QOL-CI: Development & Clinical Utility of a Pediatric Quality of Life Instrument

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3/8/18
Conflict of Interest

• AG Bell Board of Directors
• MED-EL Pediatric Advisory Board
• Research agreement - Advanced Bionics
• Research funded by:
  – NIDCD R01 DC04797
  – NIDCD R03 DC014760
  – NIDCD R21 DC016265
Learning Objectives

• Discuss the steps needed to develop health-related quality of life instruments
• List the most common concerns reported by children with CIs and their parents
• Identify ways of utilizing health-related quality of life measures in a clinical setting
Background

• Severe to profound hearing loss is associated with deficits in HRQoL, including concomitant effects on:
  – Oral language, social and emotional functioning, visual attention, and academic performance
• However, the majority of studies on children with cochlear implants (CIs) focus primarily on language and communication
  – These studies fail to capture the effects of CIs on daily functioning
• To date, there are no CI-specific HRQoL measures for young children and their parents
• This study aims to develop the first HRQoL instruments for children with CIs and their parents
Health-Related Quality of Life (HRQoL)

• HRQoL measures capture how an individual feels, functions, and survives with a chronic medical condition or disability
• HRQoL encompasses four domains:
  – 1) Disease severity and physical symptoms
  – 2) Functional status
  – 3) Emotional/cognitive functioning
  – 4) Social functioning
• FDA and NIH have pushed for the development HRQoL measures for children with chronic illnesses and disabilities using the PRO Guidance (2009)
Value of CI-Specific Measure

- Condition-specific measures are more sensitive and responsive to change than generic measures.
- They contain items that are more relevant and deemed important by patients/parents (Quittner, Cejas, Blackwell, 2013).
- A CI-specific measure can be used to:
  - Facilitate research on psychosocial outcomes
  - Compare unilateral vs. bilateral implants
  - Evaluate effects of new processors
  - Examine the efficacy of audiological, language, and behavioral interventions
Study Aims

• Develop a CI-specific quality of life (QoL) instrument using the FDA Patient Reported Outcomes Guidelines
  – Early Childhood (Birth-5)
  – School Age (6-12)
  – Adolescent- Early Adult (13-22)
i. **Identify Concepts**
   - Identify claims
   - Identify relationships among all endpoints
   - Identify concepts relevant to patients
   - Determine intended population
   - Develop expected relationships among items & concepts/domains

v. **Modify Instrument**
   - Change concepts measured, populations studied, research application, response options, recall period, or method of administration
   - Translate & culturally adapt to other languages

iv. **Collect, Analyze, & Interpret Data**
   - Prepare protocol & statistical analysis plan
   - Identify responder definition
   - Evaluate cumulative distribution curve
   - Present interpretation of treatment benefit

ii. **Create Instrument**
   - Generate items
   - Choose administration method, recall period & response scales
   - Draft instructions
   - Format instrument
   - Draft procedures for scoring & administration
   - Pilot test draft instrument
   - Refine instrument & procedures

iii. **Assess Measurement Properties**
   - Assess score reliability, validity, & ability to detect change
   - Evaluate administrative & respondent burden
   - Add, delete, or revise items
   - Confirm conceptual framework
   - Finalize instrument formats, scoring, procedures & training materials
## Development of QoL Instruments

- Open-ended interviews with stakeholders, children and parents
- Interviews are transcribed
- Content analysis: Identification of relevant items and generation of saturation matrices
- Item generation and creation of rating scales
- Creation of draft measures
- Cognitive testing
# Demographics: School-Age Version

## Open-Ended Interviews 6-12

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<td><strong>Age</strong></td>
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<tr>
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<tr>
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<tr>
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</tr>
<tr>
<td></td>
<td>Progressive</td>
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<tr>
<td></td>
<td>Bimodal</td>
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<tr>
<td></td>
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## Cognitive Interviews 6-12

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<td></td>
<td>8-9</td>
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<td></td>
<td>10-11</td>
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<td></td>
<td>12</td>
<td>1 (5%)</td>
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<td><strong>Age at Implantation</strong></td>
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<td><strong>Race/Ethnicity</strong></td>
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<td></td>
<td>White</td>
<td>11 (55%)</td>
</tr>
<tr>
<td></td>
<td>Biracial</td>
<td>1 (5%)</td>
</tr>
<tr>
<td></td>
<td>Asian</td>
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<td></td>
<td>Biracial</td>
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<td></td>
<td>Other</td>
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<tr>
<td><strong>Type of Hearing Loss</strong></td>
<td>Sudden</td>
<td>2 (20%)</td>
</tr>
<tr>
<td></td>
<td>Progressive*</td>
<td>6 (30%)</td>
</tr>
<tr>
<td></td>
<td>Congenital*</td>
<td>11 (55%)</td>
</tr>
<tr>
<td><strong>Device Type</strong></td>
<td>Unilateral</td>
<td>3 (15%)</td>
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<tr>
<td></td>
<td>Bimodal</td>
<td>4 (20%)</td>
</tr>
<tr>
<td></td>
<td>Bilateral</td>
<td>13 (65%)</td>
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</table>
Finalized Child Conceptual Framework

CI

- Cost
  - Financial burden
  - Fear over losing device

- Physical Functioning
  - Magnet falls off
  - Magnet discomfort
  - Injuries

- Oral Communication
  - Poor comprehension
  - Trouble with exp. language
  - Bilingualism

- Device Management/Usage
  - Additional accessories
  - Needing a break
  - Adapting to second device

- Behavior
  - Attention/Distractibility
  - Fatigue

- Emotional Functioning
  - Frustrated
  - Happy
  - Anxious

- Social Functioning
  - Trouble making friends
  - Social withdrawal/isolation
  - Teasing/bullying

- Hearing in Different Environments
  - Noisy Environments
  - TV, movies, videogames
  - Music

- Benefits of CI(s)
  - Comparing CI vs HA
  - Aids sleeping
  - Safety

- Family Interactions
  - Sibling relationships
  - Family interaction challenges

- Parents
  - Parent facilitating

- Child Acceptance
  - Acceptance of device
  - Future plans

- Limitations of CIs
  - Can't participate in certain activities

- Academic Functioning
  - Academic performance
  - School support
  - Teacher facilitating
Saturation Matrix for Social Functioning

|                              | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 | 11 | 12 | 13 | 14 | 15 | 16 | 17 | 18 | 19 | 20 | 21 | Total |
|------------------------------|---|---|---|---|---|---|---|---|---|----|----|----|----|----|----|----|----|----|----|----|-----|
| Self Advocacy                | 1 | 1 | 1 | 1 | 1 | 1 | 1 | 1 | 0 | 1   | 1   | 1   | 1   | 1   | 1   | 1   | 1   | 1   | 1   | 1   | 1   | 20   |
| Curiosity – Staring and      | 1 | 0 | 1 | 0 | 1 | 1 | 1 | 1 | 1 | 1   | 1   | 1   | 1   | 1   | 1   | 1   | 1   | 1   | 1   | 1   | 1   | 19   |
| Reactions                    |   |   |   |   |   |   |   |   |   |     |     |     |     |     |     |     |     |     |     |     |     |     |
| Positive Friendships         | 1 | 1 | 1 | 1 | 1 | 0 | 1 | 1 | 0 | 1   | 1   | 1   | 1   | 1   | 1   | 1   | 1   | 1   | 1   | 1   | 0   | 1   | 18   |
| Sibling Relationships        | 0 | 1 | 0 | 1 | 1 | 1 | 1 | 0 | 0 | 1   | 1   | 1   | 0   | 1   | 0   | 1   | 1   | 1   | 1   | 0   | 1   | 15   |
| Feeling Different            | 0 | 0 | 1 | 0 | 1 | 1 | 0 | 0 | 1 | 0   | 1   | 0   | 1   | 0   | 0   | 1   | 1   | 1   | 1   | 1   | 1   | 13   |
| Swimming                     | 0 | 1 | 0 | 0 | 1 | 1 | 0 | 1 | 1 | 1   | 1   | 1   | 0   | 0   | 1   | 1   | 0   | 0   | 1   | 0   | 1   | 1   | 12   |
| Social Withdrawal/Isolation  | 0 | 1 | 0 | 0 | 0 | 1 | 1 | 0 | 0 | 1   | 0   | 0   | 1   | 0   | 0   | 1   | 0   | 1   | 1   | 1   | 0   | 0   | 8    |
| Teasing/Bullying             | 0 | 0 | 0 | 1 | 0 | 0 | 1 | 0 | 0 | 0   | 0   | 0   | 1   | 1   | 0 | 0 | 1 | 0 | 1 | 1 | 1 | 0 | 1 | 8    |
| Trouble Making Friends       | 0 | 0 | 0 | 0 | 0 | 1 | 0 | 0 | 1 | 0   | 0   | 0   | 1   | 0   | 0   | 0   | 0   | 1 | 0 | 1 | 0 | 1 | 5    |
## Common Themes

<table>
<thead>
<tr>
<th>Child</th>
<th>Parent</th>
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<tbody>
<tr>
<td>2. Social Functioning</td>
<td>2. Social Functioning</td>
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<tr>
<td>3. Emotional Functioning</td>
<td>3. Academic Functioning</td>
</tr>
<tr>
<td>4. Oral Communication</td>
<td>4. Fatigue</td>
</tr>
<tr>
<td>5. Academic Functioning</td>
<td>5. Behavior Problems</td>
</tr>
</tbody>
</table>
Child Sample Quotes

- **Child Acceptance:** “Um, it doesn’t make me feel bad, but like, it’s pretty much like they want to know about it because they’ve never seen it before. So I’m pretty much happy to explain to them.”

- **Social Functioning:** “About—since—ever since I came to the school, they like to be mean to me. So only—I only just make a little bit of friends at other class. That’s it.”

- **Emotional Functioning:** “It hurt my feelings and I don’t like people to know because it bother me.”
Parent Sample Quotes

• **Emotional Functioning:** “If he feels something isn’t going his way or if someone’s mean, or if he’s just having a fit. I notice his anger side gets a little bit more aggressive.”

• **Social Functioning:** “…she might unconsciously withdraw. We definitely encourage her more. She is the kid who would want to stay home all weekend and not go anywhere.”

• **Academic Functioning:** “He doesn’t get math. We have him do his times tables every day at home, we have him write them, recite them- he doesn’t get it.”
Severity/Frequency Ratings: School-Age Version

Parents (N=21)

Stakeholders (N=36)
QoL- CI School Age Version

- **Self-report**
  - 33 Items across 8 Domains
    1. Noisy Environments
    2. Academic Functioning
    3. Child Acceptance
    4. Oral Communication
    5. Social Functioning
    6. Fatigue
    7. Emotional Functioning
    8. Device Management

- **Parent-Proxy**
  - 42 Items Across 9 Domains
    1. Noisy Environments
    2. Academic Functioning
    3. Child Acceptance
    4. Oral Communication
    5. Social Functioning
    6. Fatigue
    7. Emotional Functioning
    8. Device Management
    9. Behavior Problems
Sample Question

How hard is it for you to hear other people in the lunchroom?
Parenting Stress Module

- Standardized scores on the Parenting Stress Module ranged from 14.81 (low stress) to 79.17 (low stress), with an average score of 42.18.
- Parents’ highest rated stressors included: 1) CI/s breaking; 2) CI/s getting lost; 3) child hurt during sports (See table 2).
- The final parenting stress module consisted of 9 items rated on a scale from 1 (Not at all Concerned) to 4 (Very Concerned), with higher scores indicating more parenting stress.

<table>
<thead>
<tr>
<th>Stress Ranking (Highest to Lowest)</th>
<th>Item</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>How concerned are you about your child's CI breaking?</td>
</tr>
<tr>
<td>2</td>
<td>How concerned are you about your child's CI getting lost?</td>
</tr>
<tr>
<td>3</td>
<td>How concerned are you about your child being hurt during sports?</td>
</tr>
<tr>
<td>4</td>
<td>How stressful is it to make decisions about your child's educational placement?</td>
</tr>
<tr>
<td>5</td>
<td>How concerned are you about your child spending time away from home (i.e., sleepovers, stay with grandparents)?</td>
</tr>
<tr>
<td>6</td>
<td>How stressful is it to manage the costs of your child's CI (i.e., AV therapy, cables, upgrades)?</td>
</tr>
<tr>
<td>7</td>
<td>How stressful is it to get appropriate accommodations for your child (i.e., IEP)?</td>
</tr>
<tr>
<td>8</td>
<td>How concerned are you about splitting time between your child with a CI and siblings?</td>
</tr>
<tr>
<td>9</td>
<td>How stressful is it when someone stares or asks about your child's CI?</td>
</tr>
</tbody>
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### Demographics: Birth to 5 Version

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</tr>
<tr>
<td></td>
<td>2-3</td>
<td>3 (21%)</td>
</tr>
<tr>
<td></td>
<td>4-5</td>
<td>10 (71%)</td>
</tr>
<tr>
<td><strong>Age at Implantation</strong></td>
<td>Mean</td>
<td>20 Months</td>
</tr>
<tr>
<td><strong>Race/Ethnicity</strong></td>
<td>African American</td>
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</tr>
<tr>
<td></td>
<td>Congenital</td>
<td>10 (71%)</td>
</tr>
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<td>N/A</td>
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<tr>
<td></td>
<td>Bimodal</td>
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</tr>
<tr>
<td></td>
<td>Bilateral</td>
<td>10 (71%)</td>
</tr>
</tbody>
</table>
Severity/Frequency Ratings: Birth to 5

Parents (N=14)

Stakeholders (N=23)
Common Themes

Parents

1. Parenting Stress
2. Device Management
3. Behavior Problems
4. Communication
5. Academic Functioning
Parent Sample Quotes

• **Parenting Stress:** “Like, if we do to Disney I like to go when it’s like really *empty* ‘cuz I get anxious and I get nervous when it gets really crowded.”

• **Device Management:** “In the beginning, I feel like they were always falling off. I feel like if you were to look at our audiology report, like the first couple of times, he was like they're falling off all the time.”

• **Behavior:** “… if he really gets really upset, he'll just throw them across the room.”
## Most Common Concerns Reported by Parents

<table>
<thead>
<tr>
<th>0-5</th>
<th>6-12</th>
</tr>
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<tbody>
<tr>
<td>1. Parenting Stress</td>
<td>1. Emotional Functioning</td>
</tr>
<tr>
<td>2. Device Management</td>
<td>2. Social Functioning</td>
</tr>
<tr>
<td>4. Communication</td>
<td>4. Fatigue</td>
</tr>
<tr>
<td>5. Academic Functioning</td>
<td>5. Behavior Problems</td>
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</table>
Parenting Stress

**0-5**
1. Decision Making
2. Behavior Management
3. Educational Placement
4. Educating Others
5. Losing CI

**6-12**
1. CI Breaking
2. Losing CI
3. Child Getting Hurt During Sports
4. Educational Placement
5. Child Spending Time Away From Home
Summary

- QoL –CI - Early Childhood & School-age version
  - Next steps: psychometric validation
  - Will be available for free
- Children report social and emotional difficulties related to having a CI that are impacting their QoL
- Despite children doing well, parents report significant amounts of stress
- Discrepancy between clinicians and how patients/parents view their outcomes
Ways to Use in a Clinical Setting

• Clinical Use
  – Annual Screening
  – Use to identify children who need further intervention

• Research
  – Facilitate research on psychosocial outcomes
  – Evaluate effects of new processors/programming strategies
  – Examine the efficacy of audiological, language, and behavioral interventions
Save The Date

July 10 – 13 2019

The world famous Diplomat Beach Resort will be the host site for CI2019 which features Ft Lauderdale and Miami Beach. See you in South Florida in 2019.