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Speech articulation in children with Williams syndrome or 7q11.23 duplication syndrome.

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SPEECH ARTICULATION IN CHILDREN WITH
WILLIAMS SYNDROME OR 7q11.23 DUPLICATION SYNDROME

By

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A Dissertation
Submitted to the Faculty of the
College of Arts and Sciences
of the University of Louisville
in Partial Fulfillment of the Requirements
for The Degree of

Doctor of Philosophy
In Experimental Psychology

Department of Psychological and Brain Sciences
University of Louisville
Louisville, Kentucky

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DEDICATION

This dissertation is dedicated to my husband, Patrick without whose support, tolerance, encouragement, patience, and steadfast love I would not have succeeded in my quest.

I have carried on with this work continually grateful for the support of my daughters, Carrine P. Powers and Katelyn R. Kellems, their husbands, and my beautiful grandchildren. So often and so graciously they have taken a step back allowing me to proceed with this work.

And, to Shelley Velleman for the many years of collaboration which have afforded me enlightenment, solicitude, and fortitude.

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Our shared experiences have shaped my being. I am a better teacher, therapist, and person because of our times together. In the years to come, I look forward to enhancing our relationships and together learning the best ways for teaching our children to communicate effectively with others.

ABSTRACT

SPEECH ARTICULATION IN CHILDREN WITH
WILLIAMS SYNDROME OR 7q11.23 DUPLICATION SYNDROME

Myra J. Huffman

February 5, 2019

The present dissertation aimed to characterize speech articulation accuracy for children with Williams syndrome (WS) and children with 7q11.23 duplication syndrome (Dup7). Two studies were conducted. Study 1 addressed articulatory accuracy for each group based on citation assessment in single words. Results were compared to expected performance for same aged-peers in the general population. Study 2 evaluated variance relations among speech articulatory accuracy, phonological processing and particular cognitive and linguistic measures.

Results of Study 1 indicate that for both groups, consonant accuracy was significantly below expectations based on age norms. Accuracy was better for children in each older subgroup compared to the respective younger subgroup. The speech of children with WS were more accurate than of children with Dup7 although children with Dup7 obtained higher IQ scores. For both children in the WS group and in the Dup7 group, children with IQs at or above 70 earned significantly higher articulation SSs than did children with IQs below 70. In general, patterns of consonant accuracy as a function of several features of articulation were consistent with patterns reported for children in the general population.

Results of Study 2 indicate that for both children with WS and children with Dup7, articulatory accuracy, overall cognitive ability, spatial ability, and the combined

factor for lexical understanding and use were all moderately, to strongly, related.

Furthermore, for the children with WS, articulatory accuracy contributed unique variance to phonological processing beyond the unique variance contributed by verbal short-term memory, spatial ability, and the combined factor of lexical understanding and use.

Overall, the results showed children in both groups were significantly delayed in consonantal development. Patterns of articulatory accuracy did not differ greatly from those of younger, typically developing children. Furthermore, the findings demonstrated positive relations among articulatory accuracy, phonological processing, and intellectual abilities, and vocabulary abilities for children with these syndromes.

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CHAPTER I

GENERAL INTRODUCTION AND LITERATURE REVIEW

The overarching desire to communicate orally drives efforts to learn how to speak (Kuhl, 2007; Levelt, 1989; Locke, 1993; Oller, 2000). For most children, learning to articulate speech accurately simply involves daily practice expressing thoughts and intentions. For others, this task presents challenges. Despite ease or difficulty learning, all children should be supported in speech development because speaking is essential for social, emotional, and intellectual engagement with others.

Articulation is the technical term used to describe speech production behavior. In this dissertation I address articulatory accuracy for pronouncing English consonants for children with Williams syndrome (WS) and children with 7q11.23 duplication syndrome (Dup7). Articulatory accuracy is considered one component of articulatory competence. Competence involves both speech intelligibility and articulatory accuracy for pronouncing speech sounds in single words and in continuous speech.

Articulation results from volitional, vocal-organ activity (International Phonetic Association, 1999; Kent, 2013) and develops over years of practice speaking (Smith & Zelaznik, 2004; Walsh & Smith, 2002). Research has shown that accuracy for pronouncing consonants is positively related to (a) overall intellectual ability for English-speaking preschool children who are typically developing (Winitz, 1959),

(b) receptive (Templin, 1957) and expressive (Stoel-Gammon, 1998) vocabulary in early childhood, and (c) pre-literacy phonological awareness (McDowell, Lonigan, & Goldstein, 2007; Overby, Trainin, Smit, Bernthal, & Nelson, 2012; Vihman, 2016).

For some children, accuracy pronouncing speech sounds might develop slowly or with disorder (Velleman, 2016). The particular causes for difficulty might be associated with trouble (a) perceiving the sounds (Liu, Kuhl, & Tsao, 2003; Yoshinaga-Itano, Coulter, & Thomson, 2000; Velleman, 1988), (b) cognitively processing speech sounds in association with linguistic information (Jusczyk, 1999), and/or (c) executing the speech movements (Bauman-Waengler, 2012; Caruso & Strand, 1999; Kent, 2000; Ozanne, 2013; Smith, 2010; Vuolo & Goffman, 2017). For children with WS or Dup7, reports suggest many have difficulty learning to speak clearly (Gosch, Städing, & Pankau, 1994; Mervis et al., 2015; Morris et al., 2015; Semel & Rosner, 2003; Udwin & Yule, 1990; Velleman & Mervis, 2011). However, empirical descriptions of these disorders do not provide (a) systematic examination of articulatory accuracy at any age, (b) insight regarding the trajectory of articulatory development, or (c) description of the relation between articulatory accuracy and intellectual ability, phonological processing, or vocabulary.

To address this gap in the literature, I provide the first systematic examination of speech articulatory accuracy for children who have WS or Dup7. Articulatory accuracy was evaluated perceptually based on children's ability to pronounce speech sounds in single words on demand. The children's task was to cite words from picture cues. Thus, the assessment involved a simple, but confrontational, articulatory task. Data for the dissertation were obtained from the Sounds-In-Words subtest of the *Goldman-Fristoe*

Test of Articulation-2 (GFTA-2; Goldman & Fristoe, 2000). This subtest was designed to determine accuracy for consonants pronounced in single-word speech, instead of accuracy for both consonants and vowels or for consonant accuracy in continuous speech.

In the remainder of this chapter, I summarize the background literature that provides the foundation for the two research studies that I have conducted. In the first background section, I define speech articulation. In the next section, I describe five approaches to the study of consonant articulation that account, in part, for the complexity of this skill. In the third section, I review seminal research supporting the validity of the articulatory assessment method used in the present dissertation. In the fourth, I summarize the literature addressing the relation between articulatory accuracy and the development of phonological processing, a pre-literacy ability strongly associated with the development of reading (National Reading Panel, 2000). In the final background section, I summarize the literature regarding relations between the speech and cognitive-linguistic abilities of children with WS or Dup7. Lastly, I provide a brief outline of the two studies that I have conducted.

Articulation

Articulation is behavior performed for communicating orally; speech sounds are the medium used to encode oral language. Accurate articulation is an expected endpoint in the developmental trajectory of speech-motor behavior. The trajectory progresses from incipient immature and highly variable vocalizations to stable and adult-like articulations (Morley, 1965; Templin, 1957; Winitz, 1969). For children developing typically, an initial benchmark along this trajectory is intelligibility to listeners (Flipsen, 2006). Later, children develop the ability to articulate all speech sounds accurately in words (Templin,

1957). By the late teen years, children demonstrate mature speech-motor control for accurate articulation across all contexts of continuous speech (Walsh & Smith, 2002). In the following paragraphs, articulation is described in terms of mechanisms for learning to do it, its developmental trajectory, and its characteristic variability based on the individual speaker and the speaking context.

Articulation is purposeful behavior. It is anchored in development with physical, cognitive, social, linguistic, and phonological abilities (Beitchman, Wilson, Brownlie, Walters, Inglis, & Lancee, 1996; Eaton & Speed, 1995; Newman, Ratner, Jusczyk, Jusczyk, & Dow, 2006; Nip, Green, & Marx, 2010). Conceptualized cognitively as sensorimotor behavior, articulatory accuracy is theorized to develop as a result of learning to refine production of language-specific speech-motor routines (Green & Nip, 2010; Guenther, 2016; Ziegler & Ackermann, 2017) that form the sequences of syllables and sound patterns that constitute words and phrases in a language (Lametti Smith, Freidin, & Watkins, 2018; Rochet-Capellan & Ostry, 2011). Learning to articulate is assumed to be dependent on both implicit and explicit learning mechanisms, including (a) sensorimotor information processing (e.g., multimodal feedforward [predictive] and feedback elements of neural control; Guenther, 2016; Lametti, Nasir, & Ostry, 2012; Perkell, 2012; Riley & Smith, 2003), (b) self-monitoring for accuracy (Hashimoto & Sakai, 2003) and then executing corrections when needed (Eaton, 2015), and (c) practice and experience speaking with others (Vihman, 2016). Research has identified cortical and subcortical speech-motor networks that are dedicated to planning (Hertrich, Dietrich, & Ackermann, 2016), programming (Mersov, Jobst, Dheyne, & De Nil, 2016; Segawa, Tourville, Beal, & Guenther, 2015), and executing articulatory gestures (Behroozmand,

Shebek, Hansen, Oya, Robin, Howard III, & Greenlee, 2015; Hickok, Houde, & Rong, 2011; Houde & Nagarajan, 2016; Kumar, Croxon, & Simonyan, 2016; Simonyan & Fuertinger, 2015; Walsh, Mettel, & Smith, 2015). Studies show articulation is generated in the ventral speech-motor cortex (Ackermann, Mathiak, & Riecker, 2007; Guenther & Hickok, 2016; Riecker, Mathiak, Wildgruber, Erb, Hertrich, Grodd, & Ackermann, 2005), a motor control area of the brain that is shown in adults to be structured phonemically for both movement efficiency and speech learning (Bouchard, Mesgarani, Johnson, & Chang, 2013; Cheung, Hamilton, Johnson, & Chang, 2015; Lotze, Seggewies, Erb, Grodd, & Birbaumer, 2000; Mesgarani, Cheung, Johnson, & Chang, 2016; Terao et al., 2007).

Through development, speech is learned through links that form in memory between specific instances of speaking and relevant memories of actively articulating words (Tremblay, Houle, & Ostry, 2008; Werker & Tees, 1984). For adults, memories for isolated speech sounds are easily adapted or modified for speaking across various lexical contexts (Shiller, Sato, Gracco, & Baum, 2009). There is evidence to suggest speech-sound articulation in words is represented segmentally (Rochet-Capellan, Richer, & Ostry, 2012). However, articulation in context is likely supported by the development of dense cortical links between the primary speech motor area, lexico-semantic processing areas, the areas engaged for processing and programming phonological information, and the areas for integrating suprasegmental features of articulation (Goffman, 1999, 2004; Strijkers, Costa, & Pulvermüller, 2017). Suprasegmental features of speech include prosody, temporal patterning of speech movements, and vocal-tract subsystem activity (e.g., vocal intensity, vocal register, phonetic-voicing effects;

Behrman, 2017; Dromey, & Ramig, 1998; Guenther, 2016; Sörös, Sokoloff, Bose, McIntosh, Graham, & Stuss, 2006; Whitfield, Dromey, & Palmer, 2018).

Behaviorally, learning to articulate a single word accurately entails developing consistency in forming particular sequences of articulatory movements (i.e., target gestures) that appropriately encode aspects of the expressive lexicon. Incrementally, children build up a phonetic repertoire of sounds and syllables useful for communicating information (Scheiner, Hammerschmidt, Jürgens, & Zwirner, 2002). As the receptive lexicon develops, memories for the sound constituents of words develop (Saffran, Aslin, & Newport, 1996). First words tend to fit word forms that are constrained by the speech sounds and sequences under the child's control. The eventual development of articulatory accuracy involves learning to constrain movement precision to the degree necessary for maintaining phonemic contrast in the speech stream (Vihman, 1996).

Speech-sound segments are called *phones*. A phone is essentially a model of a distinct acoustic signal corresponding to a linguistically-relevant articulatory event. A particular phone is produced as a result of tightly coordinated maneuvers among respiratory and vocal tract structures. Specifically, during controlled expiration, articulations of particular tissues are made that involve deformations or oppositional movements of structures at key points along the vocal tract. Mechanistically, phones are induced within the vocal tract in ways similar to inducing resonance within a tube (*re*: the source-filter model; however, the simple tube model neglects both damping due to soft-tissue dynamics and the complexities of time-varying, aerodynamics inherent in continuous speech [Kent, 2013; Kent & Reed, 2002]). Within the vocal tract, resonating cavities are located at the glottis, larynx, pharynx, nasopharynx, and at particular spaces

within the mouth. Tissues capable of deformation or oppositional movement include the diaphragm; lungs; vocal cords; walls of the larynx, pharynx, and cheeks; tongue; velum; jaw; teeth; and lips. Valving, or constricting the resonating flow of air in particular ways, shapes the breath stream into distinctive sounds that have potential for contrasting linguistic meaning. Valving can occur at the vocal cords, the velum, the tongue, and the lips. From this mechanistic description, it is clear that speech articulation involves intricate control of specific sets of organs engaged intentionally for effecting sequences of linguistically-relevant resonance changes within the vocal tract.

Phones are classified as consonants (C) and vowels (V). Single phones that are articulated with a closed or nearly closed vocal tract are called consonants; single phones articulated with an open vocal tract are called vowels. A contiguous sequence of two to four consonants articulated with no intervening vowels is called a consonant cluster. Spoken words are consistent in their sequence of consonants and vowels, but they are inconsistent in kinematic detail (Kelso, Vatikiotis-Bateson, Saltzman, & Kay, 1985; i.e., movement parameters of space, time, and intergestural coordination) and in the acoustic details of pitch, duration, intensity, or timbre (Kent & Reed, 2002).

In contrast to the phone, a *phoneme* is the minimal speech-sound element that functions to signal differences in meaning (Ball, 2003; Kent, 2013). A phoneme is actually a class of phonetic variants (i.e., allophones) intuitively recognized to be equivalent by listeners of a speaking community (Kühnert & Nolan, 1999). Phonemes function to contrast meaning and to indicate morphological structure. (A morpheme is defined as the minimum meaningful element of a language.) In the generative language tradition, a phoneme is defined as a set of distinctive features (e.g., features of major

classes based on laryngeal state, manner of articulation, and place of articulation; Chomsky & Hallé, 1968). In the American structuralist tradition, a phoneme is defined according to its allophones and environments (Hockett & Hockett, 1960).

In the paragraphs that follow, articulation is described in terms of its development. Articulatory development begins very early in childhood and for most it is mastered by young adulthood (Oller, 2000; Walsh & Smith, 2002). Benchmarks in the trajectory include: (a) intelligibility to non-familiar adults by age 4 years (Flipsen, 2006; McLeod, 2013), (b) consistent accuracy in articulating consonants by age eight years (Goldman & Fristoe, 2000; Smit, Hand, Freilinger, Bernthal, & Bird, 1990; Templin, 1957), and (c) competence at variably coordinating complex articulatory gestures in continuous-speech contexts, by adolescence (Rubertus & Noiray, 2018). Often, articulatory behaviors common in early development can be observed at later ages in atypical articulation.

When learning to communicate first words, infants who are developing typically utilize their resources for attending (e.g., attention to referents and to both acoustic and visual speech information; see Burnham & Dodd, 2004; Patterson & Werker, 2003; Posner, Rothbart, Sheese, & Voelker, 2012), extracting meaning from the ambient language (Archer, Zamuner, Engel, Fais, & Curtin, 2016; Maye, Werker, & Gerken, 2002), and, concurrently, exploiting articulatory skills previously learned through babbling (Davis & MacNeilage, 1995; Menn & Matthei, 1992; Oliveira-Guimarães, 2013; Priestly, 1977/2013; Vihman & Velleman, 1989/2013; Waquier & Yamaguchi, 2013). As a result, first words are constructed with (a) the degree of articulatory control needed for communicating successfully with caregivers, (b) a number of systematic

patterns of phonetic forms selected to match particular communicative contexts (Stoel-Gammon, 1985, 1998), and (c) a variety of functional syllable shapes (Amayreh & Dyson, 1998; Chen & Kent, 2005; de Boysson-Bardies, Vihman, Roug-Hellichius, Durand, Landberg & Arao, 1992; Oliveira-Guimarães, 2013; Savinainen-Makkonen, 2013; Stoel-Gammon, 1987).

The onset of speech-like articulation usually occurs within the age range of 5–10 months. During this period, infants' marginal babbling becomes well-timed and canonical; that is, canonical babbling has patterned, syllable-like structure. But, babble's defining feature is the quickness of articulation; transitions between a consonant margin and a vowel nucleus take approximately 100–500 ms. By age 9 months, the transition time has reduced to about 50 ms (Oller, 2000), a value within the range of adult timing for syllables beginning with voiceless consonants (Kent & Reed, 2002). Importantly, delay in the onset of canonical syllable articulation, that is, onset after the age of 10 months, is one indicator of increased risk for subsequent delay or disorder in speech and/or language development (see review in Oller, 2000; Oller, Eilers, Neal, & Schwartz, 1999).

Factors important in the development of articulatory control include familiarity of words heard in the ambient language (Swingly & Aslin, 2002; Zamuner, Gerken, & Hammond, 2005), the phonetic complexity of early word shapes attempted in production (Vihman & Wauquier, 2018), and similarity of phonetic and prosodic structure among words attempted (Vihman, & Croft, 2013). Regarding familiarity, Cutler and Carter (1987) estimated the range of the number of syllables occurring in content words produced by English-speaking adults. They showed that roughly 45% of words had one

syllable, 39% had multiple syllables with strong initial syllables, and 16% were polysyllabic but with weak initial syllables. Locke (1983) examined word structure in terms of phonetic complexity. He estimated that approximately 33% of English monosyllables began with a two-consonant cluster sequence and 18% ended with one. Kehoe and Stoel-Gammon (2001) reported that the most common syllable shape produced in first speech was open and was structured with the consonant plus vowel phonotactic pattern (CV). Slightly older toddlers produced increasing proportions of CVC syllables, likely in response to the frequency of codas (final consonants) in English. By 24 months, toddlers produced more syllables with CVC shape than not (Kehoe & Stoel-Gammon, 2001). Thus, it seems likely that from the earliest ages, phonetic features of words heard have influence on articulatory learning.

For children who are developing typically or atypically, the onset of phonological systematization (Dyson, 1988; Kessler & Trieman, 1997; Locke, 1983; Shriberg, Kwiatkowski, Best, Hengst, & Terselic-Weber, 1986; Stoel-Gammon, 1985; Vihman & Greenlee, 1987; Waquier & Yamaguchi, 2013) is an apparent effect of experience producing about 25 true words consistently and intelligibly (Vihman & Velleman, 1989; Vihman, Velleman, & McCune, 1994). Prior to this point, first words are produced with apparent mimeticity (Ferguson & Farwell, 1975). Phonological systematization often becomes evident through analysis of novel words. When articulating, there is strategic interchange of consonants and vowels under personal control (CV, CVC, or VC). The systematic nature of sequencing the phones characterizes phonology (Kehoe, 2015; Menn, Schmidt, & Nicholas, 2013; Priestly, 1977/2013; Oliveira-Guimarães, 2013; Vihman & Keren-Portnoy, 2013, 2016; Vihman & Velleman, 1989/2013, 2000; Waquier

& Yamaguchi, 2013; Waterson, 1971). Some infants and young children are observed to preferentially and systematically use particular word shapes during the early word-learning period. These templatic patterns are characteristically idiosyncratic and are thought to be phonological compromises between the adult target and the child's phonetic repertoire of sounds, syllables, and phonotactic patterns (i.e., patterns developed from rules governing the possible phoneme sequences used in a particular language).

Templates have strategic function: they facilitate expansion of the lexical repertoire by reducing articulatory load. However, the strategic advantage of using familiar word-form patterns for producing new words results in a repertoire of early word forms having similar shape but inaccurate articulation (Vihman & Wauquier, 2018). Recent evidence has shown that at least some toddlers with WS strategically access familiar word forms (CV, CVC, CV.CV) and adapt phones under personal control when expanding their lexical repertoire (Garber, 2018).

Variability is evidenced in all skilled movement performance (Bernstein, 1967). However, articulatory variability does not equate with articulatory inaccuracy (Fowler & Saltzman, 1993; Kelso, Vatikiotis-Bateson, Saltzman, & Kay, 1985; Smith & Goffman, 1998; Vuolo & Goffman, 2017). Researchers have shown that when speaking in short phrases, the spatiotemporal characteristics of children's speech are both wider and slower than adult speech (Smith & Zelaznik, 2004; Walsh & Smith, 2002) and children make larger displacements of the articulatory organs relative to the size of their faces (Riley & Smith, 2003). But the variable spatiotemporal characteristics of children's speech, as evident in every reproduction of a particular word or phrase, becomes reduced in range with increasing age (Smith & Zelaznik, 2004; Walsh & Smith, 2002) and with language

learning (Heisler, Goffman, & Younger, 2010; Smith & Goffman, 2004). For infants and toddlers who are younger than two years, kinematic measurement of lower lip closing speed has been shown to be positively correlated with concurrent expressive language age equivalent scores ($r = .54, p < .001$; Nip, Green, & Marx, 2011) from the *Battelle Developmental Inventories-2* (Newborg, 2005).

Although subtle variation in the kinematic features of speech articulation is ubiquitous, well-trained listeners of a speaking community have been shown to reliably classify acceptable and unacceptable phonetic variation in targeted phonemes in words (Kent, 1996; Lieberman, Harris, Hoffman, & Griffith, 1954; Miyawaki, Strange, Verbrugge, Liberman, Jenkins, & Fujimura, 1975; but see Mowrey & MacKay, 1990). Accordingly, it is reasonable to expect that, for this dissertation, raters who are thoroughly trained and use appropriate evaluative techniques can determine phonetic accuracy based on phones articulated in single words (Kent, 1996; Shriberg, 1972). The phrase, *traditional phonetic description*, refers to a consistent way of transcribing heard speech in written form using the International Phonetic Alphabet (IPA, 2015). Traditional phonetic description is done to specify distinct phonetic events (i.e., articulations) that theoretically correspond to linguistically relevant phonemic events. Traditionally, IPA symbols are used with the assumption that the audible speech stream encodes the particulates of speech. Further, the orthographic symbols that represent segments of speech are assumed to represent articulatory targets (IPA, 1999). In speech science vernacular, the term *articulatory target* refers to a particular articulatory-movement goal. Thus, particular sequences of articulatory targets (Callan, Kent, Guenther, & Vorperian, 2000) are the volitionally—and intentionally—produced motor goals for speaking. Coding

particulate speech using the symbols and diacritics of the IPA (2015), as was done in the present research, affords evaluation of articulatory accuracy from the written record. The IPA has permitted open source reproduction of its phonetic charts; these charts are presented for reference in Appendices A and B.

The overarching theoretical perspective of my dissertation is based on a neuroconstructive view of cognition (Farran & Karmiloff-Smith, 2012). Speech-motor cognition, as reflected in the trajectory of articulatory competence through development, is easily framed within this approach because research supporting this view is designed to consider (a) emergent properties of developmental trajectories and (b) emergent biological, physical, and social outcomes constrained by ontogeny, dynamic environment-gene relations, and the probabilistic nature of broad genetic control networks (Karmiloff-Smith, 1992; Karmiloff-Smith & Farran, 2012). Consistent with this view, the present dissertation was designed to evaluate articulatory accuracy for two groups of children aged 4–17 who have reciprocal genetic disorders; that is children with WS or Dup7. In this work, perceptual judgments of articulatory accuracy have been evaluated for children with each disorder and for children in younger and older subgroups.

In the next sections, I review typical patterns of accuracy for GFTA-2 consonant items. Patterns of articulatory accuracy are discussed with regard to five features of consonant articulation. The data collected for this dissertation were analyzed in each of these five ways with the goal to determine aspects of articulation that might be more or less difficult for either group of children. Potential strengths and weaknesses found across

the constituents of the features of articulation could be exploited for customizing articulation treatment.

Age of Customary Consonant Production

An important benchmark of articulatory development is *Age-of-Customary Consonant Production* (ACCP; ASHA, 2017). This frequently used and commonly accepted referent for typical consonant *acquisition* describes a point in developmental time (i.e., “age”) at which a specified percentage of children within a particular speaking community can be said to consistently articulate a particular consonant accurately (Bernthal, Bankson, & Flipsen, 2013). Delayed ACCP is useful for gauging the status of articulatory development and for determining whether formal speech assessment should be initiated (ASHA, 2017).

In Table 1, the consonants assessed by the GFTA-2 are grouped and stratified according to three ACCP levels for consonants and consonant clusters: early-developing, middle-developing, and late-developing. In two columns, consonants are divided into singletons or clusters, and within each of these classifications, (a) consonant singletons are organized by manner and place of articulation (Ladefoged, 2005), and (b) clusters are organized by manner and common phone.

I have assigned ACCPs in Table 1 based on two large seminal studies of typical acquisition of consonants produced by English-speaking children: Templin (1957) and Smit et al. (1990). These two studies were referenced because each was conducted using citation methodology, included large numbers of child participants who spoke American English, and used the same criterion for assigning ACCP as used in the present study. ACCP was defined as the age at which a particular consonant was produced correctly by

Table 1.

Typical Age of Customary Consonant Production (ACCP) for Consonants and Consonant Clusters Assessed by the GFTA-2^a

| ACCP | Singleton Consonants ^b | Consonant Clusters |
|--|--|--|
| Early-Developing Phones: Phones acquired prior to 5 years, 0 months | <ul style="list-style-type: none"> • <i>bilabial-velar glide</i> (approximant class) /w/ • <i>nasals:</i> <ul style="list-style-type: none"> - <i>bilabial</i> /m/ - <i>alveolar</i> /n/ • <i>stops:</i> <ul style="list-style-type: none"> - <i>bilabial</i> /p, b/ - <i>alveolar</i> /t, d/ - <i>velar</i> /k, g/ • <i>fricatives:</i> <ul style="list-style-type: none"> - <i>glottal continuant</i> /h/ - <i>labiodental</i> /f/ | <ul style="list-style-type: none"> • <i>stop + bilabial-velar glide:</i> /kw/ |
| Middle-Developing Phones: Phones acquired prior to 7 years, 0 months | <ul style="list-style-type: none"> • <i>post-alveolar-palatal glide</i> (approximant class) /j/ • <i>fricatives:</i> <ul style="list-style-type: none"> - <i>alveolar, postalveolar-palatal</i> /s, ʃ/ - <i>labiodental</i> /v/ • <i>post-alveolar-palatal affricates:</i> /tʃ, dʒ/ | <ul style="list-style-type: none"> • C + /l/ clusters: /fl, gl, kl/ • /s/ + C clusters: /sp, st/ |
| Late-Developing Phones: Phones acquired prior to 9 years, 0 months | <ul style="list-style-type: none"> • <i>velar nasal</i> /ŋ/ • <i>fricatives:</i> <ul style="list-style-type: none"> - <i>alveolar</i> /z/ - <i>linguadental</i> /θ, ð/ • <i>liquids:</i> (approximant class) <ul style="list-style-type: none"> - <i>alveolar/lateral</i> /l/ - <i>postalveolar-palatal</i> /ɹ/ | <ul style="list-style-type: none"> • /s/ + C clusters: /sw, sl/ • C + /l/ clusters: /pl, bl/ • C + /ɹ/ clusters: /bɹ, dɹ, fɹ, gɹ, kɹ, tɹ/ |

^aSources: Templin (1957) and Smit et al, (1990). ^bThe alveolar fricative /z/ is not included because it is not assessed by the GFTA-2 due to rare usage by children.

at least 75% of children. If Templin (1957) and Smit et al. (1990) assigned a different ACCP to a particular item, the later age was used in arranging the item in Table 1. The only exception was medial /t/, for which Templin (p. 49) noted a very late age (likely due to dialectal variability for the stimulus word). Note that data from Goldman and Fristoe (2000) were not considered in constructing Table 1 because these authors used a different criterion for ACCP (85% of children needed accuracy for the item rather than 75%). For clusters, ACCP was defined as the age at which a particular cluster was produced correctly in the initial word position by at least 75% of children. This restriction was made because the GFTA-2 includes consonant clusters only in the initial word position.

Singleton consonants. Based on the arrangement of phonemes in Table 1, English-speaking children who have turned 5 years old are expected to accurately produce the following in all positions of words: (a) all singleton stop consonants /b, p, d, t, g, k/, (b) two of the three English nasal consonants /n, m/, (c) the fricatives /h/ (glottal continuant) and /f/ (labiodental), and (d) only one of the four English approximant consonants, that is, the bilabial-velar glide, /w/. Children who have turned 7 years old are expected to also produce the following consonants in all positions of words: (a) the approximant /j/ (a palatal glide produced initially in the word *you*), (b) several voiced and unvoiced fricatives (labiodental, alveolar, and postalveolar-palatal), (c) the palatal affricates (/dʒ, tʃ/), and (d) several /s/ and /l/ consonant clusters. Additionally, children who have turned 9 years old are expected to also produce the following consonants in all positions of words: (a) the dialectically variable nasal /ŋ/, (b) the fricatives /z/ (alveolar) and /θ, ð/ (linguadental), (c) the dialectically-dependent and articulatorily-challenging

approximant (liquid) consonants /ɹ/ and /l/, and (d) several /s/, /l/, and /ɹ/ double consonant sequences.

Clusters. The arrangement in Table 1 indicates the clusters that are included in the GFTA-2 are expected to be acquired within an extended period from < 5.00–8.99 years. Please note that many other consonant sequences that occur frequently in the speech of English-speaking children are not tested on the GFTA-2. Furthermore, the table presents a discrete range of acquisition although reports in the literature have indicated high variability among individual children for acquiring consonant clusters. Dyson (1988) reported that the developmental trajectory for clusters is characterized also by reversals and revisions. Also, the dissertation design is cross-sectional and includes some four-year-old children. Therefore, the relevant data regarding acquisition of cluster accuracy was interpreted conservatively.

Researchers have postulated plausible reasons for inconsistencies in the cluster-learning trajectory, including (a) natural variability in motor learning, immaturity, or impaired, oral-motor anatomy and/or physiology (Smith; 2010), (b) the complexity inherent in timing, sequencing, and transitioning among the double phones (Byrd & Tan, 1996), and (c) limitations imposed by delayed or impaired cognitive-linguistic development (MacNeilage, 2008). In the dissertation, clusters were combined with affricate items for the purpose of analysis. To indicate the combination, a different category title, double consonants, was applied.

Articulatory Position-In-Words

Articulatory Position-in-Words is a descriptive class that includes 37 GFTA-2 singleton items studied for the purpose of assessing articulatory accuracy at the onsets

and codas of syllables or single words (initial and final word positions). The phonetic and syllabic complexity of a word's structure affects the accuracy of its articulation (Macrae, 2013; McLeod & Hewett, 2008; Sosa & Stoel-Gammon, 2012). Syllables are units of words that usually consist of a sonorous element (typically, a vowel nucleus) with optional initial and final margins (consonants). Phonetic complexity at syllable margins contributes to articulatory load and it involves, in part, the phonotactic regularity (known familiar arrangements) of phonemic sequences in words. Languages are constrained by phonotactic structure; that is, they are constrained by the sequences of consonants that are permitted to occur between two vowels within a word. Consonant sequences that violate word-internal phonotactic constraints provide cues to where word boundaries exist (Brent & Cartwright, 1996; Jusczyk, 1999). Although many words in the GFTA-2 have multiple syllables and thus have internal consonants, the medial consonants in the GFTA-2 were not examined because the stimulus words in it were not consistent in phonotactic pattern.

Also pertinent to the present discussion is the body of research investigating the onset and decline of phonological-process errors (Hodson, 2004; Hodson & Paden, 1981). Phonological-process errors are systematic phonetic or phonemic errors that occur typically in the speech of young children (e.g., final consonant deletion; Hodson & Paden, 1981; Khan, 1982; Shriberg & Kwiatkowski, 1986) and then become increasingly rare as children grow older. For example, three errors, (a) final consonant deletion, (b) fronting of back consonants, and (c) stopping of continuants, rarely persist in typical development after children have achieved age 3.5 years (Roberts, Burchinal, & Footo, 1990). Furthermore, for native English learners, other deletions are atypical at any age, such as deletion of initial- or medial-consonant singletons, (Hodson & Paden, 1981; Khan

& Lewis, 2002; Roberts et al., 1990). Phonological-process errors that have persisted in the speech patterns of children with WS or Dup7 likely impact results of the articulatory position-in-words feature analysis conducted in Study 1 below.

In summary, justification for considering accuracy at the initial and final positions of words obtains from three lines of research. First, seminal normative reports on consonant acquisition in childhood consider phonemes classified by position in words (Goldman & Fristoe, 2000; Smit et al., 1990; Templin, 1957). Second, researchers have shown that children who are typically developing perceive and produce words (a) as structured units with vocalic nuclei marginalized by adjacent consonants (i.e., onsets and codas; Kent & Reed, 2002; Oller, 2000) and (b) within the bounds of their language's phonotactic rules (particular and predictable sequences of phones that are more likely to occur in the native language; Bruderer, Danielson, Kandhadai, & Werker, 2015; Werker & Yeung, 2005; Zamuner, Gerken, & Hammond, 2005). Third, emerging cross-linguistic research shows phonological development is characterized by an early period of whole words selected for and adapted to fit preferred word forms, or templatic patterns (Menn, 2013; Vihman & Keren-Portnoy, 2013). (*N.B.*: The use of templates, or idiosyncratic phonotactic patterns, is expected to decline early in development.) Thus, empirical and longitudinal research has shown that very young typically developing word-learners quickly learn to produce utterances with language-permitted, closed (CVC) syllable shapes (Kehoe & Stoel-Gammon, 2001).

Planes-of-Movement for Consonant Cluster Articulation

Hayden and Square (1994, 1999) suggested that the development of movement control for articulatory accuracy occurs across particular vocal tract planes-of-movement:

anterior-to-posterior, superior-to-inferior, and across diagonals. Given that some of the participants in this dissertation were known to have speech sound disorder (Huffman, Velleman, & Mervis, 2012; Velleman, Huffman, & Mervis, 2013), it was expected that movement transitions might affect accuracy for some participants to a greater degree than others. Although underspecified in the GFTA-2, items were controlled for articulatory directional plane-of-movement and thus it was possible to code each consonant cluster and each affricate for directional transition: anterior-to-posterior, posterior-to-anterior, or same place and then, to follow coding with examination of each item for accuracy. This procedure afforded a simple platform for evaluating accuracy as a function of the direction of speech movement transitions.

In the GFTA-2, expectations for consonant cluster acquisition in the general population were based on findings from the seminal literature (Smit et al, 1990; Templin, 1957). Thus, the study of planes-of-movement for consonant sequences included study of the 16 initial, consonant-cluster items and the two affricates (6 items) included in the GFTA-2. (*N.B.*: An affricate item is a phoneme that combines a plosive component with a fricative component immediately following it, and for which both components share the same place of articulation [e.g., “-dge” /dʒ/ and “-ch” /tʃ/ in the initial-, medial-, or final-word positions]).

Articulatory Targets

Articulatory targets are movement goals for pronouncing phones. In the present dissertation, they included singleton or consonant-cluster items cued for production by pictures presented in the GFTA-2. The following two subsections provide literature

support for arranging consonants in groups according to targets for (a) articulatory place-of-production and (b) articulatory manner-of-production.

Articulatory place-of-production. For the purpose of the present dissertation, articulatory place-of-production designates different locations within the vocal tract at which major articulatory events occur in English. The place locations are labeled in general agreement with the work of Ladefoged (2005) and with the IPA (2016; Appendix A). (*N.B.*: The place designations do not consider (a) the glottal source, (b) accompanying laryngeal and/or velar constrictions, or (c) those consonants with multiple articulations of the same degree or stricture; see Ladefoged & Maddieson, 1996). For the purpose of improving statistical power in the dissertation analyses, some of the nine locations (specifically in the central and posterior oral cavity) that were discussed in Ladefoged and Maddieson (1996) have been collapsed into the five places used in this work.

The place-of-production arrangement includes 55 GFTA-2 singleton items, each grouped in one of five ways: (a) *bilabial* (salient articulation at the two lips), (b) *dental* (articulation using the lips or tongue against the teeth), (c) *alveolar* (articulation of the tongue at or near the bony area immediately behind the front teeth), (d) *postalveolar-palatal* (articulation of speech organs in the area of the vault of the hard palate), and (e) *velar-glottal* (articulation of speech organs in the area of the back of the mouth or in the upper throat; Ladefoged, 2005, p. 115). The place-of-production arrangement does not include affricate or consonant cluster items.

Stoel-Gammon (1998, p. 99) reported that by age 24 months, the phonetic inventories of typically-developing English-speaking children contained both the voiced

and unvoiced labial ([b] and [p]), alveolar ([d] and [t]), and velar ([g] and [k]) stop consonants; the labial ([m]) and the alveolar ([n]) nasal consonants; the glides including that produced with bilabial-velar coordination ([w]), and that produced at the palate ([j]); the glottal continuant ([h]); and some fricatives, usually the voiceless labiodental ([f]) or the alveolar fricative ([s]). Templin (1957, p. 51) reported that 75% of the 60 children in her study aged 2 years, 11 months through 3 years 1 month correctly articulated (a) the bilabial consonants /b, p, m, w/ in the initial and medial word positions, and /p, m/ in the final position; (b) the labiodental consonant /f/ initially, medially, and finally; (c) the alveolar consonants /n, t, d/ initially, /n, d/ medially, and /t, n/ finally; (d) the velar consonants /k, g/ initially and finally, and /ŋ/ medially and finally; and (e) the glottal /h/ initially and medially in words. Templin (1957) also reported that at 49 months all bilabial, velar, and glottal consonants were produced correctly by 75% of children while some dental, palatal, and alveolar consonants had not yet been acquired.

Thus, dental consonants (/θ, ð/) and palatal consonants (/ʃ, ʒ, tʃ, dʒ, ɹ/)—with the possible exception of /j/—are acquired later than most consonants in other places of articulation. Some alveolar consonants, notably /l, z/ and sometimes /s/, are also acquired later. These later consonants all have more challenging manners of articulation.

Articulatory manner-of-production. Study of the articulatory manner-of-production consonant-group arrangement provided insight into accuracy for articulating consonants according to the ways the vocal tract can be shaped and the breath stream modified for speaking English. According to Ladefoged and Maddieson (1996), there is an interrelation between place, manner, and duration especially for the accurate articulation of fricatives. Stoel-Gammon (1985) and Smit et al. (1990) each reported that

acquisition of stops and nasals in initial and final word positions precedes acquisition of fricatives, liquids, and affricates in initial and final word positions.

In the present dissertation, the articulatory manner-of-production consonant-group arrangement included 55 GFTA-2 singleton items, each grouped in one of four ways: Nasal, Stop, Fricative, and Approximant. Ladefoged (2005) described manners of articulation as follows: *Stop* involves complete closure of the vocal tract so that breath is blocked from going out through the nose and through the mouth. *Nasal* involves closure of the oral cavity such that breath can go out through the nose, but not through the mouth. *Fricative* involves constriction of the vocal tract so that a noisy breath stream is formed. *Approximant* involves constriction of the vocal tract to a smaller extent than that required for a noisy breath stream.

For the purpose of power in statistical analysis, and for exclusivity among the consonant manner groups, neither the affricate nor the lateral manners of articulation described in Ladefoged (2005) are included as feature classes in the present dissertation. Ladefoged considered the English lateral, /l/, also an approximant consonant. So, in the present dissertation, /l/ is grouped with other consonant approximants. Ladefoged defined an affricate as “a stop followed by a fricative made at the same place of articulation” (2005, p. 117). On this basis, the two GFTA-2 affricate phonemes /tʃ/ and /dʒ/ were considered double consonant articulations produced with changes of manner (from stop to fricative) and thus excluded from the manner arrangement.

Citation Method of Articulatory Assessment

The most efficient and commonly used method for assessing speech articulatory accuracy is the citation method wherein the examinee names pictures depicting single

words. This method affords an examiner easy identification of the intended word target, permits quick transcription of several consonants per word, and does not limit the examiner who wants to capture additional information related to speech-motor timing and speech motor control for inter-word phoneme sequencing.

Findings from the US National School Speech and Hearing Survey (US NSSHS; Hull, Mielke, Willeford, & Timmons, 1976), which used the first edition of the GFTA (Goldman & Fristoe, 1969), indicated that almost all American-English-speaking 8-year-olds pronounce single words accurately (National Institute on Deafness and Other Communication Disorders, 2016). In particular, of the 2,795 8-year-olds assessed, 87.1% had mastered the repertoire of English consonants assessed on the GFTA. About 50% of the 8-year-old children did not make any errors. Overall, the US NSSHS (1976) findings are consistent with other cross-sectional speech-articulation investigations conducted within the United States (Goldman & Fristoe, 2000; Smit et al., 1990; Templin, 1957). Similar findings of adult-like speech accuracy for 8-year-old children have been found in the United Kingdom (Dodd, Holm, Hua, & Crosbie, 2003).

Related to this line of research are indicators of prevalence for speech-sound disorders (SSD) determined based on single-word citation. Approximately 24% of 5-year-olds are diagnosed with SSD. The rate of SSD rapidly decreases to approximately 2% at eight years of age (Hull, Mielke, Willeford, & Timmons, 1976; Law, Boyle, Harris, Harkness, & Nye, 2000; National Institute on Deafness and Other Communication Disorders, 2016; Shriberg, Tomblin, & McSweeney, 1999).

Association between Articulation and Intellectual or Vocabulary Abilities

Reports of analyses addressing relations between typically developing children's performance on measures of speech articulation and on measures of intellectual ability are summarized in Table 2. All correlations were positive and significant.

Articulatory accuracy was weakly to moderately correlated with intellectual ability. In Study 2, these relations will be determined for children with WS or Dup7.

Phonological Information Processing

Phonological information processing has been defined as efficient use of phonological information during processing (Torgeson, 1991). The term *processing* in this context refers to cognitive processes that are involved specifically in the storage or retrieval of phonologically coded information (Torgesen, Wagner, & Rashotte, 1994; Wagner & Torgeson, 1987; Wagner, Torgeson, & Rashotte, 1994, 1997). Accordingly, the cognitive components of phonological processing include (a) phonological synthesis, (b) phonological analysis, and (c) phonological memory. The findings from three studies that examined the relations between articulatory accuracy and vocabulary or phonological awareness are provided in Table 3.

Phonological awareness involves explicit awareness of the sound structure of language. It is viewed as distinct from word meaning and is inclusive of multilevel skills for consciously recognizing syllable structure, onset-rime relations, and phonemic categories (Gillon, 2000, 2017). From this perspective, phonological awareness can be viewed as an apex ability with developmental roots in nascent word learning (Vihman & Keren-Portnoy, 2013).

The summative report of the National Reading Panel (2000) focused on phonemic awareness, a subcomponent of phonological awareness.

Table 2

Relations Among Measures of Articulation and Measures of Cognition for Typically Developing Children

| Reference | N, Age Range | Measure of Speech Articulation | Measure of Intellectual or Language Ability | Correlation Value |
|---|--|--------------------------------|--|------------------------|
| Winitz. (1959) | 150, 5.0 – 6.0 years | TSTA RS | WISC Full Scale IQ | $r = .34$ |
| Overby, Trainin, Smit, Bernthal, & Nelson. (2012) | 272, Grade 2 | TPIAT RS | California Test of Mental Maturity (nonverbal IQ) | $r = .27$ |
| Templin. (1957) | 60, 7.0 – 8.0 years 60, 8.0 – 9.0 years | TSTA RS | Stanford-Binet Full Scale IQ Stanford-Binet Full Scale IQ | $r = .39$ $r = .29$ |

Note: California Test of Mental Maturity (Sullivan, Clark, & Tiegs, 1957); IQ = intelligence quotient; RS = raw score; r = correlation statistic; S-B = *Stanford-Binet Intelligence Scale, 2nd edition* (Terman & Merrill, 1937); SS = standard score; TPIAT = *Templin Prekindergarten Imitation Articulation Test* (Templin, 1957); TSTA = *Templin Screening Test of Articulation* (Templin, 1953); WISC = *Wechsler Intelligence Scale for Children* (Wechsler, 1949).

Table 3

Relations Among Measures of Articulation, Phonological Processing, and Vocabulary for Typically Developing Children

| Reference | N, Age Range | Measure of Speech Articulation | Correlated Variables | Correlation Value |
|--|--|--------------------------------|--|--|
| Winitz (1959) | 150, 5.0 – 6.0 years | TSTA RS | Phonological Memory Rapid Rime Naming Task RS | $r = .34$ |
| McDowell, Lonigan, & Goldstein (2007) | 700, 24 – 72 months | GFTA RS | Expressive Vocabulary: EOWPVT SS Receptive Vocabulary: PPVT-R SS Phonological Awareness Rhyme Matching Task RS Phonemic Awareness Blending Words Task RS Elision Word Task RS Phonological Memory: NWR RS | $r = .54$ $r = .55$ $r = .45$ $r = .39$ $r = .49$ $r = .58$ |
| Overby, Trainin, Smit, Bernthal, & Nelson (2012) | 272, Spring, Kindergarten 272, Spring, Kindergarten 272, Spring, Grade 1 | TPIAT RS | Receptive Vocabulary: FRPVT SS Expressive Vocabulary: BDCW RS Phonological Awareness Factor Score | $r = .19$ $r = .22$ $r = .46$ |

Note: BDCW = Berko Definitions of Compound Words (Berko, 1958); EOWPVT = *Expressive One-Word Picture Vocabulary Test* (Gardner, 1990); FRPVT = Full-Range Picture Vocabulary Test (Ammons & Ammons, 1948); GFTA = *Goldman-Fristoe Test of Articulation* (Goldman & Fristoe, 1969); NWR = nonword repetition; PPVT-R = *Peabody Picture Vocabulary Test-Revised* (Dunn & Dunn, 1981); RS = raw score; SS = standard score; TPIAT = *Templin Prekindergarten Imitation Articulation Test* (Templin, 1957); TSTA = *Templin Screening Test of Articulation* (Templin, 1953).

Phonemic awareness was defined as explicit awareness of and capacity for manipulating individual phonemes in spoken syllables and words. The National Reading Panel report stressed the importance of including explicit phonemic awareness instruction when teaching children to read because this type of instruction results in strong and significantly improved reading and spelling with effects lasting well beyond the end of training.

Implicit or explicit phonological information processing appears to be engaged whenever speech-motor potential is garnered for pronouncing new words (Guenther, Hampton, & Johnson, 1998; McCune, 2013), whenever decoding strategies are used for identifying novel written words (Lieberman, Shankweiler, Fischer, & Carter, 1974; National Reading Panel, 2000), or when one learns to reduce one's native accent in pronouncing words in a second language (Arteaga, 2000; ASHA, 2018; Brady & Shankweiler, 1991; Fowler, 2011; Locke, 1993; Munson, Swenson, & Manthei, 2005; Paul & Norbury, 2007; Torgesen, 1991). The Phonological Processing subtest of the Differential Ability Scales-II (DAS-II; Elliott, 2007) is used in the present dissertation to measure phonological information processing.

Early Phonological Development

In considering children who make articulatory errors, it is helpful to understand the patterns of errors that also are found in the early period of phonological development for typically developing children. About halfway through the first year of life, infants who are typically developing initiate vocal interactions that are characterized by speech-like syllables. Thereafter, they do so with greater frequency and across an increasing number of social settings (Cohn & Tronick, 1989; Oller, 2000). By the end of the first

year, canonical syllables serve to filter infants' selection of first words (de Boysson-Bardies & Vihman, 1991; Oller, 2000; de Boysson-Bardies, Vihman, Roug-Hellichius, Durand, Landberg, & Arao, 1992; Vihman, Velleman, & McCune, 1994). When infants filter word choices according to articulations under their personal control, it is more likely that listeners will understand what they say. The apparent existence of an early word-learning schedule influenced by experience articulating phones suggests that infants' choice of early lexical items is constrained by the articulations required to execute the items (Vihman, 1996). Thus, infants' active development of phonetic systematicity is foundational to the development of the expressive lexicon (Stoel-Gammon, 1998).

Theoretically, a period of phonetic coalescence occurs for very young word-learners who are in the age range of 12 to 22 months. This period, conceptualized by Locke (1983) and further investigated by Metsala and Walley (1998), is when implicit knowledge of allophonic variation begins to develop. That is, during this period children begin to subconsciously recognize that particular phones that have specific differences in sound fit functionally within a broader class of "phoneme" and that these phonemes distinguish words in the language that they are learning. For example, the [s] in "see" and the [s] in "I'm Sue" are both members of the noisy /s/ phoneme category despite the minute acoustic and articulatory differences that result from the subsequent vowel or, within continuous speech, from the preceding phone. Similarly, children learn that either the aspirated [t^h] or the glottal stop [ʔ] are functional phonemic choices for closing the word "coat". Thus, familiar word shapes, phonetic patterns in the language, and the communicative environment cue children regarding their choices for articulating because

the sounds and movements for particular phones come to be recognized as linguistically and/or functionally equivalent to others (i.e., allophones do not change word meaning).

For the purpose of the present dissertation, the term, allophonic variation, is restricted to indicate within phonemic-class phonetic similarities. Despite allophonic differences within a phonemic class, mature speakers within a speaking community do not necessarily discriminate among them (Kuhl, 2000). It is likely that nascent phonetic organization (or allophonic coalescence) is brought about through cognitive processes of (a) exemplar identification [i.e., use of generalization and analogy across phones (Macken, 1975)], (b) cognitive induction through action and association (Locke, 1983; McCune, 2008), and (c) procedural learning gained across ongoing experiences articulating words (Velleman & Vihman, 2002; Vihman & Croft, 2013). From this view, phonetic coalescence precipitates phonological systemization. Incipient phonological systematization is an apparent effect of experience articulating about 25 words (Vihman & Velleman, 1989; Vihman, Velleman, & McCune, 1994).

As described above, some children have been observed to make strategic use of word templates during the earliest period of word learning (Vihman & Keren-Portnoy, 2013). Although it is not known whether the application of templatic shapes across novel words is ubiquitous in phonological development (Oliveira-Guimarães, 2013), its occurrence is plausible evidence of phonological systematicity and is objective evidence of productive capacity for articulating a sequence of consonants in marginal relation with syllable nuclei. Although word templates are child-specific, they characteristically conform to a general pattern or rule (they are canonical), have a given phonotactic pattern and/or have similar sound features such as, CV; CVC; VC; or CVC.CVC (Velleman &

Vihman, 2002). The use of word templates have been observed in early speech despite the fact that some of the resulting patterns were not permissible patterns in the child's native language. Vihman and Croft (2013) argue that the word template is the primary unit of phonological representation and that knowledge of phonemes derives from it.

Most children continue to make some articulatory errors well into the preschool period. But key to children's capacity for being intelligible to unfamiliar adults is the systematicity with which they make errors. As children learn more language, as they mature physically, and as they resolve phonological error patterns, they gain important phonological competencies (e.g., phoneme matching, rime awareness) that form the foundation for the development of explicit phonological awareness (Hayiou-Thomas, Carroll, Leavett, Hulme, & Snowling, 2017).

Phonological Awareness

During the preschool years (approximately ages 3–6 years), and typically as a result of formal instruction, children become able to recognize, label, and play with speech phonemes or groups of phonemes (Burgess & Lonigan, 1998). As described above, this ability is called phonological awareness (Gillon, 2000; 2017). Linguistic environments where children are exposed to highly salient and well-spoken syllables support the development of explicit phonological awareness (Anthony & Francis, 2005; NRP, 2000; Torgesen, Wagner, & Rashotte, 1994), a benchmark of cognitive development (Scarborough, 1998a). Phonological awareness signals readiness for learning to read (Dickinson, McCabe, Anastasopoulos, Peisner-Feinberg, & Poe, 2003; National Reading Panel, 2000). At the point of acquisition of phonological awareness, two pre-literacy abilities have become well-integrated with knowledge of word meanings

(i.e., lexical semantics): speech articulatory accuracy and explicit awareness of phonological systematicity in the language (Ball, 2003; Beckner et al., 2009; Bybee, 2001). For example, children who have developed phonological awareness can list words starting or ending with the same phoneme, can identify the number of syllables in a particular word, and can rhyme words.

Carroll, Snowling, Stevenson, and Hulme (2003) assessed 67 preschool children using tasks from the Phonological Abilities Test (PAT; Muter, Hulme, & Snowling, 1997). Articulatory accuracy, computed as the percentage of consonants correct (PCC) for 21, 2- to 3-syllable word items, was assessed when the children were on average 4 years 2 months old. A confirmatory factor analysis showed that PCC was significantly and positively correlated with large segment awareness, a latent factor derived from two phonological awareness tasks ($r = .26$; PAT Syllable Completion and Phoneme Completion subtests). A structural equation model (large segment awareness factor, letter group knowledge, PCC, and receptive language factor) further indicated that PCC measured at age 4 years 2 months significantly predicted phonemic awareness (PAT Phoneme Completion, Phoneme Deletion, and Initial Phoneme) eight months later. However, the problematic results were likely impacted by the SEM's relatively small sample size. Furthermore, these analyses did not account for factors previously shown important for the development of phonology such as cognitive processes, memory-related abilities, and the integrity of the productive lexicon (Fowler, 1991; Locke, 1983, 1993; Menyuk & Menn, 1979; Metsala & Walley, 1998; Torgesen & Burgess, 1998).

Phonemic awareness. Described briefly above, phonemic awareness is considered both an advanced, metalinguistic, phonological information processing ability

and a component of overall phonological awareness (National Reading Panel, 2000). Its achievement in development signifies explicit knowledge of the segmental nature of language and the capacity for mentally manipulating speech phonemes. Examples of phonemic awareness tasks include: blending isolated phonemes to create new words or pseudowords, deleting phonemes in words to create new words or pseudowords, and segmenting words or pseudowords into constituent phonemes. Phonemic awareness is a foundational skill crucial for the development of reading and spelling (National Reading Panel, 2000).

Using the Templin Archive (Templin, 2004), Overby et al. (2012) considered the relations between articulatory accuracy assessed in the fall of kindergarten and performance on measures that have been shown to be related to the development of literacy. A concurrent statistically significant relation between articulatory accuracy and receptive vocabulary was found ($r = .19$). In addition, positive and statistically significant correlations were found between articulatory accuracy and the following variables: (a) first-grade orthographic letter knowledge ($r = .39$), (b) first-grade single-word reading ($r = .44$), (c) spring of first-grade phonological awareness composite ($r = .46$), (d) fall of second grade nonverbal cognition ($r = .27$), (e) second-grade single-word reading ($r = .42$), and (f) third-grade spelling ($r = .40$). These results are consistent with findings of a meta-analysis of relations among skill variables associated with the development of reading competency (Scarborough, 2001).

The extant literature is clear: By age 8 years most English-speaking children who are typically developing have acquired accurate speech articulation for sounds in single words and have become explicitly aware of the segmental nature of their language.

Furthermore, children's knowledge of speech sounds is related positively and significantly to elements of lexical knowledge (McDowell et al., 2007), phonological processing (Overby et al., 2012), and early literacy (Parrila et al., 2004). Key developments that contribute to this knowledge base include the following: In infancy, the production of speech articulation begins with canonical babbling; babbling is apparently a canalized behavior (evolutionarily robust; de Boysson-Bardies & Vihman, 1991; Fowler, 1991; Lee, Davis, & MacNeilage, 2010; Masataka, 2003; Oller, 2000; Oller & Eilers, 1982; Pettito & Marentette, 1991). Experience articulating supports the onset of phonology (Vihman, 1996). The two behaviors are linked reciprocally through development (Stoel-Gammon, 1998; Vihman & Keren-Portnoy, 2013). Articulatory accuracy improves as skill using the language improves (Dodd, Holm, Hua, & Crosbie, 2003; Goldman & Fristoe, 2000; Prather, Hendrick, & Kern, 1975; Sander, 1972; Smit et al., 1990; Templin, 1973), and it is positively associated with pre-literacy phonological awareness (Parrila et al., 2004; Overby et al., 2012). Phonological awareness is an ability strongly related to the development of reading (Lonigan, Anthony, Phillips, Purpura, Wilson, & McQueen, 2009; Deacon, & Kirby, 2004; National Reading Panel, 2000; Parrila et al., 2004; Vellutino, & Scanlon, 1991).

7q11.23 Neurodevelopmental Disorders

Neurodevelopmental disorders are a group of conditions primarily associated with brain or central nervous system dysfunction. Examples are attention deficit disorder, intellectual disability, communication disorders, specific learning disorder, developmental coordination disorder, and autism spectrum disorder (*Diagnostic and Statistical Manual of Mental Disorders, 5th edition*; DSM-5; American Psychiatric

Association, 2013). Neurodevelopmental symptoms often are apparent early in development and typically result in impairments of personal, social, academic, and/or occupational functioning. Observed speech and motor symptoms indicate delay or disorder (Shriberg & Mabbie, 2017). Developmental deficits can range from very specific limitations in particular areas to comorbid conditions with severe global dysfunction (DSM-5, 2013). Genetic alterations are associated with many neurodevelopmental disorders.

WS and Dup7 are neurodevelopmental disorders caused by genetic alterations of a set of 26–28 genes on the long arm of chromosome 7. Individuals with classic WS have a deletion of these genes on one chromosome so only have one copy of these genes. In contrast, individuals with Dup7 have an extra copy of these genes on one chromosome so have three copies of these genes. Both WS and Dup7 are relatively rare; the estimated prevalence is the same for each syndrome: 1 in 7500 live births (Strømme, Bjørnstad, & Ramstad, 2002; Velleman & Mervis, 2011). Each syndrome is associated with characteristic sets of symptoms; speech sound disorder is one such symptom (Mervis et al., 2015; Morris, 2017; Somerville et al., 2005; Velleman & Mervis, 2011). The two sections that follow review literature explicating the nature of symptoms having the potential to affect the developmental trajectory of articulation and review what has been found previously regarding the children’s development of articulation.

Williams Syndrome

WS is associated with cardiovascular disease (especially supra-aortic stenosis) and connective tissue abnormalities (both due to deletion of one copy of the *elastin* gene, resulting in elastin deficiency; Nickerson, Greenberg, Keating, McCaskill,

& Shaffer, 1995), endocrine abnormalities (e.g., 50% of girls have early puberty; Morris; 2017), characteristic facial gestalt (Morris, 2006), short stature, and delayed gross and fine motor milestones (Morris, 2017). Infants with WS often have difficulty gaining weight (likely due to feeding problems), oral texture aversion, gastroesophageal reflux, and constipation (Metcalf, 2012; Morris, 2017; Pagon, Bennett, LaVeck, Stewart, & Johnson, 1987). In infancy and early childhood, difficulty masticating food textures is related to problems managing food consistencies and textures (Morris, 2010, 2017). Low muscle tone is the most common central nervous system finding (Mervis & Morris, 2007). Hypotonia early in life can affect the development of standing posture (Harris, 2008) and impact development of the muscles and joints supporting the spine and extremities (Carboni, Pisani, Crescenzi, & Villani, 2002). These conditions have the potential to affect, in turn, the depth and control of respiration needed for physical activity and for learning to speak (Boehme, 1990). In combination with low tone, elastin deficiency contributes to chronic vocal hoarseness (Vaux, Wojtczak, Benirschke, & Jones, 2003). Sensory integration difficulties, including hypersensitivity to sound, are a problem for many children (John & Mervis, 2010). Farsightedness is an issue for about half of children with WS as is chronic otitis media. Older children and adolescents have an increased risk for sensorineural hearing loss (Marler, Elfenbein, Ryals, Urban, & Netzloff, 2005). Many struggle to regulate and inhibit personal emotions (Klein-Tasman, Lira, Li-Barber, Gallo, & Brei, 2015). Attention deficit hyperactivity disorder and anxiety are very common (especially specific phobias; Leyfer, Woodruff-Borden, Klein-Tasman, Fricke, & Mervis, 2006; Rhodes, Riby, Matthews, & Coghill, 2010; Woodruff-Borden, Kistler, Henderson, Crawford, & Mervis, 2010).

Intellectual abilities. WS is associated with mild to moderate intellectual disability, although the full range is from severe intellectual disability to average intellectual ability (e.g., Mervis & Pitts, 2015; see review in Martens, Wilson, & Reuters, 2008). The cognitive profile involves relative strengths in verbal and nonverbal reasoning and considerable weakness in visuospatial construction (Mervis & John, 2010). Receptive and expressive vocabulary abilities are typically in the borderline to low average range; the full range of abilities is from severe disability to high average ability (Mervis & John, 2010). Understanding of relational vocabulary typically is at the mild to moderate disability level, with the range from severe disability to average ability (Mervis & John, 2010). Grammatical understanding is typically at the borderline level, with the range from moderate-severe disability to high average ability (Mervis & John, 2010). Children with WS demonstrate relative strengths in phonological processing and verbal short-term memory (Mervis, 2009). For most children with WS, reading skills are stronger than mathematics skills (Mervis, 2009). Adaptive behavior skills are limited, with daily living skills typically at the mild to moderate disability level, with the range from severe disability to low average ability, and social interaction and communication skills typically at the borderline level, with the range from moderate disability to average ability (Mervis & Pitts, 2015).

Sociocommunicative characteristics. Behavioral descriptions of individuals with WS often include phrases such as socially engaging, gregarious, and sensitive to the feelings of others (Klein-Tasman & Mervis, 2003). At the same time, research suggests that many individuals with WS have problems maintaining relationships with peers (Järvinen-Pasley, Korenberg, & Bellugi, 2013; Laws & Bishop, 2004); the difficulty

likely stems from impaired social judgment and difficulty understanding complex social nuance (Gosch & Pankau, 1997; Thurman & Fisher, 2015). Many behavioral characteristics commonly shared among children with WS also are associated with autism spectrum (AS) symptomology (Klein-Tasman, Mervis, Lord, & Phillips, 2007; Klein-Tasman, van der Fluit, & Mervis, 2018), and the prevalence of ASD is greater than expected for children in the general population (Klein-Tasman et al., 2018; Leyfer et al., 2006; Lincoln, Searcy, Jones, & Lord, 2007).

Speech articulation. Delayed speech development was noted in the initial published reports describing WS (Beuren, 1962; Williams, Barratt-Boyes, & Lowe, 1961). To date, the development of speech articulatory accuracy for individuals with WS has been minimally described (but see Semel & Rosner, 2003; Udwin & Yule, 1990). Regarding the articulatory abilities of toddlers with WS, Velleman, Jones, Varley, Huffman, and Mervis (2013) reported on the relation between babble and words for eight toddlers with WS at ages 18 and 24 month. Compared with expectations for 14- to 18-month-old toddlers who were developing typically, toddlers with WS demonstrated a similar trajectory of babbling development that culminated in single-word articulations. Compared to performance at 18 months, productive gains in the phonetic repertoire were noted at 24 months with significant increases in the mean babble level in words, number of different vowels in the phonetic repertoire, and the percent of words structured with both a consonant and vowel (complexity of phonotactic shape).

Semel and Rosner (2003) reported anecdotal evidence suggesting that articulatory performance for school-aged children with WS ranged from fluent and intelligible to marginally intelligible depending on the circumstance. The authors indicated that most

children with WS produce intelligible speech and that articulatory load likely impacts speech clarity. Articulatory load involves complexity manifested in at least three ways: articulatory complexity (motoric challenge for greater or lesser engagement of muscle groups), prosodic complexity (motor timing challenge for variably executing place-to-place movement transitions), and sequencing complexity (memory and synthesis challenge for accurate organization of phoneme sequences). Kent (2000) suggested management of articulatory load is dependent on the degree to which neural systems regulate speech production (i.e., speech motor control).

Recent evidence has supported an impression of a relation between articulatory movement control and articulatory accuracy for young children with WS who have learned to speak. Huffman, Velleman, and Mervis (2012) assessed the speech of 31 children with WS aged 4–7 years using two speech measures: the *Hodson Assessment of Phonological Processes-3* (HAPP3; Hodson, 2004) and the *Verbal Motor Production Assessment for Children* (VMPAC; Hayden & Square, 1999). The HAPP-3 is a standardized measure for determining phoneme error patterns in children’s productive speech. The HAPP-3 ability score (AS; $Mean = 100, SD = 15$) measures speech accuracy in single words. The VMPAC is a standardized assessment of neuromotor integrity of the speech production system. The VMPAC Focal Oromotor Control subarea measures basic motor speech control, and the VMPAC Sequencing subarea measures simple speech-movement sequencing. Results indicated statistically significant, positive, and strong correlations among the measures of speech accuracy in single words (HAPP-3 AS), basic speech-motor control (VMPAC Focal Oromotor Control subarea), simple speech-

movement sequencing (VMPAC Sequencing subarea), and overall intellectual ability (DAS-II General Conceptual Ability score; similar to IQ).

Udwin and Yule (1990) examined speech behavior in 43 children with WS aged 6–16 years. Semi-structured, time-limited conversations were audio recorded. Of the participants, 84% had fluid and intelligible speech with occasional misarticulations of multisyllabic words and of words with complex coarticulations. The remaining 16% of participants did not use fluent speech.

Hargrove, Pittelko, Fillingane, Rustman, and Lund (2012) evaluated six speech skills in transcripts of spontaneous speech from 12 adolescents with WS and 12 adolescents who were typically developing and were matched for age (10–17 years) and sex. Adolescents with WS produced significantly fewer accurate whole words, attempted significantly fewer multisyllabic words, produced significantly fewer multi-syllabic words correctly, and had a significantly shorter modified phonological mean length of utterance. There were no significant differences between groups for intelligibility or PCC.

In summary, many toddlers with WS follow the same early trajectory of phonetic acquisition as toddlers in the general population although at a delayed pace. Toddlers develop a speech-sound repertoire, with regard to size, variety, and complexity, at a slower rate than children who are typically developing, likely due in part to hypotonia. Although school-aged children with WS evidence occasional episodes of false starts, pauses, and non-speech interjections, most of these children articulate sounds in words accurately except for words with complex speech sequences or with multiple syllables.

Phonological information processing. As discussed above, speech articulatory accuracy is positively related to phonological processing abilities for children who are developing typically. There is no information on this relation for children with WS. However, there have been two studies examining the performance of children with WS on standardized measures of phonological processing.

Mervis (2009) reported results for children with WS, aged 6–12 years, for the DAS-II Phonological Processing subtest ($Mean T = 50, SD = 10$). This measure includes rhyming, blending, elision, and phoneme identification and segmentation tasks. For the 55 children, $mean T = 40.24, SD = 13.28, Range = 10–62$, fourteen children (25%) earned T -scores at or above the mean for the general population.

Levy, Smith, and Tager-Flusberg (2003) administered three subtests of the *Comprehensive Test of Phonological Processing* (CTOPP; Wagner, Torgesen, & Rashotte, 1999; $Mean = 100, SD = 15$) to 20 adolescents with WS. Mean performance on three CTOPP subtests was in the borderline to low average range: Segmenting Words ($Mean = 81.50, SD = 11.01$), Segmenting Nonsense Words ($Mean = 77.00, SD = 11.17$), and Elision ($Mean = 70.50, SD = 12.97$). In contrast, the participants' intellectual ability as assessed by the *Kaufman Brief Intelligence Test* (K-BIT; Kaufman & Kaufman, 1990) was in the mild intellectual disability range ($Mean = 57.05, SD = 12.99$). The results of Levy et al. provide evidence that phonological processing is a relative strength for individuals with WS. Participants with KBIT Composite IQs above 70 ($n = 3$) performed in the low-average to average range on all phonological processing tasks. The participants with IQs in the range of 50–69 obtained higher percentile rankings for each of the three CTOPP subtests than for the KBIT Composite IQ.

7q11.23 Duplication Syndrome

Morris et al. (2015) reported on the many complex ways in which Dup7 affects multiple bodily functions, internal organs, endocrine function, and musculoskeletal structure and function. Participants included 53 individuals with Dup7, aged 1.25–21.25 years (*Mean* = 8.12 years, *SD* = 4.87); all children completed neurological examination and their parents completed in-depth interviews. Cardiovascular disease was common, with 46% evidencing aortic dilation. Mild craniofacial anomalies including facial asymmetry were very common; additionally, macrocephaly was present in 50%; micrognathia in 30%; and diastema in 30% (even though the craniofacial constellation included high-arched palate). Feeding issues were common among infants and toddlers; dysphagia persisted into childhood for some; and 7.5% required gastrostomy feeding. Most individuals showed behavioral signs of central nervous system involvement such as atypical standing posture and/or atypical walking patterns: hypotonia (60%); cranial nerve disturbance (e.g., hemifacial spasm, asymmetric smile, unintentional tongue rolling); atypical upper body movements (e.g., over-flow with intentional movement, tics, intention tremor, and synkinesia); and epilepsy or seizure disorder (20%). Of the 53 children and adolescents, 75% met the DSM-5 (American Psychiatric Association, 2013) criteria for Developmental Coordination Disorder. Recurrent otitis media requiring surgery for ventilation tubes was reported for 15% of the children, although hearing impairment was reported for just 5%. Some children experienced issues with eye misalignment. Anxiety disorders including Social Phobia and Selective Mutism were common, as was ADHD (Mervis et al., 2015).

Intellectual abilities. Mervis et al (2015) reported the intellectual abilities of very young children with Dup7 who completed the standardized *Mullen Scales of Early Learning* (MSEL; Mullen, 1995; *Mean* = 100, *SD* = 15). Overall ability was in the low average range of intellectual ability (*Mdn* = 80.88); MSEL Expressive language was significantly lower than either nonverbal reasoning or receptive language. Mervis (2018) reported results for 80 school-aged children with Dup7 who completed the DAS-II. The group's overall intellectual ability was in the low average range (*Mean* = 80.23), although the full range was from severe intellectual disability to superior intellectual ability. SSs for working memory and processing speed were in the low average range. Receptive and expressive vocabulary abilities were in the average range with the full ranges spanning severe disability to superior ability. Understanding of relational vocabulary was in the low average range, with the full range from severe disability to average ability (Mervis, 2018). A subgroup of the children with Dup7 in Mervis et al. (2015; *n* = 37) completed five subtests of the *Wechsler Individual Achievement Test-3* (WIAT-III; Wechsler, 2009). For most, reading skills were stronger than mathematics skills. Median SSs for the three reading subtests were in the average range. Median SSs for the two mathematics subtests were in the low average range. Performance on each of the five achievement measures was significantly strongly and positively correlated with overall intellectual ability.

For most children with Dup7, adaptive behavior skills were limited. Mervis et al. (2015) reported results of the *Scales of Independent Behavior-Revised* (*Mean* = 100, *SD* = 15; Bruininks et al., 1996). Broad Independence was mildly impaired with the range from severe adaptive impairment to average adaptive ability, daily living skills were at the mild to moderate disability level with the range from severe disability to low average

ability, and social interaction and communication skills were at the borderline level with the range from moderate disability to average ability.

In summary, there was a wide range of individual variability reported on measures of intellectual, vocabulary, academic and adaptive abilities among children with Dup7. Across all measures of the DAS-II, SSs for the majority of children were within the range of performance expected for children in the general population.

Sociocommunicative and psychological characteristics for Dup7. Early case report series of children with Dup7 suggested that the prevalence of ASD in children with Dup7 was significantly higher than in the general population. Berg et al. (2007), van der Aa et al. (2009), and Sanders et al. (2011) showed that Dup7 was a risk factor for ASD. More recently, Klein-Tasman and Mervis (2018) reported that the prevalence of ASD among children with Dup7 aged 4–17 years was 19%. This finding was based on gold-standard assessments for ASD.

Speech articulation. Since the first report of a child with Dup7 (Somerville et al., 2005), the most often cited developmental concern has been severe speech or expressive language difficulty (see also Berg et al., 2007; Merla et al., 2010; Mervis et al., 2015). Diagnostic classifications for speech and oral motor skills of toddlers with Dup7 were explored by Currier, Huffman, Velleman, and Mervis (2011). During administration of the *Communication and Symbolic Behavior Scales* (Whetherby & Prizant, 2002), 11 toddlers, aged 1 year 6 months–3 years 11 months were observed while eating a snack and playing in semi-structured interaction with an examiner. Verbal utterances were transcribed online for determining overall intelligibility, syllabification, babble and word shapes, phonetic repertoire, and word classes used. The speech of most was too limited

for full SSD diagnosis or for complete determination of phonological disorder. (For similar reasons, a comprehensive oral-mechanism exam was not applicable.) Speech symptoms indicated 64% of the children had mixed motor speech disorder characterized in part by oral apraxia, childhood apraxia of speech (CAS), and dysarthria. (*N.B.*, CAS is an SSD of significant severity and is defined as, “...*a neurological childhood SSD in which the precision and consistency of movements underlying speech are impaired in the absence of neuromuscular deficits...The core impairment in planning and/or programming spatiotemporal parameters of movement sequences results in errors in speech sound production and prosody.*” CAS Position Statement, ASHA, 2007.)

Huffman, Velleman, Morris, Osborne, and Mervis (2014) reported descriptive statistics for phoneme production accuracy on the HAPP-3 for 15 children with Dup7 aged 5–7 years. Children’s major phonological deviations were in the moderate disability range, *Median* HAPP-3 AS = 54.0 (lowest possible AS), *IQR* = 54.0–70.0. Only one child obtained a HAPP-3 AS within the typical expected range (e.g., 85–115) for children in the general population.

Parents’ ratings of their children’s speaking ability were reported in Velleman, Huffman, and Mervis (2013) for 27 children with Dup7 (*Mean* age = 8.9 years). The Speech subscale from the standardized *Children’s Communication Checklist-2* (CCC-2; Bishop, 2006; *Mean* scaled score = 10, *SD* = 3) was used. Overall, children obtained below average ratings (*Mean* scaled score = 4.88, *SD* = 2.69, *Range* = 1–8). Eleven of 26 children obtained CCC-2 Speech scaled scores within the range of average ability expected for the general population but all of these were below the general population mean. Nine children (33%) were rated below the 5th percentile suggesting moderate-to-

severe speech disability. Thirteen (48%) children were diagnosed with CAS based on five known differentially diagnostic features (see Strand, 2012). Not surprisingly, the group of children diagnosed with CAS scored significantly lower than the remaining children with Dup7 on the CCC-2 Speech scaled score. For the subgroup diagnosed with SSD-CAS, correlations between age and both intellectual ability and expressive vocabulary were significant but negative. This result indicated that for the children in the subgroup with SSD-CAS, the SSs on measures of intellectual and expressive vocabulary abilities decreased as age increased. This pattern was not found for the full sample of children with Dup7 or for the subgroup of children with SSD-phonological disorders.

Mervis et al. (2015) reported speech-diagnostic determinations for a cohort of 63 English-speaking children with Dup7, aged 4–17 years, using the criteria set forth in the DSM-5. The DSM-5 indicates that SSD is an appropriate diagnosis when speech-sound production is not what would be expected based on a child's age and developmental stage (APA, 2013). Fifty-one children (82.2%) met SSD criteria; most of these had symptoms of both phonological and mixed speech-motor symptoms (i.e., symptoms that fit both dysarthric and apraxic conditions), and 11 children (17.7%) had symptoms of SSD under challenging speaking conditions. One child was excluded from the study due to selective mutism. All participants in the study, except for the youngest two, were receiving or had received speech-language pathology services in the past. All of the 25 youngest participants (< 6.78 years) were diagnosed with SSD. In contrast, of the 12 oldest participants (>12.18 years), only 5 (41.7%) were diagnosed with SSD.

Phonological information processing. Children in the Mervis et al. (2015) study also completed the CTOPP-2 Nonword Repetition subtest (Wagner, Torgeson, &

Rashotte, 1999). Overall, children performed in the low average range (*Mean* scaled score = 5.38, *SD* = 2.09, *Range* = 2–9). Although 11 of the 26 children (42%) obtained CTOPP-2 nonword repetition scaled scores within the average range, all scores were below the general-population mean.

In summary, the large majority of children with Dup7, aged 4–17 years, met DSM-5 criteria for SSD diagnosis. More than half of the children aged 5–8 years were diagnosed with severe SSD. Evidence was presented of mixed speech symptomatology consistent with CAS, dysarthria, and phonological deficiency (Velleman et al., 2013). Mixed symptomatology suggested a combination of limiting factors based on disorders of speech motor control, errors of speech timing and sequencing, and inaccurate or incomplete application of phonological patterns.

Summary

The present review explicated the theoretical framework for this project's two empirical studies. It presented support from the literature for the evaluative approach using citation assessment and for relating articulatory accuracy to children's phonological processing skill. The standardized GFTA-2 was shown to be a reasonable and valid tool for examining speech sound production accuracy for children with WS or Dup7. The review also provided detailed rationale in support of the method chosen for exploratory examination of GFTA-2 items by means of independently grouping the consonant items according to five features of consonants previously shown to be important for the development of articulatory accuracy.

For children who are developing typically, articulatory accuracy was shown to be an important contributor to the early development of phonological processing and it was

shown to be related moderately to overall intellectual ability. Evidence was reviewed indicating that (a) implicit knowledge of phonetics is fundamental to developing phonological systematicity, (b) the expressive development of early lexical items is constrained by the speech articulations which execute them, and (c) especially within relevant contexts, phonological information processing ability contributes significantly to word learning and literacy development at any age (Liberman, Shankweiler, Fischer, & Carter, 1974; Maye, Werker, & Gerken, 2002; National Reading Panel, 2000; Werker & Tees, 1984). The final two subsections reviewed briefly what is known about the neurodevelopment of children with WS or Dup 7 that potentially impacts the development of speech articulation; the sections also addressed what is known regarding articulatory development for children with these syndromes.

The Present Project

A schematic of the dissertation's two studies is presented in Figure 1. As indicated in the figure, articulatory accuracy is examined in Study 1 for the two groups: children with WS and children with Dup7. Further, the graphic shows that in Study 2, correlations among the standardized variables of articulatory accuracy, phonological information processing, vocabulary, and intellectual abilities are examined for the children in each syndrome group. In addition, for children with WS, articulatory accuracy SS was evaluated to determine its potential for making a unique contribution to the explained variance in phonological processing SS beyond the contributions of vocabulary SS, nonverbal reasoning SS, spatial SS, and verbal short-term memory SS.

Predicted Findings: Study 1

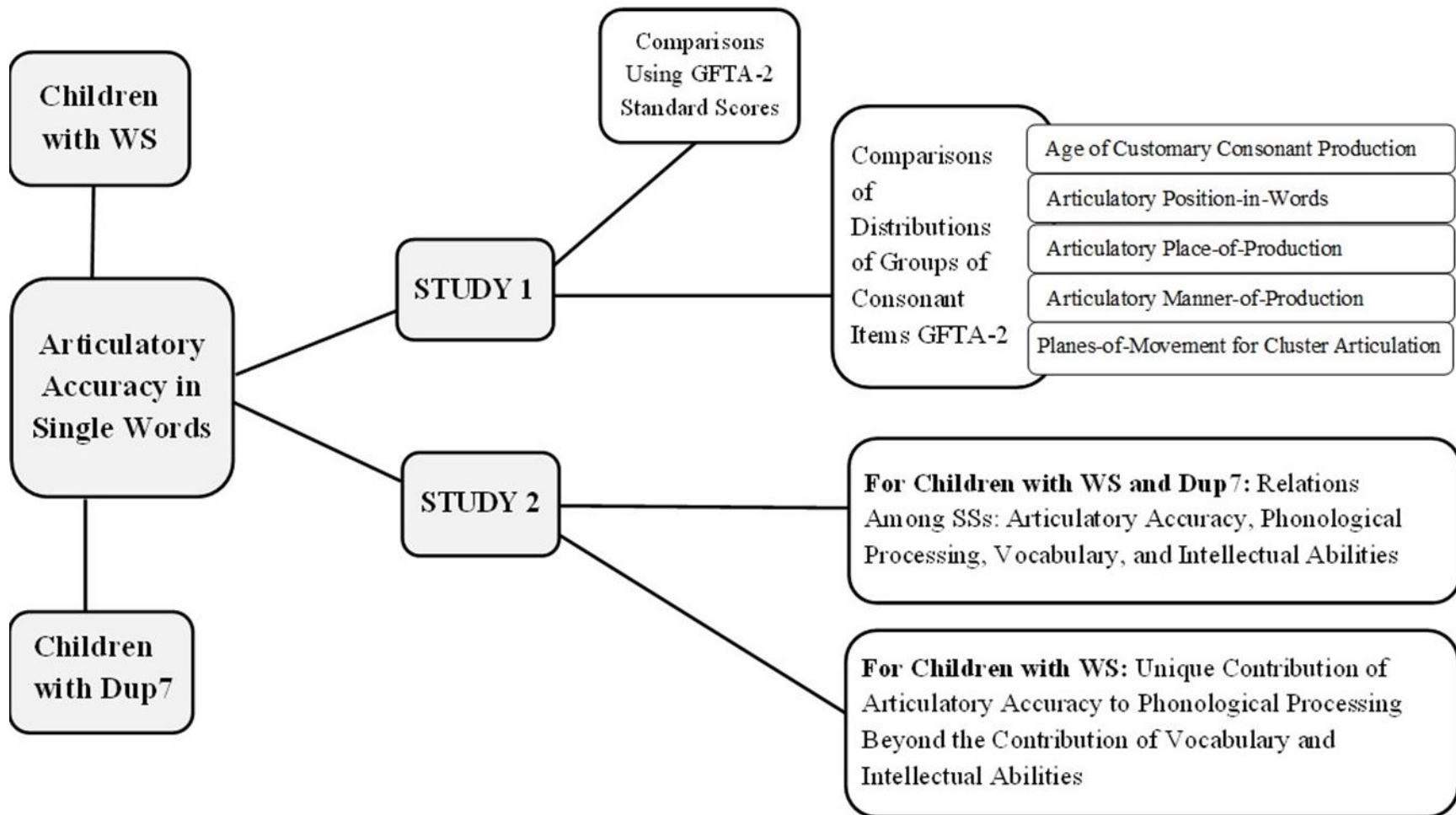


Figure 1. Speech Articulatory Accuracy in Children with Williams Syndrome and Children with 7q11.23 Duplication Syndrome

The predicted findings for Study 1 are as follows. I expected that both children with WS and children with Dup7 would articulate consonants with significantly less accuracy than same-aged children in the general population. Also, I expected that the group of children with WS would show significantly greater accuracy for articulating consonants than the group with Dup7.

Further, I expected children with WS in the older subgroup to obtain significantly higher GFTA-2 SSs than the children in the younger subgroup. I expected also significantly higher GFTA-2 SSs for older children with Dup7 compared with the younger children with Dup7. For both children with WS or Dup7, I expected that SSs for articulatory accuracy would correlate significantly and positively with SSs for general intellectual ability.

Based on Winitz (1969, p. 143), I expected that children with IQs ≥ 70 would have significantly higher GFTA-2 SSs than children with IQs < 70 . This prediction was expected to hold for both children with WS and children with Dup7.

In addition, I expected that tests computed for patterns of differences across sets of consonants arranged for ACCP (see Table 1) would show a profile of acquisition that is similar to the profile for English-speaking children in the general population. Specifically, for each test and for both groups of children, I expected the overall effect to be significant and I expected that post hoc analyses would indicate that the proportion of accurately produced early-acquired consonants would be significantly higher than the proportion for middle-acquired consonants, which, in turn, would be significantly higher than the proportion for late-acquired consonants.

I expected that the outcome of the overall test for differences across distributions of proportion correct for sets of consonants arranged for position-in-words would be significant for both groups of children. Based on the literature for younger children in the general population, I expected that for children in both younger subgroups, the proportion of accurately produced Initial Consonants would be significantly higher than the proportion for Final Consonants.

Based on the literature reviewed, I expected to find statistically significant overall effects for articulatory place and articulatory manner features of articulation both for the children with WS and the children with Dup7. Specifically, I expected to find results consistent with the literature for younger children who are developing typically: (a) the proportion of bilabial and velar consonants would be articulated with significantly higher accuracy than the proportion of consonants articulated in the central oral (especially palatal) areas, and (b) the proportion of nasal and stop consonants would be articulated with significantly greater accuracy than that for fricative and approximant consonants.

I expected that the groups of participants would produce double consonants (consonant clusters and affricates) based on ACCP (see Table 1) and that tests for differences in distributions of proportion correct based on directional planes-of-movement would result in no significant differences across the directional planes-of-movement.

Predicted Findings: Study 2

I expected correlations among all standardized variables in Study 2 to be statistically significant and positive both for children with WS and for children with Dup7.

Furthermore, I expected that for the children with WS, GFTA-2 SS would explain significant and unique variance in DAS-II Phonological Processing SS over and above the unique and statistically significant contributions made by DAS-II Recall of Digits Forward SS, DAS-II Spatial Ability SS, Composite Vocabulary SS, and DAS-II Nonverbal Reasoning SS. (This model could not be tested for the children with Dup7 because the sample size of these children was too small.)

CHAPTER II
ARTICULATORY ACCURACY DETERMINED
USING A CITATION METHOD OF MEASUREMENT

The principal objective of the present dissertation was to evaluate articulatory accuracy for children with WS and Dup7, aged 4 to 17 years. I have used the standardized GFTA-2 to address this goal by considering articulatory accuracy for consonants in single words cited on cue. Literature reviewed in Chapter I showed that for children in the general population, single-word articulation is typically inaccurate or inconsistent early in development but improves over time (Morley, 1965; Templin, 1957; Winitz, 1969). Second, it showed that most children who are developing typically have achieved articulatory accuracy before they turn nine years old (Goldman & Fristoe, 2000; Smit et al., 1990; Templin, 1957). Third, it showed that articulatory accuracy positively correlates with intellectual ability for children who are developing typically (Overby et al., 2012; Templin, 1957; Winitz, 1969). And fourth, it showed that children with deletion or duplication of the WS region are expected to develop articulatory accuracy more slowly than children who are developing typically (Mervis & Becerra, 2007; Mervis et al., 2015; Semel & Rosner, 2003; Udwin & Yule, 1994; Velleman & Mervis, 2011). Study 1 is described in the present chapter and is designed to examine these issues for children with WS or Dup7.

All participants completed the standardized GFTA-2. Results were analyzed in several ways: (a) in relation to typical expectations for articulatory accuracy, (b) for differences in articulatory accuracy between children with WS and children with Dup7, (c) in relation to overall intellectual ability, (d) with regard to ACCP for children in the general population, and (e) for four particular features of articulation shown previously important for the development of articulation. The articulatory feature analyses included tests for differences in proportion correct across sets of GFTA-2 consonants arranged for: consonant accuracy at syllable margins (i.e., initial or final word positions; Oller, 2000; Stoel-Gammon, 1998; Vihman, 1996), articulatory place-of-production (IPA, 1999; Kent, 2013), articulatory manner of vocal tract modification (Grunwell, 1981; IPA, 1999), and double consonant (cluster and affricate) articulation across planes-of-movement (McLeod, van Dorn, & Reed, 2001).

Method

Participants

The final sample for Study 1 included 118 children with WS (57 girls, 62 boys), aged 4.01–17.98 years (*Median* = 7.17 years, *IQR* = 4.55–11.17) and 50 children with Dup7 (22 girls, 28 boys) aged 4.01–17.70 years (*Median* = 9.32 years, *IQR* = 6.14–12.26). All children were participants in studies of cognitive, linguistic, and behavioral development conducted by Dr. Carolyn Mervis (Principal Investigator of the Neurodevelopmental Sciences Lab; NSL) at the University of Louisville. Authorization for the studies was granted by the University of Louisville Institutional Review Board. All children were monolingual English speakers. No child had any additional genetic diagnosis. All participants were receiving speech-language intervention services at the

time of the study and/or had had speech-language services in the past, including goals focusing specifically on speech production.

Sociodemographics. Parents with WS were asked to provide three types of demographic information: the state or country in which they resided, the highest level of education completed by the child's mother, and the child's race and ethnicity. For the 118 children with WS, the distribution of the participants' places of residence with regard to US Census regional divisions was as follows: 3 children (2.5%) were from Northeastern New England states, 26 children (22.0%) were from Northeastern Middle Atlantic states, 32 children (27.1%) were from Southern Atlantic states, 9 children (7.6%) were from Southern East South-Central states, 4 children (3.4%) were from Southern West South-Central states, 17 children (14.4%) were from Midwestern East North-Central states, 12 children (10.2%) were from Midwestern West North-Central states, 4 children (3.4%) were from Western Mountain states, and 9 children (7.6%) were from Pacific states. Two children (1.7%) were from Canada. Childrens' mothers' educational attainment was as follows: 28 mothers (23.7%) did not have a bachelor's degree and 90 mothers (76.3%) attained a bachelor's degree or higher. Childrens' reported racial and ethnic affiliation was as follows: 94 children (79.7%) were white, non-Hispanic; 9 children (7.6%) were white, Hispanic; 1 child (0.8%) was Asian, non-Hispanic; 3 children (2.5%) were African American, non-Hispanic; 10 children (8.5%) were biracial or triracial, non-Hispanic; and 1 child (0.8%) was biracial or triracial, Hispanic.

For the 50 children with Dup7, the distribution of the participants' places of residence with regard to US Census regional divisions was as follows: 4 children (8%) were from Northeastern New England states, 7 children (14.0%) were from Northeastern

Middle Atlantic states, 13 children (26.0%) were from Southern Atlantic states, 5 children (10.0%) were from Southern East South-Central states, 2 children (4.0%) were from Southern West South-Central states, 5 children (10.0%) were from Midwestern East North-Central states, 5 children (10.0%) were from Midwestern West North-Central states, 2 children (4.0%) were from Western Mountain states, and 3 children (6.0%) were from Pacific states. Four children (8.0%) were from English-speaking countries outside of the US (3 from Canada, 1 from the United Kingdom). Childrens' mothers' educational attainment was as follows: 27 mothers (54.0%) did not have a bachelor's degree and 23 mothers (46.0%) attained a bachelor's degree or higher. Childrens' reported racial and ethnic affiliation was as follows: 40 children (80%) were white, non-Hispanic; 3 children (6%) were white, Hispanic; 2 children (4%) were Asian, non-Hispanic; 2 children (4%) were African American, non-Hispanic; 2 children (4%) were biracial or triracial, non-Hispanic; and 1 child (2%) was biracial or triracial, Hispanic.

Inclusion and exclusion criteria. Participant inclusion criteria were the following: (a) genetically-confirmed classic-length deletion or duplication of the WS region; (b) no comorbid genetically-confirmed diagnosis; (c) chronological age within the range of 4–17 years; (d) typical (or corrected to typical) visual acuity and typical hearing acuity; and (e) exposure exclusively to the English language in the home environment.

Some children included in the present study were assessed multiple times as part of a longitudinal study. For these children, the data from the first usable GFTA-2 assessment were used. For six children with WS, the initial GFTA-2 administration was not used because the measure's administration criteria were not met. Specifically, the children did not have enough expressive vocabulary for valid determination of

articulatory skill. In these instances, the second GFTA-2 assessment was used. For all children with Dup7, the initial GFTA-2 assessment was used.

Nine additional children with WS were excluded from the final sample. Five were excluded because two or more languages were spoken in the home, one was excluded due to hearing impairment, and three were excluded because they had not achieved enough language to meet GFTA-2 administration criteria. Seven additional children with Dup7 were excluded from the final sample. One was excluded because of selective mutism, one was excluded because of a tongue laceration, and five were excluded because they did not have enough language to meet GFTA-2 administration criteria. All children included were physically healthy based on caregiver report and a brief screening of social affect and physical abilities that I conducted prior to the time of articulatory assessment.

Measures

Articulatory accuracy. The standardized GFTA-2 (Goldman & Fristoe, 2000) was used to measure speech articulatory accuracy in single words. The GFTA-2 provides raw scores based on production errors and SSs for accuracy articulating the 77 GFTA-2 consonant items. This articulation assessment was standardized on a representative sample of 2,350 children aged 2.00–21.99 years who resided across the four major geographic regions of the US. Participants in the standardization sample were from diverse ethnic and economic backgrounds. Children with special educational needs were included in numbers proportional to US population statistics (US Census Bureau, 1998).

The research has shown that speech articulatory skill typically approaches the stability of mature performance with increasing age (Templin, 1957). Expectedly, children in the GFTA-2 normative sample produced a distribution of scores that was

skewed in this direction. Older children articulated all 77 consonants accurately or made only a small number of errors. Only in the youngest age intervals did raw-score distributions approach a normal distribution. Because the GFTA-2 was used primarily for determining the need for speech therapy and/or for determining appropriate treatment goals, Goldman and Fristoe (2000) solved the problem of developing SSs ($Mean = 100$, $SD = 15$) based on nonparametric data by transforming the raw data using Johnson curves (Hill, Hill, & Holder, 1976; Johnson, 1949). Johnson curves were chosen because they preserved the skewness and kurtosis of the raw data. Essentially, the solution involved stratifying the raw data according to 16 age intervals and then estimating percentiles for each stratum. SSs were derived from the percentiles. So, by design, the relation between the published percentile ranks and the linear SSs varies for each GFTA-2 age interval.

Goldman and Fristoe (2000) selected the words containing consonant items assessed using the GFTA-2 based on common usage in childhood, consistency of recognition when portrayed in pictures, and consistency of targets produced in prevocalic-, postvocalic-, and intravocalic-word positions. The GFTA-2 is structured to assess accuracy for 77 items. These include: 23 singleton consonants (55 items), two affricates (six items), and 16 prevocalic consonant clusters. The consonant items occur in 53 single words. The single words consist of names of objects, activities, and descriptors familiar to young children (nouns, verbs, adjectives). The protocol requires response elicitation using 34 colored-picture plates depicting the target words. Spontaneous naming of the pictures is encouraged, but sentence completion or direct prompting for imitation is permitted. The GFTA-2 is a relatively balanced measure in terms of stimulus-item phonotactics. The aspects of balanced complexity include the following: 16 of the

53 stimulus words (30%) begin with an initial consonant cluster, 27 of the 53 stimulus word are monosyllabic, and 26 words are multisyllabic. Of the 26 multisyllabic words, 23 are bisyllabic and 3 are trisyllabic. The phonotactic patterns of the multisyllabic words have dissimilar shapes across the medial transitions.

According to the test manual (Goldman & Fristoe, 2000), the GFTA-2's internal median alpha reliability for females was .96 and for males it was .94. Overall standard error of measurement (*SEM*) was 3.0 for females and 3.7 for males. (*SEM* decreased with increasing chronological age.) Median values of test-retest reliability (measuring consistency identifying error sounds across positions-in-words) were 98% for the initial position, 98% for the medial position, and 98% for the final position. Overall median interrater reliabilities (IRR) across all sounds as a function of word position were 93% initially, 90% medially, and 90% finally. Isolated phoneme IRR exceeded 83% for all but three of the 77 items (/s/ medial, /ɪ/ final, and /tɪ/ initial). Content validity included detailed construct definitions and descriptions of phoneme development; additional content support was discussed in Eisenberg and Hitchcock (2010). Five other articulation measures have reported good concurrent validity with the GFTA-2 (see Flipsen & Ogiela, 2015). In the 15-year span of the second edition's use, the GFTA-2 was arguably the most commonly used measure of speech-sound articulation across the US and Canada (Skahan & Watson, 2007). It was used routinely for special-service eligibility consideration.

Intellectual ability. Overall intellectual ability was measured using the DAS-II General Conceptual Ability composite (GCA; similar to IQ). The DAS-II was normed for children aged 2.50–17.99 years (Elliott, 2007). In this dissertation, children aged 4.00–

8.99 years completed the Early Years battery; children aged 9.00–17.99 years completed the School-Age battery. Both levels of the DAS-II provide SSs (*Mean* = 100, *SD* = 15) for the GCA, which is based on performance on the Verbal, Nonverbal Reasoning, and Spatial core subtests. The GCA indicates the capacity for children to perform complex mental processing involving conceptualization and transformation of information (Elliott, 2007).

The psychometric properties of the DAS-II are very good to excellent. The standardization sample included 2,775 children demographically representative of the US (US Census Bureau, 2004) and included children with mild intellectual disabilities. Internal consistency of the subtests is good (see Keith, Low, Reynolds, Patel & Ridley, 2010). Test-retest reliabilities of the composite scores are excellent (.91–.98; determined using the split-half method and corrected using the Spearman-Brown formula) and adequate to excellent for the subtest scores (.80–.98). Internal consistency measures (split-half method) range from adequate to good (Elliott, 2007).

Coding

All individual GFTA-2 assessments were coded after the assessment session in a quiet room free of distraction. The audio-video records were replayed for coding using a Dell Inspiron 5759 laptop computer (Intel (R) Core i5-6200U, 2.3 GHz, 8 GB, 64-bit) and SteelSeries Siberia 350 Over-Ear Headset. Scoring followed the GFTA-2 Level 2 scoring rules described in the examiner's manual. Coding decisions for questionable articulations were made based on the rules provided in Appendix C. Diacritics were applied to transcribed segments as necessary using narrow phonetic transcription techniques (Ball & Rahilly, 1996, 2002; Duckworth, Allen, Hardcastle, & Ball, 1990;

Powell, 2001; Stoel-Gammon, 2001). The presence of articulation errors was indicated by marking the appropriate space on the GFTA-2 protocol by type of error: substitution, omission, distortion, or addition. Response forms were scored in the standard manner with all data entered in Excel spreadsheets by the present author. Accuracy of entered items was verified by having a lab transcriptionist independently re-enter all data on a separate worksheet. Errors were identified automatically using a third worksheet to subtract the second sheet from the first and then corrected.

Consonant-group proportion correct. As described in Chapter I, articulatory accuracy was examined for the 77 GFTA-2 consonant items by arranging the consonants in particular groups based on Age of Customary Consonant Production and on four features previously shown to be important for the development of articulatory accuracy: Articulatory Position-In-Words, Articulatory Place-of-Production, Articulatory Manner-of-Production, and Planes-of-Movement for Double Consonant Articulation. The consonant groups were divided further into constituent subgroups for the analyses. The following sections delineate each GFTA-2 consonant-group's organization.

Age of Customary Consonant Production. Age of Customary Consonant Production (ACCP) refers to three sets of GFTA-2 items arranged based on relative timing of acquisition by children in the general population: Early-developing, Middle-developing, and Late-developing consonants. This ordering is likely constrained by physical maturation and experience talking (ASHA, 2018). As shown in Table 4, all 77 GFTA-2 items are included in the ACCP analyses.

Table 4

Consonant Groups for Age of Customary Consonant Production

Early-Developing Consonants (30 items)

1. Singleton consonants (29 items)
 - a) Initial position (11 items): /b, p, w, n, m, t, d, k, g, h, f/
 - b) Medial position (9 items): /b, p, n, m, t, d, k, g, f/
 - c) Final position (9 items): /b, p, n, m, t, d, k, g, f/
2. Initial double consonant (1 item): /kw/

Middle-Developing Consonants (21 items)

1. Singleton consonants (10 items)
 - a) Initial position (4 items): /j, v, s, ʃ/
 - b) Medial position (3 items): /v, s, ʃ/
 - c) Final position (3 items): /v, s, ʃ/
2. Affricates (6 items)
 - a) Initial position (2 items): /tʃ, dʒ/
 - b) Medial position (2 items): /tʃ, dʒ/
 - c) Final position (2 items): /tʃ, dʒ/
3. Initial consonant clusters (5 items): /sp, st, kl, gl, fl/

Late-Developing Consonants (26 items)

1. Singleton consonants (16 items)
 - a) Initial position (5 items): /z, θ, ð, l, ɹ/
 - b) Medial position (6 items): /ŋ, z, θ, ð, l, ɹ/
 - c) Final position (5 items): /ŋ, z, θ, l, ɹ/
2. Initial consonant clusters (10 items): /sw, sl, bl, pl, bɹ, dɹ, fɹ, gɹ, kɹ, tɹ/

Table 5

Consonant Groups for Articulatory Position-in-Words

Initial Consonants (20 items)
/b, p, w, n, m, t, d, k, g, h, f, j, ʃ, ð, θ, ɹ, l, v, s, z/.

Final Consonants (17 items)
/b, p, n, m, t, d, k, g, f, ʃ, ð, θ, ɹ, l, v, s, z/.

Articulatory Position-In-Words. Articulatory position-in-words refers to the position of the target consonant as it occurs at the margins of syllables: Initial-position and Final-position consonants. As shown in Table 5, 37 GFTA-2 singleton consonants were included. GFTA-2 medial consonants or double consonants (clusters or affricate items) were not included in this consonant-group arrangement.

Articulatory Place of Production. Articulatory Place-of-Production refers to five consonant-groups with items articulated at salient points within the vocal tract:

Table 6

Consonant Groups for Articulatory Place-of-Production

| | |
|---|--|
| Bilabial Consonants (10 consonant items) | |
| a) | Initial position (4 items): /p, m, w, b/ |
| b) | Medial position (3 items): /p, m, b/ |
| c) | Final position (3 items): /p, m, b/ |
| Dental Consonants (11 consonant items) | |
| a) | Initial position (4 items): /f, v, θ, ð/ |
| b) | Medial position (4 items): /f, v, θ, ð/ |
| c) | Final position (3 items): /f, v, θ/ |
| Alveolar Consonants (18 consonant items) | |
| a) | Initial position (6 items): /n, d, t, s, z, l/ |
| b) | Medial position (6 items): /n, d, t, s, z, l/ |
| c) | Final position (6 items): /n, d, t, s, z, l/ |
| Postalveolar-Palatal Consonants (7 consonant items) | |
| a) | Initial position (3 items): /j, ʃ, ɹ/ |
| b) | Medial position (2 items): /ʃ, ɹ/ |
| c) | Final position (2 items): /ʃ, ɹ/ |
| Velar-Glottal Consonants (9 consonant items) | |
| a) | Initial position (3 items): /h, g, k/ |
| b) | Medial position (3 items): /g, k, ŋ/ |
| c) | Final position (3 items): /g, k, ŋ/ |

Table 7

Consonant Groups for Articulatory Manner-of-Production

| | |
|----------------------------------|--|
| Nasal Consonants (8 items) | |
| a) | Initial position (2 items): /m, n/ |
| b) | Medial position (3 items): /m, n, ŋ/ |
| c) | Final position (3 items): /m, n, ŋ/ |
| Stop Consonants (18 items) | |
| a) | Initial position (6 items): /p, b, t, d, k, g/ |
| b) | Medial position (6 items): /p, b, t, d, k, g/ |
| c) | Final position (6 items): /p, b, t, d, k, g/ |
| Fricative Consonants (21 items) | |
| a) | Initial position (8 items): /f, v, θ, ð, ʃ, s, z, h/ |
| b) | Medial position (7 items): /f, v, θ, ð, ʃ, s, z/ |
| c) | Final position (6 items): /f, v, θ, ʃ, s, z/ |
| Approximant Consonants (8 items) | |
| a) | Initial position (4 items): /w, j, l, ɹ/ |
| b) | Medial position (2 items): /l, ɹ/ |
| c) | Final position (2 items): /l, ɹ/ |

Bilabial, Dental, Alveolar, Postalveolar-Palatal, and Velar-Glottal, as shown in Table 6.

Fifty-five GFTA-2 singleton items (no double consonants) were included.

Articulatory Manner-Of-Production. Articulatory manner-of-production refers to the ways in which the breath stream is modified when articulating. In this feature group,

the GFTA-2 items are arranged in four consonant groups: *Nasal*, *Stop*, *Fricative*, and *Approximant*. As shown in Table 7, 55 GFTA-2 singleton consonants were included.

Articulatory Planes-of-Movement for Double Consonant Articulation. The articulatory planes-of-movement arrangement refers to the direction of transition between the two consonants in a cluster or the two consonant components in an affricate (Hayden & Square, 1999). The groups are: Back-to-Front, Front-to-Back, and Same Place. As shown in Table 8, the 22 GFTA-2 double consonant items were included.

Table 8
Double Consonants for Articulatory Planes-of-Movement

| |
|--|
| Group 1: Back-to-Front (7 items) |
| Initial position: /kw, kl, kɪ, gl, gɪ, sp, sw/ |
| Group 2: Front-to-Back (7 items): |
| Initial position: /bl, pl, fl, bɪ, dɪ, fɪ, tɪ/ |
| Group 3: Same Place (8 items) |
| Initial position: /st, sl, tʃ, dʒ/ |
| Medial position: /tʃ, dʒ/ |
| Final position: /tʃ, dʒ/ |

Procedure

Study 1 was community-based, empirical, and cross-sectional. All participants visited the NSL for the purpose of completing a battery of cognitive, language, psychosocial, academic, and speech-motor assessments. These measures were administered in accordance with instructions published in the respective assessments' manuals. All children completed the full assessment battery within three days except for one child who began intellectual assessment prior to the 2-week university winter break and completed speech assessment on the first day following the break.

DAS-II GCA data were obtained from the NSL database. All GFTA-2 assessments were audiovideo recorded for coding later using a picture-in-a picture video format. The larger picture provided a close-up of the child's face. The smaller picture

provided a close-up of the GFTA-2 stimulus picture. Audio was captured in stereo at 44,000 kHz. Raw footage was formatted and compressed at 1340 kbps using Pinnacle Systems software (Studio HD, Version 14, Avid Technology, Inc., 2009; www.pinnaclesys.com).

Reliability. All of the GFTA-2 assessments were coded by the dissertation author. To compute reliability, two other well-trained coders independently coded 52 randomly chosen records from children with WS (44% of 118) and 15 randomly chosen records from children with Dup7 (30% of 50). Second judges followed the coding procedures outlined in Appendix C. Practice coding was completed prior to coding the reliability sample using 12 GFTA-2 records not included in the present dissertation.

Reliability statistics for GFTA-2 SSs were good. For scores from children with WS, GFTA-2 second-judge SSs fell within the 95% confidence interval of the dissertation author's SSs 91% of the time. For scores from children with Dup7, GFTA-2 second-judge SSs fell within the 95% confidence interval of the dissertation author's scores 100% of the time.

As a further check on reliability, the second-judge's reliability sample of GFTA-2 consonant items was tested for differences across distributions of sets of consonants arranged in five separate ways shown important for the development of articulation. After these tests, the same five were repeated but instead using the dissertation author's reliability-sample. Outcomes of the second judge's sets of analyses were compared directly to those of the dissertation author. The overall effects were identical for each of the five comparisons.

Data Analysis

Statistical exploration of the distributions of the GFTA-2 SSs obtained from children with WS and from children with Dup7 revealed violations of the parametric assumption of normality. Therefore, statistical analyses of the GFTA-2 SSs, DAS-II GCA, and proportion correct for distributions of sets of GFTA-2 items arranged according to features of articulation were computed using the appropriate nonparametric tests in IBM SPSS 25.

Results

In the first section of the Results, I report the outcomes of analyses using the GFTA-2 SSs. Findings for the performance of the children with WS and the children with Dup7 were first compared to the level of expected performance for children in the general population. The performance of the children with WS was then compared to the performance of the children with Dup7. Third, I computed the correlation between each group's articulatory accuracy scores and their overall intellectual ability scores. In later sections, I considered whether the pattern of ACCP was the same as has previously been found for children in the general population. Finally, I tested for differences in distributions of sets of consonant-groups arranged according to the four features previously shown important for developing articulatory accuracy. For each of these analyses, I first considered the entire sample of children within a group, and then separately, I considered the performances of the younger children (aged 4.00–9.99 years) and of the older children (aged 10.00–17.99 years).

Articulatory Accuracy: GFTA-2 SSs

GFTA-2 SSs ranging from 85–115 are considered to be in the expected (typical) range. In the present study, 52 children with WS (44% of 118) achieved GFTA-2 SSs

within the expected range including 33 children in the younger subgroup (39% of 84) and 19 children in the older subgroup (56% of 34). Twenty-five children with WS (21%) made four or fewer errors out of the 77 GFTA-2 items and seven (6%) obtained the lowest possible SS (< 40). Ten children with Dup7 (20% of 50) achieved GFTA-2 SSs within the expected range, including two in the younger subgroup (7% of 29) and eight in the older subgroup (38% of 21). Seven children with Dup7 (14%) made four or fewer errors and seven (14%) obtained the lowest possible SS.

To determine if articulatory accuracy for the participant groups differed significantly from expectations for children in the general population, the median GFTA-2 SS for each participant group was compared to the median GFTA-2 SS for children in the general population (100) using one-sample Wilcoxon Signed-Rank tests. Separate analyses were computed also for younger and older children. As indicated in Table 9, all of the median GFTA-2 SSs were significantly less than 100, $p < .001$, two-tailed tests.

Descriptive statistics and test results for between-group differences for GFTA2 SSs and DAS-II GCA SSs are presented in Table 10. As indicated in the table, the distribution of GFTA-2 SSs was significantly higher for the full sample of children with WS ($IQR = 28.25$) than for the full sample of children with Dup7 ($IQR = 42.45$). Comparison of the spread of SSs (IQR s) indicated articulatory accuracy for the group of children with WS was less variable than for the group with Dup7. This is particularly noteworthy given that the distribution of DAS-II GCA SSs was significantly higher for the children with Dup7 ($IQR = 16.50$) than for the children with WS ($IQR = 15.00$). The same pattern of findings was found for the younger subgroups. For the older subgroups, the distribution of DAS-II GCA SSs was significantly higher for the children with Dup7

Table 9

Study 1: Wilcoxon One-Sample Signed-Rank Tests Based on GFTA-2 SS: Children with WS or Children with Dup7

| Group | <i>n</i> | Obtained Median: GFTA-2 SS | GFTA-2 Median = 100 | | | Group | <i>n</i> | Obtained Median: GFTA-2 SS | GFTA-2 Median = 100 | | |
|--------------|----------|----------------------------------|---------------------|----------|----------|----------------|----------|----------------------------------|---------------------|----------|----------|
| | | | <i>z</i> | <i>p</i> | <i>r</i> | | | | <i>z</i> | <i>p</i> | <i>r</i> |
| WS – All | 118 | 82.00 | 9.43 | <.001 | -0.60 | Dup7 - All | 50 | 68.00 | 6.16 | <.001 | -0.61 |
| WS - Younger | 50 | 80.50 | 7.96 | <.001 | -0.78 | Dup7 - Younger | 29 | 54.00 | 4.71 | <.001 | -0.62 |
| WS - Older | 34 | 88.50 | 5.09 | <.001 | -0.60 | Dup7 - Older | 21 | 80.00 | 4.03 | <.001 | -0.59 |

Note. Younger = 4.00–9.99 years; Older = 10.00–17.99 years. Dup7 = 7q11.23 duplication syndrome; GFTA2 = *Goldman-Fristoe Test of Articulation* (2nd ed., Goldman & Fristoe, 2000); WS = Williams syndrome.

Table 10

Study 1 Descriptive Statistics and Comparisons for GFTA-2 SS and DAS-II GCA: Children with WS or Dup 7

| Group | <i>n</i> | GFTA-2 SS | | Mann-Whitney U | | | DAS-II GCA | | Mann-Whitney U | | |
|----------------|----------|------------|---------------|----------------|----------|----------|------------|---------------|----------------|----------|----------|
| | | <i>Mdn</i> | <i>IQR</i> | <i>z</i> | <i>p</i> | <i>r</i> | <i>Mdn</i> | <i>IQR</i> | <i>z</i> | <i>p</i> | <i>r</i> |
| WS - All | 118 | 82.00 | 63.75 – 92.00 | 3.06 | .002 | 0.24 | 66.00 | 58.00 – 73.00 | -6.40 | <.001 | -0.42 |
| Dup7 - All | 50 | 68.00 | 39.00 – 81.25 | | | | 83.00 | 74.75 – 91.25 | | | |
| WS - Younger | 84 | 80.50 | 63.25 – 89.00 | 3.71 | <.001 | 0.35 | 66.00 | 54.25 – 73.00 | -5.58 | <.001 | -0.52 |
| Dup7 - Younger | 29 | 54.00 | 42.00 – 75.50 | | | | 85.00 | 78.50 – 92.50 | | | |
| WS - Older | 34 | 88.50 | 64.25 – 95.25 | 0.76 | .445 | 0.10 | 65.00 | 59.00 – 73.00 | -3.22 | .001 | -0.50 |
| Dup7 - Older | 21 | 80.00 | 39.00 – 95.50 | | | | 82.00 | 69.50 – 89.50 | | | |

Note. Younger = 4.00–9.99 years; older = 10.00–17.99 years. DAS II = *Differential Ability Scales* (2nd ed., Elliott, 2007); Dup7 = 7q11.23 duplication syndrome; GCA = General Conceptual Ability (similar to IQ); GFTA2 = *Goldman-Fristoe Test of Articulation* (2nd ed., Goldman & Fristoe, 2000); *IQR* = interquartile range; *Mdn* = median; SS = standard score; WS = Williams syndrome.

than for the children with WS. However, the test of differences in distributions of GFTA-2 SSs between the older subgroups was not significant.

In order to determine the relation between articulatory accuracy and overall intellectual ability, I computed Spearman correlations separately for each group of children. For the children with WS, GFTA-2 SSs were significantly correlated with DAS-II GCA SSs, $r_s = .59$, $p < .001$. The correlation also was significant for the children with Dup7, $r_s = .47$, $p = .001$. Scatterplots examining the relation between GFTA-2 SSs and DAS-II GCA are presented in Figure 2 for the children with WS and Figure 3 for the children with Dup7. As indicated in Figure 2, all but one of the 45 children with WS who had DAS-II GCA ≥ 70 earned a GFTA-2 SS ≥ 70 . In contrast, for the 73 children with WS who had DAS-II GCA < 70 , GFTA-2 SSs were evenly dispersed across the range of obtained values (39–110). The children with Dup7 showed a different pattern of performance. Figure 3 shows GFTA-2 SSs were evenly dispersed across the range of obtained values (39–110) for the children with Dup7 who had DAS-II GCA ≥ 70 . In contrast, for all eight children with Dup7 who had DAS-II GCA < 70 , the figure shows GFTA-2 SSs also were < 70 .

To confirm the significance of associations between the high/low ($< 70/ \geq 70$) classifications for the GFTA-2 and DAS-II GCA observations, I computed Fisher Exact Tests separately for each group of children. The observed distributions of scores are presented in Table 11. For children with WS, the test revealed that GFTA-2 classification differed significantly by DAS-II GCA classification, $p < .001$, $\phi = .48$. For children with Dup7, the test revealed also that GFTA-2 classification differed significantly by DAS-II GCA classification, $p = .004$, $\phi = .42$.

To test Winitz' (1969) hypothesis that children with IQs ≥ 70 had significantly better speech articulation than children with IQs < 70 ,

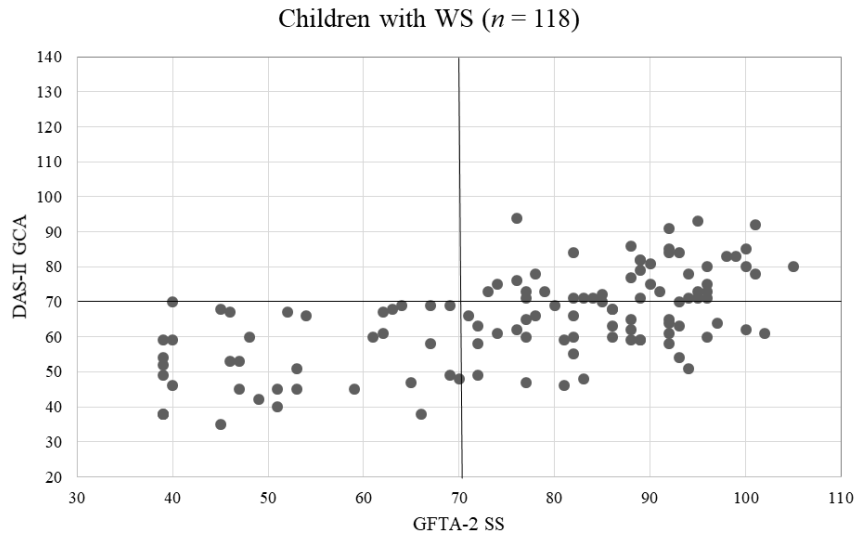


Figure 2. Scatterplot of GFTA-2 SSs and DAS-II GCA for children with WS

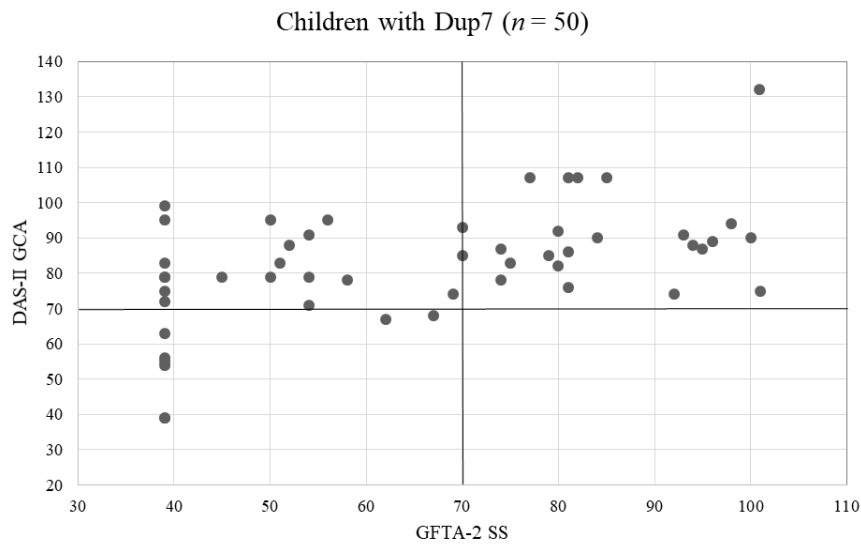


Figure 3. Scatterplot of GFTA-2 SSs and DAS-II GCA for children with Dup7.

the children in each syndrome group were divided into a higher-IQ group (DAS-II GCA ≥ 70) and a lower-IQ group (DAS-II GCA < 70). Separate Mann-Whitney U tests for each syndrome group were computed that compared the GFTA-2 SSs of the children in

the higher-IQ and lower-IQ groups. For the children with WS, the distribution of GFTA-2 SSs (*Median* = 82.00, *IQR* = 63.75–92.00) was significantly higher for the higher-IQ group (*Median* = 88.50, *IQR* = 80.75–94.00) than for the lower IQ-group (*Median* = 50.00, *IQR* = 40.00–62.00), $z = 5.64$, $p < .001$, $r = .52$. For the children with Dup7, the distribution of GFTA-2 SSs (*Median* = 68.00, *IQR* = 39.00–81.25) was significantly higher for the higher-IQ group (*Median* = 81.50, *IQR* = 77.50–94.75) than for the lower-IQ group (*Median* = 42.00, *IQR* = 39.00–54.00), $z = 3.02$, $p = .002$, $r = .43$.

Table 11
Percentages of Children with Standard Scores Above or Below 70

| | Children with Williams Syndrome ($n = 118$) | | | | Total |
|-----------------|---|-----------|------|----------|--------|
| | <i>n</i> | GFTA-2 SS | | <i>n</i> | |
| | | < 70 | ≥ 70 | | |
| DAS-II GCA ≥ 70 | 1 | 0.8% | 44 | 37.3% | 38.1% |
| DAS-II GCA < 70 | 35 | 29.7% | 38 | 32.2% | 61.9% |
| | 36 | | 82 | | |
| Total | | 30.5% | | 69.5% | 100.0% |

| | Children with 7q11.23 Duplication Syndrome ($n = 50$) | | | | Total |
|-----------------|---|-----------|------|----------|--------|
| | <i>n</i> | GFTA-2 SS | | <i>n</i> | |
| | | < 70 | ≥ 70 | | |
| DAS-II GCA ≥ 70 | 18 | 36.0% | 24 | 48.0% | 84.0% |
| DAS-II GCA < 70 | 8 | 16.0% | 0 | 0.0% | 16.0% |
| | 26 | | 24 | | |
| Total | | 52.0% | | 48.0% | 100.0% |

Note. Younger = 4.00–9.99 years; older = 10.00–17.99 years. DAS II = *Differential Ability Scales-II* (Elliott, 2007); GCA = General Conceptual Ability (similar to IQ); GFTA-2 = *Goldman-Fristoe Test of Articulation-2* (Goldman & Fristoe, 2000); SS = standard score.

Articulatory Accuracy: Proportion of Items Correct

As described in Chapter I, movement trajectories for achieving articulatory targets (i.e., gesture patterns for pronouncing phones) stabilize through years of practice (Walsh & Smith, 2002). To determine if articulation of older children with WS or Dup7 was better than articulation of younger children, separate Mann-Whitney U tests for children

with WS and children with Dup7 were computed to examine between age-group differences in GFTA-2 proportion of items correct. The descriptive statistics for proportion of GFTA-2 items correct and the test results for between-group differences are presented in Table 12. As indicated in the table, the distribution of GFTA-2 items proportion correct was significantly higher for the older children with WS than for the younger children. The same outcome was found for the subgroups of younger and older children with Dup7.

Table 12
Study 1 Comparisons for GFTA-2 Proportion of Consonant Items Correct by Age Group

| Group | <i>n</i> | <u>GFTA-2 Consonant Items Correct</u> | | <u>Mann-Whitney U</u> | | |
|----------------|----------|---------------------------------------|------------|-----------------------|----------|----------|
| | | <i>Mdn</i> | <i>IQR</i> | <i>z</i> | <i>p</i> | <i>r</i> |
| WS - Younger | 84 | .67 | .48 - .87 | 6.36 | <.001 | 0.59 |
| WS - Older | 34 | .95 | .87 - .96 | | | |
| Dup7 - Younger | 29 | .55 | .36 - .77 | 4.71 | <.001 | 0.67 |
| Dup7 - Older | 21 | .91 | .77 - .97 | | | |

Note. Dup7 = 7q11.23 Duplication syndrome, WS = Williams syndrome.

Age of customary consonant production. As indicated in Chapter 1 for children in the general population, consonants can be divided into those that are acquired early, in the middle, or late in the development of articulation. To determine if children with WS and children with Dup7 articulate GFTA-2 consonants with the same pattern as ACCP, the proportion correct for the consonants arranged in each of the three periods was calculated separately for each child. For both the children with WS and the children with Dup7, Table 13 presents descriptive statistics for median proportion correct for the sets of Early-, Middle-, and Late-developing GFTA-2 consonant items, the stepwise step-down follow-up results (indicated by subscripts), the Friedman ANOVA Chi square statistic, and the overall significance of the effect.

Table 13

Friedman ANOVAs for Age of Customary Consonant Production based on GFTA-2 Performance

| Group | <i>n</i> | Early Consonants | | Middle Consonants | | Late Consonants | | Overall Effect ^a | |
|--|----------|-------------------|------------|-------------------|------------|------------------|------------|-----------------------------|----------|
| | | <i>Mdn</i> | <i>IQR</i> | <i>Mdn</i> | <i>IQR</i> | <i>Mdn</i> | <i>IQR</i> | χ^2 | <i>p</i> |
| Children with Williams Syndrome | | | | | | | | | |
| WS - All | 118 | .94 _a | .83 - .97 | .75 _b | .45 - .90 | .64 _c | .30 - .88 | 146.76 | < .001 |
| WS - Younger | 84 | .89 _a | .81 - .97 | .60 _b | .30 - .85 | .46 _c | .23 - .81 | 116.12 | < .001 |
| WS - Older | 34 | 1.00 _a | .97 - 1.00 | .90 _a | .80 - .96 | .92 _b | .81 - .96 | 31.26 | < .001 |
| Children with 7q11.23 Duplication Syndrome | | | | | | | | | |
| Dup7 - All | 50 | .94 _a | .81 - 1.00 | .70 _b | .35 - .95 | .58 _c | .19 - .81 | 62.59 | < .001 |
| Dup7 - Younger | 29 | .87 _a | .71 - .99 | .35 _b | .10 - .73 | .23 _c | .14 - .58 | 44.68 | < .001 |
| Dup7 - Older | 21 | 1.00 _a | .92 - 1.00 | .95 _a | .75 - 1.00 | .81 _b | .66 - .94 | 19.13 | < .001 |

Note. Results of stepwise step-down post hoc tests ($\alpha = .05$) following a significant Friedman ANOVA test are indicated by subscripts; consonant-group distributions in each row that differ significantly *do not* share a subscript. Younger = 4.00–9.99 years, older = 10.00–17.99 years. Dup7 = 7q11.23 duplication syndrome; GFTA-2 = *Goldman-Fristoe Test of Articulation-2* (Goldman & Fristoe, 2000); *IQR* = interquartile range; *Mdn* = median; WS = Williams syndrome.

For the full sample of children with WS, the Friedman ANOVA test showed a statistically significant difference in the distributions of proportion correct for the sets of consonant items as a function of Age-of-Customary Consonant Production. Significant differences identified by stepwise step-down follow-up analyses ($\alpha = .05$) are indicated by subscripts in the table. These results revealed that the distribution of consonant-group proportion correct for the Early-developing consonants was significantly higher than the distribution for the Middle-developing consonants which in turn was significantly higher than the distribution for the Late-developing consonants. The pattern of findings for the younger group of children was the same as for the full sample of children. For the older group, the distributions of proportion correct for the Early-developing consonants and the Middle-developing consonants were significantly higher than the distribution for the Late-developing consonants. The distributions of proportion correct did not differ significantly for Early- and Middle-developing consonants.

The findings for the children with Dup7 are presented also in Table 13. The results of the Friedman test for the full sample and the post hoc analyses were the same as for the full sample of children with WS. The findings for the older and younger samples of children with Dup7 were also the same as those for the older and younger samples of children with WS.

Table 14 presents the distributions of the number and percentage of the younger and older children who correctly articulated at least 95% of the consonants (mastery criterion) within each sub-category of the ACCP. As reflected in the Friedman follow-up tests, more children in both syndrome groups articulated early consonants masterfully than either middle consonants or late consonants.

Table 14

Number of Children Who Mastered Consonants Based on GFTA-2 Performance as a Function of ACCP Consonant Group

| Group | <i>n</i> | <u>Early Consonants</u> <i>n, (% Subgroup)</i> | <u>Middle Consonants</u> <i>n, (% Subgroup)</i> | <u>Late Consonants</u> <i>n, (% Subgroup)</i> |
|--|----------|---|--|--|
| Children with Williams Syndrome | | | | |
| WS - All | 118 | 60 (51%) | 18 (15%) | 19 (16%) |
| WS - Younger | 84 | 32 (38%) | 6 (7%) | 7 (8%) |
| WS - Older | 34 | 28 (82%) | 12 (35%) | 12 (35%) |
| Children with 7q11.23 Duplication Syndrome | | | | |
| Dup7 - All | 50 | 22 (44%) | 12 (24%) | 6 (12%) |
| Dup7 - Younger | 29 | 9 (31%) | 2 (7%) | 0 (0%) |
| Dup7 - Older | 21 | 13 (62%) | 10 (41%) | 6 (29%) |

Note. Consonant mastery = 95% accurate articulation for consonants in class. Younger = 4.00–9.99 years, older = 10.00–17.99 years. Dup7 = 7q11.23 duplication syndrome; GFTA-2 = *Goldman-Fristoe Test of Articulation* (2nd ed., Goldman & Fristoe, 2000); WS = Williams syndrome.

Articulatory position-in-words. To compare the distributions of proportion of items correct for consonants in marginal relation with syllable nuclei, sets of Initial- and Final-position GFTA-2 consonant items were examined using related-samples Friedman ANOVA by ranks tests. The GFTA-2 medial consonants were not included in the analyses because these items were not similar phototactically. The findings for both the children with WS and the children with Dup7 are presented in Table 15 for the descriptive statistics for proportion correct and the Friedman ANOVA tests.

The Friedman test results showed the distributions of proportion correct between the sets of Initial- and Final-position consonants did not differ significantly for the full sample of children with WS or for either the younger or older subgroups. However, as indicated in Table 15 for the full sample of children with Dup7, the distribution of proportion correct for Initial-position consonants was significantly higher than the distribution for Final-position consonants.

Table 15

Friedman ANOVAs for Articulatory Position-in-Words Based on GFTA-2 Performance

| Group | <i>n</i> | <u>Initial Consonants</u> | | <u>Final Consonants</u> | | <u>Overall Effect</u> | |
|--|----------|---------------------------|------------|-------------------------|------------|-----------------------|----------|
| | | <i>Mdn</i> | <i>IQR</i> | <i>Mdn</i> | <i>IQR</i> | χ^2 | <i>p</i> |
| Children with Williams Syndrome | | | | | | | |
| WS - All | 118 | .82 | .64 - .95 | .82 | .63 - .89 | 3.06 | .080 |
| WS - Younger | 84 | .73 | .59 - .86 | .74 | .58 - .89 | 1.60 | .317 |
| WS - Older | 34 | .95 | .91 - 1.00 | .89 | .83 - 1.00 | 3.24 | .072 |
| Children with 7q11.23 Duplication Syndrome | | | | | | | |
| Dup7 - All | 50 | .80 | .65 - .95 | .76 | .59 - .94 | 10.52 | .001 |
| Dup7 - Younger | 29 | .70 | .50 - .85 | .65 | .50 - .79 | 5.83 | .016 |
| Dup7 - Older | 21 | .95 | .80 - 1.00 | .94 | .76 - .94 | 4.77 | .029 |

Note. Younger = 4.00–9.99 years, older = 10.00–17.99 years. Dup7 = 7q11.23 duplication syndrome; GFTA-2 = *Goldman-Fristoe Test of Articulation* (2nd ed., Goldman & Fristoe, 2000); *IQR* = interquartile range; *Mdn* = median; WS = Williams syndrome.

This same pattern was found for both the younger and the older subgroups of children with Dup7.

Articulatory place-of-production. To examine accuracy for targeting phones according to the articulatory place feature, the distributions of proportion of items correct for the sets of Bilabial, Dental, Alveolar, Postalveolar-Palatal, and Velar-Glottal GFTA-2 consonant items were compared using Friedman ANOVAs. The findings for both the children with WS and the children with Dup7 are presented in Table 16 and include the descriptive statistics for proportion correct, the Friedman ANOVA tests, and the stepwise step-down follow-up analyses.

The Friedman test result for the full sample of children with WS showed that the distributions of proportion correct differed significantly as a function of articulatory place-of-production. Stepwise step-down follow-up analyses showed

Table 16

Friedman ANOVAs for Articulatory Place of Production Based on GFTA-2 Performance

| Group | <i>n</i> | Bilabial | Dental | Alveolar | Postalveolar Palatal | Velar- Glottal | Overall Effect ^a | |
|--|----------|------------------------------------|------------------------------------|-----------------------------------|-------------------------------------|-------------------------------------|-----------------------------|----------|
| | | <i>Mdn</i> [<i>IQR</i>] | <i>Mdn</i> [<i>IQR</i>] | <i>Mdn</i> [<i>IQR</i>] | <i>Mdn</i> [<i>IQR</i>] | <i>Mdn</i> [<i>IQR</i>] | <i>X</i> ² | <i>p</i> |
| Children with Williams Syndrome | | | | | | | | |
| WS - All | 118 | 1.00 _a [.80 - 1.00] | .73 _c [.45 - .91] | .78 _{b,c} [.56 - .89] | .86 _b [.57 - 1.00] | 1.00 _a [.86 - 1.00] | 160.43 | < .001 |
| WS - Younger | 84 | .90 _a [.80 - 1.00] | .55 _c [.36 - .82] | .67 _{b,c} [.50 - .89] | .71 _b [.43 - 1.00] | .89 _a [.78 - 1.00] | 121.28 | < .001 |
| WS - Older | 34 | 1.00 _a [1.00 - 1.00] | .91 _b [.82 - 1.00] | .93 _b [.87 - 1.00] | .90 _{a,b} [.80 - 1.00] | 1.00 _a [1.00 - 1.00] | 44.75 | < .001 |
| Children with 7q11.23 Duplication Syndrome | | | | | | | | |
| Dup7 - All | 50 | 1.00 _a [.90 - 1.00] | .73 _b [.53 - 1.00] | .81 _b [.56 - .94] | .57 _b [.14 - .90] | .95 _a [.78 - 1.00] | 61.22 | < .001 |
| Dup7 - Younger | 29 | .90 _a [.80 - 1.00] | .55 _{b,c} [.23 - .87] | .67 _b [.47 - .86] | .29 _c [.14 - .57] | .89 _a [.56 - 1.00] | 51.08 | < .001 |
| Dup7 - Older | 21 | 1.00 _a [.90 - 1.00] | .91 _{a,b} [.73 - 1.00] | .89 _b [.78 - 1.00] | 1.00 _{a,b} [.71 - 1.00] | 1.00 _{a,b} [.89 - 1.00] | 18.19 | .001 |

Note. Results of stepwise step-down post hoc tests ($\alpha = .05$) following a significant Friedman ANOVA test are indicated by subscripts; consonant-group distributions in each row that differ significantly *do not* share a subscript. Younger = 4.00–9.99 years, older = 10.00–17.99 years. Dup7 = 7q11.23 duplication syndrome; GFTA-2 = *Goldman-Fristoe Test of Articulation* (2nd ed., Goldman & Fristoe, 2000); *IQR* = interquartile range; *Mdn* = median; WS = Williams syndrome.

that the distributions of proportion correct for the Bilabial and Velar-Glottal consonants were significantly higher than the distributions for the Postalveolar-Palatal, Dental, and Alveolar consonants. The distribution of proportion correct for the Postalveolar-Palatal consonants was significantly higher than the distribution for the Dental consonants. The distributions for the Bilabial and Velar-Glottal consonants did not differ significantly nor did the distributions differ significantly for the Postalveolar and Alveolar consonants or for the Alveolar and Dental Consonants. The pattern of findings for the younger subgroup of children with WS was the same as for the full sample of children. For the older subgroup of children with WS, the distributions of proportion correct for both the Bilabial and the Velar-Glottal consonants were significantly higher than the distributions for the Alveolar and Dental consonants. The distributions of proportion correct for the Bilabial, Velar-Glottal, and Postalveolar-Palatal consonants did not differ significantly nor did the distributions of proportion correct for the Alveolar, Dental, and Postalveolar-Palatal consonants differ significantly.

For the children with Dup7, significant differences were found among distributions of proportion correct for sets of consonants as a function of the articulatory place-of-production arrangement. For the full sample of children, the stepwise step-down follow-up analyses indicated that the distributions of proportion correct for both the Bilabial and the Velar-Glottal consonants were significantly higher than the distributions for the Postalveolar-Palatal, Alveolar, and Dental consonants. The distributions for the Bilabial and for the Velar-Glottal consonants did not differ significantly nor did the distributions for the Postalveolar-Palatal, Alveolar, and Dental consonants differ significantly. For the younger subgroup, the stepwise step-down results indicated that the

distributions of proportion correct for Bilabial and Velar-Glottal consonants were significantly higher than the distributions for the Alveolar, Dental, and Postalveolar-Palatal consonants. In addition, the distribution of proportion correct for the Alveolar consonants was significantly higher than the distribution for the Postalveolar-Palatal consonants. The distribution of proportion correct for the Bilabial consonants did not differ significantly from the distribution for the Velar-Glottal consonants, nor were there significant differences in the distributions for the Alveolar and Dental consonants or for the Dental and the Postalveolar-Palatal consonants. For the older group of children, stepwise step-down tests indicated that the distributions of proportion correct for the Bilabial consonants was significantly higher than the distribution for Alveolar consonants. No other differences were significant.

Articulatory manner-of-production. To examine accuracy for targeting phones according to the articulatory manner feature, the distributions of proportion of consonants correct for the sets of Nasal, Stop, Fricative, and Approximant GFTA-2 consonants were compared using related-samples Friedman ANOVAs. The findings for both the children with WS and the children with Dup7 are presented in Table 17 and include the descriptive statistics for proportion correct, the Friedman ANOVA tests, and the stepwise step-down follow-up analyses.

For the children with WS, the Friedman test showed that there was a significant difference in the distributions of proportion correct as a function of the articulatory manner-of-production arrangement. Stepwise step-down follow-up analyses showed that the distributions of proportion correct for the Nasal and Stop consonants were significantly higher than the distributions for either the Fricative or Approximant

Table 17

Friedman ANOVAs for Articulatory Manner of Production Based on GFTA-2 Performance

| Group | <i>n</i> | Nasal | Stop | Fricative | Approximant | Overall Effect ^a | |
|---------------------------------|----------|------------------------------------|--------------------------------------|----------------------------------|----------------------------------|-----------------------------|----------|
| | | <i>Mdn</i> [<i>IQR</i>] | <i>Mdn</i> [<i>IQR</i>] | <i>Mdn</i> [<i>IQR</i>] | <i>Mdn</i> [<i>IQR</i>] | χ^2 | <i>p</i> |
| Children with Williams Syndrome | | | | | | | |
| WS - All | 118 | 1.00 _a [.88 - 1.00] | .94 _a [.83 - 1.00] | .71 _b [.47 - .90] | .75 _b [.50 - .88] | 182.42 | < .001 |
| WS - Younger | 84 | .88 _a [.75 - 1.00] | .89 _a [.78 - 1.00] | .57 _b [.38 - .76] | .63 _b [.38 - .85] | 144.83 | < .001 |
| WS - Older | 34 | 1.00 _a [1.00 - 1.00] | 1.00 _a [.94 - 1.00] | .90 _b [.84 - .95] | .88 _b [.88 - 1.00] | 38.92 | < .001 |
| 7q11.23 Duplication Syndrome | | | | | | | |
| Dup7 - All | 50 | 1.00 _a [.88 - 1.00] | .94 _b [.82 - 1.00] | .70 _c [.44 - .95] | .50 _c [.38 - .88] | 73.89 | < .001 |
| Dup7 - Younger | 29 | .88 _a [.75 - 1.00] | .88 _a [.62 - 1.00] | .55 _b [.25 - .78] | .38 _b [.25 - .57] | 53.83 | < .001 |
| Dup7 - Older | 21 | 1.00 _a [1.00 - 1.00] | 1.00 _{a, b} [.88 - 1.00] | .95 _b [.78 - 1.00] | .88 _b [.69 - 1.00] | 22.45 | < .001 |

Note. Results of stepwise step-down post hoc tests ($\alpha = .05$) following a significant Friedman ANOVA test are indicated by subscripts; consonant-group distributions in each row that differ significantly *do not* share a subscript. Younger = 4.00–9.99 years, older = 10.00–17.99 years. Dup7 = 7q11.23 duplication syndrome; GFTA-2 = *Goldman-Fristoe Test of Articulation-2* (Goldman & Fristoe, 2000); *IQR* = interquartile range; *Mdn* = median; WS = Williams syndrome.

consonants. The distributions for the Nasal and Stop consonants did not differ significantly nor was there a significant difference in the distributions for the Fricative and Approximant consonants. The pattern of findings for both the younger and older groups was the same as for the full sample.

For the full sample of children with Dup7, the Friedman ANOVA test showed that there was a significant difference in the distributions of proportion correct as a function of articulatory manner-of-production. Stepwise step-down analyses showed that the distribution of proportion correct for Nasal consonants was significantly higher than the distribution for Stop consonants which in turn was significantly higher than the distributions for both the Fricative and the Approximant consonants, which did not differ significantly. For the younger subgroup of children, the distributions of proportion correct for the Nasal and Stop consonants were significantly higher than the distributions for the Fricative and the Approximant consonants. The distributions for the Nasal and Stop consonants did not differ significantly nor did the distributions for the Fricative and Approximant consonants. For the older subgroup of children with Dup7, the distribution for the Nasal consonants was significantly higher than the distributions for either the Fricative or Approximant consonants. The distributions for the Nasal and Stop consonants did not differ significantly, nor did the distributions for the Stop, Fricative, and Approximant consonants.

Planes-of-movement for double consonant articulations. As described in Chapter I, accuracy for articulating consonant clusters requires rapid movement transitions. To examine accuracy for articulating double consonants with regard to planes-of-movement transitions, the distributions of proportion correct for double

consonants correct arranged for *Back-to-Front*, *Front-to-Back*, and *Same Place* movements were compared using related-samples Friedman ANOVAs. The findings for both children with WS and children with Dup7 are presented in Table 18 and include the descriptive statistics for proportion correct, the Friedman ANOVA tests, and the stepwise step-down follow-up analyses.

For the full sample of children with WS, the Friedman test showed that the distributions of proportion correct among the sets of GFTA-2 double consonants examined as a function of articulatory planes-of-movement differed significantly. Stepwise step-down analyses indicated that the distribution of proportion correct for Same Place double consonants was significantly higher than the distributions for both Front-to-Back and Back-to-Front double consonants, which did not differ significantly. This pattern was found also for the younger subgroup of children. Test results showed the distributions of proportion correct for the three types of double consonants did not differ significantly for the older children with WS.

For the children with Dup7, there was a statistically significant difference in the distributions of proportion correct for double consonants as a function of articulatory planes-of-movement. Stepwise step-down analyses indicated that the distribution for Same Place double consonants was significantly higher than the distributions for both the Back-to-Front and Front-to-Back double consonants, which did not differ significantly. The same pattern obtained for the younger children with Dup7. For the older children with Dup7, the Friedman test indicated that the distributions of proportion correct as a function of double consonant group did not differ significantly.

Table 18

Friedman ANOVA for Planes-of-Movement for Double Consonant Articulations

| Group | <i>n</i> | Back-to-Front | | Front-to-Back | | Same Place | | Overall Effect ^a | | |
|--|----------|------------------|------------|------------------|------------|------------------|------------|-----------------------------|----------|--|
| | | <i>Mdn</i> | <i>IQR</i> | <i>Mdn</i> | <i>IQR</i> | <i>Mdn</i> | <i>IQR</i> | <i>X</i> ² | <i>p</i> | |
| Children with Williams Syndrome | | | | | | | | | | |
| WS - All | 118 | .57 _b | .29 - .86 | .71 _b | .14 - 1.00 | .75 _a | .50 - .88 | 19.50 | < .001 | |
| WS - Younger | 84 | .43 _b | .14 - .82 | .36 _b | .14 - .82 | .63 _a | .25 - .88 | 23.22 | < .001 | |
| WS - Older | 34 | .86 | .71 - 1.00 | 1.00 | .86 - 1.00 | .88 | .85 - 1.00 | 4.57 | .102 | |
| Children with 7q11.23 Duplication Syndrome | | | | | | | | | | |
| Dup7 - All | 50 | .50 _b | .00 - .75 | .43 _b | .00 - .86 | .75 _a | .25 - 1.00 | 20.37 | < .001 | |
| Dup7 - Younger | 29 | .14 _b | .00 - .57 | .00 _b | .00 - .43 | .38 _a | .00 - .82 | 16.98 | < .001 | |
| Dup7 - Older | 21 | .86 | .57 - 1.00 | .86 | .71 - 1.00 | 1.00 | .75 - 1.00 | 4.80 | .091 | |

Note. Results of stepwise step-down post hoc tests ($\alpha = .05$) following a significant Friedman ANOVA test are indicated by subscripts; consonant-group distributions in each row that differ significantly *do not* share a subscript. Plane-of-movement groups consist of GFTA2 initial clusters and affricates. Younger = 4.00–9.99 years, older = 10.00–17.99 years; Dup7 = 7q11.23 duplication syndrome; GFTA2 = *Goldman-Fristoe Test of Articulation-2* (Goldman & Fristoe, 2000); *IQR* = interquartile range; *Mdn* = median; WS = Williams syndrome.

Discussion

As predicted for both children with WS and children with Dup7, articulatory accuracy was significantly below expectations for same-aged children in the general population even though all children were enrolled in speech services at the time of the study or had had speech intervention services in the past. As a group, children with WS articulated with significantly better accuracy than did children with Dup7. This is particularly striking given that the children with Dup7 had significantly higher IQs than the children with WS. The younger children with WS also articulated with significantly better accuracy than did the younger children with Dup7. However, the difference in proportion correct did not differ significantly between the two older groups of children.

Importantly, the test for differences between the older compared with the younger children with WS revealed significantly higher median GFTA-2 SS for the older children. The same significant effect was found between the older and younger groups with Dup7. These findings suggest that both children with WS and children with Dup7 continue to refine articulation given time and ongoing practice speaking.

As predicted, the correlation between articulatory accuracy and intellectual ability was significant for both children with WS and children with Dup7. Similarly, as predicted by Winitz (1969), for both syndrome groups, children with IQs at or above 70 had significantly higher GFTA-2 SSs than children who had IQs below 70.

At the same time, the scatterplots of the relation between articulatory accuracy and intellectual ability showed distinct patterns as a function of syndrome. In particular, for children with WS, all but one child with IQ at or above 70 earned GFTA-2 SSs at or above 70, while for children with IQ below 70, GFTA-2 SSs were distributed across the

entire range. In contrast, for children with Dup7, for children with IQs at or above 70, GFTA-2 SSs were distributed across the full range but for children with IQs below 70, GFTA-2 SSs also were below 70 (six of eight children with GFTA-2 SS <70 obtained the lowest GFTA-2 SS). Thus, while IQ in the normal range appears to support speech articulation for children with WS, it does not appear to do so for children with Dup7.

As predicted, the pattern of consonant accuracy for both the WS group and the Dup7 group fit the developmental pattern previously identified for children in the general population (Smit et al., 1990; Templin, 1957). In particular, children in both groups produced a significantly higher proportion of the Early-developing consonants correctly than the Middle-developing consonants and a significantly higher proportion of the Middle-developing consonants than the Late-developing consonants.

Templin (1957) and others have indicated that nearly all English-speaking children in the general population who are aged eight years are expected to have mastered articulation of all English consonants. Of the 84 children with WS who were older than aged 10.00 years, the large majority obtained 95% accuracy (mastery) for the Early-developing consonants and about one-third of them had mastered the Late-developing consonants. The pattern of mastery for ACCP consonants was less positive for the older children with Dup7. Less than three-fourths of these children obtained 95% mastery for the Early-developing consonants and less than one-third had mastered the Late-developing consonants. Variability across the ACCP classes was apparent in the spread of the data reported as descriptive statistics with the results of each Friedman's ANOVA (*IQR* = middle 50% of the observed data). For example, for the older children with Dup7,

the *IQR* for the Late-Developing consonants ranged from .66–.94 (.28). For the older children with WS, the *IQR* for the Late-Developing consonants was from .81–.96 (.15).

Children in the general population aged 8 years or younger are more likely to produce Initial consonant sounds with greater accuracy than Final consonant sounds (Stoel-Gammon, 1985; Templin, 1957). As predicted, children with Dup7 showed the same pattern. However, the proportion of Initial and Final consonants produced correctly by the younger and older age groups did not differ significantly for children with WS.

As predicted, consonants produced at the front (bilabial) or back (velar-glottal) of the mouth were produced with significantly greater accuracy than consonants produced in the central oral area (dental, alveolar, and postalveolar-palatal). This was true for both children with WS and children with Dup7. This is the same pattern shown in a table of sound elements for younger children in the general population (Templin, 1957, p. 51). Templin considered word position in her description of consonants; specifically, consonants in her study could have as many as three elements, such as a /t/ that is articulated in the initial, medial, or final word position. In particular, Templin reported that all bilabial and velar-glottal consonants in English were produced correctly by at least 75% of children aged 49 months, but that several dental, palatal, and alveolar consonants had not yet been acquired.

Also as predicted, nasal and stop consonants were produced significantly more accurately than were fricatives and approximants by both children with WS and children with Dup7. This is the same pattern as is shown by younger children who are developing typically (Smit et al., 1990).

The pattern of findings for double consonants also fit the predicted pattern for the full samples of children and for the younger samples. Double consonants that were articulated at the same position were produced with significantly greater accuracy than double consonants requiring lingual transitions to a new position for the second consonant. This is the same pattern that was found by both Templin (1957) and Smit et al. (1990) for younger children in the general population. Across the planes-of-movement classes, the tests for differences in distributions of proportion correct were not significant for children in either of the older groups.

Articulatory accuracy may contribute significantly to phonological processing, a skill that has been shown repeatedly to be crucial for learning to read (National Reading Panel, 2000). In the next chapter, I investigate the relations between phonological processing, articulatory accuracy, and a variety of other cognitive-linguistic variables. This second study contributes to the literature because it is the first systematic investigation of the relation between articulatory accuracy and these cognitive and linguistic variables for the children with WS or the children with Dup7. In addition, for the children with WS only, I provide the first systematic study of the possible contribution of articulatory accuracy to phonological processing ability beyond the contribution of these other cognitive and linguistic variables, all previously found to be significant contributors to phonological processing. I was not able to address this question for the children with Dup7 because the sample size was too small.

CHAPTER III
MODELING RELATIONS BETWEEN ARTICULATORY
ACCURACY AND PHONOLOGICAL PROCESSING

Literature reviewed in Chapter I showed that cognitive processes involved in learning to speak are dependent on physical movement integrity (Green, Moore, Higashikawa, & Steeve, 2000; Kent, 2000; Smith & Zelaznik, 2004), sensory perception (Werker & Yeung, 2005), a capacity for learning language (Bloom & Lahey, 1978; Locke, 1993), and productive interaction among these factors (Nip, Green, & Marx, 2011). With ongoing word-learning and language development (Lee, Davis, & MacNeilage, 2010; Locke, 1993), and given specific instruction (Torgesen & Burgess, 2013), young school-age children acquire explicit understanding that phonemes are constituents in words (National Reading Panel, 2000). Children with this knowledge can accurately count syllables in words (segment constituents); state and match word onsets or rimes; recall similar speech sounds or similar speech-sound patterns (alliterate, match, complete); blend sequences of phonemes to form a word; and delete word parts with memory for the remainder (elision). The achievement of explicit phonological processing supports the development of literacy. Phonological processing is thus multifaceted and hierarchical in nature (ASHA, 2018; Brady & Shankweiler, 1991; Fowler, 2011; Gillon, 2017; McDowell et al., 2007; National Reading Panel, 2000;

Nittrouer, Shune, & Lowenstein, 2011; Overby et al., 2012; Parrila et al., 2004; Thomas & Senechal, 2004; Wagner, Torgesen, Rashotte, & Pearson, 2013).

Mervis (2009) reported that children with WS demonstrate relative strengths in phonological processing and verbal short-term memory. Velleman, Huffman, and Mervis (2013) reported that phonological processing skills for children with Dup7 varied widely about the average skill expected for children in the general population. For children with WS and children with Dup7, the present study is the first designed to address the relation between articulatory accuracy and phonological processing. In addition, for children with WS, the present study is the first to address the question of whether variation in articulatory accuracy accounts for unique variance in phonological processing over and above that explained by variations in verbal short-term memory, nonverbal reasoning, spatial ability, and vocabulary ability.

Method

Participants

The final participant sample included 76 children with WS (40 girls, 36 boys) aged 6.01–12.77 years (*Mean* = 7.94 years, *SD* = 2.05) and 30 children with Dup7 (13 girls, 17 boys) aged 6.00–12.95 years (*Mean* = 9.11 years, *SD* = 1.97). All of the children also participated in Study 1. For the present study, the first assessment that included administration of the GFTA-2 within the 6.00–12.99 year age range was used for each child. For 55 children with WS and all but one of the children with Dup7, this was the same assessment as was included in Study 1. For the remaining children, all of whom were 4 or 5 years old at the time of the Study 1 assessment, a later assessment was used. Authorization for the study was granted by the University of Louisville Institutional

Review Board. All participants were receiving speech-language intervention services at the time of the study and/or had had speech-language services in the past.

Sociodemographics. For the 76 children with WS, the distribution of place of residence with regard to US Census regional divisions was as follows: 3 children (3.9%) were from Northeastern New England states, 18 children (23.7%) were from Northeastern Middle Atlantic states, 20 children (26.3%) were from Southern Atlantic states, 6 children (7.9%) were from Southern East South-Central states, 3 children (3.9%) were from Southern West South-Central states, 10 children (13.2%) were from Midwestern East North-Central states, 5 children (6.6%) were from Midwestern West North-Central states, 3 children (3.9%) were from Western Mountain states, and 6 children (7.9%) were from Western Pacific states. Two children (2.6%) were from Canada. Childrens' mothers' educational attainment was as follows: 20 mothers (26.3%) did not have a bachelor's degree and 56 mothers (73.7%) attained a bachelor's degree or higher. Childrens' reported racial and ethnic affiliations were as follows: 58 children (76.3%) were white, non-Hispanic; 8 children (10.5%) were white, Hispanic; 3 children (3.9%) were African American, non-Hispanic; 3 children (3.9%) were biracial or triracial, non-Hispanic; and 3 children (3.9%) were biracial or triracial, Hispanic.

For the 30 children with Dup7, the distribution of place of residence with regard to US Census regional divisions was as follows: 2 children (6.7%) were from Northeastern New England states, 5 children (16.7%) were from Northeastern Middle Atlantic states, 10 children (33.3%) were from Southern Atlantic states, 2 children (6.7%) were from Southern East South-Central states, 5 children (16.7%) were from Midwestern East North-Central states, 2 children (6.7%) were from Midwestern West North-Central

states, 1 child (3.3%) was from a Western Mountain state, and 2 children (6.7%) were from Western Pacific states. One child (3.3%) was from the United Kingdom. Childrens' mothers' educational attainment was as follows: 17 mothers (56.7%) did not have a bachelor's degree and 13 mothers (43.3%) had attained a bachelor's degree or higher. Childrens' reported racial and ethnic affiliations were as follows: 25 children (83.3%) were white, non-Hispanic; 2 children (6.7%) were white, Hispanic; 1 child (3.3%) was African American, non-Hispanic; and 2 children (6.7%) were biracial or triracial, non-Hispanic.

Inclusion and exclusion criteria. Participants were included in Study 2 if they (a) had genetically-confirmed classic length deletion or duplication of the WS region, (b) met the age-range criterion of 6.00–12.99 years, and (c) completed all of the standardized assessments used in the study as part of the same assessment. No child meeting these criteria was excluded. All participants were receiving or had received speech therapy.

Measures

Dependent variable. Phonological skill was measured by performance on the DAS-II Phonological Processing subtest (Elliott, 2007). Phonological processing measures awareness of, memory for, and access to the phonological structure of oral language. Children aged 6.00–8.99 years completed the Early Years Phonological Processing subtest and children aged 9.00–12.99 years completed the School-Age Phonological Processing subtest. Skills assessed on both versions were the same: rhyming, syllable and phoneme blending, syllable and phoneme elision, and identifying first, last, or all phonemes in words provided by the examiner. Each measure provided *T*-scores ranging from 10–90 (*SD* = 10). *T*-scores were transformed to *SSs* (*Mean* = 100,

$SD = 15$) for consistency of scores in the analyses. The mean internal consistency reliability coefficient for the Early Years item set was $r = .90$, $SEM = 2.91$. The average corrected stability coefficient was $r = .93$, $SDiff = .13$ indicating excellent temporal stability for retesting. The mean internal consistency reliability coefficient for the School Age item set was $r = .91$ $SEM = 2.72$. The average corrected stability coefficient was $r = .86$, $SDiff = .30$ indicating very good temporal stability for retesting.

Independent variables. Five independent variables reported previously to have been related to phonological processing ability were included in this study ($Mean SS = 100$, $SD = 15$): speech articulatory accuracy, nonverbal reasoning ability, spatial ability, verbal short-term memory, and vocabulary.

Speech articulatory accuracy. The standardized GFTA-2 (Goldman & Fristoe, 2000) measures phone accuracy for articulating 77 GFTA-2 consonant items in single-words on cue. Details of the GFTA-2 and its psychometrics were discussed in Chapter II.

Nonverbal reasoning. The DAS-II Nonverbal Reasoning cluster SS was used to measure nonverbal, inductive reasoning. Children aged 6.00–8.99 years completed the Early Years cluster consisting of the Matrices subtest (analytical reasoning) and the Picture Similarities subtest (visual integration). The mean internal consistency reliability coefficient for the Early Years item set was $r = .89$, $SEM = 5.15$. The average corrected stability coefficient was $r = .77$, $SDiff = .43$ indicating good temporal stability for retesting.

Children aged 9.00–12.99 years completed the School-Age Nonverbal Reasoning cluster consisting of the Matrices subtest and the Sequential and Quantitative Reasoning subtest (inductive problem solving and verbal mediation). The mean internal consistency

reliability coefficient for the School Age item set was $r = .92$, $SEM = 4.22$. The average corrected stability coefficient was $r = .89$, $SDiff = .34$ indicating excellent temporal stability for retesting.

Spatial ability. The DAS-II Spatial Ability cluster SS was used to measure visual-spatial processing. Children aged 6.00–8.99 years completed the Early Years Spatial Ability cluster consisting of the Pattern Construction (visual-spatial analysis and synthesis) and Copying (spatial imagery and orientation) subtests. The mean internal consistency reliability coefficient for the Early Years item set was $r = .95$, $SEM = 3.40$. The average corrected stability coefficient was $r = .89$, $SDiff = .23$ indicating excellent temporal stability for retesting.

Children aged 9.00–12.99 years completed the School-Age Spatial Ability cluster consisting of the Pattern Construction subtest and the Recall of Designs subtest (memory for orientation and visual-spatial matching). The mean internal consistency reliability coefficient for the School-Age item set was $r = .95$, $SEM = 3.45$. The average corrected stability coefficient was $r = .88$, $SDiff = .30$ indicating excellent temporal stability for retesting.

Verbal short-term memory. The DAS-II Recall of Digits-Forward subtest measures short-term auditory-sequential memory for strings of digits produced by the examiner at a rate of two digits per second. Children aged 6.00–8.99 years completed the Early Years Recall of Digits-Forward subtest and children aged 9.00–12.99 years completed the School-Age Recall of Digits-Forward subtest. Each version of the measure provided T -scores ranging from 10–90 ($SD = 10$). T -scores were transformed to SSs ($Mean = 100$, $SD = 15$) for consistency of scores in the analyses. The mean internal

consistency reliability coefficient for the Early Years item set was $r = .91$, $SEM = 2.83$. The average corrected stability coefficient was $r = .80$, $SDiff = .19$ indicating good temporal stability for retesting. The mean internal consistency reliability coefficient for the School Age item set was $r = .92$, $SEM = 2.87$. The average corrected stability coefficient was $r = .71$, $SDiff = .00$ indicating good temporal stability for retesting.

Vocabulary. Two standardized vocabulary measures were administered. The *Peabody Picture Vocabulary Test-4* (PPVT-4; Dunn & Dunn, 2007) is a single-word, receptive vocabulary measure for assessing knowledge of English vocabulary; Form B was used. Pictured items broadly sample nouns, verbs, and attributes across increasing levels of difficulty. It is appropriate for use with individuals 2.5–90 years and for administration to children who are both typically developing and with special needs. Standardization occurred with a sample of 3,540 individuals similar in sociodemographic characteristics to the US population (US Census Bureau, 2004). Mean split-half internal reliability of the items for Form B = .94, $SEM = 3.6$; mean test-retest reliability $r = .93$.

The *Expressive Vocabulary Test- 2* (EVT-2; Williams, 2007) is a single-word expressive vocabulary and word retrieval measure of English. Children are asked to cite nouns, verbs, or attributes in response to pictured stimuli or to provide a synonym for a word provided by the examiner. Form B was used. The EVT-2 was standardized for use with individuals aged 2.5–90 years. It is appropriate for use with both children who are typically developing and children with special needs and was co-normed with the PPVT-4. Mean split-half internal reliability of the items for Form B = .93, $SEM = 3.9$; mean test-retest reliability $r = .95$.

Because of the very high correlation between the PPVT-4 SS and the EVT-2 SS for children with WS, multicollinearity was likely a threat to the outcomes of planned multiple regression analyses. For this reason, a composite vocabulary measure was formed using the formula: $(PPVT-4\ SS + EVT-2\ SS) / 2$. The composite vocabulary variable SS was used in the following analyses. (Note that the correlation between PPVT-4 SS and EVT-2 SS also was very high for the children with Dup7.)

Research Design

The present study is an empirical, community-based, and cross-sectional examination of the correlations among SSs on speech articulation, cognition, and language measures. In addition, for children with WS, hierarchical multiple regression analysis was used to examine the possibility of a unique contribution of speech articulation to the variance in phonological processing ability over and above that contributed by other cognitive and linguistic variables.

Procedure

All participants visited the NSL for the purpose of completing a battery of cognitive, language, psychosocial, academic, and speech-production assessments. All standardized measures were administered in accordance with the test authors' instructions. All children completed the full assessment battery within three days except for one child who began intellectual assessment prior to the traditional, 2-week university winter holiday and completed speech assessment on the first day following the break.

Data collection. Continuous variables were measured as SSs ($Mean = 100, SD = 15$). All GFTA-2 assessments were audiovideo recorded for coding later using recording

procedures described in Chapter II. Difficult-to-code items were resolved successfully according to procedures described in Chapter II and outlined in Appendix C.

Data Analysis. Statistical analyses were conducted using IBM SPSS 25 statistical software. Expectedly, for children with WS and for children with Dup7, statistical exploration of the distributions of the GFTA-2 SSs revealed violations of the parametric assumption of normality. Therefore, Spearman correlations were used. To partially adjust for the number of correlations computed, α was set at .01.

Reliability. The reliability sample included 19 randomly chosen records from children with WS (25%) and 7 randomly chosen records from children with Dup7 (23%). Second-judge coding occurred independently from the dissertation author. Second judges followed the coding procedures outlined in Appendix C. A total of 12 practice coding efforts, using GFTA-2 records not included in the present study, were performed prior to working on the reliability sample. Reliability statistics for GFTA-2 SSs were excellent. For children with WS, GFTA-2 second-judge SSs fell within the confidence interval of the dissertation author's scores 94% of the time. For children with Dup7, GFTA-2 second-judge SSs fell within the confidence interval of the dissertation author's scores 100% of the time.

Correlational analyses. In order to test bivariate relations between the study variables, nonparametric Spearman rank order correlations were computed separately for the children with WS and the children with Dup7. Variables tested included chronological age and seven standardized variables: phonological processing (DAS-II Phonological Processing SS), articulatory accuracy (GFTA-2 SS), nonverbal reasoning

ability (DAS-II Nonverbal Reasoning SS), verbal short-term memory (DAS-II Recall of Digits-Forward SS), spatial ability (DAS-II Spatial SS), and composite vocabulary SS.

Multiple regression. Hierarchical multiple regression was used to test whether the measure of articulatory accuracy contributed unique variance to the measure of phonological processing over and above that contributed by the combined effect of the remaining variables described above. These variables are known to be importantly related to phonological processing. A plausible theoretical relation between the dependent variable and the explanatory variables is crucial for using the hierarchical multiple regression procedure (Osborne, 2017; Petrocelli, 2003). Thus, the present choice for the set of Model I variables was based on strong support in the literature indicating their importance in the development of phonological processing and on positive correlations shown between them and phonological processing.

The regression procedure forced the statistical software to show the unique incremental contribution of articulatory accuracy to phonological processing in the following ways: (a) the F change increase and the significance of the F change, (b) the change in significance for the b coefficients in Model 2 due to the addition of articulatory accuracy, (c) the reduction in residual sum of squares from Model 1 to Model 2 (indicating improvement in the fit of the predicted to the observed data), and (d) the increase in R^2 from Model 1 to Model 2 (Petrocelli 2003). To measure the local effect of the independent variables for predicting phonological processing, the effect-size index, f^2 , was used: $(R^2 \text{ inclusive model} - R^2 \text{ restricted-variable model}) / (1 - R^2 \text{ inclusive model})$. The recommended classification scheme for interpreting the effect-size index quotients is: .02 = small effect; .15 = medium effect, and .35 = large effect (Cohen, 1988, p. 413).

Results

Performance on Standardized Assessments

Descriptive statistics for the dependent and independent variables are presented in Table 19. Children with WS were significantly younger and obtained significantly lower SSs for nonverbal reasoning ability, spatial ability, and composite vocabulary than children with Dup7. As expected, children with WS obtained significantly higher SSs for articulatory accuracy and phonological processing. The difference between groups on the measure of verbal short-term memory was not significant.

Correlational Analyses

Two separate series of correlational analyses were conducted, one for the children with WS and one for the children with Dup7. Results are presented in Table 20. As SSs (which are based on reference groups, distributed along the normal curve, and consistent across designated age ranges) were used to measure the dependent and independent variables, no significant correlations with age were expected, or were found, for either syndrome group.

For the children with WS, all remaining correlations were positive and significant ($\alpha = .01$) except for two: the correlation between articulatory accuracy SS and nonverbal reasoning SS, and the correlation between verbal short-term memory SS and nonverbal reasoning SS. Similarly, for the children with Dup7 all of the remaining correlations except for the correlation between speech articulatory accuracy SS and verbal short-term memory SS were significant. Although the latter correlation was not statistically significant, its effect size was moderate (implicating small sample size).

Table 19

Study 2 Descriptive Statistics and Independent Comparisons for Age and Standardized Measures: Children with WS or Dup7

| Variable | WS (n = 76) | | Dup7 (n = 30) | | Mann-Whitney U | | |
|-------------------------|-------------|----------------|---------------|----------------|----------------|-------|-------|
| | Median | IQR | Median | IQR | z | p | r |
| Age | 7.20 | 6.13 – 9.26 | 9.03 | 7.77 – 10.81 | -2.76 | .006 | -0.27 |
| GFTA-2 SS | 83.00 | 62.25 – 92.00 | 71.50 | 74.50 – 81.00 | 3.15 | .002 | 0.31 |
| DAS-II NVR SS | 79.50 | 70.25 – 89.75 | 88.00 | 83.00 – 93.50 | -2.49 | .013 | -0.24 |
| DAS-II SA SS | 52.00 | 38.00 – 66.75 | 86.50 | 76.75 – 96.50 | -6.20 | <.001 | -0.60 |
| DAS-II DigFwd SS | 75.00 | 65.00 – 89.00 | 83.00 | 70.75 – 94.00 | -1.53 | .126 | -0.14 |
| DAS-II PhP SS | 92.00 | 73.75 – 101.00 | 84.50 | 72.25 – 89.75 | 2.05 | .041 | 0.20 |
| Composite Vocabulary SS | 83.75 | 75.50 – 92.75 | 93.75 | 85.88 – 100.00 | -3.41 | <.001 | -0.33 |

Note. Composite Vocabulary SS = ([Peabody Picture Vocabulary Test-4 SS + Expressive Vocabulary Test-2 SS] / 2); DAS II = *Differential Ability Scales-II* (Elliott, 2007); DigFwd = Recall of Digits-Forward subtest; Dup7 = 7q11.23 duplication syndrome; GFTA2 = *Goldman-Fristoe Test of Articulation-2* (Goldman & Fristoe, 2000); IQR = interquartile range; *Mdn* = median; NVR = Nonverbal reasoning ability cluster; PhP = Phonological Processing subtest; SA = Spatial ability cluster; SS = standard score; WS = Williams syndrome.

Table 20
Bivariate Spearman Correlations Among CA and Assessment SSs

| Measure | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
|-------------------------------------|------|-------|-------|-------|-------|-------|---|
| Children with WS (<i>n</i> = 76) | | | | | | | |
| 1. CA | - | | | | | | |
| 2. GFTA-2 SS | .13 | - | | | | | |
| 3. DAS-II PhP SS | .08 | .60** | - | | | | |
| 4. DAS-II NVR SS | -.19 | .29 | .62** | - | | | |
| 5. DAS-II DigFwd SS | .04 | .47** | .56** | .29 | - | | |
| 6. DAS-II SA SS | .13 | .49** | .66** | .71** | .33* | - | |
| 7. Composite Vocabulary SS | -.08 | .54** | .74** | .65** | .49** | .65** | - |
| Children with Dup7 (<i>n</i> = 30) | | | | | | | |
| 1. CA | - | | | | | | |
| 2. GFTA-2 SS | .22 | - | | | | | |
| 3. DAS-II PhP SS | .22 | .57* | - | | | | |
| 4. DAS-II NVR SS | .02 | .52* | .60** | - | | | |
| 5. DAS-II DigFwd SS | -.05 | .44 | .65** | .70** | - | | |
| 6. DAS-II SA | -.10 | .55* | .48* | .60** | .70** | - | |
| 7. Composite Vocabulary SS | .03 | .48* | .55* | .73** | .59* | .76** | - |

Note. Composite Vocabulary SS = ([Peabody Picture Vocabulary Test-4 SS + Expressive Vocabulary Test-2 SS] / 2); DAS-II = *Differential Ability Scales-II* (Elliott, 2007); DigFwd = Recall of Digits-Forward; Dup7 = 7q11.23 duplication syndrome; GFTA2 = *Goldman-Fristoe Test of Articulation-2* (Goldman & Fristoe, 2000); NVR = Nonverbal reasoning ability cluster; PhP = Phonological Processing SS; SA = Spatial ability cluster; SS = standard score; WS = Williams syndrome.

p* < .01. *p* < .001.

Multiple Regression Analyses

A hierarchical multiple regression was computed for children with WS to determine if articulatory accuracy contributed unique variance to phonological processing over and above that contributed by four cognitive and linguistic variables previously shown to be important to the development of phonological processing (see Chapter I). Assumptions of the regression were met. Results of the analyses are presented in Table 21. Model 1 explained significant variance in phonological processing, $F(4, 71) = 46.09$, $p < .001$, adj. $R^2 = .71$. Articulatory accuracy, added in Model 2, contributed significantly and uniquely ($f^2 = .11$) to the variance in phonological processing beyond the contributions of verbal short-term memory, nonverbal reasoning, spatial ability, and composite vocabulary, $F(5, 70) = 41.86$, $p < .001$, adj. $R^2 = .73$; $R^2\Delta = .027$, $p = .007$.

Discussion

Based on information discussed in Chapter I, variables were included in Study 2 that have been shown previously to be significantly related to phonological processing. These included age and the standardized measures of articulatory accuracy, verbal short-term memory, composite vocabulary, spatial ability, and nonverbal reasoning ability (Ehri, Nunes, Willows, Schuster, Yaghoub-Zadeh, & Shanahan, 2001; Overby et al., 2012; Scarborough, 1998a, 1998b; Torgesen & Davis, 1997). Descriptive statistics and tests for differences between groups showed that both groups of children obtained median GFTA-2 SSs below the range expected for children in the general population. Recall that all children were enrolled in speech services at the time of the study or had had speech intervention services in the past. The children with WS articulated consonants

Table 21
Multiple Regression Analysis Predicting DAS-II Phonological Processing Standard Scores for Children with WS

| | b_i | $SE \beta_i$ | t | p -value | Semi-partial r | Cohen's f^2 ^a |
|---|-------|--------------|------|------------|------------------|----------------------------|
| Model 1 | | | | | | |
| DAS-II Digits Forward SS | .348 | .075 | 4.50 | < .001 | .28 | .31 |
| DAS-II Nonverbal Reasoning SS | .141 | .101 | 1.39 | .168 | .08 | .03 |
| DAS-II Spatial Ability SS | .235 | .091 | 2.58 | .012 | .29 | .09 |
| Composite Vocabulary SS | .332 | .103 | 3.22 | .002 | .20 | .15 |
| Model 2 | | | | | | |
| DAS-II Digits Forward SS | .296 | .074 | 4.03 | < .001 | .24 | .23 |
| DAS-II Nonverbal Reasoning SS | .172 | .098 | 1.77 | .082 | .11 | .04 |
| DAS-II Spatial Ability SS | .186 | .089 | 3.00 | .040 | .14 | .06 |
| Composite Vocabulary SS | .244 | .113 | 2.35 | .022 | .14 | .08 |
| Goldman-Fristoe Test of Articulation-2 nd Edition SS | .215 | .078 | 2.77 | .007 | .17 | .11 |

Note. $n = 76$. Variables were converted to z-scores prior to analysis. Composite Vocabulary SS = ([Peabody Picture Vocabulary Test-4 SS + Expressive Vocabulary Test-2 SS] / 2); DAS-II = *Differential Ability Scales-II* (Elliott, 2007); SS = standard score; WS = Williams syndrome.

^aCohen (1988); f^2 for predictors: $(R^2 \text{ inclusive model} - R^2 \text{ restricted-variable model}) / (1 - R^2 \text{ inclusive model})$.

with significantly greater accuracy and were significantly more aware of phonological information than were children with Dup7. Children with Dup7 had significantly higher nonverbal reasoning ability, spatial ability, and vocabulary abilities than did the children with WS.

As expected, all correlations among the standardized variables included in Study 2 were positive both for the children with WS and for the children with Dup7, and almost all were statistically significant. However, for the children with WS, two correlations were not statistically significant: one between nonverbal reasoning SS and articulatory accuracy SS and one between nonverbal reasoning SS and verbal short-term memory SS. The effect size for these correlations was small. For the children with Dup7, only the correlation between articulatory accuracy SS and verbal short-term memory SS was not statistically significant. Although this correlation was not statistically significant, the effect size was moderate suggesting that had the sample size been larger, the correlation could have resulted in statistical significance.

The pattern of statistically significant correlations involving phonological processing found for both children with WS and children with Dup7 is consistent with patterns previously reported for children who are developing typically (i.e., phonological awareness: McDowell et al., 2007; phonological awareness: Overby et al., 2012; phonemic awareness: Parrila et al., 2004). Specifically, findings from studies of children in the general population have indicated that phonological processing ability is significantly related to verbal short-term memory (Hintze, Ryan, & Stoner, 2003; Wagner et al., 1997; see also National Reading Panel, 2000), vocabulary (Dickinson, McCabe, Anastasopoulos, Peisner-Feinberg, & Poe, 2003), and spatial ability (Krajewski &

Schneider, 2009). The pattern of statistically significant correlations involving articulatory accuracy, found for both children with WS and children with Dup7, is consistent with patterns previously reported for children who are developing typically (i.e., nonword repetition: Gathercole, Service, Hitch, Adams, & Martin, 1999; phonological processing skills: McDowell et al., 2007; and phonological awareness or single-word reading: Overby et al., 2012).

The hierarchical multiple regression modeling procedure was chosen to explain whether the incremental effect of articulatory accuracy, which was added in the second model, contributed uniquely to variance in phonological processing over and above that contributed by the cognitive and linguistic variables for children with WS. In Model 1, a large amount of shared variance was explained in the standardized measure of phonological processing (adj. $R^2 = .71$). In Model 2, the incremental effect of the standardized measure of articulatory accuracy contributed positive, significant, and approximately 2.9% unique variance to phonological processing beyond that which was contributed by the same cognitive and linguistic variables included in Model 1. Although the increase in shared variance explained by the addition of articulatory accuracy was significant ($p = .007$), the unique effect was small ($f^2 = .11$; Cohen, 1988).

In the full model, verbal short-term memory contributed significantly to phonological processing, adding approximately 5.8% unique variance. Also, spatial ability and composite vocabulary each contributed 2.0% unique variance to phonological processing over and above that contributed by articulatory accuracy, verbal short-term memory, vocabulary, and nonverbal reasoning. Unexpectedly, nonverbal reasoning

ability did not contribute significant variance to phonological processing in either the full Model 2 or the reduced Model 1.

The regression result showing that articulatory accuracy made a significant contribution to phonological information processing for children with WS fits with the hypothesized relation between phonological processing and articulatory accuracy examined in Overby et al. (2012). These authors found that articulatory accuracy assessed in the Fall of kindergarten explained significant variance in second-grade single-word reading (for children who had not been exposed to explicit phonological awareness instruction in school) and that articulation's early moderate contribution was in part mediated by a factor score based on component phonological awareness tasks administered by emaniners in the Spring of first grade. Letter knowledge was a strong contributor in this model while nonverbal intellectual ability and receptive and expressive vocabulary explained minimal variance.

CHAPTER IV

GENERAL DISCUSSION

The purpose of the present dissertation was to examine and characterize speech articulatory accuracy for children with WS and children with Dup7. The project involved the first systematic examination of articulatory accuracy for children with these syndromes. The strategy for characterizing articulation for the children was to administer a standardized citation-method assessment (GFTA-2) and then to compare both SSs and proportion correct for specific features of articulation (ACCP, position in words, place and manner of articulation, and movement transition across consonants in clusters) for the full sample of children in each syndrome group and for younger and older subgroups.

The result of articulatory assessment showed the SSs obtained by children with WS and by children with Dup7 were significantly lower than expected for same aged children in the general population. I found that 56% of children with WS and 80% of children with Dup7 obtained SSs below the expected range for typical performance. The proportion correct of GFTA-2 consonant items correctly produced by children in the younger subgroups was significantly lower than that obtained by children in the respective older subgroups. This finding was important because it showed overall consonant accuracy was better for the older children with WS or Dup7 as it is also for older children in the general population (Goffman & Smith, 1999; Goldman & Fristoe, 2000; Smit et al., 1990; Smith & Zelaznik, 2004; Templin, 1957).

Reports from the literature on articulatory acquisition indicated that children with IQs of at least 70 had more accurate articulation than did children with IQs below 70 (Winitz, 1959). Test results were consistent with this assertion. For both children in the WS group and in the Dup7 group, children with IQs at or above 70 earned significantly higher GFTA-2 SSs than did children with IQs below 70. In addition, correlational analyses indicated a strong association between articulation and overall intellectual ability for the group of children with WS and a moderately strong association for the group of children with Dup7. These positive and statistically significant findings point to the need for further research regarding the influence of cognition across the full trajectory of articulatory development.

An important expectation of the dissertation, both for children with WS and for children with Dup7, was that all standardized variables in Study 2 would be significantly correlated. With few exceptions, this expectation was supported by the data. For both groups of children, the correlation between articulatory accuracy and phonological processing was the strongest, although the significant results showed articulatory accuracy, overall cognitive ability, spatial ability, and the combined factor for lexical understanding and use were all moderately, to strongly, related. Interestingly for the children with WS, nonverbal reasoning was not correlated significantly with either articulatory accuracy or verbal short-term memory. For the children with Dup7, articulatory accuracy and verbal short-term memory were not correlated significantly; although this correlation with verbal short-term memory was not statistically significant, the value of effect was moderate and was almost identical to the statistically significant value obtained for the WS group. The lack of statistical significance between articulatory

accuracy and verbal short-term memory for the Dup7 group could be due to the relatively small sample size or to other factors not included in the study. This result indicates further study is needed regarding both the nature of articulatory development and the cognitive-linguistic factors that contribute to phonological processing for children with Dup7.

A final expectation of the dissertation was that GFTA-2 SS would explain significant and unique variance in DAS-II Phonological Processing SS over and above that contributed by DAS-II Recall of Digits Forward SS, DAS-II Spatial Ability SS, Composite Vocabulary SS, and DAS-II Nonverbal Reasoning SS. Results of an hierarchical multiple regression model for children with WS suggested that speech sound accuracy plays a unique and statistically significant part in its development, as do the remaining Study 2 variables, except for nonverbal reasoning. The strong and unique contribution made by the DAS-II Recall of Digits Forward subtest SS was an anticipated finding given information regarding the strength of this ability for children with WS (Mervis, 2009). An unexpected finding was that nonverbal reasoning was a very minimal and non-significant contributor to the variance explained by the model. One might surmise that children must use analytical and inductive reasoning processes to complete explicit phonemic awareness tasks, such as elision. This result suggests there is an urgent need to address phonological information processing for children with WS using a measure that probes the ability in a deeper way.

Other research has implicated articulatory ability as a factor important in the development of early literacy for both children developing typically (McDowell et al., 2007; Overby et al., 2012) and for children with speech sound disorder (Lewis et al.,

2006). Research reviewed above showed that word learning and phonological coalescence supports the development of implicit phonological knowledge which, in turn, supports the expansion of the lexical repertoire. In Study 2, both speech sound accuracy and vocabulary ability explained significant variance in phonological processing. Thus, in ways similar to relations among these variables for children in the general population, the multiple regression results showed that for children with WS the phonological domains of processing and production are interrelated with the domains of receptive and expressive lexical ability and that these abilities are supported, in large measure, by verbal short-term memory.

Other analyses in the dissertation explored specific aspects of speech articulation as a function of features of articulation and the constituent factors of each feature. The features explored included ACCP (ASHA, 2017), accuracy as a function of consonant position-in-words (Smit et al., 1999; Templin, 1957), and accuracy as a function of targeting specific parameters of consonant articulation: place-of-articulation, manner-of-articulation (Smit et al., 1999; Templin, 1957), and transitional movement across consonant sequences (Hayden & Square, 1999). For each of these analyses, the patterns of proportion of consonants correct for both the WS group and the Dup7 group were generally consistent with patterns of accuracy previously reported for children in the general population. However, there were some notable differences, such as delayed acquisition of accuracy for consonant articulation and highly variable performance among individuals within the groups (both younger and older subgroups).

Findings consistent with expected patterns of articulatory development for children in the general population include the following. Both for the children with WS

and for the children with Dup7, articulation was significantly more accurate for consonants that are known to be acquired early in the development of articulation. This was in the presence of results for lesser accuracy for consonants that are expectedly acquired in the middle period and of results for the least accuracy for consonants that are expectedly acquired in the late period of articulatory development. Several authors have investigated potential reasons why final consonants, articulated by children who are typically developing, are less accurate than initial consonants. Some obvious reasons include perception difficulty, production difficulty, or both. Archer, Zamuner, Engel, Fais, and Curtin (2015) demonstrated that twelve month-old infants who are developing typically were able to perceive voiced stops but not unvoiced stops in the coda position. Redford and Diel (1999) showed that college students identified final consonants with less accuracy than initial consonants when targets were embedded in naturally occurring frame sentences and structured as CVC words. Thus, there is literature to support the conclusion that children who are typically developing are able to perceive highly salient differences in phonetic contrasts in the final word position but that both perception and production processes are likely to contribute to production accuracy for coda consonants that are articulated in natural speaking contexts (Edwards, Fox, & Rogers, 2002). For children with WS, one small word-learning study has shown evidence that children with WS successfully perceive and remember consonant-feature contrasts (Havy, Moukawane, & Nazzi, 2010). In this study, feature contrasts were studied only for initial consonants and vowels in CVC or CV.CV word forms. No literature has been found suggesting children with WS accurately (or inaccurately) perceive consonants in the coda position.

As reported in seminal normative studies (Goldman & Fristoe, 2000; Smit et al., 1999; Stoel-Gammon, 1987; Templin, 1957), children who are typically developing articulate consonants at word onsets and codas with respectively greater or lesser degrees of accuracy. Templin noted a clear separation in accuracy for consonants articulated at the beginning of words compared to those articulated at the end of words by children learning English and who were developing typically. She reported this for every age tested from 3 years until 7 years, after which time the differences in her data were minimal. In the present dissertation, analyses were computed to examine articulatory accuracy in the initial position and in the final position of words. Ingram (1979) suggested variability in accuracy “is motivated by the tendency for younger children to simplify” (p. 139). No significant difference in the distributions of median proportion correct for position-in-word was found for any group with WS. However, for the children in the older subgroups (CA: 10–17 years) median proportion correct was somewhat lower for consonants in the final word position. It is reasonable to conclude that word position is not a key factor impacting the consonantal accuracy of children with WS. (*NB*: The analysis excluded medial consonant articulation.)

However, for the children with Dup7, significant differences were found in the distributions of proportion correct as a function of consonants articulated in the initial or final word position. The patterns fit Templin’s reported pattern: higher accuracy through development, until the age of mastery, for consonants in the initial word position compared to the final word position. Importantly, for 25% of the children in the younger subgroup with Dup7, accuracy at both the initial and the final word position was less than 50%. One conclusion from the results for the children with Dup7 is that articulation for

these children develops in accord with the typical word-position pattern but at a delayed pace.

The findings from tests for differences across factors of articulatory place-of-production revealed children in both groups were significantly more accurate articulating consonants produced anteriorly at the lips or posteriorly at the velum or in the glottal area than they were articulating consonants in the central oral areas (at the teeth, alveolar ridge, or palate). This finding is consistent with Templin's norms regarding acquisition for "consonant elements" as a function of word position (1957, p. 51; see Discussion, Chapter II). Although some experts (Dyson, 1988; Stoel-Gammon, 1987) have implicated alveolar consonants (e.g., /t/) emerge early in development and thus they would be expected to be articulated with equal accuracy as bilabial consonants (e.g., /b/), the results suggested manner of articulation affected accuracy at different places of articulation. Overall the results showed that alveolar stops such as /t, d/ and the nasal /n/ were more accurate than alveolar fricatives such as /s, z/ and the alveolar approximant /l/. As reviewed earlier, for many fricatives, and for some approximants, articulatory load is greater. This is due to greater articulatory precision required for their accurate execution (see description in Stevens, 1998).

Regarding the tests computed for accuracy as a function of articulatory manner-of-production, the patterns of significant post hoc differences in proportion of consonants correct for both the WS group and the Dup7 group fit the typical pattern of greater accuracy for nasal and stop consonants (e.g., /m/ and /d/, respectively) and lesser accuracy for fricative and approximant consonants (e.g., /s/ and /l/, respectively; Smit et al., 1990). Importantly, a review of the raw data was consistent with what was suggested

in the *IQR* spreads for both manner and place of articulation. All children in the older subgroups with WS or Dup7 accurately articulated the following classes: all nasal consonants; all, or all but one, of the bilabial and velar-glottal consonant(s), and nearly all, or all but one or two, of the 18 stop consonants. The patterns of proportion of consonants correct for place and manner articulation was the same for children in the younger groups; however, accuracy was much lower. A clear conclusion from these results is that, overall, articulation of nasal and stop consonants required less precision than did the fricative and approximant consonants. The extant literature supports this impression also for children in the general population. For example, consider the Standard American English approximant consonant, /ɹ/ (e.g., her, sir, fur). The consonant has required complex description with respect to its manner of articulation (Stevens, 1998) and no single place in the central oral area has been shown where all individuals articulate it (Kent, 2013). Furthermore, speakers have been shown to use several different tongue configurations for /ɹ/ articulation (ASHA, 2017). Lastly, in typical development, articulatory experience over several years of speaking is needed before an accurate /ɹ/ is learned across all speaking contexts.

The expectation of the dissertation for children's accuracy articulating double consonants (clusters and affricates) was based on ACCP; children in both groups were expected to accurately articulate the GFTA-2 double consonants as indicated in Table 1. Not surprisingly, the pattern of results for proportion of double consonants correct across articulatory planes-of-movement indicated no significant differences across the factors (same place, front-to-back, and back-to-front) for children in either the older subgroup with WS or the older subgroup with Dup7. However, children in both younger subgroups

were significantly more accurate articulating double consonants at the same place than they were articulating across directional planes of movement, either front-to-back or back-to-front. Importantly, for at least one-fourth of the younger children with Dup7, no attempt to articulate a double consonant was accurate. Also for these younger children with Dup7, at least half were not able to accurately articulate double consonants requiring front-to-back transition by the articulators. One clear conclusion from this finding is that younger children with Dup7 have issues with speech motor control.

Implications

Overall, the results of the dissertation suggest English-speaking younger and older children with WS or Dup7 articulate consonants with varying degrees of accuracy. Individual variability was reflected in the wide ranges of GFTA-2 SSs that are presented, for example, in the descriptive statistics of Table 10. Furthermore, patterns of accuracy showing high or low center values, and relatively narrow or wide spread of the middle 50% of the data, were tabulated and described for sets of consonants within each feature of articulation (class). This variability is evident in the descriptive statistics presented in Tables 13, and Tables 15–18. The patterns of accuracy, reflecting development of articulation for children in the younger and older subgroups and examined using the Friedman ANOVAs, were generally consistent with patterns of articulatory development previously reported in the literature for younger children who are developing typically, with noted exceptions.

The findings from the dissertation give direction for interventionists and researchers interested in facilitating improvements in consonant articulation for children with these syndromes. Goal selection based on patterns of accuracy as a function of

features of articulation has utility because a child's accuracy across the factors of each feature indicates the child's knowledge about articulating consonants and the child's movement competency for executing that knowledge when speaking.

From the dissertation results shown for patterns of accuracy as a function of articulatory features, there appear to be many children with WS or Dup7 who were more accurate producing consonants that (a) are typically acquired very early in development, (b) occur in the initial position in words, (c) are articulated at the extremes of the mouth (with mixed results for alveolars depending on manner of articulation), (d) involve nasality or stopping the breath stream, and (e) especially for the younger children with Dup7, are clustered at the same place of articulation. Based on information in Table 1, in Appendix A, and reviewed in Chapter I, singleton consonants that fit these parameters include, /b, p, t, d, w, m, n, k, g, h, f/ and the GFTA-2 consonant cluster that fits these parameters is /kw/. It is suggested that the professional's knowledge of the child's patterns of articulatory accuracy as a function of features of articulation is useful for determining needs for frequency and intensity of treatment; informing decisions related to prioritizing treatment goals; for selecting for treatment either specific consonant targets or multiple consonants with an indirect emphasis on the features of consonant articulation (consonant classes); and for providing particular schedules of feedback during treatment (Maas, Gildersleeve-Newman, Jakielski, & Stoeckel, 2014). For example, if careful assessment indicates that a child with either syndrome has issues of speech motor control and, as a result, misarticulates some of the early consonants, an intervention program might address remediation for multiple consonants in this group of phonemes in functional ways.

In the first few treatment sessions, the therapist might consider organizing treatment targets in lists of words embedded in phrases that have the targets arranged in both in the initial or final word position. Consider ACCP for prioritizing items in the early stage of treatment. To highlight the features of articulation, first address the most visible and salient consonants that are likely accurate (or at least stimulable) and typically are developed earlier (e.g., the voiced bilabial stop and the nasal /m/). Do this before adding words to the list of select phrases that consist of less visible, less salient consonants (e.g., voiceless bilabial and the palatal approximant /j/) but also likely to be accurately articulated. The relevant research indicates that from early in the trajectory of articulatory development, children develop phonological competence through attention to and experience with the lexicon and by bootstrapping patterns of familiar word forms to less familiar forms with the goal to reduce articulatory load (Vihman & Velleman, 2002). A next step would be to have children participate in activities using target words in meaningful phrases and short sentences that expose them to similarities among articulatory features of place and manner. Alliteration and rhyming activities come to mind. Treatment should involve abundant opportunities for repeating targets and including regular recycling through previously addressed targets. If some targets prove difficult to produce in simple, continuous speech contexts, brief periods of focused attention to the targets in isolation should be considered. For this work and for all articulation treatment, principles of motor learning embedded in multisensory practice should be followed (touch cues, mirrors for visual feedback, and focused auditory feedback through devices such as a “Talk-Back” toy or a microphone with speakers on low volume). (*NB*: Basic tenets of the principles of motor learning are found in Maas,

Robin, Austermann Hula, Freedman, Wulf, Ballard, & Schmidt, 2008). As children gain some ability to produce consonants in words in phrases across the consonant classes, children should have treatment experiences that permit self-monitoring for accuracy in phrases formed with less consistent phonotactic patterns and with specific feedback regarding accuracy. Feedback should be provided by the therapist on no greater than 60% of target trials (Mass et al., 2014). It might be productive to follow focused practice with multisensory articulation activities as described above and routinely probe to document progress by having children recite short lists of words with targets in various word positions embedded in novel sentences (for children unable to read, picture description is advised). Clinical evidence is needed in support of these suggestions for children with these syndromes.

Suggestions follow in the wake of the present findings, of Huffman et al. (2012), of Velleman et al. (2013), and of Mervis et al. (2015). Results from the latter three studies showed that children with WS or Dup7 made articulatory and other speech errors which reflected issues of speech motor control. Many children in Mervis et al. (2015) were identified with CAS and/or dysarthria, or showed symptoms of these. The diagnoses cued examiners to the potential for speech-motor disorder. Commercially available programs and published intervention strategies have been designed with principles of motor learning embedded in multisensory treatment plans. Some commercial programs include Word FLIPS (Granger, 2005), Moving Across Syllables (Kirkpatrick, Stohr, & Kimbrough, 1990), and Speech Therapy for Apraxia (Blue Whale Apps Inc., 2017). Examples of peer-reviewed published programs include (a) the PROMPT Conceptual Framework and the Motor-Speech Hierarchy (Hayden, 2004; Hayden & Square, 1994,

1999), and (b) Rapid Syllable Transitions treatment for apraxia of speech (Murray, McCabe, & Ballard, 2015; Thomas, McCabe, & Ballard, 2014). Velleman and Vihman (2002) and Velleman (2016) have organized assessment, appropriate therapy goals, and explicit intervention strategies for addressing various phonotactic limitations. These intervention strategies and techniques have proven to be valid extensions to standard articulation therapy. Therapists will find that the methods directly address the absence of consonants in the final word position and the difficulty children experience when articulating clusters or multisyllabic words.

Results of Study 2 indicated that articulatory accuracy is closely associated with phonological processing, intellectual abilities, and lexical abilities. Literature reviewed showed that phonological processing is intimately associated with success learning to read (National Reading Panel, 2000). Moriarity and Gillon (2006). Therefore, interventionists must assess also the phonological abilities of children with speech motor disorder and articulatory inaccuracies (Bernthal, Bankson, & Flipsen, 2013). If thorough assessment indicates children with WS or Dup7 have poor phonological processing abilities, then the intervention protocol recommended herein should be expanded to incorporate explicit phonological awareness tasks such as games requiring phoneme blending, word segmentation, elision (dropping syllables and sounds from larger words with the result of smaller words and placing emphasis on articulatory targets), and phonological judgment activities (Carson, Gillon, & Boustead, 2013; Gillon, 2000, 2005, 2017; Sutherland & Gillon, 2005). Activities such as these also tax verbal short-term memory and stimulate lexical abilities. One commercially available program, the Lindamood Phoneme Sequencing Program for Reading, Spelling, and Speech (LiPS) is

well-supported in the literature (Murdaugh, Maximo, Cordes, O’Kelley, & Kana, 2017; Sadoski & Wilson, 2006) and might prove beneficial to time-strapped therapists in need of published intervention materials. LiPS is designed to teach children to speak accurately using oral-motor, visual, and auditory feedback information while simultaneously training them to identify and describe the phonemes in syllables and words. These are skills that children need to decode written words and to identify individual speech sounds and clusters in words for spelling accurately.

Limitations of the Research

The major limitation of the dissertation was the small sample size of children with Dup7, although the 50-child sample was comparably larger than all known studies conducted outside the NSL to date. Furthermore, the study was cross-sectional. A longitudinal design would provide clearer information regarding the trajectory of articulatory development and clearer indications of potential individual differences within and between groups.

A second limitation was that the assessment of articulatory accuracy was based only on perceptual measurement. Perceptual impressions of phoneme quality are known to vary among individuals (Cucchiarini 1996; Shriberg & Lof 1991), even among listeners well-trained to transcribe the sounds of languages. To counter this limitation, considerable effort was devoted to carefully operationalizing the procedures for coding the consonants and to ensuring reliability among judges.

Lastly, Study 1 was restricted in scope for examining articulatory accuracy as a function of the features of articulation because the standardized assessment that was used assessed articulations of only a single exemplar for each item. Even with this inherent

limitation of the instrument, the GFTA-2 is psychometrically sound and is well-accepted by researchers and clinicians alike. Several reports have documented validity for using it (Eisenberg & Hitchcock, 2010).

Future Directions

The present dissertation was the first to characterize the capacity for articulatory accuracy for children with WS or Dup7 and to show that for children with WS, articulatory accuracy predicted unique variance in phonological processing. However, the full story is incomplete. Many children with WS or Dup7 find that learning to read is challenging. Given the cross-sectional design of the present study, it is crucial to develop a longitudinal study of construct relations that facilitate the literacy skills of children with these syndromes. However, before conducting longitudinal research, a larger sample of children with Dup7 must be recruited so as to complete the current investigation of the relation between articulatory accuracy and phonological processing.

It would benefit interventionists and families of children with WS or Dup7, and the children themselves, if research was done regarding predictive relations between articulatory accuracy and single-word reading ability. Recent research on young school-age children who were developing typically indicated phonological processing mediated the effects of articulatory accuracy on reading and spelling (Overby, et al., 2012). The importance of understanding the relations between articulatory accuracy, phonological processing, and single-word reading for children with WS or Dup7 cannot be understated. The end goal of this work is to customize and refine literacy instruction by capitalizing on the children's characteristic strengths.

Second, for both children with WS and children with Dup7, the perceptual assessment of articulatory accuracy should be followed up with (a) instrumental assessment of speech and voice characteristics and (b) comprehensive behavioral assessments for determining (b.1) underlying speech motor abilities (structure and function of the speech production mechanisms), (b.2) articulatory accuracy in continuous speech, (b.3) the trajectory of phonological error patterns over time, (b.4) the full range and trajectory of phonological information processing, and (b.5) omnibus measurement of receptive and expressive language ability. Following the completion of the full range of speech and language baseline assessments, study regarding interrelations among these variables should be conducted.

Lastly, the development of potential profiles of the full range of speech production abilities for children with these syndromes should be created (see Kent & Vorperian, 2013). Speech production should be examined as a function of five major areas of performance: speech motor skills, voice, speech sounds in context, fluency, and prosody. Such information would greatly support interventions devoted to determining best practice methods for improving expressive communication ability.

Summary and Conclusion

In the present dissertation I considered speech articulatory accuracy within a neuroconstructive view of speech motor cognition. I asked whether there were differences in consonant articulatory accuracy, as produced in single words by children with WS or Dup7, compared to that expected for children in the general population. I sought also to characterize any differences that I discovered both between and within the groups of children, and then to follow these analyses with specific examination of

accuracy as a function of features of articulation. My final goals were to investigate relations among articulatory accuracy, cognitive ability, phonological processing, and vocabulary abilities for the children with these syndromes.

I have shown that speech articulatory accuracy for the groups of children with WS or Dup7 remained delayed well into middle childhood for more than half of children with WS, and for more than two-thirds of children with Dup7. For some of the children, inaccuracies in articulation persisted well into adolescence. Overall, consonants were acquired through development according to the same temporal pattern as shown for children in the general population, but with delay and with strikingly variable individual accuracy (see Overby et al., 2012 for standard deviations reported with descriptive statistics for children who were developing typically). The dissertation findings revealed also that double consonants (initial clusters and affricates) were very difficult for younger children with Dup7 to articulate.

As a group, children with WS had better ability to accurately articulate than did children with Dup7. Contrastively, children with Dup7 had higher IQs than did children with WS. For the children with WS, the findings confirmed expectations that the older children, and the children with higher IQs, articulated with greater accuracy. Differently from the group with WS, older children with Dup7 articulated also with greater accuracy compared with the younger children with Dup7, but only half of the children with higher IQs obtained higher articulation scores.

The findings showed also that for both groups, articulatory accuracy was correlated with overall intellectual ability, phonological processing, spatial ability, and composite vocabulary. This outcome was consistent with previous reports of these

relations for children in the general population. The dissertation findings showed that for the children with WS, articulatory accuracy contributed significant and unique variance to phonological processing over and above that contributed uniquely by the cognitive and linguistic variables.

In conclusion, the results of this dissertation contributed to a deeper understanding of the nature and the characteristics of speech articulation for children with WS or Dup7. The findings explicated positive relations among articulatory accuracy, intellectual ability, phonological processing, and vocabulary abilities for these children.

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Appendix A

THE INTERNATIONAL PHONETIC ALPHABET (revised to 2015)

CONSONANTS (PULMONIC)

© 2015 IPA

| | Bilabial | Labiodental | Dental | Alveolar | Postalveolar | Retroflex | Palatal | Velar | Uvular | Pharyngeal | Glottal |
|---------------------|------------|-------------|------------|------------|--------------|------------|------------|------------|------------|------------|------------|
| Plosive | p b | | | t d | | ʈ ɖ | c ɟ | k ɡ | q ɢ | | ʔ |
| Nasal | m | ɱ | | n | | ɳ | ɲ | ŋ | ɴ | | |
| Trill | ʙ | | | r | | | | | ʀ | | |
| Tap or Flap | | ⱱ | | ɾ | | ɽ | | | | | |
| Fricative | ɸ β | f v | θ ð | s z | ʃ ʒ | ʂ ʐ | ç ʝ | x ɣ | χ ʁ | ħ ʕ | h ɦ |
| Lateral fricative | | | | ɬ ɮ | | | | | | | |
| Approximant | | ʋ | | ɹ | | ɻ | j | ɰ | | | |
| Lateral approximant | | | | l | | ɭ | ʎ | ʟ | | | |

Symbols to the right in a cell are voiced, to the left are voiceless. Shaded areas denote articulations judged impossible.

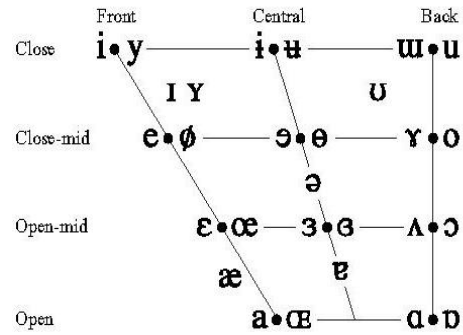
CONSONANTS (NON-PULMONIC)

| Clicks | Voiced implosives | Ejectives |
|---------------------------|--------------------------|------------------------------|
| ɔ Bilabial | ɓ Bilabial | ʼ Examples: |
| ɠ Dental | ɗ Dental/alveolar | pʼ Bilabial |
| ǀ (Post)alveolar | ɟ Palatal | tʼ Dental/alveolar |
| ǃ Palatoalveolar | ɠ Velar | kʼ Velar |
| ǂ Alveolar lateral | ʙ Uvular | sʼ Alveolar fricative |

OTHER SYMBOLS

| | |
|--|---|
| ʍ Voiceless labial-velar fricative | ç ʝ Alveolo-palatal fricatives |
| ʋ Voiced labial-velar approximant | ɺ Voiced alveolar lateral flap |
| ɥ Voiced labial-palatal approximant | ɥ Simultaneous ʃ and x |
| ħ Voiceless epiglottal fricative | Affricates and double articulations can be represented by two symbols joined by a tie bar if necessary. |
| ʕ Voiced epiglottal fricative | |
| ʡ Epiglottal plosive | |

VOWELS



Where symbols appear in pairs, the one to the right represents a rounded vowel.

SUPRASEGMENTALS

| | |
|-------------------------|---------------------------|
| ˈ Primary stress | ˌ Secondary stress |
| ː Long | ̥ Shortening |

t͡s k͡p

ˈfounəˈtʃən

*Source: International Phonetic Association. (2016). *The international phonetic alphabet*. Retrieved from www.InternationalPhoneticAlphabet.org

Appendix B

extIPA SYMBOLS FOR DISORDERED SPEECH (Revised to 2008)

CONSONANTS (other than on the IPA Chart)

| | bilabial | labiodental | dentolabial | labioalv. | linguolabial | interdental | bilabial | alveolar | velar | velophar. |
|--------------------------|----------|-------------|-------------|-----------|--------------|-------------|----------|----------|-------|-----------|
| Plosive | | p b | p̄ b̄ | p b | t d | t̄ d̄ | | | | |
| Nasal | | | m̄ | m | n | n̄ | | | | |
| Trill | | | | | r | r̄ | | | | |
| Fricative median | | | f̄ v̄ | f v | θ ə | θ̄ ə̄ | h̄ h̄ | | | ɣ |
| Fricative lateral+median | | | | | | | | ɬ ɮ | | |
| Fricative nasal | m̃ | | | | | | | ɳ | ŋ | |
| Percussive | ʘ | | | | | | ɸ | | | |
| Approximant lateral | | | | | l̥ | l̥̄ | | | | |

Where symbols appear in pairs, the one to the right represents a voiced consonant. Shaded areas denote articulations judged impossible.

DIACRITICS

| | | | | | | | | |
|----|----------------------|----|----|-------------------------|----------|----|-------------------------|----|
| ◌̠ | labial spreading | ◌̠ | ◌̠ | strong articulation | ◌̠ | ◌̠ | denasal | ◌̠ |
| ◌̡ | dentolabial | ◌̡ | ◌̡ | weak articulation | ◌̡ | ◌̡ | nasal escape | ◌̡ |
| ◌̢ | interdental/bidental | ◌̢ | ◌̢ | reiterated articulation | P̣ P̣ P̣ | ◌̢ | velopharyngeal friction | ◌̢ |
| ◌̣ | alveolar | ◌̣ | ◌̣ | whistled articulation | ◌̣ | ◌̣ | ingressive airflow | ◌̣ |
| ◌̤ | linguolabial | ◌̤ | ◌̤ | sliding articulation | ◌̤ | ◌̤ | egressive airflow | ◌̤ |

CONNECTED SPEECH

| | |
|--|---------------------------------------|
| () | short pause |
| (.) | medium pause |
| (...) | long pause |
| f | loud speech [(<i>f</i> loud)] |
| ff | louder speech [(<i>ff</i> louder)] |
| p | quiet speech [(<i>p</i> quieter)] |
| pp | quieter speech [(<i>pp</i> quieter)] |
| <i>allegro</i> | fast speech [(<i>allegro</i> fast)] |
| <i>lento</i> | slow speech [(<i>lento</i> slow)] |
| <i>crescendo, raientando</i> , etc. may also be used | |

VOICING

| | | |
|----|---------------------------|----|
| ◌̥ | pre-voicing | ◌̥ |
| ◌̦ | post-voicing | ◌̦ |
| ◌̧ | partial devoicing | ◌̧ |
| ◌̨ | initial partial devoicing | ◌̨ |
| ◌̩ | final partial devoicing | ◌̩ |
| ◌̪ | partial voicing | ◌̪ |
| ◌̫ | initial partial voicing | ◌̫ |
| ◌̬ | final partial voicing | ◌̬ |
| ◌̭ | unaspirated | ◌̭ |
| ◌̮ | pre-aspiration | ◌̮ |

OTHERS

| | | | |
|------------------|---|----|--|
| ◌̯, (◌̰), (◌̱) | indeterminate sound, consonant, vowel | ◌̲ | Velodorsal articulation |
| (◌̳), (◌̴), (◌̵) | indeterminate voiceless plosive, nasal, etc | ◌̶ | sublaminal lower alveolar percussive click |
| (◌̷) | silent articulation (◌̸), (◌̹) | ◌̺ | alveolar and sublaminal clicks (cluck-click) |
| (◌̻) | extraneous noise, e.g. ((2 sylls)) | * | sound with no available symbol |

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Appendix C

- I. Procedural rules for determining continuous speech PCC used in the present dissertation follow.
 - a. Principal guidance obtained from Shriberg and Kwiatkowski [1982, p.267].
 - b. Particular modifications obtained from SALT 2012 Research version (Miller & Iglesias, 2012).
 - c. Particular modifications obtained from NSL transcription guidelines.
 - d. Orthographic transcripts will be reevaluated for number of words after standard NSL transcription reliability has been confirmed by the lab manager.
 - i. Two trained transcribers, or consensus coders, will reevaluate all utterances in each participant's transcript.
 - ii. Transcripts will be collated prior to reevaluation with random ordering relative to age, sex, and diagnosis.
 - e. Consensus coders qualifications
 - i. The author has certification from ASHA and has been licensed to practice speech-language pathology for a period greater than 30 years
 - ii. Coder 2 is a college graduate, has completed formal training in phonetic transcription, and has been NSL lab manager for three years with greater than four years transcription experience.
 - iii. Consensus coders are both familiar with the speaking style of children with 7q11.23 disorders and with the speaking style of TD English-speaking children from many regions of the US, Canada, and from English-speaking individuals from non-US countries.
 - f. Prior reevaluation practice will include the following.
 - i. Jointly, both coders will complete six transcript reevaluations using transcripts that do not meet inclusion criteria for the present dissertation.
 - ii. The six practice transcripts will be randomly chosen from the following pools:
 - a) Two younger children with WS
 - b) Two older children with WS
 - c) One younger child with Dup7
 - d) One older child with Dup7
 - g. Two coders will reevaluate each continuous speech sample jointly. Samples will be reevaluated utterance-by-utterance.
 - h. Each coder will perform a single task when reevaluating transcripts but both will mark copies of each transcript independently.
 - i. Each coder will have a personal copy of each transcript for independently marking
 - ii. Coder 1 will identify intelligible words in a single utterance and underline each word.
 - iii. Coder 2 will confirm words above (i.) and raise issue for discussion any unintelligible word.
 - iv. Consensus will be achieved regarding utterance intelligible word count.
 - v. Coder 2 will identify phones in each utterance and circle error phones on the transcript.
 - vi. Coder 1 will confirm phones above (v.) and raise issue with any questionable error. The transcript will be marked to indicate any change in initial decision of erred phone.
 - vii. Consensus will be achieved regarding phone accuracy prior to Coder 2 marking the PCC scoring form.
 - viii. Discussion regarding (ii.) and (v.) above likely will necessitate replaying the audio-video record. Up to 3 replays will be permitted. If no agreement is reach after 3 replays, the word is considered unintelligible or the phone is considered an error.
 - ix. Both coders will sign the completed PCC scoring form attesting to the accuracy of phones and the accuracy of tally counts.
 - x. Coders will switch tasks on every subsequent reevaluated transcript.

Appendix C (continued)

- I. Sampling Rules
- a. The response definition is: score as incorrect unless heard as correct.
 - b. Only consonants in words are considered; vowels are not scored.
 - c. Syllabic consonants **are not scored** because they function as vowels, e.g., [ŋ] in *kitten*, [l] in *bottle*.
 - d. A consonant *addition* is considered an error unless the addition is appropriate for the speaker's dialect or culture; e.g., [h] in *it* articulated as [hit] or [ʔ] in *on* articulated as [ʔɔn].
 - e. Vocalic /ɪ/ scoring (excepting dialectical and cultural variation or coarticulation):
 - i. **Post-vocalic /ɪ/**, as in *fair*, [feɪɪ], is considered a consonant and thus it is scored,
 - ii. **Stressed vocalic [ə]**, as in *work*, is considered a syllabic consonant that is not scored,
 - iii. **Unstressed vocalic [ə]**, as in *furrier* [fɜːiə] is considered a vowel that **is not scored**.
 - f. Words that are incomplete or partially unintelligible **are not scored**.
 - g. The following words **are not counted or scored**: *the, a, and*.
 - h. Consonants in multiple successive repetitions of a *syllable* **are not scored**. For example, when the transcript and video record indicate stuttering, e.g., *ba-balloon--* only the first /b/ is scored.
 - i. Target consonants in the third or successive repetitions of adjacent *words* **are not scored** unless the articulation changes across exemplars. For example, the consonants in only the first two words of the series [kæt], [kæt], [kæt] are counted. However, the consonants in all three-word positions are counted if the series were [kæt], [kæk], [kæt].
 - j. The following types of connected-speech consonantal changes are scored as *incorrect*:
 - i. Omission of a consonant (initial /h/ deletion, unless appropriate dialectally: *he* = [i])
 - ii. A non-target consonant substitution: (final /ŋ/ → /n/ substitutions (*ring* = [rin])
 - iii. Addition of a consonant phone to a word, e.g., *cars* said as [kɑɪks].
 - iv. Distortions of a consonant, *no matter how subtle*; including the following:
 - a) Partial voicing or devoicing of consonants (unless dialectally appropriate) and
 - b) Stressed-syllable errors of distortion.
 - v. Unstressed syllables distortions must be considered always for dialectical norms and coarticulatory assimilations, e.g., *running north* [rʌ.nɪ.'nɔθ] and *feed her* = [fi.də].
 - vi. Clusters produced epenthetically will be scored as incorrect
 - k. Observe the following:
 - i. Phone accuracy will be identified with consideration for salient cues from the prosodic frame, respect for cultural, social, and dialectical difference, and acknowledgement of common, continuous-speech patterns of coarticulation. The following are some examples of correct phone articulations.
 - a. Final /p/ → /p̚/ (*top* = [tɑ:p̚])
 - b. Final /t/ → /t̚/ (*carrot* = [kɜː.rɔ̚t̚])
 - c. Final /dʒ/ → /dʒ̚/ (*budge* = [bʊ:dʒ̚])
 - d. Initial /tɹ/ → /t̚ɹ/ (*tree* = [t̚ɹi]), and Initial /dɹ/ → /d̚ɹ/ (*drink* = [d̚ɹɪŋk])
 - e. Initial /ʔ/ added to a word with a vowel onset. (e.g., *anyone* articulated as [ʔɛ.nɪ.wʌn])
 - f. Initial /ð/ → /n/ (*and then* = [æ.nɛːn]; e.g., continuous-speech coarticulation results in assimilatory processes for medial [n] and final [n] [casual speech: score correct]).
 - ii. Consonant productions, considered appropriate in particular ethnic, social, dialectical, or regional parlance, are transcribed as pronounced by the child, e.g., *picture* = [pɪ.t̚ʃə]; *ask* = [æks], etc. Allophonic variants that do not change word meaning are counted as correct.
 - iii. The prosodic frame will identify **word count** when coarticulatory change occurs (e.g., the following both are correct examples: [də.'noʊ] = two words, *there's* [ðe.ɪz] = one word)
 - iv. Complementary allophonic changes are scored as correct, e.g., *water* = [wa.rə], *tail* = [teɪ].
 - v. Rapid speech or casual consonantal assimilations are transcribed as the child pronounced them and scored correct, e.g., *don't know* = [də.'noʊ]; *good morning* = [gʊ.'mɔɪ.nɪŋ]

II. Appendix C (continued)

| PERCENTAGE CONSONANTS CORRECT (PCC) SCORING FORM | | | | | | | | | | | |
|--|-------------------|------------------------------|-----------|-----------------|-----------|----------|-----------|-----------------------------|--------------|----------------------------|--------------|
| Adapted from Shriberg & Kwiatkowski (1982) | | | | | | | | | | | |
| NSL Child ID: | | Severity Adjective: (circle) | | | | | | Sample Duration: | | | |
| DOB: | | PCC | | | | | | SALT 2012 PCIU: | | | |
| CA: | | 85 – 95% | | Acceptable | | (Code 1) | | Total Words (Analysis Set): | | | |
| Child Gender: | | 65 – 84% | | Mild/Moderate | | (Code 2) | | Total Number of Utterances: | | | |
| Date of Sample: | | 50 – 64% | | Moderate/Severe | | (Code 3) | | | | | |
| Date Scored: | | < 49% | | Severe | | (Code 4) | | | | | |
| Consonant Class | Position-In-Word: | Initial | | Medial | | Final | | Number Consonants Correct | In Class PCC | Total Number of Consonants | Phonemes PCC |
| | Phoneme | Correct | Incorrect | Correct | Incorrect | Correct | Incorrect | | | | |
| Nasals | /m/ | | | | | | | | | | |
| | /n/ | | | | | | | | | | |
| | /ŋ/ | | | | | | | | | | |
| Plosives | /p/ | | | | | | | | | | |
| | /b/ | | | | | | | | | | |
| | /t/ | | | | | | | | | | |
| | /d/ | | | | | | | | | | |
| | /tʃ/ | | | | | | | | | | |
| | /k/ | | | | | | | | | | |
| | /g/ | | | | | | | | | | |
| | /ʔ/ | | | | | | | | | | |
| Fricatives | /s/ | | | | | | | | | | |
| | /z/ | | | | | | | | | | |
| | /ʃ/ | | | | | | | | | | |
| | /v/ | | | | | | | | | | |
| | /f/ | | | | | | | | | | |
| | /ʒ/ | | | | | | | | | | |
| | /θ/ | | | | | | | | | | |
| | /ð/ | | | | | | | | | | |
| Approximants | /h/ | | | | | | | | | | |
| | /w/ | | | | | | | | | | |
| | /j/ | | | | | | | | | | |
| Affricates | /l/ | | | | | | | | | | |
| | /ɹ/ | | | | | | | | | | |
| | /ɹ̥/ | | | | | | | | | | |
| Clusters | /tʃ/ | | | | | | | | | | |
| | /dʒ/ | | | | | | | | | | |
| Clusters (All) | | | | | | | | | | | |
| Number in Position: | | | | | | | | | | | |
| PCC: | | | | | | | | Total | | Total | PCC |

Appendix D

- I.** Decision logic for *scoring* exceptional GFTA-2 items (c.f. Shriberg & Kent, 2013, pp. 131-135.)
- a. Administration procedures published in the GFTA-2 examiner’s manual will be strictly followed. Procedures are specified in the manual on pp. 20–25.
 - b. The video and the audio record must be considered for scoring.
 - c. A correct response is defined as a sound production that conforms to Standard General American Speech (SAE).
 - i. Allophonic variance is acceptable and will be scored as “correct”.
 - ii. Differences in pronunciation due to dialect will not be counted as errors. Specifically, dialectically different words from SAE will be transcribed in the appropriate space on the GFTA-2 record accurately using IPA notation (functionally indicating an error). Immediately, a descriptive notation will be made indicating the dialectical nature of the code. Upon scoring the record, the allowable item(s) will be ignored when tallying errors.
 - iii. Clusters produced epenthetically will be scored as incorrect.
 - iv. Item scoring applies (correct exemplars in non-targeted words do not affect scoring of the assessed target).
 - v. Compensatory articulatory movements due to structural differences (overjet, underjet, semi-paresis, etc.) that result in a production that sounds correct (undistorted acoustically but looks incorrect) will be marked with appropriate diacritics, and upon scoring will be scored as incorrect.
 - d. Any response requiring diacritic marking will be scored online as “incorrect”.
 - i. After the GFTA-2 has been administered, the video should be reviewed for dialectical differences; these are acceptable and will be scored as “correct”.
 - ii. Diacritic marking applied to sounds because of unanticipated oral movement will be scored as “incorrect” (i.e., groping movements).
 - e. Correct responses must be “socially” acceptable; that is, neither drawing attention to the speaker nor interfering with communication.
 - i. In the case when it is clear that an examinee purposefully distorted a response, a second exemplar can be scored. For repeated responses elicited due to previous socially-unacceptable responses and that occur in the reliability sample, these instances must be noted and agreed upon through consensus (The examiner should have paused or discontinued the assessment if the examinee’s behavior negatively impacted the accuracy of scoring.)
 - f. All questionable responses should be scored as “incorrect.”
- II.** Decision logic for *transcribing* narrative items or marking errors using *diacritics*
- a. IPA phonetic symbols (2016) will be used in all transcriptions (see Appendix C and <http://www.InternationalPhoneticAlphabet.org>)
 - b. A standard audio file will be accessed for transcribing questionable items: IPA 2.1 HELP program (SIL International, 2008, Consonants or Vowels pages). A free download of this program is available at: http://www.sil.org/resources/software_fonts/ipa-help).

If a response does not match the SIL International 2008 standard audio-file example, the first thing to do is to mark the item with diacritic symbols (c.f. IPA HELP 2.1, Diacritics page). When these are insufficient to describe the perceptual impression, one other source will be sufficient and necessarily adequate: Extended IPA Symbols for Disordered Speech (see attached; ICPLA, 2008, Appendix D herein).

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EDUCATION

University of Louisville
Louisville, KY
Ph.D., Experimental Psychology 2018

University of Louisville
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M.S., Experimental Psychology 2013

Certificate of Clinical Competence 1987
American Speech-Language Hearing Association

NOVA Southeastern University
Davie, FL
M.S., Speech-Language Pathology 1986

Eastern Kentucky University
Richmond, KY
B.S., Speech Pathology and Audiology 1978

PROFESSIONAL HISTORY

| | | |
|---|----------------|---|
| Speech-Language Pathology | 1988 - present | Kentucky Board of Occupations and Professions (#138101) KY License to Practice |
| Graduate Research Assistant | 2011 - present | University of Louisville, Neurodevelopmental Sciences Laboratory |
| Graduate Teaching Assistant | 2010 | University of Louisville, Dept. of Psychological and Brain Sciences |
| Graduate Research Consultant | 2008 - 2009 | University of Kentucky, Brain, Cognition, and Development Laboratory (Jane E. Joseph, Ph.D.) |
| Speech-Language Pathologist (Agency Owner) | 1996 - 2007 | Huffman and Associates, Inc. Kentucky First Steps Early Intervention Program Jefferson County Public Schools, KY (Preschool Program) |
| Speech-Language Pathologist (Independent Practitioner) | 1986 - 1996 | Myra J. Huffman, CCC-SLP Kosair Children's Hospital (University of Louisville Foundation) Audubon Hospital Frazier Rehabilitation Institute Spectra Care Home Health, Inc. Jefferson County Public Schools, KY (Preschool Program) |
| Speech-Language Pathologist | 1978 - 1986 | Palm Beach County Public Schools, FL (Language-Learning Disorders Classroom Instructor) Garrard County Schools, KY (Itinerant Speech-Language Pathologist) |

AWARDS

- 2018 Doctoral Dissertation Completion Award, University of Louisville
- 2014 Travel Award, International Professional Conference on Williams Syndrome
- 2011 Travel Award, Symposium on Research in Child Language Disorders

DOCTORAL DISSERTATION

Huffman, M. J. (2018). *Speech articulation in children with Williams syndrome or 7q11.23 duplication syndrome* (Unpublished doctoral dissertation). University of Louisville, Louisville, KY.

RESEARCH PUBLICATIONS

Velleman, S. L., Huffman, M. J., & Mervis, C. B. (in press). Childhood motor speech disorders in a child with 7q11.23 duplication syndrome. In R. Branski & S. Molfenter (Eds.), *Speech-language pathology casebook*. New York: Thieme Medical Publishers.

Mervis, C. B., Klein-Tasman, B. P., Huffman, M. J., Velleman, S. L., Pitts, C. H., Henderson, D. R., Woodruff-Borden, J., Morris, C. A., & Osborne, L. R. (2015). Children with 7q11.23 duplication syndrome: Psychological characteristics. *American Journal of Medical Genetics Part A*, 167A, 1436-1450.

RESEARCH PRESENTATIONS

Velleman, S. L., Huffman, M. J., & Mervis, C. B. (2018, November). *Babble characteristics as predictors of concurrent lexical volubility in Williams syndrome*. American Speech Language Hearing Association, Boston, MA.

Velleman, S. L., Huffman, M. J., & Mervis, C. B. (2018, June). *Speech and grammar issues in children with 7q11.23 duplication syndrome*. Duplication Cares National Convention, Crystal City, VA.

Velleman, S. L., Huffman, M. J., & Mervis, C. B. (2017, November). *Early phonological systems: Children with Williams syndrome*. American Speech Language Hearing Association, Los Angeles, CA.

Velleman, S. L., Huffman, M. J., & Mervis, C. B. (2017, September). *Early phonological systems of children with Williams syndrome*. Asia Pacific Conference on Speech, Language, and Hearing, Narita, Japan.

Price, E., Velleman, S. L., Huffman, M. J., & Mervis, C. B. (2016, November). *Babbling and later speech abilities in children with Williams syndrome*. American Speech-Language Hearing Association, Philadelphia, PA.

- Huffman, M. J., Freeman, K., Velleman, S. L., & Mervis, C. B. (2015, November). *Narrative language in children with Williams syndrome and children with 7q11.23 duplication syndrome*. American Speech-Language Hearing Association, Denver, CO.
- Morris, C. A., Mervis, C. B., Huffman, M. J., Velleman, S. L., Klein-Tasman, B. P., Tam, E., & Osborne, L. R. (2015, August). *Genotype phenotype correlations in 7q11.23 Duplication syndrome*. David W. Smith Workshop on Malformations and Morphogenesis, St. Michaels, MD.
- Huffman, M. J., Velleman, S. V., & Mervis, C. B. (2014, July). *7q11.23 Duplication syndrome and childhood apraxia of speech*. National Conference on Childhood Apraxia of Speech, Nashville, TN.
- Velleman, S. V., & Huffman, M. J. (2014, July). *Differential diagnosis in complex cases*. National Conference on Childhood Apraxia of Speech, Nashville, TN.
- Huffman, M. J., Bellamy, J. D., & Mervis, C. B. (2014, June). *Development of speech production accuracy in children with Williams syndrome*. International Professional Conference on Williams Syndrome, Anaheim, CA.
- Velleman, S. L., Jones, L., Varley, L., Huffman, M. J., & Mervis, C. B. (2013, November). *Early speech development in Williams syndrome*. American Speech-Language Hearing Association, Chicago, Illinois.
- Huffman, M. J., Velleman, S. V., Morris, C. A., Osborne, L. R., & Mervis, C. B. (2013, June). *Speech, language, and intellectual abilities of children with Williams syndrome (7q11.23 deletion) or 7q11.23 duplication syndrome*. Symposium on Research in Child Language Disorders, Madison, WI.
- Velleman, S. L., Huffman, M. J., & Mervis, C. B. (2013, June). *Speech, language, and cognitive abilities of children with 7q11.23 duplication syndrome*. International Child Phonology Conference, Nijmegen, Netherlands.
- Huffman, M. J., Velleman, S. L., & Mervis, C. B. (2012, July). *Relations among speech and intellectual abilities in children with Williams syndrome*. International Professional Conference on Williams Syndrome, Boston, MA.
- Huffman, M. J., Velleman, S. L., & Mervis, C. B. (2012, June). *Relations between motor speech skill and single-word accuracy in children with Williams syndrome*. International Clinical Phonetics and Linguistics Association, Cork, Ireland.
- Velleman, S. L., Currier, A., Huffman, M. J., Matthews, K., & Mervis, C. B. (2012, June). *Early speech development in Williams syndrome*. International Clinical Phonetics and Linguistics Association, Cork, Ireland.

Velleman, S. L., Huffman, M. J., & Mervis, C. B. (2012, June). *Relations between speech and motor-speech performance in children with 7q11.23 duplication syndrome*. International Child Phonology Conference, Minneapolis, MN.

Currier, A., Huffman, M. J., Velleman, S. L., & Mervis, C. B. (2011, November). *Speech and oral motor skills in 7q11.23 duplication syndrome*. American Speech-Language Hearing Association Annual Conference, San Diego, CA.

Huffman, M. J., Velleman, S. L., & Mervis, C. B. (2011, June). *Motor speech characteristics of children with Williams syndrome*. Symposium on Research in Child Language Disorders, Madison, WI.