Speech Production in Children and Adults with Down Syndrome: Perceptual and Acoustic Data

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Down syndrome (DS) - Trisomy 21

1. The most common form of intellectual disability (1 in 700-800 births).

2. One of the most complex genetic perturbations compatible with survival.
   ➢ In only 2 generations, life expectancy increased from 12 to nearly 60 years.

3. Speech intelligibility often is compromised and can be a lifelong problem
   (Kent & Vorperian, 2013; Kumin, 1994; Wild et al., 2018).
   ➢ Voice enabled technology -- Google initiative: “Project Understood”.

4. The speech impairments relate to multiple factors, such as:
   • motor impairments (hypotonia, dysarthria, apraxia of speech)
   • phonological delay or disorder
   • hearing loss
   • intellectual disability
   • craniofacial and laryngeal dysmorphologies
Dysmorphologies and Dysfunctions

- Small midface
- Short and narrow palate
- Pseudomacroglossia
- Stage III malocclusion and anterior open bite
- Constricted airway
- Laryngomalacia
- Hypotonia*
  - Labial
  - Lingual
  - Laryngeal

Conflicting Reports on Speech Disorder in DS--
A Few Examples...

1. Articulatory working space (e.g. vowel space area):
   - **Reduced** (Abolhasanizadeh & Olyiaiee, 2018; Bunton & Leddy, 2010; Moura et al. 2008)
   - **Increased** (Rochet-Capellan & Dohen, 2015)

2. Phonatory dysfunction:
   - **Vocal hyperfunction** (Pebbili et al., in press)
   - **Vocal hypofunction** (Wold & Montague, 1979, Moran & Gilbert, 1982)
   - **Nonmodal phonation** (Jeffery, Cunningham, & Whiteside, 2018)

3. Oral-nasal resonance:
   - **Hypernasality** (Montague & Hollien, 1973; Rolfe, Montague, Tirman, & Vandergrift, 1979)
   - **Hyponasality** (Jones et al., 2019)
   - **Other atypical resonance** (Fourakis, Karlsson, Tilkens, & Shriberg, 2010; Jones et al., 2019)

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Present Study – Research Questions

Overarching Question:
What is the speech subsystem profile in Down syndrome?

1. What are the dominant perceptual features for sustained vowels and short sentences?

2. What are the acoustic characteristics of vowels and fricatives?

How can we distinguish **functional** from **structural** aspects of the speech disorder?
Methods: Participants and Speech Samples

**Down syndrome**
- 82 children and adults
- Ages 3 to 53 years
- 40 females
- 42 males

**Neurotypical**
- 407 children and adults
- Ages 4 to 92 years
- 212 females
- 195 males

**Speech sample**: Sustained vowels, monosyllabic words, short sentences

Appropriate for individuals with limited cognitive and language abilities over the age range
Speech Samples for this Report

Five words for each corner vowel*

/i/- bead, bee, eat, sheep, feet  
/u/- boo, boot, zoo, hoot, shoe  
/æ/- bath, bat, cat, hat, sad  
/ɑ/- dot, hop, pot, top, hot

*Also used in a single-word intelligibility study (Wild et al., AJSLP, 2018)

Three short phrases or sentences

The blue duck quacks, Pop the bubble, Cars go beep beep

Sustained vowel  /ɑ/

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Perceptual Ratings – 22 Features in 4 Subsystems

1) Phonation/respiration:
   - Roughness
   - Breathiness
   - Strain
   - Pitch variability
   - Pitch break
   - Loudness variability
   - Dysphonia severity

2) Articulation and resonance:
   - Imprecise consonant articulation
   - Distorted vowels
   - Irregular articulatory breakdown
   - Atypical resonance
     - Hyponasality
     - Hypernasality
     - Cul de sac resonance
     - Other atypical resonance

3) Suprasegmental:
   - Disturbance of speech rhythm
   - Atypical intonation

4) Overall communication effectiveness:
   - Reduced intelligibility
   - Atypical overall quality of speech
   - Dysfluency
Perceptual ratings

• Perceptual ratings were completed by 3 listeners who were highly familiar with speech production in DS and who participated in consensus training.

• Features were rated on a monopolar 5-point equal-appearing interval scale. Ratings were made for both sustained vowels and sentences.

Vowel Rating Scale

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Ratings of Vowels – Principal Components Analysis

**Principal Component 1**
High ratings of:
- Roughness
- Dysphonia severity
- Strain
- Pitch variability
- Loudness variability

PC1 = vocal hyperfunction

**Principal Component 2**
High ratings of:
- Breathiness
- Atypical resonance

and low rating of:
- Strain

PC2 = vocal hypofunction

**Principal Component 3**
High rating of *Pitch breaks* and
low rating of *Loudness variability*

PC3 = pitch & loudness control
Ratings of Sentences – Principal Components Analysis

Principal Component 1
High ratings of:
- Imprecise consonants
- Distorted vowels
- Disturbance of speech rhythm
- Atypical intonation
- Breathiness
- Atypical resonance
- Dysphonia severity

PC1 = severity across systems

Principal Component 2
High ratings of:
- Roughness
- Dysfluency
- Dysphonia severity

Principal Component 3
High ratings of:
- Irregular articulatory breakdown
- Strain
Acoustic Measures

- Formant frequencies (F1-F4) of corner vowels
- Spectral moments (M1-M4) of the fricatives /s/ and /ʃ/
- Multidimensional Voice Program (MDVP)
- Cepstral Spectral Index of Dysphonia (CSID)

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Differences in:
- Dispersion of F1 and F2
- Overlap of vowels, esp. low vowels /æ/ & /ɑ/

Interpretation:
Anatomic restriction of tongue position in DS

/æ/- /ɑ/ difficulty:
- Perceptual study by Wild et al (*AJSLP*, 2018)
- Acoustic study by Carl (*JSLHR*, in press)
For /ʃ/ both M1 and M2 are larger for DS than TD.
For /s/ M1 is equal for DS and TD, but M2 is larger for DS.

Inference:
Palatal dysmorphology affects the palatal fricative /ʃ/.
Acoustic Analyses of Phonation

• Background studies
  • SLPs can identify DS from sustained vowels (Moran, J. Com. Dis., 1986).

• Current study assessed phonation using MDVP & CSID.
  ➢ Dysphonia severity score correlated mildly but significantly with:
    ▪ MDVP Fundamental Frequency Variation (r=.373, p=.006) &
    ▪ CSID (r=.392; p=.001).
  ➢ MDVP measures significantly different between DS and TD:
    ▪ Fundamental Frequency Variation (t-test p=.000)
    ▪ Peak Amplitude Variation (t-test p=.000)
  ➢ CSID also significantly different between DS and TD (t-test p=.000).

There may be different acoustic signatures of the voice disorder in DS
Conclusions

The Speech Disorder in Down Syndrome:

• Reflects the phenotypic heterogeneity in the syndrome.
• Results from impairments distributed across the systems of speech production.
• Is rooted in both dysmorphology and disordered motor control.
  • Hypothesis: Structure-function interaction through the lifespan.
• Can be better understood through a combination of methods (anatomic, physiologic, acoustic, and perceptual)
  • More interdisciplinary research!
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Thank you for your attention!
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