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VOCAL QUALITY DEVELOPMENT IN CHILDREN WITH AUTISM: THE ROLE OF FLUTTER

by

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Abstract

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Background: Children with autism spectrum disorder (ASD) are frequently described as having unusual prosody. Studies in vocal quality in children with ASD have been limited in number and are generally descriptive in nature. The objective of this study was to examine the occurrence of a specific vocal quality, flutter, in children with ASD. **Method**: Vocal flutter was examined via subjective ratings and through acoustic analyses to determine if flutter occurred more frequently in children with ASD than in language delayed or typically developing peers.

Conclusion: Vocal flutter was heard more frequently in children with ASD via the subjective ratings. The occurrence of flutter overall was associated with a measure of fundamental frequency fluctuation.

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Chapter 1

Introduction

Autism Spectrum Disorder (ASD) results in a pervasive developmental delay that develops during early childhood. It is characterized by impairments in social interaction, deficits in communication, and repetitive, stereotyped behaviors and interests that manifest prior to the child's third birthday (American Psychological Association, 2000). Reported prevalence rates have seen a dramatic increase in recent years. Current estimates from the Center for Disease Control suggest that ASD affects 1 in every 110 children (Autism and Developmental Disorders Monitoring Network, 2009). It is unclear whether this is truly an increase in the number of individuals affected by the disorder, or is a reflection of increasing knowledge and awareness of the disorder, or both. In either case, the popularity and interest surrounding this disorder have resulted in rising attention from both researchers and the mass media alike.

The deficits associated with ASD severely restrict the majority of the affected population's ability to successfully and independently function as members of the social community. Therefore, research has been driven by the need to develop accurate identification measures and treatment methodologies in order to improve the outcome for individuals with ASD. Trends in research have shifted to attempt to diagnose children with ASD as early as possible. This might be, in part, the result of a criterion from the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV-TR) that symptoms of ASD must develop prior to when a child is three years old. This exclusionary restriction in the diagnosis of ASD implies that the behaviors associated with the disorder can be clinically detected in children under three years old (APA,

2000). Additionally, parents frequently report concerns about their child's development in areas related to ASD within the first year of life (Dawson & Osterling, 1997; Orntiz, Gutherie, & Farley, 1977). Retrospective analyses of home videos from children with ASD's first birthdays revealed evidence that support suspicions that some symptoms of ASD develop before the child's first birthday (Werner, Dawson, Osterling, & Dinno, 2000). The goal of early identification is not an arbitrary race to the finish line of who can achieve an accurate diagnosis of ASD first. Instead, it is based on the need for early intervention.

Early intervention has been demonstrated to increase the long-term outcomes for individuals with ASD. For example, treatment of children with ASD administered before a child is 60 months old yields better outcomes than when treatment begins after a child turns 60 months old (Feneske, Zalenski, Kranz, & McClannahan, 1985). This effect can be specifically appreciated in the area of language development. The acquisition of language skills in early childhood (4-6 years old) are strongly linked to greater positive predictive outcome measures later in development (Szatmari, Bryoson, Boyle, & Duku, 2003). Therefore, it might be critically important to identify children with ASD within the first few years of life due to the impact of early intervention on the potential for later success. Studies in how the areas of language development, vocal development, and prosodic development differ in typically developing children from children with ASD have suggested the importance of exploring vocal quality as a diagnostic marker in children with ASD.

Chapter 2

Literature Review

Language Development in Typically Developing Children

Language acquisition in children is an extremely complex topic. Yet, the importance of obtaining an understanding of the topic for clinical application makes examining these intricacies worthwhile. Language development is a lifelong process that begins in the earliest stages of life. Typically developing children are exposed to an abundance of language long before they produce it themselves. For years, language theorists have attempted to describe events that must occur in order for children to acquire language. Precursors for language acquisition have been attributed to two types of sources: innate predispositions (i.e., biological) and functional experiences in development (especially in the social context).

Theorists who emphasize innate foundations for language acquisition suggest that the child assumes a passive role of language development. These theorists attempt to describe a system contained innately within the child that allows language to develop. The constructs used to describe the nature of this innate language system can be described from a biological approach or from theoretical (linguistic) approach. The biological approach attempts to describe a series of anatomical/physiological changes that take place which allow language to develop. For example, biological language research examines genetics, neural connections, and the formation of brain structures to attempt to reveal an anatomical basis for language milestones (Clancy & Finlay, 2001).

Other innatist theorists take a more linguistically driven perspective in describing the internal system that they propose governs language acquisition. Under this

description, every child is said to have a Language Acquisition Device (LAD; Chomsky, 1965). The LAD allows children to take instances of language exposure from environmental stimuli and intuitively recognize grammatical patterns to acquire their own language system. Proponents of this view suggest that the LAD will be automatically (i.e., with no action needed from the child) enabled when placed in an appropriate environment (Chomsky, 1993). Under this view, language development happens without much necessary social interaction. Chomsky suggests that language development will occur in the presence of an appropriate environment, the same way that a child will grow when given the proper nutrition (Chomsky, 1993).

Opponents of the innate view of language development contend that language does not serve a purpose without social interaction; therefore, it would not (and likely cannot) develop without it. Social interactionist theories of language development stress the important influence of human interaction on language development. These theorists believe that children begin to learn language in an attempt to engage with their environment (Bloom, 2000). In order for children to develop an accurate understanding of language, they must experience a series of events. First, a child must be presented with topics that are interesting and *relevant* to his or her desires or needs (Bloom, 2000). In addition, they must formulate their initial concepts. Because the child is unlikely, at first, to create conceptualizations that correspond directly to the concepts relevant to the language they are learning, *discrepancies* in understanding can occur. (Bloom, 2000). The child must become *motivated* to seek new information in order to resolve these contradictions (Bloom, 2000). Seeking clarification allows children to develop *categorization* skills that are required for language aquisition (Bloom, 2000). In contrast

to the innatist viewpoint, children are instead seen as active learners in the process of language acquisition.

For example, a child might inaccurately assume that the word *dog* refers to all four legged animals. This assumption breaks down when the child hears the word *cat* in reference to another four legged animal. This contradiction to the original construct (i.e., all four legged animals are *dogs*) results in a discrepancy in understanding. The child will become motivated to seek clarification. This is frequently done via social interaction (e.g., seeking clarification from a caregiver). The child will gain a stronger foundation of concept for *animals* as a result of this process (i.e., categorization).

Under this model, social interaction is key to the development of language in that in provides both the initial motivation for children to engage with their community, and it provides an outlet for language learning to occur. As demonstrated in the previous example, the process of word learning through explicit social interaction frequently occurs during moments of joint attention. In typically developing children, joint attention occurs when two individuals direct their gazes at an object simultaneously and begins between when a child is 9 to 12 months old (Tomasello & Akhtar, 2000). This is vital to the development of language because it provides the opportunity for adults to provide labels to specific objects in a child's environment while the child is attending to the object (Bloom, 2000). Furthermore, joint attention provides a platform by which children develop an understanding that language can be shared with others. While this model provides a strong foundation to *why* social interaction is important to language acquisition, another theory is needed to support *how* social interaction is used to develop the lexicon.

When typical children construct their knowledge of language, they continuously build and rebuild their understanding as exposure to new concepts increases. As previously described, this process is primarily guided by social interaction. The rules children use to construct both the initial concepts and subsequent modifications can be described using the Two Tiered Developmental Lexical Principles Model (Hirsh-Pasek, Golinkoff, & Hollich, 2000). The first tier of the model describes the system of rules used in order to develop early concepts of language. First, children must learn that words represent an object or concepts which pertain to individuals (for proper nouns) or to categories of entities or events in the world (Hirsh-Pasek et al., 2000). Furthermore, children learn that words are used to represent or describe objects everywhere (Hirsh-Pasek et al., 2000). In the early stages of language acquisition, children use a set of assumptions in order to guide their understanding. For example, early word-learners assume new words describe concrete items rather than more abstract ones (e.g., a rock is an object, not a description of the feeling one had when viewing the rock). Also, children map new words to concepts relating to whole objects rather than its parts (Hirsh-Pasek et al., 2000). For example, *dog* refers to the whole animal, and not only the animal's tail. After children master the fundamental principles described in tier one of the model, they will again redefine language knowledge using the principles of tier two.

In the second tier of language acquisition children, typically beginning when a child is two years old, sharpen their understanding of language in order to be able to extend their language knowledge to multiple social settings (Hirsh-Pasek et al., 2000). After children learn that words represent objects in their own environments, they must extend their depth of understanding so that their lexicon can be extended for use in all

settings for items of a similar category (Golinkoff, Hirsh-Pasek, Mervis, Frawley et al., 1995). For example, children must learn that the word *apple* can be used to represent any round, red, fruit whether they are at home, at day care, at the park, etc. Additionally, the child must learn that there are restrictions for the words that might be used to describe these concepts in order to be conventionally understood in one's shared social group (Golinkoff et al., 1995). For example, a child who uses the term "up-pants" for "shorts" will eventually abandon the created term for the one that is more socially understood. In order to continue to learn new words children must also follow the assumption that an unfamiliar word refers to a new object (Mervis & Bertrand, 1994). Typically, words and their referents have a one to one relationship. Therefore, when a child hears a new word he or she will match the new label with a novel object rather than renaming a previously constructed concept. Mervis et al.'s (1994) principle of *novel-name nameless category* (N3C principle) is explicitly used in fundamental language acquisition through the process of fast mapping.

Our earliest concepts of words and their meanings develop through the process of fast mapping (Carey, 1978). Fast mapping is also described as *quick incidental learning* (Dockrell & Messer, 2004). Fast mapping can occur with explicit indication (e.g., pointing to a referent) or without explicit social interaction (Mervis & Bertrand, 1994). Supporters of the N3C principle state that not only can a child map a new word to a novel category, but also typically developing children assign meaning to a new term after their *first* contact with the word (Carey, 1978). As exposure continues, a child will modify his or her understanding of the word when discrepancies in the initial label occur. These socially guided principles result in a child's ability to rapidly acquire new vocabulary

within the first few years of life. Significant deficits in language acquisition might occur in children who do not develop these prerequisite principles for language development (Tomasello & Akhtar, 2000).

Language Development in Children with ASD

ASD is defined by marked deficits in both social interaction and communication (APA, 2000). Based on the previous discussion of the social explanation for language acquisition, it is likely that the deficits seen in these are related. For example, children with ASD frequently demonstrate a lack of spontaneous attempts to share interests with other people (APA, 2000). This is evident through a lack of spontaneous showing, bringing, and pointing (APA, 2000). This specific deficit in social interaction severely decreases the likelihood of children with ASD initiating moments of joint attention (Mundy, 1990). Furthermore, children with ASD might also lack social reciprocity (APA, 2000). Therefore, children with ASD are also less likely to respond to bids of joint attention (Mundy, 1990). Joint attention (JA) can be used to facilitate initial language learning and to clarify discrepancies in understanding. Without the tool of JA at their disposal, children with ASD have significantly fewer opportunities to acquire and develop language. Because of deficits in fundamental skills used to acquire language, children with ASD frequently demonstrate language that is delayed, disordered, or atypical.

Evaluation of communication plays a significant role in the early diagnostic stages of determining if a child has ASD. Filipek et al. (1999) suggest that a child who does not babble by 12 months, does not produce single words by 16 months, or does not produce spontaneous two word combinations by 24 months warrants *immediate* further

investigation. However, the level of communication skill across children with ASD is highly variable. For example, some children with ASD never acquire any expressive language (APA, 2000). Others develop language but experience extreme delays in achieving the communicative milestones expected in typical development (APA, 2000). Still others develop adequate spoken language ability (APA, 2000). This is not to suggest that some children with ASD demonstrate completely typical language. Children with ASD who demonstrate adequate spoken language, frequently experience difficulties in pragmatic communication (e.g., topic maintenance) and in comprehension (especially in complex/critical thinking situations; Filipek et al., 1999).

Some children with ASD exhibit abnormalities in the course of language development and in their use of language. For example, children with ASD might appear to develop language the same as their typical peers and then experience a sudden regression in language skills. Any loss in language is also considered to be an immediate *red flag* for ASD (Filipek et al., 1999). Additionally, children with ASD are frequently noted to demonstrate stereotyped and repetitive use of language (APA, 2000). This is commonly observed in the use of echolalia. Echolalia is considered to be a typical phase in language development until a child is two years old (Filipek et al., 1999). The use of echolalia is considered atypical when it dominates a child's expressive language and/or persists beyond the child's second birthday (Filipek et al., 1999). Historically, controversy exists in the research about the function of echolalia in children with ASD. However, other work demonstrates that echolalia might be a reflection the cognitive processes children with ASD use to understanding language (Prizant, 1983). In either case (i.e., functionality or not), echolalia beyond the age of two years is atypical.

Negative indicators (i.e., missing or delayed skills and milestones), dominate the current diagnostic symptomatology for ASD. This is especially true in the area of communication. These negative linguistic markers have been the cornerstone for justifying further testing and for supporting behavioral observations for a diagnosis of ASD. While developmental language deficits have historically been one of the largest contributing features for diagnosing ASD, new focus has shifted to examining vocalizations as well as pre-lingual development and babbling as a positive diagnostic indicator for ASD.

Phonological Development in Typical Children

The study of vocal development in typical children experienced an important shift in focus around the mid to late twentieth century. At that time, many believed that phonological development in its earliest stages (i.e., babbling) was, in effect, a manifestation of the child producing, at random, all possible speech sounds from all the possible languages of the world (Jakobson, 1941). Furthermore, babbling was said to have no connection to the sounds used in meaningful speech (Jakobson, 1941). Later research studies of babbling demonstrated that the opposite was true. Babbling is now viewed in a more restrictive and predictive manner. Babbling is governed by the tendency for a child to produce relatively universal syllables. Furthermore, the same sounds found in babbling are also used later in meaningful speech (Oller, Wieman, Doyle, & Ross, 1975). Proving this relationship between infant vocalizations and more sophisticated phonological development allows pre-lingual development and speech sound acquisition to be studied in a manner that suggests a predictable pattern exists in typical development.

Children begin to vocalize within the first moments of life. For the first month of life children's vocalizations include reflexive (e.g., cries) and vegetative noises (Stark, 1980). During this time, children are also observed to produce a type of sound known as quasi resonant nuclei or quasivowels that are characterized by normal phonation; however, no attempt is made to shape the vocal tract (Oller, 1980). Over the course of the next few months of development children add *gooing* to their sound repertoire. Gooing differs from quasivowels in that children begin to shape the vocal cavity and frequently have velar-tongue contact during phonation (Oller, 1980). Next, children enter a phase of vocal play (Stark, 1980). During this phase the child experiments with the capacity to manipulate his or her vocal mechanism to achieve changes in pitch (e.g., growls and squeals) and amplitude (e.g., whispers and yells; Oller, 1980). After this expansion children begin to produce true speech-like sounds. This is first noted during the Canonical Stage (Oller, 1980). During this stage, children begin to produce consonantvowel (CV) units either in isolation (i.e., canonical babbling) or in repetition (i.e., reduplicated babbling; Oller, 1980). During the last few months of the first year of life, children begin to babble using a varied string of CV combinations (i.e., variegated babbling). Every advance in the complexity of pre-lingual development is a vital step to the development of language because they lay the foundations from which first words, word combinations, and a complex syntax and semantic systems develop.

These predictive patterns of phonological development continue beyond the prelingual stage of development and extend to individual speech sound acquisition and to phonological process extinction. Previous research has shown that certain speech sounds (e.g., /b/, /m/, /w/) emerge earlier than others (e.g., /r/, /ʃ/, /ʒ/; Hare, 1983). However,

controversy exists when attempting to pinpoint the exact ages each individual speech sound should emerge. For example, many normative charts depict broad age ranges to describe when a speech sound should be mastered.

Research has not only demonstrated a pattern in individual sound acquisition, but also established predictable rules for speech errors. In the clinical setting, consistent patterns of sound substitutions are known as phonological processes. These processes are considered to be part of typical phonological development (Locke, 1980). However, controversy exists in the underlying cause for why children make these systematic errors. Some believe that these error patterns are programmed into one's innate language code (Stampe, 2004). Proponents of this view suggest that phonological processes must be suppressed in order to achieve adult speech (Stampe, 2004). Others reject this claim (Locke, 1980). For example, children might produce a /w/ for /r/ in *rabbit* because of production difficulties for /r/ or because of difficulties in perception of the contrast between /w/ and /r/. The child simply produces the closest approximation of the target sound (possibly due to ease of production) in order to attempt to convey their meaning (Locke, 1980). In either case, there is general agreement for when (i.e., order) to expect certain errors to occur. Therefore, when evaluating speech disorders, it might be important to examine the order of acquisition/extinction rather than simply the child's chronological age. This is especially true in children with ASD.

Phonological Development in Children with ASD

A debate exists among researchers about how pre-lingual/phonological development differs in children with ASD. Some researchers claim that phonological development is normal yet delayed in the ASD population (McCleery, Tully, Sleve, &

Shreibman 2006). One study compared the use of early developing versus later developing sounds between children with ASD and typically developing peers (McCleery et al., 2006). The results of this study indicate children with ASD produced fewer latedeveloping sounds than their TD peers, which suggests a phonological delay rather than impairment (McCleery et al., 2006).

Yet, other research has indicated that children with ASD do not follow the same patterns of pre-lingual development as their typically developing peers. The majority of research focused on phonological development in children with ASD was conducted through descriptive studies (Wetherby, Yonclas, & Bryan, 1989; Wolk & Edwards, 1993, Wolk & Giesen, 2000). Even with a limited number of subjects, these studies still provide strong support for the idea that phonological development is atypical (rather than delayed) in children with ASD. For example, one study noted that children with ASD produce a greater number of vocal atypicalities (e.g., raspberries, trills, clicks, and growls) seen during vocal play than typical and developmentally delayed peers (Wetherby et al., 1989). The same study also reported that children with ASD exhibit fewer consonant productions in the pre-lingual (i.e., canonical) stages of development (Wetherby et al., 1989). Not only have findings of increased vocal atypicalities been replicated in other studies, they have been found to correlate with other developmental features including mental age (Sheinkopf, Mundy, Oller, & Steffens, 2000). These comparative studies not only demonstrate that children with ASD differ in pre-lingual development from typically developing children, but also suggest the possibility of using features of vocal development as a form of differentiating ASD from other childhood disorders.

An important methodological consideration in the case studies of phonological development is based on the idea of a chronological mismatch in children with ASD and typical development (Wolk & Giesen, 2000). Chronological mismatch refers to the emergence of later developing skills before the development of earlier developing skills in phonological development (Grunwell, 1981). The concept of chronological mismatch has been applied to both the development of individual speech sounds, as well as to the emergence and extinction of phonological processes. For example, one child in the case study presented with labialization of consonants well beyond the age at which the process is expected to be extinguished (Wolk & Giesen, 2000). These findings emphasize the point that studies of phonological development in children with ASD should focus on order of acquisition or extinction rather than merely age. This idea is further illustrated by recent studies in automated vocal analysis of children with ASD. Algorithms for predicting chronological age based on naturalistic speech samples accurately predicted development for both typical children and children with specific language impairments, but showed weak correlation between children with ASD's utterances and chronological age (Oller et al., 2010).

Additionally, the results of these studies do support the notion that children with ASD exhibit some of the same phonological processes as their typically developing peers including labialization, liquid cluster reduction, initial voicing, velar fronting, liquid gliding, and final consonant deletion (Wolk & Giesen, 2000). Interestingly, some processes were noted in the children with ASD that do not frequently occur in typical development including velarization, and frication of stops and liquids (Wolk & Giesen, 2000). Vocal development is an obvious precursor to natural expressive language

development (e.g., barring the use of an augmentative communication device). However, its study for the purposes of diagnosing ASD has been underrepresented in the literature until recently. It is important to note that studies in atypical phonological development in children with ASD have been primarily conducted without any objective acoustic data (i.e., using mostly transcription/descriptive approaches) to support their findings. Further research is needed to investigate the acoustic measures associated with these findings. *Function of Prosody in Typical Children*

Prosody refers to the stress, tone and intonation patterns for utterances that are implemented on an utterance by varying the pitch, amplitude and duration of syllables and phrases. Prosody is said to be suprasegemental in nature due the fact that it occurs at the syllabic, phrasal and utterance level rather than at the level of phonemic segments. Research indicates that prosodic development begins within the first months of life. For example, children's cry and babbling contours have been correlated with the intonation patterns found in adult speech to convey negation, interrogative, and imperative intensions (Moerk, 1977). Prosody is intricately woven into our language system. It is suggested the prosody plays such an important role in linguistic development, that it is actually used by children to acquire initial language (i.e., Prosodic bootstrapping hypothesis; Hoff, 2005). For example in English, words generally carry stress on the first syllable (e.g., *re*-cord), while verbs tend to have stress placed on the second syllable (e.g., re-cord; Kelly, 1996). Children learn these prosodic rules in order to help them develop more precise categorization skills.

Furthermore, prosodic development is vital to the development of a more complex language system, because it allows speakers to manipulate language for a variety of

functions. Prosody applies to three different domains: grammatical, pragmatic, and affective (Paul, Augustyn, Klin, & Volkmar, 2005). Grammatical prosody is used to provide the listener with syntactic information regarding grammatical class of a word (Paul et al., 2005). Pragmatic prosody refers to how stress is placed within an utterance that allows the listener to identify the important parts of the message (Paul et al., 2005). Affective stress describes the speaking rules used in situations when a person must change his or her prosody in order to conform to the rules of a situation (e.g., tone used when speaking to an employer; Paul et al., 2005). These prosodic functions help portray the semantic meaning of utterances. However, prosody can also be used to help convey the meaning of an utterance beyond the simple level of semantics.

Illocutionary force is the social intent transmitted by a speaker (Austin, 1962). Specifically, the use of illocution in language development illustrates that a child has the knowledge of why people communicate. For example, asking someone, "Are the cookies cool yet?" is a question (a kind of illocution) but it might not reflect a desire to obtain knowledge about the temperature of the cookies. Instead, the ultimate illocutionary force of the question might entail a request (an additional illocution) to be given cookies. Prosodic variations play an important role in distinguishing the semantic meanings from the intended illocutions that can be associated with an utterance which has a single semantic content (i.e., "Are the cookies cool yet"?). Prosodic manipulations represent a critical feature of language competence. Deficits in prosody significant limit an individual's ability to successfully interact with the social world.

Prosodic Deficits in Children with ASD

One of the most stereotypical features associated with ASD is unusual prosody. These descriptions of atypical prosodic use in children with ASD have been present since the very onset of the description of the disorder (Asperger, 1952/1991; Kanner, 1943). Researchers have identified a variety of prosodic characteristics found in children with ASD. For example, children with ASD have been described to have restrictive variations in pitch (e.g., robotic sounding), have unusual intonation patterns (e.g., monotone), and have irregular rhythm (Wetherby et al., 2004). Still others have described seemingly contradictory patterns of unusual prosody in other children with ASD (e.g., using singsong variations) (Peppe, McCann, Gibbon, O'Hare & Rutherford, 2007). Because no single unusual prosodic feature has been identified across all children with ASD, at best, researchers have subjectively identified prosody as bizarre when compared to typical peers (Fay & Schuler, 1980). While these global descriptions can be useful for describing the general effect of prosodic limitations, further research has been conducted on the specific impact of these limitations on both expressive and receptive prosody.

There have been several investigations of how children with ASD are able to manipulate the varying domains of prosody (i.e., grammatical, pragmatic, affective). The conclusions drawn have been limited in studies of children with ASD due to limitations of eliciting prosody in a natural manner. However, some general conclusions can be drawn from such research. Studies have shown that children with high functioning ASD demonstrate difficulties with grammatical phrasing on spontaneous productions (Paul et al., 2000; Shrigberg et al., 2001). Interestingly, children with ASD seem to demonstrate typical prosodic variations for imitation (e.g., echolalia) suggesting that children are

physically able to use prosodic variations (Local & Wootton, 1996). The deficits seen in the use of expressive prosody in children with ASD might be related to the deficits in receptive prosody.

Research has indicated that children with ASD have a receptive chunking deficit. This deficit is characterized by incorrectly interpreting (or missing) the meaning behind pause boundaries used in grammatical prosody. This negatively influences an individual with ASD's ability to identify when a speaker is finished an utterance (Paul et al., 2000). However, these prosody deficits potentially impact more than a child's communicative competence. For example, a receptive chunking deficit could offer a possible explanation for why children with ASD are unable to follow the social rules for turn taking within a conversation, thus making them appear socially inappropriate.

Theory of Mind (ToM) provides a possible explanation for why children with ASD have difficulty using and interpret prosody. Advocates of ToM state that an individual must understand that other people have various mental states similar to his or her own (Firth, 1989). ToM is used when attempting to interpret the suprasegmental meaning intended by the use of prosody. A listener must successfully infer the sender's intent (i.e., illocutionary force) in order to correctly identify meaning and must have common ground with the speaker in order to determine semantic intent in most cases of natural language (Tager-Flusberg, 1997). An inability to produce linguistic stress might result in difficulties or inabilities to interpret stress (McCann & Peppe, 2003). Prosodic expression and interpretation both require a high level manipulation of the foundational skills obtained during language acquisition and pre-lingual development. Therefore, it can be logically concluded that children with ASD, who have significantly altered

foundations in terms of ToM and language development, would exhibit problems and abnormalities with prosody. The question becomes, how early do these prosodic deficits manifest during development and can they be acoustically detected for diagnostic purposes?

Rationale

As previously described, the current Diagnostic and Statistical Manual of Mental Disorders-Fourth Edition (DSM-IV-TR) outlines areas of delay in three areas that must be observed in order for a child to receive a diagnosis of ASD (APA, 2000). These areas include impairments in social interaction, impairments in communication, and restrictive repetitive and stereotyped behaviors (APA, 2000). Additionally, the DSM-IV-TR requires that evidence of these deficits be present prior to the child's third birthday (APA, 2000). This caveat in the diagnosis implies that these behaviors are present and, therefore, theoretically could be clinically detected in children under three years old. Current estimates places for the average age of diagnosis for ASD at 5.7 years (Shattuck et al., 2009). The average age of diagnosis is even higher for females (6.1 years) and for children without a cognitive impairment (6.6 years; Shattuck et al., 2009). The disparity that exists between when the symptoms presumably manifest (i.e., within the first 3 years of life) and the average age of diagnosis suggests that the current methods for ASD screening and diagnosis have difficulty identifying children's behaviors at an early age.

The diagnostic criterion for ASD is primarily dominated by a list of negative indicators. This is especially true in the social and communication domains. For example children with ASD are said to lack the following: gestures, appropriate peer relationships, spontaneous initiation of sharing, emotional reciprocity, spoken language, ability to

initiate conversations, and make-believe play (APA, 2000). When observing children, it is possible that a variety of factors (e.g., fatigue, hunger, fear) might skew the results of an evaluation. Therefore, focus on primarily negative indicators, in any disorder, should be interpreted with caution.

In addition to the internal factors that might alter a child's performance, external (i.e., situational) factors must be considered. The implications of these external factors can be appreciated by examining the current standards used. The Autism Diagnostic Observation Schedule (ADOS) is a standardized observation tool that examines the social and communicative behaviors associated with ASD (Lord, Rutter, DiLavore, & Risi, 1999). It uses structured and semi-structured play tasks to evaluate the presence of ASD (Lord et al., 1999) The ADOS may be administered in either a clinical or home setting (Lord et al., 1999). However, specific instructions are given for modifications that must be made to the home environment (e.g., removing distracting items including televisions and siblings; Lord et al., 1999). This diagnostic tool has proven social validity. It is also important to consider environmental impact on the child's performance. For some children, clinical settings (or even modified home settings) might not elicit a true portrayal of the child's behaviors.

A recent study of automated vocal analysis offers a possible supplemental solution to some of the limitations of current diagnostic batteries for ASD. In the study, children wore pocket recorders that allowed all day recordings of the child's vocalizations to be collected in a natural setting (e.g., the child's home; Oller et al., 2010). An algorithm was developed which examined 12 acoustic parameters (Oller, et al., 2010). Automated analysis of the recordings using this algorithm accurately predicted group membership

between typically developing, language delayed, and children with ASD (Oller, 2010). The authors acknowledged the need for future studies of additional parameters to attempt to enhance the accuracy of the algorithm derived to predict group classification (Oller et al., 2010).

Research Objectives

A preliminary examination of samples from recordings used in the original automated analysis study suggests the possibility of a new parameter for analysis that might be specific to children with ASD: vocal flutter (Oller et al., 2010). The possibility of differentiation by means of this feature is of particular interest due to the fact that a review of the current literature revealed that this type of acoustically based analysis for this particular vocal pattern in studies of ASD has not been conducted before. If vocal flutter can indeed be found to exist with inordinate frequency in children with ASD, it will suggest the possibility of a new positive marker for identifying young children with ASD via a natural sample. The goal of this study is to determine the following:

- (1) Can vocal flutter be reliably detected auditorily?
- (2) Does vocal flutter appear more frequently in children with ASD?
- (3) Can flutter be acoustically quantified for the purpose of automated analysis?

Chapter 3

Methods

Participants

Vocal analysis was conducted for sixteen children. These children were participants in a larger study conducted through the LENA Foundation in Boulder, Colorado. The participants consisted of nine males and seven females. The children were between the ages of 6 and 42 months with a mean age of 27.5 months old. The participants included eight typically developing children, four children with ASD, and four children with a language delay. Demographic information for each group is displayed in Table 1. The LENA Foundation required documentation from qualified professionals (e.g., psychologists, neurologists, speech language pathologists) to determine that each child was accurately identified as a member of each diagnostic category. Standardized scores on various diagnostic and screening evaluations were provided by the LENA Foundation to support each diagnosis. Each child was randomly assigned with a subject number at the onset of the study to allow the investigators to be blind to the child's diagnosis throughout the research process.

Table 1

Demographic	Typical (N=8)	Language Delay (N=4)	Autism Spectrum Disorder (N=4)
Mean age (in	30.25		
months)	(SD = 9.4)	19.5 (<i>SD</i> = 11.6)	30 (SD = 10.3)
Range	18 - 42	6 - 31	15 - 41
Gender			
Male	4	3	2
Female	4	1	2

Demographic Information by Group

Sample Set

A 5-minute audio file was provided for each of the sixteen children by the LENA Foundation. These samples were taken from a longer, all day recording collected via pocket recorders worn by the children in a natural environment (e.g., home). These samples were reviewed by the LENA Foundation to ensure that all identifying information was removed and to determine that each sample included a minimum number of utterances for analysis.

Preparing the Sample Set

First, utterance boundaries for each of the sixteen recordings were identified. Boundaries were marked to include only the egressive portion of a breath group. Vegetative sounds (e.g., coughs, burps, hiccoughs) were not marked or included for analysis. After the boundaries were set, two levels of coding were applied. First, each segmented unit was judged to be either speech or not speech. Speech was defined as any well-formed, intentional segment consisting of at least one consonant-vowel (e.g., canonical) combination. An utterance did not have to have an interpretable meaning in order to be classified as speech. Each segment (i.e., both speech and not speech) then received a classification by vocalization type. Vocal classifications included cry, fuss, chuckle, laugh, or unspecified.

Phase 1 Subjective Analysis

Two doctoral students at the University of Memphis participated in a thirty-minute training session to introduce the concept of the vocal flutter. The students were presented with exemplars of four levels of flutter. These instances of flutter were taken from a typical child who was not associated with this study. These instances were discussed with the trainer and it was concluded that recognition of flutter should be relatively easy. Consequently the classification of utterances from the 5-minute samples of LENA data began without further training.

In the coding of the LENA data, the students were asked to subjectively rate each of the breath groups using the following scale: no flutter, slight flutter, mild flutter, moderate flutter, strong flutter. The coders were instructed to listen to the utterance up to three times before making their judgment. The coders did not have access to the speech/not-speech codes, the vocal type codes, the acoustic waveform or the child diagnoses. The coders were also unaware of the hypothesis of the study. The coders had access to the exemplars used in training throughout the coding process to be used as needed. The samples were presented and the codes were recorded using the Action Analysis Coding and Training (AACT) system.

Phase 2 Acoustic Analysis

The flutter codes obtained from the subjective phase were reviewed to determine the accuracy of the flutter ratings. Upon review of the flutter ratings, the trainer

determined that one coder understood the concept of flutter more accurately than the other. One coder (Rater 1) appeared to have misinterpreted the concept of flutter. After a brief discussion with the trainer, Rater 1 was allowed to review and change her ratings. The codes from the other of the two coders (the key coder) whose flutter codes appeared to most accurately reflect true instances of flutter were used to determine the selection of items for acoustic analysis. The highest three instances of flutter for those labeled as speech and the three highest instances of flutter for those labeled as not-speech (six total) were selected for each of the 16 children when possible. It should be noted that this could not be obtained for each child (e.g., there was one child with all utterances coded as 'no flutter'). Additionally, four instances judged by the key coder to have 'no flutter' (i.e., two 'speech' and two 'not speech') for each of the 16 subjects were also analyzed for comparison.

The TF32 software in the AACT program was used to track the fundamental frequency for each of the selected segmented utterances. Tracking the changes in the F0 results in the formation of pitch traces. Six parameters must be set in order for a pitch-determining algorithm (PDA) to be created. These parameters were set in three phases. First, global parameters were selected that provided the most accurate pitch trace for the entire five minute recording. Next, local parameters were adjusted for areas where the global parameters did not accurately trace the F0 (e.g., the PDA begins to track F1). Last, hand edits were made to adjust the pitch trace for segments where neither global nor local parameters resulted in an accurate trace. Hand edits consisted of marking the individual glottal pulses, interpolating pitch between known and unknown values, and zeroing (i.e., deleting) areas where pitch was traced inaccurately (e.g., traces of the mother's voice).

Special consideration was given when selecting the minimum correlation interval parameter. Adjustments to this parameter result in smoother pitch contours by ignoring rapid fluctuations in pitch. In order to prevent this parameter from smoothing segments of interest and to allow continuity between the 16 samples, this parameter was set to be no lower than 6.0 and no higher than 10.00 ms. The author of the paper was responsible for constructing the pitch contours for each utterance of interest. The author received frequent, individualized training sessions regarding how to set parameters and how to obtain accurate pitch contours. The pitch traces were used to objectively quantify the degrees of flutter subjectively noted during the first phase of the analysis.

After the contours were constructed for the entire utterance of interest, data were extracted for the pitch measures for the vowel portion of the utterance. The vowel was isolated in order to obtain a level of consistency with measures of similar vocal behavior across steady state vowels in Parkinson's patients (Winholtz & Ramig, 1992). The pitch measures were used in order to construct a best-fit line for the data points. Theoretically, within-syllable changes in pitch (e.g., flutter) might occur at the same time as more global changes in pitch (e.g., prosodic inflections). In order for the degree of flutter to be accurately measured, the slope of the best-fit line must be removed (i.e., de-trend). Calculations for constructing the de-trended, line of best fit were made using Microsoft Excel. The depth of change from the fundamental frequency was measured over the duration of a flutter 'cycle' in order to objectively quantify vocal flutter. A cycle consisted of any dramatic shift either completely across the de-trended best-fit line (e.g., from positive to negative) or an extreme shift taking place completely in one domain (e.g., high positive value to a lower positive value).

Chapter 4

Results

Subjective Flutter Rating Analysis by Coder

A chi-square analysis was conducted to evaluate whether vocal flutter could be significantly detected. The evaluation compared the flutter ratings of the two coders. The two variables were Rater1's codes (no flutter, slight flutter, mild flutter, moderate flutter) and Rater 2's codes (no flutter, slight flutter, mild flutter, moderate flutter). Flutter ratings between coders were found to be significantly related, Pearson's Chi-Square (12, N = 1,313) = 96.76, p < 0.001.

A similar chi-square analysis was conducted by collapsing the ratings given by each rater into either no flutter or flutter (i.e., all utterances coded as having slight, mild, moderate, strong flutter). Again, the ratings given by each rater were shown to be significantly related, (df = 2, N = 1313) = 38.83, p < 0.001, kappa = 0.116790. This very low kappa suggests that in spite of a high significance level, the two coders agreed at a fair to poor level. Table 2 displays the numbers and proportions of each flutter code as they were entered into the chi-square analysis. Note that the both total number of observed agreements on "No Flutter" and the total number of observed agreements on "Flutter" were only about 20 more than the expected value. Also note that there were many disagreements, where one coder indicated some degree of flutter and the other did not.

Table 2

Observed Values	Flutter (1)	No Flutter (1)
Flutter (2)	2.5 % (33)	20.6% (271)
No Flutter (2)	1.9% (25)	79.4% (984)
Expected Values	Flutter (1)	No Flutter (1)
Flutter (2)	0.9 % (13)	22.2 % (291)
No Flutter (2)	3.4 % (45)	73.4 % (964)

Expected and Observed Percentages of Flutter Ratings between Raters 1 and 2

Subjective Flutter Rating Analysis by Group

A one way analysis of variance (ANOVA) was conducted for the subset of flutter codes deemed by the key coder (Rater 2) to have the strongest cases of flutter for each child to determine if vocal flutter occurred more frequently in children with ASD. The ANOVA for Rater 1 did not yield a statistically significant relationship for this subset of the data, (df = 2, N = 184) F = 1.879, p = 0.156). Although statistical significance was not achieved for Rater 1, it should be noted that the flutter average remained highest for the ASD group (see Figure 1). The ANOVA test for Rater 2 did yield a significant relationship between diagnostic group and mean flutter ratings, again demonstrating highest flutter averages for the ASD group, (df = 2, N = 184), F = 5.821, p = 0.004). Figure 2 depicts the flutter averages for Rater 2 by group.



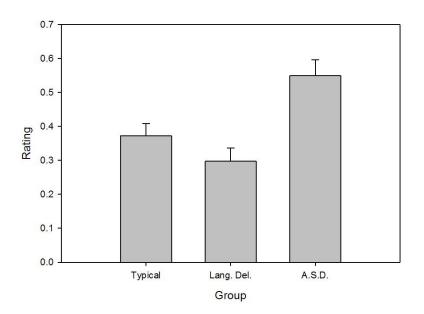
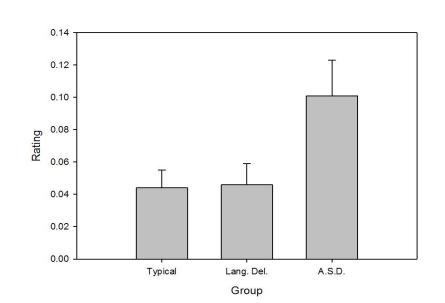


Figure 1. Rater 1 Average Flutter Ratings by Group



Rater 2

Figure 2. Rater 2 Average Flutter Ratings by Group

The relationship between diagnostic groups and flutter ratings was examined further using a Chi-square test. This was done by collapsing the subset of tokens deemed by the key coder to have the highest flutter by child into two categories: 'no' flutter and 'yes' flutter (i.e., tokens given a rating slight flutter or stronger). Group identity and instances of flutter were found to be significantly related for Rater 2, Pearson Chi-Square (df = 2, N = 181) = 11.131, p = 0.004 with more instances of flutter occurring in ASD. However, analysis of the chi-square for Rater 2 revealed that the significance level for differentiation between groups by flutter was actually driven by the number of 'no flutter' codes given for the LD group (i.e., N = 27, 66%). A statistically significant relationship was also found for Rater 3 (the author), Pearson Chi-Square (df = 2, N = 181) = 9.809, p = 0.007). Examination of the chi-square for Rater 3 demonstrated that not only did more instances of flutter occur in ASD, but also the number of instances of flutter (N = 13, 27%) for ASD accounted for the significant relationship between group and instances of flutter. This finding supports the hypothesis that flutter can be used to differentiate between groups and the possibility that it occurs more frequently in the ASD group.

Last, the relationship between group identity and flutter ratings was evaluated by examining the rank order of each child using the average flutter ratings determined by Rater 1 and Rater 2. Using this method, Rater 1 placed the children with ASD as having the first, fifth, sixth, and ninth highest flutter averages of the 16 children. Rater 2 placed the children with ASD has having the first, fifth, sixth, and fourteenth highest flutter averages for the 16 children. Place ranking for the all groups are displayed in Table 3.

Rank	Rater 1	Rater 2
1	ASD (0.3)	ASD (1.013)
2	Тур (0.194)	Тур (0.962)
3	Тур (0.172)	LD (0.889)
4	LD (0.091)	Тур (0.813)
5	ASD (0.083)	ASD (0.627)
6	ASD (0.064)	ASD (0.512)
7	LD (0.053)	Тур (0.446)
8	LD (0.049)	LD (0.247)
9	ASD (0.027)	Тур (0.247)
10	Тур (0.018)	Тур (0.162)
11	Тур (0.013)	Тур (0.152)
12	Тур (0)	Тур (0.097)
13	Тур (0)	Тур (0.096)
14	Тур (0)	ASD (0.083)
15	LD (0)	LD (0.053)
16	Тур (0)	LD (0)

Rank Order of Flutter Average by Group

Subjective Flutter Rating Analysis by Signal Type

A chi-square analysis was conducted to determine if flutter was related to signal type (speech/not speech, affect-laden/neutral). A significant relationship was found between flutter and speech/not speech signals for Rater 1, Pearson Chi-Square (df = 1, N = 184) = 5.850, p = 0.016, but not for Rater 2. Flutter appeared in 4% of the not-speech signals and in 14% of the speech signals for Rater 1. A significant relationship was found

between flutter and affect-laden/neutral signals for Rater 2 (Pearson Chi-Square = 35.877df = 1, N = 184, p < 0.001), and for Rater 3 (Pearson Chi-Square = 11.166, df = 1, N = 181, p < .001), but not for Rater 1. Flutter appeared more frequently in the affect-laden signals (81%) than in the neutral signals (37%) for Rater 2. For Rater 3, flutter occurred in 6% of the neutral signals and in 24% of the affect-laden signals.

Group Analysis by Signal Type

A chi-square analysis was conducted to determine whether the groups (ASD, TD, LD) differed by vocalization type (affect-laden, neutral). The analysis revealed a significant relationship (df = 2, N = 181) = 11.946, p = 0.003 with affect-laden signals occurring almost twice as frequently in children with ASD (62%) as in TD (34%) and in LD (33%).

Relationship between Subjective Flutter and Affect for Group Differentiation

A chi-square analysis was conducted to determine the contribution of affect-laden signal on the subjective perception of flutter in order to investigate whether flutter is truly more frequent in ASD or whether the effect was driven possibly by having twice as many affect –laden utterances for ASD in the sample set. The two variables for analysis were diagnostic group (ASD, LD, TD) and signal type (affect-laden, neutral). There was no significant relationship between group and subjective flutter noted in the neutral signals for either Rater 2 (Pearson's chi-square = 3.4, df = 2, N = 106, p = .177) or for Rater 3 (Pearson's chi-square = 5.187, df = 2, N = 75, p = .075). It should be noted that while the relationship was not significant, Rater 3 did demonstrate a tendency

which showed more flutter in ASD (37%) than in LD (7%) or TD (19%). There was a significant relationship between group and subjective flutter within the affect-laden tokens for Rater 2 (Pearson's chi-square = 12.03, df = 2, N = 75, p = .002). Again, this effect is driven by the high occurrence of 'no flutter' in LD (50%) rather than more flutter in ASD. Although these results do not rule out the possibility that subjective flutter might be the result of the affect-laden signals, the data support the possibility that the flutter heard in affect-laden signals might be different in ASD (evidenced by Rater 3's *p* value approaching significance).

Acoustic Quantification of Flutter

A series of *t* tests were conducted in order to determine the relationship between three acoustic measures and flutter. The acoustic measures included the mean fundamental frequency, standard deviation of the fundamental frequency (F0SD), and standard deviation of the de-trended fundamental frequency (DTSD). No significant relationship existed between mean F0 and flutter, or F0SD and flutter. A significant relationship was revealed between both DTSD and tokens coded by Rater 2 as flutter (t =3.75, p < 0.001). The DTSD was observed to be higher for tokens rated as having some flutter (20.3 Hz) than those having no flutter (12.5 Hz).

The same series of *t* tests were conducted to evaluate whether these acoustic parameters (mean F0, F0SD, DTSD) also were related to the various signal types (speech/not speech, affect-laden/neutral pooled, affect-laden/neutral not speech, affect-laden/neutral speech). No significant relationship was determined between any acoustic parameter and speech/not speech signals. A significant relationship was noted between

DTSD and all affect-laden and neutral signals (t = 2.85, df = 137, p = .005) with DTSD being larger in the affect-laden signals.

The relationship between the acoustic parameters and affect-laden/neutral signals was further examined by determining if a relationship existed between affectladen/neutral non-speech tokens and affect-laden/neutral speech tokens. A marginally significant relationship was found between F0SD and affect-laden/neutral non-speech tokens (t = -1.94, df = 66, p = .057) with larger standard deviations in the affect-laden, non-speech tokens. A similar relationship was noted between affect-laden signals and DTSD (t = 2.02, df = 66, p = .047) with larger DTSD in the affect-laden, non-speech tokens.

A significant relationship was also found between the acoustic parameters and affect-laden/neutral speech tokens for F0 mean (t = 2.49, df = 69, p = .015) and DTSD (t = 1.9, df = 69, p = .051). It should be noted that greater variability (i.e. higher F0 mean, F0SD, DTSD) was observed for all affect-laden versus neutral signals. This series of t tests demonstrates that DTSD was most significantly related to flutter. Values for all acoustic parameters and signal types are displayed in Table 4.

Relationship of Acoustic Parameters and Signal Type

Acoustic Parameter in Hz	High Flutter	No Flutte r	Spe ech	Not Speech	Affected Signal	Neutral Signal	Affected Not Speech	Neutral Not Speech	Affected Speech	Neutral Speech
			402.							
F0Mean	409.6	398.4	4	400.6	420.2	390.2	406.9	396.3	460.8*	387.1*
F0SD	30.5	23.6	23.8	25.8	28.2	22.9	27.1	19.3	31.2	24.6
		13.0*	15.9							
DTSD	20.3**	*	7	14.8	18.6**	13.4**	18.1*	13.1*	20.2*	13.6*

*p < .05

**p < .01

Chapter 5

Discussion

This study investigated three questions regarding the relationship between vocal flutter and children with ASD. First, the study analyzed subjective ratings of the occurrence and strength of flutter to assess the ability to detect vocal flutter auditorily. Next, the subjective ratings were examined for frequency of occurrence for three populations (i.e., ASD, TD, LD) to determine if vocal flutter occurs more often in one population than another. Last, acoustic measures were extracted based on fundamental frequency to determine if vocal flutter could be acoustically quantified. The following conclusions can be drawn from these findings:

- Agreement on flutter ratings between raters was statistically significant, but the proportion of disagreement was still very high.
- High ratings of vocal flutter occurred more frequently in children with ASD.
- Flutter appeared more frequently in affect-laden signals.
- Affect-laden signals occurred more frequently in children with ASD.
- Occurrence of vocal flutter was positively related with DTSD (de-trended standard deviation) of the F0.

This discussion highlights the implications and possible explanations for each of the findings. Limitations to the study, clinical implications, and directions for future research are also discussed.

Auditory Detection of Vocal Flutter

Agreement between the two coders was shown to be relatively poor. The chisquare analysis revealed that the two coders failed to agree in a large proportion of observations. The kappa value of 0.11 suggests that they agreed only 11% more often than would be expected by chance. However, there was evidence that each coder did notice the flutter phenomenon (demonstrated in the significant relationship between DTSD and flutter codes). This finding suggests that although high agreement was not achieved between the two raters, flutter was subjectively detected.

One possible explanation for this finding is that there was simply not enough training provided to reliably detect vocal flutter between two coders. One 30-minute training session was conducted with the raters at the onset of the study. The raters were presented with only one exemplar for each level of flutter. The trainer mistakenly concluded that no further training was needed based on the understanding demonstrated by the raters. Therefore, ratings were given without further training or sampling of flutter ratings throughout the coding process.

Another possible explanation for the discrepancies noted between the two coders could be the result of a misunderstanding by either rater of the concept of vocal flutter. The raters were allowed to use as much or as little of the 5-point scale was they wished. Rater 1 only used the code indicating flutter for 4% of the available tokens, while Rater 2 used a code indicating flutter in 23% percent of the available tokens. This suggests that Rater 1 might have believed vocal flutter was a rare phenomenon while Rater 2 believed that it occurred more frequently and subsequently 'heard' flutter more often. Instructing the raters to use the whole scale during the subjective phase might have increased the level of agreement.

Additionally, the raters were asked to only use the acoustic signal when assigning their flutter judgments. Not having visual feedback for the acoustic signal might have

limited the rater's ability to rule out other vocal phenomenon that might have been confused for vocal flutter. For example, many instances that were subjectively judged as vocal flutter were determined to actually be the result of glottal fry during the pitch analysis. Furthermore, no attempt was made to control for the quality of the tokens presented to the raters. This might have resulted in tokens being judged as having flutter that were actually the result of recording artifacts (e.g., two speakers at one time, background noise, fabric rubbing the microphone). Increasing the amount of training provided and improving the quality of the tokens presented might have improved the results on agreement for subjective flutter judgments.

Association of Vocal Flutter and ASD

Vocal flutter was shown to occur more frequently in children with ASD than in either of the groups. This finding is consistent with previous research suggesting atypical vocal quality development in children with ASD (Oller et al., 2010; Sheinkopf et al., 2000; Wetherby et al., 1989). This finding suggests that vocal flutter could specifically account for a portion of the pre-lingual vocal atypicalities revealed in previous research.

One possible explanation for why vocal flutter appears more frequently in children with ASD might be due to the neurological component of the disorder. Research has demonstrated a variety of possible regions and structures of the brain that are dysfunctional in individuals with ASD. For example, one such study outlined four portions of the brain (i.e., hippocampus, amygdala, oxytocin-opiate system, and association cortices of the temporal and parietal lobes) that show anomalies in individuals with ASD that might account for the behavior abnormalities observed (Waterhouse, Fein, & Modahl, 1996). Specifically, deficits of activity in these regions of the brain have been

shown to relate to problems with emotional regulation, which might account for the lack of social reciprocity noted in ASD (Waterhouse et al., 1996). A similar explanation could be applied to this particular phenomenon because similar phenomena (e.g., vocal tremor) have been observed in association with other neurological pathologies including Parkinson's disease and cerebellar ataxia (Aronson, 1985). Therefore, it is possible that neurological deficits might result in atypical vocalizations, specifically, vocal flutter.

Other studies have investigated the relationship between sustained fundamental frequency and ASD. These studies explored the effect of delayed auditory feedback on sustained F0 tasks for children with low functioning and high functioning ASD (Russo, Larson, & Kraus, 2008). The result of the investigation demonstrated that children with ASD can produce steady F0. However, the children with ASD had difficulty interpreting the auditory feedback in a way that allowed for successful control of the vocal mechanism (evidenced by sharp pitch-shift reflexes). Therefore, vocal flutter in children with ASD might be the result of an underlying auditory-feedback system deficiency.

Recent studies in vocal development of children at risk of developing ASD hypothesize that the underlying reason for atypical vocal development might be the result of the social impairment noted in children with ASD (Paul, Fuerst, Ramsay, Chawarska, et al., 2010). These authors theorize that because children with ASD are less likely to participate in early turn-taking (due to a lack of social reciprocity), they miss opportunities for practicing typical vocal skills acquired during pre-lingual development, which might result in an increase of atypical vocalizations (Paul et al., 2010). This theory might also explain the specific increase of atypical vocalization, flutter, noted in this study.

A last possible explanation for why instances of vocal flutter were significantly more frequent in children with ASD is that vocal flutter might be a type of stereotypic and repetitive behavior. These behaviors provide a source of sensory input and can be used by individuals with ASD to alert, relax, or avoid other environmental stimuli (APA, 2000; Stevens, Tidman, Glasgow, 2004). Intentionally-produced vocal flutter might provide children with ASD with a unique/interesting form of sensory input via the auditory system. Viewing vocal flutter as a self-stimulating behavior implies that it is produced under the child's control. This is not supported strongly by the results of this study. DTSD was significantly related to affect-laden/neutral signals (p = .005) with greater variability (i.e., larger DTSDs) seen in affect-laden tokens. Recall that affectladen tokens included any utterance that was judged to contain laugh, chuckle, fuss, or cry. These types of vocalizations usually convey an underlying emotional component. Generally, emotional states occur without direct control. If flutter occurs more frequently in affect-laden (less regulated) utterances, then it is less likely that vocal flutter demonstrated in ASD is used as a self-stimulating (controlled) behavior.

Another result of the study indicated that the affect-laden/neutral signal type differed significantly between the groups. It was observed that children with ASD produced significantly more affect-laden vocalizations than either the TD or LD children. This finding should be interpreted with some caution due to the fact that the affect-laden signals accounted for more than half of the available tokens for analysis for the ASD group. However, all vocalization codes were given with the author being blind to the subject's group identity.

One possible explanation for the fact that the groups differed in vocalization type might be an underling effect of an increased occurrence of vocal flutter in the ASD population. The changes in pitch seen in cases of vocal flutter might have been subjectively interpreted to be associated with vocalization types such as fuss or chuckle. Indeed, the utterances with the highest ratings of vocal flutter frequently received a subjective code of chuckle or fuss.

A series of chi-square analyses were used to examine the possibility that the greater use of affect-laden signals in ASD caused an increase in subjective flutter. This analysis indeed left open, the possibility that the higher degree of flutter in ASD could be accounted for by greater use of affect-laden signal type in ASD. This finding may of course be the result of the small sample size used for analysis which limited power to detect differences between groups in the neutral utterances.

Acoustic Quantification of Vocal flutter

This study investigated three acoustic parameters (F0 mean, F0SD, DTSD) to determine if vocal flutter could be acoustically quantified. It was hypothesized that DTSD would be the greatest indicator of vocal flutter. This hypothesis was confirmed by the results. In fact, DTSD was the only parameter found to be significantly higher in utterances with the subjective flutter ratings (p < 0.001).

This finding is most likely due to the fact that DTSD accounted for and removed the effect of global changes in pitch across an utterance. It was expected that an utterance could contain both global (e.g., rising intonation) and within syllable (e.g., flutter) pitch variability. The mean fundamental frequency and standard deviation of the fundamental frequency were computed without making any adjustments for the global pitch changes.

Therefore, it was not possible to accurately measure and compare changes within utterances in fundamental frequency without being influenced by global changes (which are not what flutter entails). This finding confirmed the wisdom of the preferred method (i.e., DTSD) for the acoustic analysis in the study.

Limitations

Certain limitations exist and should be considered when interpreting the findings and implications of this study. While this study was able to compare the findings found in ASD to both typically developing and language delayed groups, the subject size for each group, and for the study in general was very small. Additionally, all utterances came from a single 5-minute period for each child (selected as a period of high vocal activity by a LENA Foundation engineer). The ASD group was limited to only four subjects. Although flutter was demonstrated to be higher in these four children, one cannot definitively conclude that this same trend would apply to a larger sample. Additionally, the participants had twice as many TD children as any other group. Further investigations are needed with a larger, balanced population in order to draw conclusions about the role of flutter more globally in ASD.

While the LENA system pocket recorders allow for a natural representation of the child's vocal development, it does not have any visual (i.e., video) media to supplement the data. Therefore, it was occasionally difficult to distinguish the targeted child's utterance from other speakers or background noise (e.g., television) in the room. Although every effort was taken to ensure that only the targeted child's utterances were coded for analysis, it is possible that some superfluous tokens could have been analyzed.

Last, while vocal flutter was found to occur more frequently in children with ASD, it was not a phenomenon specific to the ASD group. Therefore, this study failed to find a feature exclusively present in ASD. This is consistent with other research findings documenting the heterogeneity of the symptoms of ASD. The goal of this study was not to find a symptom unique to ASD. Instead, the goal of this study was to find a supplementary positive behavior to add to those already associated with ASD that could help enhance diagnosis specificity for ASD.

Clinical Implications

It was predicted that the vocal flutter would be found to occur more frequently in children with ASD both through subjective and objective measures. The purpose of this study was to investigate a positive indicator of ASD that is present early in development. The results of this study indicate that vocal flutter can be (at least subjectively) associated more frequently in children with ASD than their TD or LD peers. The design and results of this study support the possibility of identifying behavioral symptoms of ASD before a child is 30 months. The subjects for this study were selected to have a mean age of 30 months or younger. Furthermore, there was no significant relationship between DTSD and speech/not-speech segments. This suggests that vocal flutter can potentially be identified at a young age without a dependence on the stage of vocal development (i.e., pre-lingually). This has important clinical applications due to the fact that delayed and absent speech and language development are key to the diagnostic criteria for ASD (APA, 2000).

The LENA recording system provides a naturalistic sample of the child's vocal behavior. This study was able to use this type of naturalistic recording sample to identify

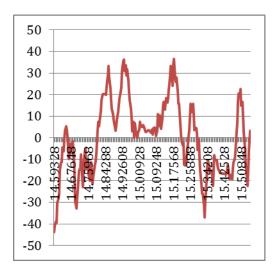
a positive behavior present more frequently in children with ASD. This, too, has important clinical implications. Such pocket recordings have already been collected at a massive scale for analysis by the LENA foundation. This precedent suggests that LENA recordings could be conducted on an even large scale, and possibly be used to develop a screening measure for all pre-school children (similar to newborn hearing screenings). *Implications for Future Research*

This study demonstrated vocal flutter was significantly related to DTSD of the fundamental frequency. A cursory observation noted during pitch analysis revealed two additional measures that warrant further investigation: pitch cycles and amplitude. A cycle for this study was defined as a dramatic shift from one quadrant to another or within one quadrant. Each token subjectively determined to have high flutter (i.e., those rated as moderate or strong flutter by Raters 2 and 3) had at least two or more cycles. Pitch and cycle data for the seven highest utterances for the children with ASD are shown in Tables 5 and 6. The coordinating de-trended pitch traces are demonstrated in Figures 3 and 4. The same data for utterances noted to have no flutter by the same raters are demonstrated in Table 7 and 8 with associated pitch trace images in Figures 6 and 8 (in the appendix). This cycle count was not formally investigated for statistical significance., Measuring the number cycle repetitions, the depth of the cycles, and the duration of these cycle might help to further characterize vocal flutter in the pitch domain.

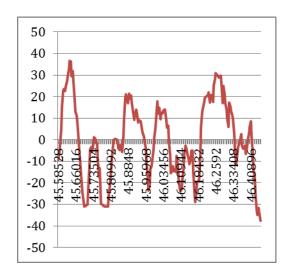
	Utterance 3	Utterance 10	Utterance 4	Utterance 76
F0 Mean	442	342	505	426
F0SD	18.2	18	24.2	13.6
DTSD	17.1	17.7	23.2	9.2
Cycles	4	4	3	3
Average Duration	240.75	220	221.6	120.3
Average Depth	48.25	49	51.3	16.03
Rater 1	2	0	0	0
Rater 2	4	4	3	3
Rater 3	4	4	3	3

Preliminary Cycle Data for Child 7 Rated as High Flutter

Utterance 3



Utterance 10



Utterance 4

Utterance 76

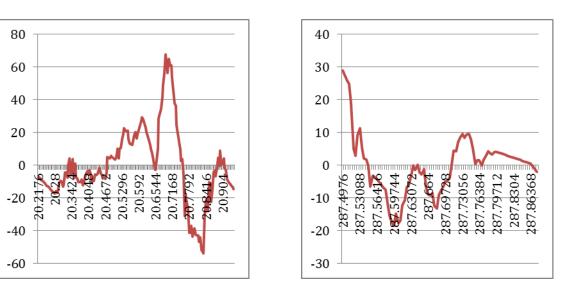
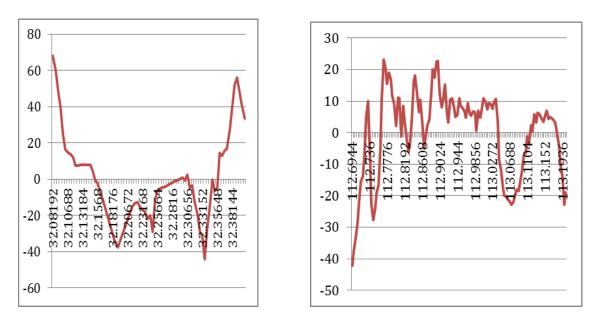


Figure 3. De-trended Pitch Analysis for Child 7



Utterance 44



Utterance 11

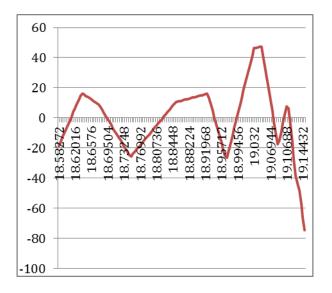


Figure 4. De-trended Pitch Analysis for Child 4

	Utterance 18	Utterance 44	Utterance 11
F0 Mean	476	454	398
F0SD	25.5	54.7	25.7
DTSD	24.3	13.7	20.8
Cycles	3	5	4
Average Duration	101.3	103.6	146.75
Average Depth	50.6	36.4	42.75
Rater 1	2	0	0
Rater 2	3	3	3
Rater 3	4	4	3

Preliminary Cycle Data for Child 4 for Utterances Rated as High Flutter

It is also possible that while DTSD of pitch correlates with vocal flutter, it might not be the truest reflection of flutter acoustically. In the preliminary stages of the investigation, it could not be determined completely auditorily if the source of variability noted it vocal flutter should be attributed to fluctuations in pitch or in amplitude. It was hypothesized that these type of change would occur more frequently in pitch than it amplitude. However, after conducting the pitch analyses, it became apparent that amplitude changes frequently co-occurred with pitch. In some instances, very little variability was observed in the pitch trace. Instead, the shifts in amplitude could account for the fluttered sound detected auditorily. Early evidence of the contribution of amplitude variability to vocal flutter are demonstrated in Figures 5 and 7 in the appendix. Future investigations in the role of amplitude change in the phenomenon of vocal flutter are needed.

Conclusions

This study provides additional support to the previous research concluding that atypical vocal development exists in children with ASD. Furthermore, this study strongly supports that specific atypicalities (i.e., vocal flutter) can be associated with ASD early in development. Future research in vocal flutter, and possibly other specific vocal anomalies, might increase the likelihood of early identification of ASD via a positive indicator.

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Appendix A

Supplementary Data for Child 7



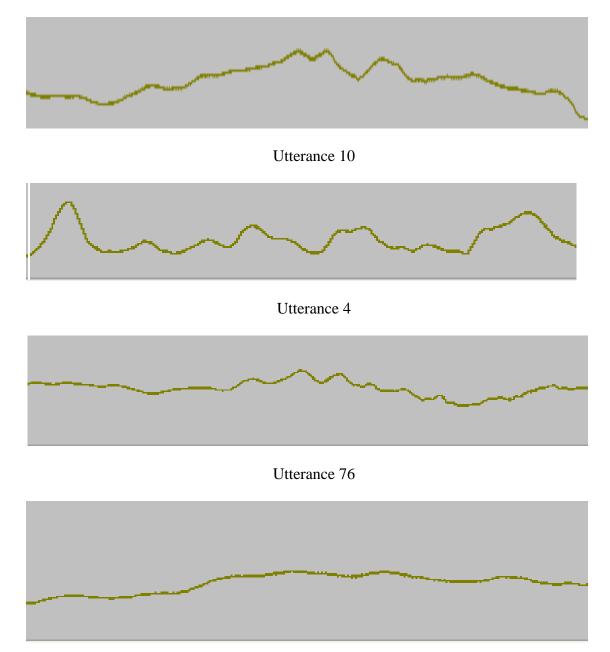
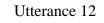


Figure 5. Amplitude Traces for Child 7

Child 7	Utterance12	Utterance 34
F0 Mean	537	406
F0SD	22	19.7
DTSD	3.3	11.9
Cycles	2	1
Average Duration	155.5	197
Average Depth	10	26.5
Rater 1	0	0
Rater 2	0	0
Rater 3	0	0

Preliminary Cycle Data for Child 7 for Utterances Rated as No Flutter





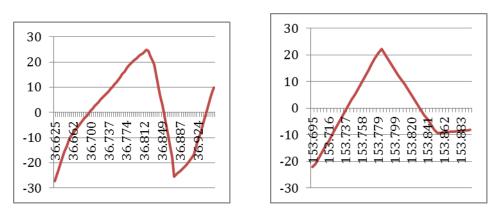
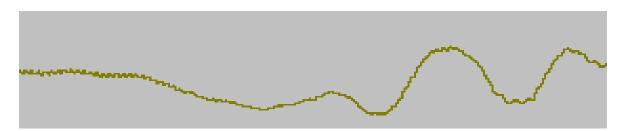


Figure 6. De-trended Pitch Analysis for Child 7

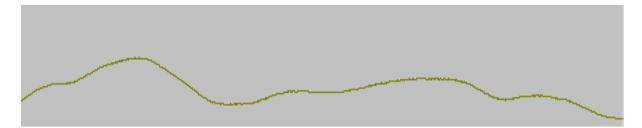
Appendix B

Supplementary Data for Child 4

Utterance 4



Utterance 18



Utterance 44

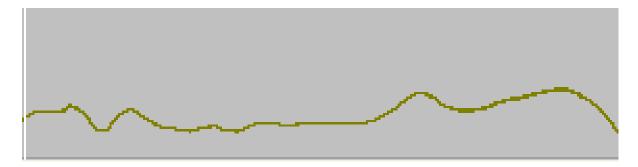
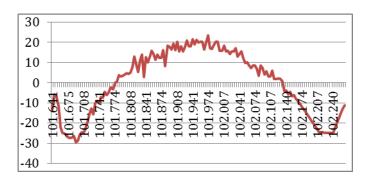


Figure 7. Amplitude Traces for Child 4

Child 4	Utterance 40	Utterance 1
F0 Mean	281	437
F0SD	15.9	107.1
DTSD	15.7	24.1
Cycles	1	2
Average Duration	627	228
Average Depth	38.5	71
Rater 1	0	0
Rater 2	0	0
Rater 3	0	0

Preliminary Cycle Data for Child 4 Utterances Rated as No Flutter

Utterance 40



Utterance 1

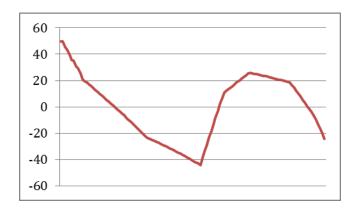


Figure 8. De-trended Pitch Analysis for Child 4

IRB Determination Form: *Planned Activity Does Not Involve Human Subjects or is Not Research*

Principal Investigator: Note: The federal regulations include a very specific definition for what constitutes "research" (see 45 CFR 46.102(d)) and for what is meant by a "human subject" (see 45 CFR 46.102(f)). If there is any doubt as to whether your project qualifies as human subject research you must obtain a formal determination from the IRB as to whether or not project either is not research and/or does not involve human subjects (e.g., as may be required by a student's doctoral dissertation committee, a funding agency, or a journal editor). Note: The IRB will not provide a formal written determination after the project has been initiated. The following application will permit the IRB to make this determination: A. Answer Each of the following questions: 1. Will any information from this project be submitted to the FDA or held for inspection by the FDA? No □ Yes □ 2. Are the data or specimens studied as part of this project obtained in a systematic manner? No □ Yes □ 3. Is the intent of this data collection to contribute to 'generalizable knowledge' - that is to disseminate the knowledge obtained to others outside the University of Memphis/UofM? No □ Yes □. 4. If 'no', and □ this project is being conducted at a UofM facility? No □ Yes □. 5. Will the study involve accessing (i.e., looking at or reviewing) identifiable private information? No □ Yes □ 7. Are the data coded in such that a link exists that could allow the data to be re-identified? No □ Yes □. 7. Are the data coded in such that a link exists that could allow the data to be re-identified? No □ Yes □. 8. Are a an en	Title of Study:
 "research" (see 45 CFR 46.102(d)) and for what is meant by a "human subject" (see 45 CFR 46.102(f)). If there is any doubt as to whether your project qualifies as human subject research you must obtain a formal determination from the IRB as to whether or not project either is not research and/or does not involve human subjects (e.g., as may be required by a student's doctoral dissertation committee, a funding agency, or a journal editor). Note: The IRB will not provide a formal written determination after the project has been initiated. The following application will permit the IRB to make this determination: A. Answer Each of the following questions: Will any information from this project be submitted to the FDA or held for inspection by the FDA? No Yes	Principal Investigator:
 A. Answer Each of the following questions: Will any information from this project be submitted to the FDA or held for inspection by the FDA? No ☐ Yes ☐ Are the data or specimens studied as part of this project obtained in a systematic manner? No ☐ Yes ☐ Is the intent of this data collection to contribute to 'generalizable knowledge' – that is to disseminate the knowledge obtained to others outside the University of Memphis/UofM? No ☐ Yes ☐. If 'no', and this project is being conducted at a UofM facility? No ☐ Yes ☐. Will the study involve intervention or interaction with living persons (i.e., human subjects)? No ☐ Yes ☐ Will the study involve accessing (i.e., looking at or reviewing) identifiable private information? No ☐ Yes ☐ Are the data coded in such that a link exists that could allow the data to be re-identified? No ☐ Yes ☐. Are the data coded in such that a link exists that could allow the PI and the research staff access to the link? No ☐ Yes ☐ 	"research" (see 45 CFR 46.102(d)) and for what is meant by a "human subject" (see 45 CFR 46.102(f)). If there is any doubt as to whether your project qualifies as human subject research you must obtain a formal determination from the IRB as to whether or not project either is not research and/or does not involve human subjects (e.g., as may be required by a student's doctoral dissertation committee, a funding agency, or a journal editor). Note: The
 Will any information from this project be submitted to the FDA or held for inspection by the FDA? NoYes	The following application will permit the IRB to make this determination:
be collected, from what source, and who will collect the sample?	 Will any information from this project be submitted to the FDA or held for inspection by the FDA? NoYes

- 1. Study Aims
 - (a)What is this project intended to accomplish?
- 2. Background and Significance
 - (a) What observations or prior scientific findings serve as the basis for this project?
 - (b) Why is it important to conduct this project?
- 3. Study Design and Methods
 - (a) How will the project be conducted?
 - (b) How will results be analyzed to determine that study aims have been met?
- 4. Types of information to be studied:
 - (a) What data will be accessed?
 - (b) Describe PI's right to access this data.
 - (c) How and where were data collected originally (if applicable)?

5. Summarize the qualifications and experience of the Principal Investigator that are relevant to the conduct this project:

6. Additional Information, Clarification, or Comments for the IRB Reviewer:

CERTIFICATION OF INVESTIGATOR RESPONSIBILITIES

By submitting this form to the IRB, I certify and agree that :

- 1. I am cognizant of, and will comply with, current federal regulations and IRB requirements governing human subject research.
- 2. I have reviewed this protocol submission in its entirety and that I am fully aware of, and in agreement with, all submitted statements.
- 3. I will conduct this research study in strict accordance with all submitted statements.
- 4. I will ensure that all co-investigators, other personnel assisting in the conduct of this research study have been provided a copy of the entire current version of the research protocol.
- 5. I will request and obtain IRB approval of <u>any proposed modification to the research protocol</u> <u>that may affect its designation as an exempt or 'no human subjects' application</u> prior to implementing such modification.
- 6. I will ensure that all members of the research team have satisfactorily completed the) webbased training program accessible on the Module for IRB Applications ("MIA").
- 7. Neither I, nor any member of my research team, will intervene or interact with the humans whose information is being studied in this research project.
- 8. Neither I nor members of my research team will have access to identifiable personal information.
- 9. I will not begin conducting analyses until the status of this application has been determined by the IRB and I have been informed in writing.
- 10. I will respond promptly to all requests for information or materials solicited by the IRB.
- 11. I will maintain adequate, current, and accurate records of research data.
- 12. I will not knowingly include data from prisoners.

Х

PI Acknowledgement & Assurance -(Researcher please type your name above)

