Speech-, Fine- and Gross- Motor Control in Children with Autism Spectrum Disorder

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PhD Thesis

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Abstract

Motor impairment is argued to be central to ASD, however, its interaction with speech motor control has not been studied in-depth. This study examined this interaction by investigating why higher rates of speech sound errors are identified in children with ASD and whether this could be related to a single underlying motor impairment. A small number of studies found residual and non-developmental speech errors are significantly higher in children with ASD (33-40%) than the normal adult population (1-2%; Cleland, Gibbon, et al., 2010; Shriberg et al., 2001). Others argue that speech follows a typical developmental trajectory (Kjelgaard & Tager-Flusberg, 2001). In this study ten children with ASD and ten age and gender-matched typically developing peers aged 6-12 years were compared. Behavioural assessments were carried out to examine nonverbal IQ, language ability, gross, and fine motor control. These were correlated with clinical assessments of speech in both single syllabic and multisyllabic contexts. Speech motor control was measured using a Diadochokinesis (DDK) task recorded with simultaneous ultrasound tongue imaging and acoustic recordings. The analysis carried out looked at tongue shape variation and mean syllable duration at slowest and fastest syllable repetition rates.

There were no correlations between DDK measures in the ASD group with movement and language, speech, non-verbal IQ, and autistic symptomatology. However, correlations were found within the subtests. There were no significant differences between the TD and ASD group in maximum rate, consistency, or accuracy of the DDK tasks. When using ultrasound to measure tongue shape variance, surprisingly, the TD group had more significant differences of tongue shape in the more motorically complex sequences (tk and ptk) than the ASD group. While children in the ASD group had significantly poorer motor performance in the movement assessment, this did not correlate with the in-depth analysis of speech motor control. Children in the ASD group often performed with less variability in the DDK tasks than the TD group, suggesting rigidity in motor plans. The results indicate that while no speech motor impairment was present, there were indicators that children with ASD had difficulty with speech attunement, being unable to sufficiently attune to the ambient speech environment. The presence of a significant fine and

gross motor impairment as well as impairment in language may further impede speech sound development.

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List of Abbreviations

ASD	Autism spectrum disorder
TD	Typically Developing
SSE	Speech Sound Errors
SLT	Speech and Language Therapist
CELF	Clinical Evaluation of Language Fundamentals
Leiter	Leiter International Performance Scale
DEAP	Diagnostic Evaluation of Articulation and Phonology
CUW	Clinically Useful Words
DDK	Diadochokinesis

Chapter 1 Introduction

1.1 Rationale

There are higher rates of speech sounds errors (SSEs) in children with autism spectrum disorder (ASD), but we do not understand why. This thesis studies these higher rates of SSEs found in children diagnosed with ASD compared to typically developing peers (TD). Through this study, I seek to understand the interactions between speech sound production and other behavioural domains such as movement, language, and non-verbal cognition to understand why these higher rates of SSEs may be occurring.

Children with ASD have difficulties with social interaction and communication. For some of these children, despite having good language skills, their speech is characterised by errors normally found in younger children (speech delay); unusual errors; or imprecise speech (Cleland, 2010; Shriberg et al., 2011; Wolk & Brennan, 2013). While these speech sound errors are around three times more often found in children with ASD than those without, the cause is not well understood. One theory suggests that the SSEs might be caused by a difficulty coordinating the muscles for speech, in other words, a speech motor control problem (Belmonte et al., 2013). Difficulties with coordinating the rest of the body, for example coordinating finger movements (i.e., fine motor control) and larger body movements in running, jumping (i.e., gross motor control) are already well described in ASD. However, no research links difficulty with speech movements to difficulty with fine or gross motor control in children with ASD. This is further compounded by our lack of understanding of whether there is a direct link between the motor systems required for speech (speech motor control) and the gross and fine motor control of other parts of the body. It is still in debate whether the movements required for speech are task-specific (Ziegler, 2003b) or share the same neurological pathways and correlates as fine and gross motor control (Ballard et al., 2003). This study aimed to take measures of speech motor control and compare the consistency, accuracy, and rate of speech tasks to the overall performance of fine and gross motor control to try to identify any correlations that may reveal a link between these domains.

This thesis also explored why there may be discrepancies in the literature. In part, the difficulties in investigating speech motor control for comparison with fine and

gross motor control may be due to the difficulty in accurately measuring speech movements. The tongue, the main articulator, is largely hidden from view during speech and requires specialist technology to capture these movements for measurement. Until recent years, clinicians and researchers have relied on perceptual assessment to identify symptoms of speech motor control difficulties rather than making a direct observation. To take us further in our understanding of speech motor control in ASD, this project used perceptual assessments that are used as standard in Speech and Language Therapy clinics and compared these to the instrumental analysis of speech sound production and speech motor control. This allowed direct comparison of these two techniques to explore if instrumental analysis through ultrasound tongue imaging is more effective at identifying SSEs in children with ASD than commonly used perceptual assessment.

Furthermore, this project studied if an interaction exists between speech sound production and other behavioural domains such as non-verbal cognition, language, and movement (both fine and gross motor control). Understanding if there is a correlation between speech sound production and these behavioural domains allows us to see if there is a cognitive domain that may be causal or have a key role in the SSEs present in the speech of some individuals with ASD. Determining the nature of SSEs in ASD will help speech and language therapists (SLTs) choose appropriate speech therapies for children with ASD as it allows clinicians to make choices of intervention based on the root cause of the SSEs rather than the presentation of symptoms. The findings of the project contribute to our understanding of the underlying cause of SSEs in ASD.

1.1.1 Speech Sound Errors

ASD is a neurodevelopmental disorder characterised by persistent differences in social communication and social interaction across multiple contexts (World Health Organisation, 2017). There have been conflicting findings on whether children with ASD have significant numbers of SSEs or typical speech.

In this study, I define SSEs as any non-adult-like production of sounds in words, including the substitution of another sound, omissions, additions, distortions or syllable-level errors (American Speech-Language-Hearing Association (ASHA), 2007). Errors may be articulatory (phonetic) or linguistic (phonological). Articulation

errors are motor-based impairments in which the SSEs are a result of the difficulty in having the speech articulators' function to produce the target sound in multiple speech contexts. Examples include distortions such as a lateral lisp. Phonological errors are rule-based errors, i.e., velar fronting where a sound typically produced further back in the mouth (/g/) is produced at the front of the mouth [d]. These phonological errors occur when the child has difficulty organising the patterns of sounds at a cognitive level and are not a result of difficulty with the speech motor system. SSEs of both articulatory and phonological basis were included within this project's definition of SSEs and later each SSE that was present were analysed individually and determined whether the error was likely to be articulatory or phonological.

Another important distinction that is made when analysing SSEs is whether it is delayed or disordered. This study followed the guideline set out by Dodd (2011) in which she studied the differentiation between speech delay and speech disorder. She defined that a speech delay was the presence of a pattern of SSEs that was typical for children younger than the speaker. Whereas a speech disorder was a pattern of SSEs in which errors are not typical for any age group with normal speech development. Table 1 displays common SSEs, what age they are expected to be eliminated by and the nature of these errors developed from multiple sources and used as a basis for analysis and discussion of SSEs in this project (Hodson, 2004; McLeod & Baker, 2017; Peña-Brooks & Hedge, 2015). SSEs that are atypical fall into the speech disorder category, and any produced after the age of elimination as speech delay.

Table 1: *Typical and Atypical Speech Sound Errors*

Speech Sound Error	Definition	Age of Elimination (years)
Backing	The sound produced at front of mouth is produced further back in the mouth	Atypical
Fronting	The sound produced at the back of the mouth is produced further forward in the mouth	3.5
Gliding	Specific consonant is replaced by /w/ or /j/	6
Stopping	Continuant consonant is replaced with a stop	/f/ and /s/ 3 /v/ and /z/ by 3.5 /ʃ/ and /t͡ʃ/ by 4.5 /θ/ by 5
Vowelisation	Sounds replaced by a vowel	Atypical

Affrication	Sound replaced by an affricate	3
Deaffrication	An affricate is replaced by a stop or fricative	4
Alveolarization	Sound replaced with an alveolar	5
Depalatalization	Palatal sound replaced with nonpalatal sound	5
Labialization	Sound replaced with labial sound	6
Cluster Reduction	Cluster of consonants is reduced to a single consonant	5
Final Consonant Deletion	Final consonant is omitted	5
Initial Consonant Deletion	Initial consonant is omitted	Atypical
Weak Syllable	Weak syllable is omitted	4
Epenthesis	A sound is added between consonants	8
Assimilation	Target consonant sounds like another sound in the word	3
Denasalization	Nasal sound is replaced by a non-nasal sound	2.5
Final Consonant Devoicing	Final consonant replaced by a voiceless consonant	3
Prevocalic Voicing	Voiceless consonant replaced by voiced	6
Coalescence	Sound is substituted for a sound with similar features	Atypical
Reduplication	Repetition of sound or syllable	3

1.1.2 Speech Sound Errors in ASD

Small studies of between ten to twenty children, including case studies of three children, have revealed that children with ASD exhibit a variable pattern of SSEs. This includes phonological processes and articulatory errors that are common in typical speech development; persistence of these SSEs beyond the expected age, and unusual speech sound changes (Wolk et al., 2016; Wolk & Brennan, 2013). When phonetic and phonological inventories of children with ASD have been examined, there were high levels of speech delay in which developmentally earlier sounds are produced more often than developmentally later sounds (McCleery et al., 2013). This may be due to later sounds being more articulatory complex, requiring a mature and efficient speech motor system. It also suggests that at a group level phonological development may be delayed and a subset of children with ASD have SSEs.

Larger scale studies have also shown similar patterns of SSEs in children with ASD (Cleland et al., 2010; - 69 children, Rapin et al., 2009 - 62 children; Schoen et al.,

2011 - 64 children). A higher prevalence of speech delay and an increased number of SSEs have been noted in children with ASD (Shriberg et al. 2011). In contrast, a larger-scale study of 89 children carried out with children with ASD aged between 4-14 years concluded that their speech skills were relatively spared (Kjelgaard & Tager-Flusberg, 2001). However, in this study, they only examined isolated speech sounds in single words and did not examine SSEs in other speech contexts. To challenge this, the current study looks at speech in various contexts (single words, multisyllabic words, and maximum performance tasks) and describes the type of SSEs produced by categorising them into typical SSEs, delayed SSEs and disordered or atypical SSEs. These are indicated in table 2. An SSE that is “typical” is produced by the child of the age stated in the table or younger, e.g., a child of three years fronting /g/ to /t/. An SSE that is “delayed” is produced by a child that is older than the age defined in the table, for example a child of five years producing the same error, fronting /g/ to /t/ but is now at an age where this is expected to have been resolved. An atypical SSE is producing an error that is not a part of typical speech development, these are indicated in table 1, for example, backing /t/ to /g/.

Previous studies have used perceptual methods of assessment, where the SLT listens to the speech and transcribes errors. This is problematic, as perception-based phonetic transcription is known to be unreliable (Howard & Heselwood, 2011). Identification of speech errors using instrumental analysis leads to more accurate diagnosis and better therapy planning (Howard & Heselwood, 2011). This study used ultrasound tongue imaging to make direct observations of the tongue during speech. Perceptual analysis is unable to identify silent movements of the tongue or some atypical SSDs (McCann & Wrench, 2007). Ultrasound tongue imaging (UTI) allows us to investigate the presence and type of speech errors produced by imaging from near the tongue tip to the root. I compared the speech profile of children with ASD to typically developing children. Speech development of typically developing children is already well described in the literature, therefore providing an effective comparison for my smaller study where it was not possible to recruit large numbers of typically developing children (Dodd et al., 2004). However, this is only in normed behavioural tests and not the instrumental measures applied in this study (e.g., ultrasound). Therefore, the instrumental measures were carried out with both groups of children.

1.1.3 General Motor Abilities in ASD

Movement (or motor) abnormalities have been reported across a wide range of children with ASD (Esposito & Pasca, 2013). The motor differences that have been identified do not directly depend on a linguistic or social development issue, arising in their own context (Esposito & Pasca, 2013). Motor differences have been consistently reported across the spectrum but are not listed as a core diagnostic criterion. Some of the differences identified have included differences in fine and gross motor control. Fine motor control being the coordination of muscles, bones, and nerves to produce small, exact movements and in gross motor control which is the ability to control large, general movements and balance. Motor difference in ASD was confirmed in a meta-analysis, finding the presence of substantial motor coordination deficits in ASD (Fournier et al., 2010). In this meta-analysis, they found that ASD was associated with higher rates of clumsiness, motor coordination differences, instability in posture, and out with the norm performance on standardized tests of motor functioning (Fournier et al., 2010). Paediatric studies have found that both small (fine motor) and large (gross motor) movements are impaired in this group (Hellendoorn et al., 2015). There is no universal motor symptom or motor bioindicator that identifies ASD, but studies have suggested that motor dysfunction may play a significant role. One theory is that impairments in ASD are rooted in the incapacity to assemble and directly grasp the intrinsic goal-directed organisation of motor behaviour (Esposito & Pasca, 2013). Therefore, in addition to looking at SSEs, this study examines the interaction of speech sound production with general motor difficulties.

The production of speech sounds involves intricate coordination of the speech muscles (including the tongue) and is required for producing accurate, clear speech (Gracco, 1994). Movement atypicality observed in ASD, therefore, may also be evident in their speech motor control and speech sound production, however, this has not been investigated robustly. This study investigates this using standardised assessments of movement used in clinics. The Movement Assessment Battery for Children, 2nd edition (Brown & Lalor, 2009) allows examination of movement abilities at both the gross motor and fine motor level to determine if there is a breakdown in either or both domains. Comparing results to speech sound production through

statistical analysis allows us to determine if an interaction exists. If an interaction is found it indicates that the higher rates of SSEs found in children with ASD could be due to a speech motor impairment, similar to that present in childhood apraxia of speech, where there is a breakdown in the control and planning of speech motor systems resulting in a speech impairment (American Speech-Language-Hearing Association (ASHA), 2007). Finding a relationship between speech motor control and gross/fine motor control might indicate that there is an overlap of the sensory-motor subsystems required for coordination of muscles for speech and other movement tasks involved at other parts of the body (Ballard et al., 2003). However, if no interaction is found, it provides support for the task-specific theory of speech, in which speech articulators have sensory-motor subsystems that are task-specific (Ziegler, 2003a). These subsystems have unique properties that have specialised neural circuitry that does not directly overlap with the sensory-motor subsystems used for different aspects of fine and general motor control.

1.1.4 Interaction with other Behavioural Domains

In addition to motor abilities, this study investigates the interaction between SSEs and language in children with ASD. There is an inherent link between expressive language abilities and speech sound development. Research suggests that infants develop language-specific speech sound perception as early as the first year of life (McCleery et al., 2006), However, there is conflicting evidence regarding whether language skills impact speech sound development in children with ASD (Cleland, 2010; Wolk & Edwards, 1993; Wolk & Giesen, 2000). It has been suggested that children with ASD who are more severely language-impaired exhibit increased rates of SSEs (McCleery et al. 2006). However, very little research has been carried out to investigate this.

Therefore, this study uses standardised language assessment in the form of the Clinical Evaluation of Language Fundamentals 4th Edition (Semel et al., 2003) to determine the interaction between speech sound production and language in children with ASD. Examining language and movement planning skills in children with ASD helps describe accurately the range of mental processes that may be impacting their speech sound production. Determining the nature of the SSEs and

underlying causes in ASD will help speech and language therapists to choose appropriate speech therapies for children with ASD. The findings of this project will contribute to our understanding of the underlying cause of these higher rates of SSEs in this population.

1.2 Contribution to Research

SSEs can have profound effects socially and educationally. Even mild speech difficulties can have a negative effect on how children are perceived by their peers (Peterson et al., 2009). There are significantly higher rates of SSEs in ASD compared to the general population, the reason for this is still unknown (Cleland et al., 2010; Shriberg et al., 2011). This study seeks to determine if SSEs in ASD are due to an underlying speech motor control impairment by exploring maximum performance tasks, where the child produced target syllables and sequences at increasing speeds.

Ultrasound is non-invasive and produces images of the tongue during speech. It shows a live image of the tongue in the midsagittal view and records tongue movements during continuous speech that is synchronised with an audio recording. This allows analysis of otherwise unobservable movements of the tongue at sub phonemic levels of speech. This has not previously been used to research speech motor control in ASD.

1.3 Aims of Thesis

This thesis aimed to determine whether the higher rates of SSEs found in children with ASD were a result of a speech motor disorder and whether this was related to a general movement disorder. This study used varied analysis techniques, both behavioural and instrumental, to determine if children with ASD produce significantly higher rates of SSEs than typically developing (TD) children and if there were any correlations with movement, language, and non-verbal cognition.

The behavioural assessments aimed to first analyse whether there were higher rates of SSEs in children with ASD in multiple speech contexts (single words and

multisyllabic words) compared to TD children. Care should be taken in the interpretation and generalising of these findings due to the small sample size (ten children with ASD) and due to the heterogeneity of the condition of ASD itself, where individual children's presentation of autistic symptomatology can vary significantly (Pelphrey et al., 2011). The instrumental assessments aimed to analyse whether instrumental analysis of speech reveal subtle articulatory differences between children with ASD and TD children. Second, in combination with the speech results in the behavioural assessment analysis, it aimed to determine whether this sample of children with ASD presented with speech motor impairment symptoms.

I focused this study on two theories that may offer explanation as to why higher rates of SSEs are present in ASD: a) the speech attunement framework and b) deficits in speech motor control. Both of these perspectives intersect and result in SSEs in individuals with ASD and this is discussed in relation to the key findings. Both perspectives could have a relation to the comorbidities of motor deficits and perceptual issues often identified in individuals with ASD. In order to understand this further, the study presented here addressed three research questions, discussed below.

Research Question 1: Do children with ASD produce significantly more speech sounds errors (SSEs) compared to typically developing children?

Hypothesis: Children with ASD produce more SSEs than typically developing children

Analysis: I compared percentage consonants correct (PCC) from the Diagnostic Evaluation of Articulation and Phonology (DEAP; Dodd, 2002) between groups. I then carried out an independent samples t-test to compare the ASD group and the control group for this analysis as and predicted that children with ASD would have significantly lower PCC.

Research Question 2: Does instrumental analysis of speech reveal subtle articulatory differences between ASD and TD groups?

Hypothesis: Instrumental analysis reveals more subtle SSDs than perceptual methods of assessment.
Analysis: To answer this research question identification of speech problems using the instrumental method (ultrasound tongue imaging) and the perceptual method (DEAP; Dodd, 2002) was required. To determine this, I analysed variation of tongue curves using ultrasound data taken from the DDK task of children with ASD and typically developing children and highlight if subtle speech motor difficulties or impairments are identified using UTI but not in the speech perceptual results of the DEAP and DDK.

Research Question 3: Do children with ASD present with speech motor impairment symptoms?

Hypothesis: There are a subset of children with ASD who present with speech motor control difficulties
Analysis: I compared rate, accuracy, and consistency perceptually across groups (and to published norms) in a diadochokinesis test (DDK- rapid alternating syllables such as pa ta ka). I then carried out an independent samples t-test for this analysis. I predicted children with ASD would have lower consistency and accuracy scores than TD children.

Chapter Two Literature Review

2.1 Introduction

People with autism spectrum disorder (ASD) experience higher rates of speech sound errors (SSEs) than their peers (Cleland, Gibbon, et al., 2010; Shriberg et al., 2011) but the reasons why are unknown. This chapter explores the current literature on the condition of ASD, as well as the literature on SSEs produced by individuals with ASD. This study aimed to understand why higher rates of SSEs occur in ASD and to move the debate forward from not just whether they exist but to why they exist. Recent studies using varied instrumental analysis techniques show that children with ASD produce significantly higher rates of SSEs than typically developing (TD) children. These are discussed in detail alongside a critique of the methods historically used to assess SSEs in this population.

This chapter proposes two theories that may offer explanation on why these higher rates of SSEs are present in ASD: a) the speech attunement framework and b) differences in speech motor control. This chapter discusses how both of these perspectives may intersect and result in SSEs in individuals with ASD. Both are discussed in relation to the comorbidities of motor differences and perceptual issues often identified in individuals with ASD.

This literature is then grounded in the research questions proposed for this current study and the chosen methods to answer these questions are discussed. This chapter concludes that there is a need to look at both motor and speech production abilities in individuals with ASD so we can describe accurately why SSEs are occurring in the context of the cognitive and neurophysiological processes that may be impaired.

2.2 Current Understanding of Autism spectrum disorder (ASD)

ASD is a neurodevelopmental disorder characterised by persistent differences in social communication and social interaction across multiple contexts (World Health Organisation, 2017). The DSM-5 (American Psychiatric Association, 2013) focuses on a dyad of symptoms: a difference in social interaction and communication and

restricted and repetitive patterns of behaviour. There is a frequent co-occurrence of verbal and non-verbal impairment in children with ASD (Noterdaeme et al., 2002). This includes difficulties in speech, language, and movement. The challenge for clinicians and researchers is to identify syndrome specific differences, particularly in a heterogenic condition like ASD. Understanding syndrome specific differences may help our understand of whether subtypes exist within ASD.

First described in seminal reports in 1943 by Leo Kanner (1943) and in 1944 by paediatrician Hans Asperger (Asperger, 1991) understanding of ASD has substantially grown since. It is now recognised as a neurodevelopmental disorder that is a highly heterogeneous and can occur with multiple comorbidities. Despite progress in understanding of ASD, a cause has not yet been identified. Multiple aetiologies have been suggested but no single environmental or genetic cause has been identified (Abrahams & Geschwind, 2008). Common genetic variants can only be used to explain a fraction of the phenotypes (Stein et al., 2013). The heterogeneity in ASD is perhaps one reason why is a high incidence condition. The first epidemiological study of ASD reported an incidence of 4.1/10000 in the UK (Lotter, 1966). The rates of ASD have been increasing in recent years, which may be a result of changing diagnostic process and criteria (Fisch, 2012).

Prognosis without early intervention for individuals with ASD is poor. Studies carried out before widespread early intervention programmes revealed 58-78% of individuals with ASD had poorer outcomes than the typical population in: independent living, educational attainment, and employment and peer relationships (Lai et al., 2014). Better outcomes have been predicted for children who have developed communicative phrase speech before six years and have fewer social impairments (Billstedt et al., 2005; Howlin et al., 2004, 2014). Early identification of ASD can improve opportunities for young children to benefit from intervention, preventing avoidable mental health problems and reducing considerable family and societal costs (Järbrink & Knapp, 2001; The National Autistic Society, 2015).

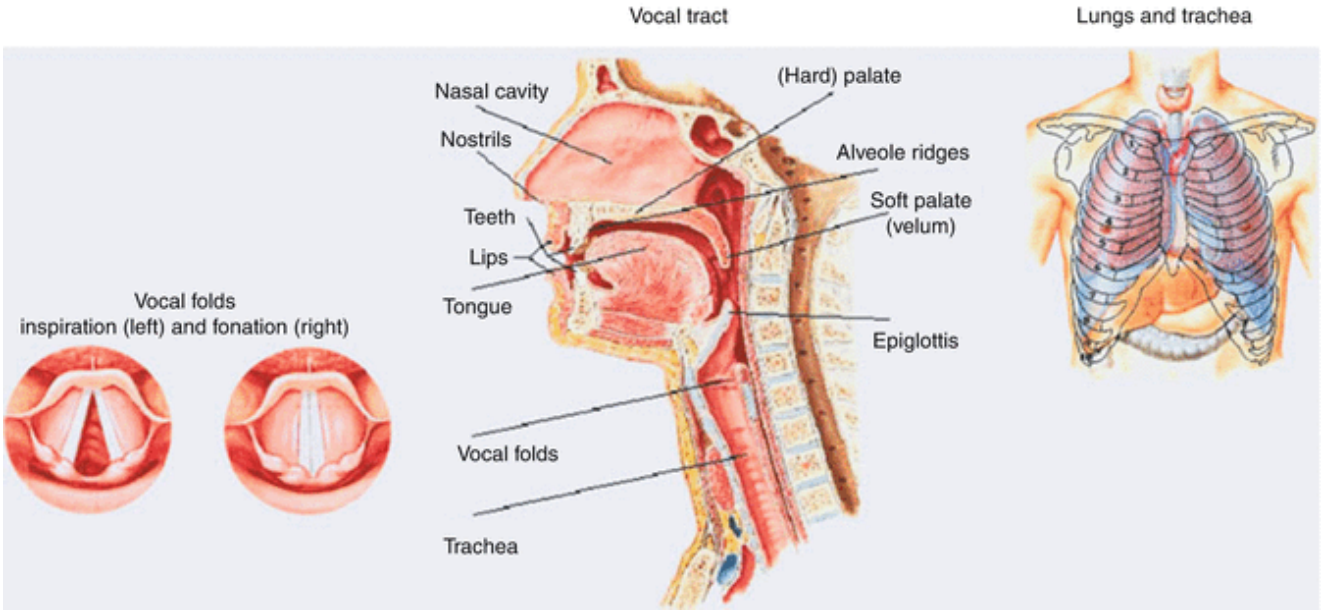
Early identification allows placement of early intervention to improve outcomes. Early indicators such as delayed verbal and non-verbal communication and motor delay contribute to screening of ASD in toddlers (Zwaigenbaum et al., 2009). It is vital we

improve our understanding of other early indicators such as speech, language, and motor impairments and how they interact so we can produce screening tools that capture these characteristics accurately. By examining speech using ultrasound it allows us to understand if this tool is more effective than perceptual speech assessments in identifying different in children with ASD. This is a tool that could be used by speech and language therapists in the diagnostic process for identifying speech differences not identified perceptually.

2.3 Nature of Speech Sounds Errors (SSEs)

In order to understand the nature of SSEs in ASD, it is important to have an understanding of what is expected in typical speech development and production, what SSEs are and why they occur. Speech production and perception break down at multiple levels can reduce the effectiveness of the final goal of fluent speech (Ferrand, 2014). The cognitive and neural processing required of speech perception and production is still not fully understood and speech production itself has been identified as one of the most complex motor skills as it requires functioning of multiple subsystems that must effectively coordinate and communicate together (Baghai-Ravary & Beet, 2013; Duffy, 2000). If we take the phonatory system as an example, in order to produce effective voicing, the vocal folds, the larynx and other muscles need to work in coordination as well as coordinate with other speech subsystems such as the respiratory system (Figure 1). In addition, before reaching the stage of speech production, there are multiple complex cognitive and perceptual processes along the auditory pathway that are required for effective speech perception. The speech perception system relies on effective auditory perception in order to transform acoustic signals into meaningful representation of spoken language in order to have accurate speech production (Gandour & Krishnan, 2016). In addition, motor speech representations are also vital for effective speech perception and production (Ravizza, 2005).

Figure 1: *Anatomy involved in speech production including vocal folds, vocal tract, and respiratory system.*



I have chosen to use the term “Speech Sound Errors” as it can be used to describe any impairment or difference in the production of speech sounds or speech segments without necessarily fitting strict definitions of “speech disorder” (American Speech-Language-Hearing Association, 2017). SSEs can come in the form of speech sound substitutions, omissions, distortions or persistent or residual SSEs. The type and nature of SSEs can vary widely, for example rhotic or sibilant distortions which can have little impact on the intelligibility or fluency of speech and are usually not associated with any language or intelligibility difference (Shriberg et al., 2011). Whereas other SSEs can cause children to become unintelligible, including to close members of their family. SSEs in children can include articulation errors which are often a result of a speech motor impairment and/or phonological errors, where there is a breakdown in the understanding and use of speech sounds (Eadie et al., 2015). Articulatory SSEs can be distinguished as they often come in form of distortions of a speech sound whereas a phonological SSE are often in the form of a deletion or substitution within the target word, these are called “speech processes”, some examples of these speech processes are; stopping of fricatives, velar fronting, consonant cluster reduction and final consonant deletion as described in table 2. Both articulation and phonological errors can occur in an individual child’s

speech profile and are not necessarily mutually exclusive, similar to the underlying causes of SSEs.

Table 2: *Speech Terms and Processes*

Speech Term	**Definition**
Speech Sound Error (SSE)	Any non-adult-like production of sounds in words
Speech Delay	Speech is characterised by errors normally found in younger children
Speech Disorder	A pattern of SSEs in which errors are not typical for any age group with normal speech development
Phonetics	The production of speech sounds by humans, often without prior knowledge of the language being spoken.
Phonological	The classification of the sounds within the system of a particular language or languages
Articulatory Error	A motor-based impairments in which the SSEs are a result of the difficulty in having the speech articulators' function to produce the target sound in multiple speech contexts
Linguistic Error	Rule-based errors, occurring when the child has difficulty organising the patterns of sounds at a cognitive level and are not a result of difficulty with the speech motor system.
Substitution	Substitution or systemic processes describe when there are changes to sounds within the word.
Omission	An SSE where certain sounds are omitted or deleted from a word.
Distortion	A distortion error is one that a child makes when they so not correctly produce a sound (e.g., frontal lisp)
Sibilant	Sibilant is a consonant sound, in which the tip, or blade, of the tongue is brought near the roof of the mouth and air is pushed past the tongue to make a hissing sound e.g., s or sh
Rhotic	Refers broadly to the sounds of the "r" family.
Persistent SSE	Speech sound distortions that have been present from an early age and have not resolved. Interestingly it has been residual SSEs that have been found to be common in children with ASD

Residual SSE	Leftover SSEs of an early speech delay of unknown origin that persist beyond the age of typical speech development, often defined as from age 9 onwards.
Labiodental	A sound made with the lips and teeth, e.g., /f/ and /v/
Lateral Sound	A sound produced by raising the tip of the tongue against the roof of the mouth so that the airstream flows past one or both sides of the tongue, e.g., /l/

The majority of SSEs present in young children's speech resolve with age, problems start to occur when these SSEs do not resolve and remain as residual or persistent SSEs. Residual SSEs are defined as leftovers of an early speech delay of unknown origin that persist beyond the age of typical speech development, often defined as from age 9 onwards (Preston & Koenig, 2011). Persistent SSEs are speech sound distortions that have been present from an early age and have not resolved. Interestingly it has been residual SSEs that have been found to be common in children with ASD. Shriberg et al. (2001) found residual SSEs in 33% of a sample of adolescents and adults with ASD compared to a TD sample in which only 1-2% presented with residual SSEs. Residual SSEs often come in the form of differences in late acquired and/or motorically complex speech sounds such as /s/ or /r/, for example /r/ can be labiodentalised or lateralised (table 2). Our understanding of why this happens more in individuals with ASD is unclear, Shriberg et al. (2011) suggests it may be a result of individuals with ASD not fine tuning their speech to the ambient speech model of their environment. Exploring cognitive theories of ASD may help our understanding on why these differences in SSEs occur.

2.4 Neural Mechanisms and Psycholinguistic Pathways of Speech

The cerebellum has been shown to play a significant role in efficient speech motor control (Ackermann, 2008). The cerebellum has shown to sequence syllables into fast and smooth large utterances as well as controlling the temporal organisation of internal speech. Difference in cerebellar functioning may account for why often subtle speech differences and higher rates of SSEs are observed in ASD, a condition known for showing altered cerebellar structures (Ackermann et al., 2007;

Belmonte et al., 2004; Gowen & Miall, 2007; Jaber, 2017). Furthermore, the network of cortical and subcortical structures controls the cooperation of around one hundred muscles used during efficient and fluent speech (Berg & Levelt, 1990). Reduced performance in maximum performance tasks such as DDK have been used to identify speech impairment in individuals with altered cerebellar function effectively (Ackermann, 2008). Ackermann et al., (2008) proposed that cerebellar disorders may give rise to speech motor disorders such as ataxic dysarthria, speech differences would include explosive syllable stress, loudness and pitch outbursts, abnormal prolongations of phonemes & intervals between sounds & words. However, it would spare perceptual and cognitive aspects of verbal communication. Regular practice of vocalisations and maturation of cortical and subcortical structures contribute to the formation of pathways required for fine level of speech motor control of the articulators (Williams, 2015). Children with ASD may be disadvantaged at this stage of early speech motor development due to potential reduced social motivation, limiting vital communicative interactions required for effective maturation of the speech motor control systems (Chevallier et al., 2012).

Understanding of the neural pathways of speech motor control and its relation to motor speech disorders such as apraxia of speech (AOS) have heavily focused on the neuromotor pathways as just described (Ziegler, 2002a). However, there are learnings to be taken from psycholinguistic pathways of speech motor control. Ziegler (2002a) carried out a review on these psycholinguistic and motor theories, specifically focusing on apraxia of speech but which can also be applied to our wider understanding of speech motor control. He discussed two important paradigms of motor programming: reaction timing and generalised motor programs. Motor programming based on reaction times stipulates that reaction time differences can occur due to the delay between the imperative signal and the motor response to that signal (Klapp & Erwin, 1976). DDK tasks can give indication of whether breakdown occurs here by looking at measures such as rate. The second theory proposed by Schmidt (2000) is that motor learning results in generalised motor programs (GMPs). GMPs are a combination of motor movements rather than a single individual movement. The execution of efficient speech motor control requires effective planning and production of GMPs that are specific to the context (e.g., speech). Our understanding of these models is that they differ in their understanding of the nature

of motor goals that guide speech movements and do not take into huge consideration the implied effect of auditory goals and perception (Ziegler, 2002a).

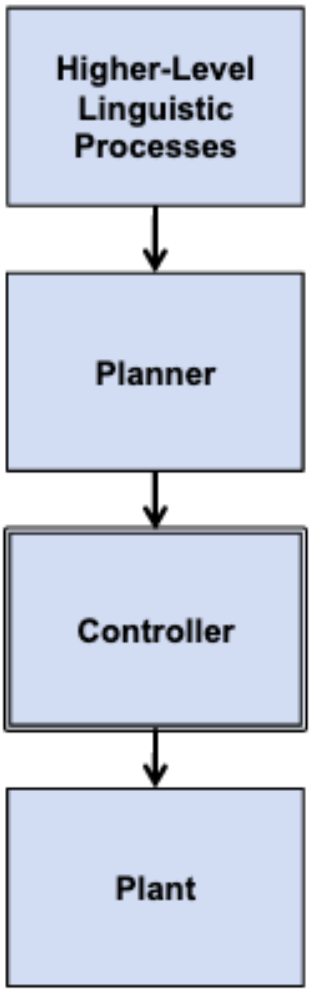
Guenther (1999) proposed the neural network model for speech motor control which is based around auditory goals. This model stipulates those auditory targets are mapped onto sensory motor representations and these are then mapped onto articulatory representations. This complex chain may explain the “speech attunement” framework, in which a child with ASD does not “tune in” or “tune up” to their ambient environment. If the child does not map these sensory motor representations to the correct speech motor execution, then this could result in altered articulatory representations and have higher rates of SSEs. According to this theory, speech motor control is still controlled on a more abstract level and relies of effective coordination between each element of the pathway and the initial speech perception being intact. The speech motor control system itself is left to its own intrinsic mechanism that adapt to the speech context.

2.4.1 Models of Speech Motor Control

Speech motor control is a combination of complex sensorimotor tasks and efficient and fluent production of speech requires fine speech motor timing and coordination of the articulators, which themselves have complex mechanical properties (Parrell et al., 2019). Speech motor control is also influenced and controlled by higher level cognitive processes that control not only motor planning but also the organization of the semantic and phonological aspects of speech sound production. It requires the integration of auditory, somatosensory and motor information which is represented in the temporal, frontal and parietal cortex as well as connected sub-cortical structures (Ghosh et al., 2008).

Figure 2 from Parrell et al. (2019) is used to help show the processes involved in effective speech motor control.

Figure 2: *Model of Processes Involved in Speech Motor Control (Parrell et al., 2019)*



Higher-level linguistic processes: These processes determine the motor planning and mediate semantic, syntactic, prosodic, and phonological organization. Language networks involves the inferior frontal cortex and the temporal cortex evidenced to support syntactic processes whereas less lateralized temporo-frontal networks serve semantic processes (Friederici, 2011). Areas of the brain that are in the language relevant cortex include parts of the middle temporal gyrus, Wernicke's areas in the superior temporal gyrus and Broca's area in the inferior frontal gryus (Kearney & Guenther, 2019). Declarative memory is rooted in the temporal lobe and the procedural memory in the frontal cortex and basal ganglia (Ullman, 2001).

Planner and Controller: The planner takes in a speech plan and potentially feedback from the plant and issues motor commands to the plant. They ensure that all formal models of sensorimotor control for speech have defined architectures that govern its functionality. The controller is integrated with feedback loops through subcortical

structures; a loop that through the pons, cerebellum and thalamus and a loop through the putamen pallidum and thalamus (Kearney & Guenther, 2019).

Plant: Speech synthesizers, the diverse articulatory structures of the tongue, lips, jaw, velum, and larynx. In the motor cortex there are distinct areas that represent each of these speech articulators (Jürgens & Ploog, 1970).

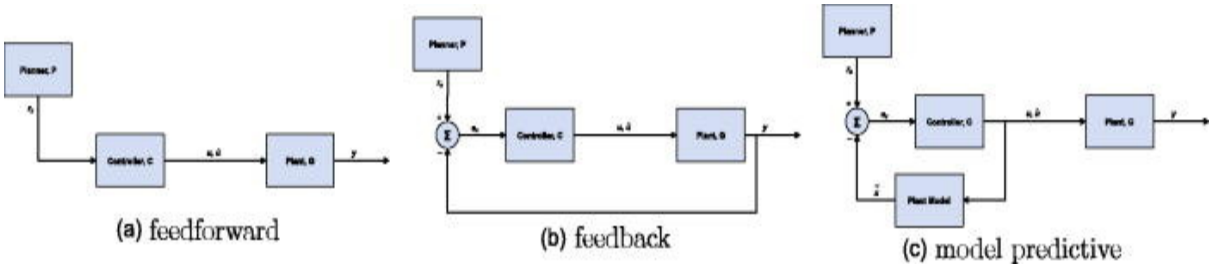
Parrel et al. (2019) summarized and compared the current models present in the literature that aim to define and understand speech motor control. This includes DIVA (Tourville & Guenther, 2011) Task Dynamics (Saltzman & Munhall, 1989), State Feedback Control, JASA/Speech Motor Control Models (Houde & Nagarajan, 2011), ACT (Kröger et al., 2009), GEPPETO (Perrier et al., 2005) and FACTS (Ramanarayanan et al., 2016). These models have been reviewed and discussed in the literature and there has been a surprising amount of variety (Bohland et al., 2010; Byrd et al., 2009; Civier et al., 2013; Saltzman et al., 2008). Whilst it is not within the scope of this study to discuss these different models in detail, one interesting discussion point as found by Parrell et al. (2019) is that one of the main differences between these models is how speech motor control is influenced by feedback signals originating from the plant, i.e., the articulators and speech synthesizers.

Feedback systems require careful coordination in order to have efficient speech motor control. If there is incorrect attunement in these feedback systems, this can result in altered movement patterns at the speech motor control level. The speech attunement framework focuses on the tuning in and tuning up the speech in the perceptual domain, but we have not yet investigated how internal feedback from the articulators in ASD is functioning. If feedback signals are impacted this may lead to inaccurate movements of the articulators or altered movement patterns as a result of delays or deficiencies in the neural processing of sensory feedback (Iarocci and McDonald, 2006; Ramanarayanan *et al.*, 2016).

What DDK tasks tells us is how these articulators are functioning and give indicators of where breakdown is occurring at the “planner” and “controller” levels. It cannot however determine accurately where this breakdown is occurring. This requires a multi-faceted assessment approach in which speech is examined at all four of these levels. This study has tried to cover these four steps but focused mainly on the

controller level. The behavioural assessments discussed in chapter five and six allowed us to observe how higher linguistic processes may be impaired in this model in groups of children with ASD.

Figure 3: *Three architectures of feedback that can be used during the speech motor control process, this includes feedforward, feedback, and model predictive.*



2.4.1.1 Feedforward control

Feedforward control is a way to cut out any delayed or noisy sensory information. In this sequence, the signals issued from the planner cuts out the use of feedback entirely, the motor commands depend only on the reference signal. In terms of speech, the feedforward control mechanism generates predictive motor commands based on past experiences with the speech target (Edwin et al., 2015). A cortico-basal ganglia loop is responsible for the launch of a motor program at the right time using an initiation map, which is a supplementary motor area found in the media wall of the frontal cortex (Kearney & Guenther, 2019).

2.4.1.2 Feedback control

In the feedback control system, this architecture uses the outputs of the plant for control, using feedback signals with sensory information and comparing them to current stored motor plans. If these signals are slow to propagate or process when received, this can result in slower performance. Additionally, if these signals are corrupted then this can cause inaccurate movements (Parrell et al., 2019). Auditory feedback control involves axonal projections from an auditory target map from higher-order auditory cortical areas in the posterior auditory cortex (Kearney & Guenther, 2019). Furthermore, if an error is made, the auditory error map in the posterior auditory cortex is initiated which projects the feedback control map in the right ventral premotor cortex.

This works in conjunction with the somatosensory feedback control subsystem. Kearney and Guenther (2019) hypothesised the main components are located in the central postcentral gryus and adjoining supramarginal gryus. This feedback arrives from the cranial nerve nuclei in the brainstem via the ventral posterior medial nucleus of the thalamus. Errors are detected by somatosensory error maps which are activated in event of a mismatch between somatosensory states and auditory subsystem maps. This activation becomes a corrective motor command via feedback control in the right ventral premotor cortex (Kearney & Guenther, 2019).

2.4.1.3 Model predictive control

An alternative architecture to the feedforward and feedback control is the model predictive control. Similar to the feedforward control, it does not use the sensory outputs produced by the plant to maintain control. It does however take motor commands from input and uses them to make predictions about the speech motor control system's state and can then predict the effects of the motor command on the plant. This reduced the need for direct feedback from the plant, replacing it was a prediction from the controller (Miall & Wolpert, 1996). This produced an error signal that can be provided to the controller.

2.4.2 Motor Speech Disorders

Speech sound errors can be a result of underlying motor, neurological and/or structural causes. They can also be idiopathic and have no known cause (Shriberg et al., 2019). Motor based SSEs tend to take shape in production of individual speech sounds, for example, substitutions or distortions. Whereas, phonological based SSEs are more often rule-based and predictable, for example, stopping, fronting and final consonant deletion (Peña-Brooks & Hedge, 2015). It is important to distinguish between the different causes of SSEs, for example, those caused as a result of a motor speech disorder or a phonological difficulty. Children with motor speech disorders tend to present with additional speech differences such as imprecise and/or inconsistent spatiotemporal distortions of sounds, prosody and voice differences (Shriberg, et al., 2019). These can also occur simultaneously with phonological errors, for example, there is evidence in the literature that children who present with idiopathic speech delay have motor speech differences that impact speech development (Shriberg et al., 2019). However, estimates of idiopathic motor

speech disorders are not available, impeded by lack of widely used measures for diagnosis and classification of motor speech disorders in children. (Shriberg et al., 2019). A key difficulty in distinguishing whether SSEs are phonological, or motor based is the overlap of the mechanisms involved in phonological processing and motor planning (Adam et al., 2017; Galluzzi et al., 2015). Diagnosis such as dysarthria and apraxia of speech are speech sound disorders that are distinguished by the disruption to the motor component of speech and have set criteria as described below:

2.4.2.1 Childhood Dysarthria

Childhood dysarthria is regarded as the most common context for childhood speech motor disorders (Shriberg, Campbell, et al., 2019). It occurs in children who have an acquired disorder such as traumatic brain injury or a congenital neurodevelopmental disorder such as cerebral palsy. Dysarthria is characterised by weakness, abnormal muscle tone and impaired articulation (Duffy, 2000). The type depends on the lesion site and by perceptual speech differences. Damage to the cerebellum can result in dysarthric speech and can affect both the feedforward and feedback control systems (Parrell et al., 2017). The cerebellum plays an essential role in speech motor timing, accuracy and coarticulating feedforward commands to the plant (Kearney & Guenther, 2019). Furthermore, the cerebellum is hypothesised to generate precisely timed auditory and somatosensory targets in feedback control of speech sounds and in the production of corrective commands.

2.4.2.2 Childhood Apraxia of Speech

Childhood apraxia of speech (CAS) is a speech disorder in the which the accuracy and consistency of speech movements are impaired in the absence of a neuromuscular disorder (American Speech-Language-Hearing Association (ASHA), 2007). It is often studied as an idiopathic disorders, with more recent research focusing on a genomic aspect of CAS (American Speech-Language-Hearing Association (ASHA), 2007). It is often associated with damage or different to the ventral precentral gyrus, anterior insula and/or inferior frontal gyrus. Damage to these areas impact the speech sound map, a core component of motor programs for frequently produced sound sequences. Damage in these areas will strongly impact feedforward commands.

Review of the literature has indicated that there is no set list of biomarkers and diagnostic symptoms of CAS (ASHA, 2007). However, there are three diagnostic indicators that are commonly used in the literature which include.

1) Inconsistent speech errors in repeated production of syllables and words
2) Lengthened and disrupted coarticulatory transitions between sounds and syllables
3) Prosody differences, particularly in stress

It is stressed that these differences alone are not sufficient for a diagnosis of CAS and that the overall behavioural features should eb taken into account, including speech, expressive language and literacy.

2.4.2.3 Speech Motor Delay

Speech motor delay is the presence of a “motor component” in the speech of children with idiopathic speech delay. Children who have presented with speech motor delay tend to also present with a general motor delay (Duchow et al., 2019). Subgroups of children with motor differences have presented with idiopathic speech delay, performing lower than age-sex norms on gross, fine and/or oral motor tasks (Archibald & Alloway, 2008; Nip et al., 2011; Richtsmeier & Goffman, 2015).

The main characteristics of speech motor delay is imprecise, or unstable speech, prosody and voice that does not meet the criteria for childhood apraxia of speech or childhood dysarthria (Shriberg, Campbell, et al., 2019). Shriberg et al. (2019) carried out an in-depth study to identify early signs of speech motor delay, finding it was characterised by an “across-the-board’ delay in speech development. The following speech signs were identified;

1) Increased duration for mid-vowels and diphthongs
2) Reduced speech consistency and accuracy in both vowels and consonants
3) Increased average syllable duration

2.4.3 Multisyllabic Speech Contexts

Complex articulatory gestures consist of multisyllabic or polysyllabic sequences e.g. complex consonant productions (Adams, 1998b). Increasing the complexity of an

articulatory gesture may increase the processing demands on motor performance of children (Maner et al., 2000). The more complex the articulatory gesture, the further heightened the requirement there is to select and sequence phonemes under high motor demands (Lewis & Freebairn, 1997). As children with autism present with higher rates of motor impairment they may have increased variability and error in production of complex articulatory gestures due to increased motor demand (Maner et al., 2000; Sadagopan & Smith, 2008). It is important to observe if there is variability in results as this will tell us.

This study used multisyllabic speech, the increasing structural complexity of sounds both in real words and non-linguistic sounds to observe if the increased motoric complexity had an impact on speech production, particularly accuracy. Multisyllabic speech was used to observe if increasing the length of the work and/or sound to test the increasing complexity of motor planning and production, helping to identify subtle speech differences not found in single word contexts. However, multisyllabic speech differences are not only indicative of speech motor difficulties, low accuracy of multisyllabic speech has also been linked to differences in phonological processing abilities (Masso et al., 2017) and poorer later literacy development (Larrivee & Catts, 1999). A study carried out by Masso et al. (2017) observed frequent deletion, a phonological SSE, in a multisyllabic context. This suggests that while using multisyllabic speech as a strategy to identify speech motor differences, it should also include wider assessment of phonological speech, expressive and receptive language abilities, all of which may also impact multisyllabic speech production.

2.4.4 Oral Motor Functioning

Though this study did not have the capacity to carry out further speech evaluation due to time constraints, future research into speech motor differences could include assessment of oral motor skills. It is possible assess oral-motor skills using clinical assessments such as the Robbins and Klee (1987) clinical assessment of oral motor development in children. It could indicate further differences between children with ASD and the TD group. Oral motor skills are strongly associated with speech fluency (Alcock, 2006). This has been demonstrated in children with autism (Amato & Slavin, 1998) and their scores distinguished them from their typically developing peers (Adams, 1998). Assessing oral motor skills will help determine if any speech sound

errors found can be directly attributed to structure or function of specific vocal tract structures (Kasari et al., 2013). Alcock (2006) also found that motor control was associated with an existing language impairment, particularly oral motor control. Lewis et al. (2011) found children with SSD were slower to complete DDK tasks and differences in their oral motor control compared to TD children.

2.5 Theories of Speech Impairment

Multiple studies have reported that roughly one third of children with ASD have a presentation of SSEs, abnormalities in speech production and potential speech motor impairment (Belmonte et al., 2013; Cleland, Gibbon, et al., 2010; Shriberg et al., 2011). This section will discuss two perspectives on why this occurs, concluding that these two causative explanations are potentially complementary.

2.5.1 Speech Motor Control

The first perspective to take into account for increased prevalence of SSEs in ASD is that there may be a subtle but significant speech motor control impairment present in the speech production of individuals with ASD (Adams, 1998a; Barbeau et al., 2015; Belmonte et al., 2013). As evidence of the general motor disruptions in ASD mounts, this explanation of SSEs in ASD has become more attractive. There is clear evidence of motor disruptions in purposeful movement of the arms (Crippa et al., 2015; Torres et al., 2013), in the legs and gait in individuals with ASD (Nayate et al., 2012; Rinehart et al., 2006). Fine motor control in children with ASD is also known to be disrupted, particularly in writing and object manipulation (Fuentes et al., 2009). Fournier at al. (2010) carried out a meta-analysis that suggests that a disruption in motor abilities may be a core feature of ASD and not necessarily an associated or co-morbid impairment.

At the gross and fine motor level, impairment is evident in ASD. There may be a fundamental impairment at the level of motor timing and sensory motor integration that allows the production of skilled and accurate movements, including in speech production (Beversdorf et al., 1998; Gowen & Hamilton, 2013; MacNeil & Mostofsky, 2012; Mostofsky et al., 2009; Whyatt & Craig, 2013). A disruption in timing and sensory motor integration may negatively impact the child's development, contributing to autistic symptomatology resulting in a disruption in expressive

intention and purposeful engagement with others, isolating and distressing the child (Trevarthen & Delafield-Butt, 2013). Speaking and articulating fluently requires intricate control and coordination of speech motor mechanisms (Gracco, 1994). Yet the relationship between speech and fine motor impairment is still relatively unexplored. This perspective proposes that the increased rates of SSEs identified in children with ASD may be due to underlying motor impairment. As seen with the residual SSEs reported by Shriberg et al. (2001), late acquired and motorically complex speech sounds are impaired, these sounds require intricate speech motor skills and may be evidence of a speech motor impairment in some speakers with ASD.

The study of motor differences in ASD is a growing field. Abnormalities in the cerebellum (Fatemi et al., 2012), impaired sensory input and multisensory integration (Gowen & Hamilton, 2013) and disruption in brain synchronization (Welsh et al., 2005) are all neuroanatomical correlates that have been suggested to cause the observed motor differences in ASD. It is possible that if gross motor and fine motor abilities are impaired in ASD, then a speech motor control impairment could be present also (Barbeau et al., 2015). Adams (1998) examined the speech motor control and oral motor control of four children with ASD compared to TD peers in simple and multisyllabic speech contexts. Oral motor abilities were assessed by asking the child to execute nonspeech motor movements on command or upon an imitation. A total of eleven movements were administered. Their results indicated that children with ASD have significantly more difficulty producing oral movement and multisyllabic speech compared to the TD group. The difficulties in producing complex articulatory gestures such as multisyllables show us that speech motor impairment may be present in some children with ASD. However, it is not possible to generalize these results due to the small sample size. Future research needs to look at both linguistic and motor planning skills in children with ASD. This will help to describe accurately the range of mental processes that may be impacting their speech production.

Whether speech motor control and general motor abilities are directly connected is still up for debate. For instance, the task independence hypothesis stipulates that motor control of the organs used for speech is independent of the motor task that is imposed on them. There is a “general” oral sensory-motor system which controls

activities for the muscles involved in speech and also relies on sensory feedback (Clark et al., 2001). Whereas Zieger (2003a) proposes that the motor skill of speech is linked to the auditory domain, a specific sensory modality. Whereas general motor skills such as grasping, and pointing are based on visual spatial and/or proprioceptive representations which is not required for speech encoding. The correlation between speech motor control and general motor control whilst not studied in the ASD population, has been examined within the TD population.

Nip, Green and Marx (2011) found that TD infants showed a correlation between changes in articulatory movements and development of early communication. Using motion capture with twenty-four children of the lips and jaw every three months, they found that children who had reduced speech motor control also presented with delayed communication development. They found significant correlations between standardized measures of language and cognitive skills and orofacial kinematics, even when accounting for age. This suggests there is a link between early communication development and oral motor control but does not clearly evidence a link between speech motor control and general motor control. There was a similar pattern noted in children with SSDs except their trajectory was delayed. Alcock (2006) also found that motor control was associated with an existing language impairment, particularly oral motor control. Lewis et al. (2011) found children with SSD were slower to complete DDK tasks and differences in their oral motor control compared to TD children. Peter and Stoel-Gammon (2008) also found children with SSDs had differences in paced repetitive finger tapping and clapping exercises. Children with SSDs have also been found to have reduced performance on grasping, object manipulation and visual motor skills (Newmeyer et al., 2007). Speech and motor differences may have a common fine motor difference; however, it is not clear how this would present in children with ASD.

There are few studies exploring the relationship between speech motor control and general motor control in populations with ASD. However, there are observations to be made from children with idiopathic speech disorder that may inform future research in ASD. For example, Bradford and Dodd (1994) compared the speech and motor abilities of ten phonologically delayed children, ten children with consistent phonological disorder and ten children with inconsistent error patterns. The groups did not perform differently on simple motor tasks, however the group of children with

inconsistent error patterns performed were significantly below other groups in their performance on timed motor planning tasks as well as expressive novel-word learning tasks.

2.5.2 Speech Attunement

An alternative perspective is the “speech attunement framework” developed by Shriberg et al. (2011) to explain the presence of SSEs in typically developing children (for instance, the presence of dentalised sibilants). Shriberg et al. (2011) recognised that there were impairments in oral motor, fine motor, and gross motor control in individuals with ASD that were potentially associated with speech production difficulties and set about to explore these. This framework posits that when a child is learning speech, they need to attend to their ambient speech environment, also known as “tuning in”. For instance, when a child takes on dialect characteristics of their peers, they do it by tuning in as discussed earlier in Baron-Cohen’s work on accent differences between children with ASD and their peers (Baron-Cohen & Staunton, 1994). They also need to “tune up” which involves making careful and small adjustments to their speech sound production in order to sound like the speech in their ambient environment (Shriberg et al., 2011). It is also vital at this stage in speech development that the child’s speech motor system is maturing effectively in order to make these small adjustments to their speech with adequate control. If there is a speech motor control impairment present, this could impact speech attunement and cause a heterogenic speech profile in the child. Individuals with ASD may have reduced social motivation that would encourage attention to the subtle details of articulation in the ambient environment. This would impact how they make minute adjustments to their speech sound production to produce speech similar to their social partners (Shriberg et al., 2011). In fact, children with ASD are not thought to have the appropriate psychological conditions that are required to engage socially with social partners in order to have the necessary experiences for speech development.

The ability to “tune in” and “tune up” as posited by the speech attunement framework may be impacted in individuals with ASD in the following ways: Enhanced auditory capacity, often observed in individuals with ASD (Baum et al., 2015) may lead to earlier “tuning in” when motor maturity has not been achieved. Therefore, SSEs

develop due to motor constraints. The second is constraints in affective social reciprocity, a common trait of people with ASD (Chevallier et al., 2012) may delay "tuning in" and any motor speech disorder present may impair the ability to tune up.

Shriberg et al. (2011) studied whether these SSEs present in children with ASD and if there was a comorbid speech motor control impairment in the form of childhood apraxia of speech (CAS). CAS is an impairment in the consistency and accuracy of speech movements. To understand if SSEs seen in children with ASD and whether they were related to the motor speech difficulties evident in children with CAS, Shriberg et al. (2011) studied the continuous speech of children with ASD (n=40) compared to TD control group (n=40), children with speech delay (n=13) and individuals with CAS (n=15) using the previously discussed PEPPER system (Programs To Examine Phonetic and Phonological Evaluation Records; Shriberg et al., 2011).They used the PEPPER software to acoustically analyse the speech samples using transcription and prosody coding.

The analysis carried out was specifically designed to identify symptoms of CAS or other types of speech motor differences, e.g., distorted consonants, vowel errors, slow speaking rate (American Speech-Language-Hearing Association (ASHA), 2007; Aziz et al., 2010). The findings did not support a diagnosis of CAS or any speech motor difference. However, they did find that the group of children with ASD had voice differences not found in the CAS group such as abnormally high pitch and inappropriate loudness. The children with ASD presented with appropriate stress and speaking rate which is direct contrast with the CAS group and the symptoms of CAS. Shriberg et al. (2011) concluded that these results indicated speech attunement issues rather than a speech motor impairment. Though 75% of the group of children with ASD presented with increased repetitions and revisions, a well-known symptom of CAS, indicating there may be overlap in symptoms of these conditions.
The following indicators of speech attunement issues used today are as a result of this study (Shriberg et al., 2011):

- Increased repetitions and revisions, consistent with the description of autistic speech as "disfluent."

- Misplaced stress, often described as “off” or “singsong (Peppe et al., 2007). This stress is dissimilar to the well-documented “excessive-equal” stress pattern in apraxia of speech.
- Inappropriate loudness and pitch.
- Higher rates of speech delay and speech errors relative to population estimates.

Importantly in this same study, there were double dissociations in speech, prosody, and voice differences in ASD. They did not have significantly slowed speech rate, lengthened vowels or uncommon phoneme distortions that define motor speech impairment (Duffy, 2000). They also had inappropriate loudness and pitch (suprasegmental issues) that are not typically associated with motor speech disorder. These findings are interpreted as consistent with the speech attunement framework, rather than motor speech impairment. Schoen, Paul and Chaurska (2011) had similar findings when examining phonology and vocal behaviour of thirty children with ASD compared to eleven age-matched and twenty-three language matched controls during parent-child and clinician-child interactions. After analysing and coding speech-like and non-speech like vocalisations it was found that toddlers with ASD produced significantly more atypical non-speech vocalizations compared to control groups. Findings imply children with ASD may not tune into their ambient environment, negatively impacting acquisition of language and increase in production of non-speech vocalisations.

Speech attunement issues may have been present but unrecognised in previous studies of children with ASD. For example, Baron-Cohen and Staunton (1994) found in a group of children with ASD that when mothers had non-native accented English, the child with ASD was likely to develop the same accent as their mother (83.3% of the sample) whereas TD peers were more likely to develop accents of the ambient environment of their peers.

Children without social communication impairment as seen in ASD, tend to identify with their peers rather than parents in terms of accent development. Whereas a lack of social motivation seen in ASD may result in a lack of opportunities for speech

attunement and therefore not developing the same accent as their peers. As a result, we would expect to see higher rates of SSEs in children with ASD, particularly phonetic distortions. This was the case in research carried out by Shriberg et al. (2001), the adolescent and adult speakers with ASD produced significantly more articulations distortions like rhotic and sibilant distortions. As these speakers were adolescents and adults, we would define these SSEs as “residual SSEs” as they have not resolved when reaching full speech maturity. This if similar to findings by Cleland et al. (2010) who found older children with similar residual SSEs in the ASD sample. These studies imply that SSEs that could be a result of reduced speech attunement in early speech development can continue into adolescence and adulthood. It shows that SSEs in samples of ASD may be at risk for social, emotional, and academic challenges compared to TD peers (Hitchcock et al., 2015). This would compound the social and emotional difficulties that children with ASD already experience and could require support and intervention.

2.5.3 Neuroimaging Studies

Impaired central auditory processing may contribute to the speech processing differences identified in children with ASD and in turn impact speech production. Vocal learning is dependent on the ability to perceive and categorize sounds at different times (Doupe & Kuhl, 1999). Key et al. (2016) found this was impaired in children with ASD using a passive listening paradigm that assessed speech sound differentiation by contrasting consonant-vowel. EEG (electroencephalography) was used to measure brain activity and found children with ASD had reduced differentiation in 84-308ms period. You et al., (2017) used magnetoencephalography (MEG) to compare fourteen children with ASDs’ responses to speech and non-speech stimuli compared to controls. Results showed that poor spoken language scores were associated with atypical event-related field (ERF) responses. These findings support the theory that children with ASD have an impaired auditory cortex which implicates their ability to process speech. This poor speech processing would reduce their ability to learn speech of their ambient environment. Studies using MEG and EEG in children have produced mixed results. ERG studies have found atypical responses to non-speech stimuli (Khan & Sepulveda, 2010) and speech sounds

(Roberts et al., 2008). Yet when measuring a different response, mismatch negativity (MMN) using EEG), children with ASD were found to have typical responses to speech sounds (Kemner et al., 1995).

The inconsistent responses could reflect the heterogeneity of ASD. Additionally, it may indicate that a subset of children experience poor auditory processing, resulting in impaired speech and language abilities (Rapin & Dunn, 2003; Tager-Flusberg, 2004). The mismatch in findings highlights the important of observing both instrumental and behavioural measures of speech. This ensures a clear and accurate picture of children's abilities is gained. Kuhl et al., (2005) recognised the need to combine behavioural and electrophysiological measures in studying speech perception. They examined social and linguistic processing of speech using MMN and ERP measures and compared with linguistic measure of phonetic discrimination. They found atypical results for both electrophysiological and behavioural measures. What these studies tell is that children with ASD who lack social interest in communication may be at a disadvantage in terms of speech perception and as a result speech production development. Early measures of speech perception and production may be a useful early diagnostic tool of young children with ASD. Their differences in 'tuning in' may be impacting both speech production and expressive language.

2.5.4 Speech Timing

Effective speech timing is fundamental to accurate and fluent speech production. Speech produced with fluency requires a set of quasi-autonomous articulatory systems to coordinate information in an accurate and time sensitive way (Kotz & Schwartze, 2016; Maassen & Van Lieshout, 2010). There has been little research carried out on speech timing in individuals with ASD but the few studies that have looked at general sensorimotor timing have found abnormalities in children with ASD. For example, Anzulewics, Sobota and Delafield-Butt (2016) found that children with ASD playing with an iPad game had an increase in their speed in fasts taps and swipes. Furthermore, Torres, et al. (2013) found in a reach-to-touch task with children with ASD that they increased acceleration-deceleration phases. These studies evidence there may be a significant disruption in the motor control occurring in the region of 30-70 ms, which is a temporal domain vital for speech.

What may underpin motor disruptions typically observed in ASD is over- and under-compensations in rapid shifts of force (Trevarthen & Delafield-Butt, 2013; Whyatt & Craig, 2013). These compensations are likely to impact basic perception and experience in ASD, perhaps resulting in impaired speech development due to lack of coordinated articulatory and sensory-motor systems (Colwyn Trevarthen & Delafield-Butt, 2017). Disruptions in sub-second control of velocity and accelerations have been indicated in simple arm-swing tasks in individuals with ASD (Cook et al., 2013). They recorded trajectory, velocity, acceleration, and jerk while adult participants with autism and a matched control group conducted horizontal sinusoidal arm movements. Additionally, participants with autism took part in a biological motion perception task in which they classified observed movements as 'natural' or 'unnatural'. Cook et al. (2013) found that faster timing at the sub-second level, which is required of speech motor control, is disrupted in the limb and hand movements of individuals with ASD. These studies evidencing abnormality in movement timing in ASD indicate there could be a difference in motor planning and programming, which can be associated with SSEs. It is vital to further our understanding of timing in the speech-motor domain that this is examined in detail in ASD to help determine the natures and causes of SSEs in this group.

2.6 Methodological Issues of Measurement of SSEs

There are several methodological issues when assessing SSEs in children with ASD that need to be examined. To begin, the over-reliance of assessment of speech in a single word context may skew results to mask atypical SSEs (Broome et al., 2017). Single word speech analysis does not account for the complex articulatory gestures found in multisyllabic speech and conversational speech, which is significantly more motorically complex (Adams, 1998b). When Kjelgaard and Tager-Flusberg (2001) studied the speech production of eighty-nine children aged 4-14 years with ASD they argued that while there was significant disparity in the language abilities, their speech produced was on the normal trajectory of development. However, they had only used a perceptual speech assessment that analysed the SSEs using broad phonemic transcription using the Goldman Fristoe Test of Articulation (2000). It required Cleland et al. (2010) to carry out their own phonetic and phonological

analysis to identify speech processes beyond this assessment in order to discover speech distortions in the children with ASD. Whilst Kjelgaard and Tager-Flusberg's sample of children with ASD may not have met the threshold for a diagnosis of a speech delay or disorder, there is value in identifying subtle SSEs in ASD so we can understand how speech perception and production develops in this population. Additionally, the Goldman Fristoe Test of Articulation (2000) does not share similar standardisation norms as found in other speech production assessments which may have impacted the results. Children under the age of seven should be classified with a speech disorder if they present even with only one or two disordered errors, which is not the guidance offered in the Goldman Fristoe Test of Articulation (2000).

When measuring articulatory gestures in children with ASD, results have been conflicting. McCleery, Tully and Slevc (2006) documented consonant production in fourteen children with ASD and severe language delay at the canonical babbling stage and 14 language matched TD children. Results showed children with ASD had no significant difference in speech production to TD children. However, only simple articulatory gesture productions were observed in this study. Adams (1998) observed simple and complex articulatory gestures in children with ASD and age-matched controls. There was no significant difference in simple articulatory gestures. However, children with ASD had significantly more difficulty producing complex articulatory gestures than controls. The measure was modified from the Kaufman Speech Praxis Test (Kaufman,1995). It measures simple articulatory gestures in the form of monosyllabic words and complex articulatory gestures in form of multisyllabic words. Whilst this provided some evidence for difficulty in producing complex articulatory gestures in children with ASD, the sample size of four children with ASD cannot be generalised. I therefore stipulated the importance of measuring speech in multiple modalities, both in perceptual and instrumental methods to understand whether discrepancies in the presence and type of SSEs produced by children with ASD is a result of variable speech assessment methods or the presentation of SSEs by children with ASD.

However, perceptual single-word assessments have effectively identified SSEs in some children with ASD. Rapin et al. (2009) used the photo articulation test (Lippke et al., 1997), a single word speech assessment to assess speech production of sixty-

two children with ASD. As found in the studies carried out by Shriberg et al. (2011) and Cleland et al. (2010), Rapin et al. (2009) found that 28% of the sample had a significant speech production difference, this was despite having good receptive language abilities. There was no particularly in-depth analysis carried out either, the authors rated each child's speech on a 3-point scale (0= normal to 2= severe impairment). Why Rapin et al. (2009) identified SSEs and Kjelgaard and Tager-Flusberg (2001) did not may be because Rapin et al. (2009) they used two methods of speech assessment: spontaneous speech utterances and single word object naming. It is important that research in this field continues to examine speech in multiple contexts to reveal if there are subtle SSEs in the speech profiles of children with ASD.

Using speech assessments that are not effective at revealing speech production impairment and the combination of the nature of assessment with participants with ASD may impede effective assessment further (Macrae, 2017). If a child has a severe language impairment, this may cause significant difficulty in obtaining a clear speech sample as a result of their expressive language impairment, often seen in children with ASD where children often present as non-verbal. McCleery et al. (2006) challenged this assessment issue when investigating the speech of fourteen severely language delayed children with ASD. They carried out a communicative inventory that allowed the child to produce both voiced and voiceless consonant sounds, including babbling. All vocalisations, speech and non-speech were analysed in order to create a profile of the child's consonant production abilities. McCleery et al. (2006) were able to conclude that children with ASD were able to produce the same speech sound patterns as TD and language-learning impaired groups. However, they acknowledged that the children with ASD produced atypical speech sound but did not carry out further analysis of these. Further transcription and analysis of frequency and form of these SSEs may have indicated an alternative speech profile from the control groups. Analysis of "atypical" vocalisations can provide indicators of whether SSEs are of a motoric or phonological root (Paul et al., 2011; Schoen et al., 2011).

Studies that previously investigated speech production of children with ASD, as a result of using perceptual speech assessments only, may have missed subtle SSEs due to the imprecise nature of this type of assessment. Assessment described in the

previously discussed studies are heavily reliant on perceptual measures of speech, i.e., the auditory perception of the clinician/researcher who then carries out phonetic and phonemic analysis from these perceptual judgements. Auditory perception alone cannot reveal precise information about articulatory movements during speech. It is this precise information that may give us information about speech motor control abilities in ASD and whether alternative or impaired movement strategies are used due to an underlying speech motor impairment or speech attunement difficulties. Kent (1996) explained that auditory perceptual judgments of this nature are susceptible to bias and errors from the listener. For instance, a phenomenon called "listener normalisation" can occur, in which the listener can attribute recognised phonemes to the speaker that were not actually produced. Furthermore, even if SSEs are identified in perceptual assessments, transcription techniques often used are too broad in order to distinguish these errors (Kent, 1996). The IPA chart is the most used categorisations tool of speech in broad phonetic transcription and does not account for the variation within categories for individual's speech (Mowrey & MacKay, 1990). This type of speech assessment requires observing speech in various contexts, not just single words, in which articulatory gestures are complex and the analysis looks at both phonetics and phonology.

2.7 Assessment of Speech

Researchers have identified atypical speech development in children with ASD (Cleland et al., 2015; Shriberg et al., 2001; Wolk and Giesen 2000; Wolk and Edwards, 1993). However, it has been argued this is within a sequence of typical development (delayed) rather than atypical (McCleery et al., 2006). Unreliable outcomes in the literature may be a result of inconsistent and reduced specificity of the measurements used in across studies. This section will discuss the different methods of assessing speech in relation to how these could impact assessment of speech in ASD.

2.7.1 Perceptual Assessment of Speech

There is a critical need for researchers and clinicians to address the speech sound behaviour in children with ASD (Eigsti et al., 2011; Shriberg et al., 2011). Researchers have identified deviant speech development in samples of children with

ASD (Cleland, Gibbon, et al., 2010; Schoen et al., 2011; Shriberg et al., 2001; Wolk & Brennan, 2013). However other researchers argue that children with ASD have a sequence of normal speech development (Kjelgaard & Tager-Flusberg, 2001a; McCleery et al., 2006). Discrepancy in findings may be due to perceptual measurement of speech and lack of precise and accurate instrumental measurement (Broome et al., 2017).

Speech and language therapists (SLTs) rely on auditory perceptual judgement to classify and measure speech sounds disorders (SSDs) (Kent, 1996). Assessments described in earlier studies such as the PAT and DEAP is reliant on these perceptual measures of speech. However, auditory perceptual judgements are susceptible to errors and bias of the listener (Kent, 1996). For instance, listener normalization can occur, which is when the listener mistakenly recognises phonemes that were not produced by the speaker (Buckingham & Yule, 1987)

Additionally, subtle articulatory errors are difficult to identify perceptually, particularly if silent movements of the articulators occur. Even if errors are identified, suitable transcription techniques are lacking in the ability to distinguish these errors (Kent, 1996). Broad phonetic transcription is reliant on the categories of the International Phonetic Alphabet (International Phonetic Association, 2015) chart, even though variation within each category can vary significantly across individuals (Mowrey & MacKay, 1990). Another weakness of perceptual assessment is the influence of prosody on phonetic classification (Kent, 1996). Acoustic boundaries shift when speaking rate changes (Miller & Wayland, 1993). Phonetic decisions can be influenced by suprasegmental information such as pitch and rate. It is also well established the individuals with ASD often have difficulties with prosody.

2.8 Measurement of Speech Motor Control

Speech motor control can be measured through maximum performance tasks which assess performance of rapidly alternative movements (Fletcher, 1978). This study measured rate, accuracy, and consistency of maximum performance tasks in the form of speech diadochokinesis (DDK). Whilst speech abilities required for DDK tasks differ from normal speech production, they provide information on motor speech and potential impairment (Cleland, Gibbon, et al., 2010; Thoonen et al.,

1996). The rate of production, accuracy and consistency of targets produced are indicative of different subtypes of speech disorders. For instance, slower monosyllabic repetition rates have been found to be indicative of dysarthria (Duffy, 2000). Whereas a normal rate of monosyllabic production but reduced rate of trisyllabic production and significant inaccuracy of targets produced is indicative of apraxia (Thoonen et al., 1996). Both are different types of motor speech disorders, and these symptoms were investigated for within this project.

There is an indication that DDK performance in children with ASD may be implicated but there is very little research in this area. In an unpublished thesis Deshmukh, Mccauley, Wagner, & Rabidoux (2012) found children with “high functioning ASD” performed faster rates of oral DDK tasks but had significantly reduced accuracy and consistency that the TD group. These results may indicate presence of speech motor coordination and control difficulties in children with ASD. There is little research on oral DDK performance in children with ASD, there has been work in the assessment of fine motor DDK tasks in ASD. There are limitation to the use of maximum performance tasks for assessing speech motor control. It is not an effective measure with very young children, in a study by Diepeveen et al., (2019) they found in a sample of 1,524 children aged 2-7 years that Less than 50% of the 2-year-olds could produce >1 monosyllabic sequence correctly. Children who could not correctly produce ≥2 monosyllabic sequences could not produce any of the multisyllabic sequences and it was only suitable for children aged 3 years and older. They also found that the effect of instruction ("faster" and "as fast as possible") was small, and multiple attempts yielded a faster MRR in only 20% of the cases. MRRs did not show clinically relevant differences when calculated over different numbers of repeated syllables (Diepeveen et al., 2019). Where MPT are useful is providing a picture of the mechanical rate, accuracy and consistency of repeated targets but does not provide information about speech motor control in the context of additional cognitive loads such as expressive language.

2.8.1 Measurement of Fine Motor Control

The PANESS (Denckla, 1974) is of particular interest in understanding fine motor skills as it analyses these using repetitive timed movement of the fingers, a task comparable to speech DDK. The PANESS (Denckla, 1974) has been normed on 168

children aged 5-10 years and examines both simple repetitive timed movements of the fingers and patterned timed movements using finger apposition. It allows measurement of speed and dysrhythmia, two skills observed to be impaired in ASD. Jansiewicz et al. (2006) found in a group of forty boys aged six to seventeen to have slower speed, increased dysrhythmia with timed movements and greater overflow compared to 55 TD peers. The speed of patterned finger movements had borderline significance and was within the average range for both groups. However, the group of boys with ASD were one standard deviation below the mean in the speed of repetitive timed movements. It suggests that the boys with ASD had timing errors that were identified by this task.

MacNeil and Mostofsky (2012) examined the PANESS (Denckla, 1974) in twenty-four children with ASD, twenty-four children with attention deficit hyperactivity disorder (ADHD) and 24 TD children as well as additional assessments, of praxis and the Postural Knowledge Test, which assesses the ability to recognise and perform skilled hand gestures (Dowell et al., 2009). The children with ASD performed significantly worse than the TD group in both of these assessments. Whilst the children with ADHD performed significantly worse on the PANESS and not the Postural Knowledge Test (Dowell et al., 2009). Children with ASD performed significantly worse in all assessments than the ADHD group, except the PANESS. These results suggest that impaired fine motor control, in relation to timing, may be a more generalized finding. Whereas the impairment of forming perceptual-motor action models in order to produced skills gestures may be specific to children with ASD. This breakdown in perceptual-motor is similar to that recognised by Shriberg et al. (2011) in which speech motor control was not impaired, that the SSEs were likely due to a speech perception issue, such as the inability to effectively “tune-in’ and ‘tune-up’ to the ambient environment. By comparing measures of speech motor control using maximum performance tasks and general SSEs, it allows us to observe whether there are specific differences in ASD in speech motor control as found in their fine motor control.

Freitag et al. (2007) studied motor abilities, including fine motor DDK tasks, in individuals aged 14-22 years diagnosed with the old classifications of Asperger Syndrome (AS) and High function autism (HFA) (now encompassed within the ASD).

DDK was assessed by examining the maximum speed of pro- and supination of the dominant and non-dominant hand separately in each participant. They found impairment in DDK performance as well as dynamic balance skills. It has been suggested that differences in these skills are associated with interaction and integration of different sensory and motor functions in ASD (Gepner et al., 1995; Minshew et al., 2004; Molloy et al., 2003). I was interested in determining whether these differences in fine motor control were also present in speech motor control and in what form.

2.8.2 Diadochokinesis (DDK) Protocols

DDK is one of the more commonly used assessments for measuring speech motor control (Ziegler, 2002c). However, despite its efficacy in differentiating dyspraxia from dysarthria (Thoonen et al., 1996), it can be difficult to elicit with children, particularly those with conditions such as ASD or Down's syndrome. This is due to compounding challenges and ultrasound analysis can be time consuming, rendering it difficult to use in clinic settings where time for SLTs is limited. This study employed the method set out by McCann and Wrench (2007a), a study which set out to change DDK protocol in face of challenges such as the need for demonstration of the task, which can result in varied target production by the SLT. Difficulty for the participant to follow instructions as a result of intellectual impairment can cause further differences in assessment (Cohen et al., 1998). McCann and Wrench (2007) used electropalography (EPG) to identify errors such as silent groping that is not available from just acoustic and auditory analysis. They created computerised "prompts" in which the target syllable or sequence was recorded and then altered to differing rates. The rates for the single syllables were taken from Robbins and Klee (1987) and the sequences from Williams and Stackhouse (2000). Table 3 is taken from McCann and Wrench (2007) and gives the syllables per second for each recorded rate of the prompts.

Table 3: *McCann and Wrench (2007) Recorded DDK Prompt Rates*

	Syllables per Second					
	-3SD	-2SD	-1SD	Mean	+1SD	+2SD
p	2.09	3.01	3.93	4.85	5.77	6.69
t	2.07	2.98	3.89	4.8	5.71	6.62
k	1.87	2.75	3.63	4.51	5.39	6.27
t k	1.32	2.09	2.86	3.63	4.4	5.17
p t k	2.45	2.9	3.35	3.8	4.25	4.7

Table 1: Prompt rates in syllables per second.

McCann and Wrench (2007) found that this protocol was successful in eliciting the expected DDK rate for children who can often be unresponsive to this type of task for the reasons described above. Measuring DDK using this protocol can help determine if the child's rate is likely to be within -3 to +2SDs of that expected from TD children's which norms are reported in the literature for (Robbins & Klee, 1987; Pam Williams & Stackhouse, 2000).

2.8.3 Maximum Performance Tasks in other Populations

Maximum performance tasks are a measure of speech motor control. The development of speech motor control takes place mainly in the first years of life, where the child develops coordination and control over the articulatory subsystems (Kent, 2004; P Williams, 2015). The fine tuning of the speech motor system takes place throughout childhood, it is suggested that full maturation is not reached until 16 years old (Walsh & Smith, 2002). Children with SSEs of differing origins have been shown in the literature to have slow rates of production and inaccurate and inconsistent repetitions in DDK tasks. It may be possible to use DDK to distinguish between children with articulatory errors and phonological errors (see chapter one for discussion) (Bradford & Dodd, 1996; Preston & Edwards, 2007; Wren et al., 2012). Yoss and Darley (1974) were one of the first to propose speech DDK tasks as a way of differentiating speech motor impairments from other speech disorders. They examined speech of thirty children, aged five to nine years who presented with SSEs of unknown origin and compared them to TD peers. They carried out a battery of

speech assessments that included auditory perception and discrimination test; non-speech oro-motor tasks (isolated and sequenced volitional oral movements); DDK tasks and speech production tasks (real and nonsense words, conversational speech and a narrative task). Children with speech disorders performed significantly poorer in all measures than the TD group.

Thoonen et al. (1996) later expanded on this work but investigating whether it was possible to distinguish children with developmental apraxia of speech (also known as childhood apraxia of speech, a motor speech disorder) from other speech disorders on the basis of maximum performance tasks (similar to DDK tasks). Thoonen et al. (1996) looked at DDK mono-syllabic repetition rate and tri-syllable repetition as well as vowel and fricative prolongation. This was carried out with three groups of children aged four to 12 years: TD children, children with spastic dysarthria and children with developmental apraxia of speech (DAS). The maximum performance tasks successfully differentiated children with spastic dysarthria from the other two groups on only two tasks: poor maximum vowel prolongation and slow monosyllabic production. The DAS group were differentiated from the TD group on maximum fricative prolongation and tri-syllable repetition rate as well as greater number of sequencing errors and required more attempts before an accurate sequence was produced. The protocol of this study was tested again by Thoonen et al. (1999) and in addition they examined a group of children with non-specific speech disorders. They found significant dysarthric or apraxic in this group, however, these findings are not generalizable due to the small sample size (n=11) and would require further research. What these studies do tell us is that not only are maximum performance tasks such as DDK able to identify speech motor impairment, but they are also able to differentiate between different types of motor speech disorder, including apraxia of speech and dysarthria. This differentiation was important to my study because I was wanted to understand the nature of the SSEs in children with ASD, DDK provided additional information around speech motor control and where the breakdown may be occurring.

Murray et al. (2015) used DDK to investigate childhood apraxia of speech (CAS) in seventy-two children, assessing them with a battery of speech assessments that included oro-motor, speech and language tasks. From all the assessment carried out

they found that it was polysyllabic word accuracy and an oral motor assessment, including DDK tasks that reliably differentiated CAS and ruled out other speech motor or structural impairments such as dysarthria. They concluded that both DDK tasks and polysyllabic tasks were motorically complex enough to challenge the underlying motor programming and planning deficits central to CAS.

2.8.4 Rate, Accuracy and Consistency

Williams and Stackhouse (2000) developed the proposal that is may be possible to differentiate between sub-groups of children with speech difficulties by assessing rate, accuracy and consistency of their DDK performance. The study was carried out with 30 normally developing children, aged three to five years. They found that in general, accuracy and consistency were more sensitive measures of DDK for younger children, however from five years on, when consistency and accuracy reach ceiling level, then rate becomes a useful indicator. They concluded that DDK tasks provided a rich source of information for assessing the speech motor skills of children. As my study was observing DDK in children aged 6-12 years, rate was included in the analysis of DDK tasks.

Norms of DDK rates of TD children were taken from a study carried out by Fletcher (1978). They suggested using a time-by-count method in which the number of syllables to be produced (e.g., five) were predetermined and the time taken to produce these syllables was recorded. Fletcher (1978) used this measure to then produce DDK rate norms from 384 children aged 6-13 years, a similar aged group to the one used in this study. The consonant targets were the same as used in this study; /p/, /t/, /k/, /tk/ and /ptk/ as well as four additional targets not relevant to this study. This set a standard for DDK rates which has enabled an effective comparison for what should be expected from school aged children in terms of their DDK rate and whether the rate produced by the children with ASD in this study is within the normal range or whether it indicates a speech motor problem.

As inconsistency is seen as a marker of CAS, this was an important aspect of the DDK tasks carried out in this study (American Speech-Language-Hearing Association (ASHA), 2007). Holm et al. (2007) studied and identified the difference

between variability and inconsistency in typically developing young children. They defined variability as productions that differ but are attributed to normal speech acquisitions. Whereas inconsistency is where speech is characterised by a significant number of differing repetitions of repeated productions, as stimulated in DDK tasks, both at the phoneme level and segmental level (consonant-vowel sequence). They carried out a large cross-sectional study with 5409 children of the consistency of their productions of words and found that typically developing children have highly consistent speech, setting a marker that inconsistent speech production is not a typical feature of speech development (Holm et al., 2007). This provides rationale for assessment the consistency of speech in DDK tasks as an effective marker of speech disorder and can applied to children with ASD.

2.9 Cognitive Theories of ASD

In this section the cognitive theories for the potential causes of ASD are discussed, focusing on the three prominent theories: impaired theory of mind, weak central coherence theory, dysfunction of the mirror neuron system.

2.9.1 Impaired Theory of Mind

Impaired theory of mind refers to difficulties in understanding mental state of self and others. It has been specifically reported in children with ASD and was developed by Baron-Cohen et al. (1985). Theory of mind is believed to be core to social communication differences observed in ASD. This theory also explores the atypical development of the precursors to social interaction such as social perception, action-perception mirroring, biological motion processing (Pelphrey, Shultz, Hudac, & Vander Wyk, 2011). This can all have a significant impact on the development of speech, language, and motor skills.

Joint attention, a precursor to theory of mind has been associated with communication differences (Miller, 2006). The ability to respond to caregiver/s joint attention bids has been associated with vocabulary development in both typically developing children (Carpenter, Akhtar, & Tomasello, 1998; Morales et al., 2000) and children with ASD (Sigman et al., 1999). It appears that the early differences in

joint attention exhibited by children with ASD may be a crucial factor in the level of communication differences (Bruinsma, Koegel, & Koegel, 2004; Sigman et al., 1999).

Differences in theory of mind may help explain why individuals with ASD who appear to have normal language skills present with speech distortions. Baron-Cohen (1991) found that children with ASD who had non-native mothers were more likely to develop her foreign accent than the accent of the native ambient environment. These results suggest that children with ASD are less socially motivated to identify with their peers and therefore do not develop the native accent. Perhaps because children with ASD lack theory of mind, they cannot perceive their speech as different from peers. This may explain why individuals with ASD who have developmentally appropriate language skills have distortions or delays in their speech (Cleland, 2010; Shriberg et al., 2011). Differences in theory of mind may also be central to the difficulties in pragmatics of language experienced by individuals with ASD (Lord and Paul, 1997). Individuals with ASD have difficulty taking into account listener's perspectives and this may in turn affect their ability to interact with others (Tager-Flusberg, 1999, 1993). It also appears to have a negative impact on early communication development where a paucity of protodeclarative gestures (requiring joint attention) is noted to be produced by children with ASD compared to controls (Baron-Cohen, 1989). All these factors can impact how a child develops speech and language.

However, ASD is not just a disorder in the cognitive domain but may be a wider neurodevelopmental disorder. Not all symptoms presented in ASD are reflective of a difference in theory of mind. For example, differences in executive function and weak central coherence and repetitive behaviours are seen to be beyond this explanation (Tager-Flusberg, 1999). Also, language differences beyond pragmatics is present in ASD, e.g., difficulties in acquisitions of grammar and vocabulary which is not reliant on theory of mind (Lord and Paul, 1997). Therefore, it is vital to explore interactions of different neurodevelopmental domains to gain a better understanding of this disorder.

2.9.2 Weak Central Coherence Theory

Proposed by Uta Frith (1989), weak central coherence theory suggests individuals with ASD have a preference for local processing over global processing or have a "detail-focused cognitive style". More recently this view has been altered with a suggestion that it is a local processing bias rather than difference. This local processing bias leads to acknowledgment only of specific features. This can be detrimental to language and speech processing which is a multi-faceted process. The weak central coherence theory may partly explain altered auditory/speech perception in children with ASD. For example, Bonnel et al. (2003) found participants with ASD had higher pitch sensitivity. Mottron, Peretz and Menard (2000) found in thirteen individuals with ASD that they performed better in a task in which they detected local level changes in musical stimuli than global level changes when compared to thirteen controls.

Similar findings have been made in face-processing where Lopez, Donnelly, Hadwin and Leekam (2004) found participants with ASD only had 'configural process' (perceived relations among features) when cued and did not in non-cued conditions. However, individuals with ASD have been shown capable of coherence. For instance, in colour and form in a visual speech task (Plaisted et al., 1998a; 1998b) and Beversdorf et al. (1998) found no group differences in recall of coherent vs. incoherent word lists or stories.

Failure to process information globally may be a result of problems in moving from local to global levels. This may result in poor planning as working memory has a bias towards working with smaller fragments of information leading to altered speech and language perception and development. However, it is important to note this may be just one cognitive feature of ASD and could be a local processing bias rather than a global processing difference, meaning individuals with ASD are capable of global processing when cued.

2.9.3 Dysfunction in the Mirror Neuron System

The hypothesis exists that ASD may be related to abnormalities in the mirror neuron system (MNS) (Dziuk et al., 2007). The mirror neuron system includes the brain regions that are active both when an individual performs an action and observes another person perform the same action (Lei et al., 2014). The mirror neuron system

has been inconsistently implicated in imitation or observation of action or emotion in individuals with ASD (Pelphrey et al., 2011). This theory suggests there is a motor aspect of the development of social and imitative behaviour (Cattaneo et al., 2007). The role of mirror neurons is controversial, but empirical evidence of parieto-frontal mirror neuron circuits suggests this could be impaired in ASD and impact the neural substrate for action and intention understanding (Rizzolatti and Sinigaglia, 2010; Casartelli and Mloteni, 2014). The mirror neuron's system has been defined as the ensemble of cortical motor and visceromotor centres furnished with mirror properties (Rizzolatti & Sinigaglia, 2010). MNs encode basic and more complex functions depending on their anatomical location. However, the general feature of mirroring mechanisms is their ability to recognize others' behaviour based on the activation of one's own motor representations. MNs encode the execution of a specific motor behaviour and also the observation of a similar motor behaviour. Individuals with ASD have presented with differences in the ability to discern (Gallese et al., 2013) and to imitate (Hobson & Hobson, 2008) the style of action (i.e. how), while not direct evidence of mirror neuron differences, it does indicate that motor action associated with MNS may differ in individuals with ASD.

Imitation plays a crucial role in development of social communication behaviours (Ingersol, 2008) due to its impact on language, play and joint attention. However, Rojers et al. (2003) did not find a relationship between imitation skills and concurrent language age in children with ASD. This brings into questions whether imitation and motor abilities are related to social communication development. It is unclear whether dyspraxia and imitations abilities, skills associated with dysfunction in motor neuron dysfunction are directly related to the differences in social communication or is there is another developmental variable at play.

2.10 Speech Development in ASD

Individuals with ASD have been found to present with higher rates of speech sound errors (SSEs). SSEs in the case of this project were taken to mean where the target word or sound produced was incorrectly articulated (as described in chapter one introduction) than the typically developing (TD) population, but the causes of these higher rates are not well understood (Cleland, Gibbon, et al., 2010; Shriberg et al., 2011). When first encountering ASD, SSEs and disturbances in prosody are

noticeable characteristics of speech production, however, the sparsity of literature in these field does not reflect this. There has been a significantly larger focus on the arguably more important issues of social and behavioural difficulties in ASD. The speech research that does exist in this field often centres around prosodic issues whilst only a small sample of studies focuses on SSEs. Furthermore, within these studies sample sizes are small, often ranging from five to twenty children or looking at individual case studies, and methods are perceptual and/or widely vary across the literature. What we do know from these studies is that individuals with ASD can also have normal speech development or simply slightly delayed speech, contradicting findings of atypical SSEs in the same population (Kjelgaard & Tager-Flusberg, 2001; McCleery et al., 2006). These studies mostly used perceptual speech assessments with small populations. However, studies that use more in-depth speech analysis measures which go beyond the standard screening assessments by carrying out phonetic inventories have found subtle and atypical SSEs in populations of ASD than TD peers, often at higher rates than when just using perceptual assessment. These researchers have employed speech assessment measures that go beyond percentage consonants correct to find a complex presentation of SSEs in children with ASD (Cleland et al., 2010; Shriberg et al., 2011; Wolk & Brennan, 2013)

The potential causes of SSEs in ASD has not been thoroughly investigated and until recently, researchers were trying to understand whether they were present in ASD but without exploring *why* these may be occurring. Recent studies have revealed phonological patterns in children with ASD that included typical and atypical processes. However, literature is heterogeneous and lacks conceptual organisation (Shriberg et al., 2011). The conflicting result in the literature indicates that fundamental speech sound behaviour is not well understood in this population. It may be due to the existence of a subgroup of children with ASD that exhibit typical and atypical errors in their speech. There are two perspectives within the current literature base that may offer explanation (1) the speech attunement framework first described by Shriberg et al. (2011) and (2) the speech motor impairment theory set out by Belmonte et al. (2013) which was previously discussed in section 2.5.2.

2.10.1 Aetiology of Speech Sounds Errors and ASD

As discussed at the beginning of the chapter, ASD is a neurodevelopmental disorder where there can be occurrence of multiple differences in both the verbal and non-verbal domains (Mattila et al., 2011). Individuals with ASD have been found to present with difficulties in communication, language, and social behaviour, all of which may impact on the intelligibility of their speech. We will explore the current literature in this area; however, it is heterogenic and there is still difficulty drawing firm conclusions on the relationship between ASD and speech production. There is a significant need for a literature base in this field in which theoretical concepts are explored and defined, using similar instrumental measures across the board.

The intersecting of different areas of differences in cognitive domains may result in higher rates of SSEs in individuals with ASD. The dyad of impairment in ASD as discussed earlier could be related to the impairment of speech, social motivation, cognitive abilities, and perceptual control. Social motivation is defined as a set of biological and psychological processes that cause a person to move towards the social world, seeking social interactions and maintaining relationships. It has been found in individuals with ASD that there is a decrease in this social motivation which causes differences in social cognitive development (Chevallier et al., 2012). This difference in social motivation can result in a child missing important communicative cues in early development, resulting in an inability to develop speech at the expected rate. This can be further impacted by cognitive rigidity seen in ASD, which is also associated with speech impairment, evidence has been found in which children with speech disorders had poor scores on cognitive flexibility tasks compared to TD peers (Crosbie et al., 2009). This cognitive inflexibility may be a core component of the social differences and symptoms associated with ASD (Valla et al., 2013).

Differences in social motivation may also impact the development of social cognition neural networks that are responsible for face processing (Sterling et al., 2008). This “piecemeal” cognitive processing style can cause the attention to focus on individual components of the face or physical configuration while losing the social information being communicated in the interaction. This can impair the processing of speech in which the person with ASD focuses on only certain aspects of the speech such as the phonological elements while not processing phonetic information. Both segmental and suprasegmental aspects of speech can be ignored in early speech development and result in differences in speech production.

The processing of perceptual information may also be an important factor in our understanding of SSEs in individuals with ASD (Baum et al., 2015). Sensory and motor experiences are the foundation of speech development. Many young children with speech disorders have reduced functions in the vestibular, proprioceptive, and tactile sensory systems compared to normal children (Takarae et al., 2008; Tung et al., 2013). However, the impairment in sensory processing in ASD is still not well understood. Sensory representations are a cornerstone for higher-order cognitive representations. Therefore, if sensory processing is impaired, this can impair higher order cognitive processes further up the chain, this is mainly found in the form of speech and language differences in individuals with ASD. For instance, studies have shown that individuals with ASD have differences in social orientation to auditory stimulus (Kuhl, Coffey-Corina, et al., 2005; Paul et al., 2007). This is an area lacking in large scale studies and requires significantly more research before we can draw firm conclusions on whether these negatively impact social and cognitive development in individuals with ASD.

Looking at higher-order cognitive processes that may be impacted, there has been a link found between the differences in language, literacy, and SSEs in children with ASD. The relationship between SSEs and language and literacy is complex, with speech sound disorders being found to have a significant risk of poor phonemic skills, word reading and spelling (Carson et al., 2003; Goffman, 1999; Whitehurst et al., 1991; Williams & Elbert, 2003). While the presence of SSEs in ASD may have a modest effect alone on literacy development, when this is compounded with other differences in language or social motivation and cognitive processing, this can cause serious negative consequences for the development of the child. So, the presence of SSEs in ASD may increase risks of a language impairment that can predict a reading disorder (Hayiou-Thomas et al., 2017).

Our understanding of why SSEs may occur in ASD still requires a significant amount of research to be carried out before finding conclusions. Whilst research carried out in the communication field has been sparse in SSEs area, focusing primarily on language, prosody and behavioural difficulties, this has started to change with the significant presence of SSEs now being recognised and researched (Kjelgaard & Tager-Flusberg, 2001b; Owens, 2004; Paul & Norbury, 2012). However, we still have questions on the nature of SSEs in ASD which may give us insights on why they

occur in higher rate. What we do know is that some children with ASD produce speech that is different to the organisation of typical speech production. I suggest this may be a result of a developmental delay but also differences in the cognitive or neuromotor processes required to produce effective speech as well as the social motivation to use it in everyday communicative situations. To increase our understanding of the nature of SSEs in ASD, the current literature base that exists will be discussed in the following sections.

2.11 Historical Research of SSEs in Children with ASD

Over 40 years ago behavioural studies started to conclude that there was a delay in acquisition of typical speech in children with ASD as often found in children with intellectual disability (Bartolucci & Pierce, 1977). SSEs were often described as “oddities” in speech and analysed using only broad phonetic transcription (Pronovost et al., 1966). Due to the lack of in-depth phonetic analysis and little instrumental analysis available at the time, the SSEs found in this period did not reveal enough to identify the type of speech patterns children presented with and often missed the more subtle articulatory distortions present in speech. The main focus of the studies at the time were often other aspects of communication with a small section focusing on speech production. Speech was often only assessed using short perceptual tests of single words or parent reports, significantly limiting findings beyond broad diagnostic categories (Pronovost et al., 1966). This resulted in often conflicting results within the literature base. For instance, Largo and Schnizel (1985) and Weiss et al. (1980) noted a presence of articulation difficulties but carried out no detailed articulatory analysis. In contrast other researchers found normal verbal language, articulation, and phonological abilities (Bartolucci & Pierce et al., 1976; 1977). Until recently there were very few in-depth phonological analyses carried out with this population.

A study at the time that did focus mainly on speech sound production was carried out by Bartolucci and Pierce (1997). In this study single word analysis was carried out using a picture naming task. They assessed both speech perception and production to compare children with ASD to TD peers and children with intellectual disability (ID). Analysis was limited to broad phonetic transcription of speech and measured using the percentage of consonants correct metric. They found none of the children

with ASD were atypical in speech perception and production. When they returned to the same data set later using more sensitive phonemic analysis, they found there was delayed pattern of acquisition of speech sounds similar to the ID group. There was a significant difference in the number of errors in perception and production on liquids (class of consonants consisting of voiced lateral approximants like /l/) produced by children with ASD compared to the ID and TD groups. Measurement of consistency and frequency of these SSEs may have revealed more about the speech profiles of these children. What these studies show is that we require more sensitive measures of speech sound production in order to identify subtle SSEs across different groups and determine the pattern of SSEs produced.

As time moved on from initial studies on speech production in ASD, more in-depth analysis in case studies found impaired speech production in children with ASD using phonological analysis that identified specific speech processes (Wetherby et al., 1989; Wolk & Brennan, 2013; Wolk & Edwards, 1993; Wolk & Giesen, 2000). An example of the wide variation of speech processes found in this sample is in the Wolk and Giesen (2000) study. They carried out a phonological analysis of speech from various speech contexts, specifically object naming and spontaneous speech in four children diagnosed with ASD. They found both typical, delayed, and disordered speech processes in this small sample size. Most interesting were the disordered speech processes identified: chronological mismatch (where sounds are not acquired in typical development order), residual errors and unusual sound changes. Children presented with both articulation and phonological SSEs and all four were diagnosed with a mild to severe phonological disorder, one child defined as non-verbal. Child B, who was primarily nonverbal, has an almost completely absent inventory. He uses mainly vowels and has only two consonant sounds present: the velar nasal [ŋ] and the palatal glide [j]. These are two sound classes expected to occur earlier in acquisition and more likely to be produced with some degree of accuracy in disordered phonology. What small case series like this tell us is that presentation of SSEs in ASD significantly vary by each individual child and that there are likely different subtypes of speech impairment in ASD.

2.12 Atypical Speech in Young Children with ASD

Evidence from multiple studies suggests that children with ASD may have different phonatory qualities from TD children and children with ID at the prelinguistic stage of communication (Schoen et al., 2011). If we are able to identify speech production differences early in life, this would allow intervention to be placed in the optimal period of communication development, when there is significant neural plasticity at the stage of rapid brain development.

Disordered or atypical speech sound development seems to be identifiable at the prelinguistic stages of communication development in children with ASD. Small case studies have used phonological analysis on the speech of young children with ASD and found atypical SSEs rather than delayed or typical for normal speech development (Wetherby et al., 1989). Wetherby et al. (1989) analysed the speech sounds of early vocalisations produced in a 30-minute communication sample and found that compared to TD peers, the three children with ASD had a disproportionate number of vocal acts that did not contain consonants. The lack of consonants seen in this sample of children with ASD may be an early indicator that speech is not on the typical development trajectory. It is often difficult to determine if the lack of consonants is a result of articulatory or phonological impairment and further analysis of these SSEs would be necessary to determine this.

Taking samples of spontaneous communication is the mainstay of analysis of speech development in groups of young children. One notable finding in a study using this technique in with young children (9-12 months) at high risk for ASD is there was a present of “atypical vocalisations” (Schoen et al., 2011). Additionally, it was these atypical vocalisations that were the main prelinguistic communication that distinguished the group of children at high risk for ASD to those at low risk. Schoen et al. (2011) went on to further analyse this type of data with thirty toddlers aged 18-36 months who had a diagnosis of ASD. They looked at phonological and vocal behaviour, coding the non-speech vocalisations without recognisable consonants and speech like utterances through broad phonemic transcription. They found that the group of young children with ASD had a significantly limited number of consonants in their speech compared to two groups of TD peers: both age and language matched. The metric of percentage of consonants correct did not significantly differ from the TD groups but the ASD group did produce significantly less speech-like utterances overall. However, what really distinguished the ASD

group from the TD groups was the presence of “atypical vocalisations” which often came in the form of “high-pitched squeals” These studies tell us that young children with ASD may not align their pre-linguistic communicative act to the phonotactic properties or pitch of their ambient speech environment.

What we may be seeing in the speech productions of young children with ASD is that they are not tuning into the speech and language model of the ambient speech environment (Sheinkopf et al., 2000). As described earlier, their reduced social motivation may result in their failure to attend to their ambient language environment, causing a negative impact on the acquisition of spoken language, in severe cases resulting in the individual remaining non-verbal for life. Sheinkopf et al. (2000) found that the eleven children with ASD produced similar well-formed complex canonical vocalisations to the eleven children with developmental delay, but in addition produced a significant number of vocalisations with disordered vocal quality. Canonical vocalisations are defined as vocalisations that are effectively formed, consonant to vowel sequences that rapidly transition. Differences in canonical vocalisations can indicate an impairment in speech motor control, however this was not the case for these children with ASD, though their production of atypical vocalisations implies there may be a difference in speech perception. The same data set of vocalisations was reanalysed later by Wallace et al. (2008) using acoustic analysis and more defined and refined speech categorisation to discover that the group of children with ASD had more atypical phonatory qualities in their communicative acts. Whereas Schoen et al. (2011) had not found any difference using broader and less specific speech analysis. In-depth acoustic analysis of vocalisations of pre-linguistic children with ASD or at high risk for ASD can reveal vocal profiles that are not aligned to typical speech development. Further research into the cause of atypical vocalisations at this stage of communication development may reveal more about the origin of issues in speech production seen at later stages in speech development.

2.13 Speech Sounds Error Analysis in Children with ASD

As the field of research in speech production and perception in ASD has developed, more sophisticated methods of analysing children’s speech have been integrated into studies, using both qualitative and quantitative methods. For instance, Shriberg

et al. (Shriberg et al., 2001) developed “PEPPER”, a software program to analysis phonetic and phonological evaluation records (Shriberg, 2001) and found a significant number of articulatory SSEs (motor-based speech sound errors like fronting) in children with ASD. This software specialises in the analysis of the frequency and form of SSEs in conversational speech. Speech and prosody-voice profiles for fifteen male speakers with High-Functioning Autism (HFA) and fifteen male speakers with Asperger syndrome (AS) were compared to one another and to profiles for 53 typically developing male speakers in the same 10- to 50-years age range. Following this technique, they found that 33% of children with ASD had a one or more SSEs that were atypical (e.g., lateral lisps or residual SSEs). One notable finding from this research study were the significant number of residual SSEs, 33% of the ASD group whereas the typical rate in a TD population is 1-2% (Flipsen, 2015). Residual SSEs were defined if the child was older than nine years, presented with more than two residual SSEs such as derhotacisation or dentalised sibilants. These types of SSEs are significant as they can persist over the individual’s lifetime without intervention. The presence of these residual SSEs suggests that children with ASD have a disordered speech profile, rather than simply delayed, as assumed from historical research in this field (Bartolucci & Pierce, 1977; Pronovost et al., 1966). These atypicality have been noted in early communication development as well. Schoen et al. (2011) when studying phonological and vocal behaviour found thirty toddlers with ASD exhibited ‘atypical vocalisations and overall limited number of consonants when compared with typically developing children. They did not however find differences in suprasegmental production, children with ASD did not produce vocalisations different in pitch or duration than typically developing peers.

McCleery et al. (2006) investigated consonant production of fourteen severely language delayed children with ASD and ten typically developing children and found the same general patterns of speech sound production. They carried out less in-depth analysis than Shriberg et al. (2001) by using a communicative inventory (MacArthur-Bates Communicative Development Inventory; Hutchins et al., 2013) a parent report checklist, which may have resulted in subtle articulation errors not being identified. They did not identify significant differences between children with ASD and typically developing children.

Cleland et al. (2010) found further evidence of SSEs in children with ASD in which they reported atypical SSEs. Participants were sixty-nine children aged 5-13 years; thirty had high functioning autism and 39 had Asperger syndrome. Carrying out perceptual analysis using standardized clinical speech assessments, they found only 12% of the sample reached the score threshold for a speech delay/disorder. The perceptual analysis was carried out using the Goldman Fristoe Test of Articulation (Goldman & Fristoe, 2000) which assesses the speech sound production of children in the context of single words. When Cleland et al. (2010) investigated the same sample using further in-depth phonetic and phonological analysis, 41% of the groups produced SSEs indicative of both speech delay and disorder. They found that non-developmental SSEs are present despite whether the child's score in a clinical speech assessment was within the threshold for a speech disorder or not. This implies that atypical SSEs produced by individual with ASD may not be picked up by traditional speech assessment used in clinic settings. When more in-depth phonetic and phonological analysis is carried out, we start to see consistency in atypical SSEs reported in children with ASD. Future research needs to assess speech production of children with ASD beyond single word assessments and include single words of increasing motoric complexity such as polysyllables and specific speech motor tasks, such as maximum performance tasks, and spontaneous speech. This may reveal articulatory or phonological SSEs that indicate a specific speech profile of speakers with ASD.

This research is also in agreement with previous studies carried out by Rapin et al. (2009). Rapin et al. (2009) provide larger-scale support for articulatory/phonological difficulties in children with ASD. The study drew on 118 children with autism available for testing at school age. They belonged to an original cohort of 176 preschool children with DSM III-R Autistic Disorder (AD) who participated in a multi-site NINDS-supported study of autism compared to non-autistic developmental disorders. Carrying out the photo articulation test (PAT; Lippke et al., 1997) they analysed the speech and language characteristics of 62 children with ASD. They found no obvious differences in speech production between children with ASD and typically developing children. However, the PAT (Lippke et al., 1997) looks mainly at single words which is not necessarily an accurate reflection of a child's speech sound production capabilities. Additionally, Tamási (2010) looked at segment

interactions in six children with ASD and found nasal and voicing disturbances. The speech analysis of transcriptions carried out the on the speech samples produced information not just on the underlying and surface representation of speech segments but also the phonological processes in each phonotactic position. This resulted in finding that nasals were severally limited in every phonotactic position and nasals in clusters were atypical. All six children had non-developmental devoicing patterns in clusters.

In the Cleland at al. (2010) sample of children with ASD, they found their speech profiles were defined by developmental phonological errors (e.g., final consonant deletion, gliding), non-developmental SSEs (e.g., initial consonant deletion) that indicate a speech disorder. Going further than the previous studies mentioned, Cleland et al. (2010) examined reasons why there may be SSEs occurring in higher rates in the ASD group by carrying out a battery of standardised assessments in non-verbal cognition, language and speech to unveil if there were any causal links. However, they found no relationship. The higher rates of SSEs may be a result of another impeding factor than language or non-verbal cognition, such as a neuromotor difficulty as hypothesised by Cleland et al. (2010). Following Shriberg et al. (2011) work, it could also be due to speech attunement difficulties, where the child has difficulty “tuning in” and “tuning up” to their ambient speech environment.

A phonetic inventory and speech process analysis carried out by Wolk and Giesen (2000) found in four children with ASD that their speech processes found in their speech profile were indicative of a speech delay and speech disorder. There was the presence of atypical speech processes such as chronological mismatch, residual SSEs, an indicator of speech motor issues and unusual sound changes, indicative of speech perceptual issues. From the speech processes alone, it is unclear whether these atypical SSEs are a result of speech motor control impairment or speech perception issues and would have benefitted from further targeted analysis of these aspects. Future research is required with larger number of participants with further in-depth phonological and articulatory analysis. Additionally, we need to include a wider variety of variability to determine interactions with speech sound behaviour. For instance, social, cognitive, language and motor skills.

2.14 Instrumental assessment of speech

Shaping and moving the tongue involves coordination of a complex range of muscles (Dawson et al., 2015). Inaccuracy or uncoordinated movements of the tongue may indicate the presence of motor coordination impairment. Previous studies have used perceptual methods of assessment, where the SLT listens to the speech and transcribes errors. This is problematic, as perception-based phonetic transcription is known to be unreliable (Howard & Heselwood, 2011). Identification of speech errors using instrumental analysis leads to more accurate diagnosis and better therapy planning (Howard & Heselwood 2011). This study uses ultrasound tongue imaging to make direct observations of the tongue during speech. Perceptual analysis is unable to identify silent movements of the tongue (McCann & Wrench, 2007b). Ultrasound allows us to investigate the presence and type of speech errors produced by imaging from near the tongue tip to the root. The speech profile of children with ASD was compared to typically developing children.

2.14.1 Ultrasound Tongue Imaging

Instrumental analysis of speech allows us to directly measure articulatory movements during speech (Gibbon, 2009). Direct measurements allow for identification of speech motor impairments through direct observation of the tongue. However, some instrumental techniques are uncomfortable and/or invasive and therefore may not be suitable for working with children with ASD. Electropalography (EPG) enables measurement of timing and tongue location in relation to the hard palate. McCann and Wrench (2007) have successfully used this technique with children with Down's Syndrome. However, EPG requires a custom-made artificial palate to be fitted to the child's hard palate and worn throughout the speech recording. For this study that was judged to be too invasive for children with ASD who may have issues with sensory overstimulation (Roley et al., 2015) causing an inability to tolerate the mould. Other non-invasive techniques such as magnetic resonance imaging (MRI) may have been suitably non-invasive and can produce high quality images of speech in movement (Baer et al., 1991). However, MRI scanners are expensive and require specialist knowledge which was outside of the scope of this project. Additionally, MRI requires lying down in a large cylinder which

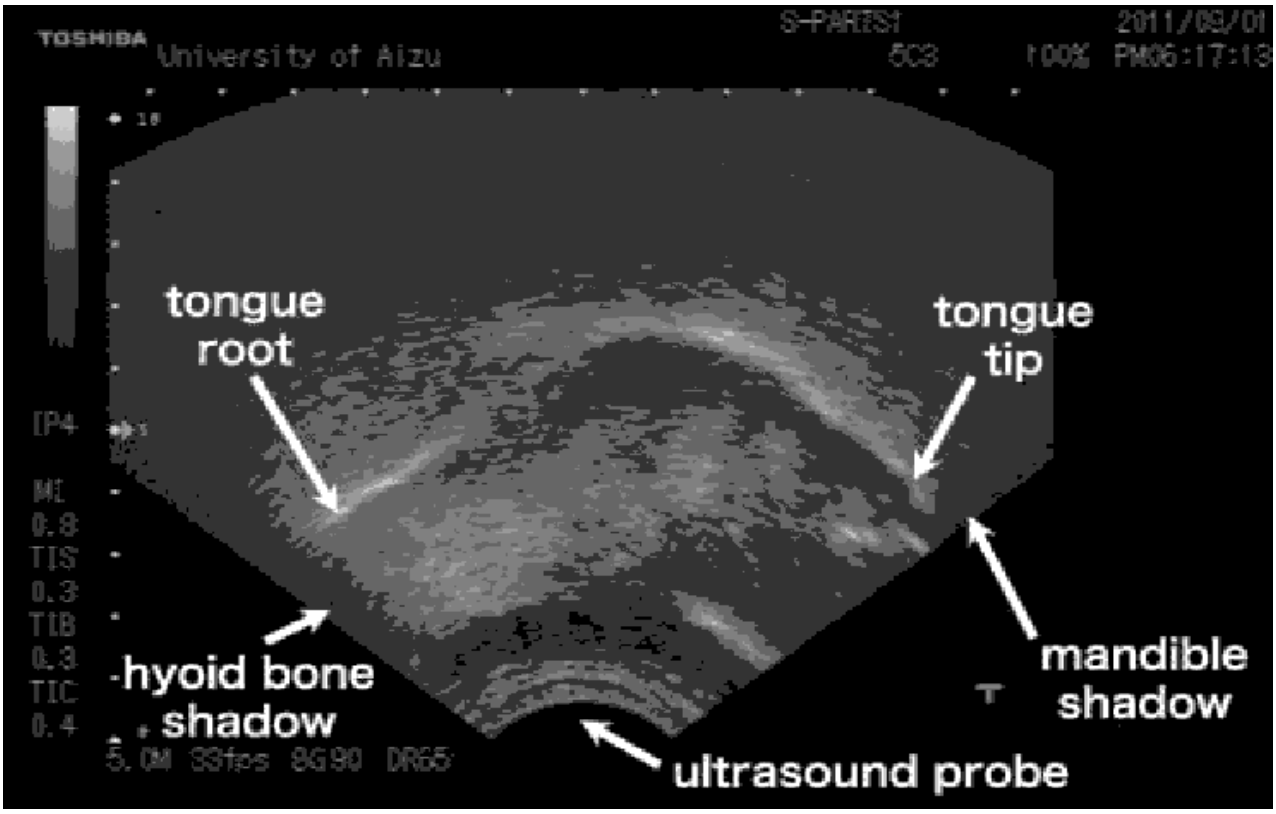
could cause distress to children with ASD due to the claustrophobic inducing environment (Gibbon, 2009).

Ultrasound used in the imaging of speech allows investigation of tongue movement. Ultrasound has been used in the field for decades with Kelsey et al. (1969) describing the application of ultrasound as a "useful tool" for speech research. However, until recently the technology was hard to gain useful data from. Now ultrasound is smaller, portable, provides fast frame rates and can synchronise ultrasound image with audio (Cleland et al. 2016). Ultrasound provides an image of the tongue surface in a safe non-invasive way. It allows application of mathematical attributes to the tongue shape to examine pattern differences produced by different clinical populations (Dawson et al., 2015).

2.14.2 Ultrasound Tongue Imaging Functioning

This study used a medical ultrasound probe (a transducer) which contains piezoelectric crystal that allow pulsing of ultrasonic waves, soundwaves that are beyond the range of human auditory perception. The ultrasonic waves which are emitted are soundwaves of 2-5MHz what travel through the body tissues. As ultrasonic waves are reflective, after every pulse that is emitted, an echo is returned from reflective surfaces. This echo is converted to a strength and distance and plotted to give an image, allowing us to view the boarders of tissues of different density. In terms of ultrasound of the tongue, it is the air gap just above the surface of the tongue which is reflected back (Figure 4). The curve created from the white line of the air above the tongue, serves as a representation of the tongue during speech in which can then be plotted onto a graph with points along the curve are given mathematical attributes.

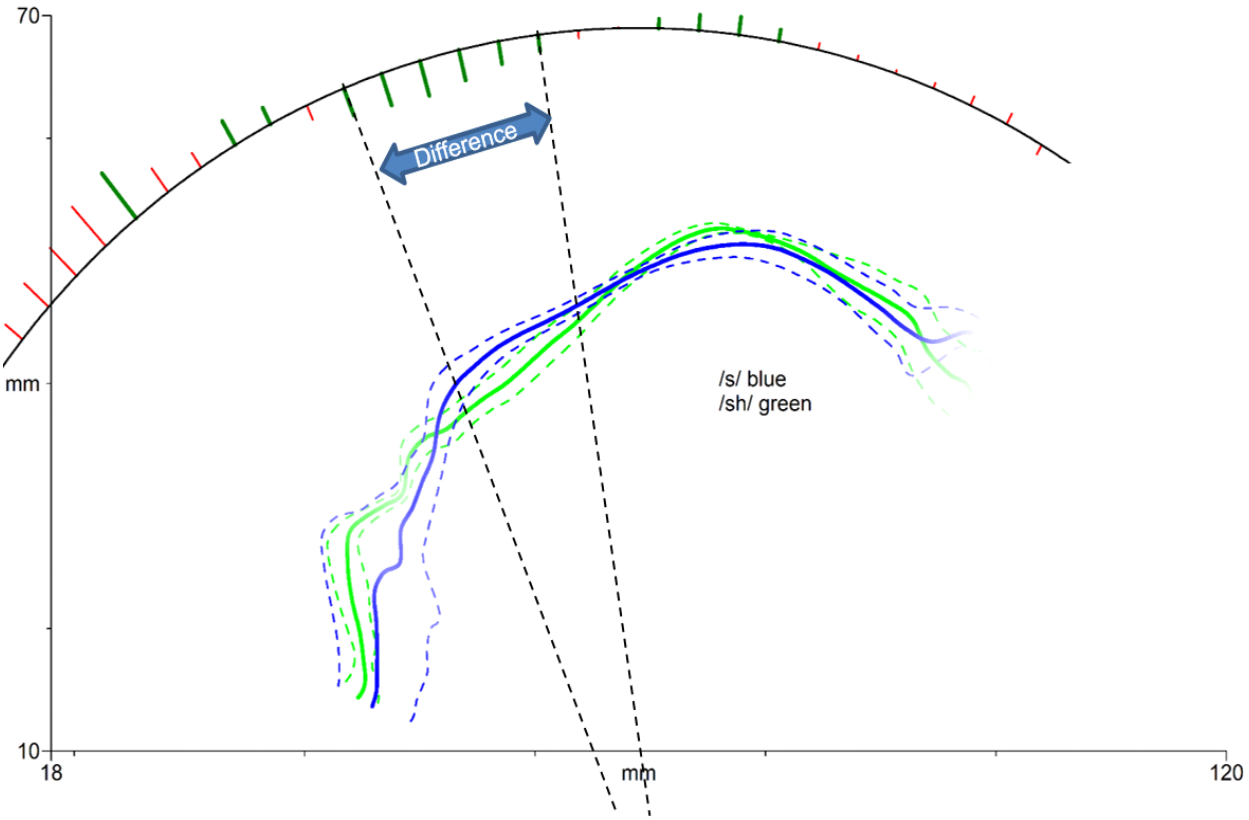
Figure 4: *Ultrasound Image of Tongue in Midsagittal Position*



2.14.3 Variability Measures using Ultrasound

This study used ultrasound to measure increased variability, where there may be different tongue shapes per repetition (Eshky et al., 2018). Increased variability at the sub-phonemic level has been suggested to indicate a subtle speech motor control problem (Zharkova et al., 2015). This variability cannot be readily identified during perceptual speech assessment as the transcriber will use the same broad phonetic symbol to describe each repetition. Whereas ultrasound shows variability through the tongue shape curves.

Figure 5: *Spline Export of Tongue in Midsagittal Position*



There has been little to no previous research carried out with children with ASD and their articulatory variability, however this has been examined in other populations using different instrumental techniques. Grigos et al. (2015) examined the spatial and temporal characteristics of the speech motor control. In eleven children aged three to seven years with CAS, eleven children with speech delay (SD) and 11 TD controls they examined articulators during a task in which the length of words increased using a motion capture system. The motion capture was used to track the upper and lower lip movements as well as the jaw. They found that movement variability was significantly higher in the CAS group compared with the SD and TD groups (Grigos et al., 2015). While both the CAS and SD group shared difference in temporal control, what differentiated these two groups was the movement duration and variability, in which the CAS group performed significantly worse. Therefore, movement variability and duration can be effective measures in which distinguish children with a motor speech disorder from those with speech delay.

There have been reports of general movement variability in this group. Manicolo et al. (2019) found in thirty-two children with ASD they had significantly higher gait (walking) variability that TD controls when assessed using the Movement

Assessment Battery for Children (Brown & Lalor, 2009). However, it is not clear whether this would translate into their speech motor control as speech may be a task dependent skill which relies on other specific neural pathways to achieve its goals (Ziegler, 2002b). Analysis of variability of speech in ASD has tended to focus on the suprasegmental aspects of speech.

Individuals with ASD have been reported to have monotonic and over-precise speech qualities (Lord et al., 1994). Kissine and Geelhand (2019) carried out a syllable-level analysis on speech data (narrative and spontaneous speech). They compared twenty adults with ASD compared with twenty neuro-typical adults. Their focus was on suprasegmental features; fundamental frequency, jitter, shimmer and the first three formants. They found the individuals with ASD showed a greater articulatory stability in their production of vowels. Results suggested the ASD group showed less variability in the vibration of their vocal folds during vowel production. Additionally, they use data from formant dispersion, the first three formants, which are determined by their position of articulators. Their results showed the ASD groups' articulatory gestures were less variable than the TD group. Despite the ASD group having longer syllable durations, leaving more room for variation, but despite this, the ASD group had increased articulatory stability. This study indicates that despite larger variation in general motor abilities this may not reflect in their articulatory and phonatory stability. The interest of this is whether this articulatory variation or lack of, is evident when directly measuring tongue movements using ultrasound.

2.14.4 Ultrasound Measurement in Motor Speech Disorders

Whilst there has been little research of speech in children with ASD using ultrasound, other clinical populations with motor speech disorders have been examined. Heyde et al. (2016) used ultrasound to assess fluency of speech in three people who stuttered and three control speakers. Despite the small sample size, they were able to identify different articulatory strategies of the two groups using ultrasound. The people who stuttered were shown to have an increased variation in tongue shapes and decreased mean peak velocity. Furthermore Zharkova, Hewlett and Hardcastle (2011) used ultrasound to examine motor control development in ten

TD children and ten adults using measure of coarticulation (the articulatory overlap of speech). Specifically, they measured the distances between tongue curves drawn from the ultrasound image and found significant differences between the groups. Children had a significantly greater amount of anticipatory lingual coarticulation, indicative of a less mature speech system.

Kocjancic (2010) used ultrasound to examine lingual movement of three teenagers with CAS compared with 10 TD children and ten adults. They used ultrasound to carry out temporal and articulatory measurement of syllable and segment duration. At a group level, they found that the teenagers with CAS differed in syllable duration and rate of tongue movement with adults but not control children. When examined individually, ultrasound showed the teenagers with CAS had different abilities in adapting tongue movement when changes in syllable structure occurs. Ultrasound allowed direct observation of these sub phonemic level changes not evident in perceptual assessment. Sreedevi and Irfana (2015) also used ultrasound to observe the articulatory pattern in a case study with one child with CAS compared to a TD control. They found variation across trials in the child with CAS but not in the TD control. The placement of the CAS speaker's tongue varied across trails, with variation evident at the front of the tongue and posterior tongue body. Variation was not significant in the TD control speaker. Whilst little research has been carried out with speakers with motor speech disorders using ultrasound to observe differences, these small studies show us it can be an effective technique in differentiating variation and rate difference in speech.

2.14.5 Syllable Durations in Other Populations

Segment and syllable durations have been identified in children with CAS which has not been found in my group of children with ASD. Maasen et al. (2001) researched syllable duration in a small sample of five children with CAS and six TD children. TD children significantly shortened /a/ and lengthened /s/ in consonantal clusters whereas the children with CAS showed no significant differences in segmental duration. Research however is conflicting and often just with small sample sizes due to the small populations of CAS that exist. Nijland et al. (2003) in another small sample (six children with CAS, six TD children) found that children with CAS

produced longer segments than TD children and also showed a different effect of syllable structure. The results from these studies imply an underlying timing issue present in CAS and evidence a speech motor control issue as duration of syllables and sequences are a result of articulatory movement.

Timing regularity in childhood apraxia of speech has been described as having staccato-like rhythmic quality and syllable segregation. Shriberg et al. (2003) examined conversational speech in fifteen children with CAS, thirty TD children and thirty children with speech delay using signal processing to identify and measure duration of speech and pause events. Children with CAS had more variation in the duration of pauses and/or less variation in duration of speech events. Shriberg et al. (2003) interpreted this as evidence that speech timing is a core feature of apraxia of speech.

2.15 Assessment of Motor Control

Weaker motor skills in ASD have been associated with greater language and social communicative differences, but the mechanisms underlying, and the nature of this relationship is still unknown (Hellendoorn et al., 2015). This study will investigate whether children with ASD have an atypical movement profile, particularly in fine motor control.

Fournier, Hass, Naik, Lodha, and Cauraugh (2010) carried out a meta-analysis, which included studies from a wide age range of participants and found that individuals with ASD exhibited difficulties with motor coordination in particular. Motor differences in ASD are often explained in terms of kinematic and sensorimotor problems (Cook et al., 2013; MacNeil & Mostofsky, 2012; Whyatt & Craig, 2013) highlighted in the paper by Trevarthen and Delafield-Butt (2013). A variety of mechanisms have been proposed for the observed difficulties in motor functioning. These range from abnormalities in the cerebellum (Fatemi et al., 2012) disruption in brain synchronization (Welsh et al., 2005) and impaired sensory input and multisensory integration.

Using assessments with a combination of gross motor and fine motor skills may not be sensitive enough to identify a specific atypical motor profile of children with ASD.

Provost et al. (2007) found no significant differences between children with developmental delay and ASD using two general motor developmental scales that combined gross and fine motor measures. Additionally, Noterdaeme et al. (2002) used a German qualitative assessment that reviewed a wide range of motor skills and found few statistically significant differences between eleven children with ASD, eleven with expressive language disorder and eleven with receptive language disorder. Results from both studies suggest there is no atypical motor profile specific to children with ASD.

In contrast, when looking at aspects of fine motor and gross motor as distinct skills using the Movement Assessment Battery for Children 2 (MABC-2; Henderson et al., 2007); found in the subtest on manual dexterity (threading lace) children with ASD performed significantly worse than a specific language impairment (SLI) group. These results suggest a motor profile specific to ASD may exist in fine motor control. The conflicting results in the literature may be due to the methodology. McPhillips et al. (2014) used the MABC-2 (Henderson et al., 2007) which looks at three areas of fine motor and gross motor in detail (manual dexterity, aiming and catching, static and dynamic balance). Future research should test specific areas of motor functioning rather than only conducting a general overview of motor skills in order to identify differences specific to ASD.

Similarly, when combining the gross and fine motor score on the MACB-2 (Henderson et al., 2007) there were no significant difference between children with ASD and children with specific developmental disorder of motor function. However, when examining subtests within fine motor functioning, there was a significant difference in manual dexterity subtests between the groups. Analysing the separate components of fine and gross motor function may allow us to develop a specific ASD motor profile to compare to speech and language abilities and indicate if any interactions exist. These conclusions are not possible when general ratings of motor are compared to general ratings of speech and language.

The ability to communicate effectively may be impacted by atypical kinematics. For instance, Cook et al. (2013) found correlation between kinematic atypicality and ASD Diagnostic Observation Schedule (ADOS; Lord et al., 2000) which scores are impacted on by effective communication through gesture and facial expression, two

components likely to be impaired through atypical kinematics. Whilst there is little research on speech DDK in children with ASD, some work has been carried out in the area of fine motor control using behavioural assessments and in neuroimaging. Examination of fine motor impairment can provide valuable insights on brain development (Denckla, 1985).

Historically fine motor differences have been identified in children using methods such as the grooved pegboard test, a small wooden board that required fine motor control to move pegs into holes. This study found children with ASD were 3-4 standard deviations below the mean in both their dominant and non-dominant hands (Knights & Norwood, 1980). Freitag et al. (2006) assessed fine motor DDK by setting a task that require pro- and supination of the dominant and non-dominant hand separately. Only two of sixteen adolescents with ASD had typical results. The performance of adolescents with ASD was significantly different from the TD controls

Muller et al. (2001) used fMRI to assess eight children with ASD and eight controls on a visually placed finger movement task. They found children with ASD had greater variety in their functional maps and less distinct regional activation patterns. This means that motor pathways may not be well organised in ASD, leading to differences in fine motor skills. Various measures have been used to examine the fine motor skills of children with ASD such as the MABC-2 (Henderson, Sugden, Barnett, & Harcourt Assessment., 2007), Zurich Neuromotor Assessment (ZNA; Largo et al., 2001) and the Physical and Neurological Examination for Soft Signs (PANESS; Denckla, 1974). The MABC-2 (Henderson et al., 2007) found children with ASD had motor impairment across manual dexterity, balance, and ball skills (McPhillips, Finlay, Bejerot, & Hanley, 2014). Lower IQ was associated with greater motor impairment.

Biscaldi et al. (2015) also assessed fine motor DDK using the ZNA (Largo et al., 2001) and found slower rates of basic movements of the fingers and hands and decreased quality of movement in children with ASD and ADHD, which significantly differed from typically developing children. They also identified strong associations of performance on fine motor performance, pegboard, and static balance task in children with ASD, suggesting an overall impairment in motor skills. However, there was no further analysis of the movement profile carried out.

Looking outside the field of ASD and with children with confirmed speech and language disorders, there seems to be overlap with motor impairment. Visscher et al. (2010) compared gross motor skills of four groups of children: children with speech disorders (n=16), those with language disorders (n=41), or those with both (n=48) and typically developing children (n=105). Significant differences in motor performance was noted between the typically developing group and the three groups with speech and language issues. Interestingly the children with only language disorders performed better than those with speech difficulties. This study suggests a link between speech and motor impairment however it is not clear-cut where the issue arises. Similarly, Bradford and Dodd (1994) assessed the motor planning abilities of speech disordered children split in three groups; ten phonologically delayed children, ten children whose phonological system was characterised by the consistent use of non-developmental phonological processes (deviant consistent group); and 10 children whose production of specific lexical items and phonological features was variable (inconsistent group). The inconsistent group performed more poorly than all the other groups on a more complex, timed motor planning task and on an expressive novel-word learning task. This suggests that the type of SSEs children with speech disorder produce may be different depending on where the breakdown is occurring in the speech processing chain and whether fine motor planning is impacted. Also, that the speech processing abilities of children may have a later effect on the motor plans they store. A similar approach could be taken when determining relationships between speech and movement in children with ASD, depending on manifestation of these difficulties, the breakdown in the speech and sensory chains may be at different points.

2.16 Interaction with Language

Researchers have found that many aspects of language are impaired in children with ASD. Discourse and pragmatics, known as the “socially motivated” domains have been found to be the most consistently impaired in ASD (Kelley et al., 2006). In addition, impairment in narrative ability has been found in children with “high functioning ASD” in which they had difficulty communicating the structure of a narrative (Kelley et al., 2006).

To gain a more complete understanding of language development in ASD, it would be useful to examine multiple measures of expressive and receptive language and its interaction with speech and motor development (Tager-Flusberg et al., 2009). This is because one process likely influences the development of another and it is difficult to decouple language, social and cognitive skills (Eigsti et al., 2011). Lewis et al. (2011) suggests that SSDs and language difference may have shared endophenotypes that may be partly genetic. Careful examination of motor performances in well-defined subgroups of children with ASD (previously defined as Asperger syndrome and High Functioning ASD), children with language impairment and TD children may enable researchers to qualify more clearly the nature of the relationship between language and motor problems in ASD.

Language development requires complex, structural, rule-governed brain operations that operate in conjunction with other networks such attention, memory, executive and motor operations (Mahon & Caramazza, 2008). It involves a one-way dedicated input-processing output system but is significantly more complex due to the feed-forward and feedback loops of every language node in the process. This ensures bottom-up and top-down processes that enable the speed and execution of language alongside other competing neural processes (Oldehinkel et al., 2019). Previously researchers have focused on the atypicality of expressive language in children with ASD and how these relate to other cognitive processes (Barett, Prior and Manjuiona, 2004). However, there has not been enough investigation in different language profiles presented in children with ASD and the interaction with other cognitive processes.

2.16.1 Receptive or Expressive Language Delay

Development of receptive language over expressive language is expected in typical infant language development (Fenson et al., 1993). Children with ASD have generally shown an impairment in expressive language and receptive language, but it is not known to what extent. Hudry et al. (2010) observed receptive and expressive language in children with ASD aged 24-59 months varying from pre-verbal to fluent speech. They used two parent report measures and one clinician report and found significant variability on all three language measures, but in particular receptive

language was lower than expressive. This was associated with non-verbal ability and affected a third of the pre-schoolers in a larger sample. These results suggest there may be subtypes of language disorders present in children with ASD, this in turn may be interacting with speech development. This language profile is different from developmental language disorder in which it is expected that lower receptive language would have a negative effect on expressive language abilities. The language profile described by Hudrey et al. (2010) was also found by Seol et al. (2014) appearing often in children with ASD.

Luyster et al. (2008) and Kjelgaard and Tager-Flusberg (2001) found that there was a relatively greater impairment in expressive language. This atypical pattern has been inconsistently identified due to different measures being used by different researchers. Even within studies, conflicting findings are found across a range of measures. Kjelgaard and Tager-Flusberg (2001) found an atypical pattern of greater impairment in expressive language using the Vineland Adaptive behaviour scale (Yang et al., 2016) and the CELF-4 (Semel et al., 2003) but no difference in receptive and expressive single word vocabulary scores using the Peabody Picture Vocabulary test (Naglieri, 2004) and expressive vocabulary test. These results suggest that differences in expressive and receptive language abilities may only be identifiable in more complex communication contexts beyond single word ability. However, Allen and Rapin (1992) found in 262 children with ASD that receptive language was deficient in all. Allen and Rapin (1992) were using the old ASD terminology in which Asperger's syndrome was not included, which is the subset of children within the diagnostic criteria whose language abilities are relatively spared. The term ASD now includes these children (World Health Organisation, 2017). What is evident from previously discussed studies is the variability in language profiles. Some children have typical profiles of receptive language, but others showed highly atypical receptive language abilities.

2.17 Summary

Studies have shown the presence of higher rates of SSEs in children with ASD (Cleland et al., 2010; Shriberg et al., 2001, 2011; Wolk & Brennan, 2013). However, other researchers have countered that these SSEs are within the sequence of typical

speech development and at most delayed, rather than disordered or atypical (Kjelgaard & Tager-Flusberg, 2001a; McCleery et al., 2006). The inconsistencies in the literature may be caused in part by the over reliance of perceptual speech measurements that can be inconsistent and lack specificity in their results. Additionally, the heterogeneous nature of ASD could be the cause of differing findings, individual children present differently. If there are differences present in speech motor control in this population then instrumental measures with higher specificity are required. This is vital as speech motor control issues can be subtle and their presence may indicate differences in motor abilities that is core to ASD. It is unlikely that perceptual measures and broad-brush analyses such as percentage of consonants correct of single words could identify these subtle speech motor control problems. However, maximum performance tasks such as diadochokinesis that specifically assess the speech motor control system may reveal more of the speech profile of children with ASD. Moreover, articulatory analysis could also identify quantitative differences in speech sound production of children with ASD compared to TD speakers. Articulatory analysis such as ultrasound tongue imaging has been successfully used to analyse and treat SSEs in children with ASD. Cleland et al. (2019) found in a study of ultrasound visual feedback, three children with ASD responded to intervention which facilitated speech sound learning. Whilst ultrasound tongue imaging is still in development as a tool for assessment and intervention, it has shown to be a promising technique for in-depth articulatory analysis of SSEs which helps identify subtle articulation errors that may be present in children with ASD.

In conclusion, it is important that we determine *why* SSEs are present in higher rates in children with ASD and whether this is a result of an impairment to speech attunement. Each aspect of speech perception and speech production can impact the other in the speech development of a child. Children with ASD may have less social motivation to “tune in” to the speech of their peers. This may cause a reduction in their motivation to attune their speech production so that is functional and intelligible for others’ comprehension. This may account for why there are prosodic abnormalities and residual SSEs which do not severely impact intelligibility, e.g., phoneme-specific nasal emission as found by Cleland et al. (2010) and difficulties in production of multisyllables (Shriberg et al., 2011). A greater

aetiological understanding of these SSEs in ASD will help determine the underlying capacities and therefore paths to effective speech intervention. The current literature does not show a clear understanding of whether either theory is applicable – speech attunement or speech motor control. Additionally, there may be a subtype of children with ASD with higher rates of SSEs within the diagnostic category. Such theories require further research and both behavioural and instrumental analysis. Understanding the cause of SSEs in ASD will improve the speech intervention offered as it could target the underlying impairment of the SSEs and not just the surface level symptoms. These could be carried out at an earlier age for more effective intervention and aid the development of biomarkers for earlier identification of ASD.

Chapter Three - Behavioural Methods, Results and Discussion

3.1 Introduction

This chapter discussed the behavioural methods applied to answer the following research questions:

- Do children with ASD produce significantly more speech sound errors (SSEs) compared to typically developing children?

Hypothesis: Children with ASD will present with more overt speech sound errors than typically developing children.

Analysis: I compared percentage consonants correct (PCC) from the Diagnostic Evaluation of Articulation and Phonology (DEAP; Dodd, 2002) between groups. I then carried out an independent samples t-test to compare the ASD group and the control group for this analysis and predicted that children with ASD would have significantly lower PCC. To explore this research question at the perceptual level, speech was assessed using single word level analysis, in the form of a phonology and articulation screening assessment (DEAP; Dodd, 2002) and then a multisyllabic word screening assessment (CUW; James, 2009) to observe performance in more structurally complex words. It involved the following levels of analyses to determine if children in the ASD group produced more speech errors than TD controls in a range of speech contexts:

- Percentage of Consonants Correct
- Speech Processes (including total errors, age-appropriate errors, delayed errors, and unusual errors which have been defined in the analysis section of this thesis)
- Inconsistency in Speech Production

3.2 Study Design

An independent measure, quantitative research design was employed to carry out this study. This study was a pilot study which observed differences between the ASD group and a group of TD developing age matched peers and also looked at case controls, observing individual children's speech and movement profiles. Information on speech, fine motor control, and language was collected and identification of any differences between the experimental and control groups was made. This project used perceptual and instrumental analysis of speech and fine motor control to investigate if an interaction exists in ASD. Perceptual analysis was carried out using standard clinic assessment of speech, language and movement. Instrumental analysis involved measuring movements of the tongue using standard medical ultrasound. Determining the nature of the speech movements in ASD will help speech and language therapists (SLTs) to choose appropriate speech therapies for children with ASD.

The goal was to recruit twenty children with ASD from NHS Speech and Language therapists in NHS Greater Glasgow and Clyde, NHS Lanarkshire and/or NHS Lothian. To match this a further aim was to recruit twenty typically developing children of the same age and gender to serve as a control group. However, there were significant difficulties in recruiting this number. This was due to lack of protocol or pathways in place for recruitment of children to research in the local authority area for the TD and ASD children. It required significant time to reach out to schools individually. No children were recruited through the NHS as a result of poor response levels from local speech and language therapists (SLTs). This may be due to time constraints as a result of large caseloads in the NHS. The final recruitment number was ten children with ASD and 10 TD children all of which were recruited from schools in the Greater Glasgow and Clyde area.

3.2.1 Ethical Approval

Ethical approval for the experimental group was granted by the West of Scotland REC 3 NHS Ethics Committee. In addition, NHS Research and Development

approval was granted through the NHS. For recruitment of children with ASD from local schools, ethical approval was granted by the University Ethics Committee from the University of Strathclyde. For recruitment of typically developing children, University of Strathclyde, School of Psychological Health, and Sciences ethics approval was granted.

3.2.2 Recruitment Procedure

For recruitment of children with ASD, letters were sent to NHS paediatric speech and language therapy managers about the project asking them to refer suitable children. They passed on information packs to speech and language therapists in the area. The speech and language therapist distributed recruitment information (Appendix 1) to potential participants, inviting them to get in touch via phone, email, or letter if they were interested in taking part. Parents/carers were then invited to contact the researcher to arrange the first research session. There were no successful responses from this method. Only one SLT responded to the call out and no parents came forward.

For recruitment of children with ASD and the control group of typically developing children we contacted schools in the area through local authorities. Once approval was granted recruitment letters (Appendix 2) was sent to headteachers from local schools asking them to distribute information to parents. The recruitment letters invited parents/carers to get in touch via phone, email, or letter if they were interested in taking part. This method of recruitment was where all twenty participants were recruited from.

Before attending the first research session, all parents were provided with and asked to fully read, the participant information sheet (Appendix 3 and 4). Parents/carers understanding of the study was confirmed in person by the researcher before informed consent was gained (Appendix 5). Also, participants in both groups were asked to read all participant information sheets and the researcher confirm their understanding of the study before starting. Participants were also given child information sheets which they explained what would happen in the study to check if they wanted to participate. It was made clear to parents/carers and children that they

could withdraw from the study at any point but that any data collected before this would be retained.

3.2.3 Inclusion Criteria

Below is a description of the inclusion and exclusion criteria for participants. Where it is not specified, the criteria applied to both groups.

3.2.3.1 Age Range

The age group of six to twelve years old was chosen due to relative stability of speech development at this stage. It is recognised that the maturation of speech motor control is a long process, often not complete until late adolescence (Smith, 2010; Walsh & Smith, 2002), however speech sounds are expected to be in place on average at seven and a half to eight years old (McLeod & Baker, 2017). To account for individual variability in speech development we expanded the age range from six to twelve. For the experimental group children with a confirmed diagnosis of ASD were recruited. Primary school children (six to twelve years) were chosen as they are more likely to have stable characteristics of ASD in comparison to younger children (Woolfenden et al., 2012).

3.2.3.2 Diagnosis of ASD

The school referring a child for participation in the study confirmed the diagnosis of ASD which had been carried out previously by a multidisciplinary paediatric team of health care professionals. A diagnosis of “ASD” encompasses both high functioning ASD (HFA) and Asperger’s syndrome (AS). Inclusions of HFA and AS in the same experimental group is to reflect the current diagnostic criteria in the Diagnostic and statistical manual (DSM-V; American Psychiatric Association, 2013) in which all of the above diagnoses are recognised as part of the “Autism spectrum disorder” (ASD).

3.2.4 Exclusion Criteria

The exclusion criteria applied to both groups.

1. No spoken English (at home or school)

The research project only had the capacity to investigate speech and language in English as translators were not available and assessments were available and carried out in English.

2. Evidence of severe/profound current hearing loss

Hearing loss has a significant effect on speech and language development. It can result in delay to receptive and expressive language and acquisition of speech sounds (ASHA, 2015). Therefore, SLTs and parents/carers confirmed there was no known diagnosis of hearing loss which could be a confounding factor to results.

3. Major physical disability or structural abnormality of vocal tract

Speech motor control and production can be severely impacted if the vocal tract is impaired and would confound results of the study. Also, as I was assessing motor control using the MABC-2 (Henderson et al., 2007) this required some basic motor abilities to attempt the tasks.

3.2.5 Sample Size

Ten children with ASD were recruited and ten age and gender matched typically developing children were recruited as controls. Due to the novel and experimental nature of this project limited effect size information was available within the published literature. Therefore, estimates of appropriate sample size and availability of children with ASD for research were based on studies that recruited participants from similar areas (Table 4). This study was unable to meet the numbers similar to these studies as it was a pilot and had limited resources to spend on recruitment.

Table 4: *Participant Numbers and Recruitment Strategies of Local Research in ASD*

Reference	Participant Numbers (Diagnosis included)	Age range (years, months)	Where recruited from
Peppé, McCann, Gibbon, O'Hare & Rutherford (2007) *Receptive and expressive prosodic ability in children with High Functioning ASD*	31 children with high functioning autism	6;1-13;6	Edinburgh area of Scotland
	72 typically developing children	4;10-11;8	Edinburgh state primary schools
Fukumura et al. (2016) *Development of audience design in children with and without Autism spectrum disorder.*	20 children with ASD	6;0-10;0	6 mainstream schools in North Lanarkshire and South Lanarkshire (both groups)
	20 typically developing children	6;0-10;0	
Alcorn et al. (2011) *Social communication between virtual characters and children with ASD*	32 children with ASD	5;0-14;0	Specialised school for children with ASD
	4 typically developing children	4;0-7;0	N?A
Cleland, Gibbon, Peppé, O'Hare & Rutherford (2010) *Phonetic and phonological errors in children with high*	30 children with high functioning autism	5;0-13;0	Registered on special needs services database. Live in Scotland (both groups)
	39 children with Asperger's syndrome	5;0-13;0	

functioning ASD and
Asperger Syndrome

Due to the number of independent sample and paired samples t-tests carried out and the small sample size, there was often not enough power present in tests to confirm statistical significance. This study is a pilot study and the data collected will be used to inform future studies. A statistical power analysis was performed for sample size estimation for future studies based on the data from this study (N=20). The effect sizes from each t-test were used along with an alpha of 0.5 and power of 0.80 using the G Power software package (Faul et al., 2009). A priori power analysis was carried out and tells us what sample size is needed to detect some level of effect with inferential statistics, in this case p-values. Appendix 6 provides this information for each t-test carried out in this study and these measures can inform future research.

3.2.6 Details of Study Participants

Table 5 gives information about the participants; speaker code, group, sex, and age (years; months).

Table 5: *Participant Information (Age in Years and Months)*

Participant	Group	Sex	Age
ASD1M	ASD	M	12;8
ASD2F	ASD	F	10;07
ASD3M	ASD	M	6;04
ASD4M	ASD	M	9;00
ASD5M	ASD	M	12;06
ASD6M	ASD	M	10;09
ASD7F	ASD	F	10;11
ASD8M	ASD	M	10;05
ASD9M	ASD	M	7;06
ASD10F	ASD	F	8;10
		Mean	9;04
		SD	1;10

		Range	6;00-12;01
TD1F	TD	F	9;08
TD2M	TD	M	11;1
TD3F	TD	F	7;07
TD4M	TD	M	7;07
TD5F	TD	F	6;08
TD6F	TD	F	7;08
TD7M	TD	M	6;00
TD8M	TD	M	6;06
TD9M	TD	M	9;04
TD10M	TD	M	12;06
		Mean	8;06
		SD	2;02
		Range	6;00- 12;05

3.2.7 Study Setting

Data collection took place over two sessions arranged at the convenience of the participants. This was either carried out at the University of Strathclyde or at the school, depending on the parent's preference. A parent or carer was present for each session.

3.2.8 Missing Data

All the children in the TD group completed all the tests set out for this group. In the ASD group, there was one participant who did not complete the full battery of assessments. ASD9M did not complete the CELF language assessment (Semel et al., 2003) but carried out all other assessments required and speech tasks.

3.3 Materials and Procedures

All children participated in assessment of their cognitive skills, speech sound production, fine and gross motor skills, and language. There were two strands to

assessment: perceptual/behavioural assessments; and instrumental assessment. Perceptual/behavioural assessments featured standardised assessments and those commonly used by SLTs or occupational therapists in clinics. The instrumental assessments were carried out using ultrasound tongue imaging (ultrasound) of the tongue during a speech repetition task which allowed quantitative analysis of the tongue during speech. Using both strands of assessment allowed comparison of how effective perceptual/behavioural and instrumental assessment are at identifying speech sounds errors (SSEs) in children with ASD.

3.3. Overview of Assessments

The behavioural/perceptual and instrumental assessments covered the following domains in order to gain a full profile of children's abilities: social communication; non-verbal IQ; language, movement, and speech sound production at both single syllable and multisyllabic levels. This range of assessments enabled me to determine whether there were underlying factors that may impede speech sound production beyond motor control. Below is a table listing the assessments carried out including time taken, procedure and analysis of results. Assessments were chosen to reduce cognitive load and time required whilst still gaining useful and comparable data.

Table 6: *Assessment Protocol for Study*

Domain	Assessment	Time (mins)	Procedure	Analysis	Group
Social communication	Social Communication Questionnaire (SCQ ; Rutter et al., 2003)	5	Parent questionnaire with 40 yes-or-no items. Current and Lifetime Forms.	Yields a total score that is interpreted with reference to cut-off scores. Scores above the cut-off of 15 suggest the individual is likely to have ASD and a more extended	ASD and TD

				evaluation should be undertaken.	
Non-verbal IQ	Leiter International Performance Scale (Leiter ; Roid et al., 2013)	25	4 subtests that yields a single ability score	Standard score calculated from manual instructions	ASD only
Language	Clinical Evaluation of Language Fundamentals 4 (CELF-4; Semel et al., 2003)	30	Subtests yields a core language score	Core language score calculated from manual instructions	ASD only
Movement	Movement Assessment Battery for Children (MABC-2; Henderson et al., 2007)	20	Subtests yields a composite score	Standard score calculated from manual instructions	ASD only
Speech Sound Production	Diadochokinesis (DDK) task. Using ultrasound tongue imaging*	5	Measures how accurately an individual can produce a series of rapid sounds. Five repetitions of single syllables (pa, ta, ka) and sequences (pataka) to be recorded at six set rates.	Rate: Average time to produce five repetitions. Accuracy: Transcribe the first syllable/sequence produced by participants. Scored 1 for correct production or 0 for incorrect. Consistency: Scored using consistency strength rating	ASD and TD

		Protocol developed by (McCann & Wrench, 2007)	designed by Williams and Stackhouse (2009)	
Clinical Useful Words Using (CUW; James, 2009) ultrasound tongue imaging*,	5	Single word picture naming task consisting of 30 words.	Measure the participants' % accuracy of words, consonants (PCC), vowels (PVC) and phonemes (PPC).	ASD and TD
Diagnostic Evaluation of Articulation and Phonology (DEAP; Dodd, 2002) Using ultrasound tongue imaging*	10	Picture naming task	Errors are classified as "typical" or "atypical" Errors are put into categories of specific speech processes e.g., fronting Calculate number of times a process occurs in each participant's speech Calculate number of children displaying a process three or more times.	ASD and TD

3.3.2 Behavioural Assessments

Below is a description of the behavioural assessments that were carried out with the ASD children. The only assessment that was carried out with TD children was the Social Communication Questionnaire (SCQ; Rutter et al. 2003) as the other behavioural assessments had norms created from large samples of typically developing children, therefore the TD group were likely to achieve ceiling effects in these assessments or score within the normal range so it was not necessary to carry out the assessment with them, having confirmed with their teachers and parents they were developing as expecting in cognition, language and movement.

3.3.2.1 Social Communication

The SCQ (Rutter et al. 2003) is a brief validated questionnaire involving parental report of ASD characteristic behaviours. It is composed of two forms: lifetime and current. Each contains forty yes or no questions. The “Lifetime” form focuses on the child’s overall development and “current” examines behaviour in the most recent three months. Parents/carers were asked to complete the questionnaire before the first session or during first session. It was used to screen participants in both groups. It has been found in the general population, four to five percent of children meet cut-offs for ASD traits (Chandler et al., 2007). The SCQ has shown strong discrimination between ASD and non-ASD traits (sensitivity 0.88, specificity 0.72) and cases of ASD and non-ASD (sensitivity 0.90, specificity 0.86). The findings are not affected by child IQ or parental education, affirming SCQ as a reliable indicator of ASD characteristics. The SCQ is suitable for screening and monitoring but not diagnosis (Rutter et al., 2003). This is because it does not examine symptomology in different contexts or include clinical observations. Children who met cut-off scores for ASD in control groups were planned to be excluded from the study, but this did not occur.

Scoring: Each of the forty items are rated as either “present” or “not present” and scored using the manual. Individuals with a score of >15 are considered to be at an increased risk of ASD (Berument et al., 1999; Rutter et al., 2003).

3.3.2.2 Non-Verbal IQ

A measure of non-verbal ability that took into account that there is a wide variation in non-verbal ability found in children with ASD was required. If not, the assessment may impede generalisation of results (Bishop et al., 2015). Overall distribution of IQ score in ASD is skewed, with varied rates of comorbid intellectual disability identified in epidemiological studies, ranging from 13-65% (Dykens & Lense, 2011). Additionally, motor delay has been found to be associated with intellectual ability, which may influence results of the movement measure if not taken into account. For instance (Smits-Engelsman & Hill, 2012) found in 460 children that lower measured IQ was associated with poorer motor performance. Additionally, the Leiter allowed me to examine IQ without language abilities significantly affecting the score. This is vital as language ability often influences results of psychological testing (Oller et al., 2000). When non-verbal IQ has been compared to language scores results, the associations have been found to be limited (Dethorne & Watkins, 2006). As I was hoping to understand correlations between these domains, a non-verbal assessment of IQ was vital.

The Leiter International Performance Scale, Third Edition (Roid et al., 2013) was used to measure non-verbal ability of participants in all groups. The test consists of four subtests to complete the cognitive core battery, these tests are: figure ground, form completion, classification and analogies, and sequential order. Testing took place on a one-to-one basis in a quiet room and was carried out according to the manual's instructions. The Leiter provides individual subtests, and numerous composite scores, that measure intelligence and discrete ability areas. These scores identify strengths and weaknesses in individual skills, as well as skill sets. Percentile and age-equivalent scores were provided.

3.3.2.3 Language

There are significant issues in the assessment of children with ASD and their language abilities due to various factors. Firstly, children with ASD have been found to struggle with motivational and attentional difficulties (Kjelgaard & Tager-Flusberg, 2001). Therefore, it is vital that language assessments are not too long and allow

time for breaks. In addition, spontaneous speech samples and standardised assessments may produce different results (Tager-Flusberg et al., 2009). I carried this out using the Clinical Evaluation of Language Fundamentals, 4th edition, (CELF; Semel, Wiig, & Secord, 2003) which assessed children's receptive language and expressive language in different contexts e.g., comprehension of words, vocabulary, sentence production etc. The CELF-4 (Semel et al., 2003) is a comprehensive tool which screens for language impairment. A first level of testing produces the "Core Language Score" which records the nature of language disorder, behaviours associated with it and effect on classroom functioning. This test took approximately 30-45 minutes and provided detailed subtest results on both receptive and expressive language for comparison to other behavioural domains.

The core language index consists of five subtests:

- Concepts and following directions. A measure of auditory comprehension and recall of utterances at increasing length and complexity.
- Formulated sentences. The child generates a sentence from a given work in reference to a picture.
- Recalling sentences. An imitation task in which sentences get longer and more complex.
- Word category. Assessment of the comprehension of relationships between words.
- Word definitions. Measures word meaning.

Children's standard scores indicate whether there is a presence of language difficulties. This information was used in the analysis to determine whether it is a confounding variable in relation to speech and motor ability.

3.3.2.4 Movement

The Movement Assessment Battery for Children, 2nd edition, (MABC-2; Henderson, Sugden, Barnett, & Harcourt Assessment., 2007) is a standardised assessment of motor skills. It comprises of eight tasks grouped into three subtests: 1) Manual dexterity, 2) Catching and Releasing and 3) Balance. Manual dexterity measures fine

motor control using tasks such as posting coins, threading beads and drawing. The other subtests involve both speeded and non-speeded tasks.

Scoring: Percentile ranks, and age-adjusted standard scores are provided for subtest scores and total impairment scores. This allowed observation of which element of motor impairment may be occurring to compare to other behavioural domains such as speech and cognition.

The MABC-2 (Henderson et al., 2007) provides a standardised measure of both fine and gross motor control. Compared to other available standardised motor assessment, the MABC-2 consists of a combination of fine motor and gross motor tasks that allow separate analyses to determine if a specific aspect of motor control may be impaired. Furthermore, the MABC-2 provides assessment in three different age bands ranging from 3-16. This ensures that motor skills being assessed are age appropriate to the participant (Brown & Lalor, 2009). Additionally, as the test focuses only on motor acts and does not include verbal instructions, the assessor can employ it in a way that ensures the assessment is suitable to be used with children with communication, intellectual, attentional and/or behavioural difficulties (Green et al., 2002). This MABC-2 (Henderson et al., 2007) also allows direct comparison of fine motor skills from the MABC-2 and the speech motor task, diadochokinesis (DDK) to determine whether there is a correlation between speech motor control and general motor control in the group of children with ASD. This will help us determine whether there are specific issues with motor control such as timing, speed or coordination that correlate with speech motor control difficulties.

3.3.3 Assessment of Speech

Speech was assessed at both the simple syllable and multisyllabic level of speech using two clinical phonology assessments often used by SLTs. This was to provide data on how children performed at differing levels of motorial complexity of speech.

3.3.3.1 Assessment of Simple Syllabic Structure

The Diagnostic Evaluation of Articulation and Phonology (DEAP; Dodd, 2002) was used to screen the participant's articulation and phonology in both groups. This

involved ten coloured pictures that the child was asked to repeat twice. The pictures contain words of two to four syllables and are used to test consonants in different positions to identify if any speech errors or phonological processes are present. To elicit the words semantic or forced choice cues were used as instructed from the manual. Each consonant sound produced was scored 1 if correct and 0 is incorrect. When the participant produced the word twice, the best attempt was scored only in this instance. Scores were then converted into percentage consonants correct (PCC) and errors patterns were also identified and noted. Audio recordings were made, and fine phonetic transcription was carried out for in-depth error analysis using Articulate Assistant Advanced (AAA) software (Articulate Instruments Ltd, 2019).

Assessment of Word Consistency:
Both groups were also assessed on consistency of word production using the inconsistency scoring of the screening assessment of the DEAP (Dodd, 2002). Participants were required to repeat the words twice when carrying out the DEAP screening assessment (Dodd, 2002). An inconsistency score was produced following instructions from the DEAP manual in which a score was tallied from the number of inconsistent words produced. The number of words produced differently was counted and divided by the number of words produced twice, the score produced was then multiplied by 100.

3.3.3.2 Assessment of Multisyllabic Speech

Measurement of multisyllabic speech was carried out using the Clinically Useful Words Assessment (CUW; James 2009). It is a short assessment consisting of ten multisyllabic words. The accuracy of this task was measured by calculating PCC (percentage consonants correct) of the best of two attempts of the target produced. Audio recordings were made, and fine phonetic transcription were carried out for in-depth error analysis using audio recordings (Articulate Instruments Ltd, 2019). Increasing complexity of an articulatory gesture increases the processing demands on motor performance of children (Maner et al., 2000). The more complex the articulatory gesture, the further heightened the requirement there is to select and sequence phonemes under high motor demands (Lewis & Freebairn, 1997). Populations that present with higher rates of motor impairment, such as in ASD, may

have increased variability and error in production of complex articulatory gestures due to increased motor demands (Maner et al., 2000; Sadagopan & Smith, 2008).

3.4 Analysis of Behavioural Assessments

Due to the nature of this study, a pilot study with a small sample size, there was not enough power to carry out a multiple regression analysis. This analysis produces a correlation coefficient to determine the strength of the estimate regression, R^2. Furthermore, the data was a mix of ordinal (MABC-2, Leiter, CELF) and interval (DEAP and POP) which is difficult to find correlations between. As a result, I chose to use scatterplots to make qualitative observations of the relationships between the ordinal data. In-depth analysis of the articulatory movements of the children is provided in chapter 4.

3.5 Analysis of Speech

A perceptual analysis of simple and multisyllabic speech as well as DDK for assessment of speech motor control was carried out. The phonological assessment was carried out using the Diagnostic Evaluation of Articulation and Phonology (DEAP; Dodd, 2002). The multisyllabic assessment was carried out using the clinically useful words assessment (CUW; James, 2009). Both of these assessments were assessed perceptually, following the same protocol as observed in clinics. The speech motor assessment was carried out using the Diadochokinesis task (DDK) and this was assessed both perceptually and instrumentally using ultrasound tongue analysis.

Analysis of the DEAP and CUW was carried out live with the child by the researcher and then verified using a recoding and spectrogram analysis from the Advanced Articulate Assistant Software (Articulate Instruments Ltd, 2019). These were phonetically transcribed using the IPA symbol chart (International Phonetic Association, 2015). From the transcriptions a score of 1 was given if a correct production of each consonant was made and 0 if incorrect. This allowed calculation of the percentage of consonants correct (PCC), an inconsistency score and classification of speech processes to be carried out for comparison across groups which is further described below.

3.5.1. Percentage of Consonants Correct (PCC)

The PCC is a metric which expresses the percentage of consonants produced in a speech sample that were accurate. Since the initial development of the PCC, there have been variations on how this measure should be used and within what speech context (Shriberg *et al.*, 1997). There have been concerns raised of using the PCC metric within conversational speech as while it would be representative of the individual, it does not guarantee validity when comparing within or across different groups (Shriberg *et al.*, 1997). In the case of this study, it had set word lists for both the DEAP (Dodd, 2002) and the CUW (James, 2009), it was considered that the use of the PCC was appropriate and followed the recommendations for calculated PCC from the DEAP (Dodd, 2002). Calculating PCC allows understanding of the severity of a disorder where > 90% = mild, 65%-85% = mild-moderate, 50%-65% = moderate-severe, and < 50% = severe (Shriberg *et al.*, 1997).

3.5.2 Inconsistency Score

The inconsistency score was calculated to determine whether children had an inconsistent speech disorder, where words are produced differently during multiple repetitions. In this study the target words in the DEAP (Dodd, 2002) and CUW (James, 2009) were produced twice and given a score of 1 if the words were produced differently within the two repetitions. If 40% or more of the words are produced differently, it suggests the child may have an inconsistent speech disorder according to Dodd's classification (Dodd, 2014). This score was used to examine whether there was a link between inconsistent speech and general motor abilities as children with inconsistent speech have been shown to perform more poorly than all the other groups on a more complex, timed motor-planning task (Bradford and Dodd, 1994).

3.5.3 Speech Processes

A phonetic and phonological pattern analysis was carried out in which SSEs identified were given speech process labels e.g., fronting (Dodd *et al.*, 2004). Speech processes were classified according to patterns described by McLeod and

Baker (2017). The number of times a speech process occurred in each participant's speech was calculated. This shows whether errors are occasional or prevalent and whether any errors are specific to ASD in this study. This allowed errors to be classified as "typical," "delayed" or "atypical." Typical SSEs were classified as SSEs that would be expected within typical development. According to Baker and McLeod (2017), a child with speech delay may exhibit systematic SSEs, such as cluster reduction or final consonant deletion, which would be typical for younger children but should be resolved so would be classified as delay due to their age. Whereas a child with atypical SSEs would exhibit patterns such as glottal insertion or initial consonant deletion that are not expected in any stage of speech development.

3.5.4 Statistical Analysis

An independent samples t-test was carried out to compare the three measures: PCC, inconsistency score and speech processes between groups. The speech processes were additionally analysed by comparing them across the two groups within the following classifications: total errors, typical errors, delayed errors, and atypical errors. A Leven's test of equality of variance was carried out to ensure that that the data did not violate the rules to then carry out an independent samples t-test. This allowed us to calculate if there was a significance difference between the ASD and TD group within PCC, inconsistency score and the speech processes.

Since the small sample size made null results more likely Cohen's effect size were also calculated. Effect size is a quantitative measure that gives understanding of the magnitude of the effect of the significance and lies between 0 and 1 (both values inclusive). The larger the number produced by Cohen's d, the stronger the relationship is between the variables. This was used in conjunction with the independent samples t-test. I followed the guidance that Cohen's effect size of d=0.2 is a small effect size, 0.5 is a medium effect size and 0.8 is a strong effect size.

To correct for multiple comparisons with the t-tests, a Bayes factor analysis was carried out using JASP (Team, 2018). Bayes factors (BF_{10}) range from 0 to infinity with a Bayes factor of 1 indicating that both the null and alternative hypothesis predicted the data equally well (van Doorn et al., 2021). Larger values of BF_{10}

indicate more support for the alternative hypothesis (H1). For example, a BF_{10} of 5 means that the data are five times more likely under H1 (alternative hypothesis) and H0 (null hypothesis). This study used guidelines set out by van Doorn et al. (2021) for interpreting the Bayes factor in which a Bayes factor of 1-2 is considered to be weak, 3-10 is moderate and 10> is strong in favour of H1. A Bayes factor of < 1/3 supports the H0.

As there has been multiple tasks carried out between the two groups, this increases chances of getting at least one significant difference by chance. There is debate on the necessity and extent required for adjustment for multiple comparisons (Perneger, 1999). The Bonferroni correction was chosen in the case of this study to control the family-wise error rate (Lee & Lee, 2018). The Bonferroni correction assume that all the hypothesis tests are statically independent, it has been critiqued that it is too conservative, since when the number of comparisons increases, the level of significance becomes very small and the power of the system decreased (Lee & Lee, 2018). The tests carried out in this study have some aspects in common (e.g., production of speech sounds) then there would be some dependence, however this cannot eb confirmed. The Bonferroni correction has been used though the probability of making at least one type I error is less than the Bonferroni assumes and may have led to over correction.

Lee and Lee (2018) recommend the following procedure:
“With an increased in the number of hypothesis tested, type I errors increases. Therefore, the significance level is divided into numbers of hypothesises tests. In this manner the type I error can be lowered.

Adjusted alpha (α) = α/k (number of hypothesis tested)
e.g., 50 t-tests, one would set each t-test to 0.05/50=0.001.”

3.6 Behavioural Assessment Results

This section discusses the results for both the ASD and TD groups in three parts: speech assessments and behavioural assessments. These measures were carried out on ten children with ASD (ASD group) and ten typically developing children (TD

group). The Leiter International Performance Scale (Leiter; Roid, Miller, Pomplun, & Koch, 2013) the Clinical Evaluation of Language Fundamentals, Fourth Edition (CELF; Semel, Wiig, & Secord, 2003) and the Movement Assessment Battery for Children, Second Edition (MABC-2; Henderson, Sugden, Barnett, et al., 2007) assessments were carried out on the ASD group only as these assessments have been standardised from large numbers of TD children by the authors of the assessments. In contrast, the other measures described below have not been standardised on large numbers of TD children for this study's age range (6-12 years) so have been carried out with a control group of TD children.

3.7 Behavioural Assessment Group Results

This section covers the results from the behavioural assessment in autistic symptomatology, non-verbal IQ, language, and movement and how these correlate with speech sound production at the single word and multisyllabic levels. Table 7 and 8 are summaries of all the behavioural assessment result for both groups.

Table 7: *Behavioural Assessment Group Results of ASD Group*

Participant	Sex	Years and Months	Leiter	CELF	MABC-2	DEAP PCC	CUW PCC	SCQ Current	SCQ Lifetime
ASD1M	M	12.08		91	4.00	97.37	97.83	20	35
ASD2F	F	10.07	78	79	6.00	97.2	98	9	24
ASD3M	M	6.04	77	56	4.00	86.1	92.3	17	22
ASD4M	M	9			1.00			17	
ASD5M	M	12.06	99	100	7.00	94.74	100	12	12
ASD6M	M	10.09	96	109	5.00	100	98.08	23	25
ASD7F	F	10.11	78	42	6.00	75	58.82	13	19
ASD8M	M	10.05	99	98	8.00	97.37	98.08	8	19
ASD9M	M	7.06	101		4.00	94.44	98.08	9	24
ASD10F	F	8.1	75	52	2.00	86.47	94.23	13	21
MEAN		9.46	84.67	78.38	4.70	93.56	96.04	14.10	22.33
SD		1.96	12.42	25.27	2.06	6.46	3.24	5.02	6.16

Note. For behavioural assessments (Leiter, CELF, MABC-2, DEAP PCC) where normative data existed, orange indicates a below the mean and red indicates significantly below the mean, indicating an impairment. In the SCQ Current and Lifetime, orange indicates a score above 15, the threshold for social communicative profile typical of ASD. No normative data was available for the CUW.

Table 8: *Behavioural Assessment Group Results of TD Group*

Participant	Sex	Years and Months	DEAP PCC	CUW PCC	SCQ Current
ASD1F	F	9.08	100	94.1	1
ASD2M	M	11.1	97.2	100	5
ASD3F	F	7.07	100	98	8
ASD4M	M	7.07	94.6	98.1	7
ASD5F	F	6.08	94	98.1	2
ASD6F	F	7.08	100	100	2
ASD7M	M	6	88.89	86.36	
ASD8M	M	6.06	97.4	100	1
ASD9M	M	9.04	100	100	12
ASD10M	M	12.06	97.3	98.1	5
Mean		8.06	96.94	97.28	4.78
SD		2.17	3.60	4.24	3.73

Note. For behavioural assessments (DEAP PCC) where normative data existed, no children were below the expected score for their age. In the SCQ Current orange indicates a score above 15, the threshold for social communicative profile typical of ASD which did not occur in this group. No normative data was available for the CUW.

3.8 Speech Assessment Results

3.8.1 Single Word Level Analysis

The Diagnostic Evaluation of Articulation and Phonology (Dodd, 2002) was used to screen articulation and phonology in both the ASD group and TD group.

- Percentage of Consonants Correct (PCC)

Analysis of speech was carried out at single word level using the Diagnostic Evaluation of Articulation and Phonology (DEAP; Dodd, 2002), this involved fine phonetic transcription of ten words that allowed analysis of consonants of participants in both groups (ASD and TD). The PCC metric analyses the intended consonants sounds in a sample of words to determine whether they were articulated correctly (Shriberg, Austin, Lewis, McSweeny, & Wilson, 1997).

An independent samples t-test was carried out to determine whether there was a statistically significant difference between the means in PCC produced by children in the ASD group and TD control group.

Results: There was no significant difference in the PCC scores between the ASD group (M=92.08, SD= 8.04) and TD group (M=96.94, SD=3.60); t(10.84) = -1.67 p=0.12, (Bonferroni = 0.02) $BF_{10}= 1.10$. Cohen's effect size value (d=-0.80) showed a large mean difference between the two groups. As the means of the two groups are different, it is likely if the sample size was larger than a significant difference may have been found when looking at previous research (Cleland, 2010; Shriberg *et al.*, 2011; Wolk and Brennan, 2013). The Bayes factor indicates weak evidence of supporting either the null or alternative hypothesis. Additionally, the DEAP screening assessment is limited due to a small sample of words. So, this result did not provide evidence that there are more SSEs produced in children with ASD.

3.8.1.1 Speech Errors Patterns

Table 9 lays out the mean and SD for the ASD group and TD group for the total number of errors and the subsets with the errors defined as age appropriate, delayed, and unusual.

Table 9: *Mean and SD of speech errors produced by ASD group and TD group*

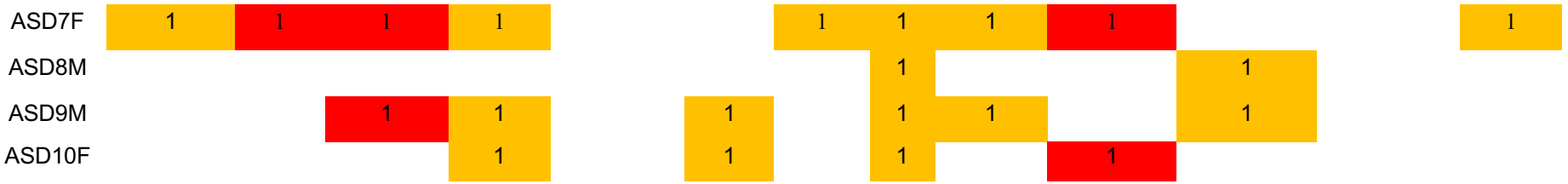
Speech Errors	Group	Mean	Std. D
Total Errors	TD	1.11	1.36
	ASD	2.9	2.23
Age-Appropriate Errors	TD	0.11	0.33
	ASD	0	0

Delayed Errors	TD	0.78	0.83
	ASD	2.2	1.55
Unusual Errors	TD	0.22	0.44
	ASD	0.7	0.82

Table 10 displays the different types of speech processes produced by participants in both groups and how many times these occurred in the DEAP (Dodd, 2002) assessment. This table shows a trend of more errors, in particular delayed errors occurring the ASD group in comparison to the TD group. Green represents age-appropriate errors, orange represents delayed errors and red represents unusual errors as defined by McLeod and Baker (2017) and Dodd (2002).

Table 10: *Speech processes produced by participants in TD and ASD group as defined by McLeod and Baker (2017)*

Participant	Velar Fronting	Backing to Velar	Backing to Pharyngeal glottal	Stopping	Deaffrication	Voicing Errors	Weak Syllable Deletion	FCD	Cluster Reduction	Assimilation	Fricative Simplification	Reduplication	Stopping of Liquids
TD1F													
TD2M											1		
TD3F													
TD5F								1			1		
TD6F						1						1	
TD7M													
TD8M	1						1	1			1		
TD9M													
TD10M					1								
ASD1M	1						1						
ASD2F	1												
ASD3M				1		1				1			1
ASD4M				1		1				1			
ASD5M													
ASD6M	1	1		1		1			1	1			


ASD7F
ASD8M
ASD9M
ASD10F

90% of the ASD group and 66% of the TD presented with at least one SSE. An independent samples t-test was carried out for total errors, age-appropriate errors, delayed errors and unusual errors to determine if there was a significant difference between the ASD and TD groups. There was a significance difference in the number of total errors produced by the ASD group (M=2.90, SD= 2.23) and the TD group (m=1.11, SD= 1.36) conditions; t(17)=-2.076, p=0.04* (Bonferroni=0.02), BF_{10}= 1.9. Cohen's effect size value (d=1.01) showed a large mean difference between the two groups. However, the Bayes factor indicates weak evidence of supporting either the null or alternative hypothesis. The t-test also did not survive the Bonferroni correction.

Age-appropriate errors

None of the ASD group and 11% of the TD (one age-appropriate SSE) had age-appropriate SSEs. There was no statistical significance in the number of age-appropriate errors between the ASD group (M=-0.00, SD=0.00) and the TD group (M=0.11, SD=0.33) conditions; t(8)=1.00, p=0.35 (Bonferroni=0.02), BF_{10}= NaN. There was only one instance of an age-appropriate error recorded in either group (the TD group) therefore the null hypothesis is accepted.

Delayed errors

90% of the ASD group and 55% of the TD presented with at least one delayed SSE. For example, 10F presented with stopping, voicing errors and final consonant deletion, all of which would have been expected to have been eliminated by her age of eight years. Only 08M in the ASD group had no delayed speech errors. There was a significant difference in the number of delayed errors for the ASD group (M=2.20, SD=1.55) and the TD group (M=-0.78, SD=0.83) conditions; t(17)=-2.45, p=0.042* (Bonferroni=0.02), BF_{10}=1.93. Further Cohen's effect size value (d=-1.01) showed a large mean difference between the two groups. The Bayes factor indicates weak evidence of supporting either the null or alternative hypothesis. If there was a strong Bayes factor, then the null hypothesis could have been rejected and found that children with ASD produced significantly more errors classified as speech delay than TD children. This would have added evidence to the hypothesis that children with ASD present with more overt speech sound errors that typically developing children, in this case, specifically delayed speech errors. This should be tested with a larger

sample size with greater power in the sample size. However, the t-test did not survive the Bonferroni correction.

Unusual errors

50% of the ASD group and 11% of the TD presented with at least one atypical SSE. For instance, both 07M and 10F presented with backing errors, which is not expected at any stage during speech development. There was no significant difference for the number of unusual errors produced by the ASD group (M=0.70, SD= 0.82) and the TD group (M=0.22, SD=0.44) conditions; t(14.04)=-1.60, p=0.06 (Bonferroni=0.02), $BF_{10}=1.49$. Cohen's effect size value (d=0.92) showed a large mean difference between the two groups. The Bayes factor indicates weak evidence of supporting either the null or alternative hypothesis. Therefore, the null hypothesis is accepted. Contrary to the hypothesis, there were not significantly more “unusual” SSEs in the speech of children with ASD. There is individual variability within the ASD group, for example 7F and 3M both have two SSEs of this type. Perhaps with a larger sample size, more SSEs of this type would have led to a significant result.

3.8.1.2 Inconsistency in speech production

An inconsistency score was taken from the DEAP assessment (Dodd, 2002) in which the child had to repeat the same word twice and was scored on whether the production was consistent both times. This project followed the guidelines set out by Dodd (2002) in which an inconsistency score higher than 40% was an inconsistent speech profile. An independent samples t-test was carried out to analyse if there was a difference between groups.

There was no significant difference between the ASD group (M=7.53, SD= 10.56) and the TD group (M=9.70, SD= 9.97) conditions; t(16) = -0.44, p=0.66, (Bonferroni=0.02), $BF_{10}=0.44$. Cohen's effect size value (d=-0.21) showed a small mean difference between the two groups. The Bayes factor indicates a move towards support of the null hypothesis. If it had met the criteria of 0.33 or lower, I would have accepted the null hypothesis that children with ASD did not present with more inconsistency in their speech than the TD children. Not all children participated in the part of the assessment which may have impacted the outcomes of the part of the assessment due to a low sample size.

3.8.2 Multisyllabic Word Level Analysis

The Clinically Useful Words Assessment (James, 2009) was used to assess ten structurally complex words containing three or more syllables. 10/10 of the TD group participated and 9/10 of the ASD group participated.

3.8.2.1 Percentage of Consonants Correct

The PCC metric was used to assess how many consonant sounds were articulated according to the adult model and given a percentage score to indicate how many were accurate (Shriberg *et al.*, 1997); An independent samples t-test was carried out to determine whether there was a statistically significant difference between the means in the ASD group and TD control group.

There was no significance difference between the PCC scores for the ASD group (M=92.82, SD= 12.97) and TD group (M=97.2, SD=4.24) conditions; t(17) = -1.03, p=0.32 (Bonferroni=0.02), BF_{10}=0.58. Cohen's effect size value (d=-0.47) showed a small mean difference between the two groups. The Bayes factor indicates weak evidence of supporting either the null or alternative hypothesis but is moving towards the null hypothesis. This in contrary to the hypothesis that children with present with more overt speech sound errors than typically developing children, in their production of multisyllabic words.

3.8.2.2 Speech Error Patterns

Speech errors were categorized into either syllable structural processes, substitution (systemic) processes and assimilation processes as defined by McLeod and Baker (2017).

Table 11 lays out the mean and standard deviation for the ASD group and TD group for the total number of errors and the subsets, with the errors defined as age appropriate, delayed, and unusual. There were no age-appropriate errors produced by either group in this test. Table 12 displays the range of processes produced by all participants and were labelled as age appropriate, delayed, or unusual as set from the DEAP criteria (Dodd, 2002).

Table 11: *Mean and SD of speech errors produced by ASD group and TD group*

Speech Errors	Group	Mean	Std. D
Total Errors	TD	0.78	0.67
	ASD	4	6.18
Age-Appropriate Errors	TD	0	0
	ASD	0	0
Delayed Errors	TD	0.67	0.71
	ASD	1.56	2.55
Unusual Errors	TD	0.11	0.33
	ASD	2.33	3.74

Table 12 displays the different types of speech processes produced by participants in both groups and how many times these occurred in the DEAP (Dodd, 2002) assessment. This table shows a trend of more errors, in particular more errors occurring the ASD group, mainly one child who had a significant number of speech processes in comparison to the TD group.

Table 12: *Speech processes produced by participants in TD and ASD group as defined by McLeod and Baker (2017)*

Participant	Velar Fronting	Backing to Velar	Backing to Pharyngeal	Gliding	Deaffrication	Voicing Errors	FCD	MCD	ICD	Cluster Reduction	Assimilation	Epenthesis	Other
01F TD							1			1			
02M TD													
03F TD											1		
05F TD				1									
06F TD											1		
07M TD													
08M TD										1			
09M TD													
10M TD											1		
01M ASD											1		
02F ASD											1		
03M ASD		1						1		2			1
04M ASD											1		
05M ASD	1	2	1	1	1	2	1	3	1	3	3		1
06M ASD										1			

Note. Green represents age-appropriate errors, orange represents delayed errors and red represents unusual errors as defined by McLeod and Baker (2017) and Dodd (2002).

An independents sample t-test was carried out for total errors, age-appropriate errors, delayed errors and unusual errors to determine if there was a significant difference between the ASD and TD groups.

Total errors

There was no significance difference in the number of total errors produced by the ASD group (M=3.10, SD= 4.01) and the TD group (M=0.78, SD= 0.67) conditions; t(17)=-1.71, p=0.11 (Bonferroni=0.02), BF_{10}=1.07. Cohen's effect size value (d=0.78) suggests there is a medium mean difference between groups. The Bayes factor indicates weak evidence of supporting either the null or alternative hypothesis. If there had been power in this sample, the null hypothesis is accepted and there was no difference in multisyllabic word production between the ASD and TD groups. It is evident there is significant variability in the scores of individual children. For example, 05M has a significant number of delayed and disordered errors within the CUW assessment, while his DEAP score was within the norm with significantly fewer errors. This indicates that this child may present with a speech motor problem, where words that are motorically complex present as a challenge to this child.

Age-appropriate errors

There were no age-appropriate errors produced by any participants in either group, so analysis was not conducted at this level.

Delayed errors

There was not a significant difference for the number of delayed errors for the ASD group (M=1.30, SD=1.49) and the TD group (M=0.33, SD=0.71) conditions; t(17)=-1.77, p=0.09, (Bonferroni=0.02), BF_{10}=1.14. Further Cohen's effect size value (d=-0.81) showed a medium mean difference between the two groups. The Bayes factor indicates weak evidence of supporting either the null or alternative hypothesis. This is in contrast with the DEAP assessment where a significant difference was found. This suggests while children with ASD produce more speech errors than TD children when looking at a wide range of consonants, this significance is reduced when the words are increasingly motorically complex and TD children produce errors as well.

Unusual errors

There was a trend towards difference but did not reach statistical significance for the number of unusual errors produced by the ASD group (M=2.33, SD= 3.74) and the TD group (M=0.11, SD=0.33) conditions; t(17)=-1.52, p=0.14, (Bonferroni=0.02), BF_{10}=0.88. Further, Cohen's effect size value (d=-0.70) showed a large mean difference between the groups. So, while the null hypothesis would not be accepted due to lack of statistical significance, it is worth noting a trend towards difference that may be revealed further with a larger sample size.

3.8.2.3 Inconsistency in speech production

An inconsistency score was assessed applying the same criteria as the DEAP (Dodd, 2002) in which the child had to repeat the same word twice and was scored on whether the production was consistent both times. An independent samples t-test was conducted to analyse if there was a difference between groups.

There was no significant difference between the ASD group (M=13.46, SD= 19.34) and the TD group (M=20, SD= 23.45) conditions; t(16)=0.55, p=0.59, (Bonferroni=0.02), BF_{10}=0.46. Cohen's effect size value (d=-0.26) showed a small mean difference between the two groups. The Bayes factor indicates weak evidence of supporting either the null or alternative hypothesis, but a trend is moving towards the null hypothesis. So, while the null hypothesis would not be accepted due to lack of statistical significance, it is worth noting a trend towards difference that may be revealed further with a larger sample size. This is contrary to the hypothesis that children with ASD present with more overt speech sound errors than typically developing children, their speech appears to be just as consistent in the case of this assessment.

3.9 Behavioural Assessment Results

The following section covers the results to answer the research question:

1. Is there an interaction between SSEs, general motor abilities, language skills and non-verbal cognition in children with ASD?

Hypothesis: Increase in SSEs significantly correlates with decreased fine motor and language skills

Comparison of behavioural assessments was undertaken at group level therefore it was first important to determine that there were no significant differences between the groups in age or sex. Age in years and months was converted into decimals and the mean and standard deviation is for the ASD group. The sex of research participants was defined from parental report.

To ensure there were no significant differences between the groups in age and sex an independent-samples t-test was conducted to compare age in the ASD group (table 8) and TD group (table 7) in which equal variances were assumed. There was no significant difference between the ASD group (M= 9.93, SD= 1.93) and TD group (M= 8.81, SD= 2.18); t (18)= -1.22, p =0.24, BF_{10}=0.65. Cohen's effect size value (d=-0.55).

Within the ASD group there were three females and seven males and in the TD group there were four females and six males. A Pearson chi-square test was conducted to determine if there was a significant difference in the distribution of participants identified as male or female between groups. The relation between these variables was not significant, X2(1, N= 20) = 0.642, p<0.423.

3.9.1 Autistic Symptomatology

Autistic symptomatology was measured using the Social Communication Questionnaire (SCQ). This was conducted with both ASD group and the TD group. The TD group was included in this assessment to ensure there were no potential participants in this group with undiagnosed ASD. This assessment consists of two parent questionnaires, the “current” form, and the “lifetime” form. The current form assesses the social communication traits that the participant expresses currently.

Whereas the “lifetime” form assesses whether the participants have expressed social communication traits throughout their lifetime. A score of 15 or above on the lifetime form is an indicator of autistic social communication traits. The current form is used to assess how the communication traits may have changed or present more recently for the child.

Results:

In the ASD group 4/10 children met the cut-off criteria of fifteen or above for indicators of “autistic traits” in the “current form” and 8/9 children met the cut-off criteria of fifteen or above in the “lifetime form”. In the TD group no children met the cut-off criteria of fifteen or above for indicators of “autistic traits” in the “current form” as expected. This was conducted to ensure that the control group did not have children present who were undiagnosed with ASD, displaying social communication traits associated with ASD.

As all the children in the ASD group have a formal diagnosis of ASD, it was expected that all children in this group would reach a score of 15 and above in both forms. However, children in the ASD group were more likely to have autistic traits presented over their lifetime than currently. Autistic symptoms remain relatively stable over time if no intervention is put in place (Matson and Horovitz, 2010). However, in Scotland with a confirmed diagnosis of ASD, children are often referred to a speech and language therapist (SLT) where a course of intervention takes place. Baghdadli et al. (2012) found that early intervention significantly benefited communication outcomes while observing these in children diagnosed with ASD over a ten-year period. This may explain why there has been a change in symptomatology in for six participants.

05M did not reach the cut-off requirement for indicators of autistic social communication traits in either the lifetime or current forms. The SCQ has shown strong discrimination validity between children with ASD and not (sensitivity 0.88, specificity 0.72) (Chandler *et al.*, 2007). However, this is not enough to determine and ASD diagnosis, the gold standard for ASD diagnosis requires a multi-disciplinary team to assess behavioural, historical and parent reported indicators using a variety of tools (Falkmer *et al.*, 2013).

3.9.2 Gross and Fine Motor Abilities

The Movement Assessment Battery for Children 2nd edition (MABC-2; Brown & Lalor, 2009) was used to measure gross and fine motor abilities.

MABC-2 Total Score

The total score is the combined composite score of the three areas assessed in the MABC-: Manual dexterity, aiming and catching and balance. This score is used to indicate whether a movement disorder may be present. Above the 15th percentile (>67 is within normal range. Between the 5th and 15th percentile (57-67) is judged as "at risk" of having a movement and at or below the 5th percentile (<56) denotes a significant movement difficulty. Ten children in the ASD group participated in this assessment and the results are described below.

Results:

- 7/10 children in the ASD group scored at or below the 5th percentile, denoting a significant movement difficulty.
- 2/10 children in the ASD group score between the 5th and 15th percentile, judging them at risk of a significant movement difficulty.
- 1/10 children scored above the 15th percentile within the normal range.

Table 13: *Scaled Scores of results from MABC-2 in form of composite scores and test total scores.*

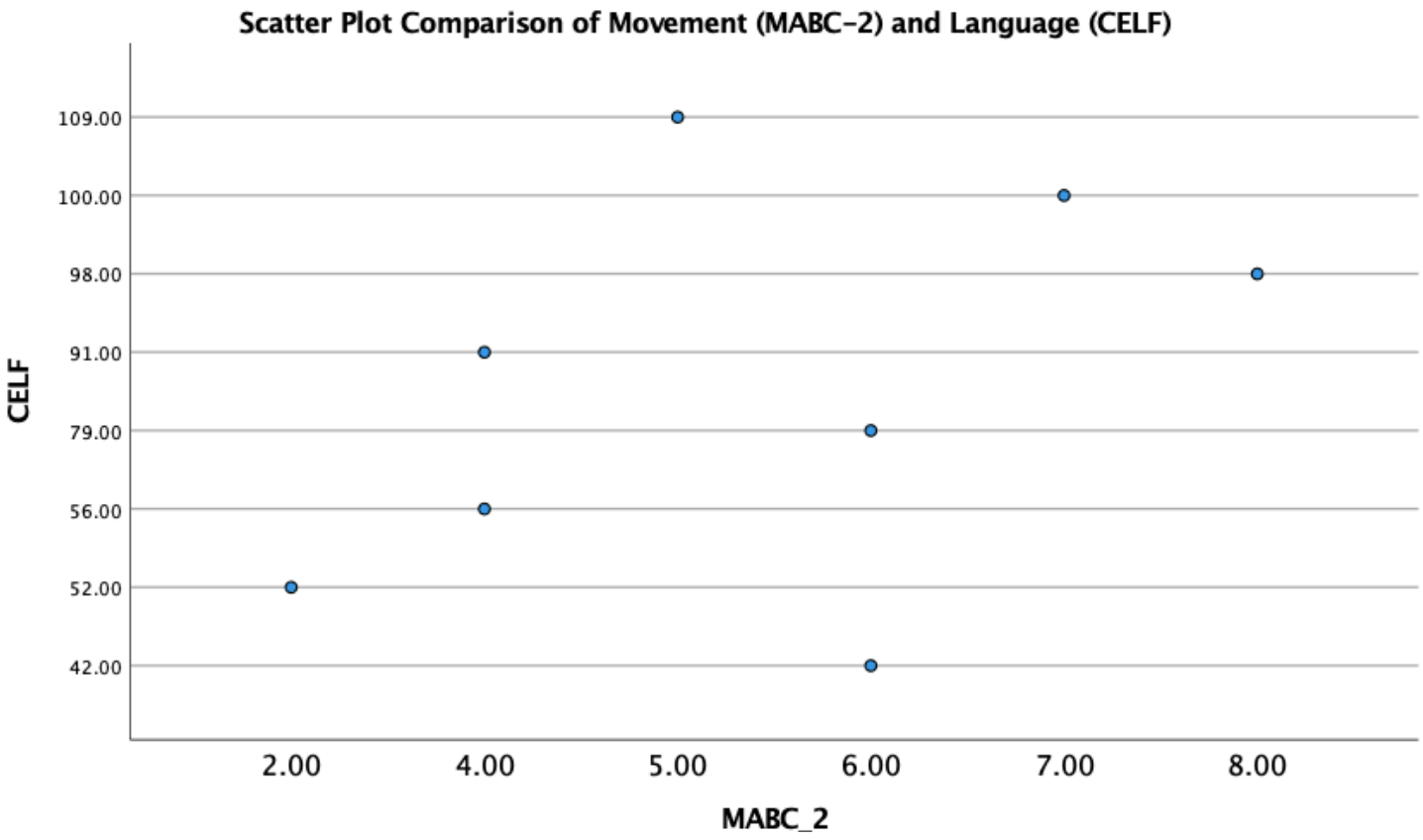
	Composite Scores			
Participant	Manual Dexterity	Aiming and catching	Balance	Test Total Score
ASD1M	3.00	4.00	7.00	4.00
ASD2F	11.00	5.00	6.00	6.00
ASD3M	2.00	10.00	6.00	4.00
ASD4M	2.00	9.00	1.00	1.00
ASD5M	6.00	7.00	9.00	7.00
ASD6M	5.00	3.00	9.00	5.00
ASD7M	9.00	6.00	6.00	6.00

ASD8M	12.00	2.00	8.00	8.00
ASD9M	6.00	5.00	5.00	4.00
ASD10F	3.00	3.00	2.00	2.00
Mean	5.90	5.40	5.90	4.70
SD	3.73	2.65	2.45	2.06

Note. Green is a score above the 15th percentile within the normal range. Orange is a score between the 5th and 15th percentile, judging them at risk of a significant movement difficulty. Red is a score at or below the 5th percentile, denoting a significant movement difficulty.

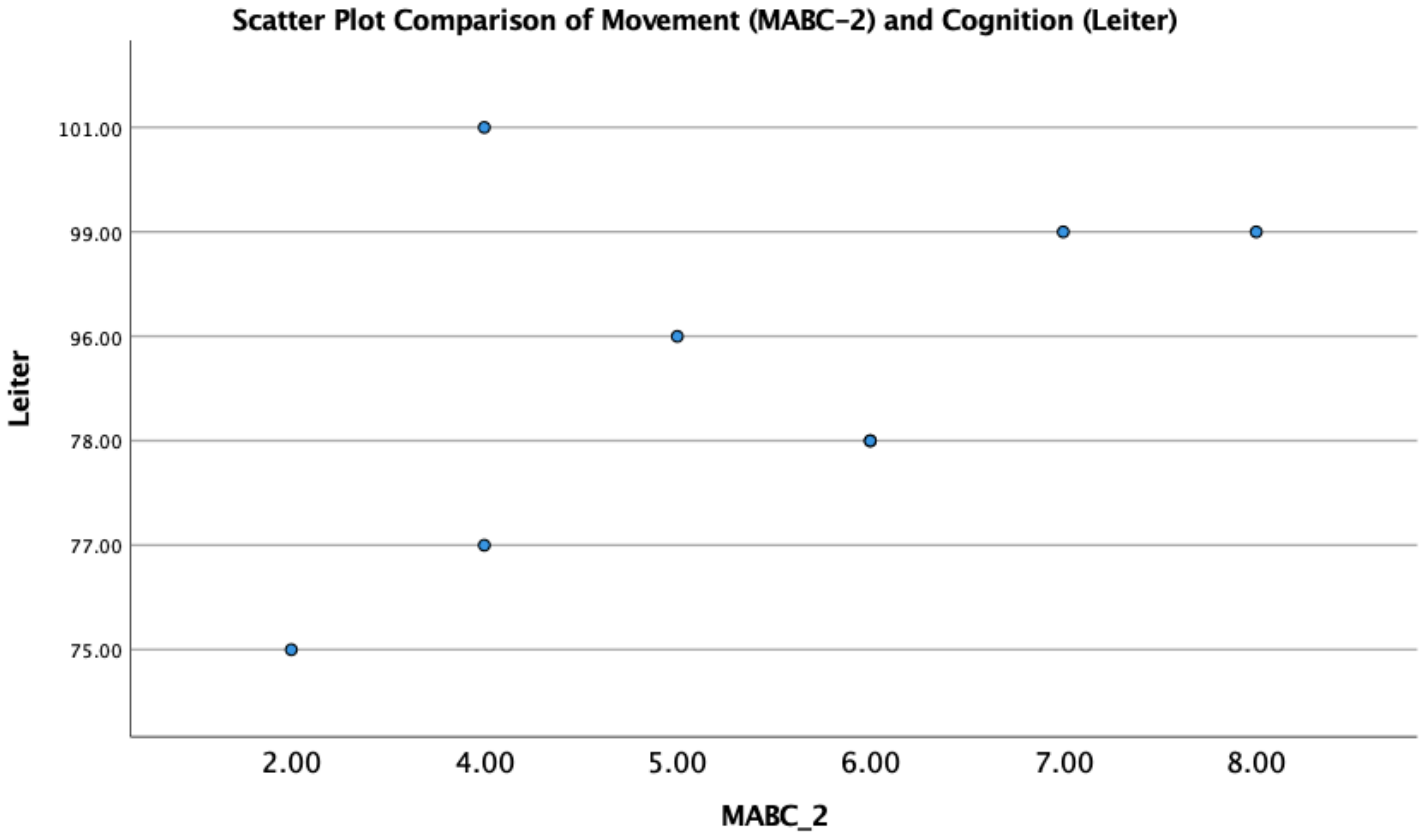
To observe potential relationships between the MABC-2 and other cognitive domains, scatterplots were created to make qualitative observations (Figure 6 and 7).

Figure 6: *Scatterplot of scores from MABC-2 (movement) and CELF (language)*



There was no obvious trend identified from this scatterplot, indicating there may not have been any correlation between the movement and language performances of children within the ASD group.

Figure 7: *Scatterplot of scores from MABC-2 (movement) and Leiter (non-verbal cognition)*



There is a trend in this scatterplot that the higher the score on the Leiter, the higher the score on the MABC-2. This suggests that there may be a relationship between movement abilities and non-verbal cognition. This would be worth exploring through a linear regression analysis with a significantly larger sample size in order to determine if a correlation exists between these two domains.

3.10 Summary of Results

- Do children with ASD produce significantly more speech sound errors (SSEs) compared to typically developing children?

Hypothesis: Children with ASD will present with more overt SSEs than typically developing children.

The key findings that answer this question were that there was no significant difference in DEAP PCC scores or CUW PCC scores between the ASD and TD group. This may have been a result of a limited sample size as there were children with ASD who had a high number of SSEs within these tests. However, when analysing speech errors by categories there was a significant difference of the number of delayed errors produced by the ASD compared to the TD group in the DEAP where more were produced by the ASD group. This would have provided evidence that children with ASD produced more delayed SSEs than TD children, similar to the findings of Cleland et al. (2010) and Shriberg et al. (2011). However, the sample size did not have enough power so results can only function as a guide for future studies.

3.11 Behavioural Results Discussion

This section discusses the results of the behavioural and perceptual assessments conducted with the group of children with autism spectrum disorder (ASD group) and the typically developing group (TD group). An overall discussion discussing all results in relation to the current literature base is presented at the end of this chapter. This section related specifically to the behavioural results including the correlations performed; and how they relate to the research questions.

3.11.1 Speech Sound Errors in Children with ASD

Studies have shown that phonological patterns involving typical and atypical processes are present in children with ASD. Nevertheless, literature is heterogeneous and lacks a set protocol for comparison of results (Shriberg et al., 2011). It has been argued that speech is relatively intact in children with ASD, yet in this study three out of ten children met the criteria for a speech delay using the

DEAP screening and all the children had delayed and/or atypical speech processes in their speech profile. In the TD group, one child was borderline for meeting the criteria of a speech delay. The literature's conflicting results suggest that speech production in children with ASD is not well understood. When assessing speech using the Diagnostic Evaluation of Articulation and Phonology (DEAP; Dodd, 2002). When looking in more detail, while the difference between the total number of errors produced by the ASD group compared to the TD group only approached significance, there was a large Cohen's effect size (0.87), suggesting a large mean difference between the two groups. The lack of a conclusive Bayes Factor result is likely due to sample size. Additionally, when analysing these results into categories of "age appropriate," "delayed errors" and "unusual errors," there was a significant difference in the number of "delayed errors" with the ASD group producing significantly more delayed errors that the TD group. There was no significant difference in "unusual errors" produced, but there were only seven unusual errors produced in the ASD group in total and only one in the TD group. In the ASD group these atypical speech processes included: backing, assimilation and stopping of liquids. Backing to velar, which is the substitution of a velar consonant sound (/k/ or /g/) for a sound further forward in the mouth (McLeod and Baker, 2017). In the TD group four children produced a speech sound with assimilation, a disordered speech error. Whilst little research has been conducted around this speech process, a study by Shriberg found that backing of obstruents (e.g., /s/ or /b/) was positively correlated with children who had a speech delay as a result of otitis media with effusion. This suggests that backing, an articulatory speech process, may be a result of impaired auditory perception (Shriberg *et al.*, 2003). In children with ASD, presence of backing could be indicative of an attunement or auditory perception issue, however this requires research to confirm.

There has been variance in the research on the type and number of SSEs produced by children with ASD at all age ranges. In this study 90% of the ASD group produced some type of SSEs in the DEAP (Dodd, 2002), whether they met a diagnosis for a speech delay/disorder or not. This is a key finding that suggests that children with ASD do produce significantly more SSEs than TD children, particularly in this sample. Shriberg et al. (2001) found a prevalence of SSEs in children with ASD utilizing the "PEPPER" programme. This software permitted investigation of the type

and recurrence of consonant and vowel errors in conversational discourse. Utilizing this strategy, they identified that 33 % of the group with ASD had at least one type of atypical speech sound disordered process (residual speech sound errors, such as lateral lisps), in this study it was 50% of the group. This is in comparison to the prevalence of speech errors at 8 years of age in the general population which is only 7.9% (Wren *et al.*, 2016). This is similar to research carried out with younger groups of children with ASD. For example, atypical vocalizations in the prelinguistic stage of communication indicates that children are at high risk for ASD (9–12 months) (Schoen et al., 2011). To investigate this further Schoen et al. (2011) examined phonological and vocal presentation utilizing broad phonemic transcription speech-like utterances and coded non-speech vocalizations without identifiable consonants. They identified that 30 young children (18-30 months) with ASD showed "atypical vocalizations" and generally produced a reduced number of consonants in contrast to two groups of TD children. While PCC was not significantly different from the TD group, the quantity of speech-like utterances delivered was significantly less. The distinction between the children with ASD and their peers was the presence of "atypical vocalizations". These atypical vocalizations were principally “piercing screeches” (Schoen et al., 2011). This implies that young children with ASD already show differences in speech production from their peers in the early years and that they do not align their speech to the duration, pitch and phonotactic properties of their surrounding language environment. This is supported by findings from Morett et al. (2016) who studied a group of adolescents with ASD and found that, unlike with TD adolescents, even in the presence of a visible listener they did not increase their speech coherency or engagement with the listener. The adolescents with ASD produced fewer gestures and sparser speech that conveyed supplementary information about what they were trying to communicate. Their findings suggest that communication differences in ASD may be caused more by social processing, as suggested in the speech attunement framework (Shriberg *et al.*, 2011)

Additional evidence of SSEs in ASD was found by Cleland et al., (2010) who reported atypical SSEs in participants with ASD in their sample. They completed a phonetic and phonological examination of speech sound production in sixty-nine children with ASD. Utilizing standardized clinical perceptual assessments, just 12 % of the sample had a diagnosis of speech delay following assessment. However,

when utilizing further in-depth phonological and phonetic analysis, they identified that 41 % produced an SSE indicative of both speech delay and speech disorder. The clinical evaluation of speech utilized was a perceptual assessment, the Goldman Fristoe Test of Articulation (GFTA-2; Goldman and Fristoe, 2000). It looks at speech sounds within single words. It is likely that further research sampling words of increasing complexity (polysyllables), maximum performance tasks, or spontaneous speech may reveal motor constraints which have a substantial negative impact on intelligibility or give rise to SSEs. However, in this study, there was no significant difference in the percentage of consonants correct (PCC) in the clinically useful words assessment (James, 2009), a measure of the accuracy of multisyllabic words. Cleland et al. (2010) found atypical SSEs occurred despite whether a child's standard score fell within normal range or not on the GFTA (Goldman & Fristoe, 2000). The CUWs does not provide norms, nevertheless it is expected that a child would have little to no SSEs by the age of seven (McLeod and Baker, 2017).

The results of the study reported here and investigations by Cleland et al. (2010) are in concurrence with other studies (Kjellmer *et al.*, 2018). Rapin et al. (2009) suggest that children with ASD produce SSEs and smaller studies such as Wolk and Brennan (2013) found both delayed phonological patterns as well as some atypical phonological processes in eight children with ASD. Similar to the results here, in which I found a significant number of delayed SSEs in the ASD group and close to a significant number of atypical SSEs in the ASD group compared with the TD group. Cleland et al. (2010) found in their sample that while speech was portrayed by developmental phonological errors (cluster reduction, gliding and final consonant deletion), atypical SSEs, characteristic of a speech disorder, were also present (for example initial consonant deletion and phoneme specific nasal emission). To understand why there might be SSEs in their sample, Cleland et al. (2010) conducted a battery of standardized assessments of language, speech, and non-verbal cognition to look for correlations. They found no correlations between speech and language or speech and cognition in their sample. Unlike in this study in which speech correlated with receptive language abilities and aspects of motor functioning. This suggests that there is individual variation in relation to ASD and SSEs. It may be that while there is commonality in children with ASD having higher rates of SSEs than TD children, the cause of these SSEs vary depending on the child. Some

children with ASD may have motor-based SSEs that could be a result of a speech motor impairment, as seen with 05M in this study's sample, where he had significantly more issues with multisyllabic words than single words. Whereas 07F showed little issues with multisyllabic words, but a significant number of phonological processes evidence in the DEAP (Dodd, 2002). It is likely that there are multiple root causes for SSEs in children with ASD and they need to be observed on an individual basis.

Whilst this study was conducted with English speakers only, the presence of SSEs also occurs in other languages. Wu et al. (2019) conducted a picture naming task to measure participants' phonology – the 21 initials, 36 finals and four tones of Putonghua of children with ASD (3-6 years) and found they were significantly lower than the TD control group. The accuracy of speech production for initial consonants and Tone 3 (the low-rising tone) in the ASD group were significantly lower than in the TD group. The children with ASD also showed atypical SSEs similar to this study and to Cleland et al. (2010). Similarly, Alqhazo et al. (2018) examined phonological and lexical abilities in children with ASD (n=39) aged 4-8 years in Jordan, using the JISH School Readiness Screening Test to measure lexical abilities and JISH Articulation Test (JAT) to measure phonological abilities compared to TD controls (n=40). Their results showed that there were impairments present in both these domains for the ASD group and that there was a greater impairment in phonological abilities.

I hypothesised that the increase of SSEs in this sample of children with ASD may be a result of an underlying neuromotor disturbance such as speech attunement difficulties, in which the child has difficulty tuning in and tuning up to their ambient language environment (Shriberg *et al.*, 2011). Further investigation of auditory perceptual capacities and speech motor abilities is required to comprehend the root cause of the SSEs in ASD, and this is recommended for further studies of SSEs in children with ASD. Wolk and Giesen (2000) completed a phonetic inventory and speech process analysis and found in four siblings with ASD and the speech process that may be indicative of delayed or disordered speech. The atypical processes they identified included unusual sound changes, chronological mismatch, and residual articulation errors. Each of the four children were also significantly delayed in their

fine and gross motor abilities. This is similar to the results reported here in which there were speech processes atypical to normal speech development such as backing, initial consonant deletion and assimilation. The combination of unusual sound changes, which are indicative of perceptual issues (assimilation) and residual articulation errors which are a sign of speech motor issues, suggests these children appear to have a combination of both speech motor control and speech perception issues. Wolk and Giesen (2002) did not find any in suprasegmental production; children with ASD did not produce speech productions significantly different in duration or fundamental frequency from the TD control group. However, in my sample one child has voicing errors (5M), but this was not present in the rest of the ASD group. This implies that this group of children with ASD are able to tune into their ambient language environment in some ways. Perhaps there are many subtypes of speech production difficulties within ASD, depending on the individual children.

Not all literature is in agreement with the presence of issues with speech attunement in children with ASD. Pomper et al. (2019) investigated in sixty-four children with ASD compared to thirty-one younger TD controls whether the children with ASD were less sensitive to mispronunciations of familiar words. They hypothesized that in ASD if the cognitive style prioritized processing local, rather than global features, as claimed by the weak central coherence theory, then children with ASD should be more sensitive to mispronunciations than typical controls. However, there were no significant differences between the two groups, even when accounting for non-verbal cognition or receptive language. Wolk et al. (2016) conducted a review of SSEs in children with ASD and while more recent studies found typical, delayed, and atypical phonological processes in some children with ASD, there were other findings suggesting that the articulatory/phonological skills in these children are relatively intact. This suggests that the literature is still unclear as to which children within the ASD diagnosis produce SSEs and why. The findings here do indicate a large presence of delayed SSEs in ASD compared to TD peers and potentially atypical SSES, however the sample size is too small to be generalisable and requires further study with larger numbers and additional study of speech perception abilities.

3.11.2 Autistic Symptomatology and Speech

The ten children in the ASD group had all received a formal diagnosis of ASD and therefore I did not conduct any assessments to give a diagnosis of ASD. However, I still wanted to examine correlations with their autistic symptomatology and the behavioural assessments conducted, therefore, the Social Communication Questionnaire (Rutter, Bailey, and Lord, 2003) was used. Interestingly from these results, only four out of the ten children met the threshold for social communication associated with ASD in the current form, which is reflective of how they are presenting now. Therefore, it may have been expected these children with ASD had lower rates of SSEs than other studies with the same groups, due to milder presentation of social communication difficulties associated with ASD. However, when using the lifetime form, which is reflective of their presentation across their lifetime, eight out of the nine children (who participated) met the threshold for social communication traits associated with ASD. What these results indicate is that there has been a change in the presentation of social communication in this group of children with ASD over time. This may indicate there was a change in the number of SSEs the children were making and has perhaps resolved with age. Future research could look at the presentation of SSEs in children with ASD over the course of their speech development as a positive shift may be noted.

Change in autistic symptomatology through the developmental period is well recorded in the literature. A longitudinal study conducted by Scheeran et al. (2020) found that 69% of children and adolescents with ASD had an overall improvement in their social behaviour. This has also been recorded at later stages of development from adolescence into young adulthood (Picci and Scherf, 2015). Fountain et al. (2012) had similar findings to this study in which children with ASD who had mixed intellectual abilities showed a developmental trend of moving from an aloof or passive social communication style to a more typical style. This was mainly noted in the first six years of life. A time in which children's speech development is also becoming more similar to the adult model (American Speech Language Hearing Association (ASHA), 2017). This might be interpreted as evidence that the change of autistic symptomatology scores from the lifetime to the current form is to be expected as it is in line with the developmental trajectory of this group of children with ASD

and does not necessarily mean there has been misdiagnosis. It should be noted, however, that atypical social interaction does form the basis of an ASD diagnosis (American Psychiatric Association, 2013) but that children with an ASD diagnosis show large individual differences and this should be accounted for when drawing conclusions from research with this group (Klin *et al.*, 2003). As seen in this study's results, there was significant variability in the number of SSEs produced by children in the ASD group. For example, 09M produced six different types of speech processes within his speech, five of which were delayed and one atypical (backing), whereas 02F only showed one speech process (velar fronting). Both of these children had the same scores on the social communication questionnaire (Rutter, Bailey and Lord, 2003). The higher rates of SSEs seen in 09M's speech may not have been a result of his social communication style but other factors.

3.11.3 Gross and Fine Motor Abilities

As 90% of the ASD group presented with SSEs in the DEAP (Dodd, 2002), it is important to understand if this high rate is also reflected in their movement abilities. Within the ASD group, there were significant movement issues found. Seven out of the ten children in the ASD group scored at or below the 5th percentile, denoting a significant movement difficulty. Two out ten of the children, 04M and 10F in the ASD group, score between the 5th and 15th percentile, judging them at risk of a significant movement difficulty. However, this was not reflected in their speech production, 10F only had two speech errors across both the DEAP (Dodd, 2002) and CUW (James, 2009). She did, however, have atypical speech processes in both assessments. So, while 10F did not have a large variation of speech processes, those that did exist were atypical. Only one child in this group, 08M, was found to have performed within the normal range for their age. He also was one of the children who had less impaired speech showing only two delayed speech processes in the DEAP (Dodd, 2002) and having a within range PCC score. It appears the children with less motor impairment had better speech production. These results agree with current research that children with ASD are more likely to show motor impairment than typically developing peers (Ament *et al.*, 2014).

Motor impairment is not currently included in the diagnostic criteria or evaluation of ASD (Licari, Alvares, Varcin et al., 2019). However, a meta-analysis of motor data in ASD suggests motor disruption may be a core feature of ASD and not merely a co-morbid or associated condition (Fournier *et al.*, 2010). Studies have shown significant rates of motor difficulties (50-79%; Dewey, Contell and Crawford, 2007; Green et al., 2019). In a comprehensive study Licari et al (2019) found in a population-based cohort of 2084 children with ASD, one-third exhibited movement difficulties. This has been one of the largest studies to date that show the significant comorbidity of movement impairment and ASD, results which are in line with the findings in this research. Higher rates reported in my study are likely to have been reported due to the use of objective assessment in the form of the MABC-2 (Brown and Lalor, 2009) unlike in Licari et al.'s (2019) study who used a parent-based evaluation. My results demonstrate high comorbidity of movement impairment, even within a small sample. Interestingly, while researchers have argued that using assessments with a combined approach to gross and fine motor assessment would not be sensitive enough to identify atypical motor abilities in children with ASD (Noterdaeme et al., 2002; Provost et al. 2006) this has not been the case in this study. The MABC-2 has shown to be sensitive enough to identify motor difficulties in this group of children with ASD, as found with other studies (McPhillips et al., 2014). What we can take from these results is that there is evidence of a significant motor impairment in this sample of children of ASD and if this was a larger sample size, which may be generalised to the group itself.

There may be a fundamental underlying problem with motor timing and integration required to produce the correct, efficient kinematic patterns required of skilled movements, including speech (Beversdorf et al., 2001; Gowen & Hamilton, 2013a; MacNeil & Mostofsky, 2012; Mostofsky et al., 2009; Whyatt & Craig, 2013). Such disruption to movement early in a child's development is thought to contribute to the broad ASD phenotype, disrupting expressive intention and purposeful engagement with others, causing frustration, distress, and isolation (Trevarthen and Delafield-Butt, 2013). In verbal expression, articulating fluently requires intricate control and coordination of speech motor mechanisms (Gracco, 1994). Therefore, this perspective proposes the increased rate of SSEs present in children with ASD may be a result of common, underlying motor difficulties. Indeed, the residual articulation

errors reported by Shriberg et al. (2001) affect the late acquired and articulatory complex speech sounds such as sibilants and rhotics; sounds that require intricate speech motor skills. Similarly, in this study there were a large number of children who produced articulatory atypical speech processes such as backing.

The evidence of motor impairment in children with ASD is now more recognised than ever. Neuroanatomical functions and structures have been proposed for the difficulties found in motor abilities, this includes abnormalities in the cerebellum (Fatemi et al., 2012), impeded tangible information, multisensory joining (Gowen and Hamilton, 2013) and disturbance in cerebrum synchronization (Welsh, Ahn, and Placantonakis, 2005). Thus, it is suggested that if motor abilities are impeded in ASD, this could bring about a speech motor control issue (Barbeau et al., 2015). Adams (1998) analysed oral-motor and speech motor production of four children with ASD compared to TD youngsters in both basic and multisyllabic speech production. Information showed that children with ASD had altogether more difficulty performing oral movements and multisyllabic speech tasks compared to the TD children. These outcomes could demonstrate a speech motor impairment. Nevertheless, because of the small sample size, these outcomes are not generalizable, and it is possible that the SSEs present in this sample are co-occurring rather than related. More research in this area is required and will be pivotal to understanding the presence of SSEs.

Chapter 4 – Instrumental Methods, Results and Discussion

4.1 Introduction

In this chapter the analysis conducted on the data collected from the instrumental assessments, diadochokinesis tasks and the statistical tests used to answer the research questions of the study is described. Furthermore, the analysis of ultrasound tongue imaging data is given in detail on how data was prepared for analysis and statistical tests used to analyse this. There is a specific focus on how statistical tests are related to measures of speech motor control and what they tell us about the articulatory data.

4.1.1 Research Questions and Analysis

RQ2. Does instrumental analysis of speech reveal subtle articulatory differences between ASD and TD groups?

Hypothesis: Instrumental analysis reveals more subtle SSDs than perceptual methods of assessment.

Analysis: To answer this research question identification of speech problems using the instrumental method (ultrasound tongue imaging) and the perceptual method (DEAP; Dodd, 2002) was required. To determine this, I analysed variation of tongue curves using ultrasound data taken from the DDK task of children with ASD and typically developing children and highlight if subtle speech motor difficulties or impairments are identified using UTI but not in the speech perceptual results of the DEAP and DDK (described in the methods chapter).

RQ3. Do children with ASD present with speech motor impairment symptoms?

Hypothesis: There are a subset of children with ASD who present with speech motor control difficulties

Analysis: I compared rate, accuracy, and consistency perceptually across groups (and to published norms) in a diadochokinesis test (DDK- rapid alternating syllables such as pa ta ka). I then conducted an independent samples t-test for this analysis. I predicted children with ASD would have lower consistency and accuracy scores than TD

children.

4.2 Diadochokinesis Speech Task

Diadochokinesis (DDK) is a type of oro-motor assessment used to study motor control by assessing the performance of rapidly alternating movements. This study assessed rate, accuracy, and consistency of DDK tasks. This allowed us to examine if there were speech motor control difficulties in any of these three measures and what that might mean in relation to speech production in children with ASD.

4.2.1 Procedure

Five repetitions of single syllables /pə/, /tə/ and /kə/ and sequences /pə tə kə/ and /tə kə/ were recorded at six set rates using the protocol developed by McCann and Wrench (2007). All prompts were recorded using neutral vowel (schwa) to control for vowel environment. Each participant received the same instructions with recorded and standardized productions of each DDK task. These target productions were recorded at -3 to +2 standard deviations of the mean rate. Mean rates for single syllables are taken from Robbins and Klee (1987) and rates for sequences taken from Williams and Stackhouse (Williams & Stackhouse, 2009).

4.2.2 Perceptual Analysis

Articulate Assistant Analysis software was used to calculate number of syllables per second produced. The number of syllables produced by each participant does not matter as a minimum of three without pauses was used. The following measures were applied:

a) Rate: Average time to produce five repetitions of syllables and sequences were calculated for each participant.
b) Accuracy: Accuracy were calculated by transcribing the first syllable/sequence produced by participants. Participants were scored 1 for correct production or 0 for incorrect (Williams and Stackhouse 2000).
c) Measure of Consistency:

Consistency of the repetitions compares the child's own baseline production to the rest of the repetitions made. The child's first production of the target is taken as the baseline. A binary scoring system was used:
1= The child produces multiple repetitions that match the first baseline production
0= Where one or more repetitions differ from the baseline production, not taking the adult model into account.

These measurements of DDK does not allow identification variability in tongue shape across repetitions also frequently associated with apraxia or motor coordination difficulties, so further in-depth analysis was conducted using ultrasound (McCann and Wrench 2007). DDK is an effective measure complex articulatory gestures (multisyllabic words) require ability to rapidly select and sequence phonemes under high motor demands (Lewis & Freebairn, 1997). The accuracy of this task reveals information on motor planning and speech. Scoring involved calculating percentage of consonants correct (PCC). It is possible to take further measures of consistency, looking at the consistency of all the repetitions and not only the first as the baseline, however due to time constraints of the analysis, this was not conducted. Future research could include this for a richer analysis of consistency.

4.3 Analysis of Diadochokinesis Task

The diadochokinesis (DDK) test was conducted to assess speech motor skills in both the ASD and TD group. This study assessed at rate, accuracy, and consistency of DDK tasks. This allowed us to examine if there were speech motor control difficulties in any of these three measures and what that might mean in relation to speech production in children with ASD. Where an independent samples t-test was conducted data was tested for normal distribution using the Shapiro Wilks test, where a significant p value would indicate that the data was not normally distributed and would require a non-parametric test. The Shapiro-Wilks test for normality is designed to detect all departures from normality. It rejects the hypothesis of normality when the p-value is less than or equal to 0.05. The p-values from this test are reported in the statistical tables, none of the Shapiro-Wilks tests in this study had a p-value less than 0.05.

4.3.1 Rate

A measure of the maximum rate of DDK productions was calculated for each child. The child was instructed to listen to the prompt and repeat the target production, this occurred at five different speeds. However, for this particular measure I examined the maximum rate produced at the fastest speed. As children often stopped before five repetitions were produced which was a common problem at the sequence level (/tk/ and /ptk) in which children stopped short in producing the 5 required repetitions. To account for this, I chose to measure DDK rate as the time taken to produce one syllable in seconds. Therefore, rate was calculated by recording the number of repetitions produced for the target DDK task, then dividing the time taken to produce these repetitions, as shown in the following formula:

Number of repetitions produced / Time took to produce these repetitions = Rate to produce one syllable/second

Both accurate and inaccurate repetitions were accepted in the rate measure. If the child stopped for longer than 0.25 seconds then it was taken as an indication that the child had finished the string of repetitions (Williams and Stackhouse, 2009). Data was inputted and analysed in IBM SPSS Version 25. I verified that the data collected met the criteria to conduct parametric statistical tests of significance by carrying out a Levene's test of Equality of Variance. This showed that there was a homogeneity of variance across groups and conditions with normality of distribution. Therefore, an independent samples t-test was conducted to investigate the differences in maximum DDK rate across the two groups.

4.3.2 Comparison to Norm

The maximum rate of DDK repetitions produced by the children were also compared to the standardised norms produced by Fletcher (1978) who derived them from syllable repetitions by 384 school-age children using the time-by count method as described in the literature review. Table 14 shows the norms that were developed by Fletcher (1978). I age matched each participant to the appropriate age of each norm set out in this table for appropriate comparison.

Table 14: *Norms for DDK (Fletcher, 1978)*

Age	pʌ	tʌ	kʌ	fʌ	lʌ	pʌtə	pʌkə	tʌkə	pʌtəkə
6 Mean	4.8	4.9	5.5	5.5	5.2	7.3	7.9	7.8	10.3
SD	0.8	1.0	0.9	1.0	0.9	2.0	2.1	1.8	3.1
7 Mean	4.8	4.9	5.3	5.4	5.3	7.6	8.0	8.0	10.0
SD	1.0	0.9	1.0	1.0	0.8	2.6	1.9	1.8	2.6
8 Mean	4.2	4.4	4.8	4.9	4.6	6.2	7.1	7.2	8.3
SD	0.7	0.7	0.7	1.0	0.6	1.8	1.5	1.4	2.1
9 Mean	4.0	4.1	4.6	4.6	4.5	5.9	6.6	6.6	7.7
SD	0.6	0.6	0.7	0.7	0.5	1.6	1.5	1.7	1.9
10 Mean	3.7	3.8	4.3	4.2	4.2	5.5	6.4	6.4	7.1
SD	0.4	0.4	0.5	0.5	0.5	1.5	1.4	1.2	1.5
11 Mean	3.6	3.6	4.0	4.0	3.8	4.8	5.8	5.8	6.5
SD	0.6	0.7	0.6	0.6	0.6	1.1	1.2	1.3	1.4
12 Mean	3.4	3.5	3.9	3.7	3.7	4.7	5.7	5.5	6.4
SD	0.4	0.5	0.6	0.4	0.5	1.2	1.5	1.1	1.6
13 Mean	3.3	3.3	3.7	3.6	3.5	4.2	5.1	5.1	5.7
SD	0.6	0.5	0.6	0.5	0.5	0.8	1.5	1.3	1.4

However, as this study used the count-by-time scale, as children often stopped before five repetitions were produced, both the means and standard deviations had to be converted from the original norms to fit with this measurement.

In order to convert this project's data for comparison with these norms, my data collected was averaged to represent the mean maximum DDK rate in a count-by-time scale. I followed a similar procedure to Robb et al. (1985) in which their data was reported in a time-by-count scale, and they converted it to a count-by-time scale. The norms were created from the time it takes (in seconds) for ten repetitions for /p/, /t/, /k/, /tk/ and for thirty repetitions of trisyllable sequence /ptk/. I performed the following transformation in order to compare to these norms, the formula below is an example of the calculation carried out on the /ptk/ sequence:

DDK Rate (syllables/s) = 30 reported average time.

If there were less than 30 trisyllable sequences produced or the sound target varied, then the number was adjusted accordingly.

4.3.3 Accuracy

Accuracy of the DDK productions were also analysed. The two measures of accuracy in this study were taken from the Williams and Stackhouse (2009) method.

- Accuracy of the first repetition of the target compared to the adult model

Each child's attempt at imitating the target once was transcribed from audio recordings using IPA symbols within the AAA software (Articulate Instruments Ltd, 2019). For single syllables (e.g., /p/) this was scored using the first syllable produced and for sequences (e.g., /tk/) this was scored using the first whole sequence (di- or tri-syllable) produced. Only consonant sounds were scored using a binary scoring method from a set list of criteria in which to gain one point, all criteria listed needed to be met (table 15).
1 point was given for a correct production and 0 point for incorrect.

Table 15: *List of criteria for correct and incorrect productions of single repetitions*

Correct 1 point	**Incorrect 0 points**
First attempt	Attempt other than first attempt
Modelled target produced	A different target produced
Correct number of syllables produced	Syllable omissions occurred
Consonants produced as in adult model - no deletion or addition errors and no substitution errors	Consonants not produced as in adult model –deletion, addition, or substitution errors occurred

- Accuracy of five repetitions of the target compared to the adult model

Each child's attempt at imitating the target in five repetitions was transcribed from audio recordings using IPA symbols. Only consonant sounds were scored in which a binary scoring method from set criteria was used. The binary scoring method used was: 1 point for a set of five correct repetitions, which met each of the five correct criteria (table 16), and 0 points for a set which included one or more incorrect productions.

Table 16: *List of criteria for correct and incorrect productions of five repetitions*

Correct 1 point	**Incorrect 0 points**
First attempt	Attempt other than first attempt
Modelled target produced	A different target produced
Correct number of syllables produced	Syllable omissions occurred

Produced run of 5 repetitions	Stopped before 5 repetitions were produced
Consonants produced as in adult model - no deletion or addition errors and no substitution errors	Consonants not produced as in adult model –deletion, addition, or substitution errors occurred

A paired samples t-test was conducted to compare the accuracy of all participants in single syllable and sequence conditions. This statistical test allowed us to examine whether there was a statistical significance difference between the accuracy of the single syllables and the sequences for both groups. In a paired samples t-test each target is measured twice resulting in pairs of observations. Following this independent samples t-test was conducted to examine whether there was a significant difference between the ASD and the TD group in the accuracy of their productions of the single syllables and sequences of the DDK task.

4.3.4 Consistency

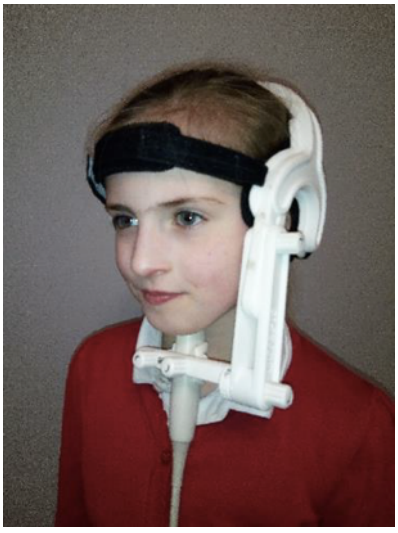
An independent sample t-test was conducted to compare the consistency of the production of single syllables of participants in the ASD group and participants in the TD group. An independent sample t-test was also conducted to compare the consistency of the production of sequences of participants in the ASD group and participants in the TD group. The same statistical test was used to compare the consistency of the overall (combination of single syllables and sequences) across both the ASD and TD group. A Shapiro-Wilks test was conducted which confirmed that data was normally distributed.

4.4 Ultrasound Tongue Shape Analysis

Ultrasound tongue imaging is non-invasive. The ultrasound probe was placed under the chin using a stabilising headset (figure 8). There are some issues with capturing data using UTI. Images obtained can vary in quality, so reliability of the identification of the tongue surface may be impacted (Dawson et al., 2015). It requires taking image quality into account when analysing tongue contours. Additionally, UTI requires stabilisations of the head and jaw in order to determine the position of the tongue relative to palatal hard structures (Dawson et al., 2015). Methods such as a

wearing a helmet to stabilise the probe were required, which can impact the naturalness of speech produced. This ensured that the ultrasound probe did not move during the recording of speech and change position of where the tongue was measured. This ensured validity in cross comparison of children's images as they were all taken from a similar, stable position. Ultrasound tongue imaging took twenty minutes per child and was recorded in a quiet room using university equipment at the school on in the University SLT department.

Figure 8: *Ultrasound Headset*

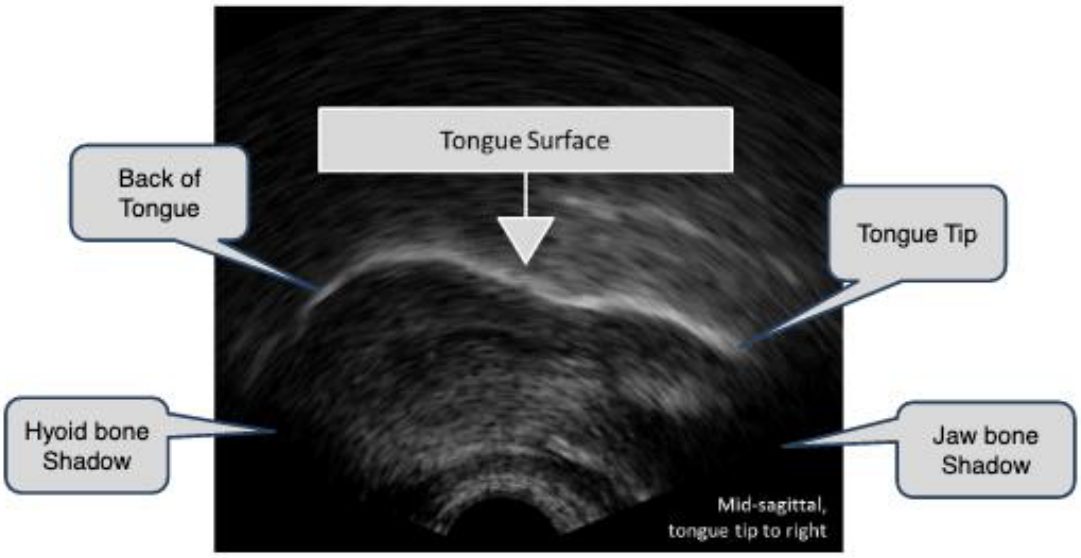


Ultrasound data was collected using a sonospeech high-speed cineloop system at 100fps over a 156-degree field of view. Data was collected in the mid-sagittal view (figure 9). Shaping and movement of the tongue involves coordination of complex range of muscles (Dawson et al., 2015). Inaccuracy or uncoordinated movements of the tongue may indicate the presence of motor coordination impairment, which can be effectively measured using UTI and compared across groups (Zharkova et al., 2011). The targets chosen were from the speech repetition task (DDK) in order to evaluate SSEs in different syllabic contexts and in an assessment focusing on speech motor control.

Automatic tracking of tongue is provided by specialised software such as Advanced Articulate Assistant (Articulate Instruments Ltd, 2012) meaning that ultrasound is an attractive method for collecting data on real-time kinematic information about tongue movement (Xu et al., 2016). Ultrasound is useful for research as you can apply

measures to reveal the mathematical attribute of the tongue shape to gain information about the trajectory of the tongue (Dawson et al., 2015).

Figure 9: *Ultrasound Image of Tongue in Midsagittal View*



To capture the subtle articulatory movements of the tongue, ultrasound tongue shape analysis was conducted in order to make quantitative analysis of variation in tongue shape in the production of the DDK task. Therefore, the DDK production task was simultaneously conducted alongside ultrasound imaging of the tongue.

4.4.1 Analysis of Ultrasound Data

Consonants were annotated using Articulate Assistant Advanced (AAA) software (Articulate Instruments, 2012). The duration of the consonant closure was annotated, and splines automatically fitted to every frame. Midpoints were extracted for single point analyses, or the sequence of frames can be extracted for dynamic analyses. Multiple splines were exported to a “workspace” to allow comparison of tongue shapes. The software uses a fan-based system, collecting data from forty-two fanlines emanating from the transducer. This allows exportation of radial or Cartesian coordinates for analysis.

4.4.2 Preparing the Data

The syllables and sequences /p/, t/, /k/, /tk/ and /ptk/ were first annotated in AAA (Articulate Instruments Ltd, 2019) acoustically and from observation of the spectrogram. This judgement was made using the acoustic waveform, spectrograms, and perceptual evaluation. The syllables were marked according to the following criteria: The beginning was marked at the start of the burst and the end marked by the appearance of a pitch period after the burst where the formant structure of the following sound is visible (figure 10). The closure phase was not annotated as this included the vowel and my study was focusing on speech motor control which is more evident in consonants. Inter-syllables (the time between target syllables) for analysis of rhythm and duration were also marked between each segment (figure 11). Total duration of the production of each target was then marked from the first burst to the end of the last vowel e.g., five repetitions of /p/ to calculate rate (figure 12).

Figure 10: *Burst of /t/*

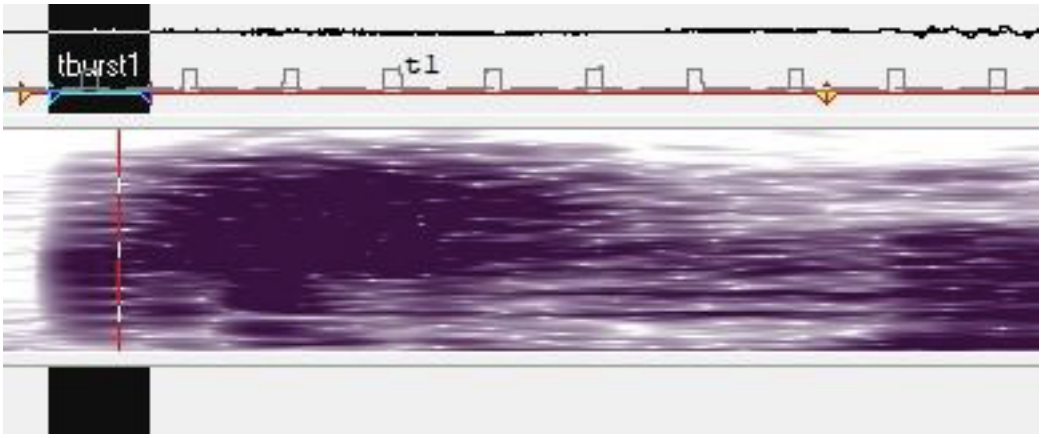


Figure 11: *Intersyllable*

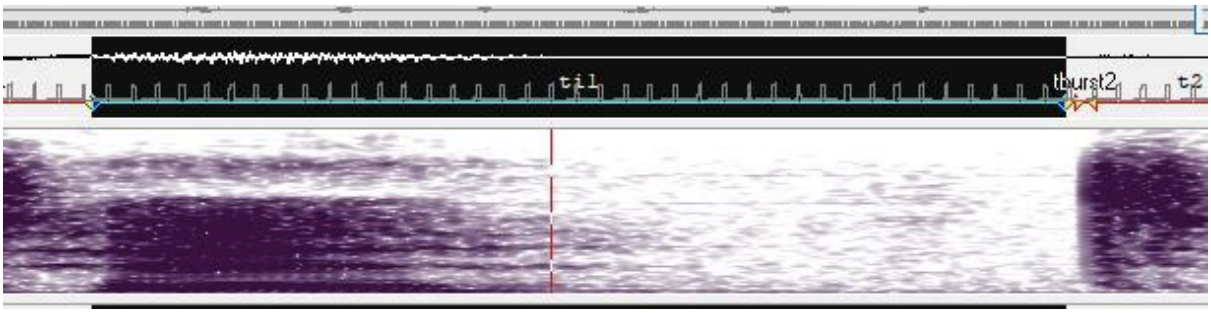
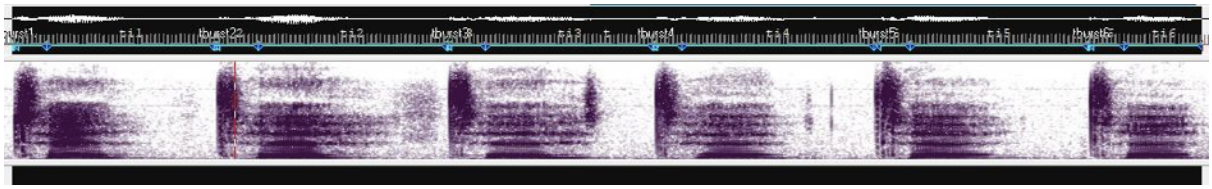


Figure 12: *Total /t/ Production*

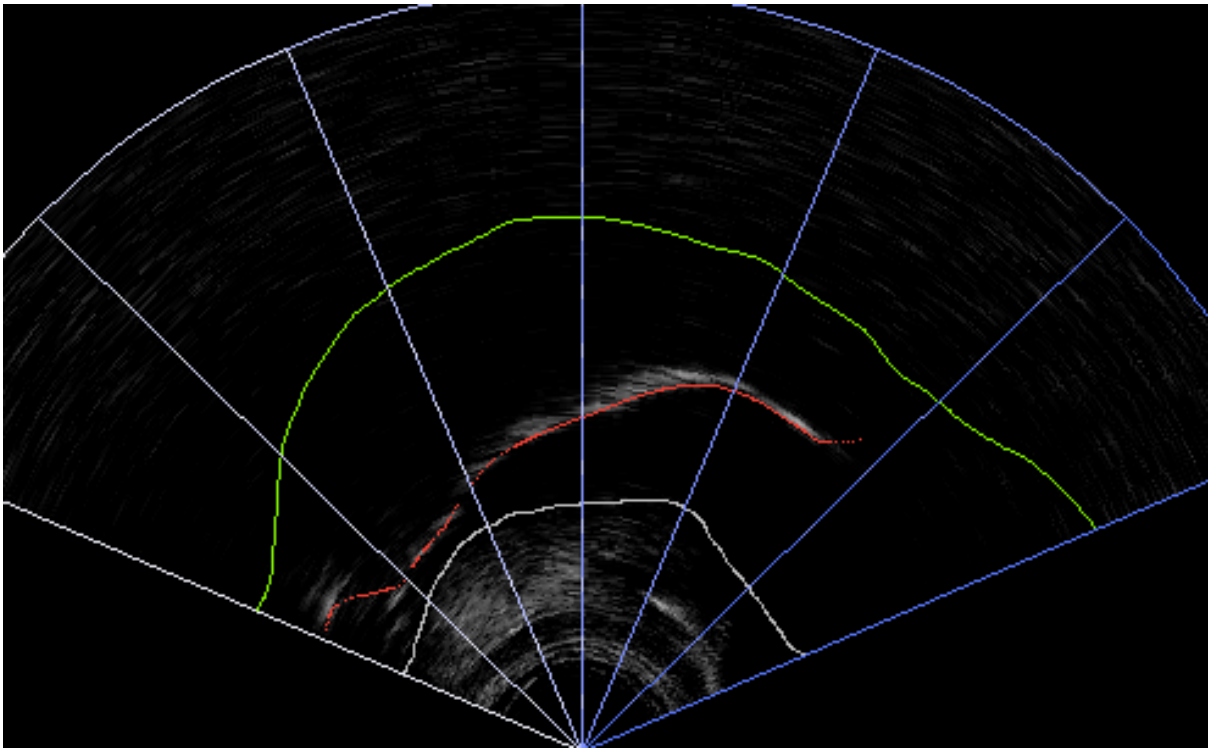


4.4.3 Batch splining

In order to determine the variability of tongue movements, the tongue contours had to be fitted to all frames of the annotated syllables. This requires fitting a tongue contour line to the white line of the tongue-air boundary. This can be conducted automatically using “batch splining” a feature of AAA (Articulate Instruments Ltd, 2019) where the white line of the tongue-air boundary is automatically tracked by the software to produce tongue contours. However, it requires a template of the tongue shape which was drawn and manually fitted for each participant by the researcher. A tongue contour template was fit to the lower edge of the bright curve representing the tongue surface manually.

Once AAA software (Articulate Instruments Ltd, 2019) can identify a bright lower edge a contour is automatically create for each ultrasound frame. Upper and lower boundaries of the space in which the tongue moves (i.e., hard palate and lower bounds of the tongue movement) were also set (figure 13). Together these three lines were used as a template to be used for automatic tracking of the tongue. All relevant frames were then batch “splined” i.e., tongue contours were added to each frame automatically (Articulate Instruments Ltd, 2019). Data was then visually inspected and manually corrected if any errors of tongue contours were produced in the automatic tracking process. These errors occur if the image of the tongue surface is not bright enough for the software to track automatically.

Figure 13: *Template with annotated tongue contour (red), upper boundary (green) and lower boundary (white)*



4.4.4 Exporting into the Spline Workspace

Once all the ultrasound frames have been fitted this creates tongue contours which can be used to compare different tongue shapes within the AAA software (Articulate Instruments Ltd, 2019). When all the ultrasound frames have been splined, the tongue contours were exported to the spline workspace for the bursts only (figure 10) to calculate the mean and standard deviation of tongue shapes. The in-built mean and SD function within AAA was used to create an average curve for the target sound e.g., /p/ in /ptk/. Having the tongue contour represented in x, y points allowed comparison of the shape of the tongue in different frames. It also allowed measurement of the distance between tongue positions from different frames. This was particularly important when measuring variability of tongue shapes.

When exporting tongue shape contours to the spline workspace, this included data points that were automatically tracked but not valid tongue contour points as seen in figure 14 at the outer edges of the tongue, particularly the anterior. These are not included in the later analysis of the tongue shape where variation of tongue shape is analysed. The mean tongue curve over multiple repetitions can then be calculated

within the spline workspace as well as the standard deviations as shown in figure 10. This allowed comparison of the mean tongue shape at the fastest and slowest productions as well as across the different sound targets within the DDK task (/p/, /t/, /k/, /tk/ and /ptk/).

Figure 14: *Mean tongue repetitions*

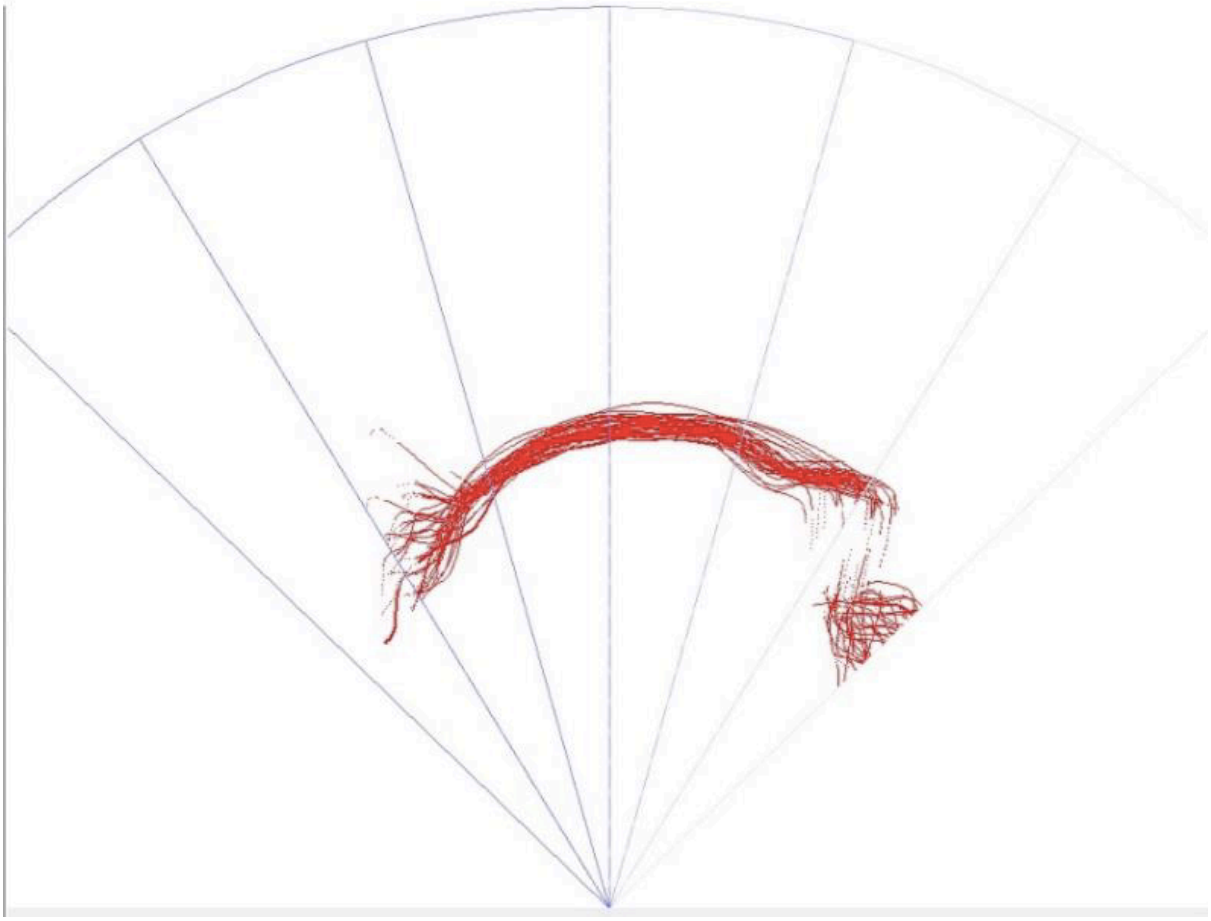
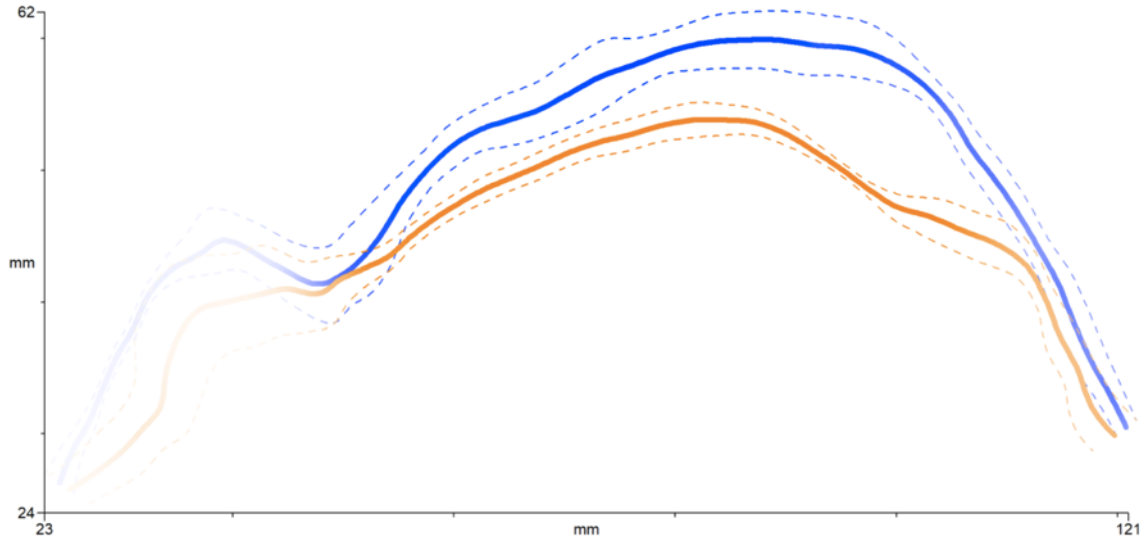


Figure 15: *Mean and standard deviation of /t/ tongue shape at slowest (blue) and fastest production (orange)*



Note. Dotted lines are standard deviations and full lines are the mean. Orange is the slowest production (-3SD) and blue is the fastest (+2SD)

4.5 Statistical Measures

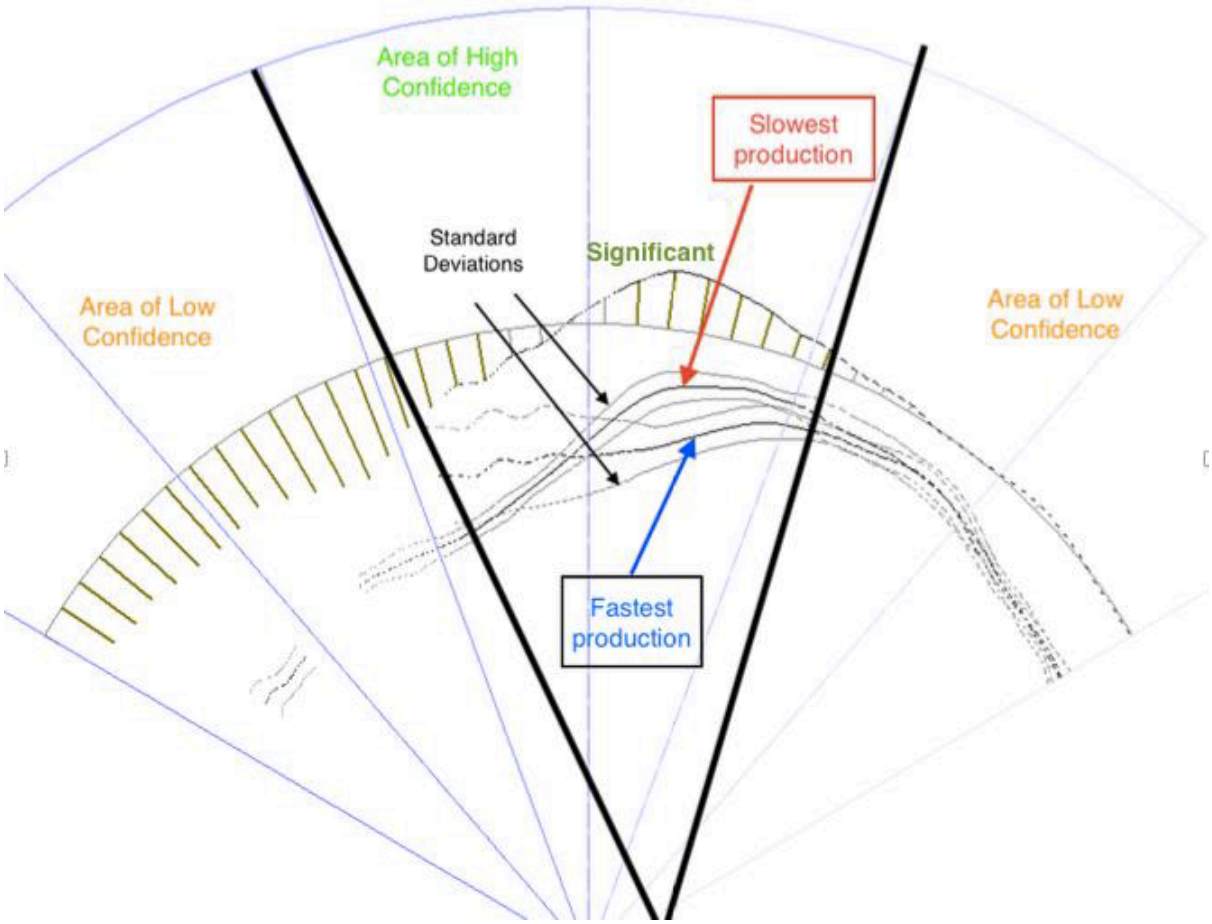
Multiple splines were exported to a "workspace" to allow comparison of tongue shapes. These measures were used to determine whether there was variability in tongue configuration during speech as this may indicate the presence of speech motor coordination difficulties. The annotated and splined ultrasound data were used to compare the results of the ASD group to the TD group using multiple statistical tests. I was particularly interested in variability during DDK tasks.

4.5.1 Independent Samples T-Tests of Tongue Shape Variation

An independent samples t-test was conducted to analyse the variability of tongue shape across the slowest and fastest productions of each DDK target. The statistical significance was evaluated along each fan line within the spline workspace. As adjacent parts of the tongue along each fan line are not independent, including their distance from the fan-grid's origin (originating in the centre of the probe) (figure 16), I

was not able to take only one point of significance as valid. Instead, this study followed similar protocol to Cleland et al. (2015) in which a part of the tongue was taken to be statistically significant if there were significance of >0.05 along a series of six adjacent points of the tongue curve. They specifically took a threshold of statistical significance at six significant adjacent fan line points on the tongue, (which would be an estimate of around 3cm of tongue length) and this study took the same threshold for my study. This ensured statistical significance was not claimed over only a few significant t-tests within a large area of non-significance of the tongue shape. The t-test took the form of comparing the mean tongue shape of all the repetitions of a sound target (/p/, /t/, /k/, /tk/ and /ptk/) for each participant and this was compared using the inbuilt t-test function in AAA within the ASD and TD group. This study looked at tongue shape variation as token-to token variation can be an indicator of reduced speech motor control (Zharkova, Hewlett and Hardcastle, 2011).

Figure 16: *Mean and standard deviation of /t/ tongue shape at slowest and fastest production in spline workspace*



Note. Labelled diagram of exported tongue shape in the spline workspace. Slowest and fastest mean production of tongue shape indicated from blue and red boxes. Long green line marked “significant” is an indicator of statistical significance at that particular point in the figure. Area of high confidence is calculated from p tests of tongue at each fanline. Area of low confidence is where the ultrasound recording may not be accurate enough to produce accurate tongue shape and is therefore not included in analysis. This is calculated from p tests.

In addition to the independent samples t-test of variation in mean tongue shape this study also assessed statistical variance using the Levene’s test of homogeneity of variance. A Levene's test is an inferential statistic used to assess the equality of variances for a variable, in this case the p-values of the comparison of the slowest and fastest productions of a target sound. Levene’s test for equality of variances was applied to examine the variability of tongue movements between the ASD and TD group (Peng *et al.*, 2004). It tests whether there is equal variance of mean values in the ASD and TD group and would affect which statistical test is chosen to compare the two groups.

4.5.2 Standard Deviations of Mean Tongue Shapes

The standard deviation of averaged sets of tongue-shapes (which was expected to be high) across multiple repetitions were calculated automatically in AAA and compared across groups using a paired samples t-test of the slowest and fastest productions of each sound target. Standard deviations of the mean tongue shape show whether there is a large variation in the tongue shape of each sound target for each participant. Large standard deviations for one participant shows there is a lack of consistency of tongue shape. It was expected to find children with ASD have a larger standard deviation in tongue shape than the TD group (Deshmukh et al., 2012).

In addition, a Levene’s test of homogeneity of variance was carried out to assess which statistical test for comparison as appropriate and whether there was equal variation of standard deviations across both groups. Furthermore, a paired samples t-test was conducted to test whether the standard deviations of tongue shape at the slowest and fastest production of sound targets are significantly different in both the TD group and

the ASD group. This was calculated separately for each group to determine whether there was any significant difference between the slowest production and fastest for each participant. It was chosen to use the slowest and fastest rate as there were five different rates and choosing the slowest and fastest enabled me to see whether a significant difference occurs as a result of a great change in rate. Any significant differences were highlighted and tallied for each group.

4.6 Syllable Durations

The duration of syllables was assessed to determine whether there was a difference in rhythm and timing of productions for the TD and ASD group. It was expected little variation if there was no presence of speech motor problems as the prompts provided were recorded using a metronome so the child's copy of this should have been evenly paced (McCann and Wrench, 2007.) Variability of syllable duration has been associated with speech motor impairment in conditions such as dysarthria (Rusz *et al.*, 2015) and to determine if a speech motor impairment was present in ASD and if it is similar to other conditions with speech motor control as a core impairment. This was conducted on both the slowest and fastest DDK productions of each sound target (/p/, /t/, /k/, /tk/ and /ptk/). The syllable duration of each sound target was annotated within the AAA software. (Articulate Instruments Ltd, 2019). A syllable was defined as the time between the burst of the plosive to the end of the adjacent vowel was measured using a spectrogram within the AAA software. The syllable durations that were calculated from each target sound or sequence were exported from the software to an excel spreadsheet. The slowest and fastest productions of DDK were exported. Following this, for each sound target the mean syllable duration was calculated as well as the standard deviations. Both accurate and inaccurate repetitions were accepted within the syllable duration measures to observe the timing of the production and not the accuracy in this measure. My method for measurement for syllable duration was informed from the literature on syllable durations in DDK tasks. This study focused on the method by Lowit et al. (2018) in which the mean syllable duration and standard deviation were found for each participant. This is a standard measure of syllable duration and helps to determine whether DDK as a simple perceptual speech assessment is sensitive enough to determine differences in rhythm between TD and ASD groups. This was to observe the regularity of these durations through the

measurement of the standard deviations as this would show the rhythmic quality of the speaker's production.

4.6.1 Statistical Analysis

A Levene's test of homogeneity of variance was conducted to assess the equality of variances of mean durations for the TD and ASD group. A low amount of variance would mean there is consistent rhythm, and a high amount of variance would mean there are irregular syllable lengths. Following this a one-way analysis of variance (ANOVA) of mean syllable duration was calculated using SPSS which also provided standard deviations of the mean syllable durations. The null hypothesis of the ANOVA is the same as an independent samples t-test in which if there is no significant difference between the ASD and TD group then syllable durations were similar across both groups and therefore similar rhythms are present in both groups. However, it was expected that there would be a significant difference between the two groups in syllable duration. After cleaning the data, it was ensured that the test assumptions of the ANOVA were met. The statistical test was run and if the p-value associated with the F is smaller than .05, then the null hypothesis is rejected, and the alternative hypothesis is supported.

Intra- and inter-rater reliability was conducted on 20% of the speech assessment data, both perceptual and acoustic to ensure reliability in the results presented. Intra-rater reliability is the consistency of the data by one rater across multiple trials. Inter-rater reliability is the consistency of data by two raters measuring the same subjects over a single trial. Both are used to determine that the measurement tool produces reliable results.

4.6.2 Inter and Intra Rater Reliability

4.6.2.1 Speech Sound Assessments

Intra and Inter-rater reliability was calculated for the single syllabic test, the Diagnostic Evaluation of Articulation and Phonology (DEAP; Dodd, 2002) and the multisyllabic test, the Clinically Useful Words (CUW; James, 2009). Four participants

were selected at random, accounting for 20% of the data. This was scored using a three-point system to rate the equivalence of transcriptions on a token-by-token basis:

0 = Different
0.5 = Almost equivalent: This includes functional equivalence, e.g., essentially equivalent phonetic transcriptions of a target behaviour that uses alternative symbolisation; and near functional equivalence, e.g., nearly equivalent phonetic transcriptions of a target behaviour in terms of place and manner features.
1 = Identical.

Table 17 shows the percentage of agreement between one rater. There was a high percentage of agreement between the two scoring trials for both speech assessments across the four participants.

Table 17: *Intra-rater Reliability, Percentage of agreement of transcriptions with one rater.*

Participant 1	***DEAP***	100%
	CUW	95%
Participant 2	***DEAP***	90%
	CUW	90%
Participant 3	***DEAP***	100%
	CUW	100%
Participant 4	***DEAP***	100%
	CUW	100%

Table 18 shows the percentage of agreement between the two raters. There was a high percentage of agreement between the raters for both speech assessments across the four participants.

Table 18: *Percentage of agreement of transcriptions between the raters.*

Participant 1	***DEAP***	95%
	CUW	75%
Participant 2	***DEAP***	85%
	CUW	85%
Participant 3	***DEAP***	100%
	CUW	90%
Participant 4	***DEAP***	90%
	CUW	80%

4.6.2.2 Diadochokinesis Task (DDK)

The Intraclass Correlation Coefficient (ICC) was used to measure the consistency of the primary rater's ratings for the subtests and was calculated using SPSSv.20. Koo and Li (2016) categorised the ICC as follows: values less than 0.50 are considered poor reliability, 0.50 to 0.75 are considered moderate reliability, 0.75 to 0.90 is considered good reliability and ICC's greater than 0.90 is considered excellent reliability

Inter-rater reliability of the DDK task was conducted for the following measures: Mean DDK Rate, Accuracy of First Production, Accuracy of Five Productions and Consistency. The interpretation of kappa, after Landis and Koch (1977) is as follows:

- <0.20 Poor
- 0.21-0.40 Fair
- 0.41-0.60 Moderate
- 0.61-0.80 Good
- 0.81-1.00 Very good

4.6.2.2.1 Mean DDK Rate

Intra-rater reliability for the mean DDK rate was calculated by comparison of two trials of the primary rater's final mean calculation. There was good reliability R= 0.944.

Inter-rater reliability for the mean DDK rate was calculated by comparison of both raters' final mean calculation. This required agreement of the raters' initial phonetics transcriptions of the DDK targets and correct calculation of the rate of each target. Cohen's κ was run to determine if there was agreement between two raters' judgement on mean DDK rate. There was very good agreement between the two raters' judgements, $\kappa = .897$, $p < .0001$.

4.6.2.2.2 Accuracy of First Production

Intra-rater reliability for the accuracy of the first production was calculated by comparison of two trials of the primary rater's judgement. There was good reliability R= 0.975. For the inter-rater reliability, the scoring of the accuracy of the first production of the DDK targets was compared for the two raters using Cohen's Kappa. The results of this interrater analysis are $\kappa = -0.013$ with $p < 0.872$ (Landis & Koch, 1977).

While this appears to be an unsuccessful result, the statistics have led to what is known as "the kappa paradox" (Bexkens et al., 2018). Bexkens et al. (2018) report that while a study may report a high absolute percentage of observer agreement, at the same time a low kappa value is produced. They report that this statistical phenomenon, the first kappa paradox, is "the effect that prevalence of the subject under study in a data set has on marginal values." This feature causes an imbalance in case distribution which would produce lower kappa value. However, they state that this paradox is not a limitation but correctly interprets agreement adjusted for agreement by chance alone. Therefore, it can still be assumed that there was high inter-rater reliability between the raters, despite the kappa paradox shown in the statistics.

4.6.2.2.3 Accuracy of five productions

Intra-rater reliability for the accuracy of the five productions was calculated by comparison of two trials of the primary rater's judgement. There was good reliability R= 0.927. For the inter-rater reliability, the scoring of the accuracy of the five productions of the DDK targets was compared for the two raters using Cohen's Kappa. There was good agreement $\kappa = 0.750$ with $p < 0.001$. This showed substantial inter-rater agreement for this measure (Landis & Koch, 1977).

4.6.2.2.4 Consistency

Intra-rater reliability for the accuracy of the five productions was calculated by comparison of two trials of the primary rater's judgement. There was good reliability R= 0.932. For the inter-rater reliability, the scoring of the consistency production of the DDK targets was compared for the two raters using Cohen's Kappa. There was high agreement with $\kappa = 0.847$ with $p < 0.001$. This showed high inter-rater reliability for this measure (Landis & Koch, 1977).

4.7 Instrumental Results

This section covers the results from the analysis of the maximum performance task, the diadochokinesis (DDK) as well as the results from the measures taken from the instrumental analysis using ultrasound tongue imaging. This specifically looked at differences in tongue shape variance as measured from the DDK task at syllable and sequence levels. Finally, the results of the mean syllable duration measure are discussed in relation to the two groups.

4.8 Diadochokinesis Task

Articulate Assistant Advanced software (Articulate Instruments Ltd, 2019) was used to annotate the acoustic data and from this the DDK rates expressed as the number of syllables per second was calculated for each child. The child listened to the model of five syllable/sequences produced by the recorded voice and was asked to repeat. These five rates were taken from the protocol developed by McCann and Wrench

(2007). Five repetitions of single syllables (pa, ta, ka) and sequences (pataka) were recorded at six set rates.

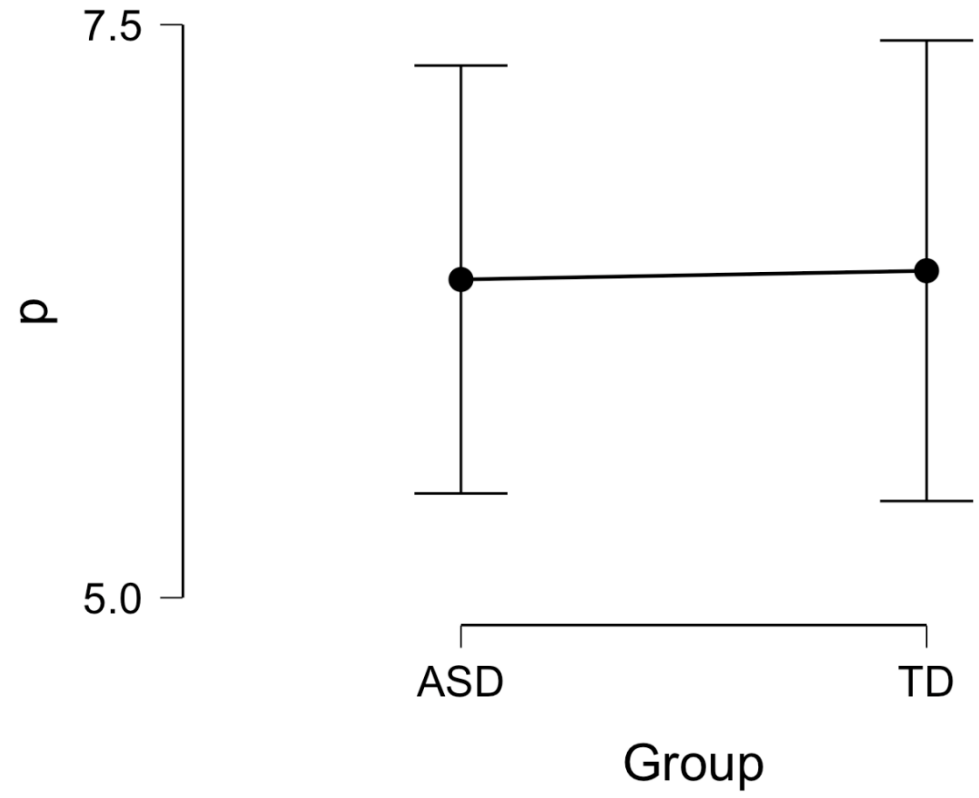
The perceptual measures that were taken from this data focused on the following parameters:

- Rate
    - Maximum rate of production compared across groups
    - Maximum rate of production compared to standardized norms developed by Fletcher (1978).
- Accuracy
    - Accuracy of a first single repetition of the target compared to the adult model
    - Accuracy of five repetitions of the target compared to the adult model
- Consistency
    - Consistency of five repetitions compared to the child's baseline production

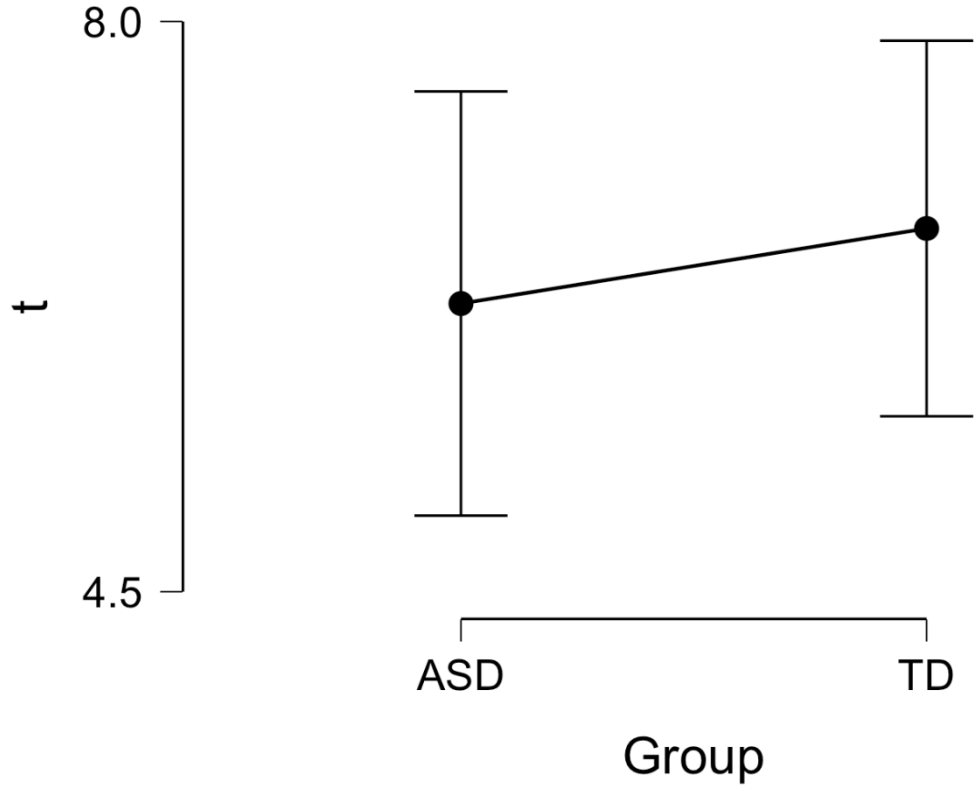
4.8.1 Rate

Maximum rate of production is the fastest speed a child produced the target syllable (p, t, k) or sequence (tk and ptk) in syllables per second (s/s). Figure 17 lays out the descriptive statistics (mean) for both groups at the five target categories.

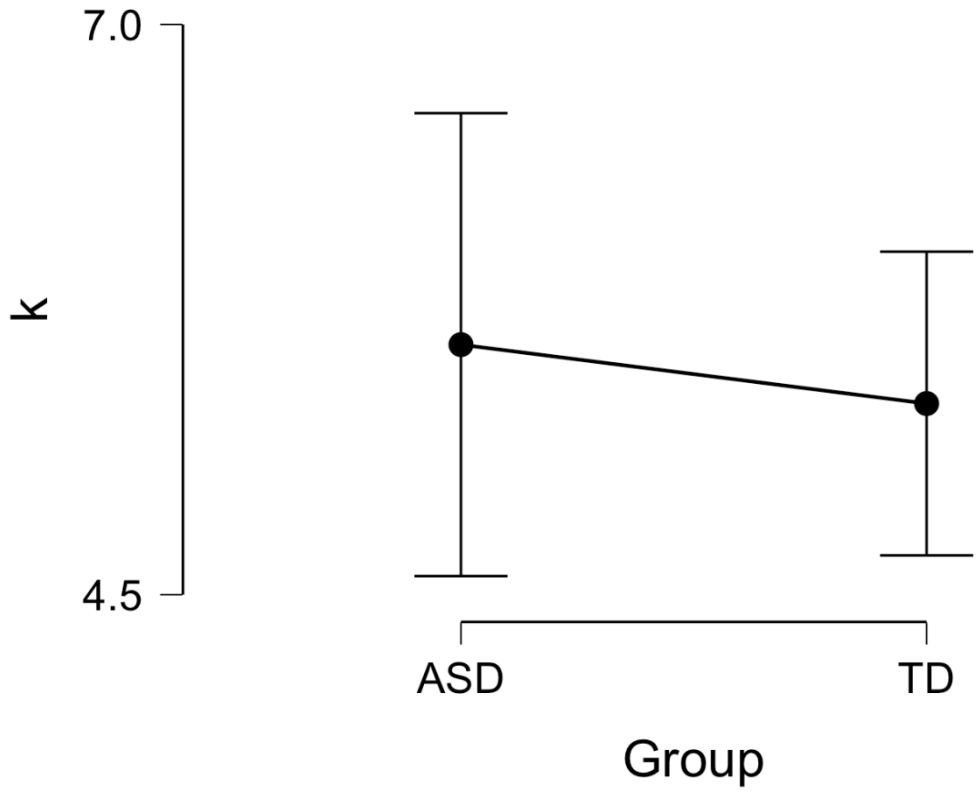
Figure 17*: Descriptive Plots of Mean of maximum rate of production (s/s) for TD and ASD groups*



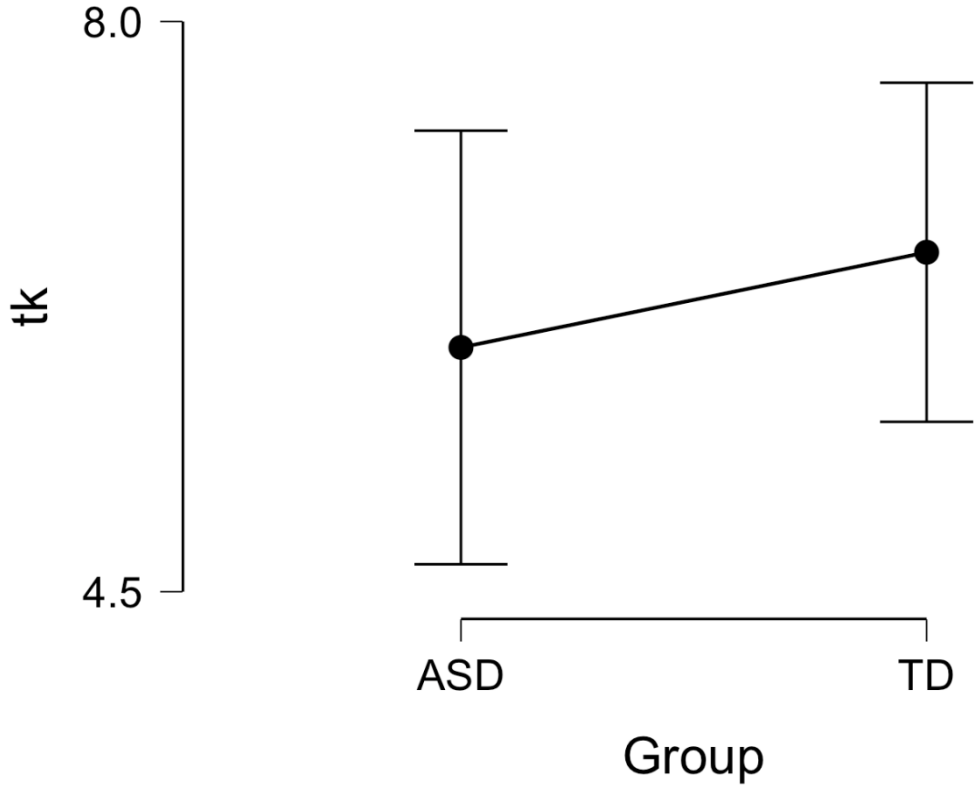
t



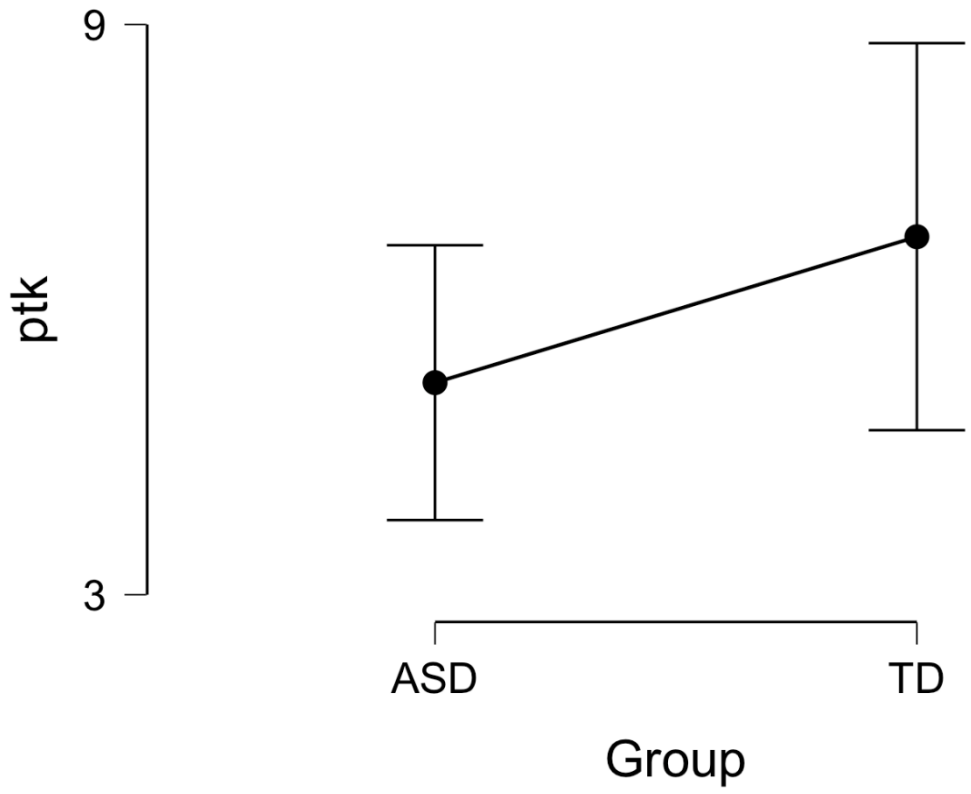
k



tk



ptk



Results:

Maximum rate of production compared across groups

An independent samples t-test was conducted, to compare the means of the two groups in order to determine if there is a significant difference between the mean of the maximum rate of syllables per second. A Levene's test for equal variance was conducted and found for all variables there was equal variance in scores within the two groups.

There was no significant difference between maximum rates of DDK for any of the syllables, or sequences of syllables, as shown in the table 19.

Table 19: *Maximum Rates of DDK for ASD and TD Group*

Target	Group	Mean (s/s)	Std. Dev	p value	BF_{10}	Cohen's d	Shapiro-Wilks (p-value)
	TD	6.43	1.31	0.95	0.43	-0.032	0.31
/p/				Bonferroni			0.16
	ASD	5.72	1.82	= 0.01			
	TD	6.73	1.5	0.32	0.49	-0.315	0.11
/t/				Bonferroni			0.19
	ASD	5.96	1.58	= 0.01			
	TD	5.34	0.87	0.76	0.47	0.266	0.88
/k/				Bonferroni			0.57
	ASD	5.49	1.06	= 0.01			
	TD	6.58	1.35	0.24	0.54	-0.42	0.61
/tk/				Bonferroni			0.56
	ASD	5.73	1.53	= 0.01			
	TD	6.77	2.65	0.15	0.78	-0.68	0.40
/ptk/				Bonferroni			0.17
	ASD	5.14	1.47	= 0.01			

4.8.2 Comparison to Norm

The maximum rate of DDK repetitions produced by the children were compared to the norms produced by Fletcher (1978) who calculated them from syllable repetitions from 384 school-age children using the time-by count method (Table 14). However, as this study used the count-by-time method, both the means and standard deviations had to be converted from the original norms to this measurement.

Results:

Tables 20 and 21 show whether the children in the ASD and TD group were within the norms for each age group according to Fletcher (1978). These are colour coded

as blue is faster than one SD above the mean, green is within the norm, orange is slower than minus one SD and red is slower than minus two SDs. Child 4M from ASD group did not participate in the DDK exercises so no data is available for this participant. Child 10F from the ASD only participated in the first target (/p/) so only this measurement is available for this participant. There were more instances of children in the ASD group being below or significantly below the norm than the TD group.

Table 20: *Maximum rate of participants in ASD group in comparison to the norm.*

Participant	Group	Age (years and months)	Target (s/s)				
			p	t	k	tk	ptk
01M	ASD	12;08	5.04	5.26	5.10	6.44	5.92
02F	ASD	10;07	8.36	9.24	7.42	6.42	5.92
03M	ASD	6;04	5.91	5.86	4.14	8.34	7.43
05M	ASD	12;06	6.20	5.80	5.23	5.82	5.16
06M	ASD	10;09	6.09	5.27	4.84	5.38	4.86
07F	ASD	10;11	6.63	5.65	6.39	3.52	2.29
08M	ASD	10;05	6.49	6.81	6.06	6.07	5.03
09M	ASD	7;06	5.13	3.77	4.71	3.87	4.52
10F	ASD	8;10	1.60				

Note. Blue cells indicate faster than the norm, green indicates within the norm and red is slower than the norm. Where cells are blank, child did not participate in exercise, so results were not collected.

Table 21: *Maximum rate of participants in TD group in comparison to the norm.*

Target (s/s)

Participant	Group	Age (years and months)	p	t	k	tk	ptk
01F	TD	9.08	6.20	5.80	5.23	5.82	5.16
02M	TD	11.40	6.64	9.07	6.03	7.08	6.05
03F	TD	7.08	9.19	9.07	4.77	8.34	13.14
04M	TD	7.08	6.49	6.81	6.06	6.07	5.03
05F	TD	6.08	4.16	4.44	4.26	4.14	7.73
06F	TD	8.50	6.94	6.65	5.93	5.81	4.07
08M	TD	6.06	5.91	5.86	4.14	8.34	7.43
09M	TD	9.05	6.43	6.50	6.61	6.25	6.15
10M	TD	12.06	5.89	6.36	5.01	7.40	6.15

Note. Blue cells indicate faster than the norm, green indicates within the norm and red is slower than the norm

A chi square test showed that there was no significant association between the groups on whether the DDK was above or below the mean, X^2 (1, N=17) = .944, p=.331.

4.8.3 Accuracy

Accuracy of first repetition across groups

A paired samples t-test was conducted to compare the accuracy of all participants in single syllable and sequence conditions. There was a significant difference in the scores for single syllable accuracy (M=99.02, SD=2.94) and sequence accuracy (M=87.17, SD=15.58) conditions; t (16)= 3.35, p=0.004* (Bonferroni = 0.01). This indicates that participants produced the single syllables with more accuracy than the sequences on the first repetition.

Accuracy of first repetition between groups

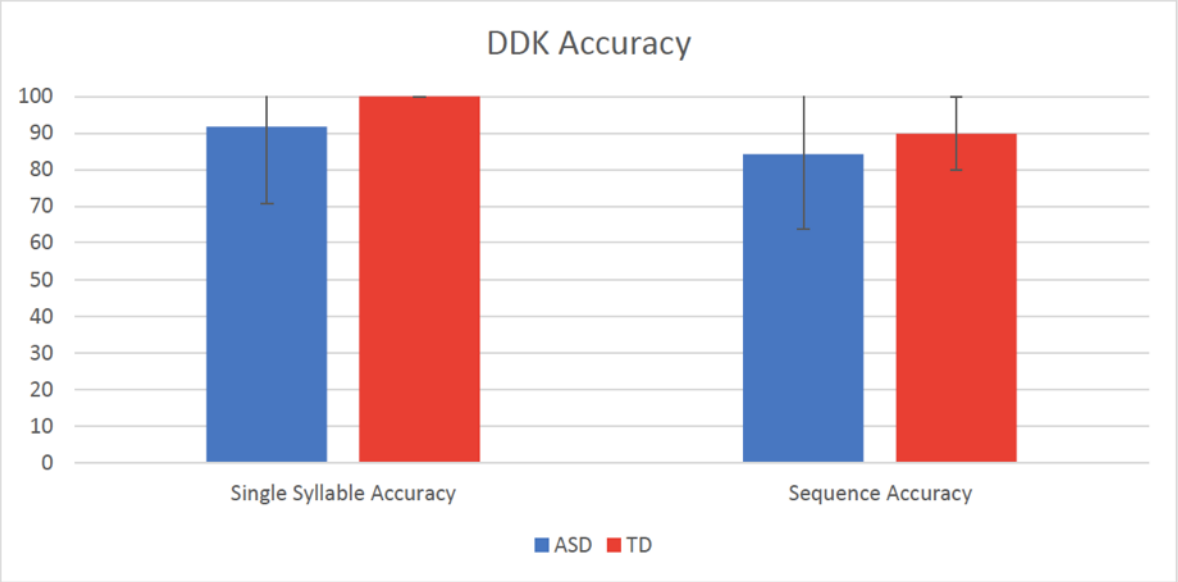
a) An independent sample t-test was conducted to compare the accuracy of the first production of **single syllables** of participants in the ASD group and participants in the TD group.

There was not a significant difference in the scores for the ASD group (M=91.67, SD=20.83) and the TD group (M=100, SD=0) conditions t (17) =-1.12, p=0.25. (Bonferroni = 0.01) (Shapiro-Wilks p= 0.18) $BF_{10}= 0.56$. Cohen's effect size value (d=1.11) showed a large mean difference between the two groups. There was no statistical significance but with a larger sample size there may have been one found indicated by the effect size. There was a ceiling effect within the TD group in this task.

b) An independent sample t-test was conducted to compare the accuracy of the first production of **sequences** of participants in the ASD group and participants in the TD group.

There was not a significant difference in the scores for the ASD group (M=84.19, SD=20.51) and the TD group (M=89.81, SD=10.02) conditions t (15) = -0.73, p=0.48 (Bonferroni = 0.01) (Shapiro-Wilks p= 0.57). BF_{10}= -1.06. Cohen's effect size value (d=-0.82 showed a large mean difference between the two groups. There was no difference in the accuracy of the first repetition of sequences between the two groups (figure 18).

Figure 18: *Comparison between ASD and TD group of accuracy of first production of syllables and sequences*



In summary, there was no significant difference between the ASD group and TD group in the accuracy of their first production or syllables and sequences. However, there was a significant difference in the first production of single syllables and sequences across both groups, children were less accurate in their first production of sequences. There was also a ceiling effect for the TD group where all the children achieved full accuracy in the tasks.

4.8.4 Consistency

My measure of consistency was taken from the William and Stackhouse (2009) method which was the consistency of five repetitions compared to the child's baseline production (the first syllable/sequence produced).

I. An independent sample t-test was conducted to compare the consistency of the production of **single syllables** of participants in the ASD group and participants in the TD group.

> There was not a significant difference in the scores for the ASD group (M=69.04, SD=19.65) and the TD group (M=85.58, SD=17.43) conditions t (15) = 1.84, p=0.08 (Bonferroni = 0.01 (Shapiro-Wilks p = 0.12). BF_{10}= 1.24. Cohen's effect size value (d=-0.89) showed a large mean difference between the two groups.

This indicates that there was no difference in the consistency of the single syllables between the two groups.

II. An independent sample t-test was conducted to compare the consistency of the production of **sequences** of participants in the ASD group and participants in the TD group.

There was not a significant difference in the scores for the ASD group (M=66.02 SD=18.6) and the TD group (M=70.37, SD=21.29) conditions t (14) = 0.43, p=0.68 (Bonferroni = 0.01) (Shapiro-Wilks p= 0.66). $BF_{10}= 0.46$. Cohen's effect size value (d=0.22) showed a small mean difference between the two groups. This indicates that there was no difference in the consistency of sequences between the two groups.

III. An independent sample t-test was conducted to compare the consistency of the production **overall (both conditions)** of participants in the ASD group and participants in the TD group.

There was not a significant difference in the scores for the ASD group (M=69.07, SD=20.08) and the TD group (M=79.46, SD=16.64) conditions t (15) =1.17, p=0.26, (Bonferroni = 0.01 (Shapiro-Wilks p= 0.35)), $BF_{10}= 0.52$. Cohen's effect size value (d=-0.37) showed a small mean difference between the two groups. This indicates that there was no difference in the consistency the overall production between the two groups.

In summary, there was no significant difference between the ASD group and TD group in the consistency of their production of syllables and sequences.

4.9 Ultrasound Tongue Shape Analysis

To measure differences in tongue shape and speech sound production at a more detailed level than allowed with speech sounds tests typically used in clinic, ultrasound analysis of the tongue shape and movement was conducted as the participants carried out the DDK task.

4.9.1 T-Tests of Tongue Shape Variance

This study used ultrasound to observe if there were sub phonemic differences in articulatory movements of children with ASD compared to TD children. The articulatory differences I was interested in observing needed to be related to speech motor control to help distinguish why there may be higher rates of SSEs in children with ASD and if the root cause was a speech motor problem. Ultrasound allows direct observation of tongue shape and consistency in the production of tongue shapes across the same repetitions is an indicator of speech motor control (Zharkova, Hewlett and Hardcastle, 2011). Therefore, using the DDK task, which elicits repetitions of the same target sound, provided a measure of speech motor control that could be observed using ultrasound. I was interested in observing whether children with ASD had a larger amount of variation in their tongue shapes than TD children. In the Articulate Assistant Advanced software (Articulate Instruments Ltd, 2019) an inbuilt t-test allows significance of difference between tongue shapes along each fan line.

Furthermore, I made this comparison with the slowest production (-3SD) of each DDK target (e.g., /p/) and the fastest (+2SD). If children with ASD had difficulty in "tuning in" to their speech as posited by Shriberg et al. (2011), then the speed and accuracy at which they process phonological information may be reduced. Observing DDK tasks at the slowest and fastest speeds allows observation of whether increase motoric complexity of faster speeds, impacts the accuracy of the child's production, indicating difficulty at the motoric and/or linguistic processing stages (Preston and Edwards, 2009).

The protocol used in this study was developed from Cleland (2015) where the threshold for reporting and estimating the size of significant difference between the tongue shape curves was more stringent than finding just a single significant difference. This protocol requires a minimum of six adjacent significant t-test results ($p=>0.05$) over a contiguous region of the tongue surface. This ensures that I do not claim any significant difference based on only a few significant t-tests from a small area of the tongue surface. As found in this study, it is possible for a pair of two different tongue shapes to have multiple defined regions of significant difference. As

shown in figure 16, areas where there are multiple areas of significance are indicated by green lines.

The area of high confidence and statistical significance of tongue shape variation is indicated in figure 16. The area of low confidence is where automatic configuration of where the tongue line was not clear enough to be traced and therefore not valid, any area of significance indicated in the area of low confidence is therefore ignored. The front of the tongue (right hand side of figure 16) where the constriction for /t/ is there is no difference whereas the tongue body is in a different position at each rate. This is very like a co-articulatory effect, i.e., the child employs a different vowel strategy.

Table 22: *P Values of differences between slowest and fastest tongue shape curves*

Group	Participant	p single	p -ptk segment	t single	t - tk segment	t - ptk segment	k single	k - tk segment	k - ptk segment
TD	TD1F	0.001	0.800	0.272	0.011	0.019	0.365	0.010	0.779
	TD2M		0.281	0.031	0.004	0.057	0.346	0.667	0.369
	TD3F		0.357	0.018	0.265		0.031	0.305	0.013
	TD4M	0.024	0.310	0.008	0.008	0.018	0.457	0.035	0.012
	TD5F	0.013	0.597	0.017	0.454	0.531	0.018	0.511	0.440
	TD6F	0.611	0.029	0.394	0.670	0.042	0.007	0.468	0.759
	TD7M								
	TD8M	0.291	0.375	0.391	0.231	0.731	0.133	0.650	0.270
	TD9M	0.001	0.030	0.015	0.466	0.006	0.620	0.443	0.004
	TD10M	0.016	0.008	0.418	0.565	0.392	0.010	0.551	0.352

	ASD1F			0.350				
	ASD2F	0.371	0.016	0.012		0.646	0.015	0.509
	ASD3M	0.023	0.331	0.005	0.017	0.442	0.529	0.568	0.443
	ASD5M	0.012	0.598	0.010	0.523	0.625	0.018	0.541	0.435
ASD	ASD6M	0.011	0.026	0.035	0.018	0.310	0.006	0.227	0.392
	ASD7F	0.200	0.018	0.005	0.010	0.397	0.008	0.002	0.448
	ASD8M	0.017	0.021	0.521	0.487	0.008	0.026	0.575	0.042
	ASD9M	0.023	0.379	0.029	0.514	0.027	0.746	0.685	0.477
	ASD10F								

Note. Green is an indicator of a significant p-value, white cells are an indicator of a non-significant p-value. Grey blank cells indicate where there was no data available.

Table 22 shows the results from the inbuilt t-test that compared the shape of the tongue during lingual movements at the slowest and fastest productions of the target. Where an area of significance was presence (six or more adjacent fan lines), the mean of these p-values was taken. If there was a significant difference in the tongue shape, this is highlighted in green. At the p single target, 75% of children in both groups produced a significant result, indicating a significant difference of tongue shape production for both groups at this level. For targets p in the /ptk/ segment, t single, t in the /tk/ segment and k single, the ASD group had more significant p values than the TD group. Where what is expected to be the motorically complex sequences in /tk/ segment the ptk segment, the TD group had more significant differences of tongue shape at the slowest and fastest production of /k/ (seven occurrences) than the ASD group (three occurrences). Overall, the TD group had

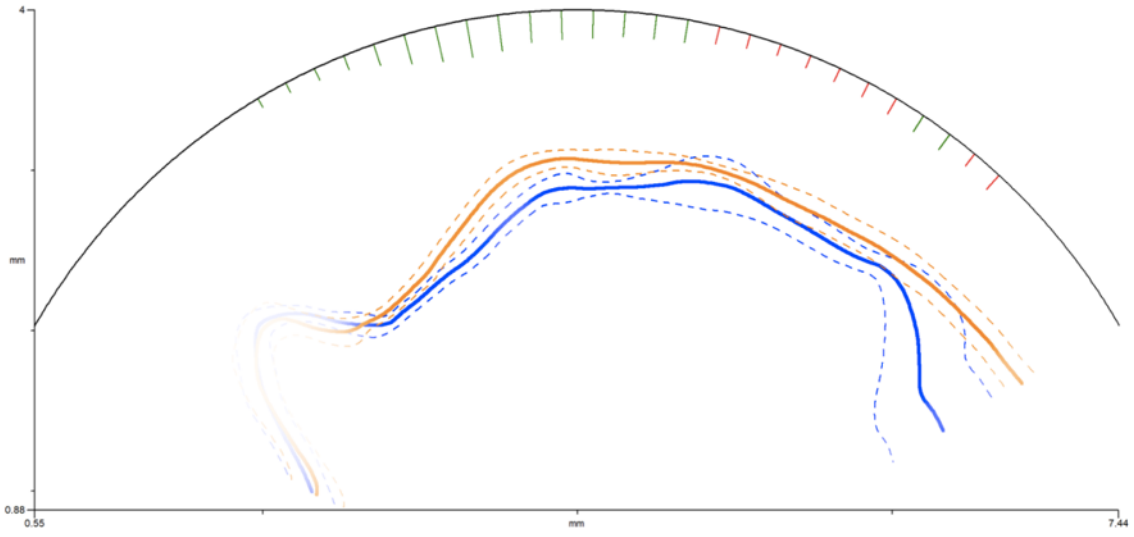
47% of the DDK targets being significantly difference at the slowest and fastest production and the ASD had slightly more at 52%.

4.9.2 Qualitative Analysis of /p/ slowest and fastest

As described above, at the p single target, 75% of children in both groups produced a significant result, indicating a significant difference of mean tongue shape production for both groups at this level. Looking at the individual speech profiles of both groups in this context helps answer RQ2, whether the ultrasound reveals subtle articulatory differences not remarked at the perceptual level of assessment. Five out of ten of the TD children (01F, 04M, 05F, 09M and 10M) and six out of ten of the children with ASD (03M, 05M, 06M, 08M, 09M and 10F) all presented with significant differences between their mean slowest and fasts /p/ tongue shapes. Therefore, each child's tongues shape comparison is given below, given qualitative judgments about tongue shape and then group summaries are made.

4.9.2.1 TD Group

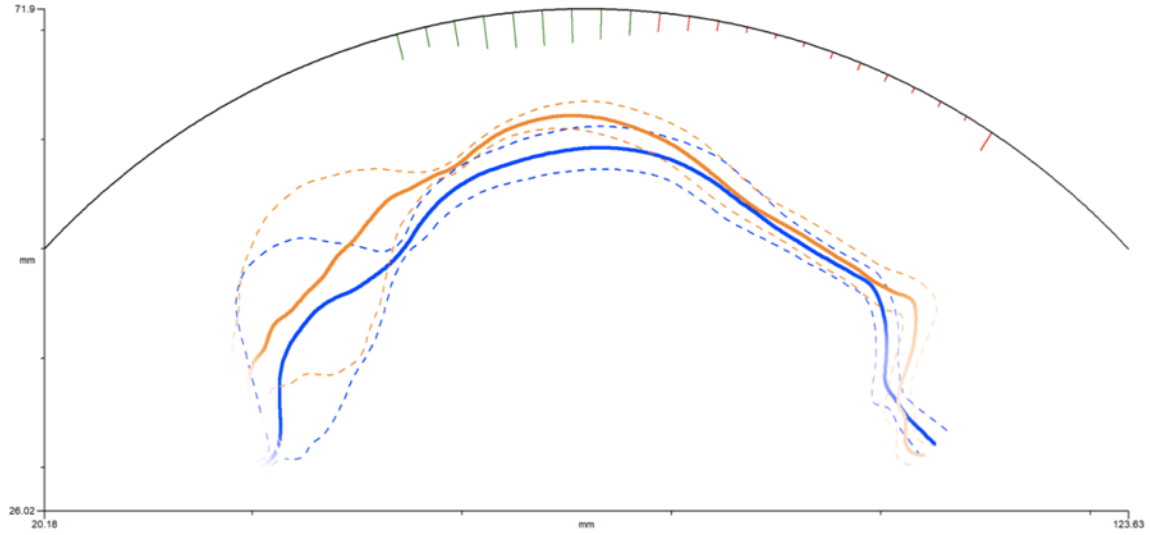
Figure 19: *TD1F tongue shape comparison of mean /p/ slowest and fastest production*



Note. Blue line represents slowest production and orange fastest. Full line represents mean tongue shape and dashed line represents standard deviations. Green lines along outer curve indicates areas of significant difference.

01F shows a significant difference in tongue shape curve at the tongue body. The significance at the tongue root and tongue tip are not dependable due to difficult in imaging the tongue accurately at these areas. At the faster production, the tongue body is higher in the mouth than the slower. There is slightly more variation in the front part of the tongue body during the slower production.

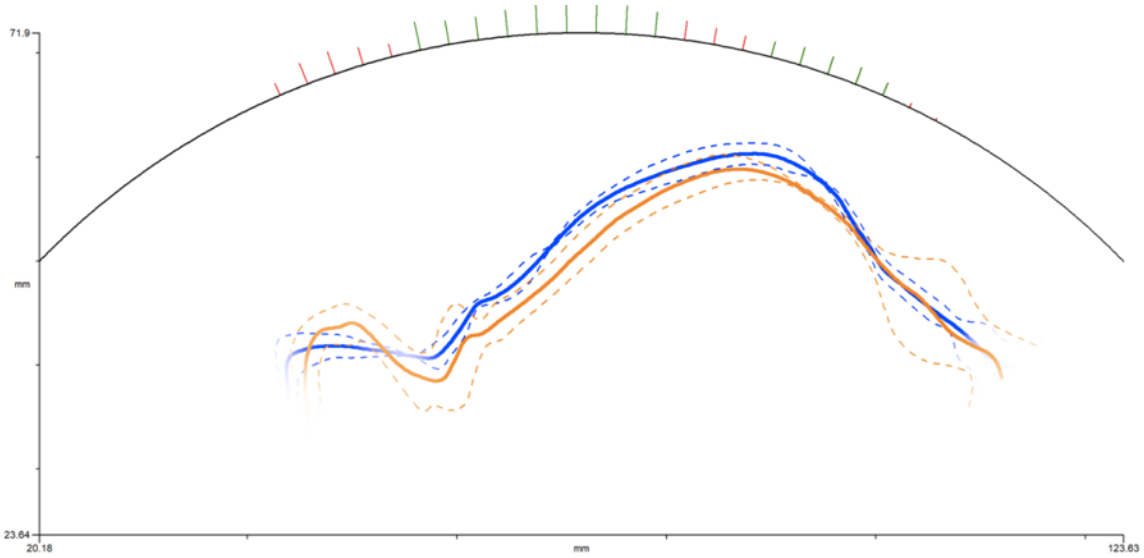
Figure 20: *TD4M tongue shape comparison of mean /p/ slowest and fastest production*



Note. Blue line represents slowest production and orange fastest. Full line represents mean tongue shape and dashed line represents standard deviations. Green lines along outer curve indicates areas of significant difference.

04M shows significantly more variation in tongue shape compared to 01F indicated by the dashed standard deviation lines. The large differences at the root and tip of the tongue are ignored as this was not accurately tracked by the software due to this region being difficult to image with ultrasound. The notable significant difference here is in the mid part of the tongue body between the fastest and slowest. Interestingly, in 04M's case, the faster speed is higher in the mouth than the slower, unlike 01F. Furthermore, there is more variation in tongue shape present at the slower speed than the faster. At the front of the tongue body there is an area overlapping where the tongue shape is the same, which is possibly due to a braced tongue at the inside of the molars.

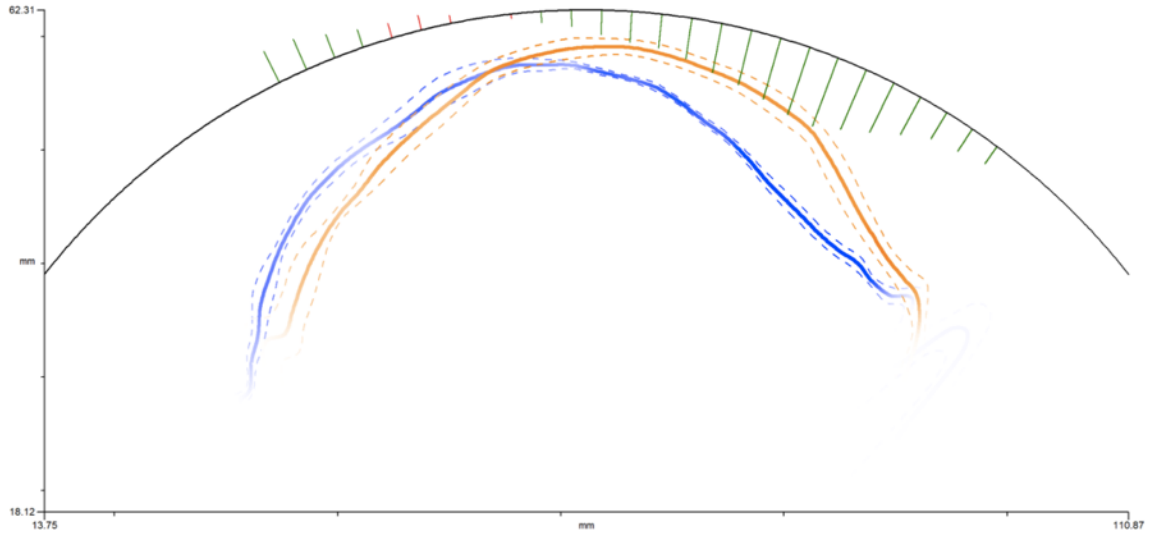
Figure 21: *TD5F tongue shape comparison of mean /p/ slowest and fastest production*



Note. Blue line represents slowest production and orange fastest. Full line represents mean tongue shape and dashed line represents standard deviations. Green lines along outer curve indicates areas of significant difference.

05F has a higher peak in the tongue body than previously discussed participants. The area of most variation is at the back of the tongue body. The tongue is lower in the mouth at the fastest production. The fastest production shows more variation in tongue shape across repetitions, shows in the standard deviations, than the slower production. Again, there is overlap at the front of the tongue which may be due to a braced tongue behind the molars.

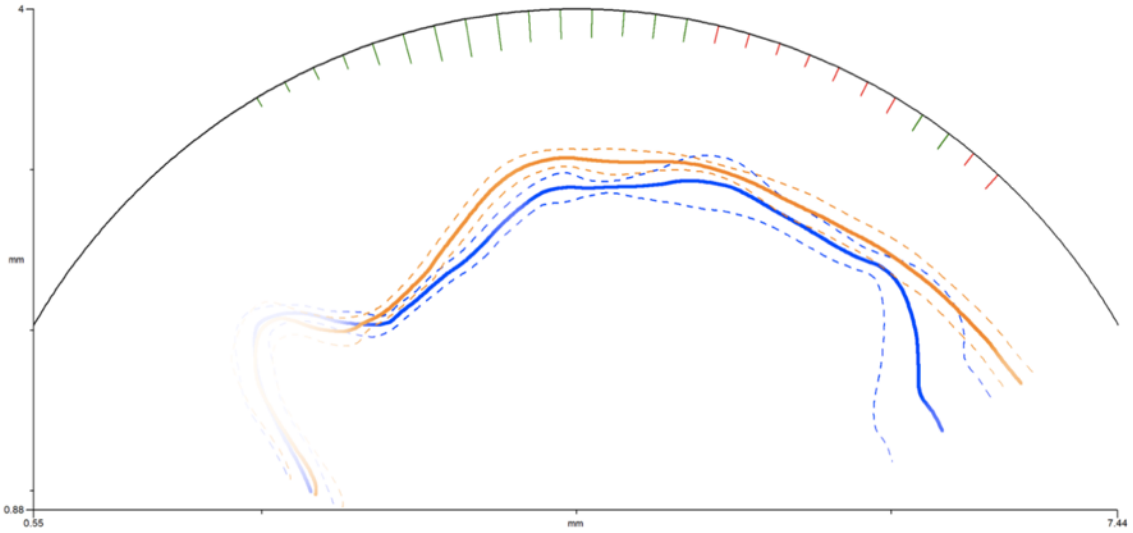
Figure 22: *TD9M tongue shape comparison of mean /p/ slowest and fastest production*



Note. Blue line represents slowest production and orange fastest. Full line represents mean tongue shape and dashed line represents standard deviations. Green lines along outer curve indicates areas of significant difference.

09M shows a significant difference in the general tongue shape at the slower and faster mean productions of /p/. The tongue if further forward on the mouth, towards the lips, in the faster production than the slower which is further back in the mouth. The faster production is made higher in the mouth than the slower. Both speeds show similar amount of variation in the standard deviations.

Figure 23: *TD10M tongue shape comparison of mean /p/ slowest and fastest production*

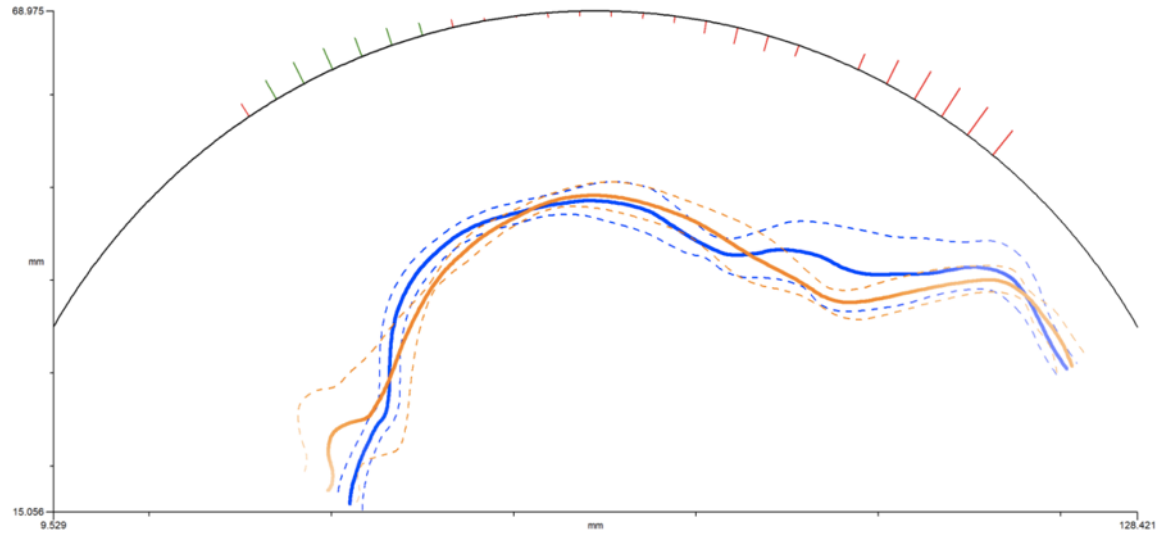


Note. Blue line represents slowest production and orange fastest. Full line represents mean tongue shape and dashed line represents standard deviations. Green lines along outer curve indicates areas of significant difference.

The front of the tongue is excluded from this analysis as it has not been accurately tracked by the software due to difficulty imaging the tongue with ultrasound. Interestingly, the slower production shows more variation than the faster, this may be due to imaging difficulties. The significant difference between these tongue shapes is that faster production is higher in the mouth than the slower.

4.9.2.2 ASD Group

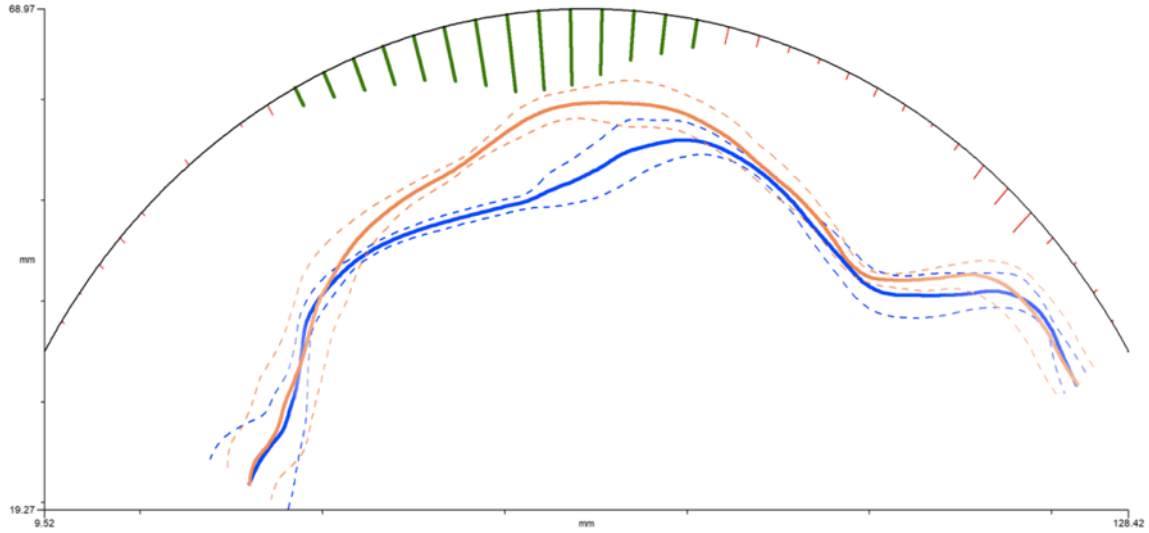
Figure 24: *ASD3M tongue shape comparison of mean /p/ slowest and fastest production*



Note. Blue line represents slowest production and orange fastest. Full line represents mean tongue shape and dashed line represents standard deviations. Green lines along outer curve indicates areas of significant difference.

The front of the tongue is excluded from this analysis as it has not been accurately tracked by the software due to difficulty imaging the tongue with ultrasound. The significant difference of interest occurs at the back of the tongue body where the slower production is slightly higher in the mouth than the faster. There is significant variation between tongue shape curves at both speeds, indicated by the dashes standard deviation lines which show large gaps from the mean line. However, compared to the TD group these tongue shapes are more similar.

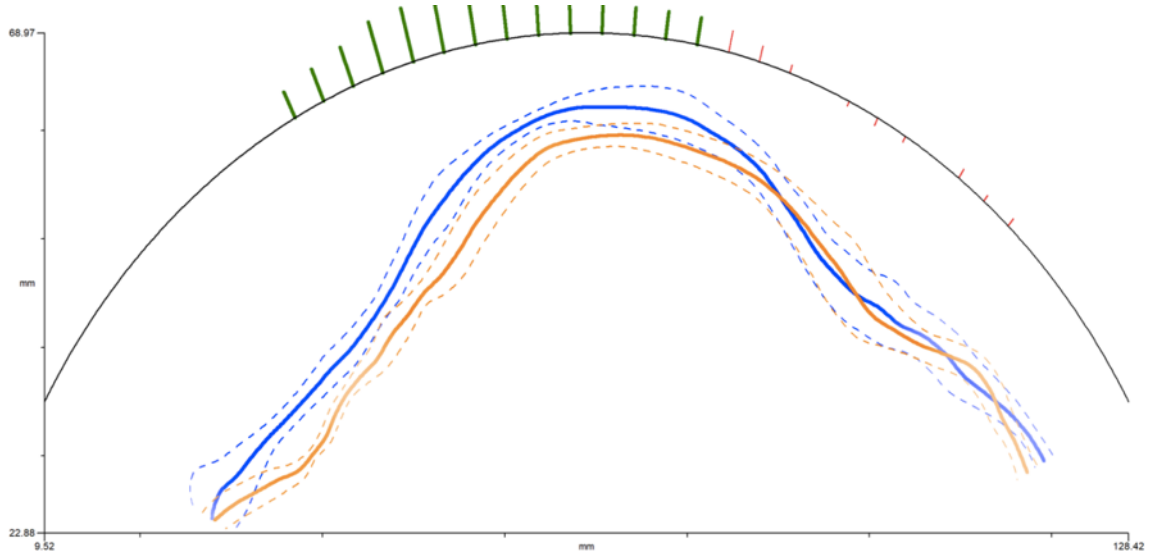
Figure 25: *ASD5M tongue shape comparison of mean /p/ slowest and fastest production*



Note. Blue line represents slowest production and orange fastest. Full line represents mean tongue shape and dashed line represents standard deviations. Green lines along outer curve indicates areas of significant difference.

Similar to the TD children, 05M shows a brace of the tongue at the front of the mouth and little variation at the front part of the tongue body. The tongue tip data is ignored as it was not traced effectively. The main variation occurs in the middle of the tongue body where the faster repetition is higher in the mouth.

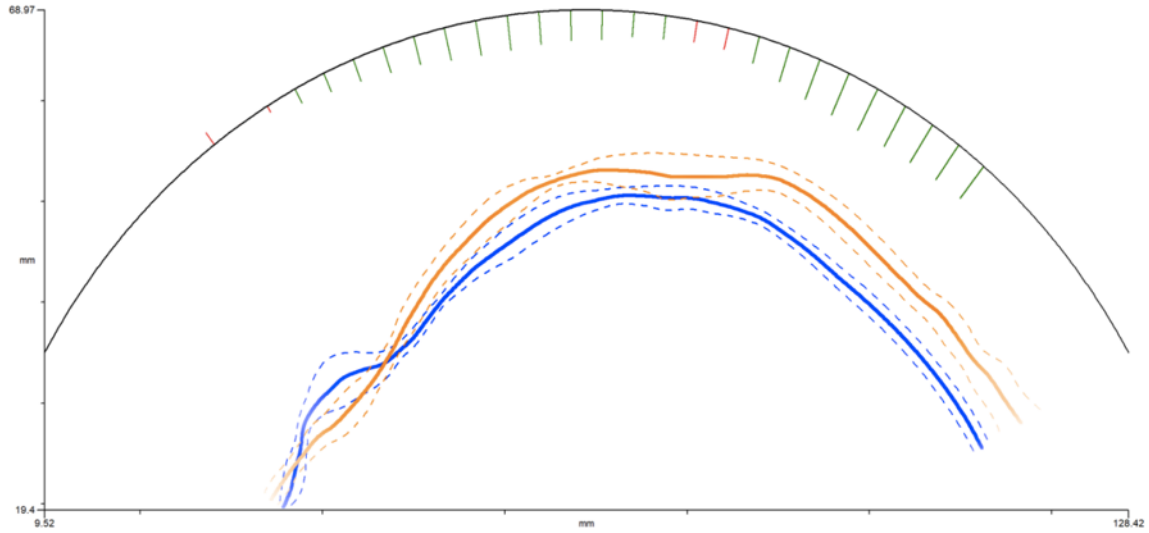
Figure 26: *ASD6M tongue shape comparison of mean /p/ slowest and fastest production*



Note. Blue line represents slowest production and orange fastest. Full line represents mean tongue shape and dashed line represents standard deviations. Green lines along outer curve indicates areas of significant difference.

The tongue shapes produced at both speeds are very similar in shape with the slowest production recorded as higher in the mouth. This may be due to a shift in the ultrasound headset and not necessarily the child's production. There is some variation present at both speeds as indicated by the standard deviation lines.

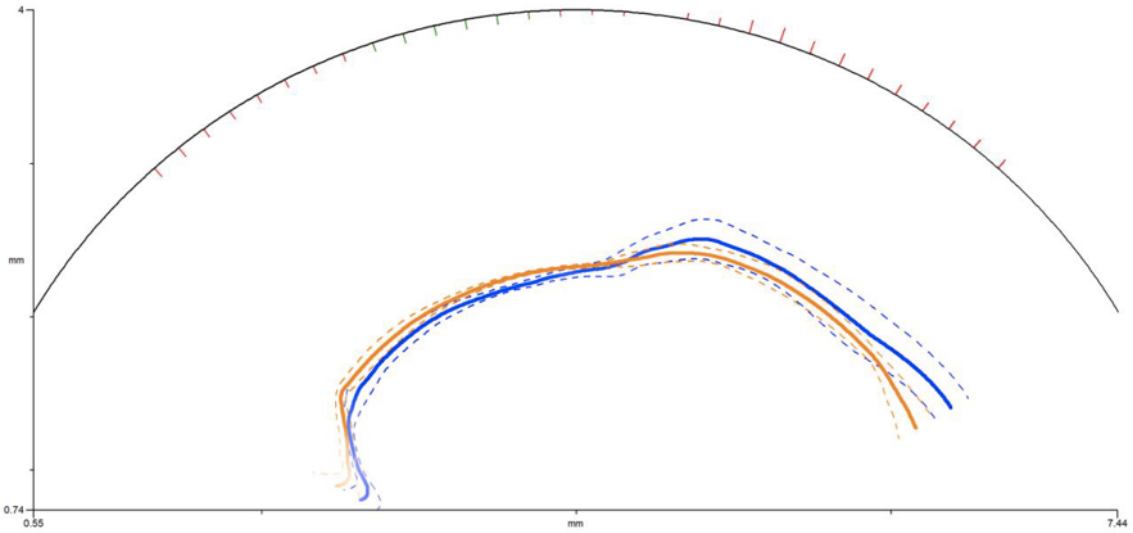
Figure 27: *ASD8M tongue shape comparison of mean /p/ slowest and fastest production*



Note. Blue line represents slowest production and orange fastest. Full line represents mean tongue shape and dashed line represents standard deviations. Green lines along outer curve indicates areas of significant difference.

08M displays very similar tongue shapes at both speeds, except the faster production is higher in the mouth than the slower. There are similar amounts of variation across both speeds. The lack of variation of 08M compared to the TD group is notable.

Figure 28: *ASD9M tongue shape comparison of mean /p/ slowest and fastest production*



Note. Blue line represents slowest production and orange fastest. Full line represents mean tongue shape and dashed line represents standard deviations. Green lines along outer curve indicates areas of significant difference.

The most notable characteristics of 09M's production is the lack of variability between the slowest and fastest curves. The tongue tip data should be ignored as this is not accurately traced using ultrasound.

4.9.2.3 Summary

What these qualitative judgements of the tongue shapes for both groups show us is that there is a larger variation in tongue shape in the TD group than the ASD group. The TD children show a wide-ranging profile from tongue bracing behind the molars (04M and 05F) to shifting tongue positions within the mouth and wide variation in tongue profiles of each child. The variation seen in both groups could be accounted for in the effect of different vocal tract morphologies, for instance, different palate shapes, individual to each child (Mugitani and Hiroya, 2012). What is notable about the ASD group is they had markedly less variation in tongue shape at the slower and faster speeds, almost identical for some of the children (09M). The children with ASD have shown to have functionally effective speech motor control and appear to use alternative motor control strategies than the TD children, with significantly more

rigidity in their tongue movement across difference speeds. In particular, reduced coarticulation and bracing.

4.9.3 Equality of Variance of Tongue Shapes p-values

A Levene's test was used to assess the equality of variances of the p-values (8 for each participant) for the TD and ASD group. A Levene's test is an inferential statistic used to assess the equality of variances for a variable (in this case the p-values of the comparison of the slowest and fastest productions of a target sound), calculated for two or more groups. It tests the null hypothesis that the population variances are equal. In terms of articulatory data this means that is there was a significant Levene’s statistic, there is potentially one group with more significantly different tongue shapes than the other.

As reported in table 23, there was no significant Levene’s statistic, therefore population variances of the p-values are equal for both groups. As indicated in table 23, both groups had equal variance of p-values, so I accept the null hypothesis for all sound targets that there is equal variance of significant p-values across both groups, there was no group that had a larger variance in p-values than the other.

Table 23: *Levene’s Test of Homogeneity of Variances for Both Groups*

Target	Levene Statistic	df1	df2	Sig.
p single syllable	1.39	1.00	14.00	0.26
p in ptk segment	0.01	1.00	14.00	0.94
t single syllable	1.79	1.00	15.00	0.20
t in tk segment	0.44	1.00	13.00	0.52
t in ptk segment	0.44	1.00	13.00	0.52
k single syllable	0.84	1.00	14.00	0.38
k in tk segment	0.09	1.00	13.00	0.76
k in ptk segment	2.91	1.00	14.00	0.11

4.9.4 Equality of Variance of Tongue Shape Standard Deviations (SD)

The standard deviations of the tongue shapes quantify how much the participants within a group differ from the mean value for the group. The standard deviation was created from an average of five repetitions. Therefore, the standard deviation of these five repetitions is a measure of articulatory stability. In this case it shows how much children within the TD or ASD group differ from the mean of the shapes at the slowest and fastest sound productions. Table 24 shows these standard deviations. In order to determine whether there was an equal or unequal variance across the two groups in their standard deviations from the mean, a comparison of standard deviations was carried out (table 25).

Table 24: *Mean p-value derived from T-Tests of Tongue Shape Variance*

Group	Participant	p single	p -ptk segment	t single	t - tk segment	t - ptk segment	k single	k - tk segment	k - ptk segment
TD	01F	0.001	0.800	0.272	0.011	0.019	0.365	0.010	0.779
	02M	0.001	0.281	0.031	0.004	0.057	0.346	0.667	0.369
	03F	0.001	0.357	0.018	0.265		0.031	0.305	0.013
	04M	0.024	0.310	0.008	0.008	0.018	0.457	0.035	0.012
	05F	0.013	0.597	0.017	0.454	0.531	0.018	0.511	0.440
	06F	0.611	0.029	0.394	0.670	0.042	0.007	0.468	0.759
	07M								
	08M	0.291	0.375	0.391	0.231	0.731	0.133	0.650	0.270
	09M	0.001	0.030	0.015	0.466	0.006	0.620	0.443	0.004
	10M	0.016	0.008	0.418	0.565	0.392	0.010	0.551	0.352
ASD	01F	0.013		0.350					
	02F	0.371	0.016	0.012		0.646	0.015		0.509
	03M	0.023	0.331	0.005	0.017	0.442	0.529	0.568	0.443
	05M	0.012	0.598	0.010	0.523	0.625	0.018	0.541	0.435
	06M	0.011	0.026	0.035	0.018	0.310	0.006	0.227	0.392
	07M	0.200	0.018	0.005	0.010	0.397	0.008	0.002	0.448
	08M	0.017	0.021	0.521	0.487	0.008	0.026	0.575	0.042
	09M	0.023	0.379	0.029	0.514	0.027	0.746	0.685	0.477

Table 25: *Standard Deviations of Tongue Shapes at Slowest and Fastest Productions*

		p single slowest	p single fastest	p -ptk segment slowest	p -ptk segment fastest	t single slowest	t single fastest	t - tk segment slowest	t - tk segment fastest	t - ptk segment slowest	t - ptk segment fastest	k single slowest	k single fastest	k - tk segment slowest	k - tk segment fastest	k - ptk segment slowest	k - ptk segment fastest
TD	01F	0.01	0.03	0.05	0.06	0.04	0.04	0.05	0.05	0.03	0.02	0.05	0.05	0.03	0.06	0.04	0.04
	02M	0.05	0.05	0.04	0.05	0.05	0.03	0.04	0.04	0.04	0.04	0.06	0.04	0.07	0.06	0.04	0.03
	03F	1.57	0.58	1.76	1.99	0.99	1.00	0.92	1.98	1.93	1.76	1.20	1.27	1.62	1.39	2.22	1.04
	04M	2.71	2.64	1.11	2.04	1.77	0.68	2.70	1.27	1.32	1.63	1.44	2.07	2.93	1.76	2.49	1.50
	05F	0.93	2.01	2.42	2.60	1.53	1.13	2.01	1.94	1.84	1.57	1.91	2.83	3.33	5.27	2.09	1.97
	06F	2.35	2.09	1.63	1.64	1.52	1.21	2.05	1.21	1.54	1.08	1.94	1.29	3.05	1.49	2.35	2.09
	08M	1.63	1.64	1.56	2.18	1.27	2.57	0.96	2.48	1.58	2.92	1.27	3.90	0.93	2.88	1.68	1.91
	09M	0.44	1.01	1.99	2.27	1.00	1.51	1.29	1.09	1.25	0.87	0.83	0.44	0.51	0.88	1.39	1.02
	10M	0.11	0.07	0.05	0.07	0.07	0.09	0.16	0.11	0.08	0.10	0.06	0.11	0.13	0.11	0.06	0.18
ASD	01F	0.04	0.04			0.12	0.11	0.04				0.09	0.14				0.12
	02F	0.71	0.58	0.47	0.71	0.63	0.59			0.71	1.20	0.59	1.16			1.05	0.54
	03M	2.15	2.16	2.04	2.06	1.20	1.38	1.00	1.21	1.63	1.15	1.62	2.19	1.50	1.79	1.93	1.08
	05M	1.49	1.82	1.97	2.18	1.98	1.33	1.99	3.23	1.75	1.53	1.90	2.83	3.29	5.38	1.88	2.00

06 M	1.73	2.18	1.64	1.69	1.52	1.65	1.92	1.16	1.21	2.85	3.60	1.66	2.28	1.31	1.68	2.07
07 F	2.21	1.62	2.37	1.33	1.45	1.94	1.96	1.11	1.71	2.25	1.40	1.71	2.02	1.14	2.21	2.06
08 M	1.13	1.43	1.10	1.53	1.17	1.52	1.25	0.87	0.90	1.51	1.17	1.26	0.88	1.43	1.14	1.60
09 M	0.08	0.04	0.07	0.09	0.08	0.09	0.11	0.11	0.09	0.09	0.09	0.15	0.10	0.13	0.10	0.12
10 F		0.07														

4.9.5 Difference in Standard Deviations at Sound Target Level

A paired samples t-test was carried out to test whether the standard deviations of tongue shape at the slowest and fastest production of sound targets are significantly different in both the TD group (table 26) and the ASD group (table 27). A significant difference means there is a larger standard deviation in the faster rate, therefore there is less motor control of the tongue.

Table 26: *Paired Samples T-Test*

	Target	t	df	Sig. (2-tailed)	Bonferroni	BF_{10}	Cohen's d	Shapiro-Wilk (p-value)
Pair 1	p single slowest - p single fastest	-0.19	8.00	0.86	0.006	0.26*	-0.08	0.20
Pair 2	p ptk segment slowest - p ptk segment fastest	-2.35	8.00	0.05*	0.006	0.55	-0.34	0.06
Pair 3	t single slowest - t single fastest	-0.01	8.00	0.99	0.006	0.25*	-0.06	0.20
Pair 4	t tk segment slowest - t tk segment fastest	0.00	8.00	1.00	0.006	0.26*	0.04	0.32
Pair 5	t ptk segment slowest - t ptk segment fastest	-0.23	8.00	0.82	0.006	0.47	-0.30	0.28
Pair 6	k single slowest - k single fastest	-1.11	8.00	0.30	0.006	0.40	-0.25	0.32

Pair 7	k tk segment slowest - k tk segment fastest	-0.36	8.00	0.73	0.006	0.30	-0.14	0.16
Pair 8	k ptk segment slowest - k ptk segment fastest	1.74	8.00	0.12	0.006	0.74	0.40	0.23

Whilst the majority of the paired sample t-tests for the sound targets in the TD group carried out were not significant, there was one significant result produced. There was a significant difference in the standard deviations of /p/ in the slowest ptk segment (M=1;18 SD=0.92) and fastest ptk segment (M=1.43, SD=1.06) conditions; t (8) =-2.35, p = 0.05. This means that for the TD group, there was a significant difference in the standard deviation from the mean in the slowest and fastest production of the /p/ in the /ptk/ segment. However, this did not survive the Bonferroni correction and needs to be carried out with a significantly larger group. From an articulatory perspective this means that for the TD group there could have been less motor control of the tongue at the faster production of /p/.

4.10 Mean Syllable Duration

To assess whether there was a difference in the rhythm of sounds produced in each target group, the mean duration for each slowest and fastest target syllable was calculated and analysed. Table 27 is a record of the mean duration of syllable produced by each participant, a syllable was defined from the burst of the plosive until the end of the following vowel.

Table 27: *Mean duration of slowest and fastest target sounds (milliseconds)*

Group	Participant	p single slowest	p single fastest	t single slowest	t single fastest	k single slowest	k single fastest	tk segment slowest	tk segment fastest	ptk segment slowest	ptk segment fastest
TD	01F	46	34	53	40	48	59	37	27	32	33
	02M	67	31	64	34	120	50	53	24	38	32
	03F	61	23	46	31	55	30	59	24	22	25
	04M	18	33	83	29	58	38	51	31	41	33
	05F	71	51	65	42	83	62	71	37	60	61
	06F	84	30	58	41	83	56	78	39	59	50
	07M	16	14	31	35	31	33	23	29	22	24
	08M	43	90	49	24	58	58	33	33	26	25

	09M	17	18	40	28	26	24	25	31	22	20
	10M	17	24	72	47	79	54	31	50	25	23
Mean		43	34	56	34	64	46	46	32	34	32
SD		25	21	15	7	27	13	19	7	14	12
	01F	28	19	51	41	48	61	34	28	57	49
	02F	22	12	83	15	68	24	-	-	51	32
	03M	16	21	56	19	43	50	30	26	32	29
	05M	20	16	53	31	68	26	25	35	26	26
ASD	06M	12	15	78	51	39	51	49	42	38	38
	07F	-	-	-	-	-	-	-	-	-	-
	08M	11	18	32	40	50	49	40	36	29	34
	09M	17	19	37	35	37	27	20	33	25	42
	10F	37	25	65	-	-	-	-	-	-	-
Mean		20	18	56	33	50	41	32	33	36	35
SD		8	3	18	12	12	14	10	5	12	7

4.10.1 Equality of Variance

A Levene's test was used to assess the equality of variances of the standard deviations of durations for the TD and ASD group, the results are reported in table 28. A low standard deviation would mean there is consistent rhythm, and a high standard deviation would mean there are irregular syllable lengths.

Table 28: *Test of Homogeneity of Variances of Mean Syllable Durations of Slowest and Fastest DDK productions*

	Levene's Statistic	df1	df2	Sig.
p single slowest	0.22	1.00	16.00	0.64
p single fastest	2.01	1.00	16.00	0.17
t single slowest	0.14	1.00	16.00	0.70
t single fastest	0.24	1.00	15.00	0.63
k single slowest	2.01	1.00	15.00	0.17
k single fastest	1.53	1.00	15.00	0.23
tk segment slowest	0.11	1.00	14.00	0.74
tk segment fastest	4.09	1.00	14.00	0.06
ptk segment slowest	0.75	1.00	15.00	0.39
ptk segment fastest	5.77	1.00	15.00	0.03*

Whilst most of the DDK productions were equal in variances across the two groups, the Levene's test indicated unequal variances in the following conditions: ptk segment fastest (F=5.77, 0.03*). This means that the TD group had more variation in the duration of the ptk segment that the ASD group. This was an unexpected finding as it indicates the ASD had more articulatory stability in this case than the TD group.

4.10.2 Independent Samples T-Test of Mean Syllable Durations

An independent samples t-test was used to determine whether there are any statistically significant differences between the means of ASD and TD groups as displayed in table 29.

Table 29: *Independent Samples T-test of Mean Syllable Durations of Slowest and Fastest DDK productions*

Target	Group	Mean (s/s)	Std. Dev	p value	Bonferroni	BF_{10}	Cohen's d	Shapiro-Wilks (p-value)
p single slowest	ASD	0.007	0.008	0.29	0.005	0.65	-0.52	0.30
	TD	0.012	0.010					0.09
p single fastest	ASD	0.007	0.007	0.18	0.005	0.79	-0.66	0.20
	TD	0.018	0.20					0.10
t single slowest	ASD	0.019	0.012	0.71	0.005	0.43	0.18	0.47
	TD	0.017	0.012					0.25
t single fastest	ASD	0.011	0.006	0.90	0.005	0.43	0.06	0.06
	TD	0.011	0.005					0.81
k single slowest	ASD	0.013	0.006	0.46	0.005	0.51	-0.37	0.43
	TD	0.023	0.036					0.10
k single fastest	ASD	0.012	0.007	0.36	0.005	0.57	-0.46	0.25
	TD	0.017	0.011					0.39
tk segment slowest	ASD	0.014	0.007	0.94	0.005	0.43	0.04	0.58
	TD	0.014	0.007					0.32
tk segment fastest	ASD	0.011	0.007	0.45	0.005	0.52	0.39	0.24
	TD	0.009	0.004					0.40
ptk segment slowest	ASD	0.016	0.007	0.38	0.005	0.56	0.45	0.50
	TD	0.013	0.006					0.19
ptk segment fastest	ASD	0.017	0.007	0.03*	0.005	2.34	1.17	0.84
	TD	0.011	0.003					0.70

There was a significant effect of the mean duration of sound productions at the $p<0.5$ level for the ptk fastest segment in the ASD (M=0.017, SD=0.007) and the TD group (M=0.011, SD=0.003) conditions t (16) = 2.37, p=0.03 $BF_{10}= 2.34$. Cohen's effect size value (d=1.17) showed a large mean difference between the two groups. However, it did not survive the Bonferroni correction.

4.11 Summary of Results

This section covered the results from the analysis of the DDK task and the results from the instrumental analysis of the tongue using ultrasound tongue imaging. With the DDK results I found there was no significant difference in the rate of production between the ASD and TD groups. There were more instances of children in the ASD group being below or significantly below the norm in the rate of DDK productions, but

this was not statistically significant. Furthermore, there was no significant difference in the accuracy of production between the ASD and TD groups. Additionally, there was no significant difference in the consistency of production between the ASD and TD groups.

The results from the ultrasound tongue imaging revealed that with the measure of tongue shape variance, the TD group had more significant difference of tongue shape in the more motorically complex segments (ptk) than the ASD group. When looking at the variation within the two groups, there was equal variance of standard deviations from the mean in both groups meaning that there was similar amount of variance of tongue shape variation across both the groups. There was a significant difference in the SD from the mean of the slowest and fastest production of /p/ in the /ptk/ segment for the TD group but none for the ASD group.

Finally, for the measure of mean duration of syllables there was unequal variance for the /ptk/ segment fastest productions across the two groups in which the TD group showed more variance in their mean syllable durations in these this sound target than the TD group. Moreover, there was a significant difference of mean duration for the same sound target (/ptk/ fastest) across the two groups from the independent samples t-test carried out in which the TD showed slower and more varied production of /p/ which may point to an over precise and restrictive rhythm of speech sound production in the ASD group.

4.12 Instrumental Results Discussion

4.12.1 Timing of Speech

Timing is an essential part of speech production, and it was surprising not to find a difference in maximum rate between the groups in relation to this, though this could also be due to a small sample size. Speech executed with fluency requires information to be chosen, sequenced, and produced in an exact and time sensitive way. A lot of semi quasi-autonomous articulatory frameworks need to work in coordination (Thoonen *et al.*, 1996; Kotz and Schwartze, 2016). While little research has been done on speech timing, current literature suggests there might be

irregularities in sensorimotor timing in children with ASD in general movement. While this may not affect the functionality of speech, this may lead to differences noted in speech patterns. This was found in my sample in which the ASD had less variation in their speech than the TD group, particularly in the motorically complex segments.

Similar to this study, Anzulewics, Sobota and Delafield-Butt (2016) found an alternative movement timing pattern in children with ASD performed faster swipe gestures when they tapped the screen, and they did so with more force and significantly shorter contact time that control children. Torres, et al. (2013) found an increase in the acceleration-deceleration phases of a reach-to-touch task in children with ASD. These tasks showed a subtle, yet critical disturbance to moment-by-moment control of movement happening in the region of 30–70 ms, a temporal domain significant for speech. However, there was no impact found in the rate of DDK production on the group of children with ASD that I worked with. This may be due to the fact the movements of speech articulators are controlled differently dependent on the purpose and goal of the motor task. In this instance speech motor control may not necessarily show the same impairments or altered movements patterns than general movement abilities.

Over-and under-compensations of such quick movements in force are thought to support the motor interruptions commonly found in ASD (Trevarthen & Delafield-Butt, 2013). The lack of variation in tongue shape identified in the ASD group, may be a result of overcompensation. These compensations may influence speech perception resulting in disturbed speech production because of the lack of coordination within articulatory systems (Trevarthen & Delafield-Butt, 2017). Cook, Blakemore, and Press (2013) discovered sub-second control of velocity and acceleration was influenced in people with ASD in straightforward arm-swing assignments. This examination showed that quick timing at the sub-second level required of speech motor control may be impacted in limb and hand movements in people with ASD. However, my findings do not support this as a speech level, suggesting there may be a level of independence between the speech motor control system and the general motor control system (Ziegler, 2003), however this would need to be explored with a large sample size.

4.12.2 Impact of Social Factors

While the literature tends to agree that motorically, speech rate in ASD is not significantly different from their TD peers, when other social demands are added, an impairment may occur. In a study carried out by Patel et al. (2020), they found a slower speech rate among individuals with ASD compared to TD controls. This was elicited from a 24-page wordless picture book used to elicit spontaneous narratives. The added cognitive challenge of interpreting stories from a picture book may have resulted in the slower rate. In this study there was no social or narrative demand as the child repeated back an audio model of the target repetitive sound. Wynn et al. (2018) carried out a study that observed speech rate during conversation entrainment, which is a phenomenon where speech behaviour is modified by the speaker in order to match their communication partner and it is key to a successful conversation. Wynn et al. (2018) suggested that when SSEs occur it is breakdown in entrainment that is being exhibited in the speech production of individuals with ASD. There are similarities with the speech attunement framework proposed by Shriberg et al. (2011) in which children with ASD do not “tune in” or “tune up” to their ambient speech environment. Wynn et al. (2018) studied speech rate entrainment in 60 participants, comprising of four experimental groups: (a) children with ASD (n = 15), TD children (n = 15), adults with ASD (n = 15), and typically developed adults (n = 15). In a quasi-conversation paradigm, the TD adults entrained their speech using a slower rate during slow speech and a faster rate during fast rate. This was not observed with the three other groups, suggesting that children and individuals with ASD do not entrain their speech rate. While it can’t be generalized due to small sample size, this study combined with my results and previous studies imply that is not necessarily an impairment in speech motor control and indicates that it is the social communication differences that plays a core role in speech production abnormalities in ASD. It provides further support for the speech attunement framework (Shriberg et al. 2011). This is further confirmed in a study carried out by Chenausky et al. (2017) who studied vocalization rate in non-speech and speech like tasks in three groups: 18 toddlers at low risk for ASD, 18 high-risk siblings without ASD and 18 high-risk siblings with ASD. They found no significant difference between the groups in the non-speech like vocalizations but the group of high-risk toddlers with ASD had slower speech like vocalizations than the control groups. It appears that rate of speech is not impacted at the motoric level in ASD but is when other social or communicative factors are included in the processing and production.

This may explain why I found no difference in DDK production. The DDK task used in this study was non-lexical (have no meaning attached to the sound), whereas the DEAP (Dodd, 2002) the words have lexical meaning. Taking a psychologistic approach, when there is no lexical meaning attached, then speech production process can pass from phonological recognition to motor programming (Stackhouse and Wells, 2001). In the ASD group, there appears to be no breakdown at this stage. Whereas when lexical meaning is attached, the speech production process requires passing through phonological representation, and often semantic representation before reaching motor programming. In the ASD group, it appears a breakdown may occur there due to the errors found in the DEAP but not the DDK task. This indicates that there may be a breakdown at the phonological planning stage rather than at the motor programming stage.

4.12.3 Insights from Neuroimaging

When looking to neuroimaging to tell us why these differences may or may not be occurring, there is a sparsity of literature as most neuroimaging investigation of communication has focused on language comprehension (Dichter, 2012; Mody and Belliveau, 2012). Studies such as Pang et al. (2016) looked at speech production in groups of children with ASD using magnetoencephalographic (MEG) to examine speech perception and found auditory processing delays that could have accounted for communication differences and difficulty attuning into their ambient speech environment. Additionally, Pang et al. (2016) used MEG with twenty-one children with high functioning ASD and twenty-one typically developing controls to look at their brain functioning during a phoneme production task, phoneme sequencing task and an oromotor task. The interesting use of MEG is that it allows examination of brain dynamics underlying speech as it can capture fast responses fundamental to the timing of speech. In the simple oromotor task they found delayed latency and an increased magnitude in the executive control area. Delayed latency is often interpreted as evidence that a task was more difficult and required more time to complete. When increased magnitudes are found it is often interpreted as a process that require more synchronized brain activity, perhaps due to a need for increased effort to complete the task (Pang et al. 2016). While these difficulties were found at the brain functioning level, this was not evident in my findings in the practice of the

oromotor task. This tells us that perhaps there could have been differences found in the DDK task if they were more challenging. Maybe the current study's group had a ceiling effect, or that within ASD there are subtypes of children with communication impairments differing in presentation and root causes.

The neural correlates underlying motor differences in ASD are relatively unknown (Mostofsky et al., 2009). However, there have been some studies that suggest dysfunction of the cerebellum may play a part in this motor disorder. The functional domains that the cerebellum serve are remarkably diverse (Courchesne and Allen, 1997). Post-mortem brain samples of patients with ASD have shown to have cerebellar alterations as one of the most replicated findings across studies (Jaber, 2017). In addition, the cerebellum which has traditionally been associated with motor control is not seen as an important structure for social circuitry, making it particularly relevant when investigate causal roots of motor dysfunction in ASD (Schmahmann and Caplan, 2006). The role of the cerebellum in ASD is not well understood and the literature can often be conflicting. Mostofsky et al. (2009) found in thirteen children with high functioning ASD that they demonstrated less activation in the ipsilateral anterior cerebellum using functional magnetic resonance imaging (fMRA) during a sequential, appositional finger tapping task. While Oldehinkel et al. (2019) carried out fMRA with 265 individuals with ASD, significantly larger than Mostofysky's study, and compared them to 218 typically developing (TD) individuals, comparing functional connectivity within twenty networks. They found increased connectivity of the cerebellum with sensory and motor networks in ASD compared with TD subjects.

However, this sample showed a lack of speech motor control-like errors in the DDK tasks, which does not particularly match the cerebellum alterations theories. Jochaut et al. (2015) used electroencephalogram (EEG) and fMRI with thirteen individuals with ASD and thirteen typically developing individuals. They found activity in the left auditory cortex failed to track speech modulations and down-regulate gamma oscillations in the ASD group but not the TD group. They found altered oscillation-based connectivity between auditory and other language cortices in the ASD group. So, the SSEs observed in ASD could be associated with an altered balance of slow and fast auditory oscillations. This could compromise mapping between sensory input and high-level cognitive representations and processes, such a motor control. This also fits the speech attunement framework in which altered processes affect the

child's ability to "tune up" and "tune in" to their ambient environment (Shriberg et al., 2011). While there is no clear conclusion on why correlation between balance and PCC of the DEAP occurred, this area is worth significant research, including neuroimaging techniques alongside speech and motor examination in order to understand how these are impacted and in relation to what brain function and structure.

One of the few studies to look at motor skills and speech delay in individuals with ASD was Barbeau (2015) who compared individuals with ASD who had speech delay (n=21), individuals with ASD without speech delay (n=18) and typically developing individuals (n=30). They found that while both subgroups of ASD showed elements of motor impairment, the subgroup of ASD without speech delay showed significantly faster reaction times, but significantly slower fine motor skills and performed poorer on bimanual coordination tasks than those with speech delay. This implies that the subgroup without speech delay may have had intact motor execution but have difficulty incorporating perceptual information during more complex fine motor tasks. This idea is in line with a review by Gowen and Hamilton (2013b), suggesting that movement atypicality in ASD are related to poor integration of information for efficient motor planning, and increased variability in basic sensory inputs and motor outputs and not to movement execution mechanisms.

There is a general consensus that the cerebellum is involved motor implementation through the construction of internal models to serve motor behaviour (Ito & Schuman, 2008; Leggio et al., 2011; Pisotta & Molinari, 2014). However, it the past decades, emerging evidence also points that the cerebellum plays a significant role in social functioning. Van Overwalle, Ma and Heleven (2020) carried out a meta-analysis that identified more than 200 fMRI studies researching the cerebellum's role in social mentalizing and emotion self-expression. Through this meta-analysis they accumulated evidence that suggests the cerebellum supports social cognition, particularly in relation to mentalizing, which refers to social understanding by directly observing human bodily motion. Social cognition is the process if perceiving and interpreting theses behaviour of self and others (Amodio & Frith, 2006) and evidence suggests that that the posterior cerebellum supports social cognition (Van Overwalle et al., 2014, 2015). The meta-analysis of Van Overwalle et al., (2020) identified over 200 fMRI studies found the bilateral Crus II areas of the cerebellum related to

‘sequencing’ during mentalizing and mere social ‘mentalizing’ or self-related emotional cognition located in the cerebellar mentalizing network. This supports the idea that the cerebellum has a significant role in social functioning, specifically in relation to mentalizing.

Neuroanatomical differences in the cerebellum are a consistent finding in individuals with ASD but little is known about the connection to the core symptoms (D’Mello et al., 2015). For example, D’Mello et al., (2015) studied the cerebellar grey matter and lobular volume of the cerebellum in 35 children with ASD and 35 TD children and found there were significant correlations between their scores on the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000) and their lobular volume. Additionally, reductions in regional and lobular grey matter in the cerebellum correlated with the severity of difference in social interaction, communication a repetitive behaviour. Furthermore, Olivito et al., (2018) found grey matter reduction correlated with the degree of ASD symptoms as measured by the autism-spectrum quotient. What these studies tell us is that it is likely the differences in the cerebellum observed in individuals with ASD likely has a significant outcome of the expression of ASD symptoms, including those related to social cognition. One possibility its distortions in mentalizing which is known to cause anomalies in social and emotion functioning. The posterior cerebellum has been found to serve the mentalizing network (Buckner et al., 2011) however significant more research is required to understand this in relation to presentation of ASD symptoms.

4.12.4 Ultrasound Tongue Shape Analysis

To measure differences in tongue shape and speech sound production at a more detailed level than allowed with speech sounds assessments typically used in clinic, ultrasound analysis of the tongue shape and movement was carried out. This allowed determination of whether subtle SSEs were identified using instrumental analysis in the speech of children with ASD, as stipulated in RQ2. Ultrasound tongue imaging analysis (ultrasound) was used to illustrate and quantify differences in tongue shape at the slowest and fasted production of each target DDK production. This allowed to carry out different statistical tests to observe whether there were differences in variations of tongue shapes produced by children in the ASD group and TD group.

4.12.5 Variability of Tongue Shape

An independent samples t-test was carried out to observe the variation of tongue shape produced at the slowest and fastest of a DDK target across the two groups. Consistency of tongue shape at differing speeds is a measure of motor control (Zharkova et al., 2011). The literature base has provided evidence that healthy mature speakers are able to produce highly consistent movements, whereas young children, aging adults, and impaired talkers produce movement that is comparatively inconsistent and used consistency as a measure of speech motor control (Green et al., 2002; Grigos, 2009; Grigos et al., 2015; Murdoch et al., 2012). However, some speakers with speech motor impairments have shown to have overly consistent speech (Mefferd, 2016), implying that impairment-related changes in movement pattern consistency can diverge from a normal level in both directions.

Within both groups of children had significant differences between their slowest and fastest speech sound targets. For example, for the /ptk/ segment target 75% of the ASD group and 75% of the TD group had a significant difference between the slowest and fastest production in which there was a larger variation of tongue shape at the fastest production compared to the slowest. There was individual variation across the TD group as shown in the tongue shape comparisons of the certain children in chapter five. The difference of tongue shape variation between targets seen between the slowest and fastest production in the TD group is likely due to a speed-accuracy trade-off where some children had wider variation in the faster productions than the slower. The motor actions required for speech are high dexterous and rapid but often a speech and accuracy trade off can occur (Lammert *et al.*, 2016) which explains why there was larger variation in the tongue shapes of /p/ between the slowest and fastest production for both groups. Variability of speech tokens like those I tested in the DDK production task have been found to be a measure of speech motor control maturity (Barbier *et al.*, 2013). Token-to-token variability has been found to be larger in children than adults. Maturation of speech motor control is a long developmental process which has been found to only reach maturity in late adolescence (Walsh and Smith, 2002; Smith, 2010). This likely explains why I found significant differences within the TD group; they are within the age range where their speech motor control has not fully matured yet. However,

there was a notable lack of variation in the ASD group, where their tongue shape was very similar across both the slowest and fastest repetitions.

What is surprising is that there was no difference between the TD and ASD group, despite the ASD group having significant general motor impairment found in the behavioural assessment results using the movement assessment battery for children (MABC-2; Brown & Lalor, 2009). When looking at individual targets of the single syllables and sequences, there is very little difference between the number of participants in each group who had significant variance of tongue shapes at the slowest and fastest production. This was confirmed in the Levene's Test for Equality of Variance which was revealed no significant results so I can conclude that there was equal variance of significance p-values across both groups. No group had more significant differences between the slowest and fastest production of any target than the other.

As discussed in the literature review, I suggested there could be overlap between SSEs seen in children with ASD and those of children with childhood apraxia of speech (CAS). CAS is a core impairment in the planning and programming of speech movement (American Speech-Language-Hearing Association (ASHA), 2007) and if there were overlaps in presentation with this condition I would have seen significantly more tongue shape variability in the ASD group than the TD group as well as significant inconsistency in the DDK task, implying an impairment in speech motor control (Grigos, Moss and Lu, 2015). Grigos et al. (2015) examined the speech motor control of eleven children with CAS, eleven with speech delay and eleven TD children using motion capture imaging of articulators. They found that the CAS group differentiated from the speech delay and TD groups in movement duration and variability. Children with CAS and children with speech delay had longer durations of jaw movement than the TD group, implying that longer durations in speech movement may be a general feature of speech impairment seen in multiple groups. However, movement variability of the jaw was only significant different for the CAS group. It appears that variability of the articulators may be unique to CAS and speech motor planning impairment, which was not evident in my group of children with ASD.

4.13 RQ3: Do children with ASD present with speech motor impairment symptoms?

Finding: Children with ASD do not present with speech motor impairment symptoms but with an alternative speech production strategy which is less variable and more rigid than the TD group.

DDK tasks tells us is how these articulators are functioning and give indicators of where breakdown is occurring at the “planner” and “controller” levels as described in the models of speech motor control in the literature review. It cannot however determine accurately where this breakdown is occurring. This requires a multi-faceted assessment approach in which speech is examined at all four of these levels. This study has tried to cover these four steps but focused mainly on the controller level. The behavioural assessments discussed in chapter five and six allowed us to observe how higher linguistic processes may be impaired in this model in groups of children with ASD.

4.13.1 Diadochokinesis (DDK) Instrumental Analysis

The DDK task was carried out with both the ASD and TD as a measure of speech motor control. Rate, accuracy, and consistency at the slowest and fastest production of each target sound was the main focus of analysis as it allowed us to observed speech motor control at a normal and motorically challenging states for comparison.

4.13.1.1 Rate

The maximum rate was assessed for each child which involved production of the target syllable (p, t, k) and sequence (tk and ptk) as fast as the child could produce them and was calculated in syllables per second (s/s) as slow DDK rates may be indicative of speech disorders (Williams and Stackhouse, 2009). Slower rates on maximum performance tasks have been used to distinguish between groups of children with different motor speech disorders. A slower maximum rate has been shown to successfully identify children with dysarthria, where there is a breakdown at the “plant” level, a weakness in speech muscles (Thoonen *et al.*, 1996). Following an independent samples t-test to compare the two groups, my findings showed that there was no significant difference across groups, meaning that children in both

groups had a similar maximum rate. I also compared the maximum rate of each child to the norms reported by Fletcher (1978) and found that there were more instances of the ASD group (3 children) being significantly below the norm in their speech rate than the TD group (1 child), however this did not impact the results of the independent samples t-test. While individual children within the ASD group were slower than the TD group, there was no group effect and generally the children in both groups performed similarly.

While studies on the general motor abilities of children with ASD have developed into a significant field within ASD research, the same has not happened with speech motor control. From the few studies looking at DDK in children with ASD, there are similar results to my own. Mahler (2012) used DDK to study speech motor control in group of children with ASD (specifically high functioning) and TD controls using similar stimuli as this study and results showed that the HFA group performed generally faster rates across the tasks. The findings showed that both groups performed significantly more poorly in multisyllabic tasks than single syllable targets, despite which group they were part of. I found no significant difference between production of single syllable and multisyllabic sequences, I found that while not statistically significant, the ASD had a slower rate that was more prominent in syllabic targets of /p/ and /t/ compared to the TD group. As this was not significant it is difficult to draw firm conclusions and would be worth investigated again with a larger sample size. Overall, there does not appear to be a dysarthric speech profile in this sample of children with ASD.

4.13.1.2 Accuracy and Consistency

Accuracy and consistency were measured to answer RQ3 as these have shown to be effective measures of speech motor control (Thoonen *et al.*, 1996). Accuracy of each target syllable /p/, /t/ and /k/ as well as the sequences /tk/ and /ptk/ were measured by comparing the accuracy of the first repetition at the start of each target production to the adult model. There was no significant difference found between the ASD and TD group in my results. However, there was a ceiling effect for the TD group which may have masked differences if they were present. As a rule, studies involving TD children demonstrated that accuracy in DDK performance increased

with age (Fletcher, 1978; Williams and Stackhouse, 2009). This suggests that DDK can be a sensitive measure of speech motor control, particularly for older age groups. Nonetheless, the less developed speech production of young children should be accounted for in order to avoid a misdiagnosis of speech motor impairment when the child is in fact comparable to their peers. In my case as children with ASD were age-matched with the control group, this accounted for this issue. My measure of consistency was taken from the William and Stackhouse (2009) method which was the consistency of five repetitions compared to the child's baseline production (the first syllable/sequence produced). There was no significant difference found between the ASD group and TD group at the single syllable level, the sequence level, and both conditions combined. Inconsistency in speech is a marker for childhood apraxia of speech and indicates a breakdown in planner aspects of speech motor control (Thoonen *et al.*, 1996; American Speech-Language-Hearing Association (ASHA), 2007)

Again, there is little research on accuracy and consistency of non-speech like tasks and DDK in children with ASD. Mahler (2012) had similar findings to my study in which there was no significant difference between children with ASD and TD controls. Adams (Adams, 1998) used perceptual measures to compare speech motor abilities and the non-speech abilities of four children with ASD (aged 9-11 years) in which they were age and gender matched with TD controls. They carried out the Kaufman Speech Praxis Test for Children (KSPT; Kaufman, 1995) to investigate whether children with ASD presented with speech motor impairment and if this was significantly different from their TD peers. Their assessment protocol included examining the accuracy and consistency of simple syllable productions, complex syllable productions and oral motor movements. Their results showed that the ASD group performed with significantly lower accuracy in the oral motor movements and also had lower accuracy on some of the speech motor tasks, e.g., the complex consonant production synthesis; blend synthesis and polysyllabic synthesis/sequencing. This is contrary to my findings in which there were no differences in accuracy this may be due to the small sample size in this study, a ceiling effect in the DDK task on accuracy or a result of different assessment protocols. The issue with the Adams (1998) findings is the nature of the stimuli in terms of their lexicality and requirements for speech processing are not evident. Also

due to the small sample size I am not able to generalize the results. They found no significant difference between the two groups on consistency of speech production.

4.13.2 General Discussion on DDK Results

I used DDK to attempt to measure whether SSEs that have been found to be produced by children with ASD could be related to motor impairment related to speech motor difficulties. I found no significant differences in the rate, accuracy, and consistency to the TD group. It appears that the SSEs found in children with ASD were not related to their speech motor control performance. Few studies have observed DDK rate, accuracy, and consistency in children with ASD, however one study similar to ours looked at the rate, accuracy and consistency of DDK performance in twelve children with high functioning ASD (HFA), eleven children with motor speech disorder (MSD) and thirteen typically developing (TD) controls (Deshmukh *et al.*, 2012). They found that although the HFA group were always performing intermediate to the TD group, but with no significant findings. However, it was the MSD that had significant differences in the three domains from the TD group. This suggests that motor delays in children with ASD may not necessarily impact their speech motor control as shown in my own results where speech motor control was not significantly more compromised at the group level. However, there may be very subtle speech motor control issues not detected within the independent sample t-tests carried. Whilst these tests can observe differences at a group level, there may be variation within individuals that show different speech patterns. The individual level was explored in the case study of chapter five of this thesis.

This is further evidenced in a study carried out by Belmonte et al. (2013) who with a cohort of 31 children with ASD assessed motor skills, receptive language, expressive language and speech, including speech motor abilities. What it revealed within their sample was there was a subtype of speech performances of children within the ASD group who presented with motor impairment and with a speech and expressive language profile out of proportion to their receptive language abilities, similar to my results. Childhood apraxia of speech is rare, and I would not expect to find high levels of comorbidity of this condition in ASD, suggesting that speech motor issues found may be a result of the ASD condition itself and not a comorbidity (American Speech-Language-Hearing Association (ASHA), 2007; Shriberg *et al.*, 2011).

Following speech and language intervention, they were found to have learned language slower than other children within the group and had reduced oral motor skills after intervention in comparison to the rest of the group and norms. Interestingly the speech motor abilities in this cohort were only weakly correlated with gross and fine motor skills at the initial level and did not correlate in rates of development (this study was carried out over a period of ten months of intervention). While my results agree with the weaker correlation with speech motor control and motor impairment, I still found a significant correlation with phonology and elements of motor control (balance). These findings show the highly varied nature of ASD and that is vital that individual assessment or oral, fine, and gross motor skills are carried out in order to create child-centered and individualized interventions.

The DDK results show that the higher rates of SSEs present in ASD are not likely due to a pure speech motor impairment and require us to observe different domains of speech perception and production such as the speech attunement theory or differences in feedback control.

4.13.3 Speech Motor Control and General Motor Control

What my findings have suggested so far is that when I used DDK to observe whether these SSEs may be a result of a speech motor impairment I found no significant differences from the TD group. This is despite the significant difference in motor control between the groups where the majority of the ASD group had significant movement impairment.

The task independence hypothesis stipulates that motor control of the organs used for speech is independent of the motor task that is imposed on them. There is a “general” oral sensory-motor system which controls activities for the muscles involved in speech (Clark et al., 2001; Clark & Robin, 1998). The hypothesis proposes that motor impairments in speech are a result of a dysfunction of a common-sensory motor system and as a result would be associated with impairment in general motor abilities as well. This relates to some parts of my findings in which there was a correlation between DEAP speech performance and elements of motor control (balance) in the ASD group. However, it does not explain why for a group of children with ASD who have significant motor impairments why this does not reflect

in their DDK production, a task designed to find speech motor impairment. Ziegler (2003) posits an alternative hypothesis, the task dependent hypothesis in which the movements of the lips, tongue and larynx are controlled differently dependent on the purpose and goal of the motor task. There are multiple sensory-motor subsystems that are task specific. These subsystems have unique properties that have specialised neural circuitry. Therefore, DDK tasks would function with sensory-motor resources unique to those of general motor skills, such as balance.

The question is how general motor skills and speech motor skills are unique from each other. Ziegler (2003) proposes that the motor skill of speech is linked to the auditory domain, a specific sensory modality. Whereas general motor skills such as grasping, and pointing are based on visual spatial and/or proprioceptive representations which is not required for speech encoding. This is evidenced in my results from the significant motor impairment in tasks such as balance, fine motor control, throwing and catching etc. Yet in DDK production there was no evidence of impairment or even significant difference from the TD group. The muscles required in speaking such as the larynx and the velum are basically inaccessible to sensory or visual-spatial representations. Instead, evidence from the literature points at speech movement planning referring to an acoustic or auditory space (Guenther, 1995; Perkell *et al.*, 1997; Guenther, Hampson, and Johnson, 1998).

4.14 Mean Syllable Duration

Syllable duration of the DDK speech sound production task was measured to assess the difference in rhythm of sounds produced in each target, to understand if a breakdown in speech motor control could be observed in the rhythmic production of DDK production, helping answer RQ3. Duration of syllables and segments would be impacted if the speaker has difficulties in producing appropriate articulatory movements (Kocjancic, 2010). In this study the mean duration of the slowest and fastest production of each of the DDK targets was calculated. The definition of a syllable was from the burst of the plosive until the end of the following vowel. A comparison of standard deviations was carried out to assess if there was variation in the mean durations produced by the two groups. In the /ptk/ segment fastest production there was unequal variance in the group, the mean durations were not equal in variance across the two groups. In this target, it was the TD group who had

longer mean durations and a larger standard deviation in the group that the ASD group. As these differences occurred at the slowest production it implies that these differences are not the result of a speed-accuracy trade off seen at faster production but a difference in rhythm of speech production.

I also carried out an independent samples t-test to determine whether there are any statistically significant differences between the mean durations of ASD and TD groups. Interestingly the TD group produced a significantly slower mean duration of both the /ptk/ segment in the fastest conditions in comparison to the ASD group. No other significant differences in other targets were found.

4.14.1 Lack of Variability in Syllable Durations

It has been suggested that speech production in ASD can be monotonic or over-precise but with a lack of robust evidence in the literature. The lack of significant variance in mean duration in the ASD group compared to the TD group who had significant variance in the /p/ single target slowest and the /tk/ segment slowest productions may be a result of the over-precise articulatory style of the group with ASD rather than an issue with the TD group. One of the few studies to was carried out by Patel et al. (2020) in which they examined the acoustic properties of the speech of 55 individuals with ASD and 39 TD controls. Similar to my study, they examined syllable duration and found no group differences in paired syllable durations between the ASD group and the TD group (Patel *et al.*, 2020). Additionally, Kissine and Geelhand (2019) examined syllable level acoustics in adults with ASD and neurotypical controls and found participants with ASD had greater articulatory stability in vowel production than controls, both in their articulatory gestures and phonation.

Variation is a ubiquitous feature of speech depending on the semantic, acoustic, and phonological context. Perceptual compensation is where the listener must account for context-induced effects to understand the message. Errors in speech perception may lead to adjustments in perception and production norms, resulting in altered speech production patterns (Yu, 2010). The difference in cognitive processing style may have caused the lack of variation in tongue shapes observed in the ASD group. The Weak Central Coherence theory argues that individuals with ASD show “detail-focused processing in which features are perceived and retained at expense of

global configuration and contextualised meaning (Happé, 1999). In contrast TD individuals tend to process information by gathering information of higher-level meaning, sometimes at the expense of memory of the details (Happé and Frith, 2006). In a similar model, the “Enhanced Perceptual Functioning” model suggests that in individuals with ASD the prioritisation of processing incoming information compared to higher-order operations can result in impairment in perception and lead to disruptive development of behaviours and abilities (Mottron *et al.*, 2006). Yu (2010) studied this phenomenon in a group of individuals with and without autistic traits, assessed according to the ASD Spectrum Quotient (ASQ; Baron-Cohen et al., 2006). They found those with high ASQ scores attended to details and patterns more and are less likely to compensate for talker voice effects. This may directly harm their social and communication abilities and result in reduced perceptual compensation.

However, lack of variability does not apply to all aspects of articulatory gestures and phonation in ASD. Randazzo (2013) studied breath control and voice onset time (VOT) in children with ASD compared to TD controls. They found VOT was not significantly different between the groups, evidencing that speech motor control was not more impaired in the children with ASD. However, they found some variation in the standard deviations of speech type in the ASD group. They had longer VOTs than the TD group. Therefore, it should not be assumed that lack of variability is present in all aspects of ASD speech and should be examined on an individual basis. Further research should look at all these different aspects of phonation and articulation in order to create a speech profile that may be widespread in the ASD phenotype.

The reason that the TD group produced a significantly slower mean duration in the /ptk/ segment fastest condition in comparison to the ASD group may be a result of coarticulation instability. Coarticulation is the overlapping of adjacent articulation, how a target phoneme is influence by surrounding phonemes (Volenec, 2015). Coarticulation on consonants impacts the articulators which are more linguistically constrained such as /k/ and /t/ (Recasens, Pallarès and Fontdevila, 1997). With /p/ there is little lingual constraint as there is no tongue-dorsum involvement in bilabials, therefore there is no clear coarticulatory direction for /p/, allowing more variation in

this tongue shape which could have occurred at the start of the production of this segment.

4.15 Summary

Whilst there was no significant difference found in the DDK analysis, children with ASD are potentially slower than TD in some aspects of their maximum performance rate but this could not be confirmed in this study and may be a result of my small sample size. However, the children with ASD presented as accurately and consistently in the DDK task as the TD group suggesting their speech profile does not indicate a comorbidity or similarity to childhood apraxia of speech. One interesting finding of this study were the significant difference in tongue shape variability between the two groups. The ASD group appears to be less variability in their tongue shape compared to the TD group, indicating they have more rigid and regular speech motor performance. This lack of variability is also evidenced in the mean syllable durations in which the TD group has slower production in the most motorically complex target than the ASD group which remains static. This aligns with common core ASD diagnostic criteria when looking at it from a behavioural level but found here specifically at the speech motor level (World Health Organisation, 2017). This may be a result of impaired speech perception, resulting in lack of attunement to the speech ambient environment and/or reduced sensory feedback control in the speech motor control processing chain. Furthermore, my results give some evidence to the task specific theory of speech production. Whilst there was significant motor impairment found in the behavioural assessment of general motor abilities (MABC-2; Brown & Lalor, 2009) this did not translate at the speech motor control level where speech motor abilities where relatively spared in the ASD group, although performing slightly differently from the TD group.

Chapter Five Case Studies

5.1 Introduction

Autism spectrum disorders (ASD) are known to be a heterogeneous condition which is commonly defined by impairment in social communication and presentation of restricted repetitive behaviours (American Psychiatric Association, 2013). While there is one diagnostic label used to describe this population, this is a condition in which individuals can have widely different clinical presentation (Lombardo *et al.*, 2016). The heterogeneity of ASD occurs in multiple domains, for example in general development, sex and gender presentation, speech, and language development as well as other clinical comorbidities (Wilkinson, 1998; Geschwind and State, 2015; Szatmari *et al.*, 2015; Lombardo *et al.*, 2016). It is now moving to an understanding that ASD is a multitude of conditions, rather than one diagnostic group with similar clinical presentation (Gillberg and Fernell, 2014). Therefore, I have chosen to present case studies of five of my participants with ASD to draw out learnings from individual presentations, as well as the group. While there was a lack of significant difference between the two groups in most of the above measurements, some children in the ASD group performed significantly below the group mean and in comparison, to the TD group. The small sample size as a result of recruitment difficulties, may have had a role in the non-significant results between the groups in most of the statistical analyses of the instrumental measures. Therefore, looking at interesting individual results gives us a window into future research directions that could be carried out with a larger sample size.

The five case studies chosen to reflect the variety in presentations within the ASD group, laid out in the table 30 below. Each case study shows a different presentation of motor and speech strengths and weaknesses, possibly sitting within different identify subsets of presentations within this diagnostic category. Each participant has been given a pseudonym indicated in table 30. The motor result is defined within the normal range according to the MABC-2 (Henderson, Sugden, & Barnett, 2007) manual which developed standardised norms from a population of children and adolescents aged between 3-16 years old. Standard scores are interpreted using percentile equivalents in which:

a) <15th percentile indicates presence of a motor impairment
b) >15th percentile indicates no motor impairment

The speech result was defined as out with the normal range based on the standardised norms defined by Shriberg et al. (1997). Any result below the 85% cut-off point was defined as out with the normal range.

a) > 90% = mild,
b) 65%-85% = mild-moderate,
c) 50%-65% = moderate-severe
d) < 50% = severe

Table 30: *Case Studies' Presentations*

Participant	Motor result within normal range	Motor result out with normal range	Speech result within normal range	Speech result out with normal range
Sophie (07F)	X			X
Harry (03M)		X		X
Sam (06M)		X	X	
Jacob (08M)	X		X	
Emma (02F)	X		X	

The five case studies were discussed in relation to the following research questions:

- RQ1: Do children with ASD produce significantly more speech sounds errors (SSEs) compared to typically developing children?

Children with ASD produce more SSEs than typically developing children. The children with ASD have a lower consonant correct (PCC) from the Diagnostic Evaluation of Articulation and Phonology (DEAP; Dodd, 2002) than the TD children

with a significant result in an independent samples t-test comparing both groups. Within the five case studies, Sophie and Harry presented with speech results out with the normal range, but both have differing motor presentations. Examining their speech motor control and other behavioural assessments allows us to determine whether the difference in motor abilities is related to their speech presentation.

- RQ2: Does instrumental analysis of speech reveal subtle articulatory differences between ASD and TD groups?

A significant difference in tongue shape variation comparing the slowest and fastest speech target reveal subtle articulatory differences. A larger equality of variance of tongue shape for the same targets test also indicate articulatory differences. Longer mean syllable durations of the slowest and fastest DDK targets were used to determine speech differences. Within the five case studies it is predicted that Sophie and Harry present with these differences compared to the other case studies. The comparison with Sam, Jacob and Emma allows observation of whether subtle speech patterns are identified using the ultrasound tongue imaging, not identified using perceptual speech assessments.

- RQ3: Do children with ASD present with speech motor impairment symptoms?

To test for this rate, accuracy and consistency was measured and compared across groups (and to published norms) in a diadochokinesis test (DDK- rapid alternating syllables such as /pa ta ka/). A lower mean rate, lower accuracy and/or lower consistency was predicted if a speech motor impairment was present. These measures help identify if a speech motor control impairment could be impacting the speech presentation, particularly in the case of Harry who presented with a speech and motor profile out with the normal range. It also allows us to understand whether the motor profile presented by Sam is related to his speech presentation.

Table 31: *Overview of Results for Case Studies*

Assessment	Sophie	Harry	Sam	Jacob	Emma	ASD Group Mean	TD Group Mean
DEAP PCC	75	86.1	100	97.37	97.2	93.6	96.9
DEAP Inconsistency	11.11	16.7	0	0	10		9.7
CUW PCC	58.52	92.3	98.08	98.08	97.83	96.0	97.3
CUW Inconsistency	40	20	0	20	0		20
MABC-2	6	4	5	8	6	4.70	-
CELF	42	56	109	98	79	78.38	-
Leiter	78	77	96	99	78	84.67	-
SCQ-Current	13	17	23	8	9	13.55	-
SCQ-Lifetime	19	22	25	19	24	21.70	-

Note. Yellow highlight indicates when the result is below the norm.

Table 32: *Maximum DDK rate for each speech target*

Target	Sophie Rate	Harry Rate	Sam Rate	Jacob Rate	Emma Rate	ASD Mean	S.D	TD Mean	S.D
p	6.63	5.91	6.09	6.49	8.36	6.39	1.04	6.42	1.31
t	5.65	5.86	5.27	6.81	9.24	6.27	1.58	6.73	1.50
k	6.39	4.14	4.84	6.06	7.42	5.60	1.06	5.34	0.87
tk	3.52	8.34	5.38	6.07	6.42	6.00	1.53	6.59	1.35
ptk	2.29	7.43	4.86	5.03	5.92	5.23	1.47	6.77	2.65

Note. Yellow highlight indicates when the result is below the norm of the subject group.

Table 33: *Accuracy of first production of single syllable and sequence targets within DDK task*

	Sophie	Harry Accuracy	Sam	Jacob	Emma	ASD	S.D	TD	S.D
Single Syllables	100	100	100	100	100	91.67	20.83	100	0
Sequences	72.73	54.55	100	91.67	100	84.19	10.02	89.81	10.02

Note. Yellow highlight indicates when the result is below the norm of the subject group.

Table 34: *Consistency of production of speech targets within DDK task*

	Sophie	Harry Accuracy	Sam	Jacob	Emma	ASD	S.D	TD	S.D
SSA	72.22	55.55	83.33	44.44	77.77	66.02	20.28	85.58	17.43
SQA	63.64	58.33	75	36.36	90.90	69.07	18.60	70.37	21.29

Note. Yellow highlight indicates when the result is below the norm of the subject group.

5.2 In-Depth Case Study: Sophie

Sophie was chosen for an in-depth case study as she presented with the lowest DEAP (Dodd, 2002) and CUW (James, 2009) percentage of consonants correct (PCC) in the ASD group but was mid-range for the MABC-2 (Brown and Lalor, 2009). Therefore, observing the types of SSEs Sophie produces as well as her tongue shape variation may indicate whether her higher rates of SSEs are a result of a speech motor impairment, indicating that this may be an issue for some children with ASD. Due to her performance in the speech assessments being out with the normal range for TD children and the ASD group, it allows an exploration of a case that may reveal a subset of children with ASD who have different speech profiles.

At the time of data collection Sophie was aged ten years and eleven months (group mean = 9;3). She completed all the behavioural assessments required which are discussed in detail below. Her results were discussed in relation to the research questions. The analysis of Sophie's behavioural, perceptual, and instrumental assessments revealed a complex behavioural and speech profile with results presented in table 31. She had low scores on both non-verbal IQ and language indicating an overall delay in cognitive and linguistic development. Her social communication questionnaire provided evidence that she has presented with autistic symptomatology, which has slightly reduced over time. The Clinical Evaluation of Language Fundamentals – 4 (CELF; Semel et al., 2003) was used to assess language abilities and was carried out with only the ASD group. Sophie's core language score placed her in the "low to severe" category as she was two standard deviations below the mean. The movement assessment revealed a significant movement impairment, where fine motor skills were slightly more spared than gross motor abilities.

5.2.1 RQ1: Do children with ASD produce significantly more speech sounds errors (SSEs) compared to typically developing children?

The DEAP (Dodd 2002) was used to give a score on general articulation and phonology and the CUW (James, 2009) was used to give an indicator of multisyllabic

production. Sophie's results for the DEAP (Dodd, 2002) and the CUW (James, 2009) are laid out in table 32. The measures shown are percentage consonants correct (PCC) and an inconsistency score, both calculated using the instructions in the speech assessments' manuals which is described in detail in chapter 3.

Sophie performed significantly lower in PCC scores compared to the ASD and TD group. Her higher inconsistency score means she was more inconsistent than the mean of both groups. Sophie performed out with the norm in the DEAP (Dodd, 2002) and CUW (James, 2009) when using both test's standard norms (James, 2009). Calculating the PCC allows understanding of the severity of a disorder where > 90% = mild, 65%-85% = mild-moderate, 50%-65% = moderate-severe, and < 50% = severe (Shriberg *et al.*, 1997). In Sophie's case, she falls into the mild speech impairment category in the DEAP (Dodd, 2002), with fairly consistent speech within this assessment, however this is quite low for her age group and would be defined as a delayed speech pattern. Whereas with the CUW (James, 2009), her multisyllabic performance is significantly poorer, with a score indicating a moderate-severe impairment and a high inconsistency score. The transcriptions for these assessments are shown in table 35 and table 36 and then a discussion of the identified speech processes follow.

Table 35: *Table of DEAP (Dodd, 2002) Transcription*

Target	IPA	Transcription 1	Transcription 2	Speech Process	No. of Consonants	No. of Consonant Correct
Watch	wɔtʃ	Correct	-		2	2
fishing	fɪʃɪŋ	Correct	Correct		3	3
gloves	glʌvz	glʌbs	glʌbs	Stopping of fricative – Speech Delay Postvocalic Devoicing – Speech Delay	4	2
spider	spaɪdə	Correct	Correct		4	4
Thank you	θaŋkju	fankju	fankju	Fronting – Speech Delay	4	3
scissors	sɪzəz	Correct	Correct		3	3

helicopter	hɛlɪkɒptə(ɹ)	hɛlɪntɔktɔ	ɛlɪntɔtɔ*	Fronting – Speech Delay (/k/ to [p]) Fronting – Speech Delay (/t/ to [p])	5	2
bridge	bɹɪdʒ	bɪd͡ʒ	bɪd͡ʒ	Cluster Reduction – Speech Delay	3	2
Umbrella	ʌmbɹɛlə	ʌmbɛla	ʌmbɛla	Cluster Reduction – Speech Delay	4	3
elephant	ɛləfənt	ɛləfən	ɛləfən	Cluster Reduction – Speech Delay	4	3
Total					36	27
PCC		No. of CC/ No. of C *100				75%
Inconsistency Score		No. of words produced differently (a) = 1 No. of words produced twice (b) = 9 Inconsistency score (a/b) x 100 = 11.1%				11.11

Note. Yellow highlight indicates where a speech sound errors has occurred

Table 36: *Table of CUW (James, 2009) Transcription*

Target	IPA	Transcription 1	Transcription 2	Speech Processes	No. of Consonants	No. of Consonant Correct
ambulance	ambjələns	ambə	ambəns *	Cluster Reduction – Speech Delay	6	4
computer	kɔmpjutə(ɹ)	kɔputə	kɔputə	Cluster Reduction – Speech Delay	6	4
vegetables	ved͡ʒtəbəls	veʃəgəl	veʃəgəl	/d͡ʒ/ to [ʃ] and loss of /s/ Cluster Reduction – Speech Delay /b/ to [g] Backing –	6	2

				Speech Disorder		
animals	anɪməls	aməls	1*		4	4
caterpillar	katəpɪlə	kapɪpɪtə	kapɪpɪta	/t/ to [p] - Regressive Assimilation – Speech Delay /l/ to [t] Sequencing Error – Speech Motor Delay	5	3
hippopotamus	hɪpopɔtəməs	hɪpohɔpəus	hɪpohɔpəus	Sequencing Error	6	3
spaghetti	spəgɛti	kəpɛti	skɛti*	Metathesis of a cluster – Speech Motor Delay	4	2
helicopter	hɛlɪkɒptə(ɹ)	hɛlɪntɔktɔ	ɛlɪntɔtɔ*	Addition of /n/ - Epenthesis	5	2
caravan	kaɹəvan	kaləvan	kaləvan	Liquid Confusion – Speech Delay	4	3
butterfly	bʌtəɹflaɪ	bʌʔəflaɪ	bʌʔəflaɪ	/t/ to [ʔ] - Glottal Insertion - Typical Loss of /r/- Postvocalic Rhoticity - Typical	5	3
Total					51	30
PCC		No. of CC/ No. of C *100				58.82
Inconsistency Score		No. of words produced differently (a) = 4 No. of words produced twice (b) = 10 Inconsistency score (a/b) x 100 = 40				40

Note. Yellow highlight indicates where a speech sound errors has occurred

Table 37: *Phonological Processes identified in Sophie's speech*

	Phonological Process	Delay or Atypical	Identified in DEAP (Dodd, 2002)	Identified in CUW (James, 2009)
Substitution Process	Stopping	Delay	1	
	Postvocalic Devoicing	Delay	1	

	Fronting	Delay	3	3
Syllable Structure Process	Cluster Reduction	Delay	3	3
Sequencing Errors	Metathesis	Delay		1
	Epenthesis	Delay		1
Assimilation	Regressive Assimilation	Delay		1
Rare or Atypical Process	Backing	Atypical		1
	Liquid Confusion	Atypical		1
Typical	Glottal Insertion	Typical		1
	Postvocalic Rhoticity	Typical		1
Total			9	14

5.2.1.1 Substitution Processes

The following speech processes are “substitution” speech processes. Substitution, or systemic, processes describe when there are changes to sounds within the word. It usually occurs when a later acquired articulatory feature of a sound is replaced by an easier feature (McLeod and Baker, 2017).

- Stopping: Stopping of fricatives, such in the first example, involves the substitution of a fricative with a plosive that is in the same or close to the place of articulation, known as a homorganic plosive (McLeod and Baker, 2017). While this process is considered typical within normal speech development, this would be expected to be eliminated around the age of four (Peña-Brooks and Hedge, 2015). Therefore, it is a persistent speech sound error for Sophie and indicative of speech delay.
- Postvocalic devoicing:
  This involves the substitution of a voice consonant with the voiceless consonant as seen in this example (McLeod and Baker, 2017). This process is reported to be one for the most frequently produced type of voicing errors occurring in speech development this would be expected to be eliminated around the age of three (Hodson, 2004; Peña-Brooks & Hedge, 2015). Therefore, it is a persistent speech sound error for Sophie and indicative of speech delay.
- Fronting:
  This is a substitution in which the consonant that is produced further back in

the mouth is replaced by a consonant produced further forward in the mouth. This process is typical of normal speech development but is expected to be eliminated around age four, therefore it is a persistent speech sound error for Sophie and indicative of speech delay (Peña-Brooks and Hedge, 2015).

5.2.1.2 Syllable Structure Processes

The following speech processes are “syllable structure” speech processes. Syllable structure processes describe when there are changes to sounds within the word. It usually occurs when a later articulatory feature of a sound is replaces by an easier feature (McLeod and Baker, 2017).

- Cluster Reduction: Cluster reduction is the deletion of a consonant within a cluster of consonants, turning it into a simplified single consonant (McLeod and Baker, 2017). This process is typical of normal speech development but is expected to be eliminated around age seven, therefore it is a persistent speech sound error for Sophie and indicative of speech delay (Peña-Brooks and Hedge, 2015).

5.2.1.3 Sequencing Errors:

These are SSEs that result in difficulty in selecting and sequencing the correct speech sounds. Sequencing speech requires a phonological encoding stage, in which the utterance is planned at an abstract level and then used to select the correct motor program for articulation and sequencing errors are indicative of speech motor control issues (Levelt, Roelofs and Meyer, 1999; Cler *et al.*, 2017)

- Epenthesis:
  This is the insertion of a sound within a word and is an uncommon structural process for typically developing English-speaking children. This sound is expected to be eliminated by age eight and is therefore a persistent speech error and indicative of speech delay (Peña-Brooks and Hedge, 2015; McLeod and Baker, 2017).

- Metathesis:
  Is the reversal or swapping of the position of two consonants in a word. This sound is expected to be eliminated by age eight and is therefore a residual speech error and indicative of speech delay (Peña-Brooks and Hedge, 2015; McLeod and Baker, 2017).

5.2.1.4 Assimilation

Assimilation processes are where the sound becomes more like another in the word (McLeod and Baker, 2017).

- Regressive Assimilation:
  This is when a sound later in the word affects the sound earlier in the word and would expect to be eliminated by age three so is a persistent speech error and indicative of speech delay.

5.2.1.5 Typical

- Glottal Insertion:
  This occurs when a glottal stop replaces a consonant, most typically /t/. This is an accepted production in Scottish English so can be expected in children within this context.
- Postvocalic Rhoticity
  Dropping of the /r/ immediately after the vowel which occurs in some varieties of English, but less so often in Scottish English.

  5.2.1.6 Rare or Atypical Processes

- Backing: Backing is another atypical speech sound process that is the substitution of a consonant sound further forward in the mouth with a consonant further back in the mouth (McLeod and Baker, 2017). This is indicative of a speech sound disorder.

5.2.1.6 Summary

In Sophie's case she confirms the hypothesis of RQ1 and does have significantly more SSEs than the TD group. Sophie's speech profile is marked by a significant speech disorder with elements of both delayed and disordered features. There were a significant number of SSEs identified from the CUW (James, 2009) compared to the DEAP (Dodd, 2002). It appears that the increase in motoric complexity resulted in the production of more atypical speech sound errors.

The ability of children to produce multisyllabic speech is an important milestone in speech development and has shown to be delayed in children with ASD, as in this instance with Sophie (Hailpern *et al.*, 2012). Multisyllabic words differ from shorter words as they comprise of more and variable phonological constituents (James et al., 2008). For Sophie it appears that the extra speech processing and timing required to produce multisyllabic words impairs production compared to simpler constructions and monosyllabic words. This has been found in children with ASD in which accuracy of multisyllabic tasks were more impaired than monosyllabic tasks (Adams, 1998; Mahler, 2012). This may suggest that the motoric complexity of the multisyllabic words may be difficult for Sophie as a result of a subtle speech motor impairment. This is evidenced further in the multitude of processes associated with speech motor delay in her speech including metathesis, epenthesis, and regressive assimilation. Typically developing children aged 1-7 years have been found to produce more consonant and vowel mismatches in multisyllabic words and single or simpler words and appears to be part of normal speech development (Vance, Stackhouse, and Wells, 2005). Impairment in polysyllabic production has also been shown to indicate the presence of childhood apraxia of speech (CAS) which may be a relevant diagnosis to explore in Sophie's case (Murray *et al.*, 2015).

The high inconsistency score in the multisyllabic assessment however may indicate difficulties with motor output of speech and other areas of fine motor control (Bradford and Dodd, 1994) and it is one to the indicators for the presence of CAS (American Speech-Language-Hearing Association (ASHA), 2007). Whilst Sophie did show subtle difficulties with fine motor control, this was not as impaired as her gross motor abilities. This indicates that perhaps Sophie has a speech delay in which breakdown in the speech processing and production chain happens earlier in the

processing and planning part, perhaps similar to CAS. A new proposed definition for this is “speech motor delay” in which the hypothesis is that children presenting with speech delay may have a motor component associated with this delay, as possibly seen in Sophie’s speech (Shriberg *et al.*, 2019). Without testing of multisyllabic speech these subtle differences would not have been revealed. This shows the importance of assessing speech in multiple domains, including multisyllabic speech.

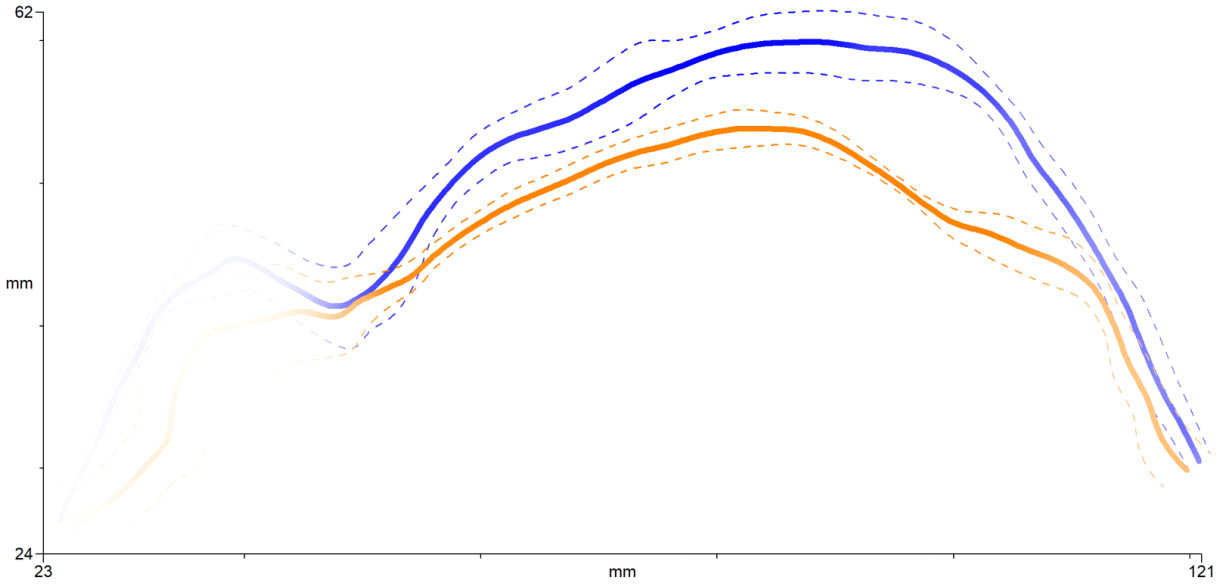
5.2.2 RQ2: Does instrumental analysis of speech reveal subtle articulatory differences between ASD and TD groups?

Ultrasound tongue analysis (ultrasound) was used to quantify tongue shapes at the slowest and fasted production of each target of the DDK production to answer this research question. When observing Sophie’s results five out of the eight sound targets were significantly different at the slowest and fastest production. An articulatory difference that was not identified from perceptual assessment alone. This includes p (ptk sequence), t (single syllable) and t (tk sequence).

The focus of the ultrasound analysis was the DDK task described above with the in-depth analysis afforded by instrumental measurement. The ultrasound data was annotated and compared to typically developing children using different statistical tests (e.g., independent sample t-tests, ANOVA, Levene’s test for equality of variance) on the following measures which account for speech motor control. However, it also afforded in-depth analysis of each participant’s tongue shapes during the DDK task.

There seems to be no trend differing the tongue shape variance at single syllable or at the more motorically complex sequence level, both have significant variability in different speech sound productions. Figures 29-31 shows this significant variance between the slowest (blue line) and fastest (orange line) production of the five significantly difference syllables and sequences. The lines at the exterior which are faded out are ignored due to lack of precise imaging possible at the root and tip of the tongue. There is a markedly different tongue shape at the different speeds.

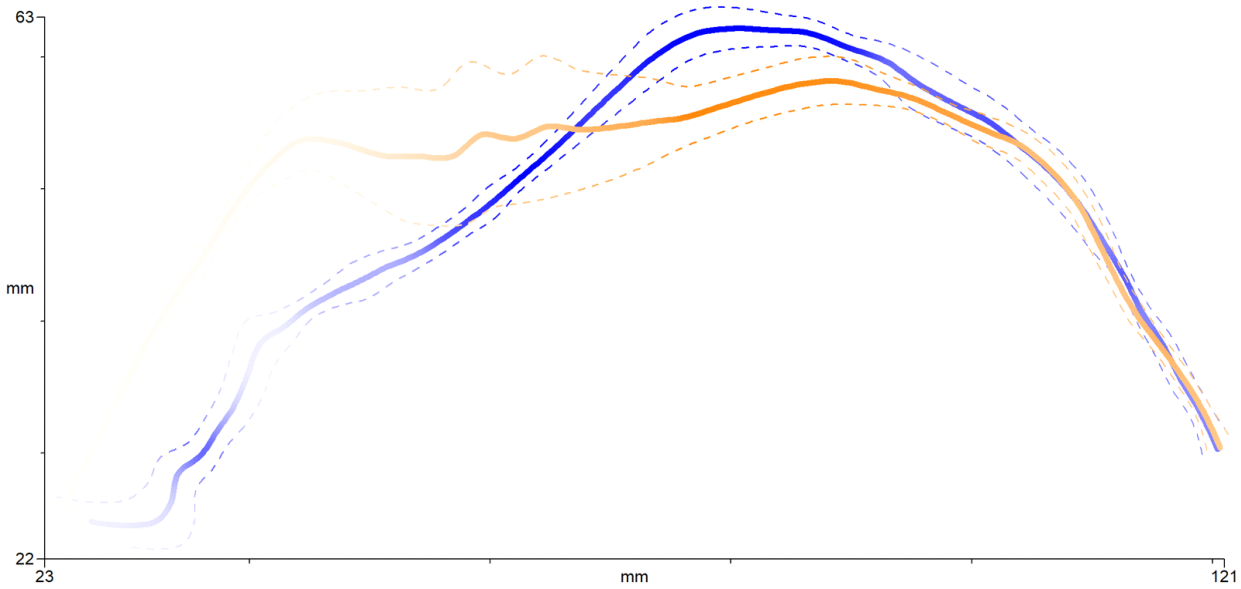
Figure 29: *Slowest and Fastest Production of /p/ in the /ptk/ sequence*



Note. Blue line is the slowest production, and the orange line is the fastest. The solid lines indicate the mean, and the dashed lines indicate the standard deviations. The tongue tip is to the right.

In these comparisons of /p/ in the /ptk/ sequence there is a large significant difference at the tongue dorsum. At the slower rate, the tongue dorsum is much higher than at the faster level. What is notable is the large variation indicated by the wide distance between the dashed standard deviation lines, particularly at the slower production. This indicates a reduction of speech motor control as consistency of tongue shape at differing speeds is a measure of motor control (Zharkova, Hewlett and Hardcastle, 2011). On further inspection of the vowels produced, an inconsistent vowel repetition was noted. In this particular sequence at the slowest repetition -3SD and -1SD, Sophie produced the vowel /ɪ/ on one repetition within the five repetitions when it should have been consistently /ə/ as modelled by the adult model provided at the start of each target sequence. This vowel difference would account for difference in tongue shape during production of /p/ which, as a bilabial stop, does not have constraints on tongue shape, instead taking on the shape of the following vowel.

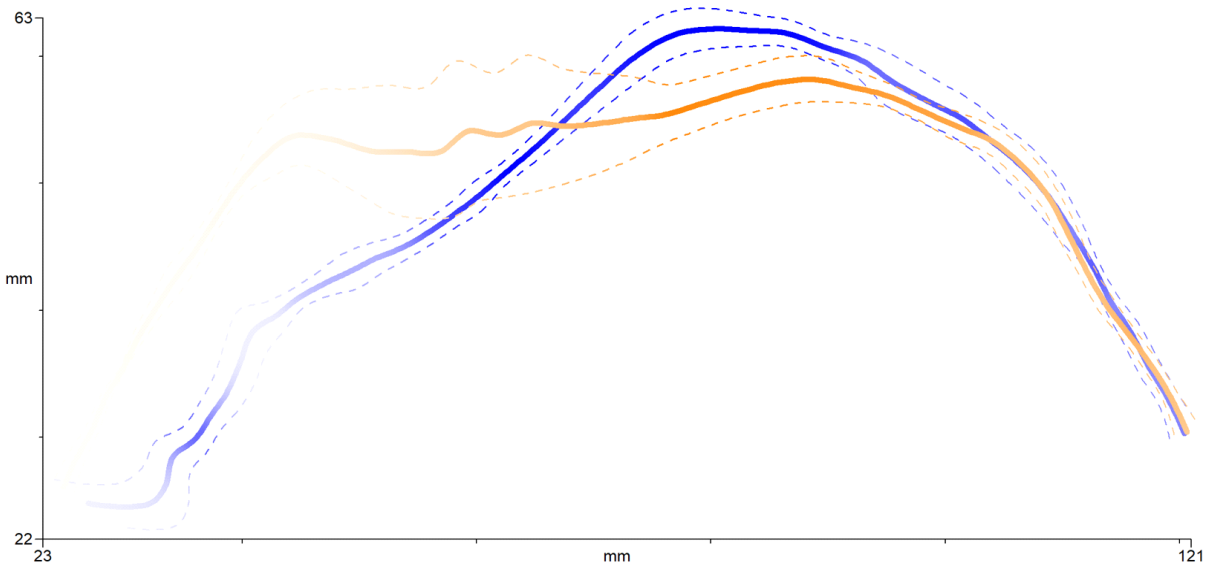
Figure 30: *Slowest and Fastest Production of /t/ single syllable*



Note. Blue line is the slowest production, and the orange line is the fastest. The solid lines indicate the mean, and the dashed lines indicate the standard deviations.

Again, in the /t/ single syllable tongue shape comparison at the slowest and fastest speeds there is a significant difference in tongue shape at the dorsum. Additionally, there is a large standard deviation of tongue shapes in the slowest but particularly the fastest in this region of the tongue. On observation of the vowels, it is noted again an inconsistent vowel strategy that changes as the speed increase. At the slower speeds -3SD, -2SD and -1SD Sophie produced /ɪ/ instead of the modelled /ə/. Then at the faster speeds of the mean, +1SD and +2SD, she produces /ə/ with two out five repetitions at the +1SD being /ɪ/. This tells us is that Sophie is employing an inconsistent vowel strategy throughout these tasks that were not identified in the clinical speech assessments. It appears at the slower rates her production of the modelled vowel is incorrect, improving as the speed increases. She is, however, very accurate at the point of constriction at the front of the tongue, in which these are the same for both speeds. This is likely due to the coarticulatory resistance that is greater for an alveolar closure, /t/ at the front of the tongue. Whereas the tongue dorsum has less constraint. The more constraint on the sound, the greater degree of coarticulatory resistance and therefore less impacted by the alternative vowel strategy (Recasens et al., 1997).

Figure 31: *Slowest and Fastest Production of /t/ in the /tk/ sequence*



Note. Blue line is the slowest production, and the orange line is the fastest. The solid lines indicate the mean, and the dashed lines indicate the standard deviations.

There is a difference in tongue shape in the dorsum region of the tongue for the slowest and fastest production of /t/ in the /tk/ sequence. There is also a large standard deviation of the tongue shapes at both speeds, significantly more for the fastest production. This is occurring mostly at the back of the tongue. Again, on observation of the vowels, there is inconsistency in her production from the slower and faster rates. However, in this sequence there is no clear pattern between the differences at slower and faster rates. At the slowest rate (-3SD) she produces /ɪ/ instead of /ə/ consistently. This becomes more inconsistent at the next fastest rate (-2SD) in which she produces three repetitions with the vowel /ɪ/ and two with the vowel /ə/. The increasing speeds following this, at -1SD and the mean are consistently produced with /ə/ vowel as modelled by the adult production. Then the inconsistency returns at the at the two fastest speeds which are an inconsistent production of /ɪ/ and /ə/ throughout the repetitions.

5.2.2.1 Summary of Ultrasound Tongue Shape Analysis

This analysis may tell us that Sophie has an inconsistent vowel strategy that is employed differently at slower and faster speeds. This was not identified in the clinical speech assessments, where speed was not a factor in the assessment. Therefore, her case confirms the hypothesis that instrumental analysis of speech

revealed subtle articulatory differences between Sophie and the TD groups. This analysis identified an inconsistent vowel strategy that was not present in the TD group or ASD group generally. Sophie's errors in vowel production and variation of this are more pronounced in the slower repetitions suggesting that this not purely a result of a speech motor control issue. Instead, it could be a difficulty in perception of the model provided by the adult production as well an impacted feedback signals from her own speech sound production, resulting in inconsistent vowel production.

The variability seen in the tongue dorsum shape in the sound targets displayed above may not only be due to an impairment but a result of a coarticulatory effect. Coarticulation is the articulatory overlapping of adjacent sounds in speech (Zharkova et al., 2014). Recasens et al. (1997) created a hierarchy of resistance to coarticulation that defined the coarticulatory potential of different consonants and vowels, i.e., how neighbouring phonemes affect the production of target sounds. For instance, in figure 31, we can see there is a wider gap between the two lines, indicating a large variation in tongue shape in the production of /p/ in the /ptk/ sequence. This is likely due to the fact that /p/ has little to no lingual constraint as it is not the tongue that is active in the production of /p/ but the lips. Therefore, the tongue adapts more to neighbouring vowels and can be influence by changes in speech as the tongue is not directly involved in the constriction formation of /p/ (Zharkova, 2008) and may not be a result of speech motor impairment. Whereas, with an alveolar closure, /t/, there is a greater degree of coarticulatory restraint, the tongue is more constrained so there is less influence of coarticulation at the front of the tongue. Though the back of the tongue is less constrained so subject to more variation (Recasens et al., 1997). So, it is possible Sophie constrains her jaw more at faster speech levels, resulting in a more accurate vowel production. It is possible she is anchoring her tongue by constraining the jaw.

5.3 RQ3: Do children with ASD present with speech motor impairment symptoms?

To answer this research question, Sophie completed the Diadochokinesis (DDK) tasks in full and her rate, accuracy and consistency were measured and compared within and across groups. The DDK task measures how accurately an individual can produce a series of rapid sounds. Five repetitions of single syllables (pa, ta, ka) and

sequences (pataka) were recorded at six set rates (McCann and Wrench, 2007). Sophie's results for rate accuracy and consistency are presented in table 32-34.

5.3.1 Rate

Compared with the other children in the ASD group, Sophie was within 1 standard deviation (SD) of the mean for the single syllables, /p/, /t/ and /k/. However, she was significantly slower in the multisyllabic targets of /tk/ and /ptk/, which were more than 1 SD below the mean of the ASD group.

When looking at the rate of Sophie's DDK performance in comparison to the norm standards, which are sampled on a large population of typically developing children (Fletcher, 1978), she was faster than the norm of her age group for single syllables /p/ and /t/ and within the norm of her age group for her production of /k/. However, she was slower than the norm for the more motorically complex /tk/ and /ptk/ syllable segments. This is not representative of the ASD group as a whole where no significant difference was found with the ASD and TD group in maximum rate. This tells us is that for Sophie, that there may been an issue of speech timing with the motorically complex multisyllabic sound targets /tk/ and /ptk/ as she was not able to produce them at the rate typical of her peers. This is further evidenced by her performance in the CUW (James, 2009) found in table 36. The PCC in the CUW (James, 2009) met the criteria for moderate-severe speech difficulty according the Shriberg et al. (1997) categorisation. Additionally, she had a high inconsistency score in the CUW (James, 2009), both scores indicating a particular difficulty with motorically complex multisyllables.

Repetitions of syllables such as those in the DDK task are dependent on the speaker's ability to precisely and rapidly produce syllables, which requires multiple complex processes of sensorimotor programming, planning and motor execution (Perkell *et al.*, 1997). Maximum performance tasks involve planning and programming the entire speech production mechanism, including respiration, phonation, articulation, and resonance. Difficulties in rate may indicate difficulties at these stages of planning or production. For instance, slower rate than age-appropriate norms has been found in both children with CAS and more so in children with dysarthria (Thoonen *et al.*, 1996; Williams and Stackhouse, 2009). Similar

difficulties have been found in groups of children with ASD in a study carried out by Boucher (2013) who found decreased rate of speech in fifteen children with ASD compared to fifteen TD peers. However, the children produced slower repetitions of /p/ and /t/, the single syllables, whereas the complex production of /ptk/ was faster. This suggests that while children with ASD as a whole seem to have no significant difference in rate, a subset of individual children within these groups may have speech motor symptoms typical of CAS or childhood dysarthria.

5.3.2. Accuracy

Accuracy of the DDK repetitions was calculated by measuring the accuracy of a first single repetition of the target compared to the adult model and, in addition, the accuracy of five repetitions of the target compared to the adult model. Sophie's results can be found in table 33. There was a similar pattern in the accuracy of Sophie's DDK productions compared with her rate. When looking at the accuracy of the first production of targets, Sophie had three inaccuracies, all of which occurred in the /tk/ and /ptk/ segments. The first two inaccuracies occurred in the two slower productions of /tk/, -1 and -2 standard deviations below the mean production. Here she had a substitution error on the first production in which she produced [d] instead of /t/, known as voicing. This particular substitution was not present in her clinical speech assessments. Her third inaccuracy occurred at a faster production of /ptk/ in which she produced [ptp] instead of /ptk/ in the first production. This could be interpreted as an assimilation speech sound error. Similar to the rate results, these inaccuracies occurred in the motorically complex segments and not in the single syllables.

It has been shown that inaccuracy in sequencing syllables is indicative of childhood apraxia of speech whereas a slowed DDK rate is indicative of dysarthria (Thoonen *et al.*, 1996). We have seen both of these characteristics subtly in Sophie's speech, however in both cases the impairment occurred at the multisyllabic level, which may be indicatory of an impairment more similar to CAS. In a study carried out by Murray et al. (2015), speech patterns of 72 children aged 4-12 years with suspected CAS were observed in hope of identifying clear diagnostic criteria that distinguished them from other groups with speech sound disorders. They found that polysyllabic

production accuracy within DDK assessments may be sufficient at reliably identifying CAS but that testing with a larger sample size was required.

5.3.3 Consistency

The consistency of the DDK tasks were measured by calculating consistency of five repetitions compared to the child's baseline (first) production. Sophie's results can be found in table 34. When measuring the consistency of Sophie's productions at the single syllable level they were 77% consistent (above the 67% group mean) and at the multisyllabic segment they were 64% consistent (below the 66% group mean). While a subtle difference, there is still an indicator that her performance at a multisyllabic level in poorer than the single syllables. Inconsistent speech can be a sign of CAS, however in the case of her consistency in the task, this appears to be within the typically developing range (Williams and Stackhouse, 2009).

5.3.4 Syllable Durations

To assess whether there was a difference in the rhythm of sounds produced in each target group, the mean duration for each slowest and fastest target syllable was calculated and analysed. A measure of the duration of syllables at both slowest and fastest productions of single syllables and segments were taken for statistical comparison. To observe this at a case study level, Sophie's results were compared to the mean within her own group (ASD group). Table 38 shows the mean duration of each syllable per second for Sophie's productions in the DDK task.

Table 38: *Mean duration of syllable in ms*

Target	Sophie	ASD Mean	S.D	TD Mean	S.D
p single slowest	0.02	0.02	0.01	0.04	0.03
p single fastest	0.01	0.02	0.003	0.03	0.02
t single slowest	0.03	0.05	0.02	0.06	0.02
t single fastest	0.03	0.03	0.01	0.03	0.01
k single slowest	0.03	0.05	0.01	0.06	0.03
k single fastest	0.03	0.04	0.01	0.05	0.01
tk segment slowest	0.02	0.03	0.01	0.05	0.02
tk segment fastest	0.03	0.03	0.01	0.03	0.01

ptk segment slowest	0.02	0.03	0.01	0.03	0.01
ptk segment fastest	0.02	0.03	0.01	0.03	0.01

Note. Red highlight indicates where Sophie produced duration of a syllable in ms slower than the mean of the ASD and TD groups.

The findings for Sophie's mean syllable durations were that in 7/10 productions, Sophie was below the mean, these include p single fastest, t single slowest, k single slowest, k single fastest, tk segment slowest, ptk segment slowest and fastest. The effect shown for the impairment only to occur in the multisyllabic sequences appears to have been lost as there is a wider impairment across all syllables in this measurement. Sophie was slower than the mean within her own group and when compared to the mean of both groups. However, this was not affected by whether it was the slowest or fastest production or if it was a single syllable or segment, the mean duration of her syllables appears to be affected across all these domains.

It has been shown that duration of syllables and sequences is impacted if the speaker has difficulties in producing appropriate articulatory movements (Kocjancic, 2010). This appears to be the case for Sophie in which 7/10 of her target productions are slower than the ASD and TD group means, though this is a subtle difference, it is indicative of a wider pattern of speech impairment that would need to be investigated more thoroughly by a clinician on a one-to-one basis with Sophie to determine whether this affects functioning. In terms of understanding Sophie's speech profile, there may be a subtle speech motor impairment present. Altered segment and syllable durations have been identified in children with CAS which has not been found in this group of children with ASD (Maassen, Nijland and van der Meulen, 2001; Nijland *et al.*, 2002) however the sample size here is small. Significantly more research needs to be carried out in relation to how syllable durations can distinguish different sub types of speech sound disorder and what this means for understanding speech motor functioning.

5.3.5 Summary

The trend appearing from Sophie's DDK results is that her rate, accuracy, and consistency were all impacted in the motorically complex sequences, /tk/ and /ptk/. Furthermore, the results from the measures of her syllable duration were below the

mean in 7/10 of her productions compared to both the TD and ASD groups. From these results it indicates that Sophie confirms the hypothesis as she presents with a speech motor impairment.

5.4 Summary of Case Study 1: Sophie

Analysis of Sophie's speech profile suggests significant speech delay with some indicators of a speech disorder. As the perceptual assessments used were screening assessments it is difficult to determine from these alone what type of speech sound disorder with which she is presenting.

The DDK analysis of rate, accuracy and consistency revealed a fairly unstable speech motor profile. Sophie was significantly slower than the norm for the more motorically complex /tk/ and /ptk/ syllable segments, and there were inaccuracies in both her production of vowels and consonants at the more motorically complex syllable segments, with a fairly inconsistent production of these target syllables. These three variables indicate the presence of a speech motor control issue, likely to be present at the planning and controller stage of the speech production chain (Ziegler, 2003b). Sophie was also slower than the mean syllable duration within her own group and when compared to the mean of both groups, indicating that she has difficulties in producing appropriate articulatory movements.

Additionally, there was significant variation in tongue shape noted for the production of several targets: /p/ in the /ptk/ sequence; /t/ single syllable; and /t/ in the /tk/ sequence/. These variations were more likely in the slowest rather than the fastest rate. On further observation, I found a significant inconsistency in the accurate production of the modelled target vowel /ə/, where a different vowel strategy was employed at the slower rates. It is likely the consonants were impacted by this changing vowel as a result of the coarticulatory effect (Recasens et al., 1997). It also appears that Sophie was not able to perceive or produce the correct adult model at slower levels, implying a speech perception issue.

Taking all of these results into consideration it would imply that Sophie presents with a speech motor impairment, which was not identified at a group level of the ASD participants. She has presented with symptoms similar to those of the class faction of "speech motor delay" proposed by Shriberg and Wren (2019), with slower rate,

inaccuracy, inconsistency in the DDK task as well as an altered vowel strategy. This shows the heterogeneity of the speech profiles of the children in this sample's diagnostic bracket and shows the importance of including case study and individual analysis within this field of research. Additionally, this case study highlights the importance of including in-depth assessment of vowels within speech analysis, which often relies on analysis of consonants. Future research should take this into account by studying how vowels and coarticulation is impacted in speakers with ASD as well as a possible subgroup of children presenting with speech motor impairments.

5.5 Case Study 2: Harry

Harry was chosen for a case study due to his speech and motor profile being out with the normal range. It allows investigation into whether differences in his speech are related to differences in motor performance. At the time of data collection Harry was aged six years and four months (group mean = 9;3). He completed all the behavioural assessments required which are discussed in detail below. His results are discussed in relation to the research questions. As shown in table 31 in his movement, language and non-verbal IQ, Harry performed significantly below his age group and was out with the norm for the ASD group. Taking these scores together, it indicates that Harry is presenting with a general developmental delay that is impacting speech, language, movement, and non-verbal abilities.

5.5.1. Perceptual Speech Assessment : Harry

Harry was chosen as he presented with a mixed speech presentation with a lower DEAP (Dodd, 2002) PCC, but within the normal range of other parts of the speech tests coupled with a low MABC-2 (Brown and Lalor, 2009). In Harry's case, he falls into the mild speech impairment category in the DEAP (Dodd, 2002) with fairly consistent speech within this assessment. He performed within the norm for the CUW (James, 2009). This indicates that his speech profile was not significantly different from the ASD and TD group averages for the more motorically complex sounds but that he presented with speech delay. When analysing the speech processes present in Harry's speech, he presented with velar fronting and stopping which are common speech processes associated with speech delay and

labialization, a less common substitution process, but still indicative of speech delay rather than disorder (McLeod & Baker, 2017). From the results of these assessments, it appears that Harry has a speech delay. This aligns with the general developmental delay he presents with in the other behavioural domains and does not indicate that speech delay is a result of a specific motor delay.

5.5.2 Diadochokinesis Tasks: Harry

Harry's results for rate, accuracy and consistency can be found in table 32-34. Compared with the other children in the ASD group (Table 32), Harry was within 1 standard deviation (SD) of the mean for the single syllables, /p/, /t/ and faster in the case of /k/. However, he was significantly slower in the multisyllabic targets of /tk/ and /ptk/ which were more than 1 SD below the mean of the ASD group.

When looking at the rate of Harry's DDK performance in comparison to the norm standards (Table 32), which is sampled on a large population of typically developing children (Fletcher, 1978), he was within the norm of his age group for single syllables /p/ and /t/. Interestingly he was slower than the norm for /k/ and the more motorically complex /tk/. However, for the most motorically complex /ptk/ syllable segments he was within the norm. It is possible that Harry performed more slowly here to account for the speed-accuracy trade off.

Accuracy of the DDK repetitions was calculated by measuring the accuracy of a first single repetition of the target compared to the adult model. When looking at the accuracy of the first production of targets. Harry produced every first target with 100% accuracy, performing above average for both groups, particularly in the motorically complex targets /tk/ and /ptk/.

Harry presents with a significantly inconsistent speech profile in these DDK tasks that was not identified in the DEAP (Dodd, 2002) or CUW (James, 2009). The inconsistency is identified in the single syllable and motorically complex segments implying a general inconsistency in speech production. Inconsistent speech can be a sign of CAS (Williams and Stackhouse, 2009).

While Harry's rate and accuracy remains relatively intact, he had significantly reduced consistency in the production of the mean targets both at single syllable and

multisyllabic levels. This confirms the hypothesis that a speech motor impairment may be present in his speech which was not found in the TD group.

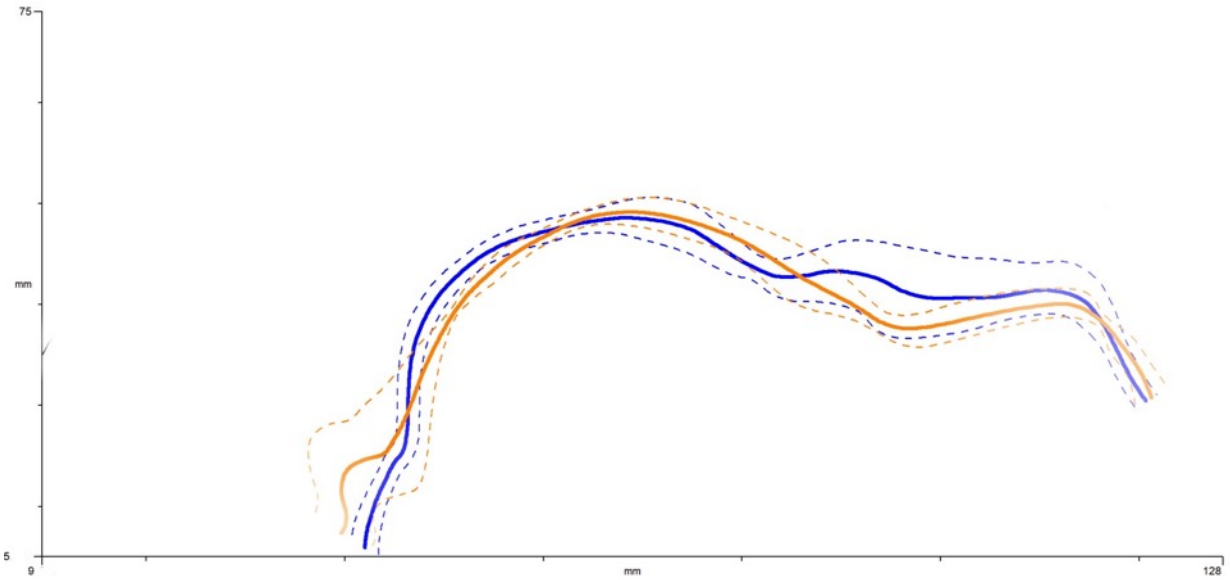
Additionally, when analysing Harry's syllable duration, he was within the ASD group and TD group means and showed no sign of impairment in syllable duration. He produced appropriate rhythm during the DDK task.

5.5.3 Ultrasound Tongue Imaging of Speech: Harry

Ultrasound was used to quantify tongue shapes at the slowest and fasted production of each target of the DDK production to answer this research question. It allowed further investigation into Harry's speech profile in order to understand if there was a motor element to the speech errors he was presenting with.

When observing Harry's ultrasound analysis, three out of the eight sound targets were significantly different at the slowest and fastest production. An articulatory difference that was not identified from perceptual assessment alone. This includes p (single syllable), t (single syllable) and t (tk sequence) Figures 32-34 show each of these.

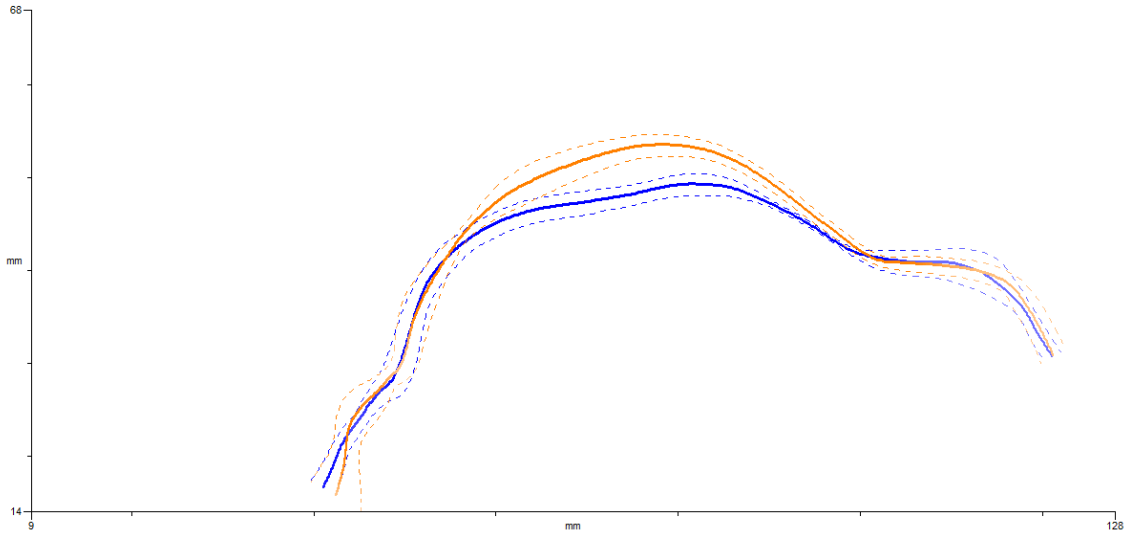
Figure 32: *Slowest and Fastest Production of /p/ in the /p/ single syllable*



Note. Blue line is the slowest production, and the orange line is the fastest. The solid lines indicate the mean, and the dashed lines indicate the standard deviations.

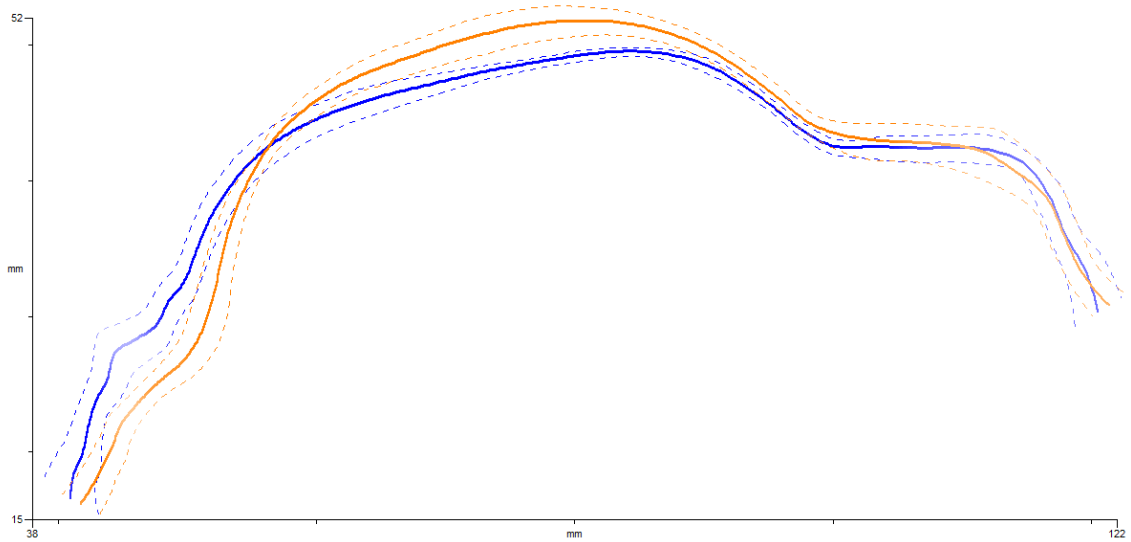
In Harry's production of the /p/ single syllable there appears to be a significant amount of variation at the front of the tongue body, particularly on the slowest repetitions shown through the large distance between the standard deviation lines (dashed lines). There is a possibility that his tongue is moving with his jaw as he opens and closes for the repetitions and is not necessarily due to a speech motor control difficulty.

Figure 33: *Slowest and Fastest Production of /t/ in the /t/ single syllable*



Note. Blue line is the slowest production, and the orange line is the fastest. The solid lines indicate the mean, and the dashed lines indicate the standard deviations.

Figure 34: *Slowest and Fastest Production of /t/ in the /tk/ segment*



Note. Blue line is the slowest production, and the orange line is the fastest. The solid lines indicate the mean, and the dashed lines indicate the standard deviations.

In both cases of production of /t/ in the single syllable and within the /tk/ segment, Harry produces the faster production of /t/ higher in the mouth in the dorsum region of the tongue than the slower production, resulting in a significant difference between the tongue shapes. There was no perceptible difference in the vowel production, indicating that there may be increased at the faster rate. Bracing refers to intentional stabilizing of tongue contact with the roof of the mouth along the upper molars or the hard palate (Tong et al., 2018). This is further evidenced by the consistency of tongue shape shown in /t/ in differing speech contexts. This indicates a different strategy in speech but does not imply a speech impairment or speech motor dysfunction. However, Harry does show lower rate, accuracy, and consistency in the perceptual speech assessment of the DDK task, the bracing in this instance may be impacting his ability to be, accurate and consistent in his production of the speech targets in the DDK task.

5.5.4 Summary: Harry

Harry presented with an overall general developmental delay that impacted his speech, language, and movement. When using speech perceptual assessments, a mild speech impairment was indicated from his DEAP (Dodd, 2002) scores.

However, in the assessment his speech appeared consistent and further analysis of speech processes indicated a speech delay rather than disorder. The consistency of speech in the speech assessments is contrary to what was revealed in the DDK task, in which Harry was below average in consistency for single syllable targets (/p/, /t/ and /k/) and significantly below in the motorically complex /ptk/. He also had a slower rate in multisyllabic production of /tk/ and below the norm for /k/. These results indicate a speech motor control issue. This was confirmed again from the ultrasound analysis in which there was significant variation in production of /t/ discovered in the dorsum region of the tongue with bracing potentially being used as a strategy. Taking these results together, Harry is presenting with speech delay with possible speech motor delay (Shriberg, Campbell, et al., 2019). The additional analysis of the DDK and ultrasound was able to identify the speech motor control element which was not found in the DEAP (Dodd, 2002) or CUW (James, 2009) Furthermore, this delay extends to Harry's language, movement, and non-verbal IQ, indicating a general developmental delay overall.

5.6 Case Study 3: Sam

Sam was selected for a case study due to his general motor result being out with the norm but with a speech profile within the norm. This allows investigation of whether ultrasound aided transcription identifies subtle covert errors that were not identified from transcription alone. The differences identified in Sam's motor performance warrant further investigation of his speech motor performance, in case there are covert errors such as increased variability and abnormal timing that was not identified in the audio-only transcriptions (Sudgen & Cleland, in press). It allows investigation into whether differences in his speech are related to differences in motor performance.

Sam was aged ten years and nine months at the time of data collection. He completed all behavioural and speech assessments. As shown in table 31 Sam was within the norm for all speech assessments, language, and non-verbal cognition. He performed out with the norm for the MABC-2, below the 5th percentile which is categorised as "denotes a significant movement difficulty" (Brown & Lalor, 2009). The greatest differences were found within the fine motor movement subtests. Sam also scored highly within the social communication questionnaire (SCQ; Rutter et al.,

2003) which both current and lifetime forms indicating a presence of a communication style associated with ASD. What these results tell us is that Sam is presenting with differences in motor abilities and a communication style typical of ASD, but no differences were found in language, speech, or cognition.

5.6.1 Perceptual Speech Assessment: Sam

Sam's speech for both single syllable and multisyllabic tests was within the norm. He was above average in performance for both the ASD and TD groups. He produced all targets accurately in the DEAP (Dodd, 2002) and only one error in the CUW (James, 2009), in which consonant harmony occurred /p/ bilabial to /t/ alveolar in the word "hippopotamus", which is a common childhood speech error.

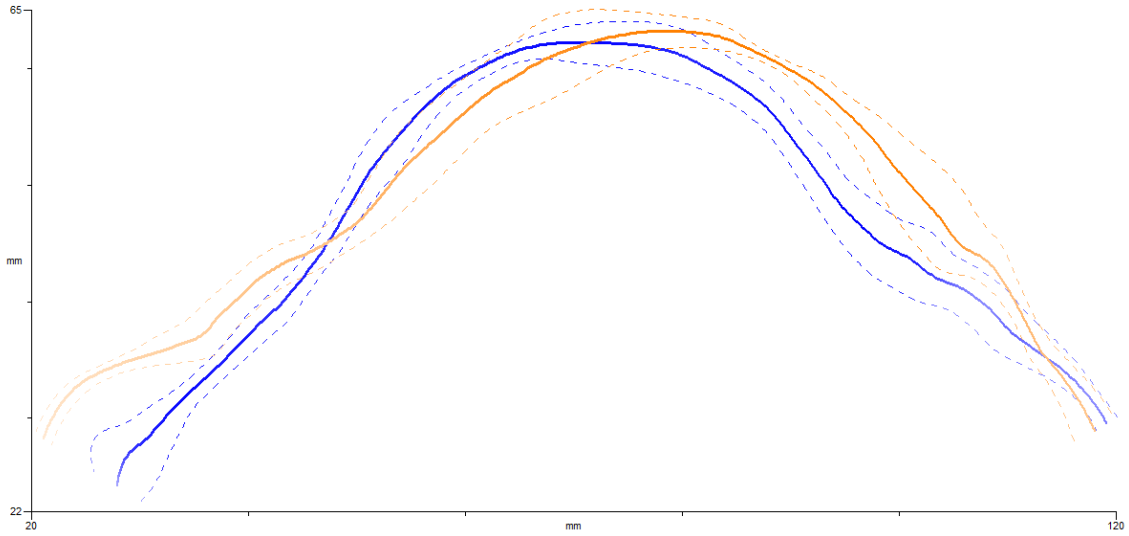
5.6.2 Diadochokinesis Task: Sam

Sam's results for rate, accuracy and consistency can be found in table 32-34. Sam's maximum DDK rate was within the norm and was above the mean for both the TD and ASD group. There was no significant difference in performance on single syllable and sequences, showing that increased motor complexity in speech did not have an impact on his performance. This was also the case with the accuracy of the production of the first syllable in which Sam was fully accurate, above the mean for the ASD group and within the mean for the TD group. Sam was more consistent in his production of single syllables than sequences in the DDK task, however this was within the norm and his performance was above the mean again for both the ASD and TD groups.

5.6.3 Ultrasound Tongue Imaging: Sam

From the perceptual assessments and the DDK tasks, Sam presented with a speech profile that was within the norm and above the means for both the ASD and TD groups. Despite this, when carrying out the ultrasound analysis, a significant difference was found for six out of eight of his DDK speech targets.

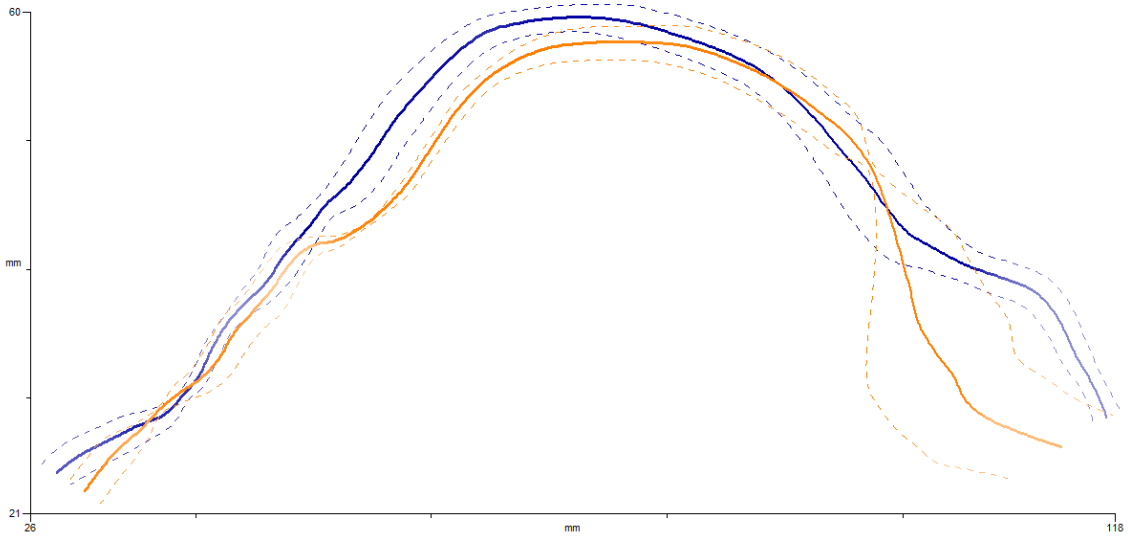
Figure 35: *Slowest and Fastest Production of /p/ in the /p/ single syllable*



Note. Blue line is the slowest production, and the orange line is the fastest. The solid lines indicate the mean, and the dashed lines indicate the standard deviations.

In the production of /p/ single syllable the tongue dorsum is raised into the velar position. However, as mentioned above in the case study of “Sophie,” during the production of /p/ the tongue is not an active articulator, instead taking on the shape of the subsequent vowel, in this case schwa. We would therefore expect the tongue to be in a similar position during the faster and slower speeds. Here there is increased variability both between the two speeds and within the repetitions themselves, indicated by the large difference in standard deviations shown through by the dashed lines.

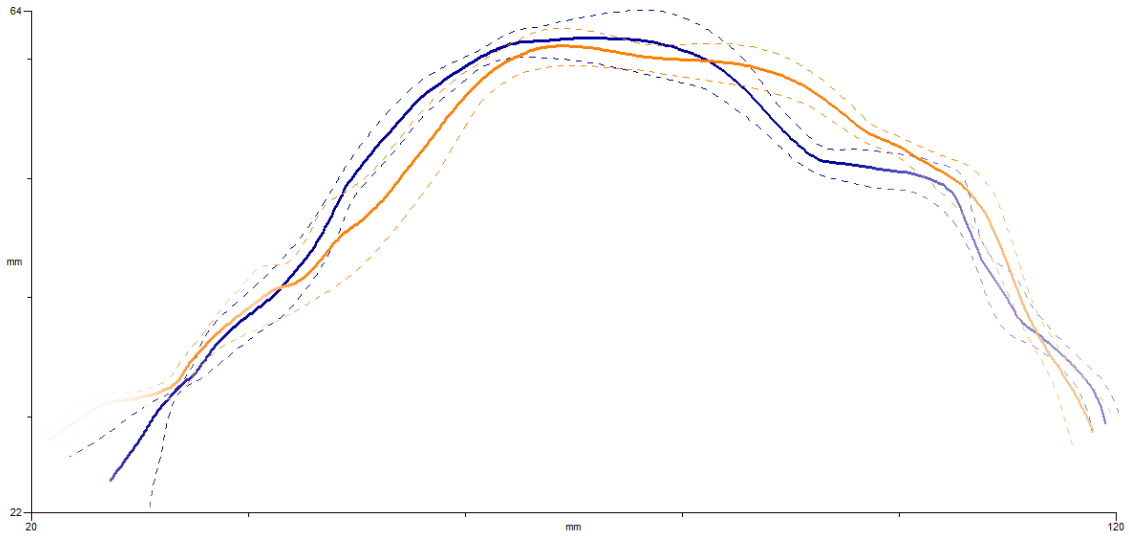
Figure 36: *Slowest and Fastest Production of /p/ in the /ptk/ segment*



Note. Blue line is the slowest production, and the orange line is the fastest. The solid lines indicate the mean, and the dashed lines indicate the standard deviations.

The increase in motoric complexity, and coarticulation changes the shape of the tongue during the production of /p/ in the /ptk/ segment. The tongue is still raised towards the velum during the slower repetitions, however at the faster production it is slightly less raised. There is also a large variation indicated through the dashed standard deviations lines for both the slow and fast productions. While it is difficult to make firm conclusion about the tongue shape during the production of /p/, as the tongue is not an active articulator for the production of this sound, it is still worth noting the increased variability as this can indicate speech motor differences (Sudgen and Cleland, in press).

Figure 37: *Slowest and Fastest Production of /t/ single syllable*



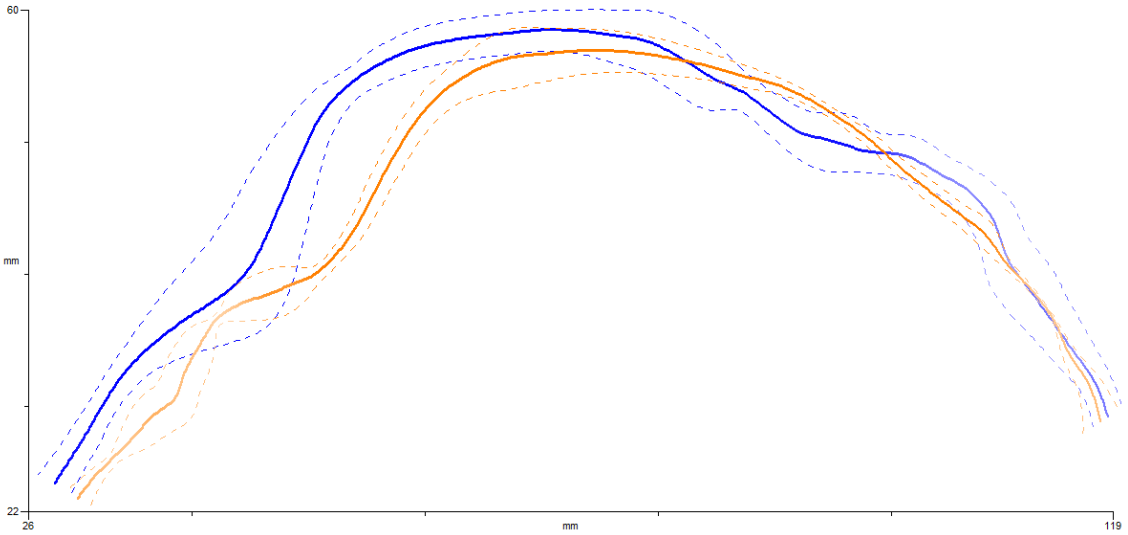
Note. Blue line is the slowest production, and the orange line is the fastest. The solid lines indicate the mean, and the dashed lines indicate the standard deviations.

For the statistical analysis of the tongue shapes, a significant difference was found between the tongue shape produced for /t/ in this context. This is despite no difference being identified in the auditory phonetic transcriptions, however there is significant variation within this faster production of tongue shape. Furthermore, there appears to be retroflexion of the tongue at the slower production. It is worth noting that Sam appears to produce a tongue shape more within the norm at during the

faster production than the slower. There is also a significant variation of tongue shape across the targets, particular at the slower production.

Figure 38: *Slowest and Fastest Production of /t/ in /tk/ segment*

Note. Blue line is the slowest production, and the orange line is the fastest. The solid



lines indicate the mean, and the dashed lines indicate the standard deviations.

In the context of /t/ the increased motor complexity of the /tk/ segment creates an evident difference between the slower and faster productions, more so than the /t/ single syllable which is not present in the faster production. Additionally, during the slower production, the tongue is retracted. There is also a large standard deviation between repetitions for the slower production. It appears that Sam has reduced speech motor control during the slower productions, which is unexpected due to the theory of the speed-accuracy trade off where we would expect with increased speed, a loss in accuracy and an increase in variability (Lammert et al., 2016).

5.6.4 Summary: Sam

Sam's performance for the behavioural assessments in language and cognition were within the norm, however, he presented with a motor profile out with the norm, with greater differences being found in his fine motor control. From perceptual speech assessments, no speech sounds errors were identified, and he was above the mean for both the ASD and TD groups. Furthermore, his performance on rate, accuracy and consistency were above average for both the ASD and TD groups. Sam's speech profile was within the norm when assessed perceptually. However, covert differences were found in the ultrasound tongue analysis. Contrary to expectations,

Sam was out with the norm for his tongue shape in the slower productions, for /t/ and evidently in /k/. It appears that an increase in speech increased the accuracy and reduced the variation of speech targets. This indicates that Sam may be using an alternative speech strategy when producing sounds that was not identified in the perceptual assessment. This is perhaps an adaptation from differences in speech motor control, further evidenced from wide variability in speech targets, an indicator of speech motor control differences (Kotz & Schwartze, 2016). This is interesting due to Sam's motor performance that was out with the norm and could be linked to the differences found in his speech motor performance. This would require further investigation to understand if there is a link between his motor performance and speech performance. However, it indicates that there may be a subset of children within the ASD diagnostic category that have alternative speech strategies, related to their motor performance.

5.7 Case Study 4: Jacob

Jacob was selected for a case study due to both his motor and speech profiles being within the norm from the assessments carried out. This allowed investigation of whether covert speech differences or alterative speech strategies could be identified within his speech profile, which was not identified from perceptual speech assessment.

Jacob was ten years and five months at the time of data collection. He completed all behavioural and speech assessments. Table 31 presents his results in which he was within the norm for all speech assessments, language, non-verbal cognition, and movement. In terms of his motor performance, Jacob was the only child in the ASD group to score within the norm with a score of 8 which is categorised as "no movement difficulty detected".

5.7.1 Perceptual Speech Assessment

Jacob was above the mean for both the ASD group in his percentage consonants correct for both the DEAP (Dodd, 2002) and CUW (James, 2009). He was fully consistent on the DEAP (Dodd, 2002) and was within the norm in consistency for the CUW (James, 2009). Two speech errors were identified that would be classified as

speech delay: one instance of cluster reduction and one instance of final consonant deletion. The only speech error that is not expected in typical development of speech was the medial consonant deletion of /l/ in the multisyllabic word “ambulance.”

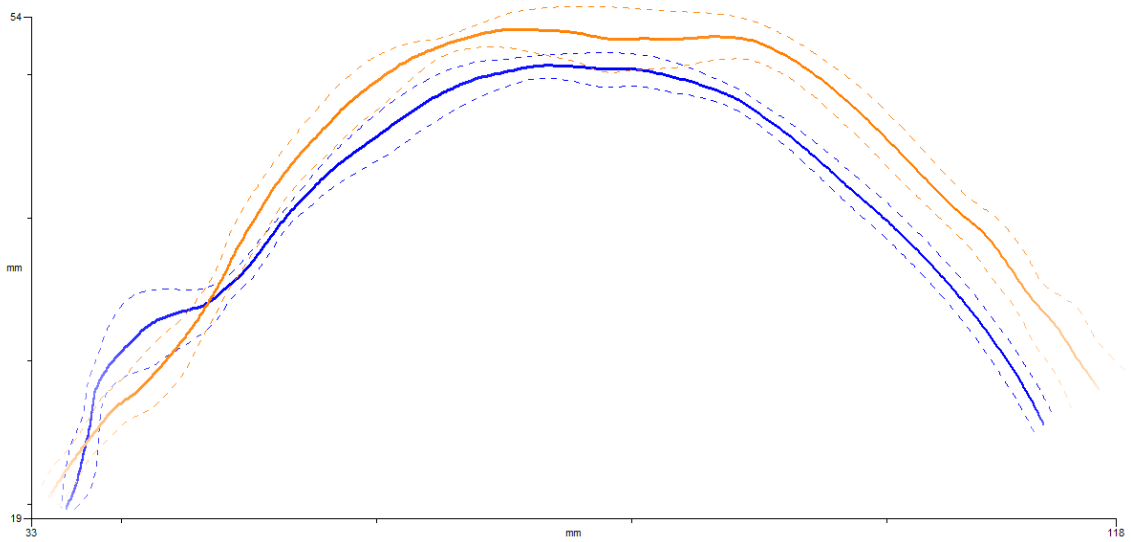
5.7.2 Diadochokinesis Task: Jacob

Jacob’s results for rate, accuracy and consistency can be found in table 32-34. Jacob’s maximum DDK rate was within the norm (Fletcher, 1978). There was no significant difference in performance on single syllable and sequences, showing that increased motor complexity in speech did not have an impact on performance. This was also the case with the accuracy of the production of the first syllable in which Jacob was fully accurate, above the mean for the ASD group and within one SD of the TD group. However, Jacob had high inconsistency on his performance of the DDK task and was significantly below both group means. He produced 44.4% of the single syllable targets consistently and 36.4% of sequences consistently. Inconsistency is a symptom of Childhood Apraxia of Speech (CAS), a neurological childhood speech disorder in which the consistency and accuracy of speech are impaired in the absence of neuromuscular deficits (Shriberg et al., 2011).

5.7.3 Ultrasound Tongue Imaging: Jacob.

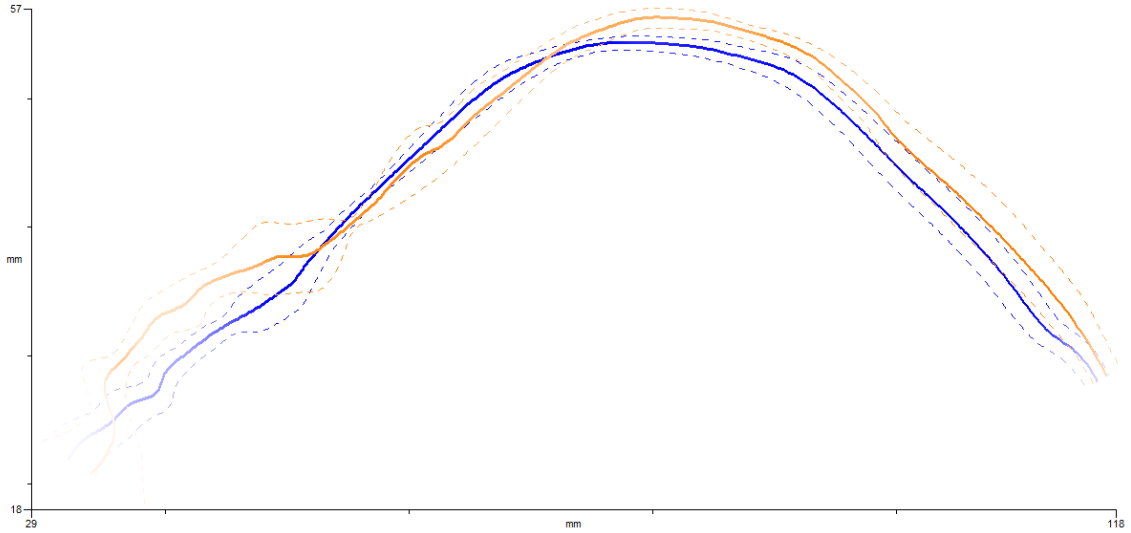
From the perceptual assessments and the DDK tasks, Jacob presented with a speech profile that was within the range for both the ASD and TD group but had high inconsistency in the DDK task for both single syllables and sequences. When carrying out the ultrasound analysis, a significant difference was found for four out of eight of his DDK speech targets. Each of these targets with significant differences are shown in Figure 39 – Figure 43.

Figure 39: *Slowest and Fastest Production of /p/ single syllable*



Note. Blue line is the slowest production, and the orange line is the fastest. The solid lines indicate the mean, and the dashed lines indicate the standard deviations.

Figure 40: *Slowest and Fastest Production of /p/ in /ptk/ sequence*

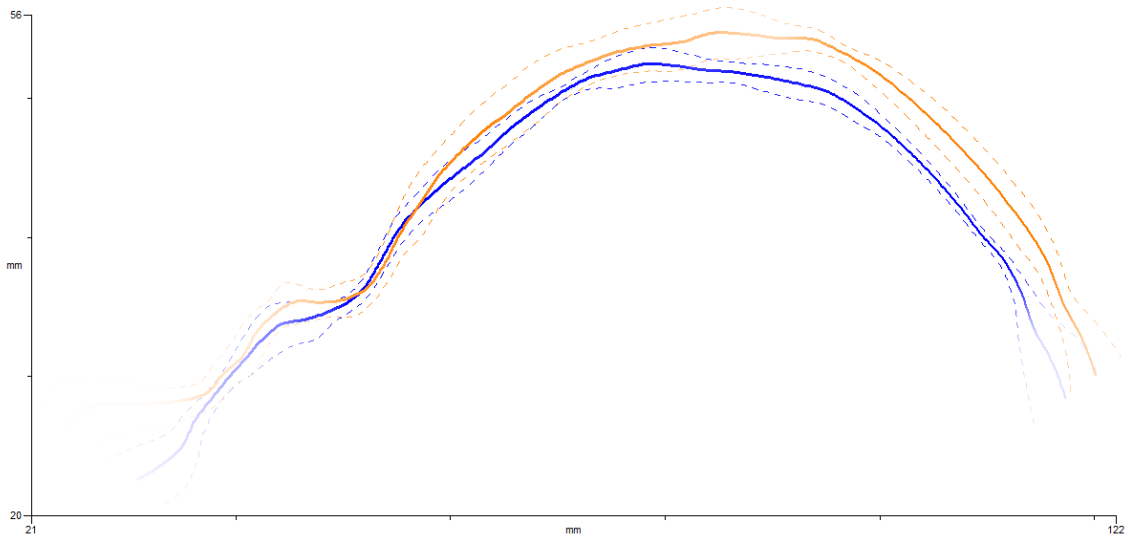


Note. Blue line is the slowest production, and the orange line is the fastest. The solid lines indicate the mean, and the dashed lines indicate the standard deviations.

In both instances of the target /p/, both as a single syllable and in the /ptk/ sequence, there is a significant difference between the slowest and fastest production. In both cases the tongue more raised at the fastest speed, this is more exaggerated in the

/p/ single syllable targets, where there is also higher variability across targets. It appears that the coarticulatory effect of the other consonants present in /ptk/ sequence enables a more consistent tongue shape.

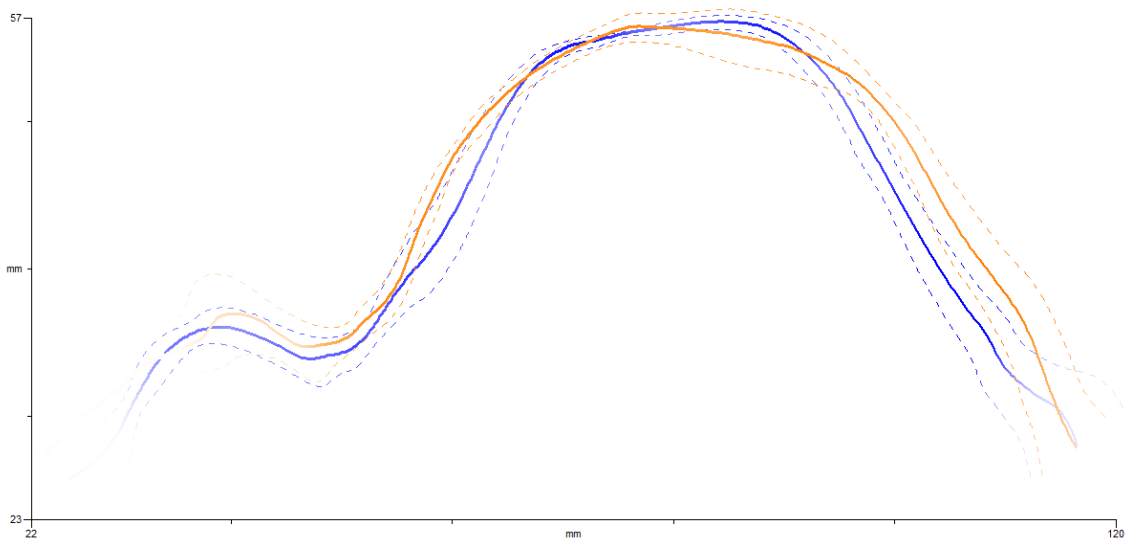
Figure 41: *Slowest and Fastest Production of /t/ in /ptk/ segment*



Note. Blue line is the slowest production, and the orange line is the fastest. The solid lines indicate the mean, and the dashed lines indicate the standard deviations.

A signficant difference was found between the slowest and fastest production of /t/ in the /ptk/ segment. The tongue is higher and further forward in the faster production, thought the tongue shapes remain relatively the same, which is why it was likely not identified in perceptual assessment.

Figure 42: *Slowest and Fastest Production of /k/ single syllable*



Note. Blue line is the slowest production, and the orange line is the fastest. The solid lines indicate the mean, and the dashed lines indicate the standard deviations.

While there was a significant difference found between the slower and faster productions of /k/ single syllable target, with a wider variation of the tongue in the mouth at the faster production, the tongue shapes remain relatively the same and would not be identified as an overt speech error. These differences may be down to speed-accuracy trade-off and not necessarily due to a speech motor control difficulty (Preston & Edwards, 2009).

5.7.3 Summary: Jacob

Jacob was selected as a case study due this his speech profile and motor profile being within the norm. The purpose was to identify any covert speech errors or differences using ultrasound tongue imaging that were not identified using perceptual speech assessment. The perceptual analysis of the DDK task showed a high level of inconsistency across targets, both at the single syllable and sequence level. On further analysis of these targets using ultrasound tongue imaging, four out of eight of the targets had a significant difference in tongue shape. All four showed a wider variation of tongue shape at the faster production. This inconsistency is likely due to a speed-accuracy trade-off that could be further exaggerated by speech motor

control differences (Preston & Edwards, 2009). However, this difference would not be significant as it does not impact the perception of Jacob's speech.

s

5.8 Case Study 5: Emma

Emma was chosen for a case study due to having speech and motor profiles within the norm. She also presented with a different behavioural profile to Jacob, which may provide avenues for future research in terms of subtypes of symptoms within the ASD diagnostic category.

Emma was ten years and seven months at the time of data collection. She completed all the behavioural and speech assessments. Table 31 presents her results in which she was within the norm for speech assessments and non-verbal cognition. In terms of her motor performance, Emma presented with borderline or mild differences. She scored 6 in the MABC-2 which is categorised as being within the "amber zone" in which she is classified as "being at risk of having a movement difficulty, monitoring required" (Brown & Lalor, 2009). The greatest differences in her motor performance were found in the balance subtests, measurements of gross motor ability. Emma also presented with "marginal/borderline" language differences in the CELF (Semel et al., 2003).

5.8.1 Perceptual Speech Assessment: Emma

Emma was above the mean for both the ASD group in her percentage consonants correct for both the DEAP (Dodd, 2002) and CUW (James, 2009). She was fully consistent on the DEAP (Dodd, 2002) and the CUW (James, 2009). She produced two speech sound errors: one instance of velar fronting in this case, 'ng' is replaced by /n/ in the word "fishing" in the DEAP (Dodd, 2002). This speech sound errors would be classified as a speech delay, as it is expected in speech development but to be remedied by age three to four years (Flipsen, 2015). There was also one instance of consonant harmony, in which the pronunciation of the whole word is influenced by the presence of a particular sound in the word (McLeod & Baker, 2017). In Emma's case, it was the influence of /t/ within the word "hippopotamus" where she produced /p/ as /t/. This speech sound errors would be classified as a

speech delay, as it is expected in speech development but to be remedied also by age three to four years (Flipsen, 2015).

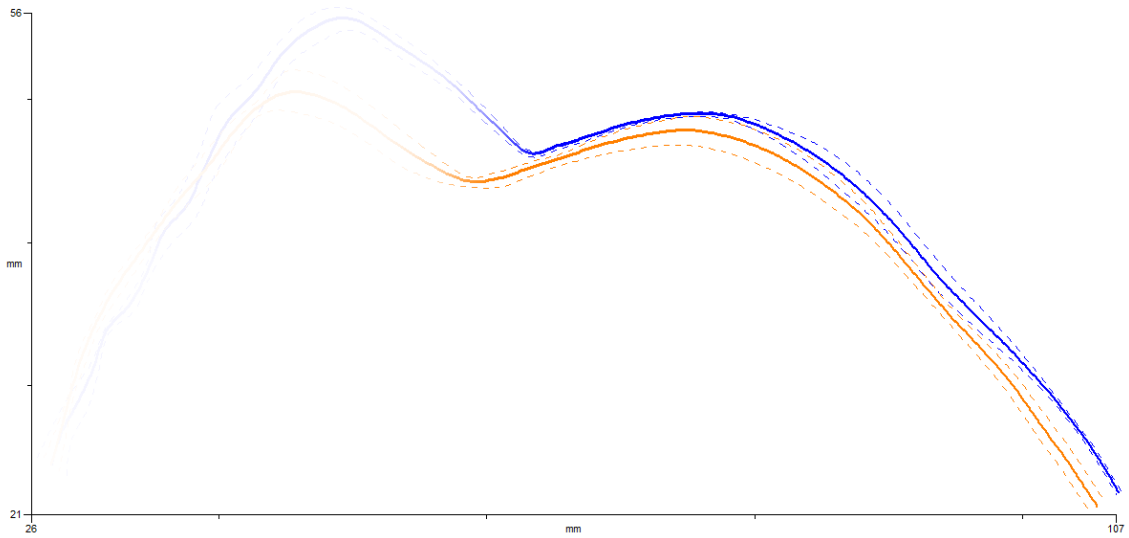
5.8.2 Diadochokinesis Task: Emma

Emma's results for rate, accuracy and consistency can be found in table 32-34 Emma's accuracy of production of the first target was within the norm (Fletcher, 1978). There was no significant difference in performance on single syllable and sequences, showing that increased motor complexity in speech did not have an impact on accuracy. This was also the case with the consistency which Emma was fully accurate, above the mean for the ASD group and within the mean for the TD group. However, Emma had a significantly slower rate on her performance of the DDK task and was significantly below both group means. DDK rates increase with age and slow DDK rates may be indicative of speech disorders (Williams & Stackhouse, 2009).

5.8.3 Ultrasound Tongue Imaging: Emma.

From the perceptual assessments and the DDK tasks, Emma presented with a speech profile that was within the norm for both the ASD and TD group with two instances of speech sound errors occurring. Her accuracy and consistency during the DDK tasks were within the norm. However, she a significantly slower rate than compared to the other children within the ASD group and was out with the norm. When carrying out the ultrasound analysis, a significant difference was found for three out of eight of her DDK speech targets. Each of these targets with significant differences are shown in Figure 43 – Figure 45.

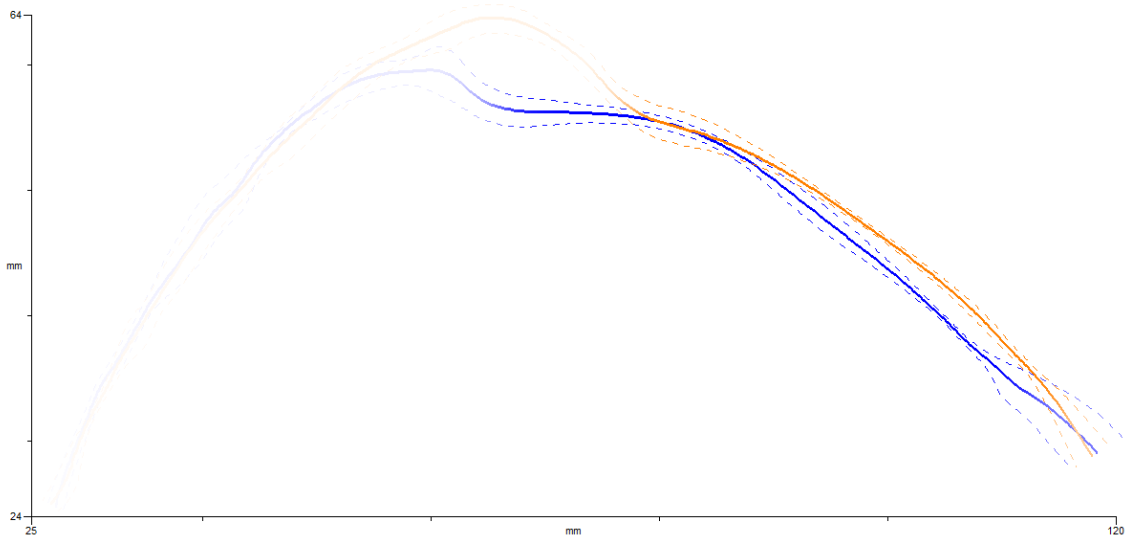
Figure 43: *Slowest and Fastest Production of /p/ in /ptk/ sequence*



Note. Blue line is the slowest production, and the orange line is the fastest. The solid lines indicate the mean, and the dashed lines indicate the standard deviations.

Emma's production of /p/ within the /ptk/ segment had a tongue shape that was within the norm Lawson, Stuart-Smith, Scobbie and Nakai. (2018). There was a significant difference between the tongue shapes at the slowest and fastest production, with the faster production being more retracted and perhaps more constrained due to the increased speed. This does not indicate a speech motor control issue but is an effect of speed of speech production. The image in the ultrasound was also rotated which has altered the diagram, with the tongue rotated to the right.

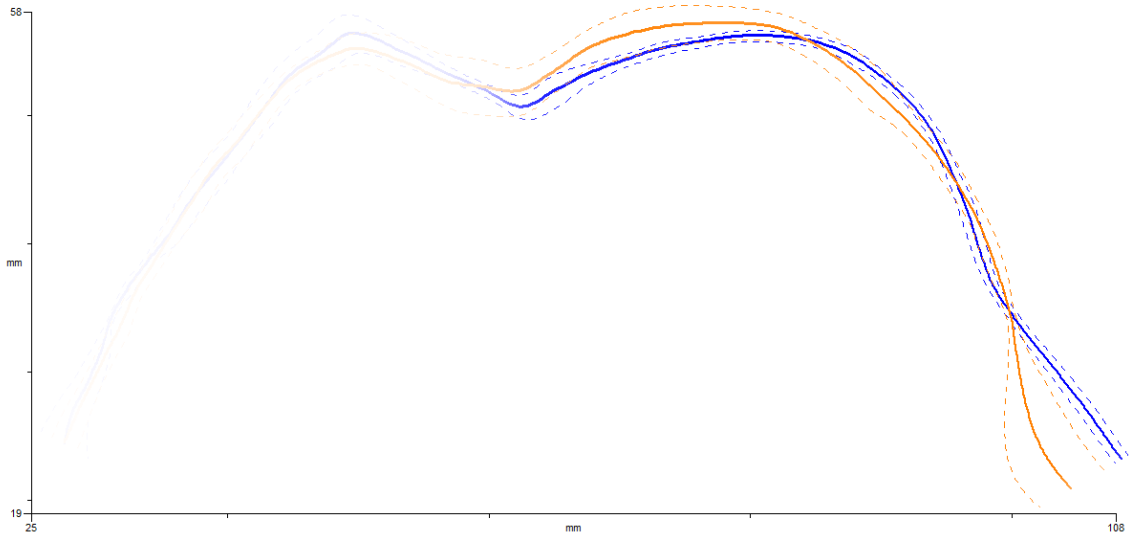
Figure 44: *Slowest and Fastest Production of /t/ single syllable*



Note. Blue line is the slowest production, and the orange line is the fastest. The solid lines indicate the mean, and the dashed lines indicate the standard deviations.

In Emma's case the tongue tip is lower than expected for the production of /t/, however the tongue body contact with the palate and teeth are in the expected position. However, it is more likely that the image is rotated when the ultrasound recording was carried out, a limitation of the procedure. There was also significant variation between the tongue shapes at the slowest and fastest productions. The differences are not surprising due to Emma's differences found in the measurement of rate during the DDK task.

Figure 45: *Slowest and Fastest Production of /k/ single syllable*



Note. Blue line is the slowest production, and the orange line is the fastest. The solid lines indicate the mean, and the dashed lines indicate the standard deviations.

Emma's production of /k/ single syllable is similar to the expected tongue shape for a velar plosive (Lawson et al., 2018). The differences between tongue shape were statistically significant but when looking at the diagram, there is minimal variation in tongue shape between the two speeds. As with other recordings for Emma, the image is rotated to the right.

5.8.4 Summary: Emma

Emma was chosen as a case study due to her speech profile and motor profile being within the norm, with some differences in language. The purpose was to identify any covert speech errors or differences using ultrasound tongue imaging that were not identified using perceptual speech assessment. The perceptual analysis of the DDK task showed a reduced rate during the production of single syllables but not during sequences, indicating that additional motor complexity of sequences was not impacting rate. On further analysis of these targets using ultrasound tongue imaging, three out of eight of the targets had a significant difference in tongue shape. All three showed a slight variation in tongue shape, while statistically significant, they would not have an impact perceptually. No significant covert errors were identified using the ultrasound tongue analysis for Emma.

5.9 Influence of Cognition

According to the Leiter international performance scale (Leiter; Roid et al. 2013) Sophie, Harry and Emma have a score that fall into the category of a mild to moderate intellectual differences. However, the profiles vary on other behavioural aspects amongst these children. Sophie and Harry have scores below the norm in speech, movement and language, indicating they may be presenting with an overall developmental delay. Whereas for Emma, her language and movement are below the norm, but speech remains intact. Looking more in-depth at their speech performance, Sophie and Harry were the only children in the group to perform below the norm in accuracy of sequences in the DDK tasks. This is notable because it suggests a difference in their accuracy of speech motor tasks compared to the rest of the group. Sophie's speech indicated extra speech processing and timing required to produce multisyllabic words as well as an inconsistent vowel strategy. Harry may have a general developmental delay that impacts his speech, language and movement as well as producing a mild speech impairment. Emma had a reduced speech rate during production of single syllables but not during sequences. There was no sign of covert errors in ultrasound analysis.

Many individuals with intellectual disability encounter severe problems in acquisition of language and communication, also impacting speech development (Vandereet et al., 2011). The speech delay present on both Sophie and Harry's speech may be relative or attributed to their differences in cognitive performance. The reduced performance in both the DEAP (Dodd, 2002), CUQ (James, 2009) and DDK tasks for Harry and Sophie may be related to this. However, for Emma, who's speech performance was within the norm, except for a reduced speech rate during production of single syllables, is not indicative of a general delay but a specific difference in speech motor performance. When looking at the language profiles of children with ASD compared to children with intellectual disability (ID). Cleland et al. (2010) found in individuals with Down's syndrome that deficits in receptive and expressive language were not fully accounted for by their cognitive delay. The majority of speech errors were developmental in nature but all of the children with Down's syndrome shower at least one atypical or non-developmental speech error.

Looking at the specific speech errors produced by Sophie, Emma and Harry, Sophie was had a speech profile marked with SSEs indicative of speech delay (stopping, postvocalic devoicing, fronting, cluster reduction, epenthesis, metathesis, assimilation) however did have on type of SSE (backing) indicative of speech disorder. Harry had a produced SSEs that aligned with a diagnosis of speech delay (velar fronting, stopping, labialization). Whereas Emma only produced two SSEs (velar fronting and consonant harmony) that would fall into the category of delay and not disorder. It is possible that the differences in cognition had a major influence on the speech production of these three children. Harry speech profile is indicative of a general speech delay, which is consistent with his behavioural profile. Sophie has also evidence of a general delay across the different behavioural domains but also speech markers that are disordered and not necessarily associated with developmental delay. Emma has little evidence of a general developmental delay impacting her speech, and one difference in speech motor performance. It is possible that speech motor differences were not a result of a general developmental delay and have closer ties to their diagnosis of ASD.

Chapter Six Concluding Discussion

6.1 Key Findings of the Thesis

Evidence from the literature has shown that individuals with autism spectrum disorder (ASD) can experience higher rates of speech sound errors (SSEs) than their peers (Cleland *et al.*, 2010; Shriberg *et al.*, 2011) yet the reasons why are unknown. This thesis aimed to determine whether the higher rates of SSEs found in children with ASD were a result of a speech motor disorder and whether this was related to a general movement disorder. This study used varied analysis techniques, both behavioural and instrumental, to determine if children with ASD produce significantly higher rates of SSEs than typically developing (TD) children and if there were any correlations with movement, language, and non-verbal cognition.

6.1.1. Research Question 1: Do children with ASD produce significantly more speech sounds errors (SSEs) compared to typically developing children?

The first research question aimed to augment current evidence of the characteristics of SSEs in children with ASD aged 6-12 years. This was a response to the paucity of information in the current literature relating to speech sound production in school-aged children with ASD. This was addressed in part one of the study by carrying out speech sound production analysis using clinical speech tests that assess speech at the single word level (Diagnostic Evaluation of Articulation and Phonology; Dodd, 2002) and in more motorically complex multisyllabic productions (Clinically Useful Words; James, 2009).

The first key finding in relation to the first research question is that there were no significant differences in DEAP (Dodd, 2002) percentage of consonant correct (PCC) scores between the ASD and TD group. However, when analysing speech errors by categories there was a difference of the number of total errors and then specifically delayed errors produced by the ASD compared to the TD group. The ASD group produced significantly more delayed SSEs than the TD group. There were no significant differences in CUW multisyllabic assessment (James, 2009) percentage of consonant correct (PCC) scores or in the categorisation of speech sound errors

between the ASD and TD group. The significant number of delayed SSEs and close to significant unusual SSEs present in the ASD group compared to the TD group is similar to findings made by Shriberg et al. (2011) and Cleland et al. (2010). In this sample I found that 90% of the ASD group produced some type of SSE, this is in comparison to the prevalence of speech errors at 8 years of age in the general population which is only 7.9% (Wren *et al.*, 2016). So, from my sample of children with ASD I was able to conclude that there were significantly higher rates of delayed SSEs, for example a significant number of children within the ASD group presented with cluster reduction and fronting.

6.1.2 Research Question 2: Does instrumental analysis of speech reveal subtle articulation differences between ASD and TD groups?

The aim of the second research question was to conduct a preliminary investigation into the accuracy of standard perceptual and instrumental assessment approaches to measure speech sound production in children with ASD. This was carried using the diadochokinesis (DDK) task, a maximum performance task that analyses syllables and sequences of speech sounds at increasing speeds. Standard perceptual analysis was carried out to determine the rate, accuracy and consistency of speech sound production in this task followed by analysis using ultrasound tongue imaging in which variation of tongue shape was analysed across and within the two groups. When speech analysis was carried out perceptually with this task, there were no significant differences in the rate, accuracy or consistency of production between the ASD and TD groups. There were more instances of children in the ASD group being below or significantly below the norm in the rate of DDK productions according to norms from the literature (Fletcher, 1978) indicating that there may be a slower rate of production in DDK detected at perceptual and acoustic analysis. However, this was not evident when comparing directly with the TD group and not statistically significant.

Instrumental analysis of the DDK tasks using ultrasound tongue imaging showed some subtle articulation differences in tongue shape variance between the ASD and TD group but not in the hypothesised way. The TD group had more significant

difference of tongue shape in the more motorically complex sequences (ptk) than the ASD group. The instrumental analysis of the DDK tasks was able to show us subtle articulatory differences, which were not found in the perceptual and acoustic analysis of the DDK data.

6.1.3 Research Questions 3: Do children with ASD present with speech motor impairment symptoms?

The third research question aimed to address the theoretical understanding of higher rates of SSEs in children with ASD by addressing whether the SSEs or the lower performance in the DDK task indicated if there was a presence of speech motor impairment and/or whether SSEs were indicators of difficulties in speech attunement. This was addressed by analysing speech at both the perceptual level using the DDK tasks and at an instrumental level using ultrasound tongue imaging to identify variation of tongue shape between repetitions. This was further highlighted in the case study of “Sophie” in which a speech pattern of a speech motor delay was further highlighted in the observations of tongue shape variation in combination with results from the phonetic analysis of her speech. Further analysis was carried out using the mean syllable duration of syllables and sequences produced by children with ASD compared to TD children.

In relation to the maximum performance tasks (diadochokinesis – DDK) there were no significant differences in the rate, accuracy, or consistency of production between the ASD and TD groups. There were more instances of children in the ASD group being below or significantly below the norm in the rate of DDK productions (Fletcher, 1978) which is the only indicator that speech motor control may have been impacted in the ASD group but due to the small sample size and the lack of significant difference with the TD control group, this is not conclusive. Another interesting finding related to this research question is that the TD group had more significant variation in tongue shape in the more motorically complex segments (ptk) than the ASD group. Token-to-token variability has been found to be larger in children than adults. Maturation of speech motor control is a long developmental process which has been found to only reach maturity in late adolescence (Walsh and Smith, 2002;

Smith, 2010). Consistency of tongue shape at differing speeds is a measure of motor control (Zharkova, Hewlett and Hardcastle, 2011) and the ASD appears to have a more rigid and less variable speech pattern than the TD group. This rigidity has also been shown in their general movement, particularly in their fine motor control (Anzulewicz, Sobota and Delafield-Butt, 2016). This rigid consistency is not necessarily a “better performance” made by the ASD group but rather expresses differences in the complex speech motor actions as performed by this group and the dynamic capability in speech of children with ASD should be further explored. These differences may be accounted for by brain tissue differences that affect behaviour, cognition and motor functions as shown in nonlinear signal processing techniques like sample entropy which characterizes temporal dynamics of brain connectivity (Maximo et al., 2021). It has been stipulated that increased entropy has been found in adults with ASD in regions such as frontal temporal and parietal lobe, corpus callosum and hippocampi (Maximo et al., 2021; Tummala, 2019). However, neuropathological mechanisms contributions to differences in motor presentation in ASD remain unclear and could also be a result of genetics or differences in brain development (Tummala, 2019).

The lack of evidence of speech motor difficulty from the DDK tasks but a significant general motor differences in this sample of children with ASD seems at odds. However, there has been an ongoing debate in relation to the special status of speech in the domain of general motor control (Ballard, Robin and Folkins, 2003; Ziegler, 2003b, 2003a; Mayer, Hannent and Heaton, 2016; Maas, 2017). Maas (2017) conducted a review of the literature examining the two differing views; the task dependent model which stipulates that speech production uses a specialised neuromotor system that is dedicated to the task of speech (Ziegler, 2003b, 2003a; Ziegler and Staiger, 2016) or the integrative model in which speech production shares the properties and skills required of other motor behaviours and that there is an overlap in the neural control systems required for these (Ballard, Robin and Folkins, 2003). Findings here indicate an intact speech motor control system despite a significantly impaired general motor system in this group of children with ASD. This lends support to the task-dependent model and following the review by Maas (2015) is in line with what is representative of the prevailing view in the current literature.

Inconsistency in speech sound production has been evidenced as a core symptom of childhood apraxia of speech (CAS) (American Speech-Language-Hearing Association (ASHA), 2007), a motor speech disorder that is caused by difficulty in programming motor commands that control speech articulators (Shriberg et al., 2012). In my sample of children with ASD, there were no significant differences in the inconsistency scores both in the single word and multisyllabic speech tests carried out. Indicating that the higher rates of SSEs are likely not due to CAS or a speech motor impairment in this sample of children with ASD.

In relation to the mean duration of syllables, the TD group had longer meant durations and a larger standard deviation than the ASD group. As these differences occurred at the slowest production it implies that these differences are not the result of a speed-accuracy trade off seen at faster production but a difference in rhythm of speech production. Interestingly, the TD group produced a significantly slower mean duration of both the /p/ single syllable at both the slowest and fastest conditions in comparison to the ASD group. No other significant differences in other targets were found.

A theory for the lack of tongue shape variability and these shorter mean duration of syllables in comparison to the TD group may have been an altered rhythmic presentation in the ASD group. However, auditory-motor rhythm synchronization has been shown to be relatively intact in children with ASD (Tryfon *et al.*, 2017). In a group of thirty-one boys with ASD compared to 23 TD boys both groups performed similarly on measures of precise temporal internal reproduction, tap synchrony, bias toward early or late response or coarse-level production of rhythmic patterns. The auditory-motor integration was relatively intact. Instead, the issue may lie in the auditory perception of speech signals. Lin et al. (2015) found speakers with ASD had an atypical delayed auditory feedback effect on speech production and Russo et al. (2008) found that individuals with ASD had an atypical audio-vocal system regulation, this could require speakers to “learn” speech patterns, relying less on direct auditory feedback and more on the auditory plans already existing.

The perspective that may explain the higher rates of delayed SSEs in my sample of children with ASD is the “speech attunement framework.” This framework posits that

when a child is learning speech, they need to attend to their ambient speech environment, also known as “tuning in”. They also need to “tune up” which involves making careful and small adjustments to their speech sound production in order to sound like the speech in their ambient environment, which is to modify speech to sound like the children around them (Shriberg et al., 2011).

The ability to “tune in” and “tune up” as posited by the speech attunement framework may be impacted in individuals with ASD in the following ways, first an enhanced auditory capacity, often observed in individuals with ASD (Baum, Stevenson, and Wallace, 2015) may lead to earlier “tuning in” when motor maturity has not been achieved. Therefore, SSEs develop due to motor constraints. Second, constraints in affective social reciprocity, a common trait of people with ASD (Chevallier *et al.*, 2012) may delay “tuning in” and any motor speech disorder present may impair the ability to tune up. A lack of social motivation may account for why these SSEs then do not remediate and persist into late childhood and beyond. The following indicators of speech attunement issues forward as a result of this study (Shriberg et al., 2011); firstly, an increase in repetitions and revisions, consistent with the description of autistic speech as “disfluent”. Second, misplaced stress often described as “off” or “singsong” (Peppé, McCann, Gibbon, O’Hare, & Rutherford, 2007). This stress is dissimilar to the well-documented “excessive-equal” stress pattern in apraxia of speech. Further there is inappropriate loudness and pitch and higher rates of speech delay and speech errors relative to population estimates.

Whilst this study has not covered the scope of the measurements that would allow us to make a judgement on these four signs, I can confirm that there were higher rates of speech delay and speech errors relative to population estimates in the absence of speech motor impairment but at a group level there was no speech motor impairment generally noted as measured by the DDK task. My group of children with ASD did not present with significantly slowed speech rate, lengthened syllables or uncommon phoneme distortions that define motor speech impairment (Duffy, 2000).

6.2 Clinical Implications

The results of this study and others described earlier (Cleland, Gibbon, et al., 2010; Shriberg et al., 2011) indicate that this population do present with higher rates of SSEs, in my case delayed SSEs, and this warrants speech perception and speech production assessments to be part of the core battery of assessments carried out in clinic with these children. If SSEs are found these should be included in a holistic speech and language therapy plan in which all parts of the child's social-communication skills are taken into account and worked into intervention. It is important that any therapy is centered around the child's ability to participate in their social environment.

The findings from my study also indicate that children with ASD should be assessed for motor skills for speech tasks separately from motor skills for non-speech related tasks. This sample of children with ASD had significant general motor impairment yet this was not reflected in their speech motor skills. Therefore, it should not be assumed that these two are interlinked and a presentation of impairment in one result in the impairment of the other. It is vital that speech and language therapists (SLTs) carry out oral motor assessment separately to speech motor tasks such as maximum performance tasks and these should be interpreted independently (Murray *et al.*, 2015).

Furthermore, it is vital when assessing the speech of children with ASD (and other populations) that it is assessed in multiple speech contexts. Some of the significant results found in this assessment were only identified in the context of sequences of the DDK task, which were more motorically complex than the syllables. Furthermore, having compared the DEAP (Dodd, 2002) and the clinically useful words assessment, an assessment of multisyllabic words (James, 2009) provided indicators that there were no significant differences between the more motorically complex speech sound production skills of the ASD and TD group, potentially ruling out a potential speech motor impairment. On a group level. This was a short, easy to administer assessment that could be included in the battery of assessments SLTs carry out in clinics that may help provide further information about the cause and nature of SSEs.

This research also sought to understand if the use of the instrumental analysis technique, ultrasound tongue imaging confirmed or disconfirmed information about the SSEs and speech motor performance of children with ASD and whether it provided additional information to the perceptual and acoustic analysis. Interestingly, subtle articulatory difference was indicated but within the TD group rather than the ASD group. These were confirmed by quantitative measurements, using paired t-tests to identify any differences in tongues shape across multiple repetitions on the DDK task. This unexpected finding helped conclude that there are likely no speech motor issues in children with ASD, which was not evident from the perceptual and acoustic analysis alone. There is a potential role for ultrasound tongue imaging in the analysis of speech with children with ASD and other populations and it may be an invaluable tool for increasing my understanding of the nature and causes of SSEs in different populations.

6.3 Strengths of the Study

This study of the speech production of children with ASD and other cognitive abilities using behavioural, perceptual, and instrumental assessments makes a novel contribution to the field in terms of my understanding of SSEs in this population. First the comprehensive study of the DDK task using standard analysis as well as ultrasound tongue imaging has shown that ultrasound is an effective tool for identifying subtle articulation differences between groups and that at least within this sample of children with ASD, there was no evidence of a speech motor impairment, adding evidence to the ongoing debate of the cause of higher rates SSEs in this group (Belmonte et al., 2013; Shriberg et al., 2011). Second, due to the range of behavioural assessments (language, non-verbal IQ, movement, and speech) I was able to find correlations between speech sound production, movement, and language. Third, including the data from the TD group allowed us to compare individual performances on the speech production tasks and determine if there was a difference between the two groups. This revealed that TD showed more variation in tongue shape and longer mean duration of syllables than the ASD group. The combination of these three strengths has resulted in a better understanding of the

nature of SSEs in children with ASD and the role of perceptual and instrumental assessments in this field.

6.4 Limitations and Future Research

Despite the comprehensive nature of assessment within this study, there are still a number of limitations that require to be discussed alongside advice for future research.

6.4.1 Sample

Due to significant difficulties in recruitment to this study because of recruitment difficulties out with control of this project, the number of participants is small and affects the significance of statistical power. While my results provide indicators of speech sound behaviour in children with ASD, this is not generalisable to the whole population and still requires significant research. Therefore, the findings of this research should be replicated with a larger population of children with ASD compared to TD children in the search for subgroups within ASD in relation to speech profiles. Therefore, the case study was carried out with Sophie in order to observe whether there were subtypes within the ASD group. This showed that Sophie potentially had a speech motor problem, which was not evident at the group level. This highlights the importance of when observing speech in large groups of children with ASD, to account for individual differences within those groups. The number of participants and the variation of age and gender was limited by the need to recruit numbers into the study of children whose parents had time and capacity to participate in the study as well as meeting the inclusion criteria. A larger sample of children with ASD with a larger distribution in gender and age may have been recruited if NHS pathways for recruitment to the study had been available. In a future study, work should be done to establish better links between research and NHS clinicians to strengthen findings with a larger, more equally distributed sample.

6.4.2 Assessment Tasks

Whilst a full battery of speech, language, non-verbal and movement assessments were carried out with the children with ASD, as a result of time constraints and the significant number of assessments carried out, I did not carry out an oro-motor assessment and instead verified there were no structural issues through parent reports and with the exclusion criteria. This means I was not able to carry out correlation analysis between non-speech motor functioning of the articulators in relation to general motor skills. In a future study, oro-motor assessment could be included alongside the DDK tasks to determine when the function of speech is removed, whether this impacts performance from an ASD group.

While multiple speech contexts were assessed within this study, due to time constraints of analysis, connected speech data was not collected either from picture descriptions or conversation. In any future study this data should be collected and possible analysed using ultrasound tongue imaging to determine whether there are subtle articulatory differences or SSEs not identified in standard clinical speech assessments from both the experimental group and a group of TD children for comparison. Furthermore, a full assessment of the DEAP (Dodd, 2002) should be carried out. Due to time constraints for this study, only the screening version of the DEAP (Dodd, 2002) was carried out which contains a limited number of words. Carrying out a full DEAP (Dodd, 2002) may reveal a higher rate of SSEs in the ASD group.

6.4.3 Ultrasound Recording

While ultrasound tongue imaging has a considerable number of advantages for assessing tongue shape during speech, the recoding procedure can cause difficulty when working with children. When carrying out the recording process, the participant is required to sit still to ensure the scanning view remains the same throughout the recording. The ultrasound probe is fixed under the participants chin using a headset which can be stressful for children with conditions such as ASD. Therefore, flexibility was required with every child that participated in my sessions, all the children wore the headset, but some were unable to stay sitting still throughout the session

resulting in a loss of data that may have contributed to the lack of significance in some of the ultrasound and DDK measures.

6.4.4 Research with Adults

Studies with adults have also shown speech differences of adults with ASD and neurotypical adults. Shriberg et al. (2001) found residual SSEs in 33% of a sample of adolescents and adults with ASD compared to a TD sample in which only 1-2% presented with residual SSEs. Additionally, Kissine and Geelhand (2019) carried out a syllable-level analysis on speech data (narrative and spontaneous speech). They compared twenty adults with ASD compared with twenty neuro-typical adults. Their focus was on suprasegmental features; fundamental frequency, jitter, shimmer and the first three formants. They found the individuals with ASD showed a greater articulatory stability in their production of vowels. Results suggested the ASD group showed less variability in the vibration of their vocal folds during vowel production. Future research can employ ultrasound tongue imaging as a tool to further explore whether these speech differences are significant and lasting into adulthood. Additionally, it would be possible to carry out longer speech assessments than with children using ultrasound with adults. So, it would be easier to examine speech differences in spontaneous speech and compare with speech motor tasks.

6.5 Summary and conclusion

The results presented in this thesis have extended previous findings and produced a number of novel results that are of value to researchers in the field, clinicians, and teachers working with children with ASD. These results also contribute to the theoretical understanding of why higher rates of SSEs occur in children with ASD and may help advance understanding of speech sound development in this population. These results indicate that while no speech motor impairment was present in this group of children with ASD, there are indicators that there are issues with speech perception, in the form of not being able to “attune effectively to the ambient speech environment, resulting in higher rates of SSEs” (Shriberg, 2011). The presence of a significant motor impairment as well as impairment in language may have further impeded their speech sound development. ASD is a

heterogeneous neurodevelopmental condition and to analyse this group as if it is homogenous may lead to contradictory findings, as found in the literature on speech sound production and development in children with ASD, particularly if perceptual abilities and socio-communicative skills are perceived as stable over time (Valla and Belmonte, 2013; Kargas *et al.*, 2015).

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Appendices

Appendix One – Recruitment Letter for ASD Group

Dear Parent or Guardian,

RE: Children with autism aged 6-12 sought for research project

University of Strathclyde are currently engaged in a study to investigate the relationship between speech, language, and movement in children with autism aged 6-12. I am sending you this letter on behalf of the research team because I think your child might potentially be suitable for the participating in the assessments.

I am holding a parents information session at **2.15 p.m.** at X on **Thursday 31st May.** Please come along to find out more about the project, see the equipment and I will answer any questions you have.

Please read the enclosed information sheet and return the cut-off slip below or email louise.mckeever@strath.ac.uk (the research Speech and Language Therapist) if you are interested in taking part.

If you are interested, please return the information below. I will contact you to arrange a time for you to bring your child to University of Strathclyde or arrange a session within your child's school. You can then discuss whether you would like to take part before making a decision.

Yours sincerely,

Louise McKeever
Speech & Language Therapist
"----------

Speech, Language and Movement in Children with Autism

Child's Name_____ Parent's Name_____

Address _____

Child's Age (Years and Month) _____ Child's GP_____

YES, we are interested in taking part please contact me by phone/ email on_____

Please return to: GH551, Graham Hills Building, 42 George Street, Glasgow, G1 1QE
Or
Email louise.mckeever@strath.ac.ukwith the above information
Or call
0141 548 4393

Appendix Two – Recruitment Letter for TD Group

Dear Parent or Guardian,

RE**: Typically Developing Children** aged **6-12** sought for research project

University of Strathclyde are carrying out a study to investigate speech, language and movement in **typically developing children** aged **6-12** and have no other diagnosis. If your child meets this criterion, please read the parent information sheet provided and decide if you would like to take part.

If you are interested, please contact me with the details below. We can discuss the project in more detail, and you can then decide whether you would like to take part.

I will then arrange with you a time for you to bring your child to University of Strathclyde **or** at your child's school when convenient to you. You are welcome to get in touch to discuss whether you would like to take part before making a decision.

Yours sincerely,
Louise McKeever
Speech & Language Therapist

Speech, Language and Movement in Children with Autism

Child's Name_____ Parent's Name_____

Address _____

Child's Age (Years and Month) _____ Child's GP_____

YES, we are interested in taking part please contact me by phone/ email on_____

Please return to: GH551, Graham Hills Building, 42 George Street, Glasgow, G1 1QE

Or
Email louise.mckeever@strath.ac.uk with the above information

Or call
0141 548 4393

Appendix Three – Parent Information Sheet (ASD Group)

Parent/Carer Information Sheet

A Study of Speech, Language and Movement in Children with Autism Aged 6-12

Your child is being invited to take part in a research study. This study is part of a PhD qualification. Before you decide whether to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully. Talk to others about the study if you wish. Contact us if there is anything that is not clear or if you would like more information. Take time to decide whether you wish for your child to take part.

I am holding a parents information session at **2.15 p.m.** at X on **Thursday 31st May.** Please come along to find out more about the project, see the equipment and I will answer any questions you have.

What is the purpose of the study?
This project aims to see if there is a relationship between speech difficulties and movement in children with autism aged 6-12. Children with autism have been shown to have higher rates of speech errors than children their own age. The accurate production of speech sounds requires intricate coordination of the speech muscles (e.g., the tongue). We hypothesize that the movement difficulties observed in autism may cause difficulties with speech sound production, however, this has not been investigated in a robust way. Understanding if there is relationship between speech and movement in children with autism compared to their peers would increase our understanding of the difficulties faced by people with autism and provide more information on why they make speech errors.

The project will use standard medical ultrasound to record the movements of the tongue during speech. We will compare these results to the results of standard assessments of speech, language and movement used in clinics.

A picture of the ultrasound setup showing ultrasound images of the tongue on a computer screen

More information on ultrasound, including pictures and videos, can be found here:
http://www.qmu.ac.uk/casl/ultra/default.htm

Why has my child been asked to take part?

- Has a diagnosis of autism
- Aged between 6-12 years at the time of the study.
- English speaker
- Do not have a diagnosis of any movement disorders e.g., dyspraxia, developmental coordination disorder
- Have no evidence of severe/profound current hearing loss
- Have no major physical disability or structural abnormality of the vocal tract

Does my child have to take part?

No, it is up to you to decide whether your child takes part. If you do decide to take part, you will be given this information sheet to keep and be asked to sign a consent form. If you decide to take part, you are still free to withdraw at any time and without giving a reason. We will keep any data you have previously provided. Deciding not to take part or withdrawing from the study will not affect the healthcare or speech and language therapy your child receives.

We will ask you to come to see us for a morning or afternoon on two different days'

- You will be asked to come to the University of Strathclyde for a speech, language, and movement assessment. This will be carried out over two sessions, and you will sit in each session with your child.
- If you are unable to travel to the University of Strathclyde, we can arrange the research session to happen in your child's school.
- Each session will take about 1 hour, including rest breaks (as requested by your child). We will arrange these at a time that is convenient for you and your child.
- This study will work with individually approximately 10-20 children with autism aged 6-12 years throughout the course of the research.

First Session (Standard clinical assessments)

- Clinical assessments: During the first session your child will participate in standard assessment of their speech, language and movement commonly used by therapists in clinics. This will be carried out with the speech and language therapist (Louise McKeever) and involves picture description tasks and movement tasks (e.g., throwing a ball, raising arms etc.).

Second Session (Experimental Research)

- Ultrasound Tongue Imaging: During the sessions, your child will be asked to sit in front of a computer screen in a sound-treated studio. Your child will wear a plastic headset, which will ensure that the ultrasound probe can be correctly positioned beneath the chin. The end of the probe will be covered in medical water-based gel. A microphone will be placed near the child, to record the voice of your child when s/he speaks.
- Your child will be asked to copy various sounds, words and sentences and drink a few sips of water. The sessions will be recorded for analysis.

Sessions can be carried out at your child's school or at the University of Strathclyde. If you and your child to travel to University of Strathclyde for the assessment and therapy. Cost of travel to and from the university will be reimbursed (public transport or petrol money). There is parking available on campus.

With your permission we will consult with your child's Speech & Language Therapist (if applicable) and report results of assessments/therapy back to her. With your permission we will inform you child's GP that he or she is taking part in the research project.

All data will be anonymised. Your child will not be mentioned by name in any report or presentation. However, if some of the data were played at a verbal presentation, there is the possibility that the voice of your child may be recognisable.

What are the possible benefits of taking part?
If you take part in the project, you will have the benefit of an in-depth speech and language assessment. All reasonable travel costs will be reimbursed.

What are the possible disadvantages and risks of taking part?
It is not thought that there are many disadvantages and ultrasound is subject to rigorous safety assessments. At all levels of intensity used for diagnostic imaging, there are no known risks associated with ultrasound and there are no specific dangers or safety requirements. The ultrasound equipment and plastic headset has been used before at Queen Margaret University and University of Strathclyde with both children and adults.

Your child may experience some mild discomfort from wearing the headset as it can start to feel heavy after around 30 minutes. The speech task only requires wearing the plastic headset for 10-15 minutes and your child may remove the headset for a rest at any time within this. The experiment can be discontinued at any point if you or your child wishes.

What happens when the study is finished?
We will write to you within two months with a report detailing your child's individual scores from the standardised behavioural assessments carried out. With your permission we will share this information with your child's GP and speech and language therapist.

Will taking part in the study be kept confidential?
All the information we collect during the research will be kept confidential and there are strict laws which safeguard your privacy at every stage. Your child's name will be removed from the data so that s/he cannot be recognised from it. Data will be kept at the University of Strathclyde. With your consent we will inform your child's Speech and Language Therapist that you are taking part.

Who will my child's data be shared with?
In order to participate in the study, the ultrasound and voice recordings will be shared with members of the research team for analysis. Results of the analysis will be published anonymously. With your permission the anonymous data collected can be used for future research projects.

Optional Data Sharing
We ask you to consider **additionally and optionally** consenting to sharing this data with students (for teaching purposes) and researchers in other universities across the world. This is entirely optional, and your child can still be included in the project if you do not wish to share the recordings in this way.

How long will you keep the data for?
We will keep the data for 15 years for future analysis. However, you (before your child is 16) or your child (at any age) may withdraw consent by writing to or emailing us. We can then destroy the raw data but will not be able to retract any published articles. We will endeavour to write to your child when s/he turns 16 to check that it is still ok for us to keep the data and/or share it with other researchers or on the internet (if consent has been given to do so).

What will happen to the results of the study?
The results of the study will be shared with the public, speech and language therapists and academics via our website, conference presentations and publication in academic journals. We will post a plain English summary of our findings on our website and to you.

Who is organising the research and why?
This study has been organised and funded by the University of Strathclyde for a PhD project and is funded by the Chief Scientist Office of Scotland. The funding runs from January 2016 for 3 ½ years.

Who has reviewed the study?
The study proposal has been supervised by the research team at the University of Strathclyde. A favourable ethical opinion has been obtained from West Scotland REC 3 and University of Strathclyde ethics committee. NHS management approval has also been obtained.

It is entirely up to you to decide whether to take part in the project. If you do decide to participate, you will be given this information sheet to keep and be asked to sign a consent form. You and your child are free to withdraw from the study at any stage without giving a reason.

If you would like to consult an independent person, who knows about this project but is not involved in it, you are welcome to contact Dr Wendy Cohen, 0141 548 3793 or **wendy.cohen@strath.ac.uk**

If you have read and understood this information sheet, and you think you might be interested in participating in the study, please now fill in the tear off slip or email **louise.mckeever@strath.ac.uk** There will be an opportunity to ask questions and sign the consent form when you come to University of Strathclyde to meet with the research SLT.

Thank you for taking the time to read this information.

Researcher Contact Details:

Chief Researcher	Dr Joanne Cleland Graham Hills Building 40 George Street Glasgow joanne.cleland@strath.ac.uk 0141 548 3037
Research Speech and Language Therapist	Louise McKeever Graham Hills Building 40 George Street Glasgow louise.mckeever@strath.ac.uk 0141 548 4393

If you wish to make a complaint about the study, please contact NHS Great Glasgow and Clyde:
NHS Greater Glasgow and Clyde Complaints Team
Phone **: 0141 201 4500**
E-Mail **: complaints@ggc.scot.nhs.uk**

Appendix Four – Parent information Sheet (TD group)

Parent/Carers Information Sheet

Speech and Movement in **Normally Developing Children aged 6-12**

Your child is being invited to take part in a research study as they have been identified as normally developing and aged between 6-12 years. Before you decide whether to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully. Talk to others about the study if you wish. Contact us if there is anything that is not clear or if you would like more information. Take time to decide whether you wish for your child to take part.

What is the purpose of the study?
This project aims to see if there is a relationship between speech difficulties in **normally developing** children aged **6-12 years** and children with autism. Children with autism have been shown to have higher rates of speech errors than children their own age.

Understanding if there is relationship between speech, language, and movement in children with autism would increase our understanding of the difficulties faced by people with autism and gather more information on why they make speech errors. We will compare the speech, language, and movement to normally developing children.

The project will use standard medical ultrasound to record the movements of the tongue during speech. We will compare these results to the results of standard assessments of speech, language and movement used in clinics.

A picture of the ultrasound setup showing ultrasound images of the tongue on a computer screen

More information on ultrasound, including pictures and videos, can be found here:
http://www.qmu.ac.uk/casl/ultra/default.htm

Why has my child been asked to take part?
Your child has been invited to take part because he or she has been identified by his or her teacher as potentially suitable for our research project because he/she is normally developing and aged between 6 and 12.

Does my child have to take part?
No, it is up to you to decide whether your child takes part. If you do decide to take part, you will be given this information sheet to keep and be asked to sign a consent form. If you decide to take part, you are still free to withdraw at any time and without giving a reason, but we will keep any assessment data you have previously provided because it will have been anonymised.

What will happen if I take part?
- You will be asked to come to the University of Strathclyde for a speech assessment using ultrasound tongue imaging.
- The session will last 30 **mins – 1 hour** and we will include rest breaks (as requested by your child). We will arrange these at a time that is convenient for you and your child.

Assessment
- Ultrasound Tongue Imaging: During the session, your child will be asked to sit in front of a computer screen in a sound-treated studio. Your child will use a headset, which will ensure that the ultrasound probe can be correctly positioned beneath the chin. The end of the probe will be covered in medical gel. A microphone will be placed near the child, to record the voice of your child when s/he speaks.
- Your child will be asked to copy various sounds, words and sentences and drink a few sips of water. The sessions will be recorded for analysis.
- We will also request for you to fill out a social communication questionnaire at the session. This is a yes/no questionnaire about your child's social communication development.

If you and your child to travel to University of Strathclyde for the assessment and therapy. Cost of travel to and from the clinic will be reimbursed (public transport or petrol money). We can organise a parking permit for you if it is helpful.

With your permission we will inform you child's GP that he or she is taking part in the research project. We can provide you and your GP a report of the results with your permission.

All data will be anonymised. Your child will not be mentioned by name in any report or presentation. However, if some of the data were played at a verbal presentation, there is the possibility that the voice of your child may be recognisable.

What are the possible benefits of taking part?
If you take part in the project, you will have the benefit of an in-depth speech assessment. All reasonable travel costs to the university of Strathclyde will be reimbursed.

What are the possible disadvantages and risks of taking part?
It is not thought that there are many disadvantages and ultrasound is subject to rigorous safety assessments. At all levels of intensity used for diagnostic imaging, there are no known risks associated with ultrasound and there are no specific dangers or safety requirements. The ultrasound equipment and headset has been used before at Queen Margaret University and University of Strathclyde with both children and adults.

Your child may experience some mild discomfort from wearing the headset as it can start to feel heavy after around 30 minutes. For this reason, we will limit wearing of the headset to a maximum of 30 minutes and your child may remove the headset for a rest at any time within this. The experiment can be discontinued at any point if you or your child wishes.

What happens when the study is finished?
We will write to you within two months with a report detailing your child's individual speech and language skills. With your permission we will share this information with your child's GP.

Will taking part in the study be kept confidential?
All the information we collect during the research will be kept confidential and there are strict laws which safeguard your privacy at every stage. Your child's name will be removed from the data so that s/he cannot be recognised from it. Data will be kept at the University of Strathclyde. With your consent we will inform your child's GP that you are taking part.

Who will my child's data be shared with?
In order to participate in the study, the ultrasound and voice recordings will be shared with members of the research team for analysis. Results of the analysis will be published anonymously. This is all you need to consent to in order to take part.

Optional Data Sharing
We ask you to consider **additionally and optionally** consenting to sharing this data with students (for teaching purposes) and researchers in other universities across the world. This is entirely optional, and your child can still be included in the project if you do not wish to share the recordings in this way.

How long will you keep the data for?
We will keep the data for 15 years for future analysis. However, you (before your child is 16) or your child (at any age) may withdraw consent by writing to or emailing us. We can then destroy the raw data but will not be able to retract any published articles. We will endeavour to write to your child when s/he turns 16 to check that it is still ok for us to keep the data and/or share it with other researchers or on the internet (if consent has been given to do so).

What will happen to the results of the study?
The results of the study will be shared with the public, speech and language therapists and academics via our website, conference presentations and publication in academic journals. We will post a plain English summary of our findings on our website and to you.

Who is organising the research and why?
This study has been organised and funded by the University of Strathclyde for a PhD project and is funded by the University of Strathclyde and Nancy Maxwell Bequest. The funding runs from January 2016 for 3 ½ years.

Who has reviewed the study?
The study proposal has been supervised by the research team at the University of Strathclyde. A favourable ethical opinion has been obtained from School of Psychological Health and Sciences (University of Strathclyde) ethics committee.

It is entirely up to you to decide whether or not to take part in the project. If you do decide to participate, you will be given this information sheet to keep and be asked to sign a consent form. You and your child are free to withdraw from the study at any stage without giving a reason.

If you would like to consult an independent person, who knows about this project but is not involved in it, you are welcome to contact Diane Dixon, 0141 548 2571 or diane.dixon@strath.ac.uk

If you have read and understood this information sheet, and you think you might be interested in participating in the study, please now fill in the tear off slip or email **louise.mckeever@strath.ac.uk**, details on recruitment letter

Thank you for taking the time to read this information.

Researcher Contact Details:

Chief Researcher	Dr Joanne Cleland Graham Hills Building, 40 George Street, Glasgow, G1 1QE joanne.cleland@strath.ac.uk 0141 548 3037
Research Speech and Language Therapist	Louise McKeever Graham Hills Building, 40 George Street, Glasgow, G1 1QE louise.mckeever@strath.ac.uk 0141 548 4393

This investigation was granted ethical approval by the University of Strathclyde Psychological Sciences and Health Ethics Committee. If you have any questions/concerns, during or after the investigation, or wish to contact an independent person to whom any questions may be directed or further information may be sought from, please contact:

Dr Diane Dixon
Psychology, Graham Hills Building, 40 George Street, Glasgow, G1 1QE
0141 548 2571
diane.dixon@strath.ac.uk

Appendix Five – Parent Consent Form

Speech, Language and Movement in Children with Autism Aged 6-12

- I confirm that I have read the information sheet dated _____ for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.

- I understand that my child's participation is voluntary and that we are free to withdraw at any time without giving any reason, without me or my child's medical care or legal rights being affected.

- I understand that any audio and visual ultrasound data can be stored and used indefinitely but anonymously for analysis.

- I understand that the anonymous results of such analyses can be disseminated freely to audiences and research users of all types.

- **I agree for myself and my child to take part in this study**

In addition to the consent above, please indicate whether you optionally consent to any of the following:

- I agree that anonymous recordings of my child's voice and visual images from ultrasound and motion capture video can be used in **university teaching**.

- I agree that that anonymous recordings of my child's voice and visual images from ultrasound can be played to a **public audience** to advance understanding of science, through broadcast, laboratory open days, science festivals and other public but non-professional talks and presentations.

- I agree that my child's anonymous raw ultrasound and audio can be **copied for analysis by other researchers** outside University of Strathclyde for their own academic research projects

- I agree to the research team to contacting my child's NHS Speech & Language Therapist to discuss my child's speech and language skills and pass on information and results of therapy (if applicable).

  Name & Address of SLT if applicable:
  _____
  _____
  _____

- I agree to the research team to informing my child's GP that he/she is taking part in this project.

Name & Address of GP if applicable:
_____
_____
_____

Name of child:_____ Age of child: Years:_____ Months_____

Signature of parent/carer: _____

Signature of researcher: _____

Date: _____

Name of researcher: _____

Address: Speech and Language Pathology,
40 George Street, Glasgow, G1 1QE
Email / Telephone: louise.mckeever@strath.ac.uk / 0141 548 4393

Appendix 6 – Sample Size Power Calculations

Independent Samples T-Tests in Chapter 3 - Speech	Effect Size	Sample Size in Each Group	Total Sample Size
DEAP PCC Scores	-0.8	26	52
DEAP Total Errors	1.01	17	34
DEAP Delayed Errors	-1.01	17	34
DEAP Unusual Errors	0.92	20	40
CUW PCC Scores	-0.47	73	146
CUW Total Errors	0.78	27	54
CUW Delayed Errors	-0.81	25	50
CUW Unusual Errors	-0.7	34	68
Independent Samples T-Tests in Chapter 4			
Maximum DDK Rate /p/	-0.32	155	310
Maximum DDK Rate /t/	-0.315	160	320
Maximum DDK Rate /k/	0.266	223	446
Maximum DDK Rate /tk/	-0.42	90	180
Maximum DDK Rate /ptk/	-0.68	35	70
Accuracy of Single Syllables	1.11	14	28
Accuracy of Sequences	0.82	25	50
Consistency of Single Syllables	-0.89	21	40
Consistency of Sequences	0.22	326	652
Consistency of both conditions	-0.37	116	232
Paired Samples T-Tests in Chapter 4			
p single slowest - p single fastest	-0.08		1229
p ptk segment slowest - p ptk segment fastest	-0.34		70
t single slowest - t single fastest	-0.06		2183
t tk segment slowest - t tk segment fastest	0.04		4908

t ptk segment slowest - t ptk segment fastest	-0.3		90
k single slowest - k single fastest	-0.25		128
k tk segment slowest - k tk segment fastest	-0.14		403
k ptk segment slowest - k ptk segment fastest	0.4		52
Independent Samples T-Tests of Mean Syllable Durations in Chapter 4			
p single slowest	-0.52	60	120
p single fastest	-0.66	38	74
t single slowest	0.18	486	972
t single fastest	0.06	4362	8724
k single slowest	-0.37	116	232
k single fastest	-0.46	76	152
tk segment slowest	0.04	9813	19626
tk segment fastest	0.39	105	210
ptk segment slowest	0.45	79	158
ptk segment fastest	1.17	13	26