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TITLE

Single-word Speech Intelligibility in Children and Adults with Down Syndrome

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Abstract

Purpose: A single word identification test was used to study speech production in children and adults with Down syndrome (DS) to determine the developmental pattern of speech intelligibility with an emphasis on vowels.

Method: Speech recordings were collected from 62 participants with DS ages 4-to-40 years, and 25 typically developing (TD) participants ages 4-to-7 years. Panels of five adult lay listeners transcribed the speech recordings orthographically and their responses were scored in comparison to the speakers' target words.

Results: Speech intelligibility in persons with DS improved with age, especially between the ages of 4-to-16 years. While consonants contribute to intelligibility, vowels also played an important role in reduced intelligibility with an apparent developmental difference in low versus high vowels, where the vowels /æ/ and/ɑ/ developed at a later age than /i/ and /u/. Interspeaker variability was large with males generally less intelligible than females, and some adult males having very low intelligibility.

Conclusion: Results show age-related patterns in speech intelligibility in persons with DS, and identify the contribution of dimensions of vowel production to intelligibility. The methods used clarify the phonetic basis of reduced intelligibility, with implications for assessment and treatment.

Introduction

Reduced speech intelligibility in persons with Down syndrome (DS) frequently results in communication difficulties and frustration for both speakers and their communication partners (Kent & Vorperian, 2013; Kumin, 1994; Roberts, Price, & Malkin, 2007). It is a challenge to study speech intelligibility comprehensively, because it is affected by many factors related to the speaker, listener, and context (Hustad, 2012; Kent, Miolo, & Bloedel, 1994). In persons with DS, factors likely to influence intelligibility are: voice, articulation, resonance, fluency, and/or prosody (Kent & Vorperian, 2013). Since many factors contribute to intelligibility, no single test can capture all aspects of intelligibility in DS. Studies of intelligibility in DS have used a variety of approaches that can be classified into two general categories: global and analytical. Hustad (2012) made a similar distinction using the respective terms "subjective" and "objective." Global approaches entail overall ratings made by listeners (such as percent of words understood, or values on an interval scale) serving as a gauge of the severity of the communication problem; while analytical approaches involve scoring a unit of analysis, such as a word, phoneme, or phonetic feature, as correct or incorrect and serve to identify the reason for reduced speech intelligibility in speakers with DS. Our use of the term "analytical" compares with the term "diagnostic intelligibility testing" which "aims to reach beyond mere quantification of degree of overall intelligibility and gain insights into specifically why someone is difficult to understand" (Miller, 2013, p. 604).

Global Approaches

Reduced speech intelligibility by both familiar and unfamiliar listeners has been documented in several studies. Questionnaires administered to parents of persons with DS make it clear that

family members recognize reduced intelligibility as an obstacle to communication and that the problem can be lifelong. Kumin (1994) reported that 95% of persons with DS across the lifespan experience some difficulty being understood. Kumin (2006) also reported that persons with DS ages 1 to 21 years had a mean speech intelligibility rating from their parents of 4.97 on a 10-point scale (1 being the least and 10 as the most), with boys with DS being less intelligible than girls with DS in the same age group. Toğram (2015) reported similar results from parents of children with DS ages 1-19 years. Reduced speech intelligibility persists into adulthood but there are few published reports that describe the problem in detail or show the general pattern of change with age (Coppens-Hofman, Maassen, van Schrojenstein Lantman-de Valk, & Snik, 2012).

Similarly, overall ratings at the word level made by unfamiliar listeners, such as percent of words correct, have often been used as an outcome measure in studies of communication in persons with DS. For example, ratings of intelligibility reported for connected speech are: 81.2% for children with DS ages 4-16 years (Barnes et al., 2009), 87% for conversation for persons with DS ages 5 to 20 years (Chapman, Seung, Schwartz, & Kay-Raining Bird, 1998), and 66.4% for males with DS ages 10-17 years (Rosin, Swift, Bless, & Vetter, 1988). Differences in methodology may account for part of the variability across these studies. These word-level analyses show that persons with DS often have difficulties producing speech that is easily understood.

Analytical Approaches

Studies using analytical approaches provide insights into the phonetic components that affect intelligibility in persons with DS, which could help in guiding treatment plans to enhance

intelligibility. Higher error rates for consonant than vowel production among children and young adults with DS were reported by Dodd and Thompson (2001) for ages 5 to 15 years and Borsel (1996) for 15 to 28 years. However, both consonant and vowel errors in all articulatory positions except for the high-front vowel /i/ have been reported in a single-word phonetic intelligibility test of five men with DS (Bunton, Leddy, & Miller, 2007). Although Bunton et al. (2007) did not report total error rates for consonants and vowels, more errors in the high-low dimension of vowel production than the front-back dimension were noted. The impact of phonological processes on intelligibility in persons with DS has not been studied systematically. However, persons with DS reportedly have both delayed and disordered phonological processes, which likely affect intelligibility (Kent & Vorperian, 2013).

Converging Conclusions from Global and Analytic Studies

The two approaches converge on some general conclusions. The main conclusion, which motivates the present study, is that reduced speech intelligibility is a common problem in persons with DS and can be a lifelong issue for some individuals. The available data address overall intelligibility but reveal little about developmental changes in factors underlying intelligibility, for example, patterns of phonetic features for vowels and consonants. Another conclusion is that speech intelligibility is highly variable within the population of individuals with DS, but little is known about the factors underlying this variability. A related conclusion, albeit somewhat tentative because of the limited data, is that boys with DS have poorer articulation and intelligibility than girls with DS (Kumin, 2006; Martin, Klusek, Estigarribia, & Roberts, 2009; Roberts et al., 2005). What is not clear is whether this sex difference persists into adulthood.

Research Strategy for a Lifespan Perspective on Speech Intelligibility

An understanding of the reduced speech intelligibility in DS, across the lifespan, requires a multi-faceted program of research that considers the nature of the deficit in speech intelligibility and the atypical phonological patterns observed at different stages of development; while taking into account the articulatory-motor difficulties suspected to occur in persons with DS as well as the anatomic features commonly associated with the syndrome.

Research on DS is further complicated because the syndrome has a complex phenotype consisting of features that are not fully penetrant (Antonarakis & Epstein, 2006; Vilardell et al., 2011). To address this issue of heterogeneity within the population of individuals with DS, recent research studies have placed emphasis to the study of developmental trajectories (Fidler, 2005), and individual profiles in development (Karmiloff-Smith et al., 2016; Tsao & Kindelberger, 2009). Such a strategy is well suited for research on a lifespan perspective of communication skills, and can encompass issues such as sex differences, time of emergence of critical skills or deviation from the norm (i.e. emergence of differences), and age intervals of greatest change.

The present study is one step in the research process and focuses on the lifespan study of speech intelligibility deficit using a single-word intelligibility test designed specifically to assess vowel production. The rationale for the methodology used is presented next.

Methods to Study Lifespan Speech Intelligibility

Tool to study lifespan speech intelligibility: A tool that is ideal for the study of the developmental trajectory of speech intelligibility is one that is suited to individuals of different ages and different levels of capability. A single-word identification test, is advantageous for

quantifying changes in speech intelligibility over development in persons with DS for several reasons: First, many persons with DS can participate in a single-word repetition task regardless of their verbal ability and/or age. Single word production is viewed as a general strength in children with DS (Grieco, Pulsifer, Seligsohn, Skotko, & Schwartz, 2015). Second, this approach provides an opportunity for systematic phonetic analysis to characterize the phonetic basis of intelligibility reduction in persons with DS of all ages. Third, the recordings can be used to determine the acoustic correlates of phonetic errors, for example, the voice-onset-time data for voiced-voiceless contrasts in consonants or the formant-frequency patterns for vowel productions offering a unique opportunity to establish correlations among acoustic, perceptual, articulatory, and even anatomic data. Fourth, a narrow phonetic focus can serve as a touchpoint for studies of acoustic and articulatory correlates (e.g., using vowel space area as an index of articulatory working space for vowels, more on this in the following section). Thus, keeping in mind the need for a lifespan perspective, the rationale for an initial focus on vowels is that (1) vowels typically are acquired early in speech development and can be followed from babbling onward, (2) vowels serve as syllable nuclei and therefore have a central role in emerging phonologies, and (3) vowels can be described in terms of articulatory-acoustic correlates such as the F1-F2 vowel plot interpreted as articulatory features.

Corner vowels depicting articulatory working space: The syndrome-specific craniofacial characteristics in DS include a smaller mid and lower facial skeleton, but no differences in tongue size (Uong et al., 2001). A typically-sized tongue inside a small oral cavity may limit articulatory performance, specifically restricting the articulatory working space for vowels (Kent & Vorperian, 2013). This idea is supported by research on acoustic vowel space area and/or

vowel overlap (Abe, 1973; Bunton & Leddy, 2011; Whitworth & Bray, 2015), and by a report of reduced acoustic contrast in the F2 values of the high vowels /i/ and /u/ (Moura et al., 2008). In fact, Moura et al. (2008) suggested that the smaller ratio of the F2 frequencies for vowels /i/ and /u/ is the consequence of anatomic features in DS and they accordingly named the relationship between these vowels the *DS vocalic anatomical functional ratio*. Aside from acoustic studies, difficulties with vowel production in DS are evidenced by parent reports (Toğram, 2015), listener judgments of prolonged vowels (Moran, 1986), and speech intelligibility tests (Bunton, Leddy, & Miller, 2007). Despite these findings indicating that vowel production may impact intelligibility, the relationship between the dimensions of vowel production (front, back, high, low) and intelligibility reduction in persons with DS has not yet been reported. Such knowledge could be invaluable in guiding speech therapy and/or assessing progress in speech treatment. Also, such information can be used in shaping future anatomic studies and/or evaluating surgical intervention strategies used in upper airway treatment.

Purpose

The purpose of this perceptual study is to use a single-word identification test with lay listeners to identify phonetic components that contribute to reduced intelligibility in female and male speakers with Down syndrome ages 4 to 40 years. Specific research questions are listed below:

1. What does an open-set word-intelligibility test reveal about the pattern of speech intelligibility (words correct scores) in males and females with DS ages 4 to 40 years? We hypothesized that the results would show (a) improving intelligibility with age, reaching asymptote at adulthood, and (b) substantial interspeaker variability, owing in part to a sex difference with males having lower intelligibility scores than females.

2. Do vowels differ in their contribution to speech intelligibility? We hypothesized that vowel errors would occur in DS and that the front-back dimension of vowel production would have more vowel identification errors than the high-low dimension because of the reduced mid and lower face skeleton associated with DS.

Methods

Part I: Speech sample production

Participants

All study participants were native speakers of English from the Midwest and followed the same procedure as approved by the University of Wisconsin-Madison health sciences Institutional Review Board (IRB). Participants (speakers) included 62 persons with DS (29 females, 33 males; ages 4 to 40 years), and a control group of 25 typically developing (TD) participants (11 females, and 14 males; ages 4 to 7 years). They were recruited via community flyers and the Developmental Disabilities registry at the Waisman Center. The participants with DS had Trisomy 21 with no other concomitant diagnosis such as autism or dementia. Elective intake information regarding speech and hearing concerns, history and/or treatment was provided by 92% of the caregivers of participants with DS and all TD participants. Findings reflected a history of speech therapy services, between the ages 4-21 years of age, for the majority of the participants with DS (92%), with 35% of those participants receiving services at the time of their speech recording; and hearing concerns was reported for 38% of the participants with 17% of those indicating it being a former concern. More formal hearing assessment (hearing screening or audiogram), available for 60% of the participants with DS, reflected hearing status to have a wide range from no hearing loss, to mild, to moderate and even severe high frequency hearing

loss in two cases (who wore hearing aids). No developmental concerns and no speech and hearing concerns or treatment was reported for any of the TD participants. Observations of the participants' orofacial structures and function revealed participants with DS to have open mouth posture at rest (37%), under bite (32%), tongue thrust with swallow (21%), and drooling (3%). No similar observations were made for any of the TD participants. The participants with DS were studied in a combined cross-sectional and longitudinal (multiple-session) design where some of them made repeat visits for speech recordings as a venue to explore intra-speaker variability during the course of development. Given that the hearing status was highly variable across participants with DS and within participants with DS who came in for multiple visits, speech recording was carried out after confirming that the participant could follow simple commands and repeat words. Each participant was involved in at least one speech recording session, with a minimum span of 9 months between recording dates for participants who had multiple sessions. Of the 62 participants with DS, 24 speakers participated in one session and 38 participated in 2 to 7 sessions, yielding a total of 143 speech recordings. As for the 25 TD participants, 23 speakers participated in one session and 3 participated in two sessions, yielding 29 total speech recordings. The limited age range of TD participants was related to the purpose of determining the age at which children are fully intelligible. TD participants of the following ages were included: age 4 years (2 girls and 2 boys), age 5 years (3 girls and 6 boys), age 6 years (3 girls and 8 boys) and age 7 years (3 girls and 2 boys).

Procedure

Speech stimuli, drawn from a larger speech task, consisted of 20 words, each word composed of one of the four corner vowels within a consonant-vowel, vowel-consonant, or consonant-

vowel-consonant syllable. Words listed by the vowel they contain are: /i/- bead, bee, eat, sheep, and feet; /u/- boo, boot, zoo, hoot, and shoe; /æ/- bath, bat, cat, hat, and sad; and /ɑ/- dot, hop, pot, top, and hot. Production of corner vowels requires that the tongue be in the most extreme articulatory positions, so these words were chosen to facilitate analysis of the four dimensions of vowel production (high, low, front, and back). Stimuli were also selected to be developmentally appropriate for children and to have a high phonological neighborhood density, as this has been shown to maximize acoustic vowel space (Munson & Solomon, 2004).

Participants were seated in a quiet room, and, when possible, completed a pure-tone hearing screening in accordance with American Speech-Language-Hearing Association (ASHA) standards (2016). Then, speech stimuli were randomly presented on a 14" laptop computer or an 11" tablet using the TOCS+ Platform (Hodge & Daniels, 2007). The printed word for each stimulus was displayed below its corresponding image, as shown in Figure 1, and a previous recording of the word, produced by a man judged to be highly intelligible and with good voice quality, was presented aurally through loud speakers at the same time. Participants were instructed to say each word after hearing it. An imitation task allowed for later comparison of listeners' responses to known target stimuli. Participants spoke into a unidirectional Shure SM48 microphone that was placed approximately 15 cm from the speaker's mouth and was stabilized using a floor stand. The microphone was directly connected to a Marantz PMD660 digital audio recorder that digitized speech at 48 kHz with 16-bit resolution. Targeted recording level on the audio recorder's volume unit (VU) meter was between 6 to 12 dB below the maximum level. Prior to producing the selected stimuli, participants counted from one to five and/or repeated two practice stimuli while adjustments were made to ensure adequate mouth-

to-microphone distance and/or recording level. If additional adjustments were needed during the recording session, repeat productions were elicited.

Part II: Intelligibility (perceptual) study

Participants

All 54 adult lay listening participants (17 men and 37 women) were native speakers of English from the Midwest and determined by self-report to be free of communication disorder. They ranged in age from 19 years to 58 years, with a mean age of 30 years (*SD* = 11.5 years).

Procedure

The stimuli for the listening task were prepared as follows. Each speech recording (as described above) was segmented at the word level, and words were saved as separate sound files using Praat (Boersma & Weenink, 2010). Each word was normalized for amplitude to an average root mean square intensity of -10 dB SPL using Sound Forge (Version 8.0; 2006) to ensure that fluctuations in speakers' sound level would not affect intelligibility ratings. Speakers' recordings were assembled into 32 groups for presentation to listeners. Each group contained speech recordings from 5 to 7 speakers. Twenty-seven groups included speakers with DS and five groups included TD speakers. Age and sex of DS speakers were balanced to ensure that recordings from children, adults, males, and females were included in each group. Similarly, TD speakers' sex was balanced to ensure that recordings from both boys and girls were presented in each group. Groups with TD speakers included either 4-to-5-year-olds or 6-to-7-year-olds. TD speakers were separated by age given inconsistent reports on the age at which children are fully intelligible to unfamiliar listeners (Coplan & Gleason, 1988; Hustad, Oakes, & Allison, 2015).

Panels of five listeners heard one group of recordings per session, and participated in 2 to 3 sessions on different days. Thus, each group of recordings was presented to five different listeners, since variability in listener performance has been reported among studies of intelligibility (DePaul & Kent, 2000; Hustad et al., 2015). Listeners were seated in a quiet room to complete the study, which began with a pure-tone hearing screening in accordance with American Speech-Language-Hearing Association (ASHA) standards (2016). If listeners did not pass the hearing screening, they completed only the first of three listening sessions and their data were not used. After the hearing screening, listeners read instructions for the listening task. They also viewed a list of the speakers' target words to reduce learning effects, since listeners can learn as a task progresses, even with no instruction or feedback (Hustad, et al., 2015). In addition, listeners were verbally instructed to type what they heard and not what they thought the speaker was attempting to say. The listening task was completed using a 15" laptop with a wireless mouse. Before beginning the task, listeners chose a comfortable listening level by manually adjusting the laptop volume while a pre-recorded voice counted from one to ten. Then, speech stimuli were presented to listeners through headphones via an in-house computer program written using Python 3.2. After each word was presented, listeners typed what they heard. Words were presented only once, and listeners controlled the rate of stimulus presentation. After typing a response, listeners clicked a 'next' button, and another word was presented after a two-second time delay. All words from each speaker were presented before listeners heard another speaker. The order of words within each speaker's recording and order of speakers' recordings within each listening group were randomized during each session to prevent potential order effects.

Data Analysis

A software program with two analysis features was written in-house using Python 3.2 to analyze listeners' responses. The first analysis option scored listeners' identification of each target phoneme and word as correct or incorrect. The program searched for the phonetic representation of each listener's response within the Carnegie Mellon University (CMU) Pronouncing Dictionary (Version 0.7a; 2008), compared it to the phonetic representation of the speaker's target word, and then scored each phoneme and word in the listener's response.

Since the CMU dictionary contains only English words, phonetic representations of some listeners' responses were not available for automatic scoring, and manual scoring was required for these responses. Manual editing was also completed as needed to verify scoring accuracy. An example of a response that would require manual scoring is 'deeb' for the target word 'bead', because the phonetic representation of 'deeb' is not in the CMU dictionary. The word, initial consonant, and final consonant would be manually scored as incorrect, but the vowel would be correct. Thus, speakers were given credit for all phonemes produced accurately in the correct word position, regardless of whether the word was correct. Additionally, listeners' responses that were blank or consisted of one letter were considered incomplete and were not included in analysis. This prevented bias due to listeners' mistakes, such as accidentally clicking the 'next' button before typing their entire response.

After manual editing, the software's second analysis feature calculated the speaker's average percentages correct across all five listeners' responses for words and each of the following phonetic components: total consonants, initial consonants, final consonants, total vowels, and individual vowels /i/, /u/, /æ/, and /ɑ/. These percentages correct, averaged across

five listeners for each speaker, are referred to as words correct or phonetic components correct, and used as the primary measure of speech intelligibility for each speaker.

Statistical Analysis

To determine the developmental pattern of speech intelligibility in males and females with DS ages 4 to 40 years (first research question), we used words correct as the primary measure of intelligibility, with total consonants, initial consonants, final consonants, total vowels, and individual vowels /i/, /u/, /æ/, and /ɑ/ correct as the phonetic components of words correct. Because words correct is a percent, we applied a generalized linear mixed model (GLMM) with a logit link and binomial errors using PROC GLIMMIX, Statistical Analysis System (SAS) software (version 9.4 by SAS Institute, Inc., 2013). As most participants with DS had multiple sessions, a random intercept for each participant was included in this model to account for correlation across multiple recordings. The model included predictors related to both age and sex (male/female) of the speaker. As scatter plots of words correct suggested a non-linear relationship between words correct and age, a fractional polynomial technique was used to select the best-fitting transformation of age (Royston & Altman, 1994). The best transformation was found to be the inverse (1/age). There was no significant interaction between 1/age and sex.

To examine differences in the frequency of errors across vowel types (second research question), we applied a different form of analysis to account for the multivariate outcome. In contrast to the words correct analysis, we now have two sources of statistical dependence in our data: (1) dependence across vowel types due to observations being collected at the same recording occasion, and (2) dependence due to the same participants providing recordings over

time. We thus specified a multivariate multilevel model in which the error frequencies of each vowel type are the outcomes and vowel-type binary indicators are entered as speaking occasion predictor variables. Due to the use of a multivariate outcome, we adopted a slightly different approach to handling proportional outcomes; specifically, we applied an arcsine transformation of the square root of proportion correct, and entered binary indicator predictors for all but the fourth vowel type /u/ which served as the reference category. The choice of /u/ as a reference category was arbitrary, and statistical equivalence involving alternative reference categories was confirmed. To apply this model, we used HLM 7.01 (Raudenbush, Bryk & Congdon, 2010) and applied follow-up contrasts within the software that conduct statistical tests of differences between all possible pairs of vowel types. Then, to compare patterns of speech intelligibility (words/vowels correct scores) between males and females with DS, in a separate analysis we entered a binary indicator predictor for speaker sex (entered at the speaker level of the model) as well as interactions between the speaker sex and vowel-type predictors. Tests of statistical significance are based on corresponding statistical contrasts of the relevant coefficient estimates in the model. Using HLM, these statistical tests are single degree-of-freedom chi-square tests.

Listener reliability for words correct scores was assessed using the intraclass correlation coefficient (ICC) (Shrout and Fleiss, 1979). Since words correct scores for each speaker were averaged across five listeners, the ICC for a mean of five random raters (1,5) was calculated for a random sample of 25 speakers.

Results

Listener Reliability of Words Correct

The average of five listeners per speaker was included in the study design to ameliorate effects related to potential differences among listeners. As noted above, the intraclass correlation coefficient (ICC) was calculated to assess listener reliability using the mean of five random raters for 25 randomly selected speakers. The ICC was 0.95 with a 95% confidence interval of 0.91 - 0.97, confirming high reliability for words correct.

Words Correct as a Function of Age and Speaker Sex

Figures 2a-b provide an overall view of the data for words correct for individual speakers with DS compared with data for the TD speaker groups. Data for persons with DS are shown by scatter plots for cross-sectional data and scatter plots connected with lines for longitudinal data. Data for the TD groups are shown by box and whisker plots in the top left corner of each graph. TD girls ages 4 to 5 had a mean percent correct of 76 (*SD* = 9), while 6-to-7-year-olds had a mean of 92 (*SD* = 7). Similarly, the mean percent words correct for TD boys ages 4 to 5 was 81 (*SD* = 11), while 6-to-7-year-olds had a mean of 93 (*SD* = 5). As expected, words correct improved with age for all speakers, both DS and TD. The 4-to-5 and 6-to-7-year-old TD children were more intelligible than children with DS of the same age and sex. Children with DS also had greater variability than TD speakers of the same age and sex, with the exception of 4-to-5-year-old TD boys. Furthermore, TD 6-to-7-year-olds were more intelligible than most children and adults with DS of the same sex, indicating markedly reduced speech intelligibility in many persons with DS. For children with DS younger than 14 years of age, 100% of males and 95% of

females were less intelligible than TD 6-to-7-year-olds. For persons with DS ages 14 years and older, 74% of males and 49% of females remained less intelligible than TD 6-to-7-year-olds.

Despite the overall trend of improved speech intelligibility with age in persons DS, there was considerable intra- and inter-speaker variability. As shown by the longitudinal data (points for the same speaker are connected) in Figures 2a-b, most persons with DS showed improved intelligibility across sessions between ages 4 and 16 years. However, after 16 years of age, intelligibility patterns across sessions were more variable. Thus, intelligibility did not always improve across sessions, and reduced intelligibility persisted into adulthood for some persons with DS. Considerable inter-speaker variability among persons with DS can be seen in Figures 2a-b for any age-sex combination.

The possibility of sex differences is also evident in Figures 2a-b, where visual examination of the data shows females with DS being more intelligible at the word level than males, particularly for persons aged 9 years and older. Words correct for females with DS was greater than 40% by age 9, but most males did not reach this level of intelligibility until age 16. Additionally, all females with DS ages 8 years and older had greater than 40% words correct, whereas some adult males with DS did not exceed 40% words correct.

Statistical testing of the developmental trajectories was conducted using the generalized linear mixed model described earlier. The coefficient for the inverse of age (1/age) was -15.64, which was highly significant ($F(1,80) = 43.36$, $p < .0001$). A significant main effect coefficient of .65 was also observed for sex ($F(1,61) = 5.39$; $p=.024$), with female speakers yielding higher proportion correct. As noted earlier there was no detectable interaction between sex and inverse of age. The resulting model based trajectories by sex are shown in

Figure 3, demonstrating that word intelligibility increased significantly with age among both males and females with DS at seemingly similar rates, and with a larger rate of increase during the developmental ages (especially 4 to 16 years).

Words Correct as a Function of Phonetic Component

As described earlier, our scoring permitted a finer assessment of how both consonant and vowel errors contributed to diminished speech intelligibility. Figures 4 and 5 illustrate that both consonants and vowel errors contribute to intelligibility as measured by percent words correct. Figure 4 highlights the relationship between words correct and total consonants correct (initial and final consonants combined; Figure 4a), and total vowels correct (Figure 4b). Figure 5 displays the finer detail of the relationships between words correct and initial consonants correct (Figure 5a) and final consonants correct (Figure 5b); as well as words correct and each of the individual vowels correct: /i/ (Figure 5c), /æ/ (Figure 5d), /u/ (Figure 5e), and /ɑ/ (Figure 5f). Closer examination of Figure 5 reveals that the relationship between percent words correct and each of the vowels correct was non-uniform, with particular vowel types associated with a greater occurrence of errors. We therefore carried out statistical analyses to test for differences among vowels (second research question), as described in the following section.

Vowel Error Frequency as a Function of Vowel Type

Statistical analysis contrasting vowel types (based on the multivariate multilevel analysis described earlier) indicated statistically significant differences in error frequencies between vowel types /ɑ/ and /u/ ($\chi^2 = 131.63, df = 1, p < .001$), /æ/ and /u/ ($\chi^2 = 53.79, df = 1, p < .001$), /ɑ/ and /æ/ ($\chi^2 = 24.82, df = 1, p < .001$), /ɑ/ and /i/ ($\chi^2 = 175.63, df =$

$1, p < .001$), and /æ/ and /i/ ($\chi^2 = 65.94, df = 1, p < .001$). The only pairwise comparison that did not yield statistical significance was between /i/ and /u/ (χ^2=.79, df= 1, p>.500). This can be seen in Table 1, where the error frequencies are compiled by vowel type and support these results with the highest frequency of errors occurring for /ɑ/, followed by /ae/, and with /i/ and /u/ having the lowest error frequencies.

Vowel Errors as a Function of Speaker Sex

Given that the GLMM analysis found a significant main effect related to sex for overall intelligibility, we also proceeded to examine potential sex differences related to vowels specifically. Our second analysis used speaker sex as an additional predictor of vowel proportion correct, as described earlier. Findings revealed a significant main effect related to sex ($\chi^2 = 5.87, df = 1, p = .015$) such that males tended to have more vowel errors. An omnibus test for interactions between sex and vowel type based on the coefficients of product variables between sex and vowel type was not significant ($\chi^2 = 1.18, df = 3, p = .276$), suggesting a similar profile of vowel errors across sex. Table 1 displays the descriptive statistics of error frequencies by vowel type for each sex.

To examine the possibility of a developmental trend for vowel intelligibility in persons with DS, percent correct (averaged across five listeners) was plotted for each of the four corner vowels, as shown in Figure 6. Individual vowels correct for persons with DS are shown by triangles for cross-sectional data and circles for longitudinal data. As shown in Figure 6, high vowels /i/ and /u/ developed at an earlier age and were more intelligible than low vowels /æ/ and /ɑ/ for persons with DS. High vowels were developed by 4 years of age for most persons with DS. Females with DS of all ages had a mean percent correct of 97 (*SD* = 4) for high vowels,

while males had a mean percent correct of 92 or greater (*SD* = 15). However, percent correct continued to increase through 12 years of age for low vowel /æ/ and 16 years of age for low vowel /ɑ/ for persons with DS. Furthermore, persons with DS had fewer vowels correct and greater variability for low vowels /æ/ and /ɑ/ at all ages. Females with DS ages 13 years and older had a mean percent correct of 93 (*SD* = 10) for vowel /æ/, while males had a mean percent correct of 86 (*SD* = 19). For vowel /ɑ/, females with DS ages 17 years and older had a mean percent correct of 84 (*SD* = 20), while males had a mean percent correct of 80 (*SD* = 23). Although high vowels were mastered at younger ages than low vowels in females and males with DS, males had fewer vowels correct and more variability than females for each individual vowel.

TD speaker groups had a mean percent correct of 95 or greater (*SD* = 7) for every vowel, except 4-to-5-year-old TD girls for vowel /ɑ/. Table 3 displays the mean percent correct and standard deviation of each vowel for TD speaker groups. Intelligibility and age of mastery for high vowels were similar for TD speaker groups and persons with DS, but TD speakers had greater intelligibility and earlier age of mastery for low vowels.

Discussion

This study supports and extends previous research showing reduced intelligibility in children and adults with DS. To our knowledge, this is the first study to analyze speech intelligibility in children and adults with DS by using a uniform methodology focusing on patterns of intelligibility at the word and phonetic levels, specifically vowels. Our results confirm increased speech intelligibility with age in persons with DS, especially between the ages of 4-to-16 years.

This 12-year window of improved intelligibility holds implications for intervention focused on speech communication. The data also reveals considerable interspeaker variability. Males with DS were less intelligible than females, particularly during early childhood. As expected, both initial and final consonants contributed to intelligibility, but vowels also contribute to intelligibility and should not be neglected, as they also provide valuable information about the phonetic bases of reduced intelligibility. High vowels developed at an earlier age than low vowels in DS.

Variability

The considerable intra- and inter-speaker variability observed in this study agrees with previous findings documenting individual variability in error patterns among men with DS (Bunton, Leddy, & Miller 2007) and variability in speech intelligibility among children with DS (Kumin, 2006). In the present study, intelligibility patterns were the most variable across sessions for persons with DS over 16 years of age, as seen in Figures 2a-b. Such intra-speaker variability could be due to a variety of factors, such as the time of day speech was recorded, physiological or psychological state, and variations in medical condition and/or speech and language abilities. Informal assessment of the time of day of speech recording did not show any consistent patterns related to speech intelligibility.

Inter-speaker variability could be rooted in factors intrinsic to the speaker, such as craniofacial dysmorphology, speech motor impairment (including hypotonia and articulatory placement and sequencing disorders), hearing loss, auditory processing deficiencies, language impairment, and/or cognitive impairment (Kent & Vorperian, 2013). Informal assessment of hearing screening results, hearing status and/or hearing history (when available) revealed no

consistent pattern between hearing loss and speech intelligibility. Variability could also be due to differences in speech therapy services received. Of the 52 participants with DS with information available, 48 speakers had a history of speech therapy. No consistent pattern of intelligibility was observed among the four participants who did not receive speech therapy. However, detailed information about type and frequency of speech therapy was not collected, and these variables likely influence intelligibility. Further research is needed to examine these factors, and their likely interactions, as they relate to intelligibility. As emphasized by Karmiloff-Smith et al. (2016), individual differences in DS are notable across genetic, cellular, neural, cognitive, behavioral, and environmental observations. The authors suggested that examination of these differences is one of the best approaches to understand genotype/phenotype relations in DS. The striking heterogeneity in persons with DS makes it difficult to classify research participants in groups according to such potentially influential factors as hearing status, type and severity of craniofacial anomaly, degree of hypotonia in the oral musculature, and dosage and type of speech therapy. Determining the effects and interactions of these variables would require a large sample.

Speaker Sex and Age

Analysis of speaker sex showed that females with DS were more intelligible than males, in agreement with results reported by Kumin (2006), Martin et al. (2009) and Roberts et al. (2005). A possible explanation is that speech intelligibility of girls with DS develops at a faster rate than that for boys with DS through age 15, as shown in Figures 2a-b and 3. This developmental trend is similar to that in TD children, as most TD girls ages 6 years and younger master phonemes earlier than TD boys (Smit, Hand, Freilinger, Bernthal, & Bird, 1990). For adults with DS, greater

variability was more evident among males than females. The influence of speaker sex on speech intelligibility in DS needs further investigation, especially with respect to causal factors that may include possible anatomic differences of the vocal tract.

A brief comment should be made on the TD data. Based on reports in the literature (Coplan & Gleason, 1988), we expected that TD 4-to-5-year olds would achieve nearly complete intelligibility for a single-word test. However, the mean words correct for TD 4-to-5-year-olds was 76% for girls and 81% for boys, while TD 6-to-7-year-old boys and girls had greater than 90% words correct. Our results support the conclusion of Hustad et al., 2015 that TD children are not fully intelligible until about 6 years of age. It should be emphasized that our results pertain to a single word test based on highly familiar lexical items and with a limited repertoire of phonetic elements. It is likely that speech intelligibility would be even lower for more complex speech materials. Despite the use of stimuli that were developmentally appropriate for speakers of all ages, TD 4-to-5 and 6-to-7-year-olds were more intelligible than most persons with DS of all ages, highlighting the speech intelligibility difficulties faced by many persons with DS.

Contribution of Consonants and Vowels to Intelligibility

As expected, total consonants correct contributed to word intelligibility in DS. Higher accuracy for vowel production than consonant production has been reported for persons with DS (Dodd & Thompson, 2001; van Bysterveldt, Gillon, & Foster-Cohen, 2010). In the present study, comparison of vowel and consonant contributions to intelligibility are limited because of the structure of the speech material. The relative distribution of consonants and vowels in the stimuli words was such that 15 of 20 words contained two consonants and one vowel.

The present study was designed to examine aspects of vowel production in relation to word intelligibility, and we were interested to see if and how vowel errors can be used as a gauge of speech intelligibility in DS. Vowel errors are likely to occur due to several factors that also contribute to inter-speaker variability, including anatomic, acoustic, and/or physiological differences in persons with DS.

Intelligibility of Individual Vowels

High vowels were more intelligible and developed earlier than low vowels for both males and females with DS (see Figure 6). For most children with DS, high vowels developed prior to 4 years of age, while intelligibility of low vowels continued to increase through 12 years of age for vowel /æ/ and through 16 years of age for vowel /ɑ/ (see Figure 6). Additionally, persons with DS of all ages had similar intelligibility to TD children ages 4 to 7 years for high vowels, although persons with DS had more variability. Furthermore, mean intelligibility of low vowels was greatly reduced for persons with DS as compared to high vowels, and vowel /ɑ/ had the greatest variability (see Figure 6). Males with DS had a lower mean intelligibility for each individual vowel and greater variability as compared to females in the present study. Bunton et al. (2007) studied five men with DS, and is the only other study that has used an analytical approach to study vowel intelligibility. The present finding of reduced intelligibility for low vowels agrees with Bunton et al.'s (2007) report that men with DS had more errors in the high-low dimension of vowel production than in the front-back dimension. Present results suggest that this trend may be true for females and males with DS of all ages. Bunton et al. also reported no errors in vowel /i/ for men with DS, and in the present study vowel /i/ had the fewest errors of the corner vowels.

The difficulty in the production of low vowels in DS may be explained as follows. Precise tongue-jaw coordination is needed to produce low vowels, but little tactile feedback is provided between the tongue and the maxilla. Low and high vowels may differ not only in sensory feedback but also in mechanical support from peripheral structures such as the molars, palate, and pharynx. The term *bracing* has been used to describe a lingual posture in speech wherein the tongue is in contact with a rigid vocal-tract surface, such as the teeth or palate (Gick, Allen, Roewer-Després, & Stavness (2017). The nature of the bracing differs with the articulatory features of vowels. For example, the high vowels are likely to be braced by the palate and the maxillary dentition whereas the low-back vowels are more likely to be braced by the pharynx. Another factor to be considered is that the articulatory distance between low vowels can be smaller than that between high vowels. Possibly, the tongue movement between low vowels may be especially restricted due to anatomic limitations in persons with DS. The result that the low dimension of vowel production is more related to intelligibility than the high dimension is in line with previous reports of a smaller mid and lower facial skeleton. This anatomic feature may contribute to a reduced articulatory working space and a consequent reduction of the vowel acoustic space in DS.

Limitations and Future Directions

To our knowledge, this is the first study to use an open-set single-word intelligibility test to determine the contribution of specific phonetic factors to speech intelligibility in children and adults with DS. Although the design of this study facilitated phonetic analysis across a wide age range, intelligibility data at the word level may not be representative of speakers' intelligibility during connected speech tasks such as conversation. Further research analyzing intelligibility at

the phrase or sentence level is needed to obtain a more comprehensive picture of intelligibility reduction in DS, and research should account for the considerable intra- and inter-speaker variability within this population. An important caveat is that children with DS appear to be capable of single-word production as compared to more complex utterances (Grieco et al., 2015), which makes these materials more suitable than complex utterances for testing basic articulation. The speech sample used in the present study was specifically designed to examine the corner vowels. It would be informative to use a speech sample that incorporates consonant contrasts and additional vowels.

Longitudinal intelligibility data from persons with DS are needed to examine individual developmental patterns using both global and analytic methods. Additionally, data from young children with DS (younger than age 4 years) should provide a comprehensive view of intelligibility during development. Other variables that warrant further study are (1) the effects of listener familiarity and the context of the utterance, (2) speaker-related factors, such as voice and prosody, and (3) the relationship between acoustic and intelligibility measures. As recommended by Miller (2013), combining "diagnostic" or analytic intelligibility testing with global measures of intelligibility is likely necessary for a full understanding of speech intelligibility in clinical populations. There are no specific data on underlying mechanisms that negatively impact speech and there is no systematic assessment procedure available for evaluating the speech of adults with Down syndrome.

Clinical Implications The word-intelligibility test used in the present research provides both an overall measure of intelligibility and an analysis of some phonetic factors that account for variations in overall intelligibility. Bunton et al. (2007) used a single-word test to determine

factors underlying reduced intelligibility in five men with DS. The most disrupted features involved: (a) word-initial and word-final cluster simplification, and (b) contrasts of tongue-posture, control, and timing (e.g., high-low vowel, front-back vowel; place of articulation for stops and fricatives). These features may be particularly important for assessing speech intelligibility in persons with DS and planning treatment that is focused on factors contributing to intelligibility.

Although the present study was not designed to examine consonant production in detail, it is clear that consonants carry a significant load in speech intelligibility. The difficulties with consonants noted in this study are especially remarkable given that stimuli words were comprised mostly of consonants that are acquired early in typical development (e.g., /b p h t d k/). Given the developmental and lifespan perspective of this general project, the selection of words was guided by the criteria of familiarity, high phonological neighborhood density, and relative ease of production. More demanding versions of the test could be developed to explore additional phonetic aspects, but even the current version appears to be applicable and informative for studies of speech intelligibility in children and adults with DS.

The protracted improvement in speech intelligibility observed in this study provides justification for treatment targeting intelligibility. It is noteworthy that many individuals with DS achieve relatively high levels of intelligibility by the age of 16 years. Although some adults with DS have persisting low levels of intelligibility, the prospects appear encouraging for many individuals to achieve speech that is reasonably intelligible to unfamiliar listeners. In view of the continued improvement in intelligibility in DS from ages 4 to 16 years, it is possible that intervention efforts during this period would accelerate intelligibility gains. This is not to say

that treatment at earlier or later ages is inappropriate, but only to suggest that individuals between the ages of 4 to 16 years appear to be good candidates for intervention focused on improving intelligibility. The protracted intelligibility improvements in DS, compared to that of typically developing children who are intelligible by age 7, may be evidence for self-adaptations and maturational processes that could be reinforced or even expanded with focused treatments.

One novel result of the present study is that reduced contrast between low-front and low-back vowels is a factor accounting for reduced intelligibility in DS. A likely explanation is that these vowels have a slower development than high-front and high-back vowels and therefore account for changes in intelligibility with maturation. The low vowels may offer insight into the developmental processes underlying intelligibility and therefore may be sensitive for purposes of assessment. They also could serve as suitable targets for early phases of treatment, not only for more correct vowel production per se but also to leverage better control of lingual articulation that could lead to more accurate consonant production. The production of consonants is predicated in part on the mastery of vowels, which require control of tongue body articulation. Strategies for the treatment of vowels in children are discussed by Gibbon (2013) and by Speake, Stackhouse, and Pascoe (2012). Mahler and Jones (2012) reported that treatment of two adults with DS using the Lee Silverman Voice Treatment (LSVT) had positive outcomes, leading to improved vowel production in one individual. More generally, there appears to be promise in the use of principles of motor learning to improve motor skills in persons with DS (Horvat, Croce, & Fallaize, 2016), and intervention for speech

could incorporate the same approach, especially if due consideration is given to the structural, motor, sensory, and cognitive issues pertaining to speech production.

It is also possible that combinations of behavioral and other kinds of treatment can facilitate gains in speech intelligibility in persons with DS. For example, interventions that directly alter the anatomy of the orofacial complex, such as a stimulating palatal plate (Matthews-Brzozowska, Cudzilo, Walasz, & Kawala, 2014), may contribute to improved intelligibility by helping speakers to overcome anatomic limitations. Concurrent behavioral intervention in concert with such anatomically directed treatment may achieve desired functional outcomes. However, it cannot be assumed that all interventions affecting the tongue or oral cavity will lead to improved speech. Partial glossectomy does not necessarily result in improved speech intelligibility in either DS or Beckwith-Wiedemann syndrome (Margar-Bacal, Witzel, & Munro, 1987; Van Lierde et al., 2012). A variety of interventions have been introduced to improve appearance, reduce drooling, and facilitate swallowing in DS, but it is important to ensure that such treatments do not have deleterious effects on speech production. Clinical speech assessment during treatment regimens is highly advisable to ensure that speech is not negatively affected. It is also imperative to keep in mind the individual differences notable in the DS population.

Bittles et al. (2007) commented on an extraordinary point of progress in improving the lives of individuals with DS: “A highly significant change in the survival of people with DS has occurred during the last two generations, with life expectancy estimates increasing from 12 to nearly 60 years of age” (p. 224). This increased life expectancy would be accompanied by improved quality of life for many individuals through enhanced communication skills.

Conclusion

This study makes a novel contribution by showing that an open-set, single-word intelligibility test can be used to determine the developmental pattern of word-level speech intelligibility and to examine phonetic factors relating to intelligibility scores. The results reveal that intelligibility gains are evident especially in the age range of 4 to 16 years, that females have higher intelligibility scores than males, and that the corner vowels make different contributions to speech intelligibility. Results of this perceptual study add to the understanding of the pervasive intelligibility difficulties experienced by children and adults with DS. Clarifying the basis of these speech intelligibility challenges is a critical step to direct future research efforts and design interventions that will enhance communication, and subsequently, quality of life.

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Tables:

Table 1. Descriptive statistics for the proportion of vowels correct across speech recordings from persons with DS (N=total number of speech recordings, generally several recordings per speaker). The table displays the statistics broken down by vowel type for all (top panel), males only (middle panel) and females only (bottom panel).

	Vowel type	N	Minimum	Maximum	Mean	SD
Overall	/i/	143	.42	1.00	.94	.11
	/u/	143	.17	1.00	.93	.15
	/æ/	143	.12	1.00	.82	.24
	/ɑ/	143	.09	1.00	.73	.25
Male	/i/	75	.42	1.00	.91	.14
	/u/	75	.17	1.00	.90	.19
	/æ/	75	.17	1.00	.74	.26
	/ɑ/	75	.09	1.00	.69	.26
Female	/i/	68	.80	1.00	.98	.05
	/u/	68	.60	1.00	.97	.07
	/æ/	68	.12	1.00	.89	.17
	/ɑ/	68	.24	1.00	.78	.23

Note. N = speaking occasions

Table 2. Mean and standard deviation (*SD*) for percent vowels correct for TD boys and girls. The only speaker group with a mean less than 95% and standard deviation greater than 7% is marked by an asterisk.

Vowel	Sex	4-to-5-year-olds: Percent Correct Mean (*SD*)	Sex	6-to-7-year-olds: Percent Correct Mean (*SD*)
/i/	F	100 (0)	F	100 (0)
	M	100 (0)	M	100 (0)
/u/	F	99 (2)	F	99 (2)
	M	98 (4)	M	98 (5)
/æ/	F	97 (5)	F	98 (5)
	M	98 (4)	M	98 (4)
/ɑ/	F	89 (10)*	F	97 (5)
	M	95 (7)	M	98 (3)

Figure Captions:

Figure 1. An example of stimuli presented to speakers to elicit the production of a target word, which consisted of an image of the target word and the printed word displayed below it, along with a played audio recording of the target word.

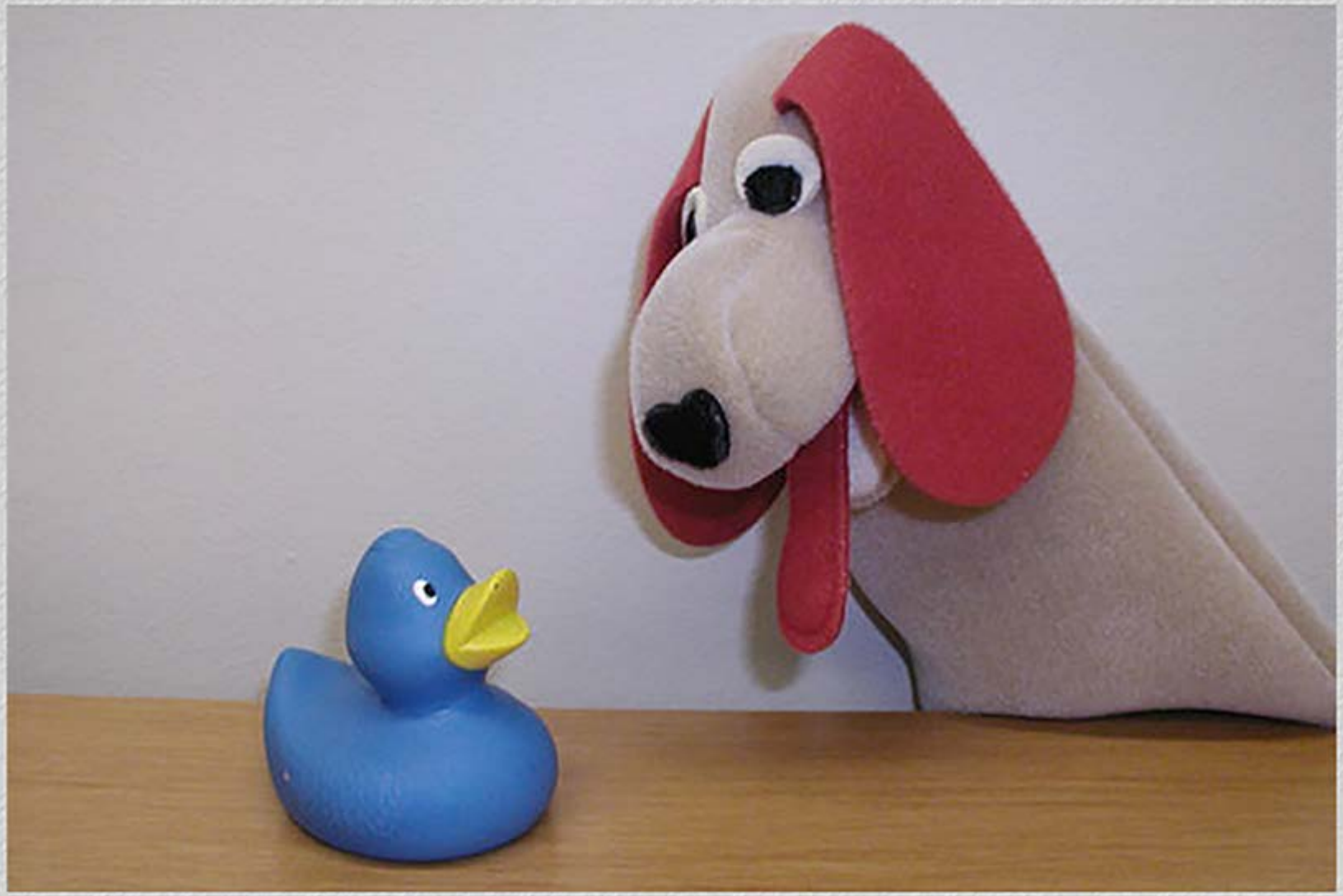
Figures 2a-b. Words correct for all speakers, with females shown in red and males in blue. Data from TD speakers are represented by box and whisker plots. The box plots show the 25th and 75th percentiles, with a dotted line representing the mean and a solid line representing the median. The whiskers show the 10th and 90th percentiles, and outlying data are shown as dots. Data from persons with DS are shown by scatter plots. Speakers with only one speech recording are represented by triangles, and speakers with multiple speech recordings are denoted by squares or circles connected by dotted or solid lines.

Figure 3. Words correct for persons with DS, with data from males shown by blue triangles and data from females shown by red circles. Colored lines indicate the equation for generalized linear mixed models for males and females, and colored bands indicate 95% confidence intervals for the models.

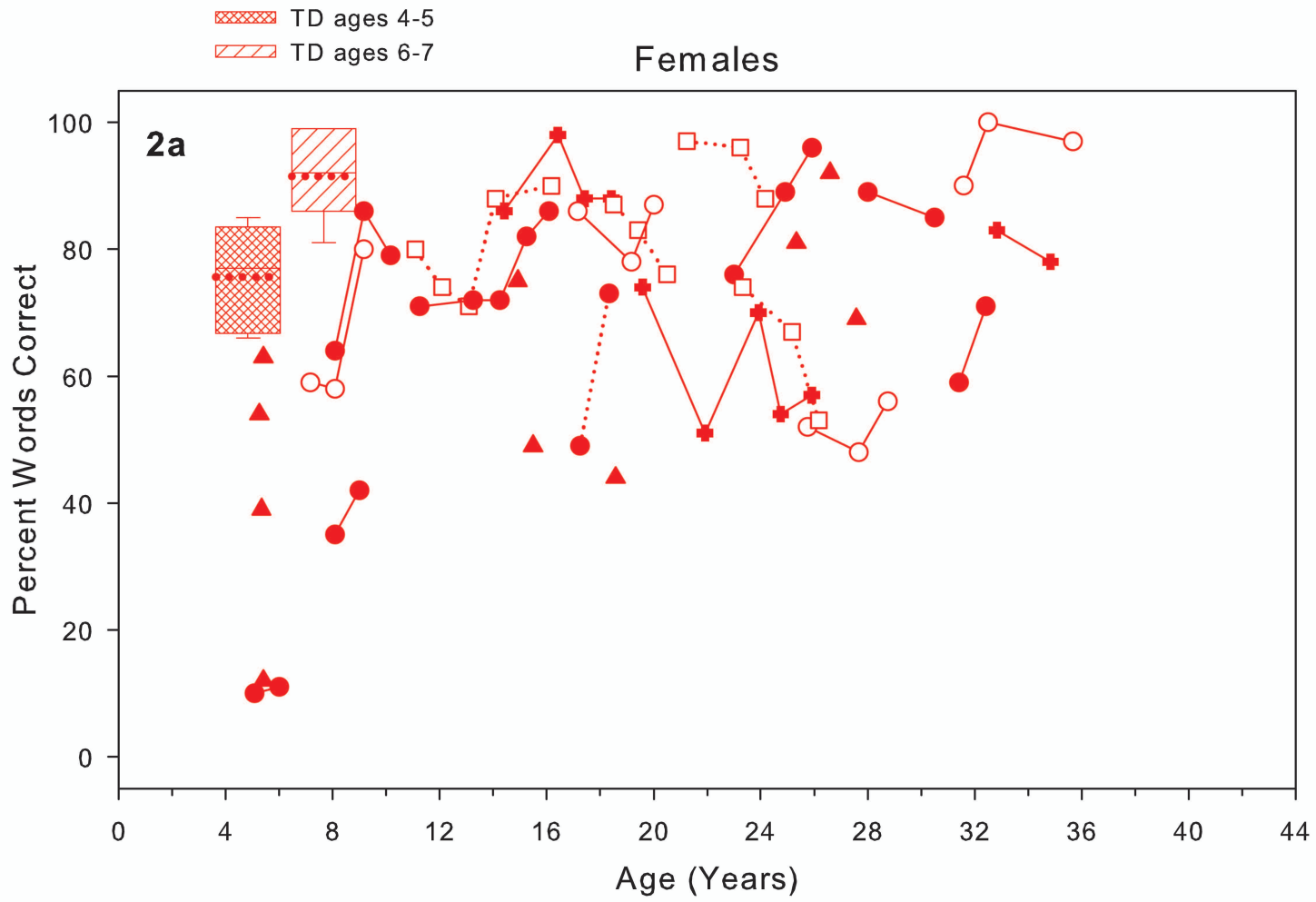
Figures 4 a-c. Relationship between percent words correct for persons with DS and total consonants correct (4a), and total vowels correct (4b). The percent of words perceived as correct was averaged across five listeners' responses, similar to the calculations used to create Figures 2 and 3.

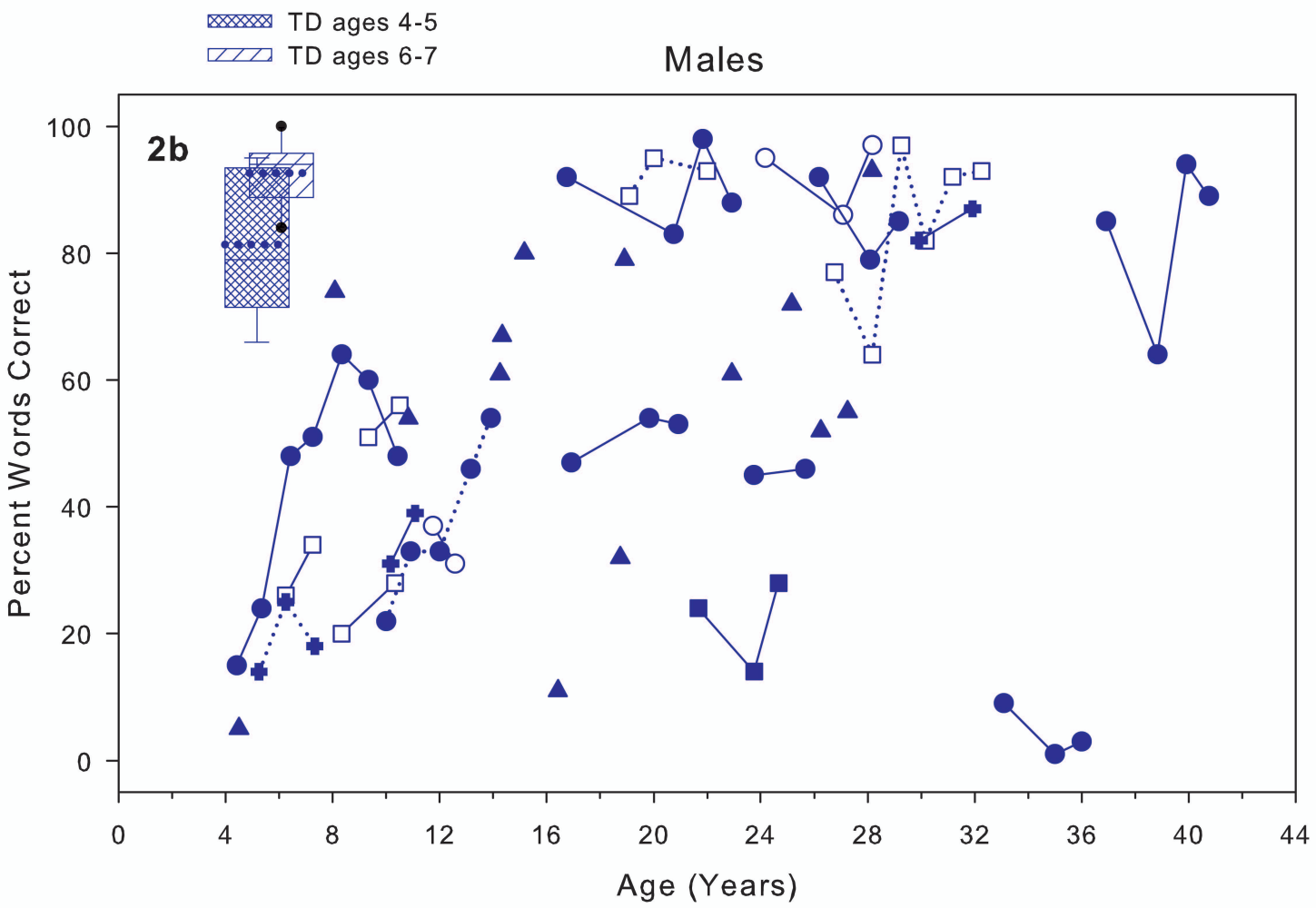
Figures 5 a-e. Relationship between percent words correct for persons with DS and initial consonants correct (5a), and final consonants correct (5b), and individual vowels correct /i/, /u/, /æ/ and /ɑ/ (5c-5f).

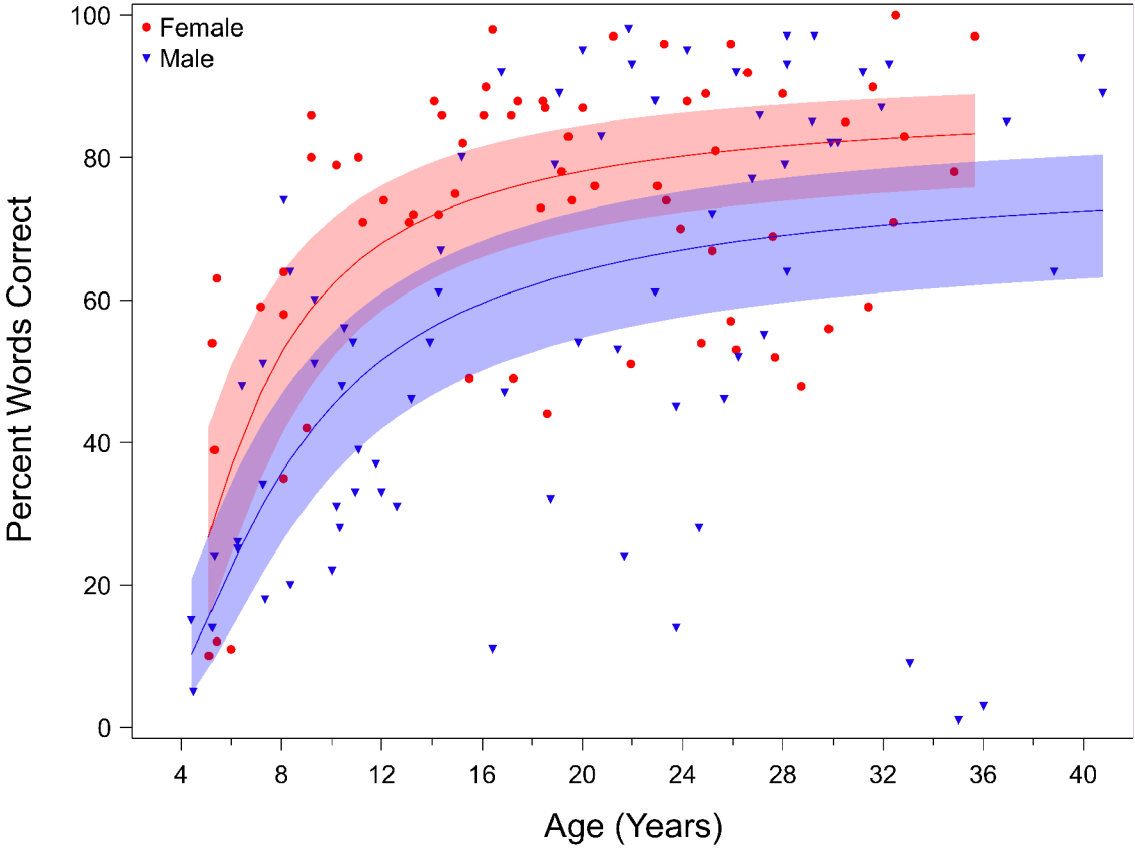
Figure 6. Percent correct for vowels /i/, /u/, /ɑ/, and /æ/ for persons with DS as a function of age, with females shown in red and males in blue. Speakers with a single session are represented by triangles, while speakers with repeat sessions are represented by circles. Refer to Table 2 for percent vowels correct data for TD speakers.

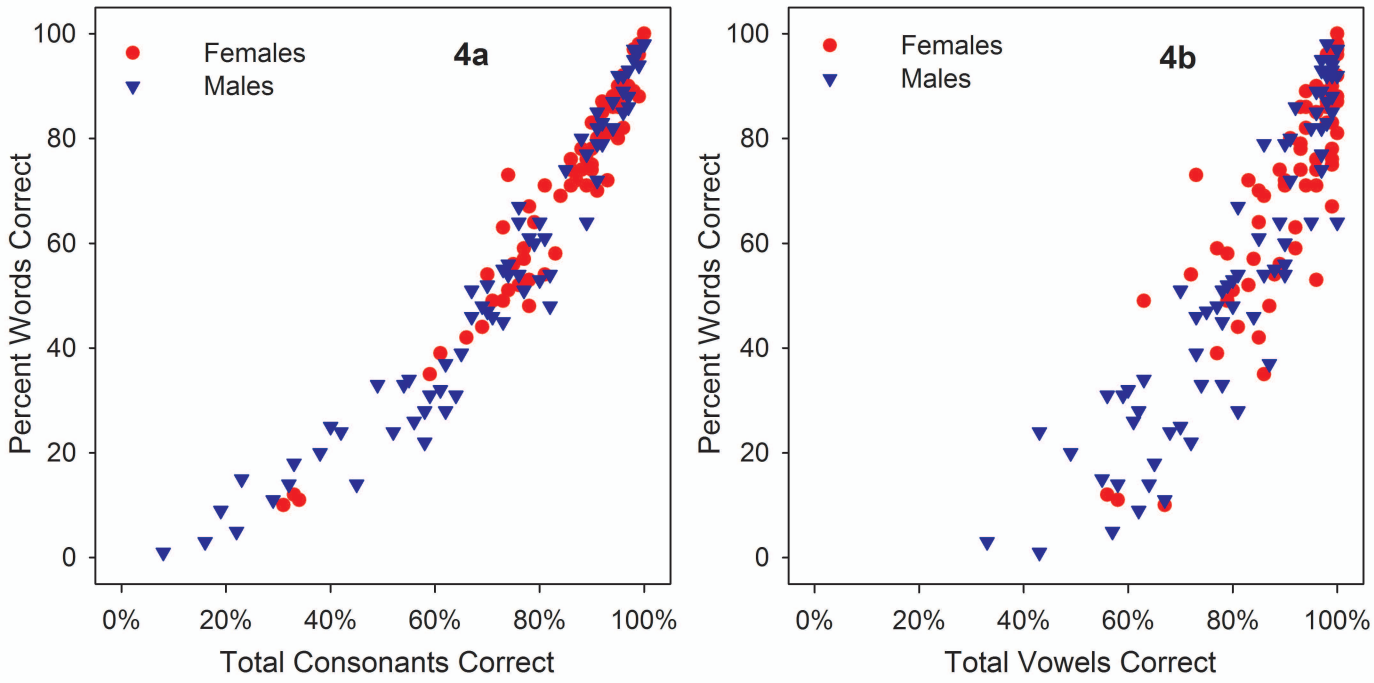


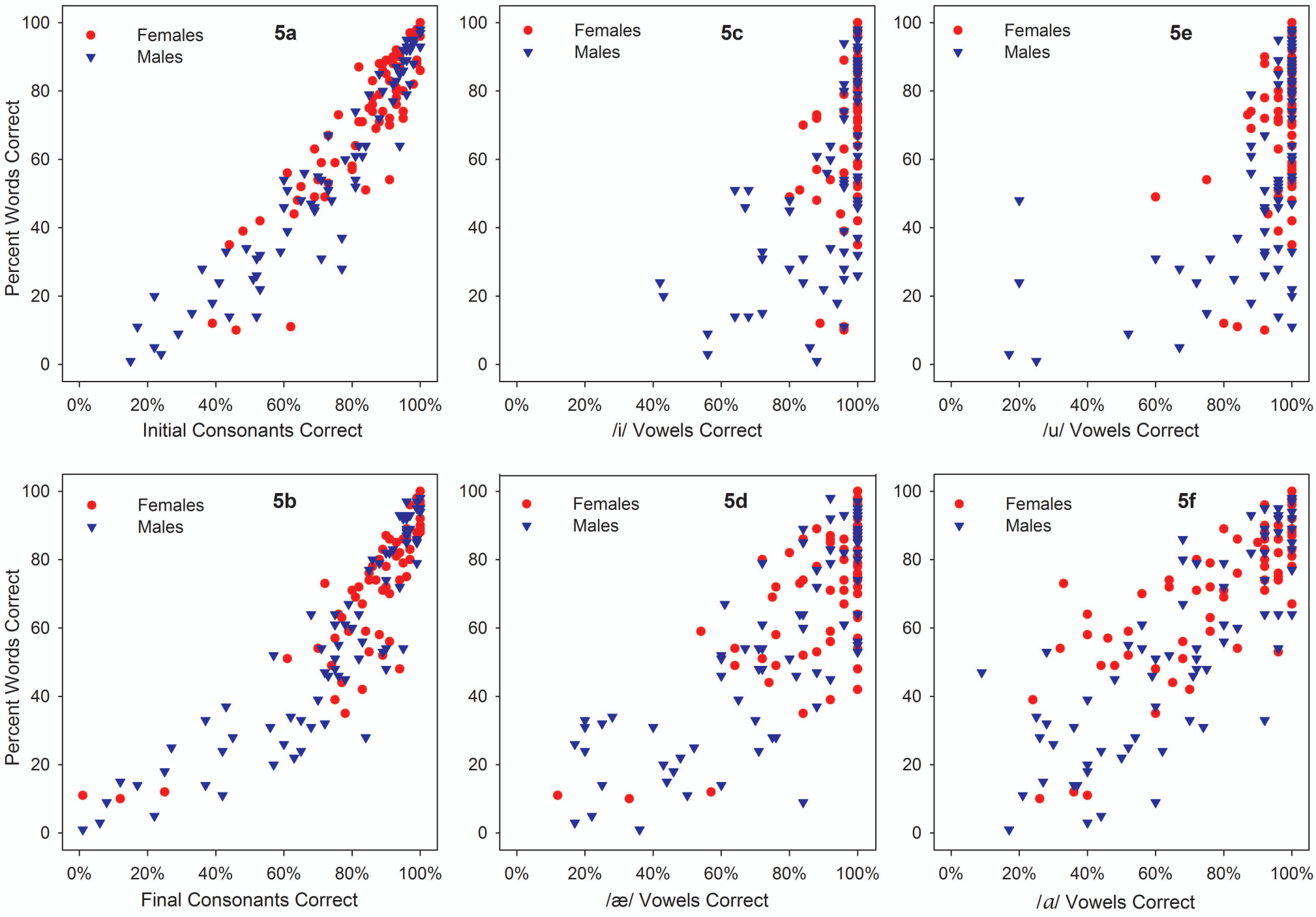
Duck

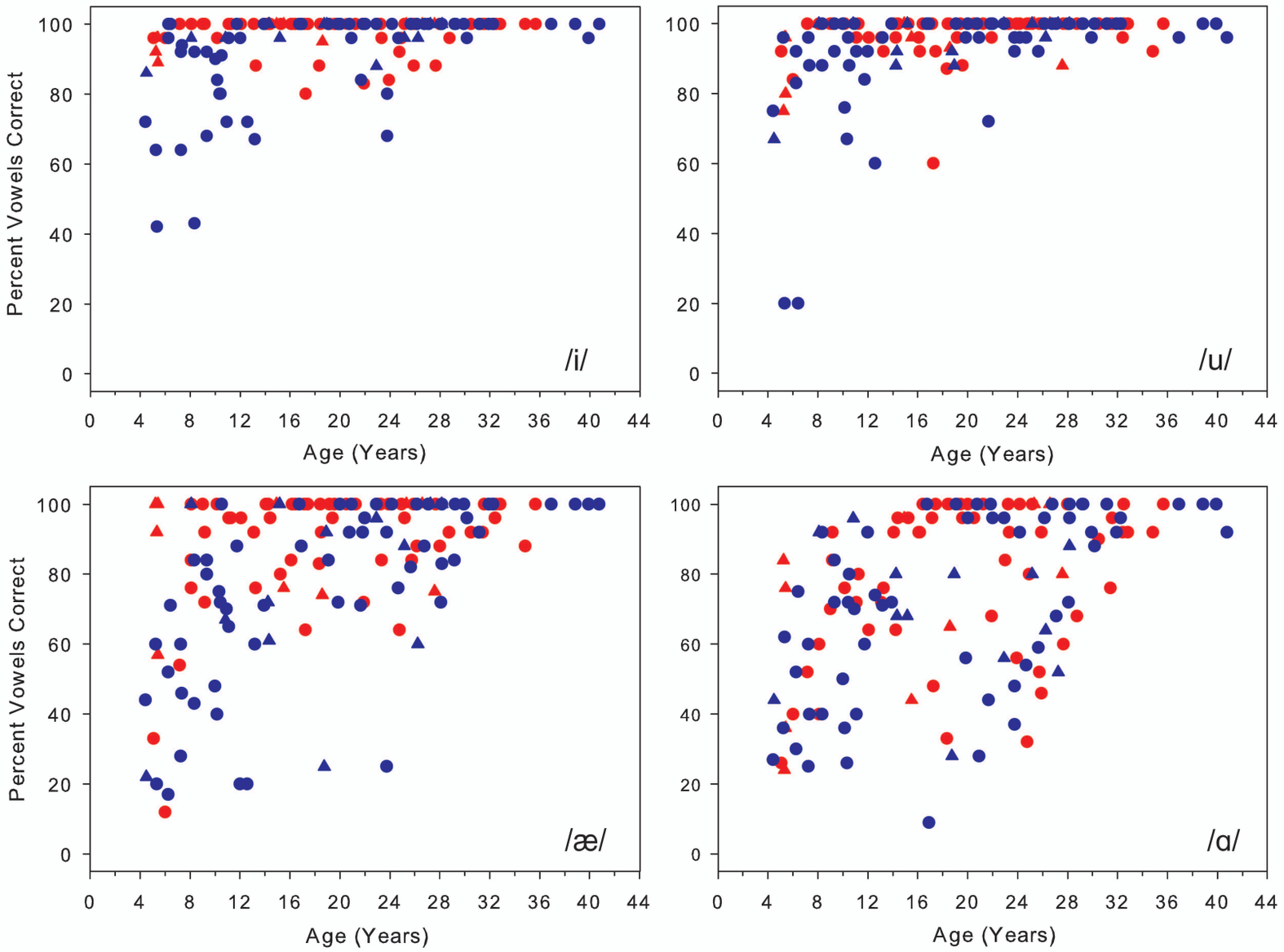

TD ages 4-5
TD ages 6-7
Females
2a
Percent Words Correct
Age (Years)
0
20
40
60
80
100
0
4
8
12
16
20
24
28
32
36
40
44


TD ages 4-5
TD ages 6-7
Males
2b
Percent Words Correct
Age (Years)
0
20
40
60
80
100
0
4
8
12
16
20
24
28
32
36
40
44


Female
Male
Percent Words Correct
Age (Years)
0
20
40
60
80
100
4
8
12
16
20
24
28
32
36
40


4a
Females
Males
Percent Words Correct
Total Consonants Correct
0
20
40
60
80
100
0%
20%
40%
60%
80%
100%
4b
Females
Males
Percent Words Correct
Total Vowels Correct
0
20
40
60
80
100
0%
20%
40%
60%
80%
100%


5a
Females
Males
Percent Words Correct
Initial Consonants Correct
5c
/i/ Vowels Correct
5e
/u/ Vowels Correct
5b
Final Consonants Correct
5d
/æ/ Vowels Correct
5f
/ɑ/ Vowels Correct
0
20
40
60
80
100
0%
20%
40%
60%
80%
100%


Percent Vowels Correct
Age (Years)
/i/
/u/
/æ/
/ɑ/
0
20
40
60
80
100
0
4
8
12
16
20
24
28
32
36
40
44