Brazilian adaptation of the Functioning after Pediatric Cochlear Implantation (FAPCI): comparison between normal hearing and cochlear implanted children

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Received 10 March 2014; accepted 11 June 2014
Available online 30 October 2014

KEYWORDS
FAPCI;
Cochlear implantation;
Verbal communication;
Normal hearing

Abstract
Objective: Enabling development of the ability to communicate effectively is the principal objective of cochlear implantation (CI) in children. However, objective and effective metrics of communication for cochlear-implanted Brazilian children are lacking. The Functioning after Pediatric Cochlear Implantation (FAPCI), a parent/caregiver reporting instrument developed in the United States, is the first communicative performance scale for evaluation of real-world verbal communicative performance of 2-5-year-old children with cochlear implants. The primary aim was to cross-culturally adapt and validate the Brazilian-Portuguese version of the FAPCI. The secondary aim was to conduct a trial of the adapted Brazilian-Portuguese FAPCI (FAPCI-BP) in normal hearing (NH) and CI children.

Methods: The American-English FAPCI was translated by a rigorous forward-backward process. The FAPCI-BP was then applied to the parents of children with NH (n = 131) and CI (n = 13), 2-9 years of age. Test-retest reliability was verified.

Results: The FAPCI-BP was confirmed to have excellent internal consistency (Cronbach’s alpha > 0.90). The CI group had lower FAPCI scores (58.38 ± 22.6) than the NH group (100.38 ± 15.2; p < 0.001, Wilcoxon test).


* Article refers to master’s project of otorhinolaryngologist medical doctor Trissia Vassoler, under orientation of professor doctor Mara Lucía Cordeiro.

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http://dx.doi.org/10.1016/j.jped.2014.06.008
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Introduction

Cochlear implantation (CI) is a treatment for severe-to-profound bilateral sensorineural hearing loss, particularly for children with congenital and perinatal etiologies. It is recommended when traditional hearing aids (sound amplification appliances) cannot enable sound discrimination. Social communication is an essential human capacity and oral language is the most used form of complex communication. ample evidence has shown that children who receive CI at a very young age are able to develop better performance in speech comprehension and production and achieve better academic and social behavior than children treated later. There is also growing evidence that children with severe-to-profound bilateral hearing loss who receive CI bilaterally can perform almost as well as children with normal-hearing (NH). Early auditory deprivation, even if partial, has a deleterious effect on language development and on the development of central auditory processing skills in young children.

Enabling hearing is the first goal of CI. Once adequate hearing has been established with CI, the development of oral language is expected to follow. Several factors can influence the outcome of CI, such as duration of deafness, age at implantation, the speech rehabilitation approach applied, and how these factors interact to influence neural plasticity. Many variables influence this process, and it is extremely important that physicians and speech therapists track the performance and progress of CI patients in the area of language development.

Several studies have investigated the effects of implantation age and the outcome of language development skills. Not surprisingly, earlier implantation has been demonstrated to lead to better language outcomes. Other factors that play a role in language development after CI include family involvement in rehabilitation therapy and the educational level of the family. Geers et al. argued that children with congenital deafness should receive CI no later than 2 years of age, while electrophysiological studies and the brain plasticity literature define the critical period for CI as extending to about 3.5 years of age. In successive studies of the long-term effects auditory deprivation on language development, Davidson et al. found that a long period of deprivation before CI had only a minor negative effect on vocabulary.
acquisition, but hindered syntax and prosody severely. The
language development of children who received stimulation
via sound amplification equipment and sign language was
better following CI than those who did not receive such
interventions, but outcomes are improved more by early CI
than by these interventions.11

Electrophysiological research by Gilley et al.6 has shown
that children with congenital deafness who receive CI within
the critical maximally plastic period for central auditory
pathway development develop cortical electrical potentials
with latencies that are close to latencies observed in hearing
children within six months of stimulation.12 In contrast,
children with congenital deafness who received CI after 7 years
of age show cortical electrical potentials with latencies that
are consistently longer than those of age-matched children
with NH; outcomes in children who received CI between 3.5
years and 7 years of age were highly variable. These findings
are consistent with prior neurophysiological and functional
imaging studies in indicating a critical period for neuroplas-
ticity of the auditory system before the age of 3.5 years.12,13

Families and physicians need to be able to deter-
mine whether or not the objectives of CI have been
met. Traditionally, clinicians have used speech percep-
tion and discrimination tests to evaluate communicative
capacity in children following CI.14 However, these measure-
ments may not reflect the child’s ability to communicate
in a real-world environment with background noise and
non-ideal listening conditions.15 The World Health Orga-
nization’s International Classification of Functioning (ICF)
distinguishes between communicative capacity, the ability
to communicate in a standardized environment, and
communicative performance, the ability to communicate in
real-world environments.16 Measuring communicative per-
formance after CI is very difficult, particularly in young
children, and this challenge has created a demand for
validated assessments tools. The widely available question-
naires used for assessment of communicative performance
after CI were designed to measure communicative capacity,
that is, the child’s ability to understand lexicon, gram-
mar, and syntax.17 Examples of this type of instrument
include the Reynell Developmental Language Scales (RDLSs),
the MacArthur Communicative Development Inventories
(MCDIs), and the Meaningful Use of Speech Scale (MUSS).
The RDLSs are used to assess expressive and receptive lan-
guage, the MCDIs are used to evaluate lexical development,
and the MUSS is used to assess oral language use in children
with hearing impairments. Communicative capacity can be
measured in a clinical setting, but such testing is not suf-
icient to establish whether patients are able to use their
communication skills well enough to function in a normal
social environment in their daily lives.15

Currently, there are no instruments with reliable para-
eters that can be used to evaluate the communicative
performance of pediatric cochlear implant users in Brazilian
Portuguese. Children who have received CI in Brazil are still
evaluated primarily in terms of the results of auditory and
language assessments applied in an isolated environment.17
Speech perception and language skills are measured by
direct behavioral observation or, more commonly, a proxy
assessment, such as the MUSS or RDLSs, mentioned above,
or the Infant-Toddler Meaningful Auditory Integration Scale
for children younger than 24 months.

To improve auditory (re)habilitation such that commu-
nicative performance of children with CI is maximized,
there needs to be a sufficient understanding of the instru-
mements’ functional limitations.18 The Functioning Inventory
after Pediatric Cochlear Implantation (FAPCI) instrument
was developed in the USA, in American English, to enable
more objective evaluation of the auditory communicative
performance of 2-5-year-old children with CI. It was
designed to probe the child’s use of communication skills in
his or her interactions with linguistically fluent individuals.18
The Speech, Spatial, and Qualities of Hearing Scale
(SSQ)19 is a widely used structured scale that evaluates hear-
ing ability in everyday situations. Originally designed for
adults, it has been adapted for use with children, parents,
and teachers.20 It is composed of three sections, A, B, and C.
Section A assesses the ability of the individual to understand
oral language in a quiet setting, with background noise, in
reverberant environments, and on the phone. Section B eval-
uates how well an individual perceives his or her position
and movement away from a sound source. Section C asks
the individual to identify sounds and voices with the aim
of determining whether sounds can be understood and seg-
regated with ease. SSQ findings are relevant for receptive
language assessment, but the SSQ does not provide infor-
mation about expressive language or the quality of oral
language communication, as the FAPCI does. Furthermore,
the SSQ was developed for adults and then adapted to a
parent/teacher version for proxy assessment of 5-11-year-
old children, and adapted to a self-report child version
for children over 11 years old. Hence, the SSQ is not suitable
for use with children in the 2-5-year-old age band, whereas
the FAPCI is.

The FAPCI models various situations of everyday life,
and allows communicative performance to be assessed by
professional care providers or family members.18 The instru-
mament consists of a 23-item questionnaire that is answ-ered
by parents or guardians probing the language development
of children with cochlear implants who are in the 2-5-
year-old age band. Respondents answer questions with a
five-point scale. The FAPCI is being utilized in several ongo-
ing NIH-funded studies of pediatric CI and has already been
translated into German.21 There is no child self-report ver-
sion of the FAPCI.

A series of studies carried out by the group that de-
veloped the FAPCI15,22,23 showed that despite the establish-
ment of good communicative capacity, children with CI were not
communicating on par with their peers and were struggling
to communicate with oral language in social environments,
including school. Therefore, the primary aim of this study
was to translate, adapt, and test the reliability of the FAPCI
for use with Brazilian children. The second aim of this
work was to test the sensitivity of the FAPCI translated and
adapted to Brazilian Portuguese (FAPCI-BP) in the evalua-
tion of language development in NH children versus children
using CI.

Methods

Participants

This research was approved by the Institutional Research
Committee for Human Subjects. The study included children
between 2 and 9 years of age, of both sexes, who were treated as outpatients at the Pequeno Príncipe Children’s Hospital in the city of Curitiba, Brazil. The CI sample included children aged 2-9 years who had undergone CI and had been living with activated implants for at least six months. The NH group consisted of similarly aged children with no otological, neurological, or neuropsychiatry complaints. Parents or guardians accompanying the children provided written informed consent and answered the FAPCI-BP. Table 1 summarizes the demographic characteristics of the children in the NH and CI groups, and Table 2 details the clinical characteristics of the CI participants. The FAPCI-BP was completed by a total of 131 parents of 144 children. The clinical characteristics of the 13 cochlear-implanted children of the CI group are summarized in Table 3.

Procedures

A two-step strategy was implemented: (1) translation, retro-translation, and adaptation of the FAPCI; and (2) administration of the FAPCI-BP to children with CI and children with NH.

FAPCI

The FAPCI is a 23-item written questionnaire designed to measure verbal communicative performance in children 2 to 5 years of age after CI. It is completed by the parents or the guardians of the subjects and can be finished in about 5-10 minutes. There are three response mode formats: frequency (response levels never, rarely, sometimes, often, always); quantity/proportion (number or percentage of occurrences, e.g. 0-4%, 5-24%, 25-49%, 50-95%, or 96-100%); and examples (responses offer descriptions or examples of behaviors, and levels correspond to an ordinal scale). Each answered item is translated into a score ranging from 1 point (e.g. for never) to 5 points (e.g. always), regardless of the type of question, and the unanswered questions are scored as 0 points, yielding a maximum total score of 115. If the number of unanswered questions exceeds two, the questionnaire is considered invalid. If more than one answer is marked, the higher answer is taken. Mean scores were compared between the two groups and reported with standard deviations (SDs). This instrument was developed to complement other tests of spoken language competence to enable assessment of communicative capacity in children with CI.

Table 1  Demographic summary of study subjects.

<table>
<thead>
<tr>
<th>Variables</th>
<th>Participants</th>
<th>NH</th>
<th>CI</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sex</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Girls</td>
<td>50</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Boys</td>
<td>81</td>
<td>4</td>
<td>9</td>
</tr>
<tr>
<td>Age (years)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>2-4</td>
<td>84</td>
<td></td>
<td></td>
</tr>
<tr>
<td>5-9</td>
<td>47</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

NH, normal hearing; CI, cochlear implantation.

Step 1: Cross-cultural adaptation and validation process

Authorization by the original instrument’s author for the translation, adaptation, and validation of the FAPCI for the Brazilian population was obtained, and the process was conducted in accordance with the guidelines established by Beaton et al. The FAPCI was translated from English to Portuguese by a professional translator familiar with both languages. Small changes were necessary in order to adapt the verbiage to Brazilian culture, but the original essence of the questions was maintained as much as possible. The FAPCI-BP is presented in its entirety as an appendix with the approval of the developers.

The adapted questionnaire was sent to another professional translator who was unfamiliar with the original questionnaire for retro-translation into English. An equivalence of construction analysis was performed in which the original and retro-translated English versions were compared to determine whether there were significant differences in the content of the questions, that is, whether the FAPCI-BP was faithful to the structure of the original questionnaire.

Table 2  Clinical characteristics of CI group participants.

<table>
<thead>
<tr>
<th>Subject</th>
<th>Diagnosis type</th>
<th>Age at CI (years)</th>
<th>Hearing time (years)</th>
<th>FAPCI score</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Idiopathic</td>
<td>6.8</td>
<td>2.3</td>
<td>43</td>
</tr>
<tr>
<td>2</td>
<td>Genetic</td>
<td>3.8</td>
<td>2.8</td>
<td>87</td>
</tr>
<tr>
<td>3</td>
<td>Idiopathic</td>
<td>3.2</td>
<td>2.1</td>
<td>35</td>
</tr>
<tr>
<td>4</td>
<td>Congenital Rubella</td>
<td>4.9</td>
<td>1.5</td>
<td>67</td>
</tr>
<tr>
<td>5</td>
<td>Meningitis</td>
<td>2.1</td>
<td>4.7</td>
<td>70</td>
</tr>
<tr>
<td>6</td>
<td>Idiopathic</td>
<td>2.5</td>
<td>1.7</td>
<td>40</td>
</tr>
<tr>
<td>7</td>
<td>Neonatal hypoxia</td>
<td>1.9</td>
<td>1.6</td>
<td>88</td>
</tr>
<tr>
<td>8</td>
<td>Genetic</td>
<td>2.9</td>
<td>4.5</td>
<td>74</td>
</tr>
<tr>
<td>9</td>
<td>Meningitis</td>
<td>2.0</td>
<td>1.1</td>
<td>31</td>
</tr>
<tr>
<td>10</td>
<td>Idiopathic</td>
<td>4.4</td>
<td>1.4</td>
<td>67</td>
</tr>
<tr>
<td>11</td>
<td>Idiopathic</td>
<td>3.3</td>
<td>1.7</td>
<td>87</td>
</tr>
<tr>
<td>12</td>
<td>Cochlear nerve hypoplasia</td>
<td>3.7</td>
<td>0.5</td>
<td>28</td>
</tr>
<tr>
<td>13</td>
<td>Idiopathic</td>
<td>2.0</td>
<td>1.1</td>
<td>42</td>
</tr>
</tbody>
</table>

CI, cochlear implantation; FAPCI, Functioning after Pediatric Cochlear Implantation.
To test for internal consistency, a subgroup of 34 parents of children with NH and 13 parents of children with CI completed the FAPCI-BP twice with a time interval of at least two weeks, but not more than one month. Cronbach’s alpha was used to verify the internal consistency of the instrument’s items between the first and second runs. A construct can be validated indirectly with an internal base of consistency or no relation between the questions that make up part of the scale, allowing the conclusion that the scale has a valid construction.\textsuperscript{15} Cronbach’s alpha coefficient is the simplest and best-known measure of internal consistency, and is the primary approach used in scale validation. In general, a group of items that explore a common factor should have a high alpha value. The minimum acceptable value for the alpha coefficient is 0.70; alpha values greater than 0.80 are preferable.\textsuperscript{25}

Step 2: Applicability of the FAPCI-BP

The FAPCI-BP was answered by parents of NH children and parents of CI children. The results were subjected to statistical analyses in R software, version 3.0.1 (R Project for Statistical Computing, University of California, CA, USA). The data were verified in relation to normality and descriptive analyses using Wilcoxon tests. Comparisons were considered significant when they had two-tailed p-values < .001.

Results

Adaptation and internal consistency

After comparison of the retro-translation to the original English and consideration of cultural linguistic use, it was determined that several items needed to be adapted and one item needed to be withdrawn (question 16), as shown in Table 1, to obtain a final version of the adapted questionnaire that was consistent with the original. The Cronbach’s alpha for internal consistency was 0.948 for the NH group and 0.964 for the CI group, with no questions observed to be outside the expected average, indicating that the instrument had good internal consistency.

FAPCI-BP trial

Comparison of the groups’ overall mean scores ± SDs revealed that the NH group performed significantly better on the FAPCI-BP (100.38 ± 23.5) than did the CI group (63.00 ± 21.0; p < .001, Wilcoxon test). The mean scores ± SDs obtained for children in the NH group divided by chronological age are reported in Table 3 together with score data for children in the CI group, separated by chronological age and the children’s ages at the time hearing was established. The mean scores by age of hearing within each age year bin are presented as means without SDs, since the subgroups were small and irregular. The group medians and distributions are illustrated in a boxplot graph in Fig. 1.

Discussion

The goal of CI is not only that children will gain functional auditory processing skills, but also that they will develop the skills needed to communicate effectively with spoken language. The FAPCI is the only currently available instrument that allows the impact of CI on communication skills to be measured in children 2-5 years old. The present study produced a Portuguese-language FAPCI version adapted for use in the Brazilian population (see appendix for the final version of FAPCI-BP). Consistent with the American\textsuperscript{18} and German\textsuperscript{19} versions, the Brazilian FAPCI had excellent internal consistency (Cronbach’s alpha > 0.90). Additionally, the expected gap in communicative performance was observed between children with CI and children with NH.

After translation and retro-translation of the original FAPCI, very few questions needed cultural adaptation, and only one item was omitted (question 16). The Cronbach’s alpha value for internal consistency that was obtained for
the 22-item FAPCI-BP was similar to that of the original 23-item version, which was validated in a study of 75 families (alpha = 0.86).\textsuperscript{18,23} The alpha value serves as an index of an instrument's reliability in situations where the researcher is not able to perform additional interviews of the individuals in question, but requires an estimate of the average degree of error.\textsuperscript{23}

Examining the scores by age enabled several inferences to be made. Firstly, it was noticed that FAPCI-BP performance was relatively stable across ages within the NH group, especially among children from age 3 through 8 years. Only the youngest (2-year-olds) and oldest (9-year-olds) had non-overlapping SDs, which is not altogether surprising given that, normally, children exhibit a great linguistic expansion between 2 and 3 years of age and are still developing basic linguistic skills. Regardless, there was at least one child within each age year bin achieved the maximum possible score (115). The greater the age, the more children achieved the maximum possible score. Yet, even in the upper age groups, there were always some children that performed below the maximum, raising the question of whether the instrument may also be valid beyond the stated upper limit of 9 years of age. Regardless, it should be noted that there were quite small numbers of children in the age 7-, 8-, and 9-year subgroups. Thus, the data for those subgroups is likely less reliable than the data for the lower age subgroups, which were substantially larger.

The comparison of scores between the NH and CI groups demonstrated a significant lingering communicative deficit among children with CI. Moreover, examining the scores of the individual children with CI, it is apparent that they had not achieved communicative performance on par with their peers. There are several factors that may affect the ability of children with CI to achieve optimum-level communicative performance, including age of onset of deafness, age at CI activation, use of speech therapy/rehabilitation, underlying pathology, and the presence of other disabilities, not to mention inter-individual variation in general, which can be substantial.\textsuperscript{26} Age of implantation appears to be a particularly important factor in language outcome. Children implanted when they were 16–24 months of age had Preschool Language Scale scores matching their hearing peers at ages 4 and 6.\textsuperscript{7} In the present study, only one child received CI before 24 months of age, and only three children received CI at approximately 24-25 months old. The majority of participants in this study received CI after 3 years of age. Thus, the comparison of NH and CI groups can be considered preliminary; more studies are needed with a larger sample of CI children receiving CI before 24 months of age.

It is expected that the sooner the CI is performed, the better the outcome will be. The present study’s CI group was quite heterogeneous in terms of age of CI, pathology, and hearing rehabilitation. Most children in the CI group were implanted and activated between 3 and 4 years of age, which is considered borderline for the critical period of auditory pathway development in the brain. Two of the children (No. 5 and No. 9) lost their hearing as a sequela of meningitis very early in life, when they were considered to be prelingual. The critical period for language development is thought to end at about 3.5 years of age for children with congenital deafness; children who received CI after their fourth birthday show greater variation in outcome than children who received CI when they were younger than 4 years old.\textsuperscript{17,21}

To minimize the duration of deafness and lack of critical auditory stimulation, CI surgery should be performed before 2 years of age.\textsuperscript{27,28} However, even when impairment is detected with a newborn hearing test, progression of referrals to hearing specialists in Brazil can be quite slow and the frequency of CIs performed is inadequate in many places (i.e. two per month in the state of Paraná, only one per month each at the Pequeno Príncipe Hospital and the Hospital das Clínicas, personal communication). Although CI is covered by the Brazilian public health system, children needing CI often have aged beyond the critical period for optimum results by the time they receive the intervention, due to long waiting times. Newborn screening programs in Brazil are also inadequate. It is particularly important for infants to be screened in neonatal intensive care units, given their elevated prevalence of affected newborns. For example, one study reported that, among 979 newborns in an intensive care unit, 10.2% presented with unilateral and 4.9% presented with bilateral auditory brainstem response impairments.\textsuperscript{29}

Furthermore, after CI, children should commence immediately with auditory rehabilitation and proper speech therapy, in addition to stimulation by the family through communicative experiences. Children are subjected to a variety of therapies, which are not always the most suitable for rehabilitation from deafness and language development. Speech therapists are not present in every city, and even when properly trained therapists are present, often there are not a sufficient number to accommodate the need. Moreover, it is important that the therapy be individualized to meet the specific needs of each child and be of sufficient duration to allow the child to absorb the treatment.

In evaluations of candidates for CI, psychologists and social workers consider the commitment and involvement of the families in the treatment, and assess whether the families are capable of bearing the expense of maintaining the appliance. Rechargeable batteries have reduced the expenses. However, in order to conserve batteries and
delay replacement, some children do not use their implants throughout the day, but rather turn them on only when they are in school. Another difficult issue for families is the cost of spare parts, such as cables and antennas, which when damaged prevent the use of the implants, leaving children without auditory stimulation.

This study has some limitations. First, because the sample of CI children was small and heterogeneous, it is not possible to extrapolate the results to all CI children. Clearly, larger studies are needed to enable multiple variables to be controlled. Also, studies comparing FAPCI-BP results with the results of traditional widely-used tests of language use are needed, since the FAPCI-BP is a new instrument. In principle, it is expected that FAPCI-BP scores should increase within children with implants over time in relation to increases in cumulative therapy and stimulation experience, as observed with the American16 and German versions.21 Likewise, the FAPCI-BP should be useful for phonological monitoring of patients, particularly in relation to revealing which communicability areas may be lagging. The aforementioned limitations notwithstanding, this study established a Brazilian version of the FAPCI with excellent internal consistency. Second, even though the FAPCI was designed originally to test the communicative skills of children in the 2-5-year-old age band, this study included older children, up to 9 years of age. This was done in order to compare their FAPCI-BP results with results obtained for NH children, similar to previous studies validating other versions the FAPCI which included subjects up to 10 years of age.6 The scores of the small number of children over 5 years old examined here did not appear to differ markedly from the scores of younger children. This result is not surprising given that, normally, the bulk of fundamental language development is thought to occur by the age of 5 years.6

In conclusion, the recently developed FAPCI-BP is the first instrument to allow functional language development to be measured in Brazilian children using cochlear implants. After translation and adaption, the FAPCI-BP showed excellent internal consistency and demonstrated the expected gap between NH and CI groups, indicating that it is valid for use in Brazilian children. This work paves the way for future studies in Brazil, such as applying the FAPCI-BP to develop score growth curves in NH children to serve as framework for interpreting scores in children with CI. Although the number of subjects with CI in this study was small, it was possible to establish that the FAPCI-BP could be very useful to Brazilian physicians and healthcare providers as a reliable metric of the development of communication skills in their CI patients. The FAPCI-BP may be particularly useful for clarifying diagnoses as well as for directing and revising rehabilitative plans, and thus bettering the prospects of a good quality of life for children with CI.

Acknowledgments

This study was supported by the Department of Science and Technology of the State of Paraná. The authors thank psychologist Cassia Benko for her assistance, and the children and their families for participating in the study. They also thank Dr. John K. Niparko for providing the opportunity to translate and adapt the FAPCI to Portuguese.

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