

Title

Trajectory of Auditory and Language Development in the Early Stages of Pre-lingual Children Post Cochlear Implantation: a Longitudinal Follow up Study

Gang Li^a, Fei Zhao^b, Yong Tao^a, Lin Zhang^a, Xinyi Yao^a and Yun Zheng^{a*}

^aHearing Center/Hearing & Speech Science Laboratory, Department of Otolaryngology/Head and Neck Surgery, West China Hospital of Sichuan University, Chengdu, China

^b Center for Speech and Language Therapy and Hearing Science, Cardiff School of Sport and Health Sciences, Cardiff Metropolitan University, Cardiff, UK.

*Corresponding Author: Yun Zheng, MD/PhD, Director, Hearing Center/Hearing & Speech Science Laboratory, Department of Otolaryngology/Head and Neck Surgery, West China Hospital of Sichuan University, No. 37 Guo Xue Xiang, Chengdu, Sichuan, China 610041, E-mail: 1141679315@qq.com

Abstract

Objectives: The aim of this longitudinal follow-up study was to explore the trajectories of early auditory and language development in Mandarin speaking children younger than 3 years of age following switch-on of their cochlear implants (CIs).

Methods: Early auditory and language development was measured longitudinally using the Infant-Toddler Meaningful Auditory Integration Scale (IT-MAIS), which is a commonly used tool for assessing early prelingual auditory development (EPLAD) in children, and the subtest (Words and Gestures, W&G) of the simplified short form version of the Mandarin Communicative Development Inventory (SSF-MCDI) to assess receptive and expressive vocabulary growths of children in 24 pediatric cochlea implant recipients at baseline, 3, 6, and 12 months following switch-on. Age at switch-on ranged from 1 to 3 years of age. Participants were divided into two groups based on age at switch-on. The IT-MAIS and SSF-MCDI (W&G) scores were analyzed with comparison to normal children, unaided hearing-impaired children, and CI children.

Results: Significant improvements in IT-MAIS and SSF-MCDI (W&G) scores from baseline to 12 months were seen after switch-on in both CI groups and were comparable to the normal hearing children in the first year of age. The IT-MAIS scores of CI children in both groups at 12 months after switch-on surpassed the average level of unaided peers with profound hearing loss and were similar to the

average level of unaided peers with mild hearing loss. SSF-MCDI (W&G) scores in word comprehension and expression were significantly different between groups at some intervals.

Conclusions: Children younger than 3 years of age with cochlear implants have similar trajectories in early auditory and language developments to normally hearing children. Moreover, early implantation is an important factor for the early auditory development when comparing EPLAD results between CI children and unaided peers with different hearing loss. Finally, it is noteworthy that CI children master the skill of word comprehension before the skill of word expression, and that word comprehension may be the basis of word expression.

[Key Words] auditory development; language skill; cochlear implant; children; longitudinal follow-up study

Introduction

A cochlear implant (CI) is an auditory prosthesis which receives acoustic signals through a microphone and transforms these signals into electrical stimulation impulses that pass through a series of electrodes to the cochlea [1]. Cochlear implants have proven to be an effective intervention for individuals with severe to profound sensory hearing loss [2]. Evidence shows positive changes in quality of life for patients with a CI, with improvements in communication, self-esteem, self-care, activity, social interactions, psychological well-being, and cognition [3-6].

For children with a CI, early intervention focuses on auditory, speech and language development, including early prelingual auditory development (EPLAD), and the development of both comprehension and production of speech and language [7]. The most commonly used tool for assessing EPLAD in children with a CI is the Infant-Toddler Meaningful Auditory Integration Scale (IT-MAIS), which is a modification of the Meaningful Auditory Integration Scale (MAIS) and designed to assess the child's spontaneous responses to sound in his/her everyday environment [8]. Currently, several studies reveal that children with a CI have rapid improvement of EPLAD over the initial 6 months or during the first year after switch-on, which further indicate the importance of early intervention [9-12]. For example, by using IT-MAIS, Robbins et al. [9] evaluated the EPLAD of 107 hearing impaired children with CIs at before implantation, 3, 6, and 12 months post implantation. They found a rapid improvement in EPLAD during the first year of device use regardless of age at implantation, although younger children achieved higher scores.

A Chinese version of the IT-MAIS has been developed to assess EPLAD in Mandarin speaking CI children. Results reveal similar patterns, i.e., the EPLAD of Mandarin speaking CI children improved rapidly during the first year [13-17]. For example, a retrospective study by Chen et al. [14] showed that EPLAD improved significantly within the first year after switch-on, and that hearing aid trial and habilitation before implantation were important factors for improvements in EPLAD. Moreover, a study by Zheng et al. [15] indicated that EPLAD followed similar trajectories during the first 12 months after switch-on regardless of culture and language, and showed the importance of early intervention. In addition, Lu and Qin [17] revealed that the EPLAD of CI children exhibited significant improvement, but age at implantation, socioeconomic status and hearing aid trial before implantation were important factors.

To assess language comprehension and expression, the MacArthur-Bates Communicative Development Inventory (CDI) is suggested as a valid tool, which enables assessment of language development in the early stages of children with CIs [18]. For example, Cuda et al. [19] assessed language development in 30 children with congenital severe to profound hearing loss fitted with CIs. The results showed that age at implantation and high maternal educational level had a positive impact on spoken language development. A recent study by Yoshinaga-Itano et al. [20] analyzed the language outcomes of 125 children with CIs and concluded that the early hearing detection and intervention, higher levels of maternal education and early cochlear implant activation had direct, positive impacts on language development outcomes.

There also is a Chinese version of the CDI developed by Tardif et al. [21]. Recent studies on Mandarin speaking CI children demonstrated considerable improvement in language development, but there are some controversial outcomes on benefits of early implantation [17,22]. For example, Li et al. [22] recruited 22 prelingually deaf pediatric unilateral CI users and divided them into two groups according to their age at implantation was more or less than 36 months. They found that early implantation had no significantly positive effect on language development until 24 months post-implantation. However, in the study by Lu and Qin [17], by analyzing the data from 132 prelingually deaf children with unilateral CIs, they reported that early implantation was a consistent predictor of improved language skills in the early stage. Although many previous studies focused on examining auditory and language skills of CI children, there is limited longitudinal study on exploring the trajectories of both auditory and language development of CI children at the same time, particularly for Mandarin speaking CI children where the age of switch-on is younger than 3 years of age.

Because of the nature of childhood auditory and language development, a longitudinal follow-up design appears more appropriate than cross-sectional studies in terms of identifying an accurate developmental trend. Therefore, the aim of this longitudinal follow-up study was to explore the trajectories of early auditory and language developments of Mandarin speaking CI children with age at switch-on younger than 3 years of age.

Materials and Methods

1.1 Participants

A total of 24 children were recruited at the Hearing Center of the Department of Otolaryngology, Head & Neck Surgery of West China Hospital of Sichuan University. All children selected as CI candidates were based on the Chinese Guideline for Cochlear Implant in Children [23]. The criteria for consideration of cochlear implant are assessment of history, ontological examination, audiological evaluation, and imaging examination, together with parental and family consultation. In addition, the candidate should have no significantly positive impacts in terms of speech and language development and responses to environmental sounds after a minimum three month trial with hearing aids.

In regard to the audiological evaluation, the audiological assessments include behavioral audiometric tests if age appropriate, acoustic immittance tests (including tympanometry and acoustic stapedius reflex measurement), Transient Evoked Otoacoustic Emissions (TEOAEs), click and tone-burst elicited Auditory Brainstem Response (ABR) tests, and speech audiometry if age appropriate.

The inclusion criteria used in the present study were:

- 1) Children with a cochlear implant between 1 and 3 years of age;
- 2) Prelingual bilateral sensorineural hearing loss without inner ear malformation or other known developmental abnormalities;

3) There are four outcome evaluation intervals, i.e., baseline, 3, 6, and 12 months after switch-on. Children should have valid assessment results at least three intervals.

Participants were divided into two groups based on the age at switch-on: 1-2 years of age and 2-3 years of age. There were the same number of children in both groups. All children were implanted unilaterally with devices manufactured either by Cochlear, MedEl, or Advanced Bionics. All children in both groups received rehabilitation at public or private centers during the first year after switch-on.

1.2 Measurements

IT-MAIS was used to assess the EPLAD of participants. It was a structured interview questionnaire via parental report, which contained 10 items and assessed three main areas: 1. First 2 items, assessment of vocalization behavior; 2. Next 4 items, evaluation of alertness to sounds; and 3. Final 4 items, assessment of the deriving meaning from sounds. Each item had a potential of 0 (lowest) to 4 (highest) points, the maximum score being 40 points [8]. In this study scores were expressed as percentages to avoid influence of the items to which parents could not respond, i.e., $\text{score} = (\text{actual score}/\text{potential maximum score}) \times 100\%$. In addition, if the number of items which the parents or caregivers could not respond to was more than 2, the score was regarded as invalid and not included in analysis.

For the Chinese version of CDI, Soli et al. [24] developed a simplified short form to meet the requirements of a busy clinic. Li et al. [25] demonstrated that the simplified of Chinese version was suitable for assessment of Mandarin language

development during the first 24 months after cochlear implantation. As a result, the simplified short form of the Mandarin communicative development inventory (SSF-MCDI) was the other assessment tool applied in this study to evaluate the receptive and expressive vocabulary growths of children. It includes two subtests, Words and Gestures (W&G) and Words and Sentences (W&S). Each comprises 50 vocabulary items. W&G was developed to assess receptive and expressive vocabulary growths of normal children between 8 and 16 months of age, and W&S was developed to assess expressive vocabulary growth of normal children between 16 and 30 months of age. Based on above description, W&G was adopted in this study. Scores were expressed as percentages to avoid influence of the items to which parents could not respond, like the criteria of IT-MAIS, only if the number of items that the parents or caregivers could not respond were more than 10, the score would be regarded as invalid and not used in analysis.

1.3 Data analysis

The scores of IT-MAIS and SSF-MCDI (W&G) were analyzed in comparison to normal children, unaided hearing-impaired children, and CI children. A number of statistical analysis tools were applied, such as Chi-square test, *t*-test, One-Way ANOVA and Post Hoc test and Mann-Whitney *U* test. Significance was set at the conventional 0.05 level in each case.

Results

1.1 Demographic data and audiological characteristics

Table 1 shows general demographic information and audiological characteristics for all included children with a CI. A Chi-squared test and Mann-Whitney analysis were performed to analyze the gender and duration of hearing aid using history respectively between the 1-2 years of age group and 2-3 years of age group. No significant differences were found (gender: $p = 1.000$; duration of hearing aid using history: $p = 0.631$).

Table 1. Age and gender distribution for the sample.

Group	ID	Birthday	Gender	Hearing status	Hearing aid fitting history (months)	Implantation Side	Chronological age (Switch-on, months)
1-2 yrs of age	1	2008/05/15	Male	Bilateral profound sensorineural hearing loss (ABR)	16.8	R	20.5
	2	2008/06/13	Male	Bilateral profound sensorineural hearing loss (Behavioral audiometry)	10.1	R	23.0
	3	2010/02/19	Female	Bilateral profound sensorineural hearing loss (ABR)	11.8	R	18.1
	4	2007/6/20	Female	Bilateral profound sensorineural hearing loss (ABR)	10.1	R	22.1
	5	2008/11/10	Male	Bilateral profound sensorineural hearing loss (ABR)	7.6	R	15.1
	6	2008/08/24	Male	Bilateral profound sensorineural hearing loss (Behavioral audiometry)	11.1	R	21.3
	7	2008/12/8	Male	Bilateral profound sensorineural hearing loss (ABR, Behavioral audiometry)	6.8	R	20.6
	8	2008/12/25	Male	Bilateral profound sensorineural hearing loss (ABR, Behavioral audiometry)	5.1	R	20.2

				audiometry)			
	9	2008/08/20	Male	Bilateral profound sensorineural hearing loss (ABR, Behavioral audiometry)	1.7	R	19.7
	10	2008/11/16	Male	Bilateral profound sensorineural hearing loss (ABR)	15.4	R	19.7
	11	2015/01/18	Female	No Records	No Records	R	21.3
	12	2014/10/26	Female	No Records	No Records	R	21.4
2-3 yrs of age	1	2007/12/28	Female	Bilateral profound sensorineural hearing loss (ABR, Behavioral audiometry)	14.6	R	32.6
	2	2008/10/22	Male	Bilateral profound sensorineural hearing loss (Behavioral audiometry)	7.0	R	32.0
	3	2007/10/15	Female	Bilateral profound sensorineural hearing loss (ABR, Behavioral audiometry)	No Records	R	25.0
	4	2007/09/26	Male	No Records	No Records	R	31.3
	5	2007/11/21	Male	Bilateral profound sensorineural hearing loss (ABR, Behavioral audiometry)	6.2	R	30.4
	6	2008/01/03	Female	Bilateral profound sensorineural hearing loss (Behavioral audiometry)	7.1	R	30.0
	7	2008/04/13	Male	Bilateral profound sensorineural hearing loss (ABR, Behavioral audiometry)	6.4	R	28.5

8	2007/12/22	Male	No Records	14.3	R	33.6
9	2008/07/15	Male	Bilateral profound sensorineural hearing loss (ABR)	17.4	R	30.1
10	2007/11/05	Female	Bilateral profound sensorineural hearing loss (ABR)	10.9	R	27.9
11	2007/11/28	Male	Bilateral profound sensorineural hearing loss (ABR)	11.7	R	29.5
12	2008/01/05	Male	Bilateral profound sensorineural hearing loss (ABR)	11.7	R	33.1

1.2 Comparison of EPLADs measured from children with a CI in both 1-2 years of age group and 2-3 years of age group

The mean IT-MAIS scores and standard deviations of children at different follow-up intervals in both groups are shown in Figure 1. The mean scores ranged from 16.3% and 21.2% at baseline to 89.5% and 85.9% at 12 months after switch-on in both groups, respectively. One-Way ANOVA and Post Hoc tests were conducted to compare the IT-MAIS scores at different intervals. Results showed that scores at baseline in both groups were significantly different to those at 3 months, 6 months, and 12 months after switch-on. Scores at 3 months after switch-on in both groups were significantly different to those at 12 months and scores at 3 months in the 2-3 years of age group were significantly different to those at 6 months after switch-on as

well ($p < 0.05$). Arrows demonstrate that scores between different intervals had statistically significant differences.

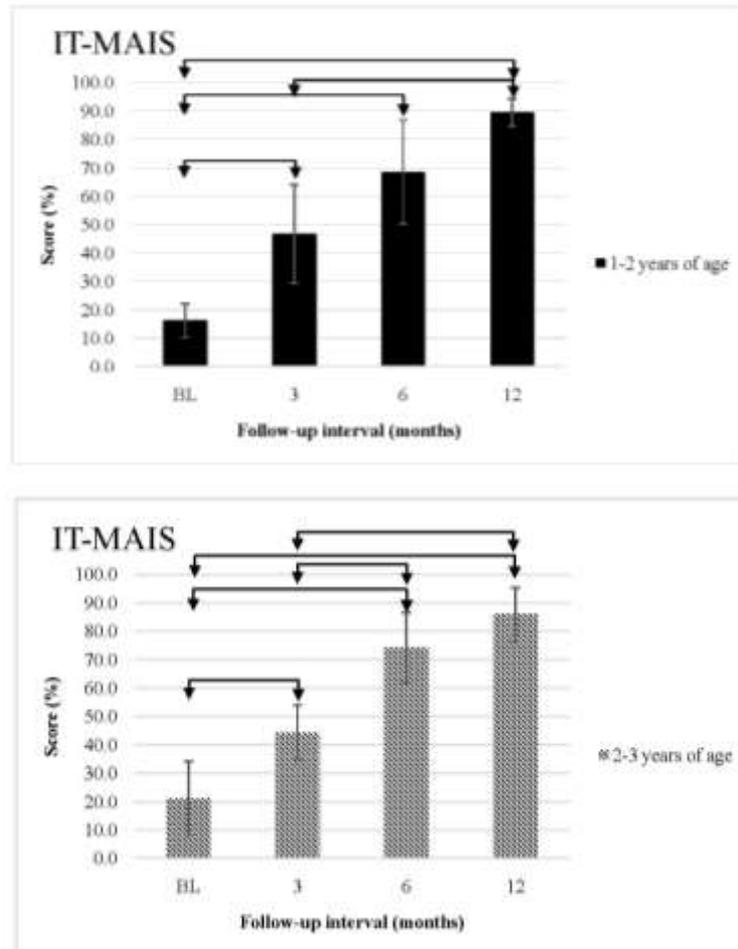


Figure 1. The EPLAD trajectories of children in both age groups. 1a. the mean scores of children in the 1-2 years of age group. 1b. the mean scores of children in the 2-3 years of age group. Arrows represent statistically significant differences ($p < 0.05$).

Scores between the two groups at the same interval were analyzed using Mann-Whitney U tests. The results showed no statistically significant differences between the two groups at the same intervals ($p > 0.05$). Further analysis was conducted to compare the mean scores measured from CI children in both groups to

those obtained from normal and unaided children with different severities of hearing loss. Figure 2 shows that the CI children in both groups had a similar EPLAD trajectory to the EPLAD from children with normal hearing in their first year of age. Although the scores of CI children in both group at baseline were at the average level of unaided peers with profound hearing loss, they had a faster improvement in terms of EPLAD than those unaided children with different severities of hearing loss in the first year of age. In the 1-2 years age group, their scores surpassed the average level of unaided peers with severe, moderate and mild hearing loss at 3 months, 6 months and 12 months after switch-on. Similarly, in the 2-3 years of age group, their scores were close to the average level of unaided peers with severe hearing loss initially, followed by a faster improvement to surpass the average level of unaided peers with moderate hearing loss at 6 months after switch-on. Their improvement in terms of EPLAD was slowed down and eventually close to the average level of unaided peers with mild hearing loss at 12 months after switch-on.

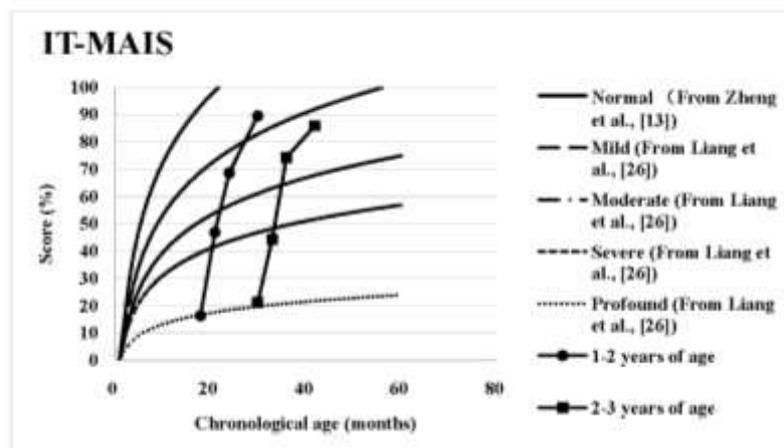


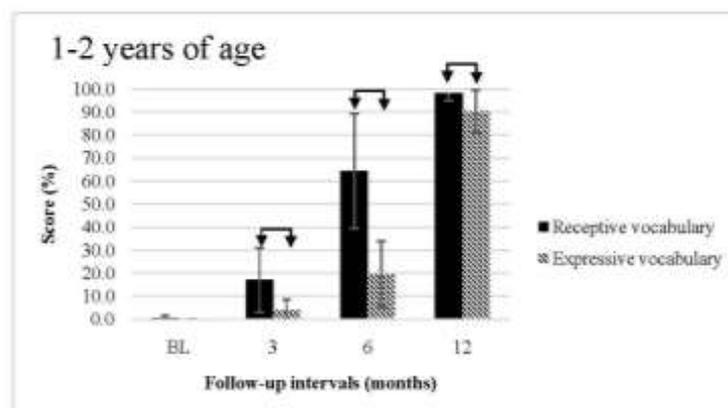
Figure 2. The EPLAD trajectories of normal children, unaided children with different severities of hearing loss from Zheng et al. [13] and Liang et al. [26], and those of children in this study.

1.3 Comparison of word comprehension and expression in children with a CI

The mean SSF-MCDI (W&G) scores showed word comprehension and expression at different follow-up intervals in both groups (Figure 3). Word comprehension and expression scores in the 1-2 years of age group ranged from 0.0 and 0.5% at baseline to 90.2 and 98.3% at 12 months after switch-on (Figure 3a). One-Way ANOVA and Post Hoc tests compared word comprehension and expression scores at different intervals in the 1-2 years of age group. The results showed that baseline word comprehension and expression scores were significantly different to those at 6 months, and 12 months after switch-on. The scores at 3 months after switch-on were significantly different to those at 6 months, and 12 months after switch-on, and the scores at 6 months after switch-on were significantly different to those at 12 months after switch-on ($p < 0.05$). In addition, Mann-Whitney U tests were used to compare scores between word comprehension and expression at each interval. The results showed that word comprehension scores at 3, 6 and 12 months after switch-on were significantly different to word expression ($p < 0.05$).

In the 2-3 years of age group, the mean scores of word comprehension and expression at different follow-up intervals ranged from 0.4 and 1.1% at baseline to 86.0 and 97.2% at 12 months after switch-on respectively (Figure 3b). One-Way

ANOVA and Post Hoc tests compared word comprehension and expression scores at different intervals. The results showed similar effects to those of the 1-2 years of age group, i.e., scores at baseline of word comprehension and expression were significantly different to those at 6 and 12 months after switch-on, scores at 3 months after switch-on were significantly different to those at 6 and 12 months after switch-on, and scores at 6 months after switch-on were significantly different to those at 12 months after switch-on ($p < 0.05$). Mann-Whitney U tests were also used to compare word comprehension and expression scores at each interval. The results showed slight difference to those of the 1-2 years age of group, i.e., the scores of word comprehension at 6 months after switch-on were significant different to word expression at 6 months after switch-on, word comprehension at 12 months after switch-on was significantly different to word expression at 12 months after switch-on ($p < 0.05$).



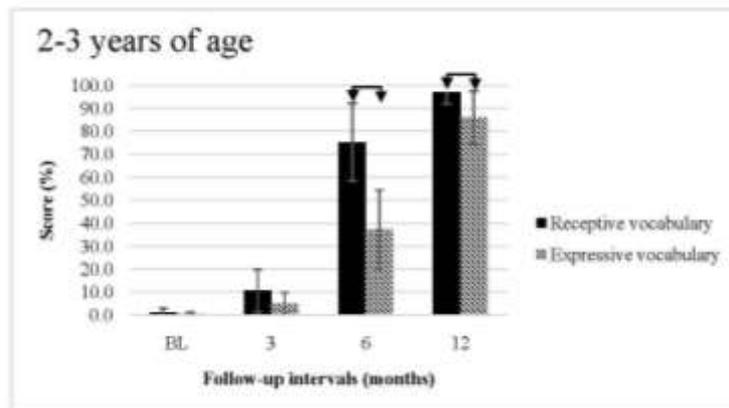


Figure 3. Word comprehension and expression trajectories of CI children in both age groups. 3a. the mean scores of CI children in the 1-2 years of age group. 3b. the mean scores of CI children in the 2-3 years of age group. The arrows show statistically significant differences in scores between word comprehension and expression ($p < 0.05$).

Further analysis was conducted to compare CI children to children with normal hearing and development (Figures 4a and 4b). Figure 4a shows word comprehension trajectories for both groups to be very similar. Figure 4b shows word expression trajectories for normal and CI children in both groups, which indicate the development of vocabulary expression in both CI groups to be slightly slower than that of normal children, although they had similar velocities.

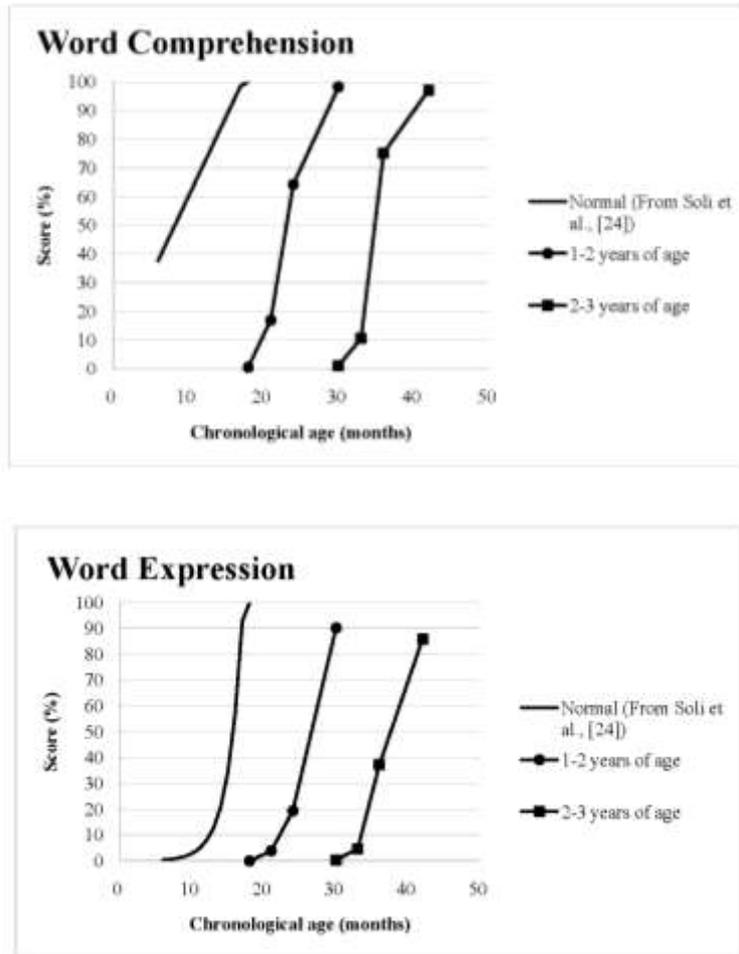


Figure 4. The word comprehension and expression trajectories of normal children from Soli et al. [24] and those of CI children in this study. 4a: the mean scores of word comprehension from normal children and CI children in this study. 4b: the mean scores of word expression from normal children and CI children in this study.

Discussion

In this longitudinal follow-up study, IT-MAIS and the W&G subtest of SSF-MCDI were used to evaluate the EPLAD, early development of word comprehension and expression in young children after receiving a cochlear implant. Although many studies have focused on the auditory and language skills of CI

children, there has been limited longitudinal study on the trajectories of auditory and language skills of CI children in the early stage when compared to those of normal and unaided children. The present results show that the auditory abilities and language skills of CI children in both age groups during the first year after switch-on significantly improved in terms of IT-MAIS and SSF-MCDI (W&G) scores with follow-up time, from baseline to 12 months after switch-on. They had similar development trajectories, which were comparable to those of normal hearing children in the first year of age. These results are in keeping with the findings in some previous studies [15,16]. For example, Zheng et al. [15] indicated that EPLAD trajectories exhibited a consistent pattern as age at implantation increased, but there were only 4 children who received cochlear implantations between 1-2 years of age. Although a cross-sectional study by Li et al. [25] demonstrated that vocabulary growth rates during the first 12 months after implantation were similar to those for normally hearing children younger than 16 months of age, the longitudinal data from this study provide more accurate developmental patterns of auditory and language abilities in profound SNHL children with a CI, indicating similar velocities of auditory improvement and language development when comparing to normal hearing children. In addition, there seems to be no difference in terms of patterns and velocities of auditory improvement and language development between the 1-2 years of age group and 2-3 years of age group. However, it is noteworthy that auditory development of children in the 2-3 years of age group trended to a lower level than that in 1-2 years of

age group. A further longer term follow-up needs to explore the difference of the EPLAD trajectories between these two groups.

Kral and Sharma [27] reviewed evidence for the existence of a sensitive period for successful cochlear implantation and found the optimal time to be within the first 3.5-4.0 years of life. They also demonstrated that early implantation within the brief sensitive period would allow more adequate cortical maturation and maximum plasticity of central auditory pathways to sound stimulation. Significant outcomes derived from early intervention were also demonstrated by comparing the auditory ability of CI children and unaided children with different levels of hearing loss in the present study. This is being one of the strengths of the longitudinal follow-up study. Evidence shows that longitudinal study provides valid milestones in terms of childhood development changes, such as memory-related linguistic skills [28].

Therefore, the current study provides more appropriate and consistent evidence than cross-sectional study to indicate the trajectories of profound SNHL children with early implantation. It provides evidence for parents or caregivers of profound SNHL children of the progress and significant outcomes to be derived from early implantation, gained from a comparison of the auditory skills of children during the first year after early cochlear implantation (before 3 years of age) to the average level of unaided peers with various degree of hearing loss.

It is noteworthy that the average scores of word comprehension in both groups were significantly higher than those of word expression in the majority of follow-up

intervals. However, the gap between word comprehension ability and expression ability reduced as the follow-up time increased. These results are consistent with the findings demonstrated in the study of Lu and Qin [17], which also showed that receptive vocabulary scores were significantly higher than expressive vocabulary scores in children with CIs in the first years after switch-on. The current results imply that CI children are able to master the skill of word comprehension earlier than mastering the skill of word expression, and that word comprehension may be as a result of mastering word expression.

Similar results were also found in normal hearing children. For example, Bornstein and Hendricks [29] found that language comprehension slightly exceeded production and correlations between comprehension and production were positive and significant. The possible rationale underlying this phenomenon is that comprehension is a process of sensory integration, while perception-for-production requires more sensory detail than perception-for-understanding [30].

Conclusions

Children with cochlear implantation at 1 and 2 years of age have similar trajectories of EPLAD, word comprehension and expression at the first year after switch-on, which are similar to normally hearing children. Moreover, the present study also indicates that early implantation is an important factor for the early auditory development when comparing EPLAD results between CI children and unaided peers with different hearing loss. It is noteworthy that CI children master the skill of word

comprehension before the skill of word expression, and word comprehension may be the basis of word expression.

Human subjects

Research protocol for the current study satisfied the appropriate ethics review policies at West China Hospital of Sichuan University.

Declaration of competing interest: The authors report no conflicts of interest.

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