

## **Aetiology of Speech Sound Errors in Autism**

### **Abstract:**

In looking at speech perception and production it is vital we understand variation in different populations in order to understand variation in what is perceived as typical speech development; develop bio-markers; and provide effective methods for diagnosis and intervention where required.

Research suggests that people with autism experience higher rates of speech sound errors (SSEs) than their peers (Cleland, Gibbon, Peppé, O'Hare, & Rutherford, 2010; Shriberg, Paul, Black, & Santen, 2011), yet the reasons why are unknown. This chapter takes an in-depth look at the current literature on SSEs produced by people with autism, from young children to young adults. It explores why these higher rates occur, moving beyond the previous debate of whether they exist at all in this population.

Recent studies using detailed articulatory analysis show that children with autism exhibited 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 perspectives that may account for these higher rates of SSEs in autism: a) the speech attunement framework and b) deficits in speech motor control. It explores how both of these perspectives may intersect to produce SSEs in people with autism. Both are discussed in relation to the comorbidities of speech perceptual issues and motor deficits often found in people with autism.

Suggestions are made for future research using sensitive articulatory analysis of assessing speech such as ultrasound tongue imaging or electropalatography. This chapter highlights the need to look equally at both linguistic and motor skills in children with autism to describe accurately the range of cognitive and neurophysiological processes that may be affecting the production of speech.

## 1.1 Introduction

People with autism present with higher rates of speech sound errors (SSEs) than their peers (Cleland, Gibbon, Peppé, O'Hare, & Rutherford, 2010; Shriberg, Paul, Black, & Santen, 2011) yet the reasons why are unknown. While SSEs and related disturbances to speech prosody might be a salient feature of autism on the first encounter, most research has focussed on the (arguably) more serious nature of social problems in autism. The small amount of research that does exist on SSE's is heterogenic, similar to the presentation of the condition itself. Early studies suggest people with autism have normal speech development or that it is simply delayed (Kjelgaard & Tager-Flusberg, 2001; McCleery, Tully, Slevc, & Schreibman, 2006). However, recent work using sensitive statistical measurements showed that children with autism exhibited significantly higher rates of SSEs than typically developing (TD) children. Moreover, researchers have identified disordered speech development when phonetic and phonological analyses go beyond percentage consonants correct measures (Cleland et al., 2010; Shriberg et al., 2011; Wolk & Brennan, 2013).

The cause of SSEs in autism has not been fully explored in the literature. Up to this stage, researchers have focused on testing whether or not SSEs are a feature of autism, without exploring *why* these may occur. Now that recent evidence demonstrates SSEs are prevalent in autism, this chapter will examine their possible causes. There are currently two perspectives on why SSEs occur in children with autism: (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). These two perspectives will be discussed in the context of autism and results from the literature will be explored regarding how each framework might intersect.

## 1.2 Speech Sound Errors

Before exploring why SSEs are present in autism, it is important to understand what is expected of normal speech production, what speech sounds errors (SSEs) are and when they occur. Speech production and perception can breakdown at multiple levels, reducing the effectiveness of the final goal of fluent speech (Ferrand, 2014). The neural processing required for speech production and perception is still only partially understood (Baghai-Ravary & Beet, 2013). Speech production has been

characterised as one of the most complex motor skills, functioning as multiple subsystems that must effectively coordinate together (Duffy, 2000). For example, the phonatory system, which consists of the larynx, muscles, vocal folds etc., must work in a coordinated manner to achieve effective voice production. Likewise, the phonatory system must also be coordinated with other sub-systems (e.g. respiratory system). Speech perception relies on the auditory system in which acoustic signals are transformed into meaningful representation of spoken language (Gandour & Krishnan, 2016). It requires various complex perceptual and cognitive tasks along the auditory pathway. Motor speech representations are important for both perception and production (Ravizza, 2005).

We use the term “Speech Sound Error (SSE)” here to describe difficulties with the production of speech sounds or speech segments (American Speech-Language-Hearing Association, 2017). Common clinical distortions or residual SSEs, such as rhotic or sibilant distortions are relatively minor and generally not associated with language or intelligibility deficits (Shriberg et al., 2011). In contrast, other children may have speech which is unintelligible even to close family members. SSEs are common in early childhood and include articulation errors (motor based production deficits) and phonological errors (knowledge and use of speech sounds) (Eadie et al., 2015). Articulation errors often come in the form of distortions whereas phonological errors come in the form of substitutions and deletions such as consonant cluster reduction, final consonant deletion, velar fronting and stopping of fricative. Articulation and phonological errors are not mutually exclusive and each can occur in a child’s speech profile.

Problems start to arise when speech errors are not resolved beyond childhood and can then be described as either residual speech sound errors (RSSEs) or persistent speech errors. Residual speech errors arise as “leftovers” from an earlier speech delay (omission or substitutions errors) that migrated closer to the norm to become distortions, whereas persistent speech errors are distortions that have been habituated from an early age. RSSEs often affect late acquired and motorically complex speech sounds such as /s/ and /r/ and are manifested as common clinical distortions of these sounds, for example lateralised or labiodentalised /r/. These types of errors might be particularly common in people with autism (33% of verbal adolescents and adults with autism compared to just 1-2% of the typical population,

Shirberg et al., 2001). Why this is the case is not known, though Shirberg and colleagues ascribe it to a difficulty in fine-tuning to the ambient speech model.

### 1.3 Aetiology of Speech Sounds Errors and Autism

Autism is a neurodevelopmental disorder in which there is a frequent co-occurrence of verbal and non-verbal deficits (American Psychiatric Association, 2013). People with autism are known to have persistent deficits in social behaviour, communication, and language, which may be entwined with their difficulties in producing intelligible speech. Evidence on speech impairment in autism is heterogenic. Researchers have found it is disrupted in speech samples of children with autism while others have found speech to be a delayed or developmentally appropriate when using perceptual and behavioural checklist assessments (Bartolucci & Pierce, 1977; Kjelgaard & Tager-Flusberg, 2001; Wilkinson, 1998). The literature currently lacks organisation of theoretical concepts, with different studies relying on different methods to measure SSEs.

Interactions between different areas of impairment in autism may cause SSEs. A triad of symptoms associated with autism could impair speech development: social motivation, cognitive (and motor) control and perceptual control. Social motivation is a set of psychological and biological mechanisms that biases a person to orient to the social world, seek social interactions and maintain social binds. In autism there appears to be a decrease in attention given to social information, causing a cascading effect on the development of social cognitive skills (Chevallier, Kohls, Troiani, Brodtkin, & Schultz, 2012). Impaired social motivation means the child may miss vital communicative opportunities in which to develop typical speech. This encompasses cognitive rigidity, a trait associated with speech disorders: one study found children with consistent speech disorder performed worse in cognitive flexibility tasks (Crosbie, Holm, & Dodd, 2009). A piecemeal cognition style alongside these social deficits may also result in autistic traits (Valla, Maendel, Ganzel, Barsky, & Belmonte, 2013). Social motivation impairment may negatively impact the development of neural networks critical to social cognition, e.g. face processing (Sterling et al., 2008). This cognitive style is described as “piecemeal” as the person’s attention is either on the individual components of the face or on the

physical configuration, losing the important social information in this interaction. This cognitive style may also have a significant effect on processing of speech, where only certain aspects of speech are attuned to, e.g. phonological elements that are necessary for differentiating meaning may be given preference over phonetic aspects which signal speaker identity. Moreover, suprasegmental aspects of speech, like prosody or pitch, go unnoticed, resulting in the production of unusual-sounding speech.

Perceptual processing may also be a cornerstone in our understanding of autism (Baum, Stevenson, & Wallace, 2015). Sensory representations form the basis of higher-order cognitive representation. However, anomalies in sensory processing in autism are not well understood. Perceptual anomalies in autism may account for commonly found traits in language and speech impairment. People with autism have been found to have difficulty in social orientation to relevant auditory stimuli such as speech (Kuhl et al., 2005; Paul, Chawarska, Fowler, Cicchetti, & Volkmar, 2007). However, significantly more research is required to determine how this affects social and cognitive development.

A link has also been found between deficits in language, literacy and SSEs (Carson et al., 2003; Goffman, 1999; Hayiou-Thomas, Carroll, Leavett, Hulme, & Snowling, 2017; Whitehurst, Smith, Fischel, Arnold, & Lonigan, 1991; Williams & Elbert, 2003). Literacy and SSEs have a complex relationship, presence of speech sound disorders have been found to have a small but significant risk of poor phonemic skills, spelling and word reading. While SSEs alone only have a modest effect on literacy development, when it is part of additional risk factors such as language delay, these can have serious negative consequences. This is consistent with the findings that multiple risks such as SSEs and language delay/disorder can accumulate to predict reading disorder (Hayiou-Thomas et al., 2017).

Previously the primary focus of research in communication in children with autism has been language, prosody and behavioural difficulties (Kjelgaard & Tager-Flusberg, 2001; Owens, 2004; Paul & Norbury, 2012). The presence of speech sound difficulties is now being acknowledged (Cleland et al., 2010; Shriberg et al., 2011; Wolk, Edwards, & Brennan, 2016) but questions remain on nature of the speech sound errors in autism. It is suggested that children with autism may exhibit

speech production that is characteristically different in its organisation from typical speech. This may be due to developmental delay, but may also be due to differences in the underlying psychological or neuromotor structures required to produce speech and to practice it regularly in everyday experience with social others. Below we discuss the presentation of SSEs in the current literature available.

#### 1.4 Historical Research of SSEs in Children with Autism

Around 40 years ago behavioural studies concluded that children with autism had a delayed pattern of acquisition of speech sounds similar to children with intellectual disability (Bartolucci & Pierce, 1977). “Oddities” in speech production were often described following broad phonetic transcriptions (Koupernick, 1971; Pronovost, Wakstein, & Wakstein, 1966; Wing, 1971). Due to the lack of in-depth instrumental analyses or narrow phonetic transcription, these errors revealed little about specific speech patterns and conspicuously absent was the level of detail required to identify the minor articulatory distortions. Speech sound production was often only assessed in addition to other aspects of communication impairment (e.g. receptive and expressive language, social communication deficit, prosody). The main purpose of analyses in these studies were often other aspects of communication impairment. The analysis of speech was therefore often only assessed in brief using parent questionnaires or short perceptual tests of single words (Pronovost et al., 1966; Shervanion, 1959; Wing, 1971). This severely limited the detail of findings beyond broad diagnostic categories.

During the last century and more recently, smaller, more in-depth case studies used phonological analysis to identify both delayed and disordered phonological processes (Wetherby, Yonclas, & Bryan, 1989; Wolk & Brennan, 2013; Wolk & Edwards, 1993; Wolk & Giesen, 2000). Wolk and Giesen (2000) carried out a phonological analysis of speech elicited with both object naming and spontaneous speech in four children with autism. They found typical, but delayed phonological processes. However, they also identified atypical processes such as residual errors, unusual sound changes and chronological mismatch (where phonemes are not acquired in the developmentally typical order). The speech profiles contained evidence of both articulation errors and phonological errors. All children had a

diagnosis of a phonological disorder ranging from mild to severe, with one child classed as non-verbal. Even within this very small sample, there is a huge variation, indicating that there are likely different subtypes of speech sound disorders in autism. One of the few studies that focused exclusively on speech sound behaviour was carried out by Bartolucci and Pierce (1977). Using a picture-naming task (single word analysis) to assess both perception and production, they compared children with autism to TD children and children with intellectual disability (ID). The analysis was limited. Twenty-four consonant sounds were broadly transcribed using the International Phonetic Alphabet (IPA) and errors were compared in terms of percentage consonants correct. They concluded that verbal children with autism failed to show any atypical traits in the production or perception of speech sounds. Additionally, when looking at the findings in further detail using more sensitive phonetic analysis they found a delayed pattern of acquisition, similar to children with ID. Conversely, there was a significant difference in the percentage of errors on liquids made by children with autism (11.4%) compared to children with ID (4.7%) and TD children (0%). Similarly, in their perception task of liquids, there was a significant difference between errors made by children with autism (16.6%) compared to TD (8.6%) and ID (5.5%). These results indicate an atypical profile rather than delayed speech acquisition. Further analysis of the consistency and frequency of errors may have revealed more about the speech sound patterns of these different groups. These studies tell us that SSEs were being identified in groups of children with autism, but we require measures that are more sensitive in order to determine the causes. Further in-depth analyses are required to determine the *pattern* of errors produced by this particular group at all stages of communication development.

### 1.5 Atypical Speech in Young Children with Autism

Findings from multiple researchers suggest that children with autism at the prelinguistic stage of communication have different phonatory qualities than their peers and children with other developmental disorders (Schoen, Paul, & Chawarska, 2011; Wallace, Cleary, Buder, Pettit, & Oller, 2008). This information could contribute to early identification that would allow intervention to be put in place during the optimal period, i.e. in the first years of life, when the brain is developing rapidly and there is significant neural plasticity.

Atypical speech development appears to be identifiable at an early stage of communication in children with autism. Smaller case studies using perceptual phonological analysis identified SSEs in young children with autism (under 5 years). Wetherby et al. (1989) analysed the syllables of vocalisations made by three children with autism (under 5 years) in a 30-minute sample of communicative behaviour. They found that the children had a deficient proportion of vocal acts that contained a consonant. This absence of some consonants in communicative acts might be an early warning sign that speech is not developing normally. It is difficult to distinguish whether this is a result of phonological or articulatory issues, thus further analysis of the speech errors made would be required. Samples of communicative interactions are often used in younger cohorts to analyse their phonological development perceptually. One interesting finding from this in-depth analysis is the presence of “atypical vocalisations” in children with autism. Atypical vocalisations were the primary aspect of prelinguistic communication that differentiated children at high risk for autism (9-12 months) from children at low risk (Schoen et al., 2011). To investigate this further Schoen et al. (2011) studied phonological and vocal behaviour using broad phonemic transcription of speech-like utterances and coded non-speech vocalizations without recognisable consonants. They found 30 toddlers (18-36 months) with autism exhibited ‘atypical vocalisations’ and overall a limited number of consonants compared to two groups of TD children (age-matched and language- matched). Whilst the percentage of consonants correct was not different from their peers, the number of speech-like utterances produced was significantly less. The main area of difference between the children with autism and their peers was the presence of “atypical vocalisations”. These atypical vocalisations came mainly in the form of high-pitched squeals (Schoen et al., 2011). What this research might suggest is that toddlers with autism do not align their speech to the duration, pitch and phonotactic properties of their ambient language environment.

Toddlers with autism may not tune into the language model of their environment (Sheinkopf, Mundy, Kimbrough Oller, & Steffens, 2000). Their failure to attend to their ambient language environment may negatively affect their ability to acquire spoken language, which in severe cases can mean people with autism remain nonverbal throughout life. In a study of early vocal behaviours in young children with autism (n=15) and children with developmental delays (n=11), the children with autism did

not differ in production of well-formed complex canonical vocalisations, but had significantly more utterances with atypical vocal quality (Sheinkopf et al., 2000). Canonical vocalizations are well-formed consonant-vowel sequences with rapid CV transitions. An impairment in these sequences serves as a sign of speech motor control impairment, which was not the case in this sample. However, the significant presence of atypical vocal quality may be an indicator of speech perception issues. Wallace et al. (2008) reanalysed this data using acoustic analysis and more refined categorisations techniques and found that children with autism produced more atypical phonatory qualities than children with developmental delay. On the contrary, Schoen et al. (2011) found that toddlers with autism followed a normal trajectory of phonological development, suggesting no issues with speech development. However there was a significant presence of atypical vocalizations in their speech, which may be due to a presence of speech attunement issues. Descriptions of vocal profiles differentiating developmental profiles could provide valuable evidence for early biomarkers of autism and could help us explain the origin of the issues in speech production demonstrated at a later age. Further research as to why atypical vocalisations occurs in young children with autism is required.

Current research of SSEs in autism has started to use technology as a means of increasing the sensitivity of analysis of speech, both qualitatively and quantitatively. Shriberg et al. (2001) found a predominance of articulation errors in children with autism using the “PEPPER” software. This software allowed analysis of the type and frequency of consonant and vowel errors in conversational speech. Using this method, they found 33% of the cohort with autism had at least one type of speech distortion error (residual speech sound errors, such as lateral lisps). These may be an indicator of a disordered speech profile, rather than delayed speech acquisition as previously assumed in earlier research.

An interesting finding from this study was the significant presence of “residual speech sound errors”. These occur when speakers older than 9 years have two or more of the same type of residual distortion errors (e.g. dentalized sibilants, derhotacization). Thirty-three percent of the children with autism presented with residual speech errors, a significant proportion compared to the expected 1-2% found in the TD population (Flipsen, 2015). RSSE’s are clinically significant as they

involve sub-phonemic changes in articulatory place and manner and can persist over the individual's lifespan.

Further evidence of SSEs in autism was found by Cleland et al., (2010) who report atypical/non-developmental SSEs in children with autism. They carried out a phonetic and phonological analysis of speech sound production in 69 children with autism. Using standardised clinical perceptual assessments, only 12% of the sample received a diagnosis of speech delay/disorder. However, when using further in-depth phonological and phonetic analysis, they found 41% of the group produced speech errors indicative of both speech delay and speech disorder.

The clinical assessment of speech used was a perceptual assessment called the Goldman Fristoe Test of Articulation (GFTA; Goldman & Fristoe, 2000). This is one of the few standardised assessments of speech sound behaviours in children. It examines speech sounds in the context of single words. Further research beyond this assessment such as single words of increasing complexity (polysyllables), maximum performance tasks or spontaneous speech may reveal motor constraints which have a substantial negative impact on intelligibility or increase the likelihood of an SSE occurring. Cleland et al. (2010) found non-developmental speech errors occurred despite whether a child's standard score fell within normal range or not on the GFTA (Goldman & Fristoe, 2000). This implies SSEs produced by people with autism may not meet the criteria for a speech disorder in clinical assessments. However, there is more to understand in relation to speech profiles of people with autism, which may reveal information about the different subtypes within autism and whether this aligns with a particular speech profile.

The study by Cleland et al. (2010) is in agreement with previous findings by Kjelgaard & Tager-Flusberg (2001) and Rapin, Dunn, Allen, Stevens, & Fein (2009) that children with autism make a number of SSEs. Cleland et al. (2010) found in their sample that while speech was characterised by developmental phonological errors (gliding, cluster reduction and final consonant deletion), non-developmental errors, indicative of a speech disorder, were also present (e.g. phoneme specific nasal emission and initial consonant deletion). To understand *why* there may be SSEs in the group, Cleland et al. (2010) carried out a battery of standardised assessments in speech, language and non-verbal cognition to determine if there are any causal

links. Interestingly no relationship between speech and language or speech and cognition was identified in this group. This indicates SSEs may be a result of another impeding factor. Cleland et al. (2010) hypothesised that the increase of SSEs may be due to an underlying neuromotor difficulty. Additionally it could also be due to speech attunement difficulties. Further analysis of auditory perceptual abilities and speech motor abilities is needed to understand the origin of the SSEs in this group.

Wolk and Giesen (2000) carried out a phonetic inventory and process analysis and found in four siblings with autism speech processes indicative of delayed speech development. In addition, they identified atypical processes such as residual articulation errors, unusual sound changes and chronological mismatch in their speech profiles. All four children were significantly delayed in gross motor and fine motor abilities. The combination of residual articulation errors (indicative of motor issues) and unusual sound changes (indicative of perceptual issues) suggests these children appear to have a combination of both speech motor control and speech perception issues. However, they did not find differences in suprasegmental production; children with autism did not produce vocalisations different in fundamental frequency or duration from TD peers, suggesting they are able to tune in to their ambient environment effectively in some ways. These children may be a different subtype of autism. These are limited measures of suprasegmental ability and would require further analysis.

### 1.6 Methodological Issues of Measurement of SSEs

Multiple methodological issues need to be taken into account when assessing SSEs. Firstly, analysis of speech in single-word contexts may be ineffective. It does not examine the effect of complex articulatory gestures during spontaneous speech, which is significantly more motorically complex than single word production (Adams, 1998). Kjelgaard and Tager-Flusberg (2001) investigated language and speech production in eighty-nine children (4;0-14;0 years) with autism. They argued whilst there was significant heterogeneity in the children's language skills, their articulation skills were relatively spared. However, this conclusion is brought into question when noting they also used the Goldman Fristoe Test of Articulation. It required further phonetic and phonological analysis for Cleland et al. (2010) to identify speech distortions using this assessment alone which was not carried out in this study.

Therefore, Kjelgaard and Tager-Flusberg's (2001) assessment of speech sounds may have been inadequate to determine whether their sample of children had SSEs. Clinically the children may not have met the diagnosis for speech sound disorder, but there is value in understanding if SSEs occur to gain understanding on speech perception and production in autism.

Perceptual single-sound assessment helped identify irregularities in speech sound production in some studies. Rapin et al. (2009) used a single-word assessment, the Photo Articulation Test (Lippke, Dickey, Selmar, & Soder, 1997) to analyse the speech of 62 children with autism. The test yields a score for correct speech sounds produced in naming single word objects. Similar to Cleland et al. (2010) and Shriberg et al. (2011) they found that 28% of the participant's speech was characterised by persistently and severely impaired speech sound production; this was despite better language comprehension. Additionally, they analysed the spontaneous speech samples and concluded that "several minutes of conversation provides more opportunities for mispronunciations than the single words of the Photo Articulation Test (Rapin et al., 2009). Their assessment was in agreement with the Photo Articulation Test results, finding 28% of the speech sample was characterised by severely impaired expressive phonologic skills. However, this was not an in-depth analysis, the authors rated each child's speech on a 3-point categorical scale (0= normal to 2= severe impairment). Whilst both these results indicate an abnormality in speech production of some children with autism, again it does not go beyond a quick perceptual analysis. One reason that (Wolk & Giesen, 2000) may have identified SSEs whereas McCleery et al. (2006) and Kjelgaard and Tager-Flusberg (2001) did not, is that they elicited speech using two methods: object naming and spontaneous speech utterances. Additionally, they did not rely on perceptual standardised assessments. It is vital that researchers consider speech in multiple contexts to ensure subtle articulation errors are identified.

The use of ineffective standardised assessments, and issues of the nature of autism can cause difficulty in speech sound assessment (Macrae, 2017). For instance, in children with autism with severe language impairment, there is often difficulty obtaining a speech sample due to expressive language difficulties associated with autism. McCleery et al. (2006) investigated the consonant production of fourteen severely language delayed children with autism and ten TD children. To assess

speech in the context of severe language delay, their assessment involved a communicative inventory (Fenson et al., 1993) providing opportunities for the child to produce voiced and voiceless consonant sounds. All vocalisations, including babbling, were scored in an effort to determine the child's consonant production repertoire. McCleery et al. (2006) concluded that the children with autism showed the same general speech sound production pattern as TD and language-learning impaired children. Interestingly, they acknowledged that the children with autism produced more sounds that were not classified as developmentally normal but did not carry out further analysis on these errors. Transcription and counting of these errors may have revealed an alternative speech pattern in children with autism or clinically significant errors. Furthermore, analysis of the "abnormalities" may have provided indicators of whether the nature of these errors were motoric or phonological, similar to studies of early communication behaviour (Paul, Fuerst, Ramsay, Chawarska, & Klin, 2011; Schoen et al., 2011).

Previous studies investigating speech of children with autism may have misidentified subtle articulation errors because of the imprecise nature of perceptual speech assessment. The assessments described in studies using single word analysis are reliant on these perceptual measures of speech. Precise information about articulatory movements cannot be identified from perceptual analysis alone. Yet this information may reveal more about speech motor control in autism and whether alternative movement strategies exist due to motor impairment and/or attunement issues. Auditory perceptual judgements are susceptible to errors and bias of the listener (Kent, 1996). An example of this is listener normalization where the listener mistakenly recognises phonemes that were not produced by the speaker. 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 IPA chart, even though variation within each category can vary significantly across individuals (Mowrey & MacKay, 1990). Speech assessment needs to look at a speech in multiple contexts, where articulatory gestures are more complex and using more in-depth forms of phonetic and phonological analysis. This will help to determine whether SSEs in autism are due to speech perception difficulties and/or tuning into speech or to speech motor control issues.

## 1.7 Potential Causes of SSEs in Autism

Several research groups have reported that around a third of children with autism present with oral motor or speech sound abnormalities at various levels of severity (Belmonte et al., 2013; Cleland et al., 2010; Shriberg et al., 2011). We will now discuss two potentially complementary perspectives of *why* this may be the case.

The “speech attunement framework ” was originally developed to explain common speech errors in the otherwise typically developing populations (e.g. dentalized sibilants). Shriberg et al. (2011) developed the “speech attunement framework ” due to ongoing suggestions that impairments in gross motor, fine motor and oral motor control in people with autism were associated with speech deficits frequently exhibited. The speech attunement framework posits that a child learning speech needs to attend to their ambient environment or ‘tune in’ to models in that environment. For example young children adopting dialect features of their peers. In addition, they need to make small and careful adjustments to their speech production to ‘tune up’ for accurate and socially acceptable speech production (Shriberg et al., 2011). It is also important at this stage to have a maturing speech motor system that ensures adjustments made to speech can be done so with adequate control by the child. A difficulty with speech motor control could intersect with speech attunement and cause the heterogenic speech profiles identified in speakers with autism.

As discussed earlier, people with autism may have a reduced ability and/or motivation to focus on the subtle details of articulation, due to social motivation impairment. This prevents them from making minute adjustments in order to produce speech similar to their social partners and others in their ambient environment (Shriberg et al., 2011). In essence, children with autism are thought not to have the psychological conditions necessary to engage socially with others through language to give the necessary experiences for learning speech.

Speech attunement may be affected in people with autism by various combinations of the following conditions:

- a) Enhanced auditory capacity, often observed in people with autism (Baum et al., 2015) may lead to earlier “tuning in” when motor maturity has not been achieved. Therefore, SSEs develop due to motor constraints.

- b) Constraints in affective social reciprocity, a common trait of people with autism (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) investigated whether children with autism had ‘speech attunement’ issues and a comorbid speech motor disorder, specifically childhood apraxia of speech (CAS). CAS impairs the precision and consistency of speech movements, despite the lack of any neuromuscular deficits. To determine whether the increased presence of SSEs in children with autism was a result of speech attunement issues or CAS, Shriberg et al. (2011) examined the continuous speech of 40 children with autism; 40 TD children; 13 children with speech delay; and 15 individuals with CAS. They used software PEPPER (Programs To Examine Phonetic and Phonological Evaluation Records; Shriberg et al., 2001). They used this software to perceptually and acoustically analyse continuous speech samples, transcribing and prosody-coding subsets of the speech.

This detailed analysis was designed to identify specific signs of CAS or motor speech disorder, e.g. slow speaking and articulation rate, spatiotemporal vowel errors and distorted consonants (American Speech-Language-Hearing Association (ASHA), 2007; Aziz, Shohdi, Osman, & Habib, 2010). Although, it can be argued that the findings did not support a diagnosis of motor speech disorder or CAS, children with autism had voice differences not reported in the CAS group, e.g. inappropriate loudness, abnormally high pitch. Additionally, they had appropriate rate and stress, in direct contrast to symptoms of CAS. Shriberg et al. (2001) use these results as evidence of speech attunement issues, rather than a motor-speech impairment. However, 75% of children with autism had increased repetitions and revisions; a symptom demonstrated by CAS speakers, with both groups producing these significantly more than TD children.

As a result, some indicators of speech attunement were noted:

- a) Increased repetitions and revisions, consistent with the description of autistic speech as “disfluent”.
- b) 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.

- c) Inappropriate loudness and pitch.
- d) Higher rates of speech delay and speech errors relative to population estimates.

A study by Baron-Cohen and Staunton (1994) found that children with autism whose mothers were non-native English speakers were more likely to develop their mother's non-native accent (83.3% of the sample) than that of their peers (12.5% of the sample). TD children without social communication difficulties have a strong drive to identify with peers they engage with regularly. A lack of drive to identify with peers, present in autism, could lead to weaknesses in opportunities for speech attunement. We would then expect children with autism to show higher rates of phonetic distortions. Indeed, research by Shriberg et al. (2001) has shown that adults with autism frequently produce articulation distortions, such as sibilant and rhotic distortions. It should be noted that this study included both adolescents and adults, speech distortions did not resolve with age and were classified as a "residual speech errors". Cleland et al. (2010) also found a number of older children with similar phonetic distortions. These studies tell us that speech distortions, which may be a result of poor speech attunement in childhood, can continue through to adolescence and adulthood. This is an important finding as children with residual speech sound errors face an increased risk of social, emotional and/or academic challenges relative to their peers with typical speech (Hitchcock, Harel, & Byun, 2015). These likely compounds the social and emotional disadvantages children with autism already have.

One aspect of speech attunement that requires examination is the effect on suprasegmental attributes such as pitch. Tonal languages such as Chinese and Thai rely on the ability to perceive pitch as they involve categorical distinctions of lexical tone. Lexical tones serve a phonemic role, they are vital for speech comprehension and production (Wang, Wang, Fan, Huang, & Zhang, 2017). Wang et al. (2017) found in an event-related potential (ERP) study that 16 children with autism had lexical tone processing that was impaired and likely had its root cause as a phonological deficit in categorical perception, similar to the findings of Yu et al. (2015). Bonnef, Levanon, Dean-Pardo, lossos & Adini (2011) also found abnormal speech spectrum and fundamental frequency processing in young autistic children

who spoke Hebrew. They assessed long-term average spectrum and fundamental frequency variability in 60-second speech samples of 41 children with autism using a picture-naming task. Compared to the control group, the spectra were shallower and there was less harmonic structure in the group with autism. These results imply abnormal processing of auditory feedback or elevated noise and instability in the mechanisms that control phonation. All of which could have a significant impact on the child's ability to tune into speech. Finally, Lyakso, Frolova and Grigev (2016) assessed acoustic features of speech such as fundamental frequency ( $f_0$ ),  $f_0$  range, formants, frequency and duration in emotional speech, spontaneous speech and repetitions of words in 60 Russian-speaking children with autism. Similar to previous studies in tonal languages and Hebrew, abnormal prosody was a consistent feature. All children with autism had high values of fundamental frequency, abnormal spectrum and well-marked high frequency. Stressed vowels also had higher values of fundamental frequency. Results indicated speech abnormalities in autism is reflected in their spectral content and fundamental frequency variability. Understanding and producing appropriate stress patterns appears to be difficult for people with autism.

The second perspective for increased prevalence of SSEs in autism is that a subtle, but significant, motor control impairment in autism causes differences in speech production (Adams, 1998; Barbeau, Meilleur, Zeffiro, & Mottron, 2015; Belmonte et al., 2013). This perspective is becoming increasingly attractive as evidence accumulates that motor disruptions in other domains, such as in the purposeful movement of the arms (Crippa et al., 2015; Torres et al., 2013), legs and posture movements in gait (Nayate et al., 2012; Rinehart et al., 2006). Additionally fine motor control during writing and object manipulation (Fuentes, Mostofsky, & Bastian, 2009) are disrupted in children with autism. A recent meta-analysis of motor data in autism suggest motor disruption may be a core feature of autism and not merely a co-morbid or associated condition (Fournier, Hass, Naik, Lodha, & Cauraugh, 2010).

Motor impairment is evident at both gross and fine level in autism. 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 (Beverdors et al., 2001; Gowen & Hamilton, 2013; MacNeil & Mostofsky, 2012; Mostofsky, Powell, Simmonds, & Goldberg, 2009; Whyatt & Craig,

2013)). Such disruption to movement early in a child's development is thought to contribute to the broad autism 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 autism 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.

Evidence of motor impairment in autism is growing. Neuroanatomical correlates have been proposed for the observed difficulties in motor functioning including abnormalities in the cerebellum (Fatemi et al., 2012), disruption in brain synchronization (Welsh, Ahn, & Placantonakis, 2005), impaired sensory input and multisensory integration (Gowen & Hamilton, 2013). As a result, it is suggested that if general motor abilities are impaired, this could result in a speech motor control impairment (Barbeau et al., 2015). Adams (1998) examined oral-motor and motor-speech production of four young children with autism compared to TD children in both simple and complex phonemic production. Data indicated that children with autism had significantly more difficulty performing oral movements and complex syllable production tasks compared to TD children. These results could indicate a speech motor impairment. However, due to their small sample size, these results are not generalizable. More research of speech motor control compared to general motor abilities is crucial to the understanding the case of SSEs.

The connection between speech motor control and general motor abilities has been examined in 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 a motion capture system every three months, the movements of the upper lip, lower lip and jaw were recorded from 24 children (between the ages of 9-21 months). Children who had reduced speech motor control had a delayed trajectory of communication development. Significant associations were identified between orofacial kinematics and the standardized measures of language and cognitive skills, even when age served as a covariate. This initial

evidence suggests interactions between cognition, language and speech motor skills during early communication development. Further research is required to identify and quantify causal relations among these co-emerging skills and whether this extends to general motor ability. Alcock (2006) also found that motor control was associated with an existing language impairment, particularly oral motor control. Moreover, Lewis et al. (2011) found children with SSD were slower to complete diadochokinesis tasks and had differences in their oral motor control compared to TD children. These studies provide evidence of an inherent link between speech and general motor capabilities, though whether there is a causative mechanism (in either direction) is unknown.

Little research explores the relationship between speech and general motor impairment in children with autism. Nevertheless, there have been some interesting findings in children with idiopathic speech disorder who have been found to have reduced performance on tasks that involve visual motor control and fine motor control, e.g. grasping and object manipulation (Newmeyer et al., 2007). Peter and Stoel-Gammon (2008) found children with SSEs had deficits in repetitive finger tapping and clapping exercises associated with fine motor control. Lewis et al. (2011) found children with SSE were slower to complete diadochokinesis tasks and a maximum phonation task associated with the competency of speech motor control, compared to TD children. Bradford and Dodd (1994) compared ten phonologically delayed children, ten children with consistent phonological disorder and ten children with inconsistent error patterns. Groups did not differ on simple motor tasks; however, the group with inconsistent error patterns performed significantly worse in timed motor planning tasks and expressive novel-work learning tasks than in the other two groups. These results provide support for the perspective that inconsistent error patterns are associated with a deficit in some aspects of fine motor planning, a similar pattern that has also been identified in autism (Fournier et al., 2010).

Timing is a fundamental aspect of speech production. Fluent speech requires information to be selected, sequenced and articulated in an accurate and time sensitive manner. A set of quasi-autonomous articulatory systems need to work in coordination (Kotz & Schwartz, 2016; Maassen & Van Lieshout, 2010). Whilst little research has been carried out on speech timing, studies suggest there may be abnormalities in sensorimotor timing in children with autism. Anzulewics, Sobota and

Delafield-Butt (2016) found an increase in the speed of fast taps and swipes in children with autism playing an iPad game. Torres, et al. (2013) found an increase in the acceleration-deceleration phases of a reach-to-touch task in children with autism. These tasks demonstrate a subtle, but significant disruption to moment-by-moment control of movement occurring in the region of 30-70 ms, a temporal domain important for speech. Over- and under-compensations of such rapid shifts in force are thought to underpin the overt motor disruptions typically observed (Trevorthen & Delafield-Butt, 2013; Whyatt & Craig, 2013). These compensations may affect basic perception and effect experience resulting in disrupted speech development due to lack of coordination of articulatory systems (Colwyn Trevorthen & Delafield-Butt, 2017). Cook, Blakemore and Press (2013) found sub-second control of velocity and acceleration was affected in individuals with autism in simple arm-swing tasks. This study indicated that fast timing at less than a second (sub-second) required of speech motor control might be disrupted in limb and hand movements in individuals with autism.

Future research needs to look at both linguistic and motor planning skills in children with autism to describe accurately the range of cognitive processes that may be affecting their speech production. These studies above indicate there may be a deficit in motor planning and programming associated with speech sound disorders, but the origin is still unknown (Shriberg & Kwiatkowski, 1994). Therefore, it is important we look at the studies on SSEs in autism in detail to determine what knowledge exists on their nature and causes.

## 1.8 Conclusion and Future Directions

Researchers have identified atypical speech development in children with autism (Shriberg et al. 2001; Cleland et al. 2010; Wolk & Brennan 2013). However, it has been argued these errors are within a sequence of normal development (delayed) rather than atypical (Kjelgaard & Tager-Flusberg 2001; McCleery et al. 2006). Inconsistent outcomes in the literature may be a result of inconsistent and reduced specificity of the perceptual measurements used across studies. This could also reflect the heterogeneity in the population of people with autism in their production of speech. If deficits in speech motor control mirror the deficits in fine motor control, then finer-grained techniques may be needed to identify them. This is important,

because even if speech motor control problems are subtle, their existence might indicate that an underlying motor impairment is at the heart of autism. It is unlikely that subtle speech motor control problems will be identified with judgments on the correctness of productions of single words. Instead, one needs speech tasks such as maximum-performance tasks that tax the motor system. Alternatively, it is possible that articulatory analysis will identify qualitative differences in the articulations of children with autism compared to typical speakers. Indeed articulatory analysis, namely ultrasound tongue imaging, has been used in one study to assess and treat abnormal articulations in children with autism. Cleland, Scobbie, Heyde, Roxburgh and Wrench (under revision) found that ultrasound visual feedback might facilitate speech sound learning. While the study was not focused exclusively on children with autism, three of the children presented with SSEs and autism within the sample and responded to intervention. Although ultrasound tongue imaging is at early stages of development for assessment and intervention, it is a promising method of analysing SSEs in the depth required to identify subtle articulation errors in children with autism.

In conclusion, it is vital to determine *why* SSEs may be occurring and whether such occurrences are a result of disruption to speech attunement, a disruption to speech motor issues, or, more likely, both. Each aspect of speech development and production could affect the other, the two are entwined within the life of the child. Children with autism appear to have less drive to attune to the speech of peers due to particular social impairments. This may result in a reduction of motivation to produce speech that is intelligible and functional for others to comprehend. This may explain why we see prosodic abnormalities and unusual distortions errors, which do not affect intelligibility, such as phoneme-specific nasal emission (Cleland et al., 2010) and difficulties with articulatory complex speech sounds (Shriberg et al., 2011). Conversely, the disruption in speech motor performance that thwarts its intended meaning for others can itself drive a reduction in motivation to attune, leading to the same set of autistic consequences. Either way, improved aetiological understanding will help to determine principal underlying capacities and therefore routes to more effective intervention. Current research does not provide a clear picture of what theory best applies – if indeed either theory is appropriate without consideration of the other. In addition, it may be that there are subgroups of children with SSEs within

the broad autism spectrum. Such ideas require testing. Future research also needs to look equally at both linguistic and motor skills in children with autism to describe accurately the range of mental and neurophysiological process that may be affecting the production of speech. Understanding these questions will help to improve effective speech therapy interventions to target the underlying disruptions that give rise to SSEs at an early age and develop bio-markers for earlier diagnosis of autism.

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