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Measuring Communicative Performance with the FAPCI Instrument: Preliminary Results from Normal Hearing and Cochlear Implanted Children

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Abstract

Objective—To develop preliminary “growth curves” of Functioning after Pediatric Cochlear Implantation (FAPCI) scores using a cross-sectional sample of normal hearing children and to compare these curves to trajectories of FAPCI scores in children receiving cochlear implants.

Methods—Quantile regression was used to develop growth curves from the FAPCI scores of a cross-sectional sample of 82 normal hearing children (age range 7 months – 5 years). Trajectories of FAPCI scores from a longitudinal cohort of 75 children with cochlear implants (age range 1-5 years) were compared to these growth curves.

Results—FAPCI scores were positively associated with increasing age in normal hearing children with a rapid increase in scores observed at earlier ages followed by a plateau at age 3 years. FAPCI trajectories for cochlear-implanted children varied with age at implantation and did not reach a plateau until age 5-6 years.

Conclusion—Normal hearing children demonstrated increasing FAPCI scores with age, and these preliminary growth curves allow for the interpretation of a cochlear-implanted child's FAPCI scores in comparison to normal hearing children. Additional research using a larger, longitudinal cohort of normal hearing children will be needed to develop definitive normative FAPCI trajectories.

Keywords

FAPCI; Pediatric; Cochlear Implantation; Communicative Performance

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INTRODUCTION

A child's proficiency in oral language is largely dependent on the child's experience and exposure to language during their first few years of life [1]. Pre-lingual sensorineural hearing loss (SNHL) limits this exposure resulting in a significant lag in the mastery of language in its spoken form compared to normal hearing peers [2]. Cochlear implantation (CI) enables a deaf child access to sound and audition [3], and over half of children identified with early SNHL currently receive a CI [4].

Traditionally clinicians have assessed outcomes after CI by measuring speech perception, discrimination [5], and communicative capacity, utilizing speech and language metrics such as the Early Speech Perception Test [6] and the Reynell Developmental Language Scales [7]. However, such measures may incompletely capture a child's communicative abilities in a normal listening environment, where background noise and non-ideal listening conditions predominate [8]. The World Health Organization's International Classification of Functioning (ICF) distinguishes between communicative *capacity*, an individual's ability to communicate in a standardized environment (e.g. clinic setting), and communicative *performance*, an individual's ability to communicate in real-world environments [9]. Measuring communicative performance after cochlear implantation is complicated further by the young age of CI recipients with many undergoing implantation before one year of age. This has created a demand for a validated assessment tool which fully captures speech development in these pre-verbal children [10].

A number of novel measures have been developed to better capture speech perception and language skills in this pediatric population. These measures rely on either direct measurement of the child's performance (visual-habitual procedures in the presence of speech competitors [1]), or more commonly by proxy assessment. Two of the most frequently used parent-proxy instruments include the Infant-Toddler Meaningful Auditory Integration Scale (ITMAIS) and the Little Ears Auditory Questionnaire. The IT-MAIS consists of 10 items probing a child's response to sounds in the everyday environment, and parents respond on a five level scale [11]. The well-validated Little Ears Auditory Questionnaire also surveys the auditory behaviors of children up to 24 months with 35 binary items that are based on parental report [10]. In contrast to these instruments focused on the early auditory behaviors of infants and toddlers <24 months, the Functioning after Pediatric Cochlear Implantation instrument (FAPCI) assesses the communicative abilities of children > 24 months and was designed to supplement these other instruments [12].

The FAPCI instrument is a psychometrically-validated scale that was designed from the ground up to assess the communicative performance of 2-5 year-old CI children based on the conceptual framework of the World Health Organization's International Classification of Functioning [9]. The instrument consists of 23 items (Table I) with each item having a five-level response scale (range of possible total scores: 23 to 115). The measure queries a young child's everyday expressive and receptive communicative behaviors as reported by the parent or primary caregiver. The FAPCI instrument was developed through a rigorous three phase process [12]. Phase I consisted of qualitative instrument development consisting of domain/item identification and instrument pretesting. This was achieved through discussion and interviewing experts working with CI children, parents of CI recipients, and a review of the current literature. Phase II and III consisted of quantitative instrument validation and refinement using psychometric analysis where excellent reliability was demonstrated (Cronbach's $\alpha \geq 0.86$) [12]. The FAPCI instrument has been translated into German [13] and Korean [14] and is currently being used in ongoing NIH and industry-sponsored studies of CI outcomes in young pediatric populations.

In the current study we developed a framework for the interpretation of FAPCI scores by developing preliminary growth curves of FAPCI scores in normal hearing (NH) children. We then compared trajectories of FAPCI scores among a cohort of CI children to these normal hearing growth curves.

METHODS

Study Cohort

Normal hearing (NH) cohort—The 23-item FAPCI [12] instrument and an introductory letter were distributed in two local childcare centers in Baltimore affiliated with Johns Hopkins University and the University of Maryland. Parents were asked to complete the survey and to answer optional questions regarding their level of education and annual income. Completed forms were returned via mail.

CI cohort—The FAPCI instrument along with an introductory letter and a demographic intake sheet were mailed to 102 households with a CI recipient between the ages of 2-5 years. All recipients were being followed by the Johns Hopkins Listening Center or enrolled at The River School. Seventy-five families (74%) returned completed surveys at baseline. Respondents were requested to repeat the survey approximately one year later. Fifty-four families (72%) completed this follow up survey. This study was approved by the Johns Hopkins Hospital Institutional Review Board.

Statistical Analysis

FAPCI survey data were entered into an electronic database and verified by double-data entry. FAPCI scores range from 23 to 115 with higher scores denoting better communicative performance. Missing data on items accounted for <1% of returned surveys. FAPCI scores were calculated by summing the scores of all items and assigning a score of 0 to items with no response (no surveys contained more than 1 missing item). Exploratory analyses were conducted using graphical displays and frequency distributions to identify potential outliers for further data validation. Quantile regression models were used to estimate the 10th, 25th, median, 75th, and 90th percentile of growth curve for FAPCI scores over age based on data from the normal hearing sample. Nonparametric locally weighted smoothed scatter plot (lowess) approach was used to estimate the growth trajectories in FAPCI score over age in CI children stratified by implantation age. These estimated trajectories were plotted against the growth curve percentile estimates from the normal hearing sample to examine the degree by which CI trajectories deviated from the normal hearing growth curves. Comparison of proportions between groups was performed with a Fisher exact test. Analyses were carried out using SAS 9.1.3 (SAS Institute Inc, Cary, NC).

RESULTS

Subjects

FAPCI score results were collected from 82 parents of NH children and from a longitudinal cohort of 75 CI children (Table 2). Baseline FAPCI scores from the parents of CI children were gathered at a mean of 29 months (standard deviation: 13.3) after implantation and follow-up scores were assessed 10-15 months later (mean: 13 months) with the exception of one child whose follow-up data were gathered at 24 months.

Normative curves

Cross-sectional FAPCI scores from the 82 NH children demonstrated that FAPCI scores consistently increase with older age (Figure 1), and these data were used to derive

preliminary growth curves through quantile regression (Figure 2). For NH children, communicative performance, as measured by the FAPCI, developed rapidly until age three years before reaching a stable plateau. Percentile curves were tightly clustered demonstrating near homogeneous growth in communicative performance among all NH children. Only children at the 10th percentile were delayed by approximately one year in reaching plateau FAPCI scores (Figure 2).

FAPCI trajectories in CI children

Unlike the FAPCI scores in NH children, substantial heterogeneity in FAPCI scores was present in the CI children. Some CI children rapidly developed communicative performance on par with NH children whereas others lagged far behind (Figure 3). FAPCI trajectories from 6 children were substantially lower than those from other children (Figure 3, trajectories clustered below a FAPCI score of 50). On further review of these CI recipients, it was found that four had intra or post natal infections with resultant co-morbidities and developmental delays. Of the remaining two, one had been adopted from Russia and whose full medical history was unknown, and the other had experienced technological issues with the CI device.

On average, CI children implanted at age <2 years (n = 42) began to reach plateau FAPCI scores at approximately 48-60 months of age with scores corresponding to below the 10th percentile in NH children (Figure 4). For those children implanted at age ≥2 years (n=33), plateau FAPCI scores were not reached until 60-72 months of age. CI children implanted at age <2 years, therefore, appear to be delayed by approximately 1-2 years and those children implanted at age ≥2 years are delayed between 3-4 years in the development of communicative performance compared to NH children. There were no differences in parental education (p = .9) or income (p=.8) between children undergoing earlier versus later implantation.

DISCUSSION

We have developed preliminary growth curves of FAPCI scores in normal hearing children to serve as a framework for interpreting FAPCI scores from CI children. We reasoned that graphical representations of communicative performance in NH children would be a clinically useful approach toward understanding and monitoring a CI child's growth in communication skills.

The use of these growth curves may provide clinicians and parents greater insight into whether a CI child is adequately developing communication skills. For example, trajectories in FAPCI scores from two hypothetical children (Child A [dashed line] and Child B [solid line]) are presented in Figure 5. For both children, FAPCI scores are similar before implantation and both children undergo CI at 18 months. However, Child A demonstrates rapid acquisition of communicative performance skills on par with a normal hearing child, while Child B initially demonstrates improvement but then quickly plateaus. Such information when used in conjunction with other conventional speech and language metrics likely offers substantial insight into a child's communicative development and whether further interventions (e.g. aural rehabilitative, device troubleshooting, etc) need to be considered.

Our results demonstrate that NH children typically reach maximal FAPCI scores by 3 years of age. Communicative performance in CI children by comparison remains delayed. FAPCI trajectories in CI children were found to be less steep and lag behind the trajectories of NH children by a range of 1-4 years depending on the age of implantation. These results are consistent with prior studies that have demonstrated less language delays in children

implanted earlier [2,15,16]. This observation has been attributed to a finite period of cortical plasticity in young children where the acquisition of verbal language is optimal [12,17].

There are limitations to our study. First, our normal hearing growth curves were drawn from a moderately-sized, cross-sectional sample of normal hearing children whose families were of high socioeconomic (SES) and educational status. Given the strong association between SES and child language development [18], our normal hearing growth curves likely reflect the performance of a high-functioning cohort of children. As such, future assessment of longitudinal FAPCI scores in a larger and more representative population of normal hearing children will be needed to derive true normative trajectories. However, we note that the FAPCI instrument reflects relatively rudimentary communicative skills (e.g. ability to understand speech in the car) that may not be as closely correlated with family SES as much as higher order language skills focusing on vocabulary and syntax. Children receiving cochlear implants are also more likely to live in areas of higher median incomes such that growth curves derived from normal hearing children of higher SES likely remains useful [19,20].

The generalizability of the mean trajectories of FAPCI scores in CI children in our study (Figure 4) is also limited. Our results were based on a relatively small cohort of subjects, and there was heterogeneity in the time from cochlear implantation to testing with the FAPCI. However, the main purpose of our study was to demonstrate the feasibility of FAPCI growth curves in NH children and not to precisely estimate FAPCI trajectories in CI children. We are continuing to gather longitudinal FAPCI data along with other speech and language data in NIH and industry-sponsored studies to further understand how communicative performance as measured by the FAPCI develops over time in children with cochlear implants. Finally, we observed that FAPCI scores typically reached a ceiling by approximately 36 months in NH children, possibly limiting the instrument's utility in assessing the communicative performance of children after 3-4 years.

Verbal communicative performance has been hypothesized to serve as the foundation for a cochlear-implanted child's future development of academic achievement, social versatility, and independence and quality of life in adulthood [21]. The FAPCI instrument is a psychometrically-validated metric of communicative performance that demonstrates robust validity and allows for assessment of communicative performance relative to NH peers. Continued use of the FAPCI instrument in conjunction with other speech and language measures may allow clinicians, educators, and therapists to better focus clinical, research, and rehabilitative efforts on a child's true communicative ability rather than isolated measures of speech and language obtained in clinical settings.

Copies of the FAPCI instrument and scoring manual are available for public use by contacting fapci@jhmi.edu or at www.thelisteningcenter.com.

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Figure 1. Cross-sectional FAPCI scores from 82 NH children. Solid line denotes the smoothed (lowess) average of FAPCI scores across age. FAPCI scores range from a minimum of 23 to a maximum of 115.

Figure 2.

FAPCI growth curves derived from 82 NH children. FAPCI scores can range from a minimum of 23 to a maximum of 115. The solid line is the median, the long-dashed lines are the 25th and 75th percentiles, and the short-dashed lines are the 10th and 90th percentiles.

Figure 3.
FAPCI score trajectories in CI children implanted at age <2 years (dashed) and age ≥2 years (solid) compared to growth curves from NH children.

Figure 4. Mean FAPCI trajectories for CI children implanted at age <2 years (dashed) and age ≥2 years (solid) compared to NH children. FAPCI scores range from a minimum of 23 to a maximum of 115.

Figure 5.
FAPCI trajectories for 2 hypothetical CI children (Child A [dashed] and Child B [solid line]). Both children are implanted at 18 months (arrow), and FAPCI scores are measured annually after implantation.

Table 1
FAPCI Instrument

Detailed instructions, specific examples, and tips for responding to all items are provided with questions in the actual FAPCI instrument. Response format: F = Frequency-based questions (response levels of “never”, “rarely”, “sometimes”, “frequently”, and “always”); Q = Quantity-based questions (response levels with either specific quantities or “almost none (0-4%)”, “few (5-24%)”, “some (25-49%)”, “most (50-95%)”, or “almost all (96-100%)”); E = Example-based questions (response levels contain a description or an example of a behavior, and levels correspond to an ordinal scale of functioning adjudicated by the authors).

Item	Response Format
How often does your child respond to phrases that s/he overhears from a nearby conversation?	F
Given an unlimited set of possible choices, how many age-appropriate items would your child be able to point to when they are presented in spoken language without visual cues?	Q
How many age-appropriate 2-step spoken commands presented without visual cues does your child understand?	Q
When riding in a car, my child is able to understand...	E
When listening from a different room of the house, my child is able to understand...	E
When in a noisy environment, my child is able to understand...	E
When using the telephone with a familiar caller, my child is able to understand...	E
How often does your child appropriately answer simple questions presented in spoken language without visual cues?	F
How many age-appropriate items would your child be able to identify with spoken language when they are pointed to?	Q
How much of your child's speech would an adult who is not familiar with your child understand?	Q
How does your child typically respond when greeted by a familiar person?	E
How many people's names does your child use in spoken language?	Q
Which statement best describes your child's singing?	E
What is the main way that your child communicates his/her wants when not coached by an adult?	E
How many of the following types of words/phrases does your child use in spoken language: what, where, why, inversion questions, which?	Q
How many of the following types of words/phrases does your child use in spoken language: words to describe size or color, numbers to describe how many, words to describe quantity, plural endings, possessive ending?	Q
How often does your child ask simple questions using spoken language?	F
How often does your child talk about his/her experiences during the day or about a past event using simple spoken sentences?	F
How often does your child use the past tense in spoken language?	F
How often does your child use the negative in a 2-3 word spoken phrase?	F
How often does your child correctly use pronouns in spoken language?	F
How often does your child correctly use prepositions in spoken language?	F
How often does your child initiate a spoken conversation with another child?	F

Table 2

Characteristics of Study Cohort

Characteristics		Normal Hearing N =82	Cochlear Implant N = 75
Age of child ¹	0-12	3 (3.7)	0 (0)
	13-24	20 (24.4)	1 (13)
	25-36	21 (25.6)	10 (13.3)
	37-48	17 (20.7)	15 (20.0)
	48-60	13 (15.9)	22 (29.3)
	61-72	8 (9.8)	27 (36.0)
Gender			
	Female	43 (52.4)	41 (54.7)
Age at Implantation	<24 months		42 (56.0)
	≥24 months		33 (44.0)
Parental Income ²	<\$50,000	11 (13.4)	13 (17.3)
	\$50,000-\$100,000	18 (22.0)	29 (38.7)
	>\$100,000	53 (64.6)	32 (42.7)
Parental Education	High School	3 (3.7)	8 (10.7)
	Some College	3 (3.7)	14 (18.7)
	Completed College	12 (14.6)	25 (33.3)
	Graduate School	64 (78.0)	28 (37.3)

¹ In months² 1 parent did not respond in the CI cohort