EARLY SPEECH DEVELOPMENT IN WILLIAMS SYNDROME

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• We are grateful to the children and their families for their participation in this study.
Disclosure

- None of the presenters has any conflicts of interest associated with this presentation to disclose.
Williams Syndrome

• A genetic neurodevelopmental disorder resulting from a microdeletion of about 26 genes on chromosome 7 (7q11.23)

• Occurs in approximately 1 in 7,500 births

• Supravalvar aortic stenosis

• Hypercalcemia
Williams Syndrome: Characteristics

• Intellectual ability range: severely challenged to average
• Mean full-scale IQ: 64
• Considerable weakness in visuospatial construction
• Relative strength in verbal short-term memory
• Relative strength in concrete vocabulary
• Gregarious and empathic
• Socially disinhibited
• Anxious
• ADHD
WS Previous Findings

• CDI measures of vocabulary and grammar development highly correlated with laboratory play measures (Mervis, 2012)

• Onset of meaningful words delayed
  • 10-word vocabulary: Median: 24.75 months, Range: 15.57 – 52.99 months
  • 50-word vocabulary: Median: 28.02 months, Range: 20.83 – 61.21 months
  (Pitts, Adelson & Mervis, 2014)
WS Previous Findings

Age of acquisition of 10-word expressive vocabulary strongly correlated with:
- age of acquisition of 50-word vocabulary ($r_s = .86$)
- age of first novel word combinations ($r_s = .81$).

Ages of acquisition of 10- and 50-word vocabularies and first novel word combinations all strongly negatively correlated with intellectual abilities at age 4 years ($ps < .0001$)

(Pitts, Adelson, & Mervis, 2014)
WS Previous Findings, cont.

- However, typical relations between language and other communicative milestones (e.g., pointing) do not necessarily hold

- Once vocabulary and grammatical development begin, they proceed consistently and relatively rapidly for most, but not all, children with WS (Mervis, 2003, 2006; Mervis et al. 1999; Mervis & Bertrand, 1997)
WS Previous Findings

- For example, mean expressive vocabulary size (CDI) falls at the 1\textsuperscript{st} percentile from 18 to 48 months – but increases at a consistent rate (DeLapp, Pitts & Mervis, 2013).
WS Previous Findings, cont.

• Onset of canonical babbling delayed
  Mean: 17.7 mos. (Masataka 2001)

Very little known about the nature or progression of speech production in early vocalizations in toddlers with Williams syndrome
Overview of the Study

We consider four aspects of early speech production that have been proposed to be scientifically and/or clinically useful:

- Variety of phonetic repertoires
- Typicality of phonetic repertoires
- Complexity of phonotactic repertoires
- Word-babble comparisons
Variety of Phonetic Repertoires

• Stoel-Gammon (1985): Mean number of different consonants in at least two different words increases from 15-24 months
• Stoel-Gammon (2002): Mean number of different consonants in at least one word increases from 21-24 months
• Selby et al. (2000): Number of different vowels produced at least twice by at least ¾ children in words and babble mixed increases from 15-36 months
Typicality of Phonetic Repertoire

Stoel-Gammon (1985)

• Consonants in initial position, occurring in at least two different words:
  • 90% of 18-month-olds: [b, d]
  • 50% of 18-month-olds: [m, n, w, h]
  • 90% of 24-month-olds: [b, d]
  • 50% of 24-month-olds: [m, n, w, h, t, k, g, f, s]

• In general:
  • initial C’s are voiced, anterior
  • final C’s are voiceless and alveolar
Complexity of Phonotactic Repertoire

1. Mean Babble Level (MBL; Stoel-Gammon 1989)
   Level 1: no supraglottal non-glide C
   Level 2: 1 supraglottal non-glide C
   Level 3: 2 different supraglottal non-glide Cs

Morris (2010): lower-than-expected MBL at 20 months indicates risk of not developing meaningful speech by 24 months of age (Fasolo et al., 2008; Stoel-Gammon, 1989)
Complexity of Phonotactic Repertoire

2. Percent C+V syllables (Rvachew et al. 2005)
Includes CV, VC, CVC. Related to “canonical syllable ratio” (CSR) although canonical syllables must also have mature timing and phonation (Oller 1986).

Children with Bronchopulmonary Dysplasia (BPD) have lower CSRs than healthy infants and than preterm infants w/o BPD although they do not differ statistically on % of syllables of CV shape (Rvachew et al. 2005). Children with otitis media in the first 6 months of life have lower CSRs through 18 months. Those with lower CSRs have smaller expressive vocabularies at 18 months (Rvachew et al. 1999).
Complexity of Phonotactic Repertoire

3. Mean # syllables/utterance (Rvachew et al. 2005)

- decreased at 18 months in children with BPD
- steady in TD infants and premature infants w/o BPD
- overall from 8 – 18 months no statistically significant main effects of Age or Group

Note: Age x Group interaction not tested
Complexity of Phonotactic Repertoire

4. % monosyllables
Varies by language background (American English vs. French) @14 – 17 months (in words) (Vihman, Velleman & Lleo, unpublished)

5. % closed syllables (VC and CVC syllables)
Varies by language background @14 – 17 months (in words) (Vihman, Velleman & Lleo, unpublished)
Word-Babble Comparisons

- Stops more frequent than nasals in both words and babbles (Boysson-Bardies & Vihman, 1991)
- Babbles more likely than words to be multisyllabic
- Babbles less likely to include consonants
- Consonants in babbles more varied
- Alveolar/dental consonants more frequent in babble
- Labial consonants predominate in words (Vihman et al., 1985)
- Vowels more varied in babble (Selby et al., 2000)
The Present Study

- Longitudinal speech profiles of 8 toddlers with WS at 18 and 24 months
- Words vs. babble
- Focus on four aspects:
  - Consonant and vowel variety
  - Typicality of phonetic repertoires
  - Phonotactic complexity
  - Babble-word comparisons
Participants

• 8 toddlers with confirmed genetic diagnoses of WS
• Four girls; four boys
• Focus on group data from two time points:
  • 18 months (17.4-19.4 months)
  • 24 months (24.1-25.9 months)
Participants

• # different words produced/30 minutes
  • 18 months: 0 – 3 (median: 1)
    • 3 participants produced no words
    • Words not further analyzed at 18 months
  • 24 months: 0 – 48 words (median: 15)
Participants

- Mullen Early Learning Composite (ELC; similar to DQ) at 24 months: Mean 71 (SD: 17, Range: 53 – 92)
  - General population mean ELC: 100 (SD: 15)
  - For a group of 55 children with WS aged 24 – 31 months: mean ELC 66 (Range 49 – 96)
Methods

• Recorded 30-minute play sessions with a familiar adult
• Transcribed independently by two trained research assistants; agreement on semi-fine transcription ranged from approximately 75 – 95%
• Final transcript derived by consensus
• Data derived from transcripts
• Non-meaningful (babble) and meaningful vocalizations (words) analyzed separately
• At 18 months: 3 children with no words; only 1 child had 3 different words. Thus, only babble analyzed at that age.
• One child had no words even at 24 months.
Vocalization Types by Age

- Significant increases across ages for both types (Babble: $Z=2.37$, $p=0.018$; Words: $Z=2.38$, $p=0.017$).
- Significantly more babbles than words at both ages (18 mo.: $Z=2.52$, $p=0.012$; 24 mo.: $Z=-2.10$, $p=0.036$)
### Variety Measures

#### # Different Initial Consonants in Words and Babble

<table>
<thead>
<tr>
<th>Age</th>
<th>Group &amp; Criteria</th>
<th>Babble</th>
<th>Words</th>
<th>Mixed Vocs</th>
</tr>
</thead>
<tbody>
<tr>
<td>15-24 mo.</td>
<td>TD*: 2 diff. wd.</td>
<td>mean 6.3</td>
<td>mean 2-10</td>
<td>mean 12, SD 4.35</td>
</tr>
<tr>
<td>18 mo.</td>
<td>TD+: 2+ occ.</td>
<td>median 3.5</td>
<td>median 1</td>
<td>median 3.5</td>
</tr>
<tr>
<td></td>
<td></td>
<td>mean 5</td>
<td>mean 1.3</td>
<td>mean 4.1</td>
</tr>
<tr>
<td></td>
<td></td>
<td>range 2-8</td>
<td>range 0-3</td>
<td>range 1-8</td>
</tr>
<tr>
<td>24 mo.</td>
<td>TD*: 2 diff wd.</td>
<td>mean 9.5</td>
<td>mean 4-16</td>
<td></td>
</tr>
<tr>
<td>24 mo.</td>
<td>TD#: 1 occ.</td>
<td>median 14</td>
<td>mean 13.8</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>mean 9-19</td>
<td>mean 8</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>range 3-14</td>
<td>range 0-9</td>
<td>range 3-14</td>
</tr>
<tr>
<td>24 mo.</td>
<td>WS: 2+ occ.</td>
<td>median 6</td>
<td>median 6</td>
<td>median 7</td>
</tr>
<tr>
<td></td>
<td></td>
<td>mean 7.3</td>
<td>mean 5.1</td>
<td>mean 8</td>
</tr>
<tr>
<td></td>
<td></td>
<td>range 3-14</td>
<td>range 0-9</td>
<td>range 3-14</td>
</tr>
<tr>
<td>24 mo.</td>
<td>WS: 1 occ.</td>
<td>median 7</td>
<td>mean 7.3</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>mean 3-12</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

* Stoel-Gammon (1985)
† Rvachew et al. (2005)
‡ Stoel-Gammon (2002)
# Different Initial Consonants

TD mean 24 mo. Words

18 mo. Babble  24 mo. Babble  24 mo. Words
## Variety Measures

### # Different Vowels in Words and Babble

<table>
<thead>
<tr>
<th>Age</th>
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<th>Babble</th>
<th>Words</th>
<th>Mixed Vocs</th>
</tr>
</thead>
<tbody>
<tr>
<td>18 mo.</td>
<td>TD*: 2+ occ.</td>
<td></td>
<td></td>
<td>mean 7 range 6-9</td>
</tr>
<tr>
<td>18 mo.</td>
<td>WS: 2+ occ.</td>
<td>median 9</td>
<td>median 1</td>
<td>median 9 range 4-12</td>
</tr>
<tr>
<td></td>
<td></td>
<td>mean 8.9</td>
<td>mean 1</td>
<td>mean 9.1 range 4-12</td>
</tr>
<tr>
<td></td>
<td></td>
<td>range 4-12</td>
<td>range 0-3#</td>
<td></td>
</tr>
<tr>
<td>24 mo.</td>
<td>TD*: 2+ occ.</td>
<td></td>
<td></td>
<td>mean 9 range 7-10</td>
</tr>
<tr>
<td>24 mo.</td>
<td>WS: 2+ occ.</td>
<td>median 11</td>
<td>median 7</td>
<td>median 12 range 4-12</td>
</tr>
<tr>
<td></td>
<td></td>
<td>mean 9.5</td>
<td>mean 7.3</td>
<td>mean 9.9 range 4-12</td>
</tr>
<tr>
<td></td>
<td></td>
<td>range 4-12</td>
<td>range 3-12#</td>
<td></td>
</tr>
</tbody>
</table>

*Selby et al. (2000)*
# Different Vowels in Babble & Words

TD mean 24 mo.

TD mean 18 mo.

18 mo.

24 mo.
## Typicality Measures

<table>
<thead>
<tr>
<th></th>
<th>90% of Children (7/8 of WS)</th>
<th>50% of Children [m, n, w, h]</th>
</tr>
</thead>
<tbody>
<tr>
<td>TD 18 mo. words*</td>
<td>[b, d]</td>
<td></td>
</tr>
<tr>
<td>WS 18 mo. babble</td>
<td>N/A</td>
<td>[b, d, m, n, h]</td>
</tr>
<tr>
<td>TD 24 mo. words*</td>
<td>[b, d]</td>
<td>[t, k, g, m, n, h, w, f, s]</td>
</tr>
<tr>
<td>WS 24 mo. babble</td>
<td>[d, m, n]</td>
<td>[b, j, h]</td>
</tr>
<tr>
<td>WS 24 mo. words</td>
<td>N/A</td>
<td>[b, d, m, n, h]</td>
</tr>
</tbody>
</table>

*Stoel-Gammon (1985): Occurrence in two different words
WS: Two occurrences
Phonotactic Complexity

<table>
<thead>
<tr>
<th></th>
<th>Average Mean Babble Level (s.d.)</th>
</tr>
</thead>
<tbody>
<tr>
<td>TD 18 mo. babble*</td>
<td>1.65 (0.20)</td>
</tr>
<tr>
<td>WS 18 mo. babble</td>
<td>1.49 (0.32)</td>
</tr>
<tr>
<td>TD 24 mo. babble*</td>
<td>2.03 (0.28)</td>
</tr>
<tr>
<td>WS 24 mo. babble</td>
<td>1.57 (0.32)</td>
</tr>
<tr>
<td>WS 24 mo. words</td>
<td>2.07 (0.22)</td>
</tr>
</tbody>
</table>

*Stoel-Gammon (1989)

Significantly higher MBL in words than babble at 24 months ($Z = 2.20$, $p = 0.028$).
Mean Babble Level by Age

18 mo Babble  24 mo Babble  24 mo Words
**Phonotactic Complexity**

<table>
<thead>
<tr>
<th></th>
<th>Mean % C+V Syllables (range; s.d.)</th>
</tr>
</thead>
<tbody>
<tr>
<td>TD 18 mo. words &amp; babble*</td>
<td>35.45</td>
</tr>
<tr>
<td>WS 18 mo. babble</td>
<td>41.9 (0.5-87; 27.2)</td>
</tr>
<tr>
<td>TD 24 mo.</td>
<td></td>
</tr>
<tr>
<td>WS 24 mo. babble</td>
<td>50.9 (13-91; 28.9)</td>
</tr>
<tr>
<td>WS 24 mo. words</td>
<td>92.9 (86-100; 5.6)</td>
</tr>
</tbody>
</table>

*Rvachew et al. (2005)*

Significantly more C+V syllables in words than babble at 24 months ($Z = 2.20, p = 0.028$).
Phonotactic Complexity

<table>
<thead>
<tr>
<th></th>
<th>Mean # Syllables/Vocalization (median; range)</th>
</tr>
</thead>
<tbody>
<tr>
<td>TD 18 mo.*</td>
<td>1.7</td>
</tr>
<tr>
<td>WS 18 mo. babble</td>
<td>1.39 (1.3; 1.05-2.48)</td>
</tr>
<tr>
<td>TD 24 mo.</td>
<td></td>
</tr>
<tr>
<td>WS 24 mo. babble</td>
<td>1.40 (1.3; 1.14-1.64)</td>
</tr>
<tr>
<td>WS 24 mo. words</td>
<td>1.48 (1.48; 1.3-1.76)</td>
</tr>
</tbody>
</table>

*Rvachew et al. (2005): words and babble

Note: Higher number of syllables per vocalization can be achieved via immature strategies (e.g., reduplicated babble)
Median # Syllables Per Word-like Vocalization

TD mean 18 mo. words
### Phonotactic Complexity

<table>
<thead>
<tr>
<th></th>
<th>Mean % Monosyllables (range; s.d.)</th>
</tr>
</thead>
<tbody>
<tr>
<td>TD 14-17 mo. in words*</td>
<td>53.6 (36-77)</td>
</tr>
<tr>
<td>WS 18 mo. babble</td>
<td>71.9 (16-94; 23.8)</td>
</tr>
<tr>
<td>TD 24 mo.</td>
<td></td>
</tr>
<tr>
<td>WS 24 mo. babble</td>
<td>63.3 (11-88; 25.1)</td>
</tr>
<tr>
<td>WS 24 mo. words</td>
<td>54.9 (33-67; 13.1)</td>
</tr>
</tbody>
</table>

*Vihman, Velleman & Lleo (unpublished): words at 14-17 months

NOTE: Higher # is less mature
Median Percent Monosyllables by Age

TD mean
14-17 mo. words
# Phonotactic Complexity: 18 months

<table>
<thead>
<tr>
<th></th>
<th>Mean % Closed Syllables (range)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>TD 14-17 mo. in words</strong>*</td>
<td>13.6 (0-42)</td>
</tr>
<tr>
<td><strong>WS 18 mo. babble</strong></td>
<td>4.9 (0-15; 5.3)</td>
</tr>
<tr>
<td><strong>TD 24 mo.</strong></td>
<td></td>
</tr>
<tr>
<td><strong>WS 24 mo. babble</strong></td>
<td>7.9 (0-23; 8.5)</td>
</tr>
<tr>
<td><strong>WS 24 mo. words</strong></td>
<td>15.6 (0-50; 17.5)</td>
</tr>
</tbody>
</table>

*Vihman, Velleman & Lleo (unpublished)*
Median Percent Closed Syllables by Age

TD mean 14-17 mo. words
## Babble-Word Comparisons

<table>
<thead>
<tr>
<th>Generalizations from TD Babble vs. Words</th>
<th>WS babble: 18 mo.</th>
<th>WS babble: 24 mo.</th>
<th>WS Words: 24 mo.</th>
</tr>
</thead>
<tbody>
<tr>
<td>Babble more multisyllabic</td>
<td>1.4 syll/voc</td>
<td>1.4 syll/voc</td>
<td>1.4 syll/voc</td>
</tr>
<tr>
<td>Babble fewer consonants (initial position)</td>
<td>20 tokens/85 vocs (0.24)</td>
<td>69 tokens/130 vocs (0.53)</td>
<td>34 tokens/46 vocs. (0.74)</td>
</tr>
<tr>
<td>Babble more varied consonants (initial position)</td>
<td>4 types</td>
<td>7 types</td>
<td>5 types</td>
</tr>
<tr>
<td>Babble more varied vowels</td>
<td>9.1 types</td>
<td>9.4 types</td>
<td>7.6 types</td>
</tr>
<tr>
<td>Initial stops &gt; nasals in both</td>
<td>40% vs. 31%</td>
<td>33% vs. 27%</td>
<td>41% vs. 27%</td>
</tr>
<tr>
<td>Babble more alveolar/dentals</td>
<td>4.1%</td>
<td>34.9%</td>
<td>20.3%</td>
</tr>
<tr>
<td>Words more labials</td>
<td>8.1%</td>
<td>26.1%</td>
<td>43.6%</td>
</tr>
</tbody>
</table>
### Relationships at 24 Months

<table>
<thead>
<tr>
<th></th>
<th># Words</th>
<th># Diff. #C in Babble</th>
<th># Diff. #C in Words</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mullen ELC</td>
<td>rho = .786 p&lt; .05</td>
<td>rho = .805 p&lt; .05</td>
<td>rho = .873 p&lt; .05</td>
</tr>
<tr>
<td># Words</td>
<td></td>
<td>rho = .805 p&lt; .05</td>
<td>rho = .873 p&lt; .05</td>
</tr>
<tr>
<td># Diff. #C in Babble</td>
<td></td>
<td></td>
<td>rho = .973 p&lt; .01</td>
</tr>
</tbody>
</table>
# Different Initial Consonants in Words by Mullen ELC: 24 months

Note: Child X had no words

\[ \rho = 0.89; \ p < .01 \]
Conclusions: WS vs. TD

- Toddlers with WS somewhat lower on several phonetic and phonotactic measures, though ranges overlap:
  - variety of C and V repertoires
  - MBL
  - % syllables including both a C & a V
  - # syllables/word-like vocalization
  - % longer words
  - % closed syllables
- Tendency for smaller C repertoires
- Similar babble vs. word patterns with exception of vocalization lengths in syllables
Conclusions: Babble vs. Words

- Significantly more babble than words even at 24 months

*Babble more complex/mature:*
- Tendency for more varied Cs and Vs in babble – like TD’s

*Words more complex/mature:*
- Tendency for more C tokens in words – like TD’s
- Significantly higher MBL in words than babble at 24 mos.
- Significantly higher % of words with both a C and a V at 24 mos.
Conclusions: Progress from 18 – 24 months

- Significant increases in volubility for both babble & words
- Tendency for group increases on all other measures:
  - Variety of phonetic repertoire
  - Complexity of phonotactic repertoires
  - Not statistically significant
- Correlations among the following with each other:
  - # words @ 24 months
  - # different initial C’s in words and babble @ 24 months
  - Mullen ELC @ 24 months
Causation/directionality cannot be determined
Conclusions: Relationships 18 – 24 months

• Only a few correlations between 18 mo. and 24 mo. measures (babble):
  • # syllables/word-like vocalization: stable (rho = 1.0)
  • # of different vowels (rho = 0.83; p < 0.02)
  • Mean babble level (rho = 0.89; p < 0.01)
• None of babble measures at 18 months are significantly correlated with # of words at 24 months
Implications

• Although WS group ranges overlap with TD ranges on many measures, many children with WS are below age expected ranges on many variables – including one child with no words at 24 months

• C variety is correlated with developmental level – directionality unknown

• Assessment and monitoring of speech skill development are important

• Assess and treat all aspects of child’s phonology, whether purpose is research or clinical:
  • phonotactic (word and syllable shape) complexity
  • phonetic variety and typicality
Future Directions

• Add at least 12 more children for N = 20 (or more)
• Look at reduplication and harmony
• Follow to later ages (48 months)
• Explore individual differences: individual phonetic and phonotactic profiles over time
• Explore relationships between prelinguistic and early linguistic measures with later lexical, grammatical, and literacy measures
References


References

- Pitts, Adelson & Mervis, 2014
Thank you!