

Assessing the Use of Speech and Language Measures in Relation to Parental Perceptions of Development After Early Cochlear Implantation

*Frank R. Lin, †Nae-Yuh Wang, †Nancy E. Fink, ‡Alexandra L. Quittner,
§Laurie S. Eisenberg, ||Emily A. Tobey, *John K. Niparko,
and the CDaCI Investigative Team

**Department of Otolaryngology-Head and Neck Surgery and †Johns Hopkins Welch Center, Johns Hopkins School of Medicine, Baltimore, Maryland; ‡Department of Psychology, University of Miami, Miami, Florida; §House Ear Institute, Los Angeles, California; ||School of Behavioral and Brain Sciences, University of Texas-Dallas, Dallas, Texas, U.S.A.*

Objective: Clinicians and investigators use multiple outcome measures after early cochlear implantation (CI) to assess auditory skills, speech, and language effects. Are certain outcome measures better associated with optimal childhood development from the perspective of parents? We studied the association between several commonly used outcome instruments and a measure of parental perceptions of development to gain insight into how our clinical tests reflect parental perceptions of a child's developmental status.

Study Design: Cross-sectional analysis.

Setting: Six academic centers.

Patients: One hundred eighty-eight deaf children (<6 yr) 1 year after CI activation enrolled in the longitudinal Childhood Development after CI study.

Main Outcome Measures: Measures of auditory skills, speech, and language. Parental perceptions of development quantified with a visual analogue scale (visual analogue scale–development).

Methods: Nonparametric and parametric regression methods were used to model the relationship between outcome measures and visual analogue scale–development scores.

Results: All outcome measures were positively associated with parental perceptions of development, but more robust associations were observed with language measures and a parent-report scale of auditory skills than with a selected measure of closed-set speech. For speech and language data, differences were observed in the trajectories of associations among younger (2–3 yr) versus older (4–5 yr) children.

Conclusion: Our results demonstrate the importance of measuring multiple outcome measures after early pediatric CI. The degree to which an outcome measure reflects childhood development as perceived by parents may be affected by the child's age. Measures that are based on parental report and broader outcome measures focused on verbal language offer the potential for a fuller understanding of the true effectiveness of early implantation. **Key Words:** Language—Outcomes—Pediatric cochlear implantation—Speech.

Otol Neurotol 00:00–00, 2007.

Access to sound provided by a cochlear implant (CI) can enable prelingually deaf children to attain a cascade of developmental benefits (Fig. 1) (1). Restoration of auditory inputs allows a child to develop the skills needed for sound discrimination and identification (2). Such

access can promote speech and language acquisition that then serves as the foundation for effective communication between a child and a caregiver (3–5). A child's ability to participate in bidirectional communicative interactions with parents and caregivers during critical infant-toddler stages subsequently allows children to reach critical milestones in childhood development (6,7).

Currently, most studies of pediatric CI have focused on assessing auditory skills and speech, rather than downstream targets such as verbal language ability or communication skills (4). This most likely stems from the ease of measuring these outcomes in a clinical setting

Address correspondence and reprint requests to Dr. Frank R. Lin, Department of Otolaryngology-Head and Neck Surgery, Johns Hopkins Outpatient Center, Room 6163, 601 North Caroline St., Baltimore, MD 21287; E-mail: flin1@jhmi.edu

This work was supported by the National Institute on Deafness and Other Communication Disorders (grant nos. R01 DC04797 and T32 DC00027).

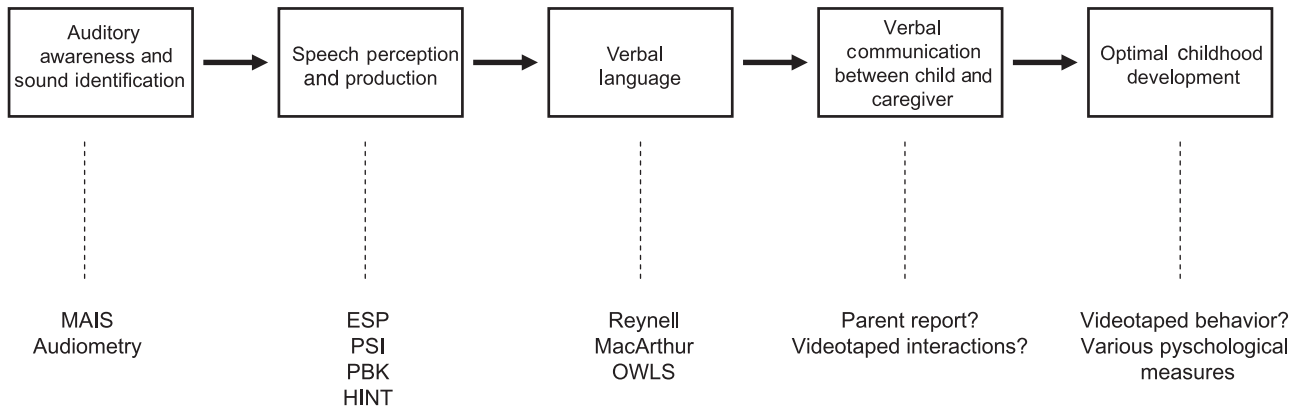


FIG. 1. Simplified model of domains of development hypothesized to result from cochlear implantation (CI) of prelingually deaf children. Outcome measures used with CI can target multiple points along this cascade, with most current measures focused on auditory skills, speech, and language. Measures of communication and childhood development are less well defined. MAIS indicates meaningful auditory integration scale; ESP, Early Speech Perception Test; PSI, Pediatric Speech Intelligibility Test; PBK, Phonetically Balanced Kindergarten Word List; HINT, Hearing in Noise Test; OWLS, Oral and Written Language Scales. The various psychological measures used to assess child development can include measures of visual attention, social skills, and child behavior.

because speech perception measures are routinely performed by most audiologists. Measuring linguistic capability requires additional time and personnel specifically trained in conducting tests of verbal language. In addition, current methods of outcomes assessment for CI children have evolved from a historical basis where many deaf children never progressed far beyond basic speech recognition and production, thereby obviating the need for sophisticated tests of verbal language ability.

The most important overarching objective of CI in children should be to achieve optimal development. Are all outcome measures that target facets of the developmental cascade of benefits (Fig. 1) equal in this regard, or are some more highly associated with global developmental outcomes?

In the current study, we evaluated the associations of performance measures of language (MacArthur Communicative Development Inventory and Reynell Developmental Language Scales), speech (Early Speech Perception Test), and auditory skills (Meaningful Auditory Integration Scale) measured 1 year after CI in a cohort of prelingually deaf children in relation to a measure of parental perceptions of development. In the absence of a criterion standard measure of optimal childhood development, we assessed parental perceptions of development because parents are uniquely situated to observe a child's developmental growth and abilities in a home setting on a day-to-day basis. Assessing the relationship between parental perceptions of development with currently used outcome measures may provide insight into how well our clinical instruments reflect a child's abilities as demonstrated in natural environments.

METHODS

Overview

Ascertaining the relationship between parental perceptions of development measured with a visual analogue scale (VAS-

development) and auditory skills/speech/language scores is complicated by the presence of effect modifiers and confounders. An effect modifier is a factor that moderates the relationship between two variables (8). In the current study, child age is an effect modifier because it influences how parents would perceive their child's level of functioning. Whereas a mother may respond with a VAS-development score of 90 or 95 if her 1-year-old child is consistently using two- to three-word sentences at home, a much lower score would most likely be reported if her child was a 5 year old. Several possible factors may also confound the relationship between VAS-development and language scores. A confounder is a factor that is associated with both variables of interest without lying in the causal pathway between the variables (8). A confounder may therefore mask real associations between the two variables or lead to a spurious association that is only present because of the confounder. We hypothesized that several factors associated with speech and language outcomes in cochlear-implanted children (parental education/income status, sex of the child, family size) may also be associated with parental perceptions and, therefore, act as potential confounders (9,10).

To account for effect modifiers and confounders when investigating the association between VAS-development and language scores, we first applied nonparametric methods (lowess smoothers) while stratifying by the effect modifier (child age). This approach allows us to observe the individual relationships between VAS-development and language scores that may be different at each age, but it does not allow us to control for multiple confounding factors. In a second step, parametric methods were used to control for multiple potential confounders and to assess the statistical significance of relationships observed with the nonparametric approach.

Study Cohort

This study used 1-year post-CI activation data gathered in the Childhood Development after CI (CDaCI) study, a National Institute on Deafness and Other Communication Disorders-funded, multisite, longitudinal study of language and developmental outcomes after CI. A detailed description of the study cohort and the design of the study is available elsewhere (11). From 2002 to 2004, 188 prelingually deaf children younger than

5 years were consecutively enrolled at 6 study sites: House Ear Institute, Johns Hopkins, University of Miami, University of Michigan, University of North Carolina, and University of Texas-Dallas. All children were implanted with a multichannel device using a transmastoid, transfacial recess approach, and data presented in this study were gathered 1 year after CI activation. Parents of all children were hearing and committed to educating their children in a verbal language environment. This study was approved by institutional review boards at all participating study centers. Brief characteristics of the study cohort at 1-year post-CI activation are presented in Table 1.

CI Outcome Measures

The MacArthur Communicative Development Inventory–Words and Gestures is a parent-reported measure of language ability that is appropriate for 8- to 16-month-old children or older children with language delays (12). Scores from Part D (vocabulary checklist) of the instrument were used consisting of 396 commonly used children’s words. Scores correspond to the number of words parents report that the child both understands and says. The Reynell Developmental Language Scales are a clinician-administered test of a child’s language abilities conducted in a standardized (clinic) environment. The Reynell assesses verbal comprehension and expressive language for children older than 1 year (13). Correct scores for each scale (Reynell Verbal and Reynell Expressive) were summed to provide an overall total score for each child. Age-standardized scores for the Reynell (normalized to a nationally representative cohort of hearing children) were not able to be used due to most CI children at 1 year still scoring less than the standardized floor score of 63.

The Early Speech Perception (ESP) Test is a closed-set test that examines pattern perception (differentiation of number of syllables and stress patterns), spondaic word identification, and

monosyllabic word identification using toy objects to represent the stimulus items (14). The Meaningful Auditory Integration Scale (MAIS) is a parent-report scale used to assess auditory behavior and is composed of 10 stimuli that assess auditory awareness, discrimination, and vocal behavior (15). For both ESP and MAIS, low-verbal versus standard editions (ESP) or infant-toddler versus preschool editions (MAIS) were used accordingly dependent on subject age. Further details of ESP and MAIS administration in the CDaCI study are available elsewhere (16).

Measure of Parental Perceptions

A visual analogue scale (VAS–development) was presented to parents of study subjects as a 10-cm horizontal line anchored at no development (0)/perfect development (100). Parents were asked to draw a line signifying their perceptions of their child’s development during the past 4 weeks while considering such factors as their child’s “ability to communicate and respond to others” and “development of language and ability to express his/her thoughts” after the Infant and Toddler Health Questionnaire domains developed by Klassen et al. (17). The use of VASs to quantify subjective perceptions has been extensively used in previous studies (18,19).

Data Cleaning and Statistical Analysis

Verification of data integrity and data cleaning was performed by the data coordination center for the CDaCI study at the Johns Hopkins Welch Center. To analyze the MacArthur and Reynell data using the same scale, data from both tests were standardized by dividing by the highest score obtained in the cohort on the MacArthur (396) and the Reynell tests (123). Language scores, therefore, ranged from 0 to 1 and represent the fraction of the highest language score obtained by a subject. Nonparametric regression methods (locally weighted least squares [lowess]) or box plots were used to analyze the shape of the relationship between VAS–development and outcomes scores (20,21). Parametric linear regression analyses were then used to test the significance of the relationships observed with the nonparametric smoothers or in the box plots, while controlling for potential confounders (household income [$<US \$50,000$, $\$50\text{--}100,000$, $>\$100,000$], child sex, parental education [college graduate, yes/no], and presence of other children in the household). Visual analogue scale–development scores were regressed against language scores and confounders using ordinary least-squares estimates of β coefficients.

For language tests in 2- to 3-year-old children, a linear spline term was introduced to test for the significance of a transition point in the observed trajectory at a language level of 0.15. For language tests for 4- to 6-year-old children and MAIS tests for all children, the significance of a linear increase in VAS–development with outcome scores was tested. Linear regression model assumptions were checked. Robust variance estimates were used in models that had heteroscedasticity in model residuals. Subjects with missing data in the studied variables due to improperly filled-out forms or other reasons were excluded from the analysis using a complete-subject analysis approach. This was less than 10% of the study subjects for all analyses except for ESP tests. For ESP testing, data were only available on 111 of 188 subjects due to respondent fatigue and behavior issues as well as some children not having the requisite cognitive skills to complete the assessment. All analyses were performed using Stata 8.2 software (College Station, TX, USA). Statistical significance was accepted at a two-sided $\alpha < 0.05$ level.

TABLE 1. Characteristics of the CDaCI study cohort at 1-year post-CI activation

	CI subjects, n = 188
Age (yr)	
2 ^a	79 (44)
3	35 (20)
4	36 (20)
5	19 (11)
6	10 (6)
Male	91 (48)
Respondent education	
<High school	14 (7)
High school graduate	26 (14)
Some college	55 (29)
College graduate	92 (49)
No response	1 (1)
Household income (US \$)	
<15,000	15 (8)
15,000–29,999	22 (12)
30,000–49,999	42 (22)
50,000–74,999	31 (16)
75,000–100,000	26 (14)
100,000+	31 (16)
Declined/do not know	21 (11)
Other children	
Yes	128 (69)

Data presented as “count (%).”

^aThis group consisted of all 2-year-olds except for 2 children who were 21 and 23 months old.

RESULTS

Visual analogue scale–development and language data were independently modeled for each age group (2, 3, 4, 5, and 6 yr) using nonparametric methods. Similar relationships were observed for 2- to 3-year-old and 4- to 6-year-old children, and data were pooled accordingly for all subsequent measures. Figure 2 depicts the relationship between VAS-development and language scores stratified by child age (2–3 versus 4–6 yr). Results from the MacArthur and Reynell demonstrated consistently similar trajectories after stratification by age despite the MacArthur being a parent-reported metric, whereas the Reynell is a clinician-administered test. Markedly different trajectories were observed for the 2- to 3- versus the 4- to 6-year-old age group. In 2- to 3-year-olds, initial increases in language scores were associated with steeply increasing VAS-development scores, but threshold VAS-development scores were reached at 15% of the total language score. In contrast, in children who are 4 to 6 years old, improvements in language ability were linearly associated with increasing VAS-development scores during the entire range of the language tests without a threshold effect. This result suggests that parents of 2- to 3-year-old children may have different (lower) expectations for language development than parents of older children. After controlling for household income, parental education, child sex, and the presence of other children, we found strong statistical evidence for the observed trends in the 2- to 3-year-old age group (MacArthur [$p = 0.03$] and Reynell [$p < 0.001$]) and in the 4- to 6-year-old age group (MacArthur [$p = 0.006$] and Reynell [$p = 0.03$]).

In contrast, the relationship between VAS-development and speech perception (ESP) test scores demonstrated that higher speech perception scores were only marginally associated with higher VAS-development scores (Fig. 3). For 2- to 3-year-old CI children, mean VAS-

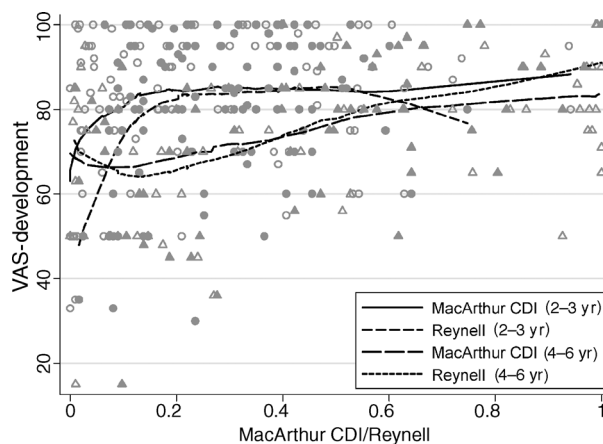


FIG. 2. Nonparametric trajectory of VAS-development over MacArthur and Reynell language scores. *Solid circle* indicates MacArthur (2–3 yr old); *hollow circle*, Reynell (2–3 yr old); *solid triangle*, MacArthur (4–6 yr old); *hollow triangle*, Reynell (4–6 yr old).

development scores were 79.2 for children with ESP category scores of 1 to 2 (none to some pattern perception) versus 84.5 in children with ESP category scores of 3 to 4 (some to consistent word identification) ($p = 0.30$). For the 4- to 5-year-old children, mean VAS-development scores were 68.9 for children with ESP category scores of 1 to 2 versus 80.5 in children with ESP category scores of 3 to 4 ($p = 0.14$). Interestingly, for the older age group, ESP scores seem to better reflect parental perceptions of development, with a clear trend toward higher VAS-development scores observed in children with better speech perception.

Assessment of auditory skills with the MAIS demonstrated robust associations between VAS-development scores and MAIS scores (Fig. 4). For both the younger and older age groups, higher MAIS scores were linearly associated with increasing VAS-development scores ($p < 0.001$ and $p = 0.03$, respectively).

DISCUSSION

We investigated the association of language, speech, and auditory skill outcome measures used with CI and parental perceptions of development. We found that all outcome measures were positively associated with parental perceptions of development, but more robust associations were observed with language measures and the parent-report MAIS scale of auditory skills.

Our results demonstrate the importance of measuring multiple outcome measures after early pediatric CI, and that an emphasis should be placed on broader downstream measures such as verbal language. This finding reflects the increasing recognition that CI of prelingually deaf children represents a profound departure from the results that would have been previously expected with conventional amplification strategies. Studies of pediatric CI, including the present CDaCI study, are increasingly focused on tracking the progress of CI children relative to normal-hearing peers, a decision that would have seemed extraordinary before the advent of current multichannel CIs.

In addition, our results demonstrate that outcome measures based on parent-report should also be increasingly emphasized. We found a strong association between a measure of basic auditory skills (MAIS) that was based on a child's behaviors at home and VAS-development scores. Language performance measures also demonstrated that the MacArthur and Reynell yielded extremely similar results (Fig. 2) despite the MacArthur being a parent-report checklist of words and the Reynell being a clinician-administered 1-hour test. Relative to clinician-administered metrics such as the MAIS and Reynell, parent-report instruments such as the MAIS and MacArthur seem to yield similar results that reflect parental observations of their child's developmental progress at home.

Speech perception results from the ESP demonstrated weaker associations with VAS-development scores. This finding likely reflects the discrepancy between

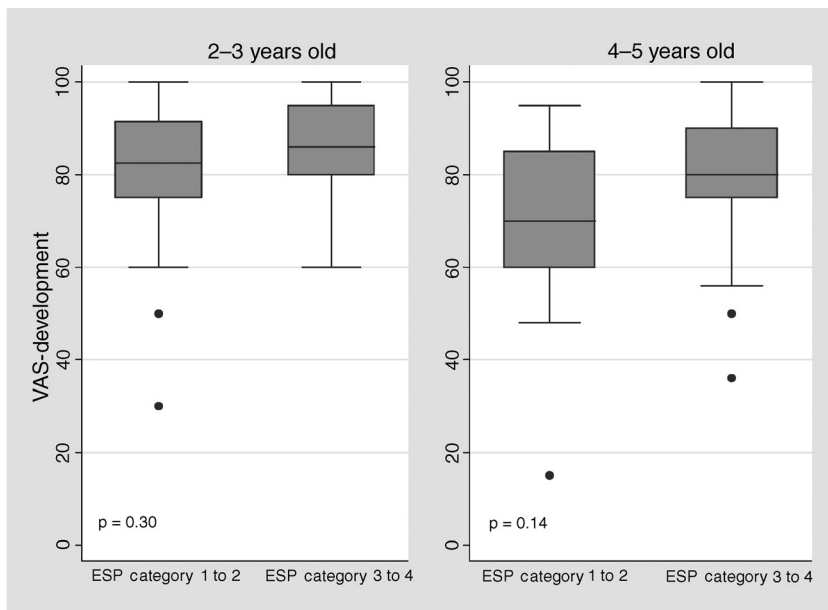


FIG. 3. Box plots of VAS-development scores stratified by ESP test scores. ESP categories 1 to 2 indicate none to some pattern perception; ESP categories 3 to 4, some to consistent word identification.

this particular clinician-administered test and a child’s functioning in everyday life, where numerous other factors (environmental noise, siblings) impact a child’s ability to communicate. Broader outcome measures focused on language or even lower-order auditory skills when based on parental report most likely better take these other factors into account. Other speech perception tests in addition to the ESP that are focused on open-set recognition would most likely demonstrate more robust associations with parental perceptions of development. Child age also seems to mediate the relationship between speech data and VAS-development scores with a clearer association observed in older (4–5 yr) than in younger (2–3 yr) children. This finding suggests that speech ability as measured by the ESP may have a differential impact on parental perceptions of childhood development that is dependent on a child’s age.

Limitations

The major limitation related to our current study relates to our use of the VAS-development metric as an indicator for childhood development. Our decision to use the VAS metric reflects the current lack of a criterion standard to practically assess a preschool-aged child’s development at home. Although one ideal alternative would be to use video recordings of child behavior at home (22,23), video-analytic techniques are less practical given the expense and time required to obtain recordings and objectively code observed patterns of behavior. The use of a parent-report metric also offers significant advantages. First, parents are uniquely situated to observe a child’s growth and abilities in a home setting on a day-to-day basis. Second, recent nonbinding recommendations from the Food and Drug Admin-

istration, emphasizing that some treatment effects are known only to the patient, have called for the use of patient-reported measures that directly reflect outcomes salient to the patient and family (24). Finally, previous works have clearly established the importance of the parent-child dyad for future language growth and optimal childhood development (6,7,25). Parental assessments of childhood development then should be a valid approach to assessment because a child’s true developmental status is likely to be reflected in parental perceptions of their interactions.

The scores from the VAS-development metric primarily reflect a child’s developmental status at home. However, given the relative simplicity of the testing metric,

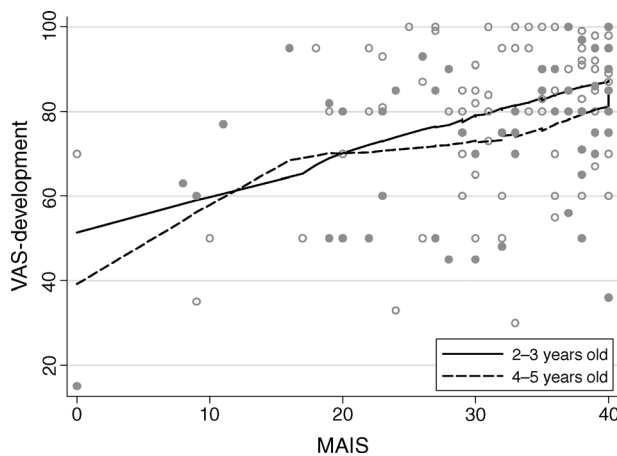


FIG. 4. Nonparametric trajectory of VAS-development over MAIS scores. *Hollow circle* indicates 2 to 3 years old; *solid circle*, 4 to 5 years old.

numerous other factors unrelated to the child's developmental status can also affect scores (e.g., parental socioeconomic status, inadvertent parental rating of developmental change during the last 4 weeks as opposed to overall developmental status, parental numeracy skills, child's motor skills). However, the impact of these other factors on VAS-development scores may be minimal given the wording of the VAS-development metric and the characteristics of our study cohort. All children enrolled in the study had no substantial cognitive or developmental delays (defined as a Bayley mental or motor development index score of >70 or a Leiter-R IQ >66), and all parents had normal hearing and, therefore, primarily focused on communicative aspects of their child's development. Most importantly, however, bias introduced by any other factors would most likely be nondifferential and bias any observed association toward the null, whereas we found strong statistically significant evidence for our observed results (8). We also confirmed that our observed results were not systematically biased by using linear regression models to control for confounding factors such as household income and parental education.

Future Directions

Current clinical measures of outcomes after early CI focus on auditory skills, speech, and language. All are useful and are reflected in parental observations of their child's development. However, as demonstrated in Figure 1, one facet of the developmental cascade that is not currently addressed by current outcome instruments is an overarching assessment of a child's communicative ability by caregivers, namely, how well a child uses verbal language in real-world situations to convey thoughts and needs. Guided by these observations, we are completing the development of a new parent-report instrument designed for 2- to 5-year-old CI children that will directly measure a child's communicative performance. In the Functioning after Pediatric Cochlear Implantation instrument, items were specifically designed using parent input and the conceptual framework of the World Health Organization's model of functioning and disability to provide salient situational and behavioral contexts for assessing verbal communicative performance (26). We think that this instrument will serve as a useful adjunct to currently available clinical measures to comprehensively assess the developmental progress of the cochlear-implanted child younger than 5 years.

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