., 1972; Rogers, Bennetto, McEvoy, & Pennington, 1996; Stieglitz Ham et al., 2011; Stone, Ousley, & Littleford, 1997; Williams, Whiten, Suddendorf, & Perrett, 2001). Likewise, some authors suggest that speech imitation is challenging for children with ASD (Abrahamsen & Mitchell, 1990; Sigman & Ungerer, 1984); however, given four studies from this systematic review successfully used imitation tasks to elicit speech samples (Demouy et al., 2011; Kjelgaard & Tager-Flusberg, 2001; McCleery et al., 2006; Riches et al., 2011), the idea that speech imitation tasks should not be used is questionable.

Data Analyses

Independent analyses are used to obtain a representative picture of a child's prelinguistic speech ability. Independent analyses focus on the sounds and syllable shapes present in a child's phonological system. This includes phonetic repertoires and analysis of syllable shapes and stress patterns (Morris, 2010; Stoel-Gammon & Williams, 2013). These data provide important information regarding a child's stage of speech development. Speech development in typically developing children follows a sequence of developmental stages from birth to first words, and these stages can be categorized according to the level and complexity of the phonetic and syllabic level of development (Mitchell, 1997; Morris, 2010; Paul, 1995; Paul & Jennings, 1992; Stark, 1980; Stoel-Gammon, 1989).

By 10–12 months of age, typically developing children begin to produce complex syllable strings with varied adult-like intonation known as jargon (Mitchell, 1997; Nathani et al., 2006; Paul, 1995). This stage can coincide with the use of first words and is an important milestone in a child's motor speech development. Therefore, to accurately report on a child's prelinguistic speech development, the child's phonetic and syllabic complexity, in addition to the use of jargon and development of stress, need to be reported.

Standardized reporting measures that have been employed in previous research with typically developing children and children with delayed expressive language include Mean Babbling Level (MBL; Stoel-Gammon, 1989), Syllable Structure Level (SSL; Olswang et al., 1987), and Canonical Babbling Ratio (CBR; Oller, Eilers, Steffens, Lynch, & Urbano, 1994). Although these measures provide information on vowel and consonant production and use of syllables, they fail to capture the importance of stress development. By 10–12 months of age, typically developing children begin to produce complex syllable strings with varied adult-like intonation known as jargon (Mitchell, 1997; Nathani et al., 2006; Paul, 1995). This often coincides with the use of first words. This developmental milestone is included in the Stark Assessment of Early Vocal Development–Revised (SAEVD-R; Nathani et al., 2006) as well as in the final stages of other published stage-based models (e.g., Oller, 1980; Paul, 1995).

Verbal Children

Assessment Methods

The use of a standardized single-word naming assessment is widely recommended (Bleile, 2002; Eadie et al., 2014; Khan, 2002; McLeod & Baker, 2014; Miccio, 2002; Stoeckel et al., 2013; Tyler & Tolbert, 2002) and commonly completed by practicing SLPs (McLeod & Baker, 2014; Skahan, Watson, & Lof, 2007), although there is no consensus on which assessment tools to use. Interestingly, surveys of practicing SLPs report that clinicians tend to select standardized tools that are developed locally (McLeod & Baker, 2014). Although standardized scores allow comparison to normative data, they do not provide adequate descriptive information on a child's phonological system, prosodic development or use of sounds across word positions, word shapes and lengths (Bernhardt & Holdgrafer, 2001; Eisenberg & Hitchcock, 2010). Additionally, very few standardized single-word naming tests contain the recommended 100 words required for an adequate sample (Bernhardt & Holdgrafer, 2001). Some experts even question the usefulness of completing a standardized assessment, preferring to focus on other speech assessment tasks (Bleile, 2002). Researchers and clinicians alike accept that standardized single-word naming tasks have limitations and further assessment tasks are required to differentially diagnose SSDs.

Connected speech samples are also recommended by the American Speech-Language-Hearing Association (2004b) and expert clinicians (Hodson, Scherz, & Strattman, 2002; Miccio, 2002; Shriberg et al., 2011; Stoel-Gammon & Williams, 2013), although due to time constraints, these may not be widely used by practicing SLPs (McLeod & Baker, 2014; Skahan et al., 2007). As previously noted, a minimum of 50 speech-like utterances is required for speech analysis. This sample provides information regarding the child's ability to produce sounds during connected speech, in addition to suprasegmental features of speech, including the use of stress, pitch, rate, intonation and loudness.

Stimulability refers to a child's ability to imitate a new sound immediately following an examiner's model (Miccio, 2002). Stimulability testing provides useful clinical information regarding prognosis and goal determination (Miccio, 2002). It is commonly completed by practicing SLPs (McLeod & Baker, 2014; Skahan et al., 2007) and expert clinicians (Bleile, 2002; Hodson et al., 2002; Khan, 2002; Miccio, 2002) and is recommended by the American Speech-Language-Hearing Association (2004b). However, stimulability testing is not frequently completed in large population research studies, reflecting a possible disparity in the objectives of the clinicians and researchers.

Less frequently recommended or completed is the assessment of polysyllabic words. Polysyllabic words provide information on the production of a variety of word shapes and stress patterns, information that is often not obtained from monosyllabic word tasks (Bernhardt & Holdgrafer, 2001; James, van Doorn, & McLeod, 2008; Murray, McCabe, Heard, & Ballard, 2015). The inclusion of a polysyllable word test may identify phonological speech errors that would not be observed when producing shorter words (James et al., 2008). This is thought to be because the production of polysyllabic words places more pressure on the speech system than monosyllablic words, as more information is perceived, stored and produced (James et al., 2008; Masso, McCabe, & Baker, 2014).

Data Analyses

As previously described, independent and relational data analyses are required to provide a complete picture of the child's speech system. Independent analyses report what a child can do and include phonetic repertoires, word and syllable shapes, in addition to stress patterns.

Relational analyses require comparison to the adult model and encompass measures of accuracy, intelligibility and error patterns. Common methods to measure a child's speech accuracy are the calculation of percentage of consonants correct (PCC) and percentage of vowels correct (PVC; Shriberg et al., 2010; Shriberg & Kwiatkowski, 1982) taken from a 5–10 min connected speech sample. The limitation of PCC is that all consonants are weighted equally, regardless of complexity (Preston, Ramsdell, Oller, Edwards, & Tobin, 2011). Therefore, additional analyses of speech error patterns and intelligibility are required. Speech error patterns are categorized as typical or atypical. As defined in Part A, these error patterns can be described as substitutions, omissions and distortions. Shriberg, Austin, Lewis, McSweeny, and Wilson (1997a) report methods for calculating a percent consonants correct–revised (PCC-R) score, where common and uncommon clinical consonant distortions are scored correct. This revision was devised to address previous concerns that equal weight was given to omissions, substitutions, and distortions when calculating a PCC score. These authors suggest using this score when comparing children at different ages and/or levels of speech development. Intelligibility refers to the child's ability to be understood by unfamiliar listeners without the use of external nonverbal aids, such as pictures, gestures or objects. Intelligibility measures range from the use of parent-report measures (e.g., McLeod, Harrison, McAllister, & McCormack, 2013) or a tally of intelligible words (e.g., Flipsen, 2006; Shriberg et al., 1997b) to simple rating scales. Intelligibility measures provide some information regarding the severity of the speech difficulty; however, they yield no information regarding the nature of the SSD.

Auditory-perceptual coding information on suprasegmental aspects of speech can be taken from a child's production of polysyllabic words and connected speech sample. Information regarding phrasing, rate, stress, loudness, pitch, voice, quality and resonance is needed (Peppé & McCann, 2003; Shriberg et al., 1990).

Summary

There are clear published guidelines for the speech assessment of a general pediatric population. These recommendations include tasks for speech sampling and data analyses appropriate for the child's linguistic level of development. Although this literature is not specific to children with ASD, it can be used to provide best practice guidelines for future research with this population (Table 4).

Recommendations

Core Components of Evidence-Based Speech Assessment of Children With ASD

Clinicians are constantly trying to balance thoroughness with efficiency (Tyler et al., 2002) and may argue that researchers are afforded luxuries in the assessment process that could not be completed in a demanding clinical setting. Although there is no doubt that managing a clinical caseload is challenging, researchers should also be challenged to complete a thorough and complete evidence-based assessment within realistic time constraints. This is particularly pertinent when assessing children with ASD, with their own challenges with compliance, joint attention, and imitation.

Speech assessment methods recommended for children with ASD are also suggested for clinical use, although it is acknowledged that it may be more challenging for clinicians to complete all of the data analyses recommended

Table 4. Guidelines for speech production assessment with children with autism spectrum disorders.

Parameter	Prelinguistic	Verbal
Data collection method		
Oral motor assessment	✓	✓
Connected speech sample (≥ 50 speech-like utterances)	✓	✓
Single-word naming (≥ 100 words)		✓
Phonetic stimulability	✓	✓
Polysyllabic words		✓
Minimum data analysis		
Phonetic repertoire	✓	✓
Syllable shapes	✓	✓
Stress pattern analysis	✓	✓
Intelligibility rating		✓
Error analysis		✓
Additional data analysis		
PCC/PVC		✓
Consistency of production		✓
Imitated vs. spontaneous speech	✓	✓
Sentence imitation		✓

Note. PCC = percentage consonants correct; PVC = percentage vowels correct.

(Table 4). For children with ASD at a prelinguistic level of development, a connected speech sample of at least 50 speech-like vocalizations and phonetic stimulability are recommended. This information would allow the child's phonetic repertoire and syllable shape inventory to be described, in addition to allowing a stress pattern analysis to be completed. Further comparisons would also be made between the child's imitated versus spontaneous speech production, which is useful when differentially diagnosing a motor speech disorder. For children with ASD at the verbal stage of development, it is recommended that a connected speech sample is collected, in addition to performing a single-word naming task, polysyllabic-word naming task, and phonetic stimulability test. A list of recommended minimum data analyses and considerations for additional analyses to aid the differential diagnosis of SSDs are listed in Table 4.

Future Research Directions

To date, very little is known about the speech of children with ASD. This is an emerging area of research, with much scope for future exploration. There is a wide range of questions that can be asked. Some of these include, can children with ASD complete the recommended speech assessment battery? Can children with ASD at a prelinguistic stage of development complete a phonetic stimulability task? What are the speech characteristics of younger and/or less verbal children with ASD? How does the speech of children with ASD change over time?

Conclusion

There is a growing body of research investigating the speech development of children with ASD, both at the prelinguistic level and in the verbal population. This review identified 21 articles that assessed the speech of children with ASD. The participants, assessment tasks, and reporting standards varied widely, making it difficult to identify trends between studies. Following a review of the pediatric SSD literature, both clinical and research guidelines for speech assessment of children with ASD are recommended. The use of standard procedures will allow future comparisons between studies. Although some research teams may have collected the recommended data, they have not reported it to date. As this body of research continues to grow and more detailed information is consistently reported, it is anticipated that future comparisons between studies will allow best practice clinical guidelines for the speech assessment of children with ASD.

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Chapter 3: Speech Abilities in a Heterogeneous Group of Children with Autism

Paper 2: Speech Abilities in a Heterogeneous Group of Children with Autism

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Author Contribution Statement

As co-author of the above paper and primary supervisor, I confirm that Kate Broome made the following contributions:

- Conception of the research questions in collaboration with co-authors
- Literature reviews
- Recruitment of participants
- All data collection
- Data entry and data analysis/interpretation in collaboration with co-authors
- Writing the first draft of the paper with subsequent drafts developed in collaboration with co-authors
- Journal submission
- Journal revisions in collaboration with co-authors
- Journal resubmission

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Date: 28 November 2021

Note. This cross-sectional study presents Time 1 data from the longitudinal study. The longitudinal results are presented in paper 3 (Chapter 4).

JSLHR

Research Article

Speech Abilities in a Heterogeneous Group of Children With Autism

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Purpose: This study aimed to provide detailed descriptive information about the speech of a heterogeneous cohort of children with autism spectrum disorder (ASD) and to explore whether subgroups exist based on this detailed speech data. High rates of delayed and disordered speech in both low-verbal and high-functioning children with ASD have been reported. There is limited information regarding the speech abilities of young children across a range of functional levels.
Method: Participants were 23 children aged 2;0–6;11 (years;months) with a diagnosis of ASD. Comprehensive speech and language assessments were administered. Independent and relational speech analyses were conducted from single-word naming tasks and spontaneous speech samples. Hierarchical clustering based on language, nonverbal communication, and spontaneous speech descriptive data was completed.
Results: Independent and relational speech analyses are reported. These variables are used in the cluster analyses, which identified three distinct subgroups: (a) children with high language and high speech ability (n = 10), (b) children with low expressive language and low speech ability but higher receptive language and use of gestures (n = 3), and (c) children with low language and low speech development (n = 10).
Conclusions: This is the first study to provide detailed descriptive speech data of a heterogeneous cohort of children with ASD and use this information to statistically explore potential subgroups. Clustering suggests a small number of children present with low levels of speech and expressive language in the presence of better receptive language and gestures. This communication profile warrants further exploration. Replicating these findings with a larger cohort of children is needed.
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Children are diagnosed with autism spectrum disorder (ASD) when they present with specific social communication impairments, in addition to restricted and repetitive behaviors (American Psychiatric Association, 2013). The heterogeneity of individuals with ASD is well documented (Boucher, 2012; Pickles et al., 2014). Language and cognitive ability do not feature in the diagnostic criteria and are the most varied comorbid characteristics of individuals with ASD (Plesa Skwerer et al., 2016; Tager-Flusberg et al., 2005). At one end, children with ASD may present with significant cognitive and language impairments and remain nonverbal. At the other end, children with ASD may present with superior function in these domains and even be verbose.

Until recently, communication research in children with ASD has focused on language and pragmatic ability with little information available regarding a child's speech development. The growing interest in the speech ability of children with ASD has emerged from several lines of enquiry. First, a number of researchers are interested in differences in the prelinguistic speech of children with ASD as a potential early behavioral marker for autism. Second, speech sound disorders (SSDs) have been reported in high-functioning, verbal, school-age children with ASD. Finally, some researchers have recently described subgroups of children with ASD with impaired speech development.

The available research investigating the speech of children with ASD is limited. Most studies have focused on homogeneous groups of children at either the very early stages of development or the higher end of functioning. These studies have included both very young children and school-age children, leaving a gap in our knowledge regarding the speech of children with ASD aged 2–6 years.

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Furthermore, few studies to date have included detailed descriptive speech data (e.g., consonant and syllable shape repertoires) as opposed to standardized scores based on accuracy of adult targets. Such data are relevant for diagnosis, monitoring, and intervention planning. In addition, participant descriptions improve researchers' ability to draw comparisons between studies.

Prelinguistic Vocalizations of Children With ASD

Prelinguistic vocalizations are sounds produced by children with expressive vocabularies of less than 10 (Stoel-Gammon, 1989; Thal et al., 1995) or 20 (Kasari et al., 2013) words and can be categorized as speechlike or nonspeechlike. The prelinguistic vocalizations of children with ASD have been studied via retrospective videos of children later diagnosed with ASD (Watson et al., 2013; Werner & Dawson, 2005) and prospective studies of children at risk of ASD, such as siblings of children with ASD (Paul et al., 2011). The finding that deficits in early vocal development correlated with poorer expressive language outcomes for children with ASD (McDaniel et al., 2018; Saul & Norbury, 2020) highlighted the importance of prelinguistic speech assessment.

The prelinguistic vocalizations of children with ASD have been shown to differ from age-matched children with typical development (Plumb & Wetherby, 2013; Schoen et al., 2011). Prelinguistic children with ASD produce a higher proportion of nonspeech vocalizations (i.e., squeals, raspberries, and hums) than age-matched children (Paul et al., 2011; Plumb & Wetherby, 2013; Schoen et al., 2011). Fewer consonants and less complex syllable forms have also been reported (Paul et al., 2011; Plumb & Wetherby, 2013; Schoen et al., 2011), although few studies have closely analyzed prelinguistic speechlike vocalizations, and information regarding phonemic repertoire and syllabic complexity is limited. Schoen et al. (2011) provide one of the most detailed descriptions of the speech of toddlers with ASD. In their study, the speech development of 30 toddlers with ASD (23 prelinguistic) was compared with language and age-matched controls. Consonant accuracy and consonant inventories categorized by order of acquisition, consonant blend inventories, and mean syllable structure level were reported. The children with ASD produced significantly fewer consonants from the early-, middle-, and later-acquired consonant groupings, fewer consonant blends, more non-English blends, and fewer consonant–vowel (CV) or complex syllable forms than the age-matched controls.

The picture is less clear when comparing the speech of children with ASD to language-matched children. A higher rate of nonspeech vocalizations in children with ASD has been reported (Sheinkopf et al., 2000), although conflicting results have also been reported (Schoen et al., 2011). The speechlike vocalizations of children with ASD were commensurate with that of children at comparable linguistic levels in two studies (McCleery et al., 2006; Schoen et al., 2011). In Schoen et al. (2011), children with ASD produced similar number of consonants and complexity of syllable forms to language-matched children. Studies that include detailed analyses of speechlike prelinguistic vocalizations are required to develop this body of research.

Speech of Verbal Children With ASD

Research investigating the speech of verbal children with ASD has focused primarily on high-functioning school-age children. Most studies have relied on standard assessment scores with little, if any, detailed descriptive information reported. Independent speech analyses describe a child's speech without reference to the adult target. Independent analyses are useful in understanding a child's speech capacity (as opposed to their speech accuracy) and represent a strengths-based approach to speech assessment. Few studies of verbal children with ASD have included detailed independent speech analyses. The nine (30%) low--verbal children in the Schoen et al. (2011) study produced between 10 and 30 words and were found to have similar speech development to children at comparable linguistic levels. Only four other studies with verbal children have reported phonetic or phonemic transcription data (Cleland et al., 2010; Riches et al., 2011; Shriberg et al., 2001, 2011). Nearly all participants in these studies were school age or older, with ages in the ranges of 4–7 years (Shriberg et al., 2011), 5–13 years (Cleland et al., 2010), 10–49 years (Shriberg et al., 2001), and 14;0–15;4 (years;months; Riches et al., 2011). There is a paucity of reported independent speech analyses of verbal children with ASD, and no study has provided detailed speech data for a heterogeneous group of young children with ASD.

Relational speech analyses reference adult targets. The few available studies presenting these data suggest older verbal children with ASD, particularly at higher levels of functioning, may present with relatively high rates of SSDs. Both articulatory (Cleland et al., 2010; Shriberg et al., 2001, 2011) and phonological (Cleland et al., 2010; Rapin et al., 2009) speech errors have been reported, although average speech skills have also been found (e.g., Kjelgaard & Tager-Flusberg, 2001). More detail is needed regarding the speech accuracy of verbal children with ASD, with particular focus on younger and lower functioning children.

Speech Subgroups Within ASD

Given the known heterogeneity in autism, the idea that definable subgroups exist within the larger cohort of children with ASD is logical. Researchers have begun to report subgroups of children with different speech sound development patterns (Chenausky et al., 2019; Rapin et al., 2009); however, varied terminology and methodological limitations make it difficult to draw comparisons between existing studies.

Kjelgaard and Tager-Flusberg (2001) were among the first to include a speech measure in a study of language profiles of children with ASD. This study categorized a heterogeneous cohort of 82 verbal children with ASD, aged 4–14 years, into "impaired" (n = 50), "borderline"

(n = 10), or "normal" (n = 22) language subgroups based on Peabody Picture Vocabulary Test (Dunn & Dunn, 1981) scores. All three subgroups scored within normal limits on the Goldman–Fristoe Test of Articulation (Goldman & Fristoe, 1986), although children in the "impaired" subgroup scored significantly lower than the other two subgroups. Average scores on the Goldman–Fristoe Test of Articulation suggested that expressive phonology at the single-word level was unimpaired in children with autism in this study. Although interesting, these results cannot be generalized to younger or less verbal children with autism. It remains unclear if the same result would be obtained if detailed independent descriptive data had been used instead of standardized scores. It is also not known if forming subgroups manually based on comprehension of single words alone is the most meaningful or empirically valid way to view the heterogeneous communication ability of children with ASD.

Rapin et al. (2009) were the first to report subgroups of children with ASD with atypical phonological development. These researchers studied the language of 62 children with ASD, aged 7–9 years, with the aim of defining clinical subtypes of language disorders. Participants were assessed for nonverbal and verbal cognition, language comprehension, receptive and expressive word-level language, single-word naming, and perceptually rated intelligibility and oromotor function. Detailed assessment and analyses of speech were not included. Four subgroups emerged from the data using hierarchical cluster analysis: (a) significant impairments on all assessments (n = 11); (b) "profoundly" impaired phonology, borderline or average comprehension, and average nonverbal IQ (n = 4); (c) adequate phonology, expressive vocabulary, and nonverbal IQ but impaired receptive language and verbal IQ (n = 40); and (d) average or above on all assessments (n = 7). Few have attempted to replicate these findings or explore whether similar results can be obtained with younger children.

Chenausky et al. (2019) studied the speech of a more homogeneous group of 54 "minimally verbal" (less than phrase-level language) and "low-verbal" (phrase-level language) individuals with ASD aged between 4;4 and 18;10. Videos of participants completing portions of a speech praxis test were coded for signs of childhood apraxia of speech (CAS; American Speech-Language-Hearing Association, 2007), and participants were manually categorized into one of four descriptive subgroups based on speech presentation. Four subgroups emerged: (a) within normal limits (n = 12), (b) non-CAS speech difficulties (n = 16), (c) suspected CAS (n = 13), and (d) insufficient speech (n = 13). These results provide preliminary support for the hypothesis that some minimally verbal and low-verbal children with ASD present with co-occurring motor speech disorders (Dawson, 2010; Prizant, 1996; Velleman et al., 2010). However, because the cohort in this study was selected based on low levels of expressive language, it is not clear whether similar subgroups are present across a more heterogeneous population. Also, it remains unknown if comparable subgroups emerge when statistical methods for clustering, based on independent and relational speech analyses, are used instead of manual categorization.

Why Study a Heterogeneous Cohort?

Although widely recognized, the heterogeneity of this population is understudied and the value of examining a heterogeneous cohort is often overlooked (Georgiades et al., 2013). A heterogeneous cohort best reflects the clinical population. Research that aims to include children at all levels of the spectrum can help to further our understanding of autism and provide useful information regarding presentation and development (Georgiades et al., 2013). Such research may highlight factors that best explain change over time. Although large population studies are needed to reach conclusions regarding prevalence, causality, and outcomes, small cross-sectional and heterogeneous cohort studies may initially be needed to highlight interesting areas for future research.

In summary, despite growing interest in the speech of children with autism, significant gaps in the research persist. The challenges in completing speech assessments with children with ASD are significant (see Broome et al., 2017, for review) and may explain the paucity of research in this area. A picture of higher rates of delayed and disordered speech in children with ASD compared with their typically developing peers is beginning to emerge in the limited published data available. A small subgroup of children with limited verbal output and impaired speech sound development has also been described. However, few studies have provided detailed descriptive information of the speech capacity of children with ASD across heterogeneous cohorts, and information on the speech of 2–6 year old children with autism, in particular, is lacking. Descriptive data are essential for differential diagnosis of communication disorders, and appropriate intervention planning and may assist comparisons between research studies. This study aimed to (a) describe in detail the speech of a heterogeneous cohort of children with ASD aged 2;0–6;11 and (b) use this detailed speech data to explore whether subgroups of children with ASD exist within the cohort.

Method

The research protocol was approved by the Human Research Ethics Committee of The University of Sydney (2012/712 and 2012/1305) as part of a longitudinal descriptive evaluation of speech development in children with ASD. Parents of all participants provided written consent to be included in this study.

Eligibility and Recruitment

Participants were recruited from three sources. Written information was placed on the ASPECT website and the Facebook site, e-mailed to early intervention service providers in the greater metropolitan area of Sydney, and e-mailed to 18 private speech pathologists in the Sydney area who listed ASD as an area of interest on the Speech Pathology Australia website or who were a member of the ASD evidence-based practice interest group in Sydney. Parents contacted the first author and were screened for eligibility over the telephone.

Inclusion criteria included the following: (a) documented evidence of a diagnosis of ASD within 12 months of recruitment by a developmental pediatrician in accordance with the *Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition Text Revision* (American Psychiatric Association, 2000) or *Fifth Edition* (American Psychiatric Association, 2013); (b) producing speechlike vocalizations; (c) aged between 2;0 and 6;11; (d) documented results of a developmental or cognitive assessment or, if this was not available at the time of recruitment, the intention to complete a developmental assessment within the time frame of the broader longitudinal study; and (e) English as the primary language of at least one caregiver and the primary language of the child.

Children were excluded from the study if they were diagnosed with any comorbid developmental disorders or genetic syndromes, were born at less than 36 weeks gestational age, or had any uncorrected hearing or visual deficits. All participants completed an oral motor assessment with the first author, a clinician with over 10 years' experience with children with diverse disorders, including ASD and cleft palate. The informal assessment included the core components of an oral motor assessment as outlined in the literature (Duffy, 2000; Kent, 2015; Strand et al., 2013). Observations of the strength, symmetry, accuracy, speed, and range of movement of the lips, tongue, face, and palate at rest, during static postures, and during nonverbal oral movements were recorded. Children were excluded from the study if there was asymmetry or weakness of the oral musculature, which resulted in significant drooling and/or dysarthria. Children were also excluded if there were any oral structural abnormalities (e.g., cleft palate). No child was excluded from this study following oral motor assessment.

Participants

Participants were 21 boys (91%) and two girls (9%) with ASD, aged between 24 and 74 months (M = 46.1, SD = 14.6). All eligible children whose parents contacted the researchers and consented were included in the study. A further child, Participant 10, was recruited, however withdrew from the study prior to the first assessment. The first author completed and scored all clinical assessments according to the assessment manuals. A description of participants is reported in Table 1.

Assessment Measures

Assessments were completed at the participants' home (96%) or at the on-campus clinic at The University of Sydney (4%). Due to the large assessment battery and the desire to capture participants' optimal communication level, most assessments were conducted over two sessions.

Table 1. Description of participants.

Variable	*n*	*M*	*SD*	Range
Age (months)	23	46.2	14.8	24–64
CDI				
Receptive Vocabulary	23	232.8	156.3	1–396
Expressive Vocabulary	23	169	175.8	0–396
Gestures	23	40.1	17.4	6–63
CSBS Behavior CC	9	54.6	8.3	47–70
PLS-4				
AC	14	66.1	14.9	50–92
EC	14	65.6	14.2	50–85
TLS	14	64.4	12.9	50–87
GMDS-ER				
Nonverbal DQ	13	59.3	23.4	21.3–108.3
Stanford–Binet				
Nonverbal IQ	3	88	14.5	74–103
WPPSI-III				
Nonverbal IQ	3	99	20.7	82–122
WISC-V				
Nonverbal IQ	1	86		

Note. CDI = MacArthur–Bates Communicative Development Inventory; Receptive Vocabulary = number of words understood, max score 396; Expressive Vocabulary = number of words produced, max score 396; Gestures = number of gestures, max score 63; CSBS = Communication and Symbolic Behavior Scales; CC = Communication Composite, sum of scaled scores based on level of functioning; PLS-4 = Preschool Language Scale–Fourth Edition; AC = Auditory Comprehension; EC = Expressive Communication; TLS = total language score; GMDS-ER = Griffiths Mental Development Scale–Extended Revised; DQ = developmental quotient; IQ = intelligence quotient; WPPSI-III = Wechsler Preschool and Primary Scale of Intelligence–Third Edition; WISC-V = Wechsler Intelligence Scale for Children–Fifth Edition.

All assessment sessions were video- and audio-recorded. Participants completed a comprehensive communication assessment battery, including direct language and speech measures, informal speech sampling, parent questionnaires, and the previously mentioned oral motor assessment.

To provide descriptive data on the communication ability of a heterogeneous cohort of children with ASD, a balance needed to be achieved between completing the same assessment with all individuals and capturing the communication capacity of every child. Therefore, although all children in the heterogeneous cohort were presented with the complete primary assessment battery, individuals were not excluded from this study if unable to complete one or more individual assessments. Instead, alternative measures were presented to provide the child with opportunities to demonstrate their optimal communication ability. This strengths-based assessment approach provides more detailed information on what a child can do and is widely used in clinical practice with children with ASD (Cosden et al., 2006).

The Preschool Language Scale–Fourth Edition (PLS-4; Zimmerman et al., 2002) was attempted with all participants. Fourteen participants (61%) completed the PLS-4, a standardized language assessment for children from birth to 6;11. This age range encompasses all participants in this study; however, previous research with children with ASD reports difficulty completing standardized language assessments particularly for young children or children at very early stages of linguistic development (Charman et al., 2003; Volden et al., 2011). For example, almost 30% of the 294 children aged 2;0–4;11 in the Volden et al. (2011) study performed at floor level on the PLS-4. Where it was not possible to obtain a basal level on the PLS-4 in this study, the Communication and Symbolic Behavior Scales (CSBS) Behavior Sample (Wetherby & Prizant, 2002) was employed as recommended by Volden et al. The CSBS is a standardized assessment of communication for children at early stages of development. The CSBS assesses language comprehension and word use, in addition to other important aspects of very early communication development, such as social-affective signaling, nonverbal communication, and joint attention. The tool can be used with children aged 8–24 months or with language developmental levels of less than 24 months. Nine participants (39%) completed the CSBS in this study. Six participants were prelinguistic, and three children were using less than phrase-level speech. Standard scores reported are based on the child's language stage as recommended in the manual.

The Polysyllable Preschool Test (POP; Baker, 2013) was attempted with all participants. Ideally, both a single-word naming task assessing all phonemes in all word positions and a polysyllabic word assessment would be administered. However, given the compliance challenges and lengthy assessment battery, a polysyllabic assessment was prioritized to add information on stress patterns and phonological ability that may not be available from a self-directed, spontaneous speech sample. Eleven participants (48%) completed the POP. Participants at early stages of verbal development were unable to complete the POP assessment. The First Words First Sentences Test (FWFST; Gillham et al., 1997) was administered to these children. This single-word naming task contains early developing consonants and vowels as well as syllable shapes, which are important to assess in very young children and children at the early stages of language development (e.g., Stoel-Gammon, 1998). The FWFST also presents photographs rather than symbolic pictures, which can present a challenge for some children with ASD (Hartley & Allen, 2015). Six participants (26%) completed this assessment. A further six participants (26.1%) were unable to complete either single-word naming task. These six children were prelinguistic, producing fewer than 10 recognizable words (Broome et al., 2017; Stoel-Gammon, 1989).

Spontaneous speech samples were collected for each participant. An utterance was defined as a string of speech-like vocalizations preceded and followed by a pause or interruption (Nathani et al., 2006; Stark et al., 1993). Utterances in this study could be babble, words, phrases, and sentences. Speechlike vocalizations produced at the same time as additional noise, such as adult speech or noisy toys, were not transcribed. Utterances were coded as "babble" if the target word(s) remained unclear after listening to the recording 3 times. For verbal children in the cohort, 50 or more utterances were collected during at least 10 min of parent–child play interactions. Prelinguistic and low-verbal participants in this study produced more phonemes during the CSBS assessment than during play with a parent, so for these children a speech sample was taken during the entire CSBS behavior sample, in keeping with previous research (Schoen et al., 2011). For children who were unable to reach at least 50 speechlike utterances, all utterances from the entire assessment battery were transcribed. One prelinguistic participant and two verbal children produced less than 50 speechlike utterances during the entire assessment, and this is reported in Table 2. Echolalia was included for two reasons. First, for children at early stages of linguistic development, it was too difficult to determine what was or was not delayed echolalia. Additionally, echolalia may represent part of a child's articulation capacity and may add valuable information regarding a child's phoneme repertoire and capacity for syllable complexity.

The MacArthur–Bates Communicative Development Inventory (CDI) Words and Gestures (Fenson et al., 2007), a parent questionnaire, was completed by all parents. The CDI Words and Gestures form is standardized on children ages 0;8–1;4, and the CDI Words and Sentences form is standardized on children ages 1;4–2;6. The CDI is a 396-item checklist of the child's receptive and expressive vocabulary. Parents in this study were asked to mark the words their child "understands" and words their child "'says" separately, rather than words the child "understands" and "understands and says," as indicated on the form. The CDI Words and Gestures form also tallies the child's use of 18 early gestures (i.e., communicative and games/routines) and 45 later gestures (i.e., actions with objects, pretending to be a parent, and imitating adult actions). CDI raw data are reported in this study as children were outside the standardized age range. Parent report measures are useful for children who may not perform well during formal standardized assessment, as is the case with a large proportion of children with ASD (Charman et al., 2003). The CDI has been used by several research groups as a measure of vocabulary in children with ASD (Charman et al., 2003; Luyster et al., 2007; Stone & Yoder, 2001). Charman et al. (2003) examined early vocabulary development in 134 preschool children with ASD, ages 18 months to 7 years, using the CDI Words and Gestures form. Like these authors, we required one instrument that could provide data on all children in our study. It was suspected that the CDI Words and Sentences form would be above the level of some participants and therefore could not be used. We also wanted a measure of nonverbal communication that could be used with all participants. Although some gestures on the CDI are communicative, other items give an indication of the child's level of imaginative and symbolic play.

It is important to highlight that comparing data from multiple assessment tools is problematic. For example, the used single-word naming tasks varied in the number, length, and complexity of words, whereas the various language assessments assessed different skills. The paucity of valid standardized assessment tools that can be used with children with ASD across ages and levels of functioning limits our profession's understanding of the nature of the heterogeneity in autism (Kasari et al., 2013; Plesa Skwerer et al., 2016). Until an appropriate measure is developed, the large proportion of

Table 2. Participant speech characteristics.

	Participants																						
Variable	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**11**	**12**	**13**	**14**	**15**	**16**	**17**	**18**	**19**	**20**	**21**	**22**	**23**	**24**
Age in months	34	48	74	30	33	55	24	37	56	35	63	36	30	63	55	43	62	38	63	43	43	28	68
Independent analyses																							
Consonants	2	9	5	10	14	18	6	16	21	4	4	6	10	23	19	12	18	16	21	18	19	8	18
Early 8	2	7	5	6	7	8	5	8	8	3	4	6	7	8	8	7	8	8	8	8	8	6	8
Middle 8	0	1	0	3	4	6	1	5	8	1	0	0	2	8	7	3	7	4	8	7	7	2	7
Late 8	0	1	0	0	3	4	0	3	5	0	0	0	1	7	4	2	3	4	5	3	4	0	3
Syllable shape—single-word naming																							
V		15.4	52.5		2.1	2.7		11.1	1.5		48.2	57.1	28.6	1.5	3		4.1	2.8	1.5	4.1	9.1		7.5
CV		84.6	47.5		49	68.5		60.3	70.6		33.3	37.1	71.4	71.6	71.6		74.3	33.3	70.2	50.7	69.7		61.3
CVC					34.7	13.7		17.5	13.2		7.4	2.9		14.9	9		10.8	33.3	14.9	27.4	10.6		a16.3
VC						5.5		6.3	4.4			2.9		6	4.4		2.7		4.5	5.5	3		3.7
CCV						8.2		3.2	10.3					6	9		9.5	2.8	7.5	2.7	6.1		6.3
CCVC								1.6			11.1							8.3		4.1			1.3
CVCC					10.2	1.4									3		1.4	5.6	1.5	5.5	1.5		3.7
CCVCC																		8.3					
VCC					2													2.8					
VCCC					2													2.8					
Syllable shape—spontaneous speech samples																							
Utterances	28	58	57	79	71	50	50	60	56	52	44	48	70	58	75	86	74	50	67	54	55	83	64
V	93.9	23.4	34.5	26.2	10.2	17.9	53.6	7.8	9.5	29.3	67.1	70.3	24.3	10.5	15.5	21.4	9.7	5.2	10.7	12.7	10.2	49.6	9.9
CV	6.1	76.6	65.5	73.8	66.2	38.5	46.4	62.8	32.6	69.6	24.1	28.4	75.2	34.3	37.9	71.3	33.3	55.2	40.9	53.2	56.7	50.4	52.7
CVC					19.4	22.2		17.8	23.6	1.1	1.2	1.3		30.2	25.6	6.1	31.8	28.5	23.3	24.6	21.3		15.8
VC					3.2	13.2		7.8	12.2		7.6			9.3	8.3	1.2	5	8.1	11.2	2.8	4.7		5.4
CCV						0.4		1.7	1.1					1.7	1.1		0.4	1.2	3.7	0.7			2
CCVC						0.9		1.1	3.5				0.5	2.9	1.8		2.7	1.7	3.5	3.5	4.7		6.9
CVCC					0.9	4.3		0.5	9.2					3.5	8.7		12.8		4	1.1	2.4		4.9
VCC						1.7		0.5	5.2					4.7	0.7		2.7		2	0.4			2.5
CCVCC						0.4			3.3					2.3			1.6		0.6	1.1			
CVCCC						0.4								0.6	0.4								
Relational speech analyses—single-word naming																							
PCC		30.5	4.9		57.1	81.7		48.1	79.7		18.8	7	29.6	92.7	76.8		74.4	64.3	91.5	72.1	58.5		63.5
PVC		90.9	10.5		82.1	91		89.4	96.7		25	35.1	68.6	100	92.5		88.1	91.4	98.5	97.2	85.1		85.1
PPC		54.7	7.4		65.8	85.9		64.1	87.4		21.3	18.1	46.8	96	83.9		80.5	63.8	94.6	81.4	70.5		73
Stress						86.8			90.2					98.5	98		88.2		100		78.3		85.6
Relational speech analyses—spontaneous speech samples																							
PCC		21.7	11.9		51.6	84.2		62.4	88.4		21.1	4.6	33	94.4	80.4		83.1	73.9	91.6	66.9	77		70.1
PVC		71.5	17.6		82	93.9		85.7	96.6		31	23.4	78.4	97.1	89.2		91.4	95	96.8	78.4	92.6		82.7
PPC		41.9	14.1		64.2	88.5		71.8	91.5		25.3	12.5	57.2	95.5	84.1		86.1	82.1	93.8	71.6	83.2		75.3

Note. The consonants were categorized as Early 8, Middle 8, and Late 8 (Shriberg, 1993). Consonants = the number of consonants in phoneme repertoire; Syllable shape = proportion of each syllable type; V = vowel; C = consonant; Utterances = number of utterances in spontaneous speech sample; PCC = percent consonants correct; PVC = percent vowels correct; PPC = percent phonemes correct; Stress = proportion of stress matched syllables in speech sample.

children with ASD who are unable to complete standardized assessments will be excluded from research (Plesa Skwerer et al., 2016).

The focus of this study was to describe the speech of a heterogeneous cohort of children with ASD and to use these data to explore the possibility of subgroups within the cohort. Given the large assessment battery and the professional scope of the authors, it was decided that repeating developmental screening was outside the scope of this study. Instead, we relied on the available data provided by the families. Some children completed a developmental or cognitive assessment at a later date and made these assessment scores available to the researchers. The results from a formal developmental or cognitive assessment were not available for three participants. The results of the Griffiths Mental Developmental Scales–Extended Revised (Luiz et al., 2004) were available for 13 participants. A developmental quotient based on age equivalent divided by chronological age and multiplied by 100 was used as many participants scored below the first percentile. A nonverbal developmental quotient was calculated from the performance scale. The verbal developmental quotient was calculated from the hearing and speech scale. The results of a cognitive assessment were available for seven participants. For these children, the verbal intelligence quotient and the nonverbal intelligence quotient are presented.

Reliability

The first author completed broad phonemic transcription for all responses on the single-word naming task and retranscribed 22% of the data to check for intrarater reliability. An independent researcher transcribed 17% of the single-word naming tasks, randomly selected using random.org. Intrarater reliability was 97.8%, and interrater reliability was 97%.

The first author transcribed the spontaneous speech samples using broad phonemic transcription and retranscribed 13% of randomly selected samples. An experienced speech-language pathologist (SLP) researcher transcribed 13% of randomly selected spontaneous speech samples. Intrarater agreement for broad phonetic transcription was 93.6%, and interrater reliability was 91.3%

The first author tallied the total number of different consonants from the entire assessment battery. The first author completed these ratings again for 22% of participants more than 6 months following the initial analysis. An independent postgraduate SLP tallied the total number of consonants for 10 (44%) participants. Intrarater reliability was 95.4%, and interrater reliability was 91.2%.

Data Analysis

All responses during the single-word naming task and spontaneous speech sample were transcribed using broad phonemic transcription and entered into Phon 3.1 computer software (Hedlund & Rose, 2020). Stress patterns and phoneme accuracy were calculated using Phon. Descriptive statistics were completed.

Independent Speech Analyses

Consonant inventories. The total number of consonants in the participants' speech sound repertoire was tallied from the single-word naming task and the spontaneous speech sample. The consonants were categorized as Early 8, Middle 8, or Late 8 consonants (Shriberg, 1993).

Syllable shapes. All speechlike utterances from the speech samples were analyzed manually according to syllable shapes. Syllables were defined as having a nucleus (i.e., vowel) and optional prevocalic or postvocalic consonants (Grunwell, 1982). Single consonants (i.e., /m/) were not recorded in this count. A consonant blend was depicted as CC or CCC, depending on the number of consonants in the blend. For example, "three" would be recorded as CCV to reflect the two consonant sounds followed by the vowel. The number and proportion of different syllable shapes was then calculated.

Relational Speech Analyses

Stress patterns. Lexical stress is a prosodic feature of speech, referring to the relative emphasis placed on syllables within a word, typically a combination of weak and strong syllables or equal stress. Lexical stress is achieved in English by changing one or more of the duration, intensity, or pitch of a syllable in relation to other syllables in a word (Ballard et al., 2010). Lexical stress accuracy was calculated as a proportion of lexical stress matches on the POP (Baker, 2013).

Phoneme accuracy. Percent consonants correct (PCC), percent vowels correct (PVC), and percent phonemes correct (Shriberg & Kwiatkowski, 1982) were calculated from the single-word naming task and the spontaneous speech sample for the 17 verbal participants.

Hierarchical Cluster Analysis

Hierarchical cluster analysis uses the variables collected, including descriptive speech data, to explore whether meaningful homogeneous subgroups exist within the data. It is a useful approach when there is good reason to believe heterogeneity among the sample, as is the case with this cohort of children with ASD. An advantage of cluster analysis is that it provides visual representation of the clustering process (e.g., two clusters, then three, then four), which can then be described and interpreted. It is a way of discovering interesting patterns within the data but cannot be used to inform the predictive value of each of the individual speech variables on outcomes (Hastie et al., 2009; James et al., 2013; Masso et al., 2017). Agglomerative clustering is the most common type of hierarchical clustering. In agglomerative clustering, the dendrogram is built bottom-up, starting from the "leaves" (participants) and combining clusters up to the "trunk" (James et al., 2013).

Given the large number of communication variables in this study and limited past research to inform our selection of variables, a number of factors were considered when choosing variables. First, we limited this cluster analysis to include data from the spontaneous speech samples and the CDI since these were the consistent assessment methods used with all participants. Additionally, it was important to include variables that describe all aspects of the child's communication profile, including understanding, expression, nonverbal communication, and various independent and relational speech measures. Six communication measures (CDI Receptive Vocabulary, CDI Expressive Vocabulary, CDI Number of Gestures, consonant repertoire, PCC, and PVC) were used as clustering variables. Syllable shapes were not included in the final cluster analysis, although including all communication variables with this data set did not change the final dendrogram. To ensure each variable was on a common scale, data were converted into z scores using Scale in R, with a mean of 0 and an *SD* of 1.

Following statistical guidelines (Hastie et al., 2009; James et al., 2013) and in keeping with past research in the field (Allison & Hustad, 2018; Strand et al., 2013), Euclidean distance with complete linkage was used. Euclidean distance is the most common dissimilarity measure and results in a participant being fused to another when the scaled data are most similar (James et al., 2013). Observations that fuse at the very bottom of the dendrogram are quite similar to each other, whereas observations that fuse close to the top will be quite different. Linkage refers to how fusions occur between groups rather than individual participants (James et al., 2013). Average and complete linkage tend to result in more balanced dendrograms. There was no change in the dendrogram when either complete or average linkage was used.

Determining the number of clusters is subjective and requires the researchers to determine if the addition of further clusters adds meaning to the results. To identify clusters, a horizontal line across the dendrogram is made. For this data set, the dendrogram visually produced two possibilities: (a) cutting the dendrogram at Height 4, producing three clusters, and (b) making a horizontal cut at Height 3 to give four clusters. Both the three- and four-cluster solutions were explored when examining the stability of the dendrogram. To investigate how robust the clusters were, variables were systematically removed and added, and the change in the dendrogram was examined (Hastie et al., 2009). A series of Kruskal–Wallis tests were used to examine if the differences between the clusters were significant for each of the communication variables. Due to the small sample size and the possibility of Type I and II errors, these analyses were considered exploratory in nature. Alpha was set at .05 for all comparisons, given the n, adjustment to a stricter alpha level would have inflated the Type II error rate, possibly obscuring interesting avenues for further exploration. Variables that were statistically different on the Kruskal–Wallis test were further examined using Dunn's test. Dunn's test is used to determine which clusters were different on the various communication variables.

Results

Speech Descriptions of the ASD Cohort

Detailed description of the participants' speech is presented in Table 2. Descriptive data were used to inform the cluster analysis and to describe the clusters that emerged following clustering. This is a heterogeneous group by design, and the large spread in the results reflects this variability. Two interesting results emerged from the descriptive data prior to clustering. First, there was a trend of higher receptive vocabulary (M = 232.8, SD = 156.3) than expressive vocabulary (M = 169, SD = 175.8) across the cohort. Second, children in this cohort tended to produce a larger number of Early 8 consonants (M = 6.7, SD = 1.8) compared with Middle 8 (M = 4, SD = 3) and Late 8 consonants (M = 2.3, SD = 2.1). Table 3 details the consonant repertoires of individual participants and illustrates this trend. Scatter plots were used to visualize the speech characteristics of participants in the cohort and are presented for interest in Supplemental Material S1.

Hierarchical Cluster Analysis

The hierarchical cluster analysis was completed to examine whether naturally occurring subgroups existed in the heterogeneous cohort. Euclidean distance and complete linkage were used, although the use of average and centroid linkage made no difference to the dendrogram. The cluster analysis is represented in the dendrogram shown in Figure 1. The dendrogram illustrates the three-cluster solution. Cluster A consists of 10 participants, Cluster B includes three children, and Cluster C includes 10 children.

Cluster Stability

The three-cluster solution remained stable when communication variables were systematically removed or added to clustering. The four-cluster solution was more volatile, although the 10 participants in Cluster A and three participants in Cluster B did not change cluster membership.

Describing the Clusters

A series of Kruskal–Wallis tests were conducted to test for significant differences between clusters on age and each of the communication variables. Results indicated statistically significant differences between clusters on all but one communication variable and are presented in Table 4. Dunn's tests were completed to determine specifically which clusters were statistically different across the variables. These results will be presented below and are available in Supplemental Material S2.

Cluster A: High Receptive, High Expressive, High Gestures, and High Speech

Pairwise comparisons using Dunn's test indicated that the 10 Cluster A children presented with statistically higher expressive vocabulary scores than Cluster B (p = .002) and Cluster C (p = .0001) participants. These children also had larger consonant inventories than either those in Cluster B (p = .0006) or Cluster C (p = .0003). All 10 Cluster A children presented with a complete repertoire of Early 8 consonants and high numbers of Middle 8 and Late 8 consonants. The participants used more postvocalic consonants than those in Cluster B (VC, p = .02; CVC, p = .008) and Cluster C (VC, p = .0003; CVC, p = .0001) and more

Table 3. Participants' consonant repertoires.

Participant	Early 8	Middle 8	Late 8	Total
1	m, j			2
2	m, b, j, n, w, d, p	t	l	9
3	m, b, j, w, h			5
4	m, b, j, w, d, p	t, k, g	s	10
5	b, j, n, w, d, p, h	t, ŋ, k, g	s, z, l	14
6	m, b, j, n, w, d, p, h	t, ŋ, k, g, f, dʒ	s, z, l, ʃ	18
7	m, j, n, w, d	t		6
8	m, b, j, n, w, d, p, h	t, ŋ, k, g, f	ʃ, s, z	16
9	m, b, j, n, w, d, p, h	t, ŋ, k, g, f, v, tʃ, dʒ	ʃ, s, z, l, ɹ	21
11	m, b, j	g		4
12	m, b, j, p			4
13	m, b, j, n, d, h			6
14	m, b, j, n, w, d, h	k, g	z	10
15	m, b, j, n, w, d, p, h	t, ŋ, k, g, f, v, tʃ, dʒ	ʃ, θ, s, z, ð, l, ɹ	23
16	m, b, j, n, w, d, p, h	t, ŋ, k, g, f, tʃ, dʒ	ʃ, s, z, l	19
17	m, b, j, n, w, d, h	t, k, g	ʃ, s	12
18	m, b, j, n, w, d, p, h	t, ŋ, k, g, f, tʃ, dʒ	ʃ, s, z	18
19	m, b, j, n, w, d, p, h	t, k, g, f	ʃ, s, z, l	16
20	m, b, j, n, w, d, p, h	t, ŋ, k, g, f, v, tʃ, dʒ	ʃ, s, z, l, ɹ	21
21	m, b, j, n, w, d, p, h	t, ŋ, k, g, f, tʃ, dʒ	ʃ, s, z	18
22	m, b, j, n, w, d, p, h	t, ŋ, k, g, f, v, dʒ	ʃ, s, z, ɹ	19
23	m, b, n, w, d, h	k, g		8
24	m, b, j, n, w, d, p, h	t, ŋ, k, g, f, tʃ, dʒ	ʃ, s, z	18

Note. The consonants were categorized as Early 8, Middle 8, and Late 8 (Shriberg, 1993).

pre- and postvocalic consonant blends compared to the children in Cluster B (CCV, *p* = .006; CCVC, *p* = .002; CVCC, *p* = .003; VCC, *p* = .008; CCVCC, *p* = .02) and Cluster C (CCV, *p* = .0007; CCVC, *p* = .0001; CVCC, *p* = .0002; VCC, *p* = .0006; CCVCC, *p* = .002). The Cluster A children scored significantly higher than the other two clusters on consonant and vowel accuracy measures. Overall, Cluster A children had significantly different results from the 10 children in Cluster C on all communication variables. Children in Cluster A did not differ from children in Cluster B by age (*p* = .43) or on measures of receptive vocabulary (*p* = .15), use of gestures (*p* = .11), or proportion of CV syllables (*p* = .33).

Cluster B: High Receptive, Low Expressive, High Gestures, and Low Speech

The children in Cluster B (*n* = 3) presented with small expressive vocabularies, not dissimilar to the expressive vocabularies of children in Cluster C on a post hoc Dunn's test (*p* = .37). Although these children produced very few

Figure 1. Dendrogram.

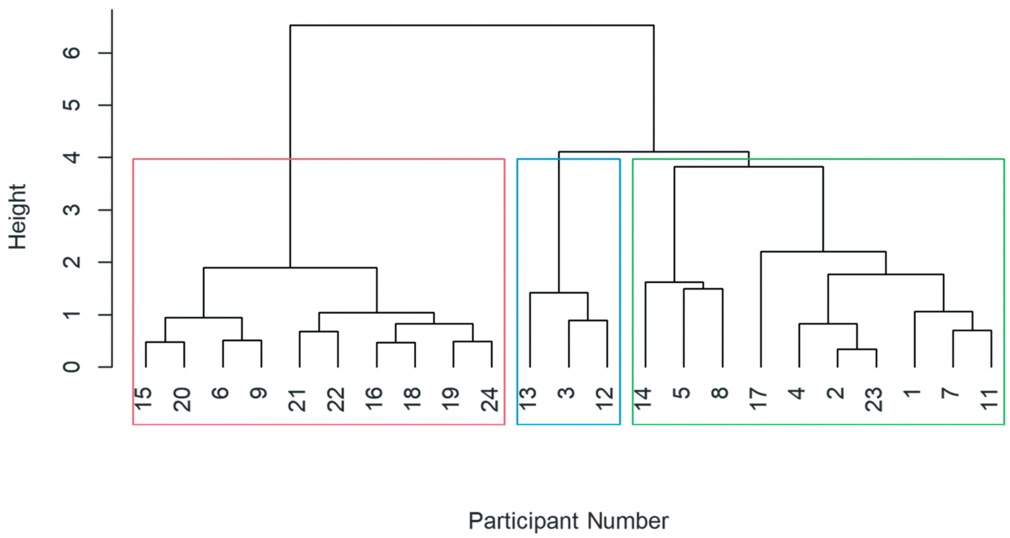

Table 4. Characteristics of clusters.

Variable	Cluster A *n*	Cluster A *M* (*SD*)	Cluster A Range	Cluster B *n*	Cluster B *M* (*SD*)	Cluster B Range	Cluster C *n*	Cluster C *M* (*SD*)	Cluster C Range	χ^2	*p*
Age in months	10	54.6 (10.1)	38–68	3	57.7 (19.6)	36–74	10	34.2 (7.1)	24–48	12.6	.002*
Receptive vocab	10	372 (20.2)	344–396	3	295.3 (64.7)	234–363	10	74.9 (88.5)	1–280	16.9	.0002*
Expressive vocab	10	361.6 (26.9)	317–396	3	3 (2.6)	0–5	10	26.3 (42.5)	0–130	16.5	.0003*
Gestures	10	54.1 (8.5)	42–63	3	45.3 (7.6)	40–54	10	24.5 (12.5)	6–43	15.2	.0005*
Consonants	10	19.1 (2)	16–23	3	5 (1)	4–6	10	9.1 (4.3)	2–16	16.9	.0002*
Early 8	10	8 (0)	8	3	5 (1)	4–6	10	5.8 (1.9)	2–8	16.3	.0003*
Middle 8	10	6.9 (1.2)	4–8	3	0		10	2.2 (1.6)	0–5	17.7	.0001*
Late 8	10	4.2 (1.2)	3–7	3	0		10	1.1 (1.2)	0–3	16.4	.0003*
Syllable shape											
V	10	11.2 (3.5)	5.2–17.9	3	57.3 (19.8)	34.5–70.3	10	34 (25.6)	7.8–93.9	11.4	.003*
CV	10	43.5 (9.8)	32.6–56.7	3	39.3 (22.8)	24.1–65.5	10	59.8 (21.5)	6.1–76.6	6.7	.036*
CVC	10	24.7 (4.7)	15.8–31.8	3	0.8 (0.7)	0–1.1	10	4.4 (7.7)	0–19.4	15.7	.0004*
VC	10	8 (3.5)	2.8–13.2	3	2.5 (4.4)	0–7.6	10	1.2 (2.5)	0–7.8	12.9	.002*
CCV	10	1.2 (1.1)	0–3.7	3	0		10	0.2 (0.5)	0–1.7	12.6	.002*
CCVC	10	3.2 (1.7)	0.9–6.9	3	0		10	0.2 (0.4)	0–1.1	17.8	.0001*
CVCC	10	5.1 (4)	0–12.8	3	0		10	0.1 (0.3)	0–0.9	14.9	.0006*
VCC	10	2 (1.8)	0–5.2	3	0		10	0.05 (0.3)	0–0.5	12.5	.002*
CCVCC	10	0.9 (1.1)	0–3.3	3	0		10	0		9.8	.007*
CVCCC	10	0.2 (0.2)	0–0.6	3	0		10	0		4.3	.12
PCC	10	81 (9.1)	66.9–94.4	3	12.5 (8.3)	4.6–21.1	4	42.2 (18.3)	21.7–62.4	12.5	.002*
PVC	10	91.4 (6.3)	82.7–97.1	3	24 (6.7)	17.6–31	4	79.4 (6.1)	71.5–85.7	10.8	.005*
PPC	10	85.2 (7.7)	71.6–95.5	3	17.3 (7)	12.5–25.3	4	58.8 (12.7)	41.9–71.8	12	.003*

Note. The consonants were categorized as Early 8, Middle 8, and Late 8 (Shriberg, 1993). Receptive vocab = number of words understood on MacArthur–Bates Communicative Development Inventory (CDI; Fenson et al., 2007); Expressive vocab = number of words expressed on CDI (Fenson et al., 2007); Gestures = number of gestures used on CDI (Fenson et al., 2007); Consonants = total number of consonants in phoneme repertoire; Syllable shape = proportion of syllable shapes in spontaneous speech sample; V = vowel; C = consonant; PCC = percent consonants correct in spontaneous speech sample; PVC = percent vowels correct in spontaneous speech sample; PPC = percent phonemes correct in spontaneous speech sample.

*$p \leq .05$, Kruskal–Wallis test.

words, they had stronger receptive skills than Cluster C children ($p = .04$). Cluster B children used more gestures than children in Cluster C, although this difference did not reach significance on the Dunn's test ($p = .08$). All three children in this group used a small number of Early 8 consonants only. These children had very low phoneme and vowel accuracy scores, significantly lower than the 10 children in Cluster A (PCC, $p = .0008$; PVC, $p = .001$). Although the children in Cluster B scored lower than the four children in Cluster C with phoneme and vowel accuracy scores, these differences did not reach significance (PCC, $p = .18$; PVC, $p = .13$). As previously described, the children in Cluster B did not differ from children in Cluster A on age, receptive vocabulary, use of gestures, or proportion of CV syllables.

Cluster C: Low Receptive, Low Expressive, Low Gestures, and Low Speech

The children in Cluster C ($n = 10$) differed from the other two clusters by age. They also had significantly different scores than the children in Cluster A on all communication variables. These children presented with lower expressive vocabularies ($p = .0001$), lower receptive vocabularies ($p = < .0001$), and overall less developed speech skills than Cluster A children. The children in Cluster C were more similar to Cluster B children compared to Cluster A children. Cluster C and B children differed on receptive vocabularies ($p = .04$), and although both clusters used predominantly V and CV syllables only, Cluster C children overall used a higher proportion of prevocalic consonants (CV, $p = .02$). Only four (40%) of the Cluster C children produced enough words to complete relational analyses. The phoneme accuracy scores are limited for this cluster of children. The remaining six (60%) children in Cluster C were prelinguistic, producing less than 10 recognizable words.

Additional Findings

One participant appeared to be an outlier. Participant 14's cluster membership changed between Cluster B and Cluster C, depending on the speech variables used. This participant was similar to the children in Cluster B in many ways. Participant 14 had a larger receptive vocabulary than expressive, used a similar number of gestures to the Cluster B children, had only 10 consonants in his sound repertoire, and had low phoneme accuracy. Contrastingly, Participant 14's vowel accuracy was higher than the children in Cluster B. In the final dendrogram, he remained in Cluster C.

Discussion

This is the first study to explore the speech abilities of a heterogeneous cohort of children with ASD aged 2;0–6;11. A detailed independent speech analysis for children with ASD at a variety of levels of functioning was feasible and yielded important information. The detailed speech data reported in this study provided a broader set of speech variables to form descriptive clusters than used in previous research. From the 23 children in this heterogeneous cohort, three distinct clusters emerged: (a) children with high receptive and expressive vocabularies, high nonverbal communication, and high speech; (b) children with high receptive vocabularies, high nonverbal communication, low expressive vocabularies, and low speech; and (c) children with low levels of language, nonverbal communication, and speech. These findings and how they add to our understanding of the speech of children with ASD are discussed.

Describing the Speech of Children With ASD

This study reports detailed speech descriptions of children with ASD, including consonant repertoires, syllable complexity, and phoneme accuracy. This level of detail is needed to link findings of existing and future studies in the field. To date, it has been difficult to draw comparisons between studies due to differences in terminology and a paucity of descriptive data. The study by Schoen et al. (2011) is unique in the detail of reported speech data for children with ASD. Although differences in the ages, linguistic level of some participants, and the method of the Schoen et al. study and this study mean it is difficult to compare results, some similarities are suggested. Overall, children with ASD in both studies followed a typical pattern of consonant acquisition, producing more Early 8, then Middle 8 and Late 8 consonants. Children in prelinguistic or early stages of language development produced very few Late 8 consonants or consonant blends in either study. The current research extends this finding and includes older and more verbal children.

Defining the Clusters

Previous research (Chenausky et al., 2019; Kjelgaard & Tager-Flusberg, 2001; Rapin et al., 2009) has identified subgroups of children with ASD with specific speech profiles. This study aimed to extend this body of research by exploring whether subgroups could be defined based on more detailed speech data in children aged 2;0–6;11. This study was also interested in exploring whether meaningful subgroups emerge from a more heterogeneous cohort, when participants were not preselected based on their speech or language ability. The results suggest that participants in this cohort can be divided into three meaningful clusters.

Cluster A: High Receptive, High Expressive, High Gestures, and High Speech

The 10 children in the Cluster A presented with relatively high language and speech ability. This cluster of children may be similar to subgroups of “average” children reported in existing literature (Kjelgaard & Tager-Flusberg, 2001; Rapin et al., 2009). Although exploring articulatory and phonological errors was out of the scope of this article, it would be interesting to examine these in the future. High rates of mild articulation errors have been reported in high-functioning children with ASD (Cleland et al., 2010; Shriberg et al., 2001, 2011) and cannot be ruled out in this study. Particular consonants (/θ/, /ð/, /l/, /ɹ/, /ʒ/) were absent from the repertoires of some Cluster A children.

Cluster B: High Receptive, Low Expressive, High Gestures, and Low Speech

The children in Cluster B (n = 3) presented with very few words, very few consonants in their phoneme repertoires, and very low phoneme accuracy. However, these children had comparable receptive vocabularies and nonverbal communication to children in Cluster A and were the same age. This profile of higher receptive and nonverbal communication ability compared with speech and expressive language is unexpected. Previous research reports higher expressive language compared with receptive language and fewer gestures in children with ASD (Mitchell et al., 2006; Osterling et al., 2002; Watson et al., 2013). The difference in the communication profile of these three children warrants further investigation. A small subgroup of children with ASD with higher receptive language compared with limited expressive language and poor speech skills has been reported in previous research (Chenausky et al., 2019; Rapin et al., 2009; Velleman et al., 2010), and it is possible that these children may be similar, but given the age and methodological difference between studies, effective comparison is difficult. This is the first study to report a subgroup of children in this age range with this communication profile using a broad set of speech variables.

The underlying communication diagnosis of these three children is unclear, although an SSD could be suspected. Differentially diagnosing an SSD in minimally verbal children is challenging (Chenausky et al., 2019; Strand et al., 2013). Like the children in the Chenausky et al. (2019) study, the three children in Cluster B were unable to complete many of the tasks that would be required for an accurate SSD diagnosis, such as diadochokinesis (repetition of syllables such as “pa,” “ta,” and “ka”), multisyllabic word naming, and speech imitation tasks. This study adds valuable descriptive information regarding the capacity of these children but does not diagnose the presence or absence of specific SSDs.

Cluster C: Low Receptive, Low Expressive, Low Gestures, and Low Speech

The children in Cluster C (n = 10) may be consistent with previous findings reporting a subgroup of children with ASD with low language and low speech ability (Chenausky et al., 2019; Rapin et al., 2009). Six participants in this cluster produced fewer than 10 recognizable words and, therefore, could be classified as prelinguistic. The participants in this cluster scored significantly lower than the other two clusters on measures of receptive vocabulary. These children also had a significantly smaller expressive vocabulary and speech capacity than the Cluster A children. The results of this study support previous findings that children at lower levels of linguistic development generally produce fewer consonants and consonant blends and fewer complex syllable forms (Schoen et al., 2011). Some children in this cluster had more than 10 consonants in their sound repertoire. It would be interesting to follow the development of their speech and to explore whether children with ASD with more advanced speech develop expressive language skills earlier than children with less developed speech, in line with Saul and Norbury (2020).

Limitations

It is important to consider the limitations of this study when interpreting the findings. By design, the sample in this study was heterogeneous in age, developmental level, and language capacity. It would be valuable to follow a similar method with more homogeneous groups of children with ASD to see if similar results emerge. It is possible that more subgroups exist within the heterogeneous cohort than the three outlined in this study. For example, some children in Cluster A reached ceiling level on the CDI vocabulary checklist, which may have obscured additional subgroups in that cluster. Given the relatively small sample and the fact that not all participants were able to complete the full assessment battery, the results need to be viewed with caution. Replicating these findings with a larger cohort of children is important, particularly given the additional subgroups or clusters reported in the previous literature (Chenausky et al., 2019; Rapin et al., 2009; Shriberg et al., 2011).

Spontaneous speech samples proved to be the only consistent speech assessment method that was feasible for all participants; however, this method is not without limitations. Children with ASD have varying vocalization rates (Chenausky et al., 2017), meaning some children require much longer to produce a comparable number of vocalizations or utterances. Speech sample analysis was also limited by the assessor's and independent reliability transcriber's ability to identify the target word. Many participants in this study used a combination of real words, jargon, and echolalia. Within the spontaneous speech samples, it was often difficult to differentiate between these vocalizations and to ascertain the target word. As may be expected, this was less of a concern in the single-word naming task. Furthermore, some children used a large proportion of general words (i.e., "there" and "yeah") or learned scripts (i.e., "open the door"), resulting in samples that may not have represented their language level. Differences in the number of utterances and phonological complexity may make it difficult to draw comparisons between samples (Stoel-Gammon & Williams, 2013) and to compare samples from the same child over time.

Furthermore, it is difficult to interpret the accuracy measures in relation to severity or presence of SSDs in this cohort. Typically, relational speech analyses are compared to age norms. For a heterogeneous cohort of children with ASD, many of whom have linguistic and developmental difficulties, this is not an appropriate approach. Previous studies have compared the speech of minimally verbal children with language-matched comparisons (Schoen et al., 2011) or the speech of high-functioning children with ASD to typically developing or speech disordered peers (Shriberg et al., 2011). Both reported that the speech of verbal children with ASD was on par with their language level. Without a control group for comparison, this study is limited in this regard. Phoneme accuracy measures were only available for the four verbal children in Cluster C. Although this is representative of the linguistic level of these children, it does limit the results and highlights the importance of remaining cautious when interpreting these findings.

Future Directions

This descriptive cohort study highlights a number of interesting areas for future research. First, longitudinal follow-up of the participants would add valuable information regarding the development of speech of children with ASD. Consonant inventory has been reported to be one of the strongest predictors of expressive language development in children with ASD (Wetherby et al., 2007; Yoder et al., 2015). Therefore, it could be hypothesized that the children in this study with few consonants may be expected to remain minimally verbal over time. Longitudinal follow-up of such children is warranted to establish an expected sequence of development. It would also be interesting to ascertain if children with ASD at all levels of linguistic development can complete a speech imitation task and more specific motor speech assessments, such as the Dynamic Evaluation of Motor Speech Skill (Strand & McCauley, 2019). With additional data, further investigation regarding the nature of the speech sound difficulties in children with ASD could be completed. Furthermore, although exploring articulation and phonological errors was out of the scope of this paper, it would be interesting to examine these in the future. High rates of mild articulation errors have been reported in high-functioning children with ASD (Cleland et al., 2010; Shriberg et al., 2001, 2011) and cannot be ruled out in this study.

The results described here may guide future research in this area. The level of detail reported in this study may help identify variables for future studies trying to predict speech outcomes for children with ASD who persistently retain limited verbal output. It is hoped that this small cross-sectional study may precede large population studies of the speech of children with ASD, the type of study required to reach conclusions regarding prevalence, causality, and outcomes.

Clinical Implications

A number of considerations for clinicians emerge from this study. First, when clinicians see a child with limited verbal output, they should not assume that the child has only a language deficit. Children with ASD with limited verbal output may vary significantly in their receptive vocabularies and nonverbal communication abilities. Some children with ASD may present with high receptive vocabulary and high use of gestures in the presence of very low expressive vocabularies. In this instance, a thorough speech assessment is recommended. It is possible that these children have a combination of speech and language difficulties, which can only be described and appropriately targeted for intervention if both speech and language assessments are completed.

Furthermore, it is important that clinicians capture what the child can do using independent speech analyses, rather than only completing standardized speech assessments and focusing on specific speech deficits. For children at very early stages of development, comparing their speech to adult targets may not be possible or informative. When clinicians shift their focus from language to speech and from deficits to strengths, it becomes clear that a speech assessment with children with ASD does not have to include a standardized measure. The speech assessment here included a spontaneous sample of the child's speechlike vocalizations and for some included echolalia. Strengths-based independent speech analyses should include phoneme and syllable shape repertoires. Ideally, a speech assessment would also include single-word naming tasks and sound stimulability; however, these tasks may be impossible for some children with ASD, and this should not result in the exclusion of a speech assessment altogether.

Finally, a detailed baseline of a child's speech capacity, rather than their deficit, is likely to inform intervention and provide measurements to record growth. The results of this study suggest a move toward focusing on each child's unique communication profile, encompassing language, nonverbal communication, and speech capacity, is needed. Clinicians are in a position to begin that shift now.

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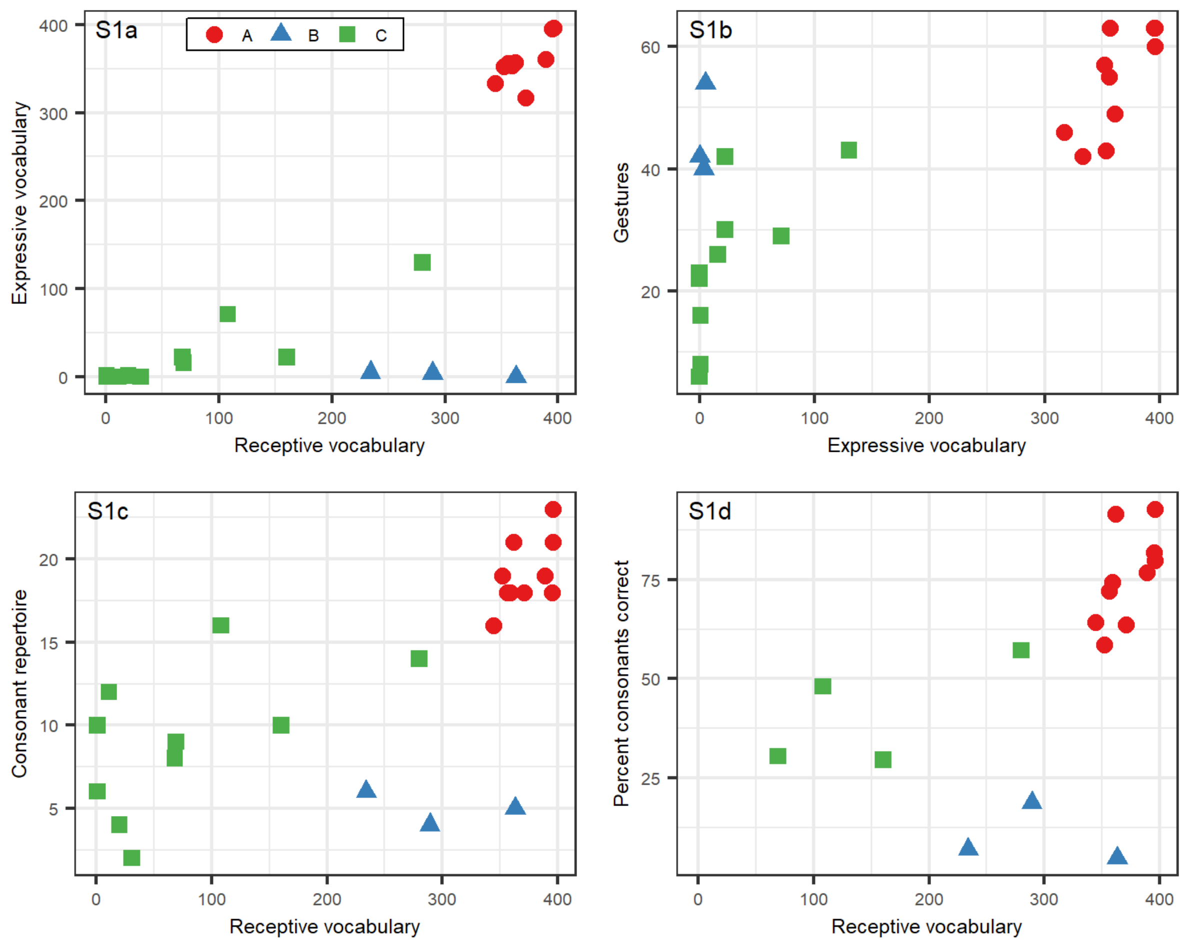
Supplementary Materials

Supplementary Table 3.1. Dunn's test results

Variable	Cluster A and B		Cluster A and C		Cluster B and C	
	z	*p*	z	*p*	z	*p*
Age	-0.2	.43	3.3	.0005*	2.4	.008*
Receptive vocab	1	.15	4.1	<.0001*	1.7	.04*
Expressive vocab	2.8	.002*	3.7	.0001*	-0.3	.37
Gestures	1.2	.11	2.9	<.0001*	1.4	.079
Consonants	3.2	.0006*	3.5	.0003*	-.89	.19
Early 8	3.1	.0009*	3.5	.0003*	-.79	.21
Middle 8	3.6	.0002*	3.3	.0005*	-1.4	.09
Late 8	3.3	.0006*	3.4	.0004*	-.96	.17
Syllables						
V	-3	.0014*	-2.5	.006*	1.3	.098
CV	0.4	.33	-2.2	.013*	-1.96	.025*
CVC	2.4	.008*	3.8	.0001*	0.2	.44
VC	2.1	.02*	3.5	.0003*	0.3	.38
CCV	2.5	.0006*	3.2	.0007*	-0.3	.37
CCVC	2.9	.002*	3.9	.0001*	-0.3	.37
CVCC	2.7	.003*	3.5	.0002*	-0.3	.37
VCC	2.4	.008*	3.2	.0006*	-0.2	.41
CCVCC	2	.02*	2.9	.002*	0	.5
PCC	3.2	.0008*	2.3	.01*	-0.9	.18
PVC	3.1	.001*	1.9	.03*	-1.1	.13
PPC	3.1	.0009*	2.2	.02*	-1	.17

$^*p \leq .05$ Dunn's test

Receptive vocab = number of words understood on CDI (Fenson, et al., 2007); Expressive vocab = number of words expressed on CDI (Fenson, et al., 2007); Gestures = number of gestures used on CDI (Fenson, et al., 2007); consonants = total number of consonants in phoneme repertoire; Early 8, Middle 8, Late 8 consonants (Shriberg, 1993); syllable shapes = proportion of syllable shapes in spontaneous speech sample; V: vowel; C: consonant; PCC: percent consonants correct in spontaneous speech sample; PVC: percent vowels correct in spontaneous speech sample; PPC: percent phonemes correct in spontaneous speech sample



Supplementary Figure 3.1. Scatterplots of descriptive data

Receptive vocabulary: raw score on CDI; Expressive vocabulary: raw score on CDI; Gestures: raw score on CDI; Consonants: number of consonants in phoneme repertoire; PCC: percentage consonants correct in spontaneous speech sample
A = Cluster A: High Receptive, High Expressive, High Gestures, High Speech; B = Cluster B: High Receptive, Low Expressive, High Gestures, Low Speech; C = Cluster C: Low Receptive, Low Expressive, Low Gestures, Low Speech.

Chapter 4: Speech development across subgroups of children with autism spectrum disorder: A longitudinal study

Paper 3: Speech development across subgroups of children with autism spectrum disorder: A longitudinal study

The paper presented in this chapter has been submitted for publication as:

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Author Contribution Statement

As co-author of the above paper and primary supervisor, I confirm that Kate Broome made the following contributions:

- Conception of the research questions in collaboration with co-authors
- Literature reviews
- Data collection
- Data entry and data analysis/interpretation in collaboration with co-authors
- Writing the first draft of the paper with subsequent drafts developed in collaboration with co-authors
- Journal submission

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Abstract

Subgroups of children with different speech profiles have been described however, little is known about the trajectories of speech development or stability of subgroups over time. This longitudinal study described both speech trajectories and subgroup stability of 22 children with ASD, aged 2;0 – 6;11 years, over 12 months. Independent and relational speech analyses, vocabulary size and nonverbal communication were used in clustering. Results suggest varied speech trajectories, particularly for children with ‘low language and low speech’ at Time 1. Receptive vocabulary and consonant inventory at Time 1 may predict speech outcomes after 12 months. A small subgroup of children (n=3) presented with low expressive vocabulary and speech but higher receptive vocabulary and use of gestures. This unique profile remained stable.

Keywords:

Speech; Autism; Child; Longitudinal

Children with autism spectrum disorder (ASD) present with a dyad of social communication difficulties and restricted and repetitive behaviors (American Psychiatric Association, 2013). Although not a core feature, many children with ASD are also diagnosed with language impairments (Levy et al., 2010). As a result, communication research with children with ASD has largely focused on the areas of social communication and language. The speech capacity of children with ASD has, until recently, been largely overlooked. The small body of research examining the speech of children with ASD outlines a few key findings: (1) prelinguistic children with ASD may produce fewer consonants and less canonical babbling than typically developing children (Paul et al., 2011; Plumb & Wetherby, 2013; Schoen et al, 2011); (2) highly verbal children with ASD present with higher rates of delayed and disordered speech (Cleland et al., 2010; Shriberg et al., 2011; Shriberg et al., 2001); and (3) a small subgroup of minimally verbal children with ASD may have a significant co-occurring speech sound disorder (SSD; Broome et al, 2021; Chenausky et al., 2019; Rapin et al., 2009).

Despite this growing body of evidence, little is known about the development of speech in children with ASD. Few longitudinal studies of communication development in children with ASD have included speech variables, and those that have are studies looking at predictors of expressive language in children with ASD who are ‘minimally verbal’-- a term generally accepted as referring to children using less than 20 words and not yet at phrase-level expressive language (Chenausky et al., 2019; Thurm et al., 2015; Yoder et al., 2015). Longitudinal research on this group of children highlights the importance of a child’s early speech capacity to later expressive language ability but adds little information regarding a child’s speech progression. For example, early vocalizations and consonant inventories have been suggested as important predictors of later expressive language ability in minimally verbal children with ASD (see McDaniel et al., 2018 for review). In the most recent study of this kind, Saul and Norbury (2020) aimed to build on findings by Yoder et al. (2015) that reported parental responsiveness, child response to joint attention, child communicative intent and consonant inventory were unique predictors of expressive language growth. Saul and Norbury (2020) examined the expressive language development of 27 minimally verbal children with ASD, aged 2-5 years, over 12 months and used the same predictors as Yoder and colleagues but with an expanded measure of phonetic repertoire. Consonant inventory and phonetic repertoire were found to be significant predictors of expressive language growth. These results highlight the importance of a child's early speech capacity to later expressive language ability, although do not add information regarding a child's speech progression.

There is a paucity of available literature detailing the speech development of children with ASD. Different subgroups of children with ASD based on their speech capacity are beginning to emerge in the literature (Broome et al., 2021; Chenausky et al., 2019; Rapin et al., 2009) but, to date, little is known about the trajectory of speech development for the different subgroups or the stability of these subgroups over time. There are some similarities between studies exploring the possibility of subgroups of children with ASD based on speech skills. Although, given the differences in method, participant characteristics, and terminology used, comparisons across studies need to be made with caution. All three studies (Broome et al., 2021; Chenausky et al., 2019; Rapin et al., 2009) report a subgroup of children with average speech abilities, a subgroup of children with very low speech and language, and one or more subgroups of children with suspected speech sound disorders (SSDs). Rapin et al. (2009) described a subgroup of four (6%) children, aged 7-9 years, with 'profoundly' impaired phonology, stronger receptive language, and average nonverbal IQ. Chenausky et al. (2019) reported two subgroups of children with suspected SSDs in their study of 54 'minimally verbal' and 'low verbal' children with ASD, aged 4;4-18;10 years. One subgroup of 13 (24%) children were suspected of having Childhood Apraxia of Speech (CAS), and a second subgroup of 16 (30%) children were described as having non-CAS speech difficulties. Broome et al. (2021) described a subgroup of 3 (13%) children with low speech and expressive vocabulary but higher receptive language and use of gestures, in a study of the speech capacity of 23 children with ASD, aged 2;0-6;11 years. The differences across studies, make comparisons across subgroups of children with suspected SSDs difficult. Prospective longitudinal speech studies with children with ASD are required in order to document speech development over time both individually and in the subgroups outlined. It is also unknown whether the emergent subgroups remain stable over time or if some children's speech progresses at a different trajectory to other group members.

Defining patterns of speech development is important to further our understanding of the different speech profiles of children with ASD and the speech outcomes for these children. Identifying the barriers to communication for children with ASD informs diagnosis and guides intervention. Some children with ASD may present with a co-occurring SSD which impacts their ability to develop intelligible speech and may require targeted intervention. Therefore, the current prospective longitudinal study aimed to: 1) examine the stability of speech subgroups over 12 months, and 2) describe which variables may explain changes in speech capacity over time.

Method

This study used a prospective longitudinal descriptive design to evaluate the speech development of children with ASD. The research protocol was approved by the Human Research Ethics Committee of The University of Sydney (2012/712) and (2012/1305) and informed consent was obtained from parents on behalf of all participants.

Participants

As described in Broome et al (2021), participants were recruited from an autism early intervention service provider in the greater metropolitan area of Sydney, and from private speech pathologists in the Sydney area who either listed ASD as an area of interest on the Speech Pathology Australia website or who were a member of the ASD evidence-based practice interest group in Sydney, Australia. Parents interested in their children participating in this study contacted the first author initially and were screened for eligibility over the telephone.

Children were eligible to participate if they (a) were aged between 2;0-6;11 years at entry to the study, (b) had a documented diagnosis of ASD in accordance with the Diagnostic and Statistical Manual of Mental Disorders - Fourth Edition Text Revision (American Psychiatric Association, 2000) or Fifth Edition (American Psychiatric Association, 2013), and (c) had a developmental or cognitive assessment or the intention to complete a developmental assessment within the time frame of the broader longitudinal study. Children were excluded from the study if they were: (a) born at less than 36 weeks gestational age, (b) diagnosed with co-morbid developmental disorders other than ASD, or genetic syndromes, or (c) had any uncorrected hearing or visual deficits. All participants underwent an oral motor screening examination with the first author at the initial assessment. Children were excluded from the study if there was asymmetry or weakness of the oral musculature resulting in significant drooling and/or dysarthria. Children were also excluded if there were any oral structural abnormalities (e.g., cleft palate). A total of 22 children (20 males and 2 females; mean Time 1 age of 46 months) participated in the study. All participants were reported to be using speech-like vocalizations and had English as their primary language. A description of participants is provided in Table 1. One participant (4%), Participant 2, was unavailable for follow-up assessment at Time 2, 12 months after initial assessment. A further participant, Participant 10, was recruited however, withdrew from the study prior to the first assessment.

Table 1. Description of participants

Variable	n	*M*	*SD*	Range
Time 1 Age (months)	22	46	14.9	24 – 74
Time 2 Age (months)	22	58.4	15	37 – 86
CSBS				
Time 1 CC	9	54.6	8.3	47 – 70
Time 2 CC	6	51.5	6.3	47 – 64
PLS-4				
Time 1 AC	14	66.1	14.9	50 – 92
Time 2 AC	16	72.7	16.7	50 – 101
Time 1 EC	14	65.6	14.2	50 – 85
Time 2 EC	16	71.3	14.6	50 – 92
GMDS-ER				
Nonverbal DQ	13	59.3	23.4	21.3 – 108.3
Stanford-Binet				
Nonverbal IQ	3	88	14.5	74 – 103
WPPSI-III				
Nonverbal IQ	3	99	20.7	82 - 122
WISC-V				
Nonverbal IQ	1	86		

CSBS CC: Communication Composite sum of scaled scores based on level of functioning; PLS-4 Preschool Language Scale Fourth Edition standard scores: AC: auditory comprehension, EC: expressive communication, TLS: total language score; GMDS-ER Griffiths Mental Development Scale – Extended Revised: DQ: developmental quotient, IQ: intelligence quotient, WPPSI-III: Wechsler Preschool and Primary Scale of Intelligence Third Edition, WISC-V: Wechsler Intelligence Scale for Children Fifth Edition.

Assessment measures

Participants completed a comprehensive communication assessment battery, including direct language and speech measures, spontaneous speech sampling, and parent questionnaires. Assessment measures were completed at both Time 1 and at Time 2, 12 months later and are detailed in Table 2. All assessment sessions were video- and audio-recorded. To capture each participant's optimal communication ability, most assessments were completed over two sessions at the participants' homes (95%). One child was assessed at the on-campus clinic at The University of Sydney (5%). Every effort was made to complete the primary assessment battery with all participants, however if a child was unable to engage with the assessment or reach basal level on an individual assessment an alternative assessment was presented. To develop our understanding of the capacity of these children, participants were not excluded from this study if they were unable to complete one or more primary assessments.

Table 2. Assessments

Variable	Primary assessment	Secondary assessment
Speech		
Single-word naming	SWPT*	FWFST
Speech sample	Spontaneous speech sample	-
Language		
Parent questionnaire	CDI	-
Standardized assessment	PLS-4	CSBS

* POP: Polysyllable Preschool Test (Baker, 2013) at Time 1
CDI: MacArthur-Bates Communicative Development Inventory – Words and Gestures (Fenson, et al., 2007); CSBS: Communication and Symbolic Behavior Scales - Behavior Sample (Wetherby & Prizant, 2002); FWFST: First Words First Sentences Test (Gillham, Boyle, & Smith, 1997); PLS-4: Preschool Language Scale - Fourth Edition (Zimmerman, Steiner, & Pond, 2002); SWPT: Single Word Polysyllable Test (Gozzard, Baker, McCabe, 2013)

Due to the professional expertise of the authors and length of the assessment battery, completing a cognitive assessment was out of the scope of this study. Instead, results from previous cognitive or developmental assessments were obtained. A formal developmental or cognitive assessment was not available for two participants. Available developmental scores on the Griffiths Mental Developmental Scales – Extended Revision (GMDS-ER; Luiz et al., 2004) were available for 13 participants and reported as a developmental quotient (DQ) as many participants scored below the 1st percentile. The DQ was calculated by dividing age equivalent by chronological age (CA) multiplied by 100. A nonverbal developmental quotient (NVDQ) was calculated from the performance scale and the verbal developmental quotient (VDQ) was calculated from the hearing and speech scale. The results of a cognitive assessment were available for seven participants. For these children, the verbal intelligence quotient (VIQ) and nonverbal intelligence quotient (NVIQ) are presented.

Capturing language ability

The Preschool Language Scales – Fourth Edition (PLS-4; Zimmerman et al., 2002) was presented to all participants. The PLS-4 is a standardized language assessment for children from birth to 6;11 years. One participant was 7;2 years at Time 2, when this tool was readministered. While this age is out of the range for the PLS-4, this participant performed at very low language levels, making the PLS-4 an appropriate assessment tool. This participant scored a standardized score of 50 (1%ile) for all scores at Time 1. His Time 2 scores were compared to children between 6;6-6;11 and again he scored 50 (1%ile) for all standard scores. It is considered that this score is reflective of his performance. Sixteen participants completed the PLS-4 at Time 2 and comparative data across time points was collected for 14 participants.

Where it was not possible to achieve a basal level of performance on the PLS-4, the Communication and Symbolic Behavior Scales - Behavior Sample (CSBS; Wetherby & Prizant, 2002) was completed. The CSBS is a standardized assessment that assesses language comprehension and word use, in addition to other important aspects of very early communication development, such as social-affective signaling, nonverbal communication and joint attention. As children were older than the published normative sample, scores reported are based on the child's language stage as recommended in the manual (Wetherby & Prizant, 2002, p. 61).

A parent questionnaire was used to ensure a consistent measure could be used with all participants. The MacArthur-Bates Communicative Development Inventory – Words and Gestures (CDI; Fenson et al., 2007) was completed by all parents at Time 1 and 2. The CDI is a 396-word checklist of a child's receptive and expressive vocabulary, in addition to the use of 18 early gestures (i.e., communicative and games/routines) and 45 later emerging gestures (i.e., actions with objects, pretending to be a parent, imitating adult actions). The Words and Gestures form provides standard scores for children aged 0;8-1;4 years. For the purposes of this study the form was used to tally the participant's vocabularies and only raw data reported. Parents were asked to separately mark their child's words 'understood' and words 'says' instead of 'words understood' and 'words understood and says' as guided on the form. This allowed for separate measures of expressive and receptive vocabulary. The CDI has been used by several research groups as a measure of vocabulary in children with ASD (Charman et al., 2003; Luyster et al., 2007; Stone & Yoder, 2001). Like these authors, we required one instrument that could provide data on all children in our study. We also wanted a measure of nonverbal communication that could be used with all participants.

Capturing speech capacity

A single-word naming task was presented to all participants. Ideally, the assessment tool would assess all phonemes in all word positions, in addition to a polysyllabic word assessment (Broome et al., 2017). However, to reduce the length of the assessment battery to improve participant's compliance, only a polysyllabic assessment was included. A child's ability to produce polysyllable words provides phonological and stress pattern data that may not be apparent from spontaneous speech samples in which a child may choose to use simpler word shapes. At Time 1, participants were presented with the Toddler Polysyllable Test – Second Edition (POP: Baker, 2013), a 20-word task. At Time 2, the Single Word Polysyllable Test (SWPT: Gozzard et al., 2004) was used to expand this data. The SWPT is a 50-word measure, including 19 of the 20 words included in the POP. At Time 2, 10 participants were able to complete the SWPT.

Participants unable to complete the polysyllabic word assessment were presented with the First Words First Sentences Test (FWFST: Gillham et al., 1997). This single-word naming task presents early developing vocabulary as photographs rather than symbolic pictures, making it easier for children at earlier levels of linguistic development to complete. Nine participants completed the FWFST at Time 2.

A spontaneous sample of speech and speech-like utterances was also collected for all participants. Speech-like utterances included babble, jargon and echolalia. Echolalia was included in the spontaneous speech samples as it demonstrated a participant's speech capacity. For children who were verbal, a minimum of 50 utterances were collected during parent-child play lasting at least 10 minutes. If children did not produce many utterances during play with a parent, the spontaneous speech sample was taken from the CSBS recording. Three participants were at a prelinguistic level, defined as producing less than 10 recognizable words (Broome et al., 2017; Stoel-Gammon, 1989) at Time 2. Vocalizations produced concurrently with background noise, such as an adult talking or dog barking, were excluded from the sample. Utterances were categorized as babble if a target word was unable to be identified after analyzing the recording three times.

Data preparation

Broad phonemic transcription was completed on all single-word naming task responses and entered into Phon 3.1 Computer Software (Hedlund et al., 2020). Independent and relational speech analyses were completed.

Independent speech analyses

Consonant inventories. The total number of consonants for each participant was tallied from the single-word naming task and spontaneous speech sample. For prelinguistic participants, the number of consonants was calculated from the entire assessment battery. Consonants were categorized as Early 8, Middle 8 or Late 8 (Shriberg, 1993).

Syllable shapes. Responses on the single-word naming task were analyzed according to syllable shapes. Syllables were those containing a nuclei vowel (V) and possibly one or more pre- or post-vocalic consonants (C). Consonant blends were represented by the number of consonants (C) within the syllable shape. For example, VCC would indicate a syllable with a vowel and post-vocalic consonant blend of two consonants (e.g. 'ink'). The number of different syllable shapes is reported.

Relational speech analyses

Phoneme accuracy. Percent consonants correct (PCC), percent vowels correct (PVC) and percent phonemes correct (PPC; Shriberg & Kwiatkowski, 1982) were calculated from the single-word naming task completed by the 19 verbal participants using Phon (Hedlund et al., 2020).

Reliability

The first author completed broad phonemic transcription for all responses on the single-word naming task and then re-transcribed 23% of the data to check for intra-rater reliability. An independent researcher transcribed 23% of the single-word naming tasks, randomly selected using random.org. Intra-rater reliability was 96.8% and inter-rater reliability was 93.1%.

The first author tallied the total number of different consonants from the entire assessment battery. The first author completed these ratings again for 23% of participants more than 6 months after the initial analysis. An independent postgraduate SLP tallied the total number of consonants for five (23%) participants. Intra-rater reliability was 98.6% and inter-rater reliability was 95.3%.

Data analysis

The primary analysis conducted in this research was a hierarchical cluster analysis (HCA). The process of analysis, from descriptive data to HCA and then to describing subgroups of children is outlined in Figure 1.

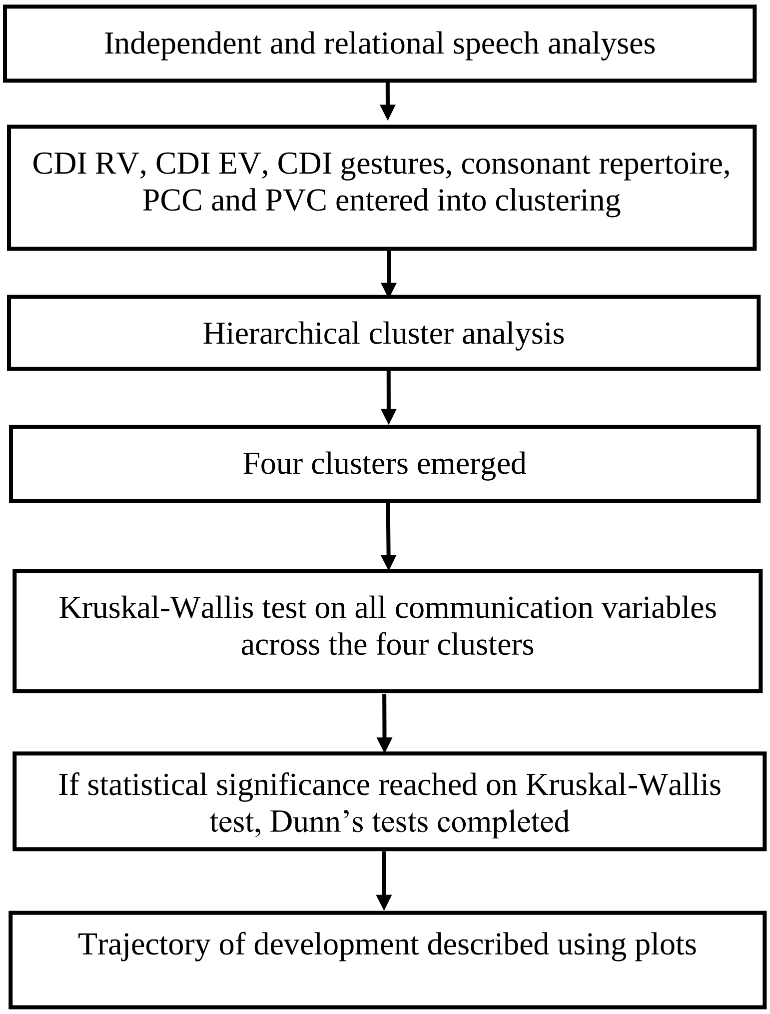

Figure 1. Process of data analysis

CDI RV = receptive vocabulary on CDI (Fenson, et al., 2007); CDI EV = expressive vocabulary on CDI; CDI gestures = number of gestures on CDI; PCC = percent consonants correct; PVC = percent vowels correct

Hierarchical cluster analysis

Agglomerative hierarchical cluster analysis with Euclidean distance (Hastie et al., 2009) was used to explore whether homogeneous subgroups exist within the cohort. In this paper, Time 2 data are analyzed in order to examine stability of subgroups from Time 1 to Time 2. The data derived from clustering is visually presented on a dendrogram, a tree-based representation of the participants. In agglomerative clustering, the dendrogram is built bottom-up. At the bottom of the dendrogram, each participant is initially in their own cluster. Participants join together hierarchically, first joining with those participants most similar, and eventually to the participants most dissimilar. The dissimilarity measure, of which Euclidean is the most common, determines the similarity of two individual participants (James, 2013). Participants most similar join at a low height on the dendrogram. A measure of dissimilarity between sets of data is needed to determine how clusters combine. This is referred to as linkage. In this study, complete linkage was used. Complete linkage, also known as furthest neighbor, defines the difference between two groups of participants as the distance between the two most dissimilar participants in those groups. Participants who merge higher in the dendrogram are less similar than those who fuse at a lower height.

Six Time 2 communication measures were used as clustering variables and entered into R (R Core Team, 2017). These included the CDI receptive vocabulary, CDI expressive vocabulary, CDI number of gestures, consonant repertoire, PCC and PVC. These variables described all aspects of a participant's communication ability, including language, nonverbal communication and independent and dependent speech measures were selected. As variables in this study are measured on different scales, Time 2 data was converted into z scores prior to clustering. This method was used previously to report on Time 1 data (Broome et al., 2021).

The number of clusters is determined by drawing a horizontal line across the dendrogram. Determining the most appropriate level to cut the dendrogram does in part require the researchers to ascertain which solution may best suit the data. For some dendrograms, researchers may explore more than one solution. Once a solution is decided upon, then it can be statistically evaluated to determine if differences between clusters on communication variables reached statistical significance. This was done for all variables in this study through a series of Kruskal-Wallis tests with alpha was set at 0.05. Due to the exploratory nature of this study and given the small n, using a stricter alpha level may result in higher type II errors. Variables that were statistically different on the Kruskal-Wallis test were then subjected to Dunn’s test across clusters. Dunn’s test analysis examined which clusters differed on which variables. This process is outlined in Figure 1.

Trajectories of speech development

Plots were used to visualize the communication profiles of the Time 2 clusters and to illustrate change in speech over 12 months. To plot variables measured on different scales and to visualize change over time, data from Time 1 and Time 2 were converted into z scores collectively. The mean z scores at Time 1 and Time 2 were displayed on separate plots. Comparing these two plots illustrates change over 12 months across the six communication variables for the Time 2 clusters.

Results

Describing the clusters

Agglomerative hierarchical cluster analysis was used to explore whether homogeneous subgroups exist within the cohort of children with ASD based on Time 2 data. Euclidean distance with complete linkage was used. By comparing dendrograms from Time 1 and Time 2 (Figure 2) the stability of cluster membership over 12 months can be analyzed. The dendrogram from Time 1 clustering is included for comparison (with permission from ASHA). This dendrogram produced a 3-cluster solution (Figure 2a).

The Time 2 dendrogram (Figure 2b) illustrates a 4-cluster solution, by horizontally cutting the dendrogram at height 3. Three clusters emerge if you cut the dendrogram higher, with the children in Cluster C and D fusing. Merging high in the dendrogram suggests a less homogeneous subgroup (James, 2013) and so was not explored further.

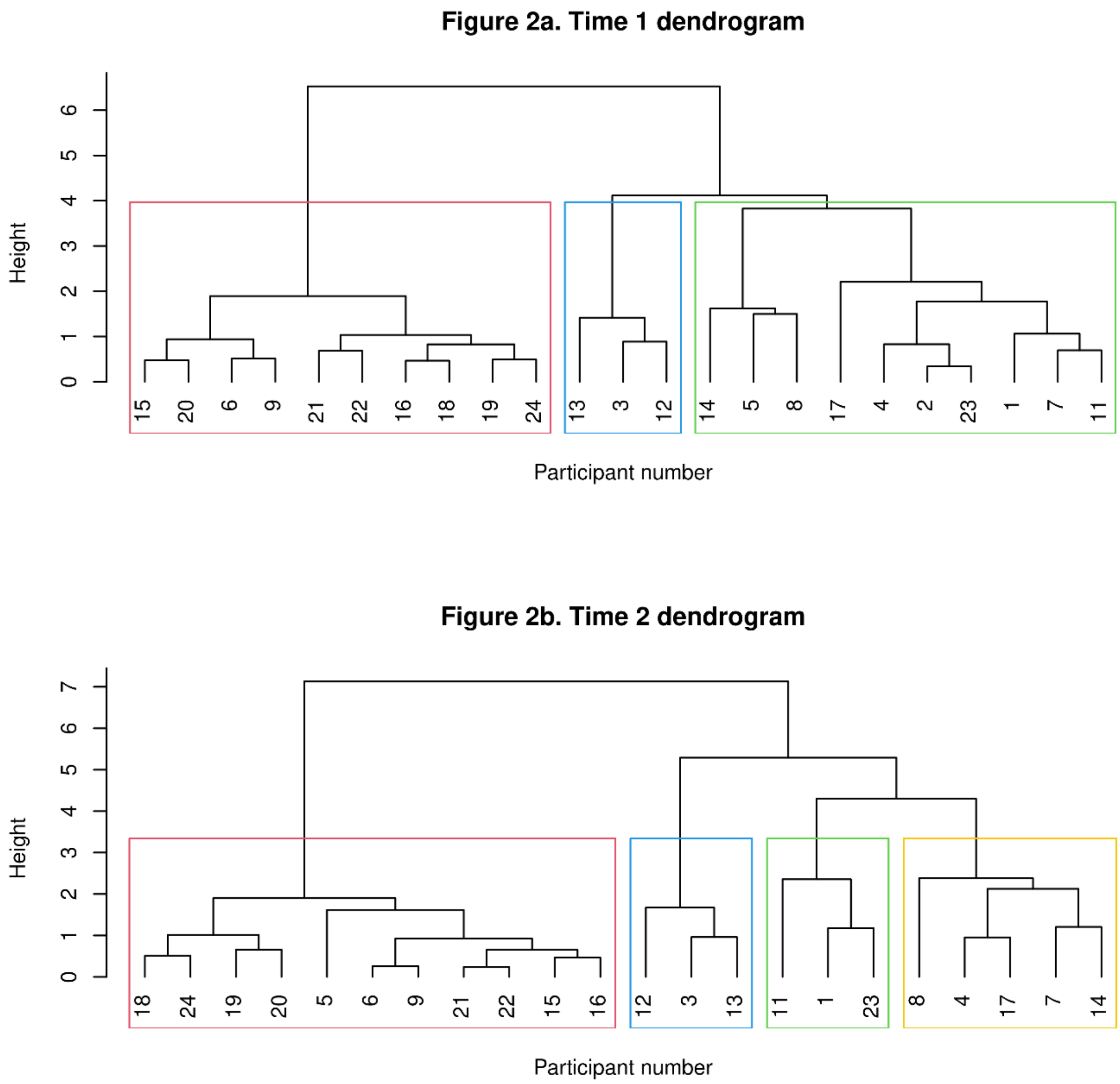

Figure 2. Time 1 and Time 2 dendrograms

Kruskal Wallis tests were completed to statistically analyze if the four clusters differed on communication variables. The results of these tests are presented in Table 3. The PLS-4 Auditory Comprehension scores, proportion of CV syllables and proportion of 3-consonant blends did not reach statistical significance on the Kruskal Wallis test and were not explored further. Dunn's tests were performed for the remaining communication variables to ascertain exactly which clusters differed on which variables. The results from the Dunn's tests are shown in Table 4. The characteristics of each cluster are described below.

Cluster A: High Receptive, High Expressive, High Gestures, High Speech

Children in Cluster A presented with high language, use of gestures and speech capacity. Pairwise comparisons using Dunn's test indicated that the 11 children in Cluster A presented with statistically higher expressive vocabularies than children in any other cluster. Interestingly, their Expressive Communication scores on the PLS-4 did not differ significantly from Cluster D, although only two participants in Cluster D completed the PLS-4. The children in Cluster A also had higher receptive vocabularies than children in Cluster C (p = .0007) and Cluster D (p = .0008), but not statistically different to the three children in Cluster B (p = .16). It is important to note that some children in this cluster reached ceiling level on the CDI and all scored highly. This measure, intended for children at early stages of linguistic development, was not sensitive enough to detect variation within this subgroup.

By contrast, scores from the PLS-4 vary widely. Both the Expressive Communication and the Auditory Comprehension scores range from 50 (floor) to scores within the normal range. The PLS-4 Auditory Comprehension score did not reach significance on the Kruskal Wallis test meaning subgroups did not differ significantly on this score and as such a Dunn's test was not completed.

Cluster A children presented with the strongest speech abilities of any cluster. Pairwise comparisons with Dunn's tests indicated that the children in Cluster A had higher scores on all speech variables compared to Cluster B and Cluster C children. The children in this cluster had the largest consonant repertoires, with all children using at least 19 consonants and some using the complete 24 consonants. The children in Cluster A scored significantly higher than the other three clusters on consonant accuracy. Children in Cluster A and D did not differ on use of Early 8 consonants (p = .28), use of CVC (p = .39), VC (p = .47), CCVC (p = .45) and CVCC (p = .12) syllables, or vowel accuracy scores (p = .057).

Table 3. Characteristics of clusters

Variable	Cluster A			Cluster B			Cluster C			Cluster D			χ^2	*p*
	n	Mean (SD)	Range	n	Mean (SD)	Range	n	Mean (SD)	Range	n	Mean (SD)	Range		
T2 Age	11	64.9 (11.8)	46-80	3	70.7 (18.6)	50-86	3	44 (4.4)	39-47	5	45.2 (7.2)	37-55	11.1	.011*
CDI RV	11	379.9 (20.6)	328-396	3	332.7 (54)	289-393	3	94 (74.6)	13-160	5	169.4 (73.4)	60-244	16	.001*
CDI EV	11	363.3 (48.7)	230-396	3	32.7 (35.5)	6-73	3	3 (3)	0-6	5	103 (68.6)	35-191	17.6	.0005*
Gestures	11	54.5 (6.6)	45-63	3	49 (7.9)	40-55	3	29.7 (8)	22-38	5	42.6 (7.2)	35-52	11.5	.009*
PLS AC	11	77.6 (14.9)	50-101	3	55.7 (9.8)	50-67				2	70.5 (24.7)	53-88	4.2	.12
PLS EC	11	76.4 (12.3)	50-92	3	50 (0)					2	75.5 (2.1)	74-77	6.1	.047*
Consonants	11	21.4 (1.5)	19-24	3	7.3 (1.2)	6-8	3	3.3 (2.5)	1-6	5	15.4 (3.4)	13-21	17.1	.0007*
Early 8	11	8 (0)		3	4.7 (2.1)	3-7	3	2.6 (2.5)	0-5	5	7.8 (0.4)	7-8	18.4	.0004*
Middle 8	11	7.7 (0.5)	7-8	3	1.7 (1.2)	1-3	3	0.3 (0.6)	0-1	5	4.4 (1.7)	3-7	18.2	.0004*
Late 8	11	5.6 (1.1)	4-8	3	1 (1)	0-2	3	0.3 (0.6)	0-1	5	3.2 (1.6)	2-6	15.1	.001*
Syllable shape														
V	11	4.9 (1.5)	2.9-7.9	3	49.5 (30.7)	14.6-72.5	3	62.5 (22)	43.2-86.5	5	9.7 (4.6)	4.7-15.8	15.3	.001*
CV	11	57 (11.4)	22.9-61.8	3	45.6 (26.1)	27.5-75.6	3	37.2 (21.9)	13.5-56.8	5	52.9 (16.8)	31.6-72.5	3.4	.33
CVC	11	21.5 (6.2)	17.2-40	3	4.8 (4.9)	0-9.8	3	0.3 (0.5)	0-0.9	5	25 (10.7)	10.1-34.2	12.7	.005*
VC	11	3.9 (1.2)	2-5.7	3	0		3	0		5	6 (5.6)	2.2-15.8	12.8	.005*
CCV	11	6.4 (2.5)	0-9.8	3	0		3	0		5	2.2 (2.1)	0-4.7	14.1	.003*
CCVC	11	2.4 (3)	0.7-11.4	3	0		3	0		5	2.5 (2.6)	0-6.3	10.1	.018*
CVCC	11	3.6 (3.6)	2.5-14.3	3	0		3	0		5	1.3 (1.8)	0-3.4	10.6	.014*
CCVCC	11	0.3 (0.9)	0-2.9	3	0		3	0		5	0.2 (0.5)	0-1.1	1.1	.77
CVCCC	11	0		3	0		3	0		5	1.1		3.4	.33
PCC	11	84.9 (8.7)	71.4-95.3	3	13.3 (9.3)	2.7-19.7				5	54.9 (17.2)	42.9-84.2	12.3	.002*
PVC	11	94.4 (3.7)	88.2-99.4	3	22.3 (3.4)	18.9-25.6				5	82.4 (13.2)	68.7-100	9.7	.008*
PPC	11	89 (6.4)	78.6-96.8	3	16.6 (6.3)	9.7-21.9				5	65 (15.1)	53.2-90.2	12.3	.002*

*$p \leq .05$ Kruskal-Wallis test

T2 Age = age in months at Time 2; CDI RV = number of words understood on CDI (Fenson, et al., 2007); CDI EV = number of words expressed on CDI (Fenson, et al., 2007); Gestures = number of gestures used on CDI (Fenson, et al., 2007); PLS AC = Preschool Language Scale – Fourth Edition Auditory Comprehension score; PLS EC = Preschool Language Scale – Fourth Edition Expressive Communication score; Consonants = total number of consonants in phoneme repertoire; Early 8, Middle 8, Late 8 consonants (Shriberg, 1993); Syllable shapes = proportion of syllable shapes in speech sample; V: vowel; C: consonant; PCC: percent consonants correct in speech sample; PVC: percent vowels correct in speech sample; PPC: percent phonemes correct in speech sample

Table 4. Multiple pairwise comparisons using Dunn's test

Variable	Cluster A and B		Cluster A and C		Cluster A and D		Cluster B and C		Cluster B and D		Cluster C and D	
	z	*p*	z	*p*	z	*p*	z	*p*	z	*p*	z	*p*
T2 Age	-0.52	.3	2.3	.011*	2.5	.007*	2.2	.013*	2.3	.011*	-0.23	.41
CDI RV	1	.16	3.2	.0007*	3.2	.0008*	1.7	.041*	1.4	.076	-0.5	.31
CDI EV	2.6	.004*	3.5	.0002*	2.5	.007*	0.7	.24	-0.5	.29	-1.3	.09
Gestures	0.8	.2	3	.0012*	2.2	.013*	1.8	.039*	0.9	.18	-1.1	.14
PLS EC	2.5	.007*			0.4	.36			-1.5	.073		
Consonants	2.8	.002*	3.5	.0003*	2	.022*	0.5	.31	-1	.15	-1.6	.05
Early 8	3.1	.001*	3.5	.0002*	0.6	.28	0.3	.37	-2.3	.011*	-2.7	.004*
Middle 8	2.9	.002*	3.5	.0002*	2.3	.011*	0.5	.3	-0.9	.19	-1.5	.07
Late 8	2.9	.002*	3.3	.0006*	1.8	.036*	0.3	.37	-1.2	.11	-1.6	.06
Syllables												
V	-2.9	.002*	-3.1	.001*	-1.8	.036*	-0.1	.45	1.3	.099	1.4	.077
CVC	2.3	.01*	2.7	.003*	-0.3	.39	0.3	.38	-2.3	.011*	-2.6	.004*
VC	2.6	.005*	2.6	.005*	-0.1	.47	0	0.5	-2.4	.009*	-2.4	.009*
CCV	2.8	.003*	2.8	.003*	2.3	.01*	0	0.5	-0.8	.22	-0.8	.22
CCVC	2.4	.009*	2.4	.009*	0.1	.45	0	0.5	-2	.022*	-2	.022*
CVCC	2.5	.006*	2.5	.006*	1.2	.12	0	0.5	-1.4	.08	-1.4	.08
PCC	3.2	.0007*			2.3	.012*			-1.2	.12		
PVC	3	.001*			1.6	.057			-1.5	.067		
PPC	3.2	.0007*			2.3	.012*			-1.2	.12		

$*p \leq .05$ Dunn's test

CDI RV = number of words understood on CDI (Fenson, et al., 2007); CDI EV = number of words expressed on CDI (Fenson, et al., 2007); Gestures = number of gestures used on CDI (Fenson, et al., 2007); Consonants = total number of consonants in phoneme repertoire; Early 8, Middle 8, Late 8 consonants (Shriberg, 1993); Syllable = proportion of syllable shapes in spontaneous speech sample; V: vowel; C: consonant; PCC = percent consonants correct; PVC = percent vowels correct; PPC = percent phonemes correct

Cluster B: High Receptive, Low Expressive, High Gestures, Low Speech

Children in Cluster B did not differ on the Dunn's test to children in Cluster A on measures of age (p = .3), receptive vocabulary (p = .16) and use of gestures (p = .2). The three children in Cluster B differed from Cluster A on all speech variables. Cluster B children's speech and expressive vocabularies were similar to children in Cluster C. Pairwise comparisons indicate that children in Cluster D differed from Cluster B children on age (p = .011), number of Early 8 consonants (p = .011) and use of post-vocalic consonants (CVC: p = .011; VC: p = .009).

Cluster C: Low Language, Low Gestures, Low Speech

The three children in Cluster C had the lowest levels of language, nonverbal communication and speech capacity of any cluster. These children could be described as prelinguistic, all producing less than 6 recognizable words. Children in this cluster were unable to complete the PLS-4, and speech accuracy scores were unable to be calculated. Dunn's comparisons indicate that children in Cluster C differed from Cluster A children on age and all communication measures. Their speech and expressive vocabularies did not statistically differ to Cluster B children, although their receptive vocabularies (p = .041) and use of gestures (p = .039) were lower. Children in Clusters C and D did not differ on age, vocabularies or use of gestures. Their speech skills did differ, however, with children in Cluster D using more Early 8 consonants (p = .004) and post-vocalic consonants (CVC: p = .004; VC: p = .009).

Cluster D: Low Language, Low Gestures, Developing Speech

The five children in Cluster D were comparable to Cluster C children on age and although their receptive and expressive vocabularies were larger than the Cluster C children, these differences did not reach significance on pairwise comparison using Dunn's test. Interestingly, the two children in Cluster D who were able to complete the PLS-4 scored similarly to Cluster A children on Auditory Comprehension and Expressive Communication. Cluster D children differed from Cluster B and C on many speech variables, such as number of Early 8 consonants and use of post-vocalic consonants. Their consonant accuracy was higher than the Cluster B children but lower than Cluster A. The accuracy of vowel production was similar to Cluster A children (p = .057).

Changes in speech capacity

The exact change across communication variables for each participant is reported in Table 5. The 11 participants in Cluster A from Time 2 include all ten children from the Time 1 Cluster A and participant 5. Participant 5 is the last child to merge with this cluster, as depicted by fusion at a higher level on the Time 2 dendrogram (Figure 2b). Participant 5 was included in Cluster C at Time 1, a cluster with low language and low speech ability. Cluster B remained stable from Time 1 to Time 2. This cluster includes three participants. Cluster C from Time 1 splits in two and forms Time 2 Cluster C (n=3) and Cluster D (n=5).

Communication profiles and trajectories of development

The communication profiles of the four clusters were plotted based on Time 1 and Time 2 scores for the six clustering variables (Figure 3). Differences in the Time 1 and Time 2 plots provides information regarding change in abilities and highlights possible predictor variables that may explain why some children in the cohort developed speech along a different trajectory to others.

Table 5. Participants' Time 2 results on communication measures and exact change over 12 months

Variable	Participants																				
	1	3	4	5	6	7	8	9	11	12	13	14	15	16	17	18	19	20	21	22	23	24
T2 Age	46	86	42	46	67	37	50	68	47	76	50	42	76	67	55	75	50	75	55	54	39	80
T2 CDI raw scores																						
RV	109	393	60	328	396	204	207	396	13	289	316	244	396	393	132	373	386	396	370	368	160	377
change	*78*	*30*	*59*	*48*	*1*	*203*	*99*	*0*	*7*	*0*	*82*	*84*	*0*	*4*	*121*	*14*	*42*	*34*	*14*	*16*	*92*	*6*
EV	0	73	35	230	396	191	161	396	3	6	19	70	396	383	58	359	383	392	367	364	6	330
change	*0*	*73*	*35*	*100*	*1*	*190*	*90*	*0*	*2*	*2*	*14*	*48*	*0*	*22*	*58*	*6*	*50*	*35*	*21*	*12*	*-16*	*13*
Gestures	29	55	37	55	63	48	14	63	22	40	52	52	57	54	35	45	51	46	60	59	38	47
change	*7*	*13*	*14*	*12*	*0*	*32*	*12*	*0*	*14*	*0*	*-2*	*10*	*3*	*5*	*29*	*3*	*9*	*0*	*5*	*2*	*8*	*1*
T2 Independent analyses																						
Consonants	1	8	13	20	21	13	21	22	6	8	6	14	24	22	16	19	19	22	22	22	3	22
change	*-1*	*3*	*3*	*6*	*3*	*7*	*5*	*1*	*2*	*4*	*0*	*4*	*1*	*3*	*4*	*1*	*3*	*1*	*4*	*3*	*-5*	*4*
Early 8	0	3	8	8	8	7	8	8	5	7	4	8	8	8	8	8	8	8	8	8	3	8
Middle	1	3	3	7	8	3	7	8	0	1	1	4	8	8	5	7	7	8	8	8	0	8
Late 8	0	2	2	5	5	3	6	6	1	0	1	2	8	6	3	4	4	6	6	6	0	6
Syllable shape																						
V	43.2	61.5	10.9	7.9	3.7	4.7	5.6	4.2	57.8	14.6	72.5	11.6	4.3	4.3	15.8	4.7	2.9	4.3	4.9	4.9	86.5	7.6
CV	56.8	33.8	67.4	58.9	59.9	46.7	46.1	62	41.4	75.6	27.5	72.5	56.4	60.7	31.6	61.7	22.9	60.9	60.5	61.1	13.5	61.8
CVC	0	4.6	17.4	21.2	21	29.7	33.7	18.7	0.9	9.8	0	10.1	19.6	19	34.2	20.1	40	19.3	20.4	19.8	0	17.2
VC	0	0	4.3	2	3.1	4.7	2.2	3	0	0	0	2.9	5.5	5.5	15.8	4	5.7	3.7	4.3	3.1	0	3.2
CCV	0	0	0	5.3	8.6	4.7	3.4	7.8	0	0	0	2.9	9.8	6.1	0	6	0	7.5	6.2	6.2	0	6.4
CCVC	0	0	0	0.7	1.2	6.3	3.4	1.8	0	0	0	0	1.8	1.8	2.6	0.7	11.4	1.9	1.2	2.5	0	1.3
CVCC	0	0	0	2.6	2.5	3.1	3.4	2.4	0	0	0	0	2.5	2.5	0	2.7	14.3	2.5	2.5	2.5	0	2.5
CCVCC	0	0	0	0	0	0	1.1	0	0	0	0	0	0	0	0	0	2.9	0	0	0	0	0
CVCCC	0	0	0	0	0	0	1.1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
T2 Relational speech analyses																						
PCC		17.6	42.9	71.4	88.4	55.9	84.2	92.7		19.7	2.7	44.5	93.7	86.8	46.8	72.1	91.3	95.3	85.6	81.1		75.6
change		*12.7*	*-*	*14.3*	*6.7*	*-*	*36.1*	*13*		*1*	*-4.3*	*14.9*	*1*	*10*	*-*	*-2.3*	*27*	*3.8*	*13.5*	*22.6*		*12.1*
PVC		18.9	85.4	88.2	95.1	88.1	100	99.4		25.6	22.5	68.7	95.8	97	69.8	89.1	94.3	98.8	96.9	92.7		91.5
change		*8.4*	*-*	*6.1*	*4.1*	*-*	*10.6*	*2.7*		*0.6*	*-13*	*0.1*	*-4.2*	*4.5*	*-*	*1*	*2.9*	*0.3*	*-0.3*	*7.6*		*6.4*
PPC		18.2	59.2	78.6	91.3	67.6	90.2	95.6		21.9	9.7	53.2	94.6	91.4	54.9	79.3	92.3	96.8	90.5	86		82.4
change		*10.8*	*-*	*12.8*	*5.4*	*-*	*26.1*	*8.2*		*0.6*	*-8.4*	*6.4*	*-1.4*	*7.5*	*-*	*-1.2*	*28.5*	*2.2*	*9.1*	*15.5*		*9.4*

T2: time two, 12 months after T1; Age in months; RV = receptive vocabulary taken from CDI at T2; EV = expressive vocabulary taken from CDI at T2; change: change between T1 and T2 (T2-T1); Consonants is number of consonants in phoneme repertoire; Early, Middle, Late 8 (Shriberg, 1993); syllable shape = proportion of each syllable type; V: vowel; C: consonant; PCC: percent consonants correct; PVC: percent vowels correct; PPC: percent phonemes correct

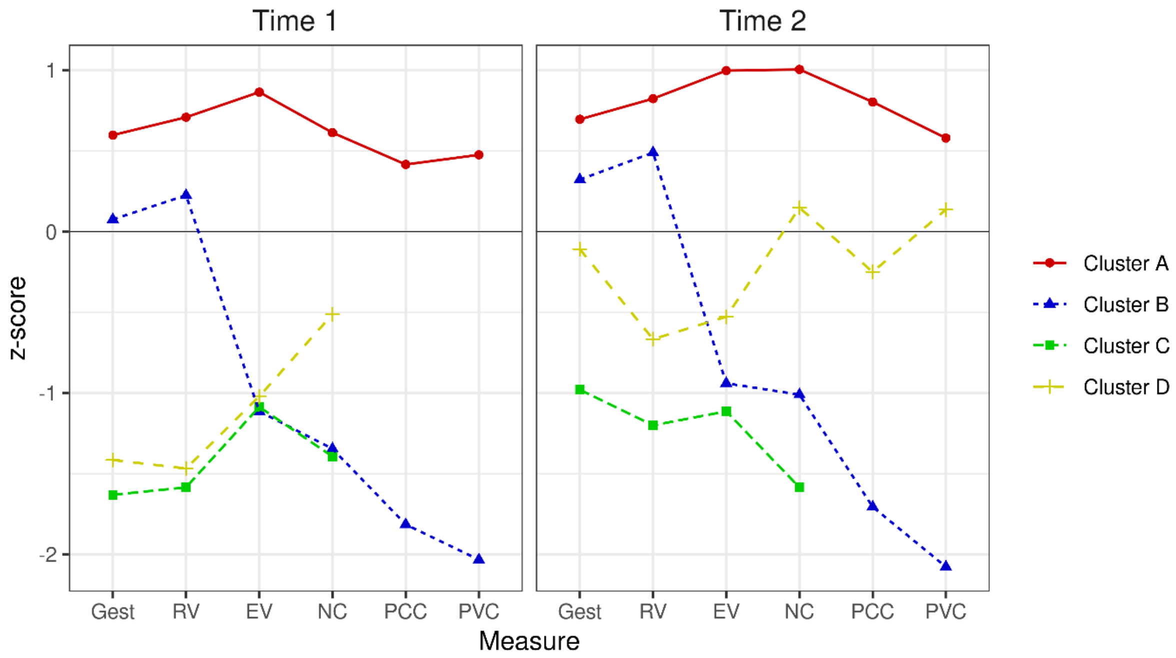

Figure 3. Communication profiles of the four clusters at Time 1 and Time 2.

Gest = number of gestures on CDI (Fenson, et al., 2007), RV = receptive vocabulary on CDI, EV = expressive vocabulary on CDI, NC = number of consonants in consonant inventory, PCC = percent consonants correct, PVC = percent vowels correct.

Cluster A

Cluster A children scored above the mean for the cohort on all communication variables at Time 1 and Time 2. Small improvements in their expressive vocabularies, consonant repertoires and consonant accuracy can be seen across 12 months. It is important to note, that these children were at or close to ceiling levels on the CDI and consonant repertoire measures. One participant (participant 5) moved from Cluster C (low verbal, low gestures, low speech) at Time 1 to Cluster A (high language, high gestures, high speech) at Time 2. A plot of his individual change demonstrates a large improvement across all communication variables (Supplementary Material). Participant 5's expressive vocabulary, gestures, consonant repertoire, and PCC went from below mean compared to the cohort at Time 1 to above mean at Time 2.

Cluster B

The three children in Cluster B presented with a unique communication profile characterized by high gestures and receptive vocabulary, with very low expressive vocabulary and speech capacity. This profile remained consistent over time. There was very limited improvement in the communication ability of Cluster B children over 12 months. Their vowel accuracy was lower at Time 2 compared to Time 1.

Cluster C

Cluster C and D children were combined into one cluster (Cluster C) at Time 1. At Time 1, these children were described as prelinguistic or minimally verbal. Visually, at Time 1, the children who ended up in Cluster C and D at Time 2 appear similar on measures of gestures, receptive vocabulary, and expressive vocabulary. The three children who ended up in Cluster C at Time 2 used fewer consonants at Time 1 than the children in Cluster D at Time 2. Over the 12 months, the children in Cluster C improved slightly in their use of gestures and receptive vocabulary but had no change in their expressive vocabularies and remained at the prelinguistic stage. The Time 1 dendrogram (Figure 2a) shows two of the children who ended up in Cluster C at Time 2, participant 1 and 7, fused to form at cluster low in the dendrogram illustrating similarity at this time. Participant 23 was different and did not fuse to participant 1 and 7 until much higher in the Time 1 dendrogram. Looking at the individual communication profile of participant 23, this child improved their use of gestures and receptive vocabulary over 12 months but regressed in their expressive vocabulary and use of consonants (Supplementary Material).

Cluster D

The five children in Cluster D at Time 2 presented as prelinguistic or minimally verbal at Time 1. These children had comparable use of gestures and receptive and expressive vocabularies but used more consonants than the Cluster C children at Time 1. Time 2 plot

(Figure 2b) shows the children in Cluster D had a very different trajectory of communication development over 12 months. The five children in Cluster D, those using more consonants at Time 1, improved significantly more than those in Cluster C. These children went from below mean to near mean on all communication measures. The children in Cluster D also became verbal in these 12 months, as can be inferred from the inclusion of consonant and vowel accuracy measures at Time 2. Once these children began using words, their consonant and vowel accuracy was at mean compared to the participants in this study.

Discussion

This is the first longitudinal study to describe different trajectories of speech development for subgroups of children with ASD. Subgroups were formed using the same set of detailed communication variables used in Time 1 clustering (Broome et al., 2021) so the stability of these subgroups over 12 months could be described. Results suggest varied trajectories of speech development particularly for the children with 'low language and low speech' at Time 1. Some children who presented with limited language and speech capacity at Time 1 improved across all communication variables over 12 months and were talking at Time 2. Other children in this subgroup remained nonverbal. A child's consonant inventory at Time 1 may predict speech outcomes after 12 months.

Stability of speech subgroups of children with ASD

Previous research (Broome et al., 2021; Chenausky et al., 2019; Kjelgaard & Tager-Flusberg, 2001; Rapin et al., 2009) has described subgroups of children with ASD with specific speech profiles but, until now, no study has investigated the stability of these subgroups over time. Although cross-sectional data provides a snapshot of a child's speech capacity, longitudinal data is needed to better understand the likely speech outcomes for children with ASD. Results of this study suggest membership of two subgroups from Time 1 remains stable, but the children in the initial third subgroup have varied outcomes.

The ten children in Cluster A at Time 1 all remained in Cluster A at Time 2. These children presented with relatively high language, use of gestures and speech capacity. Overall, as the children in this subgroup were at or near ceiling on many of the communication variables in this study, few improvements were recorded. There was slight improvement in their consonant inventories and PCC scores over 12 months. These children are possibly indicative of the ‘average’ speech subgroup of children with ASD previously identified (Chenausky et al., 2019; Kjelgaard & Tager-Flusberg, 2001; Rapin et al., 2009) and may indeed follow the speech trajectory of typical developing children already outlined in the literature. Past research reports high rates of mild articulation errors in older highly verbal children (Cleland et al., 2010; Shriberg et al., 2011; Shriberg et al., 2001). This study did not explore the presence or absence of mild articulation errors. It is therefore possible that some children in Cluster A present with mild speech errors, which would be interesting to investigate in future research.

The three children in Cluster B at Time 1 remained in Cluster B at Time 2. Cluster B children presented with a unique communication profile of high receptive vocabulary and use of gestures, but low speech and low expressive vocabulary. The communication ability of the three children in this subgroup showed very little change and the unique communication profile remained constant over 12 months. The etiology of both their speech and expressive vocabulary deficits needs to be explored further. All three children used effective augmentative and alternative communication (i.e., communication devices, Key Word Signs) suggesting their lack of verbal communication was unrelated to low communicative intent, a sentiment which echoes Saul and Norbury’s (2020) report. It is likely that Cluster B children present with a co-occurring SSD, characterized by a severely limited consonant repertoire and very low consonant and vowel accuracy. Differentially diagnosing a speech sound disorder in minimally verbal children is difficult (Chenausky et al., 2019; Strand et al., 2013).

This study expands on the previous descriptive study of the speech capacity of these children (Broome et al., 2021) and adds valuable information regarding the trajectory of their speech development. Further information regarding imitated verses spontaneous speech, sequencing of speech sounds, vowel repertoire, and consistency of production would add vital information needed to differentiate the specific speech sound disorder in this subgroup.

The nine children with available longitudinal data from Time 1 Cluster C (low language, low speech) had three different communication trajectories at Time 2. This suggests that, particularly for children at very low levels of speech and language development, there is a need to be cautious when predicting communication outcomes. While two children may appear very similar at a given point in time, they could have vastly different trajectories of speech development. In the current study, all nine children in this cluster were described as having low language and low speech at Time 1. After 12 months, one participant moved to the high speech, high language subgroup (Cluster A), possibly due to underperforming at Time 1 assessment. The eight remaining children split to form two clusters at Time 2, Cluster C and D, with vastly different communication outcomes. Three participants remained at the prelinguistic stage of linguistic development at Time 2 (Cluster C), with expressive vocabularies of fewer than six words and low speech capacity. Five children originally with low language and low speech formed Cluster D at Time 2. These children presented with the most communication growth of any subgroup. The 12-month trajectory of this subgroup, from minimally verbal to verbal, offers valuable information and hope to parents, clinicians and researchers. The results suggest there may be the potential to identify the children likely to have better communication outcomes.

Possible predictor variables for speech outcomes

Children in Cluster C and D at Time 2 differed only in the number of consonants in their sound repertoire at Time 1. This preliminary result adds support to prior research reporting consonant inventories as one of the strongest predictors of expressive language development in children with ASD (Saul & Norbury, 2020; Wetherby et al., 2007; Yoder et al., 2015). Children in this study with few consonants at Time 1 remained minimally verbal at Time 2.

No single communication variable could differentiate the four subgroups at Time 1. For example, Cluster B children and Time 2 Cluster C children had comparable consonant inventories at Time 1. Instead, a combination of a child's receptive vocabulary and consonant inventory has the potential to describe their communication profile. These preliminary descriptive results may guide future research aiming to predict speech outcomes for children with ASD. Large population studies of the speech of children with ASD are required to reach conclusions regarding prevalence, causality, and outcomes.

Communication regression

An unexpected finding in this study was communication regression. Regression in children with ASD has been reported in the literature for decades (Lord et al., 2004; Ozonoff et al., 2011, Shumway et al., 2011). Language is reported to be most frequently affected, encompassing loss of babbling, words, and word combinations (Barger et al., 2013; Borterberg et al., 2019). In a meta-analytic review of the literature, the mean age of regression in children with ASD was reported to be 21.4 months (Barger et al., 2013). In this study, participants were at least 2;0 years so regression was not expected. For some children, regression of speech capacity may in fact reflect their attempts at more complex word forms (i.e. consonant blends, polysyllabic words) rather than a true regression of skill. This would be reflected in reduced consonant and vowel accuracy, rather than a regression in consonant inventory or expressive vocabulary. This is likely true for the Cluster A children who regressed in phoneme accuracy (participants 13, 15, 18 and 21). One participant in this study regressed in speech and expressive language between 27 months (Time 1 age) and 39 months

(Time 2 age). Although older than the average age for language regression reported by Barger et al. (2013), this age falls within the 6-36 month range provided by Luyster et al. (2007). Few prospective longitudinal studies of children with ASD have captured regression (Boterberg et al., 2019) and this unexpected finding adds information to this body of research.

Limitations

Consideration should be given to the limitations of this study when interpreting the findings. Firstly, the small sample size in this study limited the statistical approaches that could be applied to the longitudinal data. As each subgroup had a small number of children, communication profiles and trajectories were represented visually. Expanding this preliminary data with larger cohorts of children is important. With a larger cohort of children, additional subgroups may emerge, subgroups that exist within the larger population.

Secondly, a number of children in Cluster A reached near or at ceiling on the CDI. As a result, growth in receptive and expressive vocabulary and use of gestures was unable to be ascertained for these children using this measure. It is possible that more subgroups exist within the high language, high speech subgroup. Larger studies using more sensitive assessment measures for children at this level of functioning are needed to explore this possibility and describe this cluster in more detail.

Finally, this study recruited a heterogeneous cohort of children. The variation in age, level of functioning and communication capacity made it difficult to select assessment tools appropriate for the whole cohort. Some participants were unable to complete the standardized language assessment (PLS-4), limiting the use of this measure in clustering. Spontaneous speech samples, collected for every participant, differed in length and phonological complexity of words. The variations between samples resulted in a limited ability to compare samples within the cohort (Stoel-Gammon & Williams, 2013) and over time for each child.

For these reasons, speech data was based on single-word naming tasks in this study. Different single-word naming measures were required to capture each participant's optimal speech capacity. Given these inconsistencies, it is important to interpret the results in this study with caution. Replicating these results with more homogeneous cohorts using consistent assessment measures is important.

Future research directions

This is the first prospective longitudinal study to detail the speech development of children with ASD. It is hoped that this preliminary data paves the way for future research in this area. Large population studies are needed to explore speech trends, trajectories, and outcomes for children with ASD. Prospective measurement of consonant inventory and receptive vocabulary in a large number of children is required to confirm prediction of speech outcomes in this population using these measures.

Future research aiming to differentially diagnosing SSDs in children with ASD is needed. It remains unknown if children with ASD can complete a speech imitation task, such as the Dynamic Evaluation of Motor Speech Skill (DEMSS; Strand & McCauley, 2019). Further information regarding their imitated compared with their spontaneous speech, sequencing of speech sounds, vowel repertoires, and consistency of productions is required to differentiate the specific speech sound disorders in this subgroup. Some children with ASD and a co-occurring SSD may require targeted speech intervention. Some researchers have begun to investigate the outcomes of speech-based intervention with these children (Beiting & Maas, 2020; Chenausky et al., 2018), and this is an important avenue for further research.

Clinical implications

Clinicians are frequently asked by parents of children with ASD, particularly those children who are less verbal, of the probable prognosis for their child's communication. Parents want to know if their children will ever talk. Although there is some literature to guide clinicians regarding the possible language outcomes (see Brignell et al., 2018 for review), there is a large gap in the literature to inform of the expected prognosis of children with ASD and a suspected co-occurring SSD. Until the body of literature detailing the trajectories of speech development grows, clinicians will be unable to provide parents with an informed response. Results from this study suggest that for minimally verbal children with ASD and a suspected SSD, very little development in speech or expressive language occurs over 12 months. Given this limited progress, clinicians should consider targeted speech interventions for these children. Some minimally verbal children with ASD can make significant improvements in their speech and expressive language in 12 months, and it appears that their early consonant inventory and receptive vocabulary may be important predictors to this growth. Clinicians should be assessing the consonant inventory and receptive language capacity of children with ASD and developing a child's capacity in these areas through intervention.

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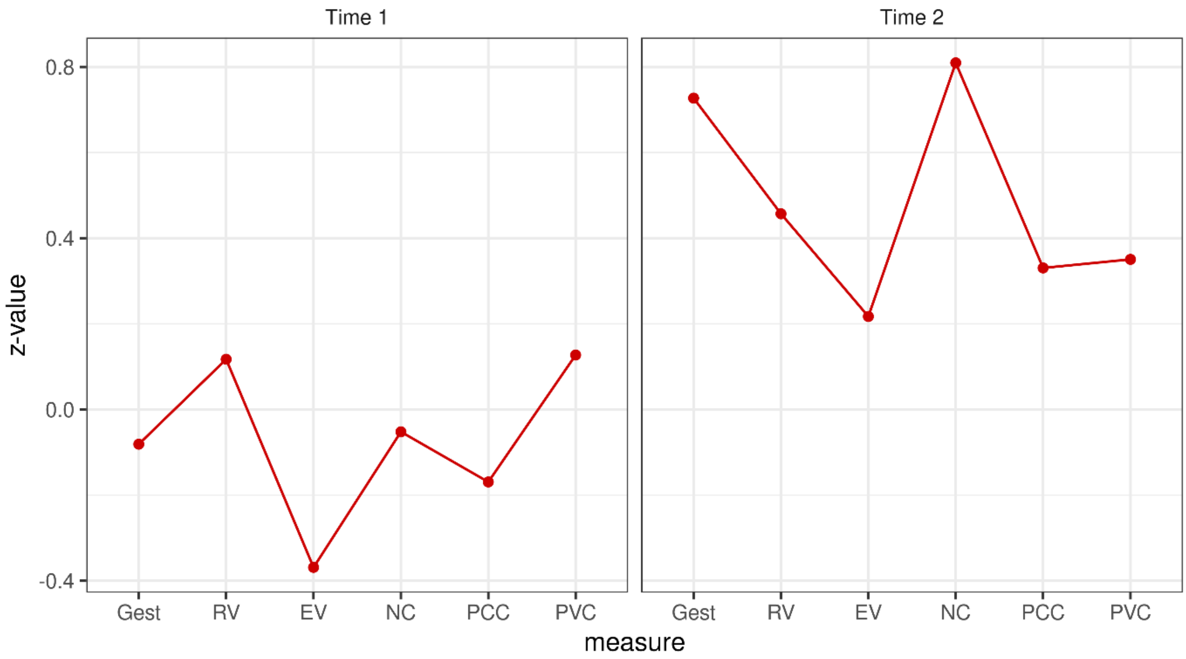
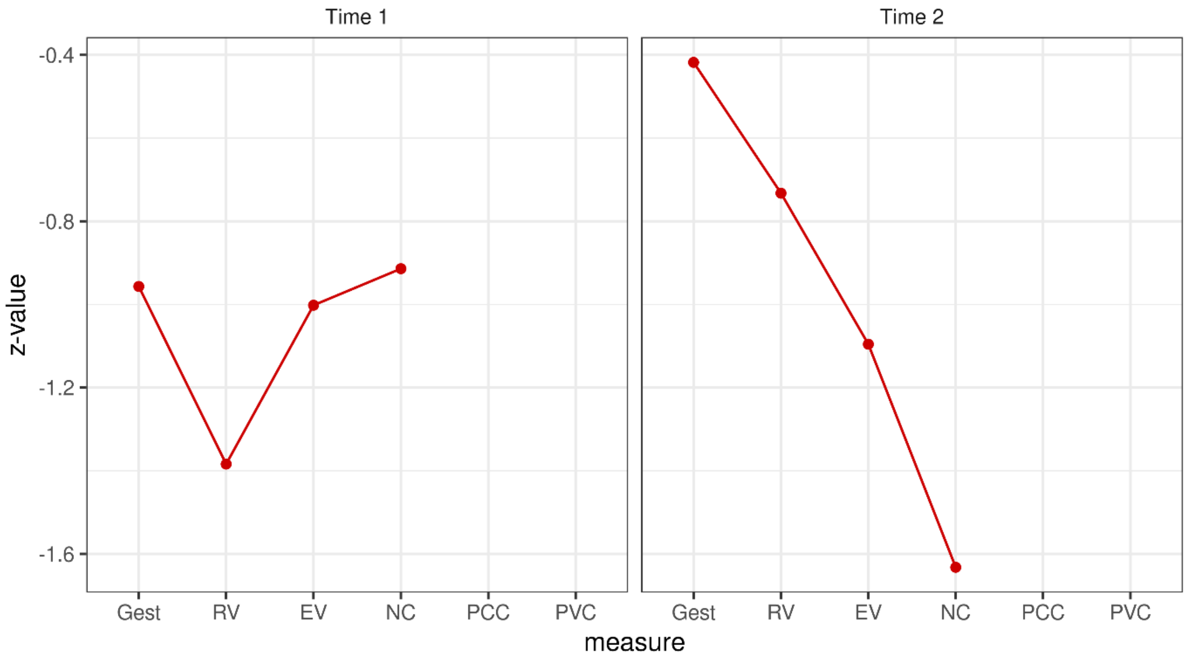
Supplementary Materials



Supplementary Figure 4.1. Trajectory of communication development of Participant 5

Gest = number of gestures on CDI (Fenson, et al., 2007), RV = receptive vocabulary on CDI, EV = expressive vocabulary on CDI, NC = number of consonants in consonant inventory, PCC = percent consonants correct, PVC = percent vowels correct



Supplementary Figure 4.2. Trajectory of communication development of Participant 23

Gest = number of gestures on CDI (Fenson, et al., 2007), RV = receptive vocabulary on CDI, EV = expressive vocabulary on CDI, NC = number of consonants in consonant inventory, PCC = percent consonants correct, PVC = percent vowels correct

Chapter 5: Taking a Closer Look at the Children in Cluster B

Although the descriptions of all children in the longitudinal study are interesting and important, this chapter focuses on the question that originally prompted this research and has been asked by clinicians for decades (Dawson, 2011; Prizant, 1996): Is there a small subgroup of children with ASD with a significant SSD? A consistent theme throughout this thesis has been the value of describing what children can do, rather than focusing solely on their speech deficits. In line with this approach, this chapter provides further descriptive details of the development and communication of the three participants in Cluster B, in an attempt to further our understanding of their abilities and differentially diagnose an SSD.

Differentially diagnosing an SSD in children with limited verbal output is challenging (Chenausky et al., 2019; Strand et al., 2013) but necessary to guide appropriate intervention. Some researchers in the ASD literature have begun to describe a subgroup of minimally verbal children with similar communication profiles to the children in Cluster B presented in Chapters 3 and 4 (Beiting & Maas, 2021; Chenausky et al., 2019; Chenausky et al., 2021; Saul & Norbury, 2020). Childhood Apraxia of Speech (CAS) has been diagnosed or suspected in three studies (Beiting & Maas, 2021; Chenausky et al., 2019; Chenausky et al., 2021). A further subgroup in the Chenausky et al (2019) study was found to have a non-CAS speech disorder, possibly reflecting a phonological disorder. For good reason, participants in these studies were selected based on low language levels or suspected SSD, so it remains unknown if similar subgroups of children with ASD and an SSD would emerge from a heterogeneous cohort.

Determining the extent to which phonological and/or motor speech deficits contribute to the child's SSD is challenging, particularly with low verbal children (Strand et al., 2013). Criteria for both diagnoses largely require at least some verbal ability, and many diagnostic features do not discriminate between diagnoses (McCabe et al., 1998). Three significant speech sound disorders can be contemplated as potentially responsible for severe impairment in the speech of children with ASD: phonological impairment, childhood apraxia of speech, and dysarthria. An impairment at the phonological level results in sound error patterns, such as omissions, substitutions, and additions (Vihman, 1996). A diagnosis of a phonological impairment typically involves comparing a child's production of sounds, syllables, and words to adult targets. This comparison may not be possible for children at very early stages of linguistic development. Childhood Apraxia of Speech (CAS) is a neurological SSD, affecting the precision and accuracy of speech production in the absence of underlying neuromuscular impairments (ASHA, 2007) which is caused by difficulties planning and programming coordinated speech movements. Finally, it is possible that speech disorders in ASD may be dysarthric in nature. Dysarthria, another motor speech disorder, occurs when there is a disruption to the final neurological pathway to the articulators and is marked by concomitant asymmetry at rest and in movement, changes to typical precision, rate, and range of speech and non-speech movement, and to changes in muscle strength and tone. As Strand (2017) discussed, a common error is to assume children can only have one specific SSD, when in fact many children have both a phonological and a motoric component to their SSD.

Further Descriptions of Children in Cluster B

The three children in Cluster B presented with a unique communication profile, with strengths in receptive vocabulary and nonverbal communication, in the presence of significantly limited speech and expressive vocabularies (Chapters 3 and 4). To expand our understanding of their individual abilities and profiles, further descriptive details are provided below. Considering the complex developmental histories of these three children, comparing their speech ability to age norms seems out of place. Instead, consonants were categorised as Early, Middle, or Late (Shriberg, 1993) to provide some context regarding the expected order of acquisition.

Participant 3

Participant 3 was a male aged 74 months at Time 1. Participant 3 reportedly babbled as an infant and spoke his first word prior to his first birthday. His parents reported a regression of his verbal skills prior to 2 years. He was diagnosed with ASD at 31 months and had attended early intervention services, including speech pathology, since diagnosis. There was a large discrepancy between his nonverbal developmental quotient (NVDQ; 76.27) and verbal developmental quotient (VDQ; 11.86) on the Griffiths Mental Developmental Scales – Extended Revised (GMDS-ER; Luiz et al., 2004). Participant 3 was engaging and communicative and augmented his limited verbal capacity with gestures. He enjoyed letters and numbers and could reportedly read and write simple words. His parents reported comprehension of spoken words as a strength, indicating that he understood 363 words on the MacArthur-Bates CDI (Fenson et al., 2007) at Time 1. By contrast, his parents reported no spoken words at Time 1, although he did use words during the first assessment (Time 1) reported in Chapter 3. His scores on the Preschool Language Scales – Fourth Edition (PLS-4; Zimmerman et al., 2002) did not capture the large difference between his language comprehension and expression. He scored a standard score of 50 (< 1st percentile) for both Auditory Comprehension and Expressive Communication at Time 1 and 2. This floor effect may underscore the challenges of completing standardised assessments with children with ASD, as well as the inherent difficulties of comparing the ability of children with developmental delays to chronological-aged norms. Specifically, Participant 3 became fixated on the page numbers during the PLS-4, was impulsive and pointed to pictures often prior to the completion of the instruction. He was unable to attend to a book-based task for any length of time. These behaviours are not uncommon for children with ASD, highlighting the importance of obtaining details of abilities through multiple methods, rather than only from standardised assessment.

On oromotor assessment, Participant 3 had difficulty imitating both non-speech and speech oral movements and so observations of his oral musculature were also taken at rest, and during other speech and non-speech activities. Participant 3 presented with symmetrical oral musculature. No structural abnormalities were evident. He had normal tone of the oral musculature, although his oral movements during speech appeared slow. Participant 3 was able to protrude his tongue and laterally move his tongue when eating but had difficulty imitating lateral tongue movement during assessment. He could lick his lips but was unable to imitate tongue tip elevation. Participant 3 could spread his lips, smack his lips closed and round his lips to drink with a straw. He was unable to imitate lip rounding during assessment. Diadochokinesis tasks (imitation of repeated syllables, such as “pa”, “ta”, “ka”) were attempted but were unable to be completed. Participant 3 looked at the examiners mouth and attempted to imitate various sounds, but produced neutral schwa (/ə/), or /mə/. His speech attempts were slow and appeared effortful. After a few moments, his attention waned.

Participant 3’s speech was severely limited. At Time 1 his consonant repertoire included five consonants (/m/, /b/, /w/, /j/, /h/) and his vowel use was dominated by schwa (/ə/) and /a/, with infrequent use of /ʌ/, /ɒ/, and /ʊ/. On one occasion at Time 1, participant 3 used a dipthong (/o͡ʊ/) accurately. He produced only single syllables and a large proportion of these syllables were single vowels. Participant 3 had profoundly limited consonant and vowel accuracy (4.8% and 10.5% respectively) during single-word naming at Time 1 (Table 5.1).

Over 12 months (Time 2), Participant 3’s communication improved slightly. His CDI scores increased to 393 words understood and 75 words spoken. The large jump in his spoken words likely reflects a combination of increased vocabulary, and slightly improved consonant and vowel production. Together these improvements may have assisted his parents to interpret more speech-like productions as attempts at real words. At Time 2 he was using eight consonants (/b/, /w/, /p/, /k/, /g/, /f/, /ʃ/, /s/); only two of the same consonants from Time

1. His vowel use continued to be dominated by schwa (/ə/), but he also used short vowels /æ/, /ʊ/, and /ʌ/, and occasionally long vowel /ɔ/. There was one instance of a dipthong /o͡ʊ/ used accurately when saying a new word ‘toast’. Despite an increase in both his consonant and vowel repertoires, his consonant accuracy (17.6%) and vowel accuracy (18.9%) remained profoundly low (Table 5.1). His speech vocalizations continued to be single syllables with a large proportion of single vowels. At Time 2, he was able to round his lips when given a visual cue. There were no other changes to his oromotor ability.

Table 5.1. Comparing speech profiles of Cluster B children

Variable	Participant 3	Participant 12	Participant 13
Time 2 age (months)	86	76	50
Consonant inventory			
Time 1	m, b, w, j, h	m, b, j, p	m, b, j, n, d, h
Time 2	b, w, p, k, g, f, ʃ, s	m, b, n, w, d, p, h, t	m, b, j, h, t, s
Vowel inventory			
Time 1	ə, a, ʌ, ɒ, ʊ, o͡ʊ	ə, ʌ, a	ə, ʌ, æ, a, ɔ, ɒ, ɪ, o͡ʊ, a͡ɪ
Time 2	ə, æ, ʌ, ʊ, ɔ, o͡ʊ	ə, ʌ, a, ɛ, ɔ, u, a͡u	ə, ʌ, æ, ɪ, ɛ, i, a͡ɪ, ɛ͡ə, e͡ɪ
PCC (%)			
Time 1	4.8	18.8	7
Time 2	17.6	19.7	2.7
PVC (%)			
Time 1	10.5	25	35.1
Time 2	18.9	25.6	22.5

PCC = percent consonants correct, PVC = percent vowels correct

Participant 12

Participant 12 was a male aged 63 months at Time 1. His parents did not recall him babbling as an infant. He reportedly used his first word around 14 months of age and verbally regressed soon after. He was diagnosed with ASD at 36 months. Participant 12 had attended early intervention since his diagnosis and was in his first year at an autism specialist school during the study. His NVDQ was 48.21 and VDQ was 21.43 on the GMDS-ER (Luiz et al., 2004). His parents reported he understood 289 words on the CDI (Fenson et al., 2007) but only had 4 words in his spoken vocabulary. His scores on the PLS-4 (Zimmerman et al.,

2002) for both Auditory Comprehension and Expressive Communication were also at floor level (standard score 50, <1st percentile) at both Time 1 and 2. Participant 12 used a voice-output device, picture symbols and key word signs to communicate.

On oromotor assessment, Participant 12 had difficulty moving his oral musculature when given a verbal instruction or a visual model to imitate. Observations of his oral movements were also taken at rest, during non-speech activities, and speech. Participant 12 presented with symmetrical oral musculature with no structural abnormalities. The tone of his oral musculature appeared to be typical, although during speech his movements were slow. On assessment, Participant 12 was able to protrude his tongue, but had difficulty imitating lateral or elevation movement. These movements were observed during eating. Participant 12 had difficulty imitating lip movements at Time 1 but could imitate lip rounding at Time 2. He continued to have difficulty imitating lip spread movement, although he could smile. He was unable to imitate sequential oral movements or sounds, and made no attempts at diadochokinesis tasks. When singing, he maintained an open mouth posture and produced a monotonous string of vocalisations. There was no variation in vowels, pitch, or rhythm.

Participant 12's speech was very limited. He produced a large number of non-speech vocalizations such as squeals and grunts. At Time 1 his consonant repertoire included four consonants (/m/, /b/, /j/, /p/) and his vowel repertoire included only three vowels (/ə/, /ʌ/, /a/). He was only able to produce single syllables. A large proportion of his speech-like vocalizations were vowels only. His consonant and vowel accuracy were profoundly limited (18.8% and 25% respectively; Table 5.1).

Over the 12 months of the study, there was some improvement in Participant 12's communication ability. His consonant repertoire increased to eight consonants (/m/, /b/, /n/, /w/, /d/, /p/, /h/, /t/) and his vowel repertoire expanded to seven vowels (/ə/, /ʌ/, /a/, /ɛ/, /ɔ/, /u/, /a͡u/). His speech included mainly CV (consonant-vowel) syllables at Time 2. Participant

12's consonant and vowel accuracy scores (19.7% and 25.5% respectively) remained functionally unchanged (Table 5.1). He only added two words to his spoken vocabulary over the 12 months, using six spoken words at Time 2.

Participant 13

Participant 13 was a male aged 36 months at Time 1. His parents reported that he had babbled as an infant and produced his first word around his first birthday but then verbally regressed. He was diagnosed with ASD at 29 months of age. Participant 13 attended speech pathology and occupational therapy since diagnosis. A developmental assessment was unavailable for Participant 13. His parents reported comprehension of 234 words but use of only 5 spoken words on the CDI (Fenson et al., 2007) at Time 1. He communicated with a combination of gestures, key word signs and pictures. His use of eye contact and facial expressions to communicate his enjoyment were strengths. On the PLS-4 (Zimmerman et al., 2002), Participant 13 scored 67 (1st percentile) for Auditory Comprehension and 55 (1st percentile) for Expressive Communication.

During oromotor assessment, Participant 13 made attempts at all non-speech and speech imitation tasks but had difficulty completing these tasks at both Time 1 and Time 2. As with the other two participants, observations of his oral musculature were also taken during rest, non-speech activities and in speech. At rest, he presented with symmetrical oral musculature with no structural abnormalities. He had typical rate and range of movement of his lips and tongue during non-speech and speech tasks and the tone of his oral musculature appeared normal. Tongue protrusion, lateral tongue movement and tongue elevation were observed and appeared normal. Lip spread and lip rounding movements were also normal. He had difficulty imitating sequential oral movements or diadochokinesis (imitation of spoken syllables) tasks, although made some attempts to imitate. Participant 13 looked at the examiners mouth and produced single vowels, predominantly /ʌ/. There was no sequencing of varied speech sounds, although he was able to imitate the same vowel on some occasions. When reduplicating vowels, the rate of his speech was normal.

Participant 13 presented with a significant speech deficit. His consonant repertoire at Time 1 included six consonants (/m/, /b/, /j/, /n/, /d/, /h/) which he used infrequently. His speech was predominantly single vowels and his vowel repertoire was dominated by /ə/, /ʌ/ and /æ/, although he also used /a/, /ɔ/, /ɒ/, /ɪ/, /o͡ʊ/, and /a͡ɪ/ infrequently. At Time 1, the accuracy of his vowel production (35.1%) was stronger than his consonant production (7%), although both were very low. It should be noted the Participant 13 had higher vowel accuracy than the other two children in Cluster B at Time 1 (Table 5.1).

By Time 2, his understanding of spoken words had increased to include 316 words and his spoken vocabulary included 19 words. Of the three children in Cluster B, Participant 13 had the least speech development across the 12 months. At Time 2 he was still only using six consonants (/m/, /b/, /j/, /h/, /t/, /s/) sparingly and nine vowels (/ə/, /ʌ/, /æ/, /ɪ/, /ɛ/, /i/, /a͡ɪ/, /ɛ͡ə/, /e͡ɪ/). The accuracy of his consonants (2.7%) and vowels (22.5%) had reduced over the 12 months (Table 5.1). This apparent regression was most likely associated with an observed increase in his overall attempts to communicate verbally. His speech continued to be dominated by single vowel syllables.

Limitation of assessments

A strengths-based assessment approach was adopted to capture the speech capacity of these children. While we are confident that the results represent the participant's optimal communication abilities, the data is limited in one regard. Only participant 13 attempted all tasks on the oromotor assessment and none of the participants in Cluster B were able to imitate non-speech and speech sequential movements. Given that these three children presented clinically with normal tone and no overt distortions of oral musculature, we can rule our dysarthria as a primary cause of their significant speech deficits. However, considering the challenges completing the oromotor assessment, concomitant mild dysarthria cannot be ruled out.

Summary of children in Cluster B

Table 5.1 summarises the speech abilities of the three children in Cluster B at Time 1 and Time 2. The similarities and differences of their communication profiles are discussed.

The three children in Cluster B presented with similar communication histories, profiles, and trajectories of development. The parents of all three children reported a history of verbal regression at various timepoints between 12-24 months of age. Whether this was a true loss of skill, or a plateau of their abilities is unknown. Verbal regression is discussed further in Chapter 7. Interestingly, none of the children in Cluster B retained all Time 1 consonants over 12 months. Instead, they replaced some consonants with new ones. This difficulty retaining previously learnt sounds may reflect instability and inconsistency of their sound systems rather than true verbal regression. It is also possible that these sounds were simply not sampled during the Time 2 assessment.

The unique common communication profile of the three children in Cluster B remained stable over the 12 months of the longitudinal study. As discussed in Chapter 3, all children presented with strengths in receptive vocabularies and nonverbal communication in the presence of few words and limited speech capacity. The three children developed their receptive vocabularies and nonverbal communication during the longitudinal study but had minimal speech or development. Ordinarily we would expect speech to develop along with language, so this decoupling is notable.

There were a number of similarities between the speech of the three children in Cluster B (Table 5.1). All three children had small consonant and vowel repertoires. At Time 1, these children used only some Early 8 consonants (Shriberg, 1993) and predominantly schwa (/ə/) and one other vowel. They all had high rates of vowel-only vocalizations, and severely reduced consonant and vowel accuracy. The speech development over 12 months of the three children in Cluster B was limited. Participant 3 made the most gains in consonant and vowel accuracy, although these scores continued to be profoundly low. Participant 12's speech accuracy remained relatively unchanged. The speech accuracy of Participant 13 reduced over the 12 months. As mentioned previously, this may reflect the instability of his sound system, but may also underscore his increased attempts to say new words, that were not attempted in his initial assessments. As children attempt more linguistic variation rather than only using learnt and practised words or scripts, it is possible that the accuracy of their speech may reduce. This is likely true for Participant 13, who was attempting more words at Time 2 compared to Time 1.

Given the disparity between the receptive language and nonverbal communication development, and limited speech growth of the Cluster B children, the presence of an SSD can be assumed. Differentially diagnosing a motor speech disorder, especially CAS, from a phonological disorder in these children is difficult. As discussed by Strand (2017), limited consonant and vowel repertoires, use of simple syllable shapes, frequent omission of sounds, and poor speech intelligibility are frequent features of both CAS and significant phonological impairments.

The American Speech-Language-Hearing Association defines CAS as a neurological SSD of childhood and lists three core features consistent with a deficit of motor planning and programming: "(a) inconsistent errors on consonants and vowels in repeated productions of syllables or words; (b) lengthened and disrupted coarticulatory transitions between sounds and syllables; and (c) inappropriate prosody, especially in the realization of lexical or phrasal stress" (ASHA, 2007, p. 4). It is important to highlight that while these three features were not reported as required or sufficient for a diagnosis of CAS, they have been used widely for description of children with CAS over the past 12 years. For low verbal children, it can be difficult to ascertain some of the above-mentioned features due to limited use of words or difficulty completing the various assessment tasks. Although phoneme imitation and repetition may be possible with minimally verbal children, all three children in Cluster B had significant difficulty completing diadochokinesis tasks during the oromotor assessment. Difficulty with diadochokinesis tasks in verbal children suspected of CAS was however a key diagnostic determiner in Murray et al. (2015). Whether the children reported here could complete speech imitation assessment tools devised specifically for children at early linguistic levels (e.g., DEMSS; Strand et al., 2013) remains unknown. Completing such a task may provide the necessary information to make a clear diagnosis of CAS however it would not confirm or negate the presence of concomitant phonological impairment.

It is important to note that the studies in this thesis did not intend to differentially diagnose an SSD in children with ASD. As a result, we didn't collect some of the data needed to differentially diagnose the specific SSD in this population, such as the inconsistency data suggested for a CAS diagnosis. It is possible that the hypothesised SSD reported in the children in Cluster B comprises both phonological and motor speech characteristics. Clinically, our instinct is to diagnose CAS as the primary SSD in these children, but at this stage we do not have the data to support this claim. Mild concomitant dysarthria cannot be ruled out but is unlikely to be a primary cause of the children's significant speech impairment. Establishing the nature of a speech disorder associated with ASD, and therefore, moving towards appropriate intervention, will require examination of larger samples of children with ASD who belong to this cluster.

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Chapter 6: Systematic Review Update

There is growing interest in the speech capacity of children with ASD, particularly those described as prelinguistic or minimally verbal. The systematic review presented in Chapter 2 and published as Broome et al. (2017) presented literature published between 1990 and 2014. This chapter presents an update of that systematic review to provide current context for the studies in Chapter 3 and 4 and the discussion which follows in Chapter 7.

Method

An update of the published systematic review was completed using the same method for identification and screening described in Chapter 2. Peer-reviewed articles published between January 2015 and August 2021 were eligible for review. The paper presented in Chapter 3 (Broome et al., 2021) was published outside of these dates and is therefore, not included in this systematic review. The initial broad search identified 1,076 articles. Four additional articles were identified through hand searches. Once duplicates were removed, 857 articles remained for further review. Titles and abstracts were initially screened and excluded based on criteria outlined in Chapter 2.

Eligibility

Fifty-eight full-text articles were assessed for eligibility. Articles were excluded if they: (a) did not include children with ASD, (b) did not report the results from a speech output assessment, (c) focused on voice or fluency, (d) did not include English-speaking children, (e) used only general terms (i.e. 'vocalizations'), or (f) were not peer-reviewed research studies. One article (Shriberg et al., 2019) presented a summary of previously reported results included in the original systematic review. Of the remaining 29 articles, only two (Peter et al., 2019; Vidal et al., 2020) did not meet 'confidence in ASD diagnosis' criteria outlined in Chapter 2.

Analysis

The review identified 27 articles that met inclusion criteria. These articles were coded according to: (a) the language stage of the participants, (b) the speech domain assessed, (c) the speech assessment used, and (d) the speech characteristics described in the article. As per the original review, articles were coded as including prelinguistic and/or verbal participants. Articles that included verbal children were further categorized as assessing segmental or suprasegmental aspects of speech. In this update, children were defined as being in the prelinguistic stage if they produced fewer than 20 recognizable words, rather than 10 words used in the original systematic review. This change was made to reflect the distinction commonly used in the current literature in this area (Chenausky et al., 2017b; McDaniel et al., 2020a; Woynaroski et al., 2016; Woynaroski et al., 2017). One study (Saul & Norbury, 2020) stretched this definition to include children using fewer than 24 words, similar to the range reported in McCleery et al. (2006). For the purpose of this review, the Saul and Norbury (2020) article was coded as including prelinguistic participants. Although the term 'minimally verbal' is more frequently used in this field of research, 'prelinguistic' was retained to be consistent with the original systematic review.

Results

The review update identified 27 articles that met inclusion criteria. The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA; Moher et al., 2009) flow diagram outlines the process of study selection and is presented in Figure 6.1. Overall, 617 participants are included in the 27 articles.

Prelinguistic participants

Children were categorized as being at the prelinguistic stage of language development in this update if they produced fewer than 20 recognizable words. Eighteen articles assessed the speech of prelinguistic children with ASD (Table 6.1). These articles contained 13

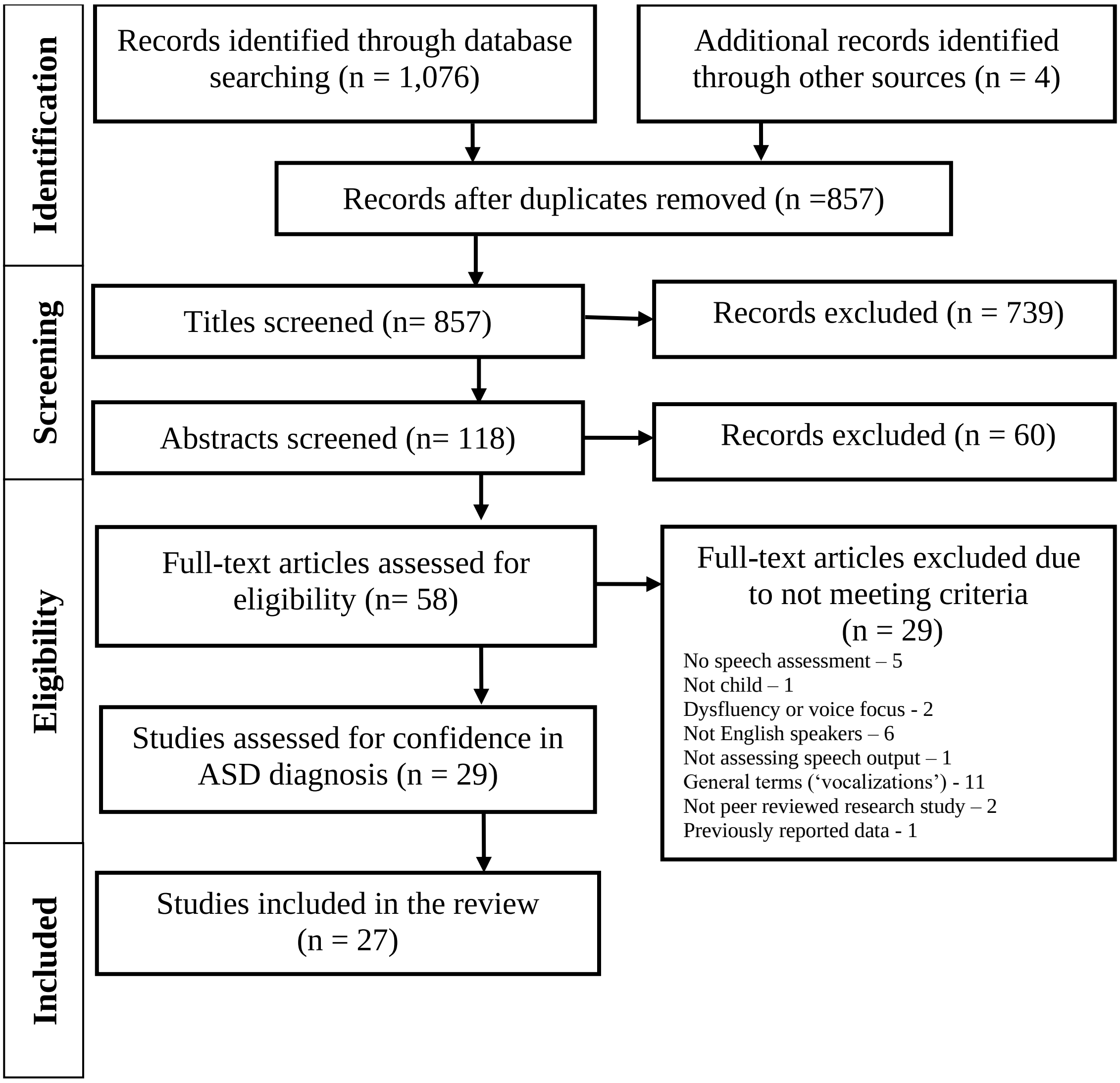

Figure 6.1. Flow diagram of study selection (adapted from PRISMA, Moher et al, 2009)

separate cohorts of children. Participants from the same cohort are reported in Chenausky et al. (2016), Chenausky et al. (2018), and Chenausky et al. (2021). Some of these children had previously been reported in Wan et al. (2011), and this article was included in the original systematic review. Another sample of children are reported in McDaniel et al. (2019), Woynaroski et al. (2016) and Yoder et al. (2015). Overall, 407 participants are included in the 13 cohorts of prelinguistic children with ASD. One article (Chenausky et al., 2019) included prelinguistic children, verbal children and one adult. It is unclear how many children were at the prelinguistic level and so all 54 participants are listed in both prelinguistic and verbal results. Two further articles (Chenausky et al., 2017b; Ellawadi & Ellis Weismer, 2015) included prelinguistic and verbal participants but presented the data according to participant's language level. This information could then be separated for the purposes of this review and presented in Table 6.1 and 6.2.

Regarding study design, three treatment studies adopted a single-subject multiple-baseline design (Beiting & Maas, 2021; Chenausky et al., 2016; Chenausky et al., 2018), one study was a prospective case study (Biller & Johnson, 2019), three were prospective cohort studies (Chenausky et al., 2019; Chenausky et al., 2021; Ellawadi & Ellis Weismer, 2015), two were prospective case control studies (Chenausky et al., 2017b; Tenenbaum et al., 2020) and six were prospective longitudinal cohort studies (Garrido et al., 2017; Heymann et al., 2018; McDaniel et al., 2019, Woynaroski et al., 2016; Woynaroski et al., 2017; Yoder et al., 2015).

Assessment methods for prelinguistic participants

Spontaneous speech samples were used to assess the speech of prelinguistic children in 13 of the 18 articles. A speech sample was collected from vocalizations made during the Communication and Symbolic Behavior Scales—Developmental Profile Behavior Sample (CSBS; Wetherby & Prizant, 2002) in seven articles (Biller & Johnson, 2019; Garrido et al., 2017; McDaniel et al., 2019, Saul & Norbury, 2020; Woynaroski et al., 2016; Woynaroski et al., 2017; Yoder et al., 2015). Two further articles collected a spontaneous speech sample from the Early Social Communication Scales (ESCS; Mundy et al., 2003) (Ellawadi & Ellis Weismer, 2015; Heymann et al., 2018). Although taking a sample during a standard assessment would suggest comparable methods, none of the studies reported number of utterances and only some included length of time of the sample. Without this detail, it is challenging to compare samples between studies. Semi-structured play interactions using a

Table 6.1 Participant information and speech assessment details from studies with prelinguistic participants with ASD

Reference	Research Design	ASD n	Mean Age (mo.)	Cognitive Level	Language Level	Speech Data Collection Method	Speech Analyses
Beiting & Maas (2021)	Single subject multiple-baseline design	3	75	Not stated	Receptive vocabulary[a] Raw scores 5-24 Expressive vocabulary 0-20 words	Speech imitation Spontaneous speech sample Single-word naming	Phonetic repertoire Percent perceptual accuracy Standard score on GFTA-2*
Biller & Johnson (2019)	Prospective case study	5	63.2	Nonverbal cognitive[b] AE 25.4 (4.04) months	Receptive language[b] AE 13.2 (9.15) months Expressive language[b] AE 11.2 (2.59) months Expressive vocabulary[c] 5 (3.24) words	Speech imitation Spontaneous speech sample	Number of speech sounds Number of syllables Categorized vocalizations
Chenausky, et al. (2016)	Single subject multiple-baseline design	23 AMMT 7 SRT	77 68	Mental age[b] 20.4 (8.1)* 22.3 (10.8)	Not stated	Speech imitation	Phoneme inventory, PCC, PVC, percent syllables approximated
Chenausky, et al. (2018)	Single subject multiple-baseline design	27 AMMT 11 SRT	80 74	NVIQ[b] raw score 29.1 (8.6)AMMT 31.7 (11.3) SRT	Expressive language[b] raw scores 10.8 (1.9) AMMT 11.7 (3.9) SRT	Speech imitation	as above
Chenausky, et al. (2021)	Prospective cohort study	38	77	NVIQ[b] raw score 30.3 (9.8)*	Receptive language[b] raw scores 19.5 (8.4)* Expressive language[b] raw scores 11.3 (2.7)*	Speech imitation	Perceptual coding of CAS features Acoustic analysis*
Chenausky, Norton & Schlaug (2017b)	Prospective case control study	2	64.5	Nonverbal cognition[b] raw score 17.5 (6.4)	Receptive language[b] raw scores 11 (1.4) Expressive language[b] raw scores 9.5 (0.7) Expressive vocab[c] raw score 2 (1.4)	Speech imitation	Phonetic inventory

Chenausky, et al. (2019)	Prospective cohort study	54**	52-226	NVIQ[d] 30-115	Receptive vocabulary[a] 0-123 Number of different words (NDW) 0-229	Speech imitation	Perceptual coding of CAS features
Ellawadi & Ellis Weismer (2015)	Prospective cohort study	19	29.21	Cognitive standard score[e] 76.84 (9.84)	Expressive language[f] AE < 15 months	Spontaneous speech sample	Consonant inventory
Garrido, et al. (2017)	Prospective longitudinal cohort study	34	13.79	Nonverbal cognition[b] T score 43.00 (12.49)	Receptive language[b] 33.21 (12.34) Expressive language[b] 34.26 (11.71)	Spontaneous speech sample	Categorized vocalizations
Heymann, et al. (2018)	Prospective longitudinal cohort study	9	14, 18, 24	Not stated	Not stated	Spontaneous speech sample	Categorized vocalizations
McDaniel, et al. (2019)	Prospective longitudinal cohort study	68	35.26	Mental age[b] 12.59 (5.11)	Receptive vocabulary[c] raw score 113 (108) Expressive vocabulary[c] raw score 19 (27)	Spontaneous speech sample	Categorized vocalizations for canonical syllables, identifiable words
Woynaroski, et al. (2016)	Prospective longitudinal cohort study	87	34.7	Mental age[b] 12.1 (4.7)	Receptive vocabulary[c] raw score 75.8 (85.4) Expressive vocabulary[c] raw score 3.7 (5)	Spontaneous speech sample	Coded for DKCC
Yoder, Watson & Lambert (2015)	Prospective longitudinal cohort study	87	34.7	as above	as above	as above	Consonant inventory
McDaniel. et al. (2020a)	Prospective longitudinal cohort study	87	23.42	DQ[b] 58.83 (17.96)	Receptive language[b] AE 10.11 (7.22) Expressive language[b] AE 11.97 (4.71)	Spontaneous speech sample Naturalistic speech sample using LENA	Coded vocalizations for DKCC, canonical syllables
McDaniel, et al. (2020b)	Prospective longitudinal cohort study	87	23.42	as above	as above	Spontaneous speech sample	as above

Saul & Norbury (2020)	Prospective longitudinal cohort study	27	49.6	NVIQ[b] 0.42 (0.17)	Receptive vocabulary[c] Raw score 150 (111) Expressive vocabulary[c] Raw score 4.5 (7.4)	Spontaneous speech sample Speech imitation Parent report measure	Consonant inventory, phoneme repertoire composite
Tenenbaum, et al. (2020)	Prospective case control study	22	26.19	Not stated	Not stated	Spontaneous speech sample	Categorized vocalizations
Woynaroski, et al. (2017)	Prospective longitudinal cohort study	20	37.9	Cognitive composite[b] AE 9.07 (3.32)	Receptive vocabulary[b] AE 4.05 (5.34) Expressive language[b] AE 7.35 (3.65) Expressive vocabulary[c] raw score 5.4 (5.23)	Spontaneous speech sample Naturalistic speech sample using LENA	Consonant inventory Proportion of canonical syllables 3 automated vocal analyses

Note. standard scores unless otherwise mentioned (standard deviations in brackets); AE = age equivalent; CAS = childhood apraxia of speech; DKCC diversity of key consonants used in communicative acts; DQ = developmental quotient; GFTA-2 = Goldman-Fristoe Test of Articulation–Second Edition (Goldman & Fristoe, 2000); IQ = intelligence quotient; LENA = Language ENvironment Analysis (LENA Research Foundation, 2016); PCC = percent consonants correct; PVC = percent vowels correct

*Data not for all participants

**Mean age for all participants with ASD, including verbal participants

[a] Peabody Picture Vocabulary Test (PPVT; Dunn & Dunn, 1981). [b]Mullen scales of early learning (MSEL; Mullen, 1995). [c]MacArthur-Bates Communicative Development Inventory (CDI; Fenson, et al., 2007). [d] Leiter International Performance Scale – Third Edition (Roid & Miller, 2013). [e]Bayley Scales for Infant Development, Third Edition (Bayley, 2006). [f]Preschool Language Scales – Fourth Edition (PLS-4; Zimmerman, Steiner, & Pond, 2002).

standard set of toys was used to collect a speech sample in three separate cohorts across four articles (Garrido et al., 2017; McDaniel et al., 2020a, McDaniel et al., 2020b; Woynaroski et al., 2017). Play interactions varied across studies by time (from 6 to 15 minutes) as well as communication partner (parent or clinician). Daylong audio recordings using the LENA digital recording device (LENA Research Foundation, 2015) were utilized in two studies (McDaniel et al., 2020a; Woynaroski et al., 2017).

Speech data reported from the spontaneous speech samples included consonant inventory, phonetic repertoire, and proportion of syllables that contained a consonant. Studies that categorized speech-like vocalizations did so according to pre-determined levels of complexity or specific speech characteristics described in the article. Biller and Johnson (2019) characterized vocalizations according to Tager-Flusberg and colleague's (2009) definitions. Other articles categorized speech-like vocalizations as containing canonical syllables and syllables without consonants (Garrido et al., 2017; Heymann et al., 2018; McDaniel et al., 2019; McDaniel et al., 2020a; McDaniel et al., 2020b; Woynaroski et al., 2017). One article made a distinction between nonword canonical syllables and canonical syllables obtained from recognisable words (Tenebaum et al., 2020).

Speech imitation tasks were used by 8 of the 18 articles that assessed the speech of prelinguistic children with ASD. Standard assessments were used in four articles. Chenausky and colleagues used the first two sections of the Kaufman Speech Praxis Test (KSPT; Kaufman, 1995) in three of their studies with prelinguistic children (Chenausky et al., 2016, Chenausky et al., 2017b; Chenausky et al., 2019). Saul and Norbury (2020) implemented an adapted version of the KSPT to ascertain children's ability to imitate single phonemes. Biller and Johnson (2019) used the Focal Oromotor Control section of the Verbal Motor Production Assessment for Children (VMPAC; Hayden & Square, 1999). The other three articles used less standardized speech imitation tasks (Beiting & Maas, 2021; Chenausky et al., 2018; Chenausky et al., 2021).

Beiting and Maas (2021) were the only authors to use a standardized single-word naming task to assess the speech of prelinguistic children with ASD. These authors used the Goldman-Fristoe Test of Articulation – Second Edition (GFTA-2; Goldman & Fristoe, 2000) with two of the three children in their study and reported a standard score of less than 40 (<1st percentile) for both children.

Verbal participants

The speech of verbal children with ASD was assessed in 12 articles and is summarised in Table 6.2. Three articles (Chenausky et al., 2017b; Chenausky et al., 2019; Ellawadi & Ellis Weismer, 2015) included both prelinguistic and verbal participants and were previously mentioned in the prelinguistic section. Two articles included both adults and children (Chenausky et al., 2019; Patel et al., 2020). Without individual scores, data could not be separated and so all participants from these studies are included in Table 6.2. The 12 articles covered 11 separate cohorts comprising 264 participants.

Segmental speech

Segmental speech refers to the sound production of speech, both phonemic and phonetic. Eight articles assessed segmental speech of verbal children with ASD. Of these articles, one was a single case study (Biller & Johnson, 2020), two articles adopted a longitudinal control study design and reported on the same children (Chenausky et al., 2017a; Chenausky & Tager-Flusberg, 2017c), two articles were prospective cohort studies (Chenausky et al., 2019; Ellawadi & Ellis Weismer, 2015), and three were prospective case control studies (Chenausky et al., 2017b; Dalton et al., 2019; Nadig & Mulligan, 2017).

Table 6.2 Participant information and speech assessment details from studies with verbal participants with ASD

Reference	Research Design	ASD n	Mean Age (mo.)	Cognitive Level	Language Level	Speech Data Collection Method	Speech Analyses	Speech Domain
Akbari & Davis (2019)	Single case experimental design	1	168*	Average[a] **	Average[b] **	Spontaneous speech sample	PVSP	Suprasegmental
Arciuli & Bailey (2019)	Prospective case control study	20	88.55	Not stated	Receptive vocabulary[c] raw score 96.55 (31.21)	Single-word naming	Perceptual and acoustic analysis	Suprasegmental
Biller & Johnson (2020)	Single case study	1	40	Nonverbal cognition[d] AE 24 months	Receptive language[d] AE 25 months Expressive language[d] AE 21 months Expressive vocabulary[e] 40 words	Spontaneous speech sample Speech imitation	Control of voicing, number of speech sounds, number of syllables, categorized stage of spoken language	Segmental
Chenausky, et al. (2017a)	Longitudinal control study	10	12, 18, 24	Not stated	24 month T scores Receptive language[d] 49.4 (10.9) Expressive language[d] 48.0 (9.2)	Spontaneous speech samples	Categorized vocalizations	Segmental
Chenausky & Tager-Flusberg (2017c)	Longitudinal control study	11	12, 18, 24	Not stated	24 month T scores Receptive language[d] 47.9 (16.6) Expressive language[d] 49.1 (11)	Spontaneous speech samples	Categorized vocalizations	Segmental
Chenausky, et al. (2017b)	Prospective case control study	2	56	Nonverbal cognition[d] raw score 36 (14.1)	Receptive language[d] raw score 21 (11.3) Expressive language[d] raw score 17 (1.4) Expressive vocabulary[e] raw score 110.5 (29)	Speech imitation	Phonetic inventory	Segmental

Chenausky, et al. (2019)	Prospective cohort study	54***	52-226	NVIQ[f] 30-115	Receptive vocabulary[c] Raw score 0-123 Number of different words 0-229	Speech imitation	Speech coded for signs of CAS	Segmental
Dalton, et al. (2017)	Prospective case control study	10	57.3	Nonverbal cognition[d] AE 58.17 (15.6)	Receptive vocabulary[c] 97 (24.35)	Spontaneous speech sample Speech imitation	VMPAC score Motor control score 0 – 7 for connected speech sample	Segmental
Ellawadi & Ellis Weismer (2015)	Prospective cohort study	86	31.23 ***	Cognition[g] First words (n=61) 84.51 (9.73) Word combinations (n=22) 94.77 (13.84)	Expressive language[h] AE > 15 months	Spontaneous speech sample	Consonant inventory	Segmental
Nadig & Mulligan (2017)	Prospective case control study	9	68.67	Not stated	Receptive language[d] Raw score 36.11 (6.37) Expressive language[d] Raw score 36.00 (8.60)	Speech imitation	PCC	Segmental
Nadig & Shaw (2015)	Prospective case control study	15	126	PIQ[i] 111 (17)	Core language[b] 108 (16)	Spoken sentences	Acoustic analysis	Suprasegmental
Patel, et al. (2020)	Prospective case control study	55***	198.84	Full scale IQ[i] 104.22 (12.03) PIQ[i] 102.88 (14.58)	Not stated	Spoken narrative	Acoustic analysis	Suprasegmental

Note. standard scores (standard deviations in brackets); standard scores and standard deviations combined for studies which reported separate diagnostic groups; WNL = within normal limits; IQ = intelligence quotient; MLU = mean length of utterance; NVIQ = nonverbal IQ; PIQ = performance IQ; PVSP = Prosody-voice coding using (Shriberg, et al., 1990).

*age not reported in months

** scores not reported

***included prelinguistic and/or adult participants

[a]Test of Nonverbal Intelligence–Third edition (TONI-3; Brown, Sherbenou, & Johnsen, 1997). [b] Clinical Evaluation of Language Fundamentals: CELF-3 (Semel, Wiig, & Secord, 2000), CELF-4 (Semel, Wiig, & Secord, 2003), CELF-P (Wiig, Secord, & Semel, 1992). [c]Peabody Picture Vocabulary Test—Revised (PPVT-R; Dunn & Dunn, 1981). [d]Mullen Scales of Early Learning (MSEL; Mullen, 1995). [e]MacArthur-Bates Communicative Development Inventory (CDI; Fenson, et al., 2007). [f]Leiter International Performance Scale – Third Edition (Roid & Miller, 2013). [g]Bayley Scales for Infant Development, Third Edition (Bayley, 2006). [h]Preschool Language Scales – Fourth Edition (PLS-4; Zimmerman, Steiner, & Pond, 2002). [i]Weschler Intelligence Scales: WISC-3 (Wechsler, 1992), WISC-IV (Wechsler, 2003), WPPSI (Wechsler, 2002), WASI (Wechsler, 1997).

Assessment methods for segmental speech of verbal participants. Connected speech samples were collected in five articles (Biller & Johnson, 2020; Chenausky et al., 2017a; Chenausky & Tager-Flusberg, 2017c; Dalton et al., 2017; Ellawadi & Ellis Weismer, 2015). All studies obtained connected speech samples during standard assessments of other skills such as language. Biller and Johnson (2020) collected a speech sample during the CSBS (Biller & Johnson, 2020). Ellawadi and Ellis Weismer (2015) tallied a child's consonant inventory from speech used during the ESCS (Mundy et al., 2003). Chenausky and colleagues (2017a; 2017c) collected 30-minute speech samples during the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000) at 18, 24 months, and 36 months. In the first article (Chenausky et al., 2017a), vocalizations were categorized as speech-like, nonspeech-like or obscured. Speech-like utterances were transcribed using broad transcription and consonant inventories tallied. In the second article, syllables that contained bilabial stops /b/ and /p/ were extracted from the samples and broadly transcribed (Chenausky & Tager-Flusberg, 2017c). Time waveforms and wideband spectrograms were used to ascertain the voice onset time for /b/ and /p/ word or word approximation. Finally, Dalton and colleagues (2017) sampled participant's connected speech during a four-part picture story retell from the VMPAC (Hayden & Square, 1999). Samples were assigned scores for language level and motor control. Number of utterances were not reported in any study.Five articles used standard assessments of speech imitation to assess the segmental speech of verbal children with ASD (Biller & Johnson, 2020; Chenausky et al., 2017b; Chenausky et al., 2019; Dalton et al., 2017; Nadig & Mulligan, 2017). Chenausky and colleagues (2017b; 2019) completed the first two sections of the KSPT (Kaufman, 1995). In the earlier study, phonetic inventory was tallied for each participant from the KSPT and a phonetic inventory test. In the later study, performance on the KSPT was coded for signs of CAS. Both Biller and Johnson (2020) and Dalton and colleagues (2017) completed the VMPAC (Hayden & Square, 1999).

Biller and Johnson (2020) reported on the participant's ability to imitate voiced [ɑ], voiceless [h], vowels, consonants, vowel sequences and syllables. Dalton and colleagues (2017) used a modified multi-modal version of the VMPAC to test a child's ability to imitate vowel and word sequences. Scores were assigned according to the assessment manual. Nadig and Mulligan (2017) reported scores from the Syllable Repetition Test (SRT; Shriberg et al., 2009). ***Suprasegmental speech***

Suprasegmental speech refers to the prosodic elements of speech, such as stress, pitch, rate, intonation, and loudness. Four articles assessed suprasegmental speech in verbal children with ASD, including three prospective case control studies (Arciuli & Bailey, 2019; Nadig & Shaw, 2015; Patel et al., 2020) and one single case experimental design (Akbari & Davis, 2019).

Assessment methods for suprasegmental speech of verbal participants. Akbari and Davis (2019) collected samples of 24-utterances and scored the participant's prosody using the Prosody Voice Screening Profile (Shriberg et al., 1990). Patel and colleagues (2020) used a wordless picture book to collect a sample of at least 20 spoken utterances for each participant. Intonation, rhythm, and rate were analyzed acoustically and perceptually in this study. Contrastive stress was studied by Arciuli and Bailey (2019) and Nadig and Shaw (2015). Arciuli and Bailey (2019) examined the use of lexical stress in a short single-word naming task of four highly familiar words. Nadig and Shaw (2015) used a referential communication task to elicit spoken sentences containing contrastive stress. Both studies reported results from acoustic analyses. **Discussion**

The purpose of this systematic review was to provide an update to the published review in Chapter 2 and to establish current research methods for assessing the speech of children with ASD. This information is important to guide future research and clinical practice in this area. A total of 27 articles including 617 participants met inclusion criteria.

There was a marked increase in the number of papers assessing the speech of prelinguistic children with ASD in this update compared with the original review. The expanded definition of 'prelinguistic' in this update may account for some of this increase, but not all. Contrastingly, fewer studies of verbal children with ASD were published in the intervening period.

Prelinguistic participants

There is a growing body of literature interested in the speech capacity of prelinguistic children with ASD, as evidenced by a more than two-fold increase in the number of articles reporting on these children in this review compared with the original review. Historically, children with ASD have been excluded from research studies if they were unable to complete standardized assessments, resulting in prelinguistic children being under-represented in the literature (Koegal et al., 2020; Plesa Skwerer et al., 2016). The increase in published articles may underscore a general shift towards understanding the capacity and trajectory of development of less verbal children with ASD. The rise of longitudinal studies stems from recent interest identifying early predictors for later expressive language development in prelinguistic children with ASD. Researchers, clinicians, and parents alike want to know which children will talk and what skills to develop to assist those at risk of remaining minimally verbal. Researchers are also beginning to examine which speech-based intervention approaches result in greater speech and/or expressive language outcomes for prelinguistic children with ASD, as indicated by a slight increase in treatment studies (Beiting & Maas, 2020; Chenausky et al., 2016; Chenausky et al., 2018).

Following the systematic review included in this thesis, best practice guidelines for a speech assessment of minimally verbal children with ASD recommend collecting data through spontaneous speech samples and phonetic stimulability (Broome et al., 2017). These were the predominant assessment methods used in articles in this review. Although one study reported results from a single-word naming task, this is currently not common practice with prelinguistic children. As children at this stage of language development produce fewer than 20 recognizable words, attempts at single-word naming will likely capture samples of prelinguistic speech (i.e. 'babble') rather than attempts to say real words. Comparing babble to adult targets is likely to yield little useful information and could result in misleading results.

Spontaneous speech samples continue to be the most common speech assessment method for children at the prelinguistic stage of development. Speech samples produce the most representative sample of prelinguistic speech (Paul et al., 2012; Rice et al., 2010). Most studies in this review collected a speech sample during a standard assessment of the child's communication ability. Less verbal children with ASD have been reported to produce more speech-like vocalisations during a standard assessment (i.e. CSBS; Wetherby & Prizant, 2002) than during unstructured play (Schoen et al., 2011) suggesting this to be an appropriate data collection method. This is in keeping with the results from the cross-sectional study presented in Chapter 3. Children with ASD at the prelinguistic stage of development in our study produced more transcribable vocalisations during the CSBS than free play, although not all prelinguistic children produced the intended 50 utterances (Broome et al., 2021). For reasons discussed in chapters 2 and 3, in this thesis, we included echolalia as it provided useful speech data. Unfortunately, no study in this updated systematic review commented on echolalia, no study reported the number of utterances in the sample, and only a few studies reported the length of time of the samples. Without standard reporting protocols it is impossible to compare samples between studies or to determine if adequate representative samples were collected.

Speech imitation tasks were used more frequently in this updated review compared to the previous one. Speech imitation may provide valuable information regarding a child's phonetic repertoire and sound stimulability that may not be ascertained during a speech sample alone. Some children with ASD may predominantly use learnt scripts with minimal phonological variation and like neurotypical children may have sound preferences and sound avoidance, both of which may limit the results of a spontaneous speech sample alone. Given that imitation deficits are often associated with children with ASD (e.g. Williams et al., 2001), speech imitation tasks may be overlooked by clinicians. This review highlights the possibility that speech imitation tasks may be a useful and appropriate speech assessment measure to complement spontaneous speech samples in prelinguistic children with ASD. Further research examining the use of appropriate speech imitation assessments for less verbal children (e.g., DEMSS; Strand et al., 2013) would be valuable.

Verbal participants

Fewer studies assessing the speech of verbal children with ASD were included in this update compared with the original review in Chapter 2. Considering 3 of the 12 studies also included prelinguistic participants (Chenausky et al., 2017b; Chenausky et al., 2019; Ellawadi & Ellis Weismer, 2015) and one study reported results of a single low verbal outlier (Biller & Johnson, 2020) from a previously discussed prelinguistic study (Biller & Johnson, 2019), the available new evidence for speech assessments with verbal children with ASD is limited.

The best practice guidelines published as part of the initial systematic review recommended collecting speech data for verbal children from single-word naming tasks, spontaneous speech samples, and speech imitation (Broome et al., 2017). Unfortunately, no study with verbal children in this update employed multiple speech assessment methods. Most published speech assessment guidelines recommend using multiple assessment measures (e.g., Bernhardt & Holdgrafer, 2001; McLeod & Baker, 2014; Stoel-Gammon & Williams, 2013). The paucity of available research using multiple assessment tasks may reflect the challenges assessing children with ASD. It is important to achieve a balance between time-efficiency and thoroughness of speech sampling. Relying on a single measure, however, may not provide the child with adequate opportunities to demonstrate their optimal communication ability.

Single-word naming tasks are recommended in a speech assessment of verbal children with ASD (Broome et al., 2017). No study in this update assessed the segmental speech of verbal children using a single-word task. Clinically, single-word naming tasks are used frequently, offering a time-efficient method of sampling a child's ability to produce phonemes in different positions in words and different word shapes (McLeod & Baker, 2014; Skahan et al., 2007). The lack of recent research implementing single-word naming tasks to assess the speech of verbal children with ASD is unexpected. There is evidence that children with ASD at this level of linguistic development can complete single-word naming tasks. In the original review, three studies used single-word naming tasks and in the longitudinal study in this thesis (Chapters 3 and 4), a number of single-word naming tasks were successfully used.

In contrast to the lack of single-word naming tasks, standard speech imitation tasks were employed in five studies providing further support for the use of these assessments with children with ASD. Comparing spontaneous and imitated speech provides important diagnostic information, only obtained if the speech imitation task is used in collaboration with either a naming task or spontaneous speech sample. Two studies of verbal children in this review (Biller & Johnson, 2020; Dalton et al., 2017) did just that, combining speech imitation assessment with samples of spontaneous speech.

Spontaneous speech samples are also recommended to assess the speech of verbal children with ASD (Broome et al., 2017). To date, the cross-sectional study in Chapter 3 of this thesis (Broome et al., 2021) is the only study to employ both single-word naming and spontaneous speech samples to assess the speech of verbal children with ASD. Five studies in this update collected speech samples during a standardised developmental or communication assessment, a method comparative to the one used in the longitudinal study reported in Chapters 3 and 4. Unfortunately, no study reported number of utterances, making it challenging to compare samples between studies. The guidelines presented in Chapter 2 and published in Broome et al. (2017) suggest collecting at least 50 utterances. This recommendation was aligned with best practice speech assessment recommendations at the time. Since then, new evidence has emerged suggesting 75 word tokens provides a sufficient sample to measure speech accuracy and complexity (Wren et al., 2020). The samples collected in the longitudinal study in this thesis (Chapters 3 and 4) far exceeded this 75-word recommendation. Given the challenges assessing children with ASD and the need for efficiency, these new recommendations provide promising evidence that shorter connected speech samples can still yield accurate measures of speech ability. It is important to note that the Wren et al. (2020) guidelines are based on a normative community sample of 776 5-year-old children from the Avon Longitudinal Study of Parents and Children (ALSPAC) and not specifically children with ASD. Although some children with autism may have been part of the ALSPAC sample, the number is likely to be small and equivalent, or possibly less, than the English prevalence figures (Roman-Urrestarazu et al., 2021).

Updated guidelines for the speech assessment of children with ASD

The results from the updated systematic review, together with findings from the longitudinal study, provide important new perspectives on speech assessment methods appropriate for children with ASD. Taken together with recent best practice guidelines for speech sampling, some adjustments to the guidelines for speech assessments with children with ASD, presented in Chapter 2, are provided below in Table 6.3.

Table 6.3 Guidelines for speech production assessment with children with ASD

Data Collection Method	Prelinguistic/ Minimally verbal	Verbal
Oromotor assessment	✓	✓
Spontaneous speech sample (≥ 50 speech-like utterances including echolalia)	✓	
Spontaneous speech sample (≥ 75 word tokens)		✓
Single-word naming (≥ 75 words)		✓
Speech imitation	✓	✓
Polysyllabic words		✓
Minimum Data Analysis		
Phonetic repertoire	✓	✓
Syllable shapes	✓	✓
Stress pattern analysis	✓	✓
Intelligibility rating		✓
Error analysis		✓
Additional Data Analysis		
PCC/PVC		✓
Consistency of production		✓
Imitated vs. spontaneous speech	✓	✓
Sentence imitation		✓

PCC = Percent Consonants Correct, PVC = Percent Vowels Correct

Conclusion

The speech of children with ASD, particularly less verbal children, has received more attention in the literature in the years between 2015 and 2021. This review identified 27 articles since 2015 that assessed the speech of children with ASD. A wide array of speech assessment measures were used, with spontaneous speech samples the most common assessment tool. Since the original review, a number of studies have employed multiple speech assessment measures, in line with best practice guidelines. Considering findings from the systematic review update and longitudinal study in this thesis, and advances in best practice speech assessment evidence, some changes have been suggested for best practice speech assessments with children with ASD. The use of standard speech assessment and reporting protocols will continue to advance this body of work in the future.

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Chapter 7: Discussion

Autism is a neurodiverse diagnosis encompassing a heterogeneous spectrum of individuals, each with their own inherent strengths and struggles. For much of history, the rigidly defined separate categories of 'infantile autism' and what became known as 'Asperger's syndrome' existed at polar ends of the continuum. Children who presented with symptoms between these two classifications were excluded from a diagnosis and thus from intervention. It wasn't until the late 1970s and early 1980s, when Lorna Wing married the diagnostic categories, that many children along the broad spectrum of autism were recognised (Wing & Gould, 1979; Wing, 1981). Scientific consensus for the autism spectrum Wing proposed wasn't reached until 2013, when the DSM-5 published the current diagnostic criteria for ASD (American Psychiatric Association, 2013). Researchers now have a responsibility to broaden our understanding of the entire autism spectrum. This is achieved only by embracing and describing the inherent variation that exists within the diagnosis.

This thesis began with my own clinical question: Do some children with ASD present with a speech sound disorder (SSD) which limits their verbal communication development? After noticing the many gaps in our knowledge regarding the speech of children with autism, this initial curiosity evolved into a desire to capture each child's optimal speech ability across the full autism spectrum and to explore what patterns emerge from this descriptive speech data. Previous speech research largely focused on infants later diagnosed with ASD, highly verbal older children, or homogenous samples. Like Georgiades et al. (2013), we saw the value in studying a heterogeneous cohort. This data has the potential to provide important information regarding the individual nuances and subgroup differences that exist within the spectrum.

The initial step in this research was to complete a systematic review of speech assessment practices from the literature. The results from this review, presented in Chapter 2, were used to provide researchers and clinicians with best practice speech assessment guidelines for children with ASD, and to guide the selection of speech assessments for the longitudinal study in Chapters 3 and 4. Chapter 5 expands on findings from the longitudinal study and details the unique communication profiles of the three children in Cluster B. An update to the systematic review in Chapter 6, examined changes in research practices since the original review, and described the current context in which to interpret the findings of the longitudinal study. Considering the findings from the longitudinal study and updated systematic review, some amendments to the speech assessment guidelines for children with ASD were proposed in Chapter 6.

Key findings

The research in this thesis adds valuable information regarding the speech capacity of children across the full autism spectrum. These studies are the first to: (1) provide best practice guidelines for the speech assessment of children with ASD, (2) report detailed speech data for a heterogeneous cohort of children with ASD and to describe subgroups based on this speech data, and (3) detail the trajectories of speech development for children with ASD. Three main findings emerged from this research:

1. A speech assessment can be completed with children on the autism spectrum, but specific modifications should be considered.
2. Speech subgroups of children exist within a heterogeneous autism cohort. A small subgroup have a unique communication profile consistent with a co-occurring SSD.
3. Speech subgroups had varied trajectories of speech development, particularly those initially described as having low language, low nonverbal communication, and low speech.

Speech assessments with children with ASD

The initial systematic review, reported in Chapter 2, examined speech assessments with children with ASD in 21 articles between January 1990 and December 2014. Wide variability in participant characteristics and reporting standards made it difficult to draw comparisons between studies and a review of the neurotypical SSD literature was also considered when compiling speech assessment guidelines for children with ASD. An update to this review, reported in Chapter 6, summarized speech assessment practices in 27 articles published between January 2015 and August 2021. What follows is separate recommendations for speech assessment of prelinguistic and verbal children with ASD based on the two reviews and reflections from collecting diverse samples from the children in the longitudinal study, reported in Chapters 3, 4, and 5.

Speech assessment recommendations with prelinguistic children with ASD

It is recommended that researchers and clinicians describe the prelinguistic speech of a child with ASD through a strengths-based assessment, rather than comparing the child's speech to an adult target. A combination of spontaneous speech samples and speech imitation tasks will likely yield the most representative sample. Traditionally, speech samples are taken during child-parent play interactions, however, Schoen et al. (2011) reported more success sampling the speech of children with ASD during a semi-structured assessment, such as the CSBS (Wetherby & Prizant, 2002). Considering this was the most successful method to obtain a speech sample in the present longitudinal study (Chapters 3, 4, and 5) and the method used in nine recent studies (Biller & Johnson, 2019; Ellawadi & Ellis Weismer, 2015; Garrido et al., 2017; Heymann et al., 2018; McDaniel et al., 2019, Saul & Norbury, 2020; Woynaroski et al., 2016; Woynaroski et al., 2017; Yoder et al., 2015), this approach appears appropriate. In the studies in Chapter 3 and 4 some prelinguistic children had difficulty reaching 50 speech-like utterances and a speech sample was taken during the entire assessment. This method is likely to be too time-consuming for clinicians, who may opt to continue to have a time limit (i.e. 20 minutes) if 50 utterances are not sampled. Although recent research suggests a minimum of 75 word tokens (different words) is sufficient for a representative speech sample, the recommendation was based on verbal 5 year old children. Given the results of the longitudinal study, a target of 75 speech-like utterances (word tokens) is out of reach for at least some prelinguistic children with ASD.

In addition to spontaneous speech samples, assessment guidelines also suggest phonetic stimulability with prelinguistic children with ASD (Broome et al., 2017). Speech stimulability is often achieved through imitation tasks. Prelinguistic participants in the longitudinal study had difficulty imitating sounds and syllables during the oromotor assessment. By contrast, speech imitation tasks were used to assess prelinguistic speech in eight articles in the updated review (Chapter 6), suggesting that speech imitation is both possible and informative with some prelinguistic children with ASD. Given this body of research, it is recommended that speech imitation at least be attempted with prelinguistic children with ASD.

A note about echolalia

Interestingly, no study in either systematic review described whether echolalia was ignored or included in speech sampling. Echolalia is defined as the immediate or delayed repetition of spoken language and is often used by children with ASD (Tager-Flusberg & Caronna, 2007). Echolalia was included in speech sampling methods in the longitudinal study here for two reasons. Firstly, determining what speech was echolalic, particularly for children at the prelinguistic stage of development or with limited speech accuracy, was predicted to be too challenging. Secondly, it was hypothesised that echolalia could represent the child's speech capacity, regardless of the linguistic and social communication differences to conversational language. These suspicions were supported by the findings of the longitudinal study and it is recommended that echolalia be used by clinicians and researchers as part of a representative sample of a child's speech.

Speech assessment recommendations with verbal children with ASD

Both independent and relational speech analyses are recommended for verbal children with ASD. As with prelinguistic children, there is value in describing the speech capacity of these children through strengths-based assessment and interpretation. For verbal children, this detailed description is largely lacking from published literature. The guidelines created and reported in Chapter 2, and updated in Chapter 6, suggest collecting representative samples of the child's speech through multiple assessment methods, including spontaneous speech samples, single word naming tasks, and phonetic stimulability. Although many studies of prelinguistic children in the updated review (Chapter 6) used multiple speech assessment methods, studies of the speech of verbal children largely relied on a single assessment measure (predominantly either speech imitation tasks or spontaneous speech samples). Multiple speech assessment measures were used to sample the speech capacity of verbal children in the longitudinal study, presented in Chapters 3, 4, and 5, and offered many benefits. Firstly, the use of multiple assessments provided detail that could not be obtained from a single measure (Morrison & Shriberg, 1992). For example, many children with ASD used learnt scripts, limiting the linguistic and phonological variation of a spontaneous speech sample. Further, some consonants and syllable shapes not sampled spontaneously were able to be captured during a single-word naming task. Finally, the variation in length and complexity of spontaneous speech samples made it difficult to compare samples from the same child over time (Stoel-Gammon & Williams, 2013). Single-word naming tasks provided a more consistent baseline for comparison.

A number of considerations emerged from the findings of the longitudinal study. While a single-word naming task was completed by all verbal children at Time 2, nine (47%)

verbal children with ASD responded best when naming tasks used photographs of common objects rather than symbolic images. Additionally, when collecting a spontaneous speech sample, some highly verbal participants produced far more than the recommended 50 utterances during a 10-minute sample, while other children required significantly longer time to reach this target. This added to an already lengthy assessment battery and resulted in less compliance from the children. Recent research suggests 75 word tokens provides a sufficient speech sample to measure phoneme accuracy and complexity (Wren et al., 2020). This is a welcome change to the published guidelines for the speech assessment of children with suspected SSD and will reduce the length of the required speech sample considerably for some verbal children. While these new sampling guidelines are yet to be researched with neurodiverse children specifically, it could be assumed that similar sampling procedures would be appropriate.

Summary of findings and implications of systematic reviews

The fundamental purpose of the original systematic review (Chapter 2) was to guide assessment selection in the longitudinal study (Chapter 3, 4, and 5). In the absence of high-level evidence on how to proceed with speech assessment of children with ASD, literature of speech assessment practices with neurotypical children was also considered. These results guided assessment methods of the primary study to focus on strengths-based assessment using independent and relational data analyses.

Since publication of assessment guidelines (Broome et al, 2017), a number of well-designed studies with best practice assessment methods are beginning to emerge, particularly with less verbal children; these are reviewed in Chapter 6. This new evidence and the results from the longitudinal study were considered and amendments to the guidelines proposed. Overall, children with ASD are not afforded the same rigour in SSD assessment as neurotypical children. This is the challenge for researchers and clinicians alike. As more research studies attempt to implement best practice methods and feedback the results, these guidelines can evolve to meet the unique needs of children with ASD.

Speech subgroups

The results from the longitudinal study, describing the speech capacity and development of a heterogeneous cohort of children with ASD, are reported in Chapters 3, 4, and 5. Findings from the Time 1 cross-sectional study, presented in Chapter 3, suggest three subgroups of children with distinct speech profiles: (a) children with high language, nonverbal communication, and speech (n=10); (b) children with high receptive vocabularies, high nonverbal communication, low expressive vocabularies, and low speech (n=3); and (c) children with low levels of language, nonverbal communication, and speech (n=10). As subgroups were formed using hierarchical cluster analysis they are referred to as Cluster A, Cluster B, and Cluster C and are described further below.

Cluster A: High language, nonverbal communication, and speech

The children in Cluster A presented with relatively high receptive and expressive vocabularies, use of gestures and speech development. As many children in this subgroup reached ceiling on measures of vocabulary and gestures and had near complete consonant repertoires at Time 1, little growth in their communication skills could be captured on these measures. The study is limited in this regard. Past research has reported a group of children with ASD with average and even advanced speech skills (Kjelgaard & Tager-Flusberg; Rapin et al., 2009), and it is likely that most children in Cluster A represent these children. It is possible that a small number of children in Cluster A may present with mild speech delays as reported in previous research (Cleland et al., 2010; Shriberg et al., 2001; Shriberg et al., 2011). Describing articulation and phonological errors in future research is important.

Cluster B: High receptive, high nonverbal communication, low expressive language, low speech

The three children in Cluster B presented with an unexpected communication profile. Specific details regarding the communication abilities of these three children was provided in Chapter 5. Their strengths were comprehension of words and use of gestures to augment their communication. Children in this subgroup used very few spoken words and appeared to have limited speech capacity. Specifically, their speech was characterized by very few consonants and vowels, high number of single vowel syllables, and profoundly limited consonant and vowel accuracy. A speech sound disorder (SSD) was likely a core barrier to their verbal communication, although differentially diagnosing a specific SSD is challenging.

Cluster C: Low language, nonverbal communication, and speech

Ten children were grouped into Cluster C at Time 1. These children presented with low receptive and expressive vocabularies, low number of gestures and low speech ability. Children in Cluster C are likely representative of children with ASD with low speech and low language reported in previous research (Chenausky et al., 2019; Rapin et al., 2009). Most of the Cluster C children could be defined as ‘minimally verbal’, a term used in a growing number of research studies and defined as using fewer than 20 spoken words (Chenausky et al., 2019; Kasari et al., 2013; Thurm et al., 2015; Yoder et al., 2015). Two participants in this subgroup used more than 20 words and some word combinations. These two children were more representative of ‘low verbal’ children in previous literature (Chenausky et al., 2019).

Trajectories of Speech Development

The 22 participants who were followed longitudinally had varied trajectories of speech development over 12 months (Chapter 4). Children in Cluster A and Cluster B remained in the same subgroups at Time 2. As the children in Cluster A reached near ceiling on many communication measures at Time 1, little growth could be captured. As mentioned above, this study is limited in this regard. The children in Cluster B improved in their receptive vocabularies and use of gestures. Their speech and expressive vocabularies developed very little across the 12 months, adding further support for a suspected SSD (Chapter 4 and 5). The most growth was seen in some of the Cluster C children, those described as having low language, nonverbal communication, and speech at Time 1.

Nine of the 10 Time 1 Cluster C children were followed longitudinally (Chapter 4) and had three vastly different communication trajectories across the 12 months: (1) one participant improved across all aspects of communication and joined Cluster A at Time 2, (2) five participants formed Cluster D at Time 2 and had the most communication growth of any subgroup over the 12 months, and (3) three participants had very little communication development and remained nonverbal at Time 2. The three children who remained nonverbal (Time 2 Cluster C) and the five children who improved across all communication domains and were verbal at Time 2 (Time 2 Cluster D) differed only in the number of consonants in their sound repertoire at Time 1. This result suggests that the consonant inventory of low verbal children with ASD may be an important predictor of later communication development, as previously suggested in the literature (Saul & Norbury, 2020; Wetherby et al., 2007; Yoder et al., 2015). Importantly, children with few consonants at Time 1 remained minimally verbal at Time 2. Consonant inventory alone did not predict cluster membership, however. For example, Time 2 Cluster B and C children had comparable consonant inventories at Time 1. Instead, a combination of a child's receptive vocabulary and consonant inventory may have the potential to predict a child's communication profile and possible speech development trajectory. These are only preliminary descriptive results and further research is needed to explore these hypotheses with larger cohorts of children.

A note about verbal regression

Regression refers to loss of previously established skill. The existing literature reports approximately one third of children with ASD have a history of regression, usually occurring in the second year of life (e.g., Boterberg et al., 2019). The parents of ten participants (43%), including one Cluster A child, all three children from Cluster B, and six from Cluster C, reported a history of verbal regression. Although most literature on regression is based on such retrospective parent report, this method is limited by several factors, such as accuracy of parent recall and the wording of the questions (Dawson, 2011; Lord et al., 2004). Very few prospective longitudinal studies of children with ASD have captured data of regression (Boterberg et al., 2019). The longitudinal study in this thesis (Chapters 4 and 5) adds valuable information. It is important to note that some apparent regression in this study was unlikely to be true loss of skill. Some children's consonant and vowel accuracy measures reduced over 12 months, most likely reflecting an increase in attempts at new sounds and word forms. For example, some children produced predominantly consonant-vowel syllables at Time 1 and very few polysyllabic words, but by Time 2 were using post-vocalic consonants, consonant blends, and attempting multisyllabic words. For some children, this development resulted in reduced speech accuracy, although this is likely to only be a temporary reduction as the child's phonological system develops. As previously mentioned in Chapter 5, all three children in Cluster B replaced some of their Time 1 consonants with different consonants at Time 2. As children in Cluster B used consonants infrequently, it is possible that previously learnt consonants were simply not captured in the speech sample at Time 2. Alternatively, difficulty retaining previously learnt sounds may reflect an underlying and significant SSD.

One Cluster C child (Participant 23) regressed verbally during the 12 months of the longitudinal study (Chapter 4). At Time 2, Participant 23 was reportedly using only six words (loss of 16 words) and produced only three consonants (loss of five consonants). No words were recorded during the Time 2 assessment. This verbal regression occurred in the context of improvements in receptive vocabulary and use of gestures. This is important data demonstrating verbal regression, between 27 and 39 months of age, to add to the limited body of prospective evidence of regression.

Limitations

There are three primary limitations to note when interpreting the findings from the studies in this thesis. Firstly, this thesis deliberately aimed to describe a heterogeneous cohort of children with ASD. To achieve this goal, assessment measures appropriate for a wide range of ages, developmental abilities, and linguistic levels were needed. Unfortunately, there was a paucity of valid language assessment tools of this nature. While the PLS-4 theoretically covered the age ranges of our cohort, children at very early stages of development were unable to engage in this assessment. Future development of a language measure appropriate for use with both prelinguistic and verbal children with ASD would further expand our understanding of the heterogeneous nature of the autism spectrum (Georgiades et al., 2013; Kasari et al., 2013; Plesa Skwerer et al., 2016). Instead, multiple assessments were required (CSBS and PLS-4). These assessments measured different aspects of language and results were unable to be compared.

To ensure one consistent language measure was used across the entire cohort, a parent checklist of the participant's vocabulary (CDI; Fenson et al., 2007) was employed. Many children in Cluster A reached high levels or ceiling on this measure, and therefore, little change in language was able to be recorded. It is possible that the children in Cluster A are more varied than what was captured in our study. Future study of these children, using assessment measures sensitive to their level of functioning, would be valuable.

Finally, the small sample size in the longitudinal study limited the statistical approach that could be employed to analyse the data. It is hoped that the preliminary descriptive results from the studies in this thesis provide a starting point for larger cohorts to be studied in the future.

Future Directions for Researchers

There is a subgroup of children with ASD for whom a speech deficit is a core feature of their communication impairment. More detailed speech descriptions of these children, including comparisons of imitated and spontaneous speech, sequencing speech sounds, and consistency of productions, are needed to differentially diagnose the specific SSD. Researchers have two options moving forward. Firstly, further prospective and longitudinal studies with larger cohorts of children are needed to expand the results presented in this thesis. It would be interesting to explore whether similar subgroups of children emerge in larger heterogeneous cohorts. The second option is to progress with treatment studies, with response to intervention informing diagnosis. Some researchers have already begun to investigate the outcomes of speech-based intervention with minimally verbal (Beiting & Maas, 2020; Chenausky et al., 2016, 2018) and verbal (Akbari & Davis, 2019; Chenausky et al., 2017b) children with ASD. Both options for future research are important and needed.

Clinical Implications

In the meantime, clinicians do not have a choice to wait to gather data and diagnose prior to treatment. For clinicians, a dynamic assessment and implementation of intervention, which may inform diagnosis, is the only option. Differentially diagnosing a specific SSD in minimally verbal children with ASD is challenging. For example, to differentially diagnose a phonological speech disorder from a motor speech disorder, it is important to complete an oromotor assessment and to obtain a child's spontaneous and imitated speech, in addition to repeated attempts at the same target. We encourage clinicians to attempt all aspects of a core assessment outlined in Chapter 6 but recognise that with minimally verbal children with ASD a complete assessment will not always be possible. Our research highlights four essential components of a communication assessment with children with ASD and a suspected SSD which should be considered by clinicians:

1. Capturing the speech capacity of a child with ASD requires a strengths-based assessment, which may not compare their speech to an adult target.
2. A speech assessment may be most important for minimally verbal and low verbal children with ASD
3. A child's early consonant repertoire and receptive vocabulary may be predictors of communication growth and should be measured.
4. A child's use of gestures and nonverbal communication is important and should be measured.

Conclusion

The research in this thesis began with a specific clinical question regarding the possibility of an SSD in some children with ASD but evolved into an exploration of the speech ability of children across the entire autism spectrum. We became interested in how to capture and describe the speech of children with autism, and the patterns that may emerge from such descriptive data. The published systematic review and updated review provide research and clinical best practice speech assessment guidelines for children with ASD. Although there is a growing body of research studying the speech of children with ASD, consistent assessment and reporting standards are needed to advance this evidence.

The longitudinal study reported in this thesis is the first to describe the speech development of children with ASD. This study deliberately recruited a heterogeneous cohort of children, rather than preselecting participants based on low language levels or suspected speech deficits. The findings provide valuable preliminary information regarding individual and subgroup speech differences in the heterogeneous cohort of children with ASD. The subgroup of children who initially presented with low language, nonverbal communication, and speech had varied communication trajectories. Some children with low communication ability made significant gains and were talking by the end of the study. Other children remained nonverbal. Further, there is a small subgroup of children with ASD who present with a unique communication profile indicative of a co-occurring SSD. The speech impairments of these children appear to be a core barrier to their verbal communication development. More information is needed to differentially diagnose the SSD of these children. The combination of a child's receptive vocabulary and consonant repertoire may predict speech development. Future research with larger cohorts is needed to explore these preliminary findings.

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