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Early intervention and assessment of speech and language development in young children with cochlear implants

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ABSTRACT

Objective: Age is one of the most important determinants of the benefit achieved in the cochlear implantation of pre-lingually deafened children. Earlier age at implantation increases the exposure of children with a hearing impairment to auditory stimuli. Earlier auditory stimulation enables children to better understand spoken language and to use spoken language themselves. Furthermore, there appears to be critical period under 2 years of age during which access to spoken language is essential in order for language development to proceed appropriately. The present study aimed to assess the impact of cochlear implantation under 2 years of age on subsequent speech and language development.

Methods: 28 children implanted with a cochlear implant prior to 2 years of age were included in this study and the effects of age at implantation were determined using a reception of grammar test, active vocabulary test and speech development test. Demographic features were described using descriptive statistics and data were compared to the normative values (*T*-values) of their hearing peers by *t*-test or Mann–Whitney *U*-test.

Results: The present data indicates that overall children with a hearing impairment implanted at less than 2 years of age perform as well as or better than their hearing peers in speech and grammar development. Word Comprehension was significantly greater in children with a cochlear implant compared to their normative peers (p = 0.003), whereas Phonological Working Memory for Nonsense Words was poorer (p = 0.031). An effect of age on grammatical and speech development could be found for younger implanted children (<12 months), who reached higher scores than children implanted after 12 months of age.

Conclusions: The data suggests that early hearing loss intervention via cochlear implantation in children benefits the speech and language development of children. A potential sensitive period exists for implantation before 12 months of age. These outcomes support the recent trend toward early cochlear implantation in pre-lingually deaf children.

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1. Introduction

Auditory deprivation caused by deafness leads to severe impairment of speech and language development in children [1]. A lack of sensory activity leading to poorer neuroplasticity is at fault [2,3]. However, the effect of sensory deprivation can be reversed by the provision of sensory stimulation, such as delivered by cochlear implantation in children with a hearing loss [2].

In the past, the majority of children with profound hearing loss were receiving cochlear implants at 2 years of age or older [4]. This narrowed greatly the window of opportunity for language learning in these children, determined to be from birth to approximately 7 years of age [5]. Improvements in neonatal hearing screening have

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greatly facilitated the early diagnosis of hearing loss in infants [6,7]. Accordingly, this has led to earlier clinical intervention and treatment of children with hearing loss [8]. The clinical evolution in hearing loss diagnosis and intervention taken together with advances in the development of cochlear implants has led to a resultant increase in the number of young patients being implanted for the treatment of profound hearing loss [3].

Cochlear implantation has shown that children with pre-lingual deafness benefit substantially from early intervention in terms of speech perception and linguistic ability [9–13]. Several studies illustrate that cochlear implantation of pre-lingual deaf children, which gives greater access to spoken language via hearing, promotes an increase in auditory skills, speech understanding and oral linguistic development [9,14,15]. Furthermore, the evidence indicates that the negative effects of auditory deprivation through hearing loss on language acquisition can be reversed by cochlear implantation [2]. This has been demonstrated for speech perception and speech production [10,16].

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An important determinant of the benefit achieved in spoken language through pre-lingual cochlear implantation is age at implantation [17]. The younger a child is exposed to auditory stimuli the more likely they appear to be able to understand spoken language and to use spoken language themselves [18-20]. Tomblin et al. demonstrated age alone at stimulation accounts for over 14% of the variance of the individual differences in expressive language growth rates and suggests that delays in implantation result in an ever-increasing gap between implant candidates and their hearing peers in terms of language status [21]. Likewise, investigation of language development in young children with cochlear implants indicates age at implantation is a significant determinant in auditory and speech development [8,22,23]. Preliminary studies indicate that early cochlear implanted children develop language skills more efficiently than their non-implanted peers and that their development is comparable to that of hearing children [5,14]. Thus, there is a current trend to decrease the age at implantation. By providing the child with access to spoken language at an earlier age it is believed that rapid learning and language development is facilitated [21]. Earlier intervention attempts to limit the gap between the chronological age and the age appropriate development of language skills in deaf children [5,14].

Further to the perceived benefits of early implantation, numerous studies indicate that there is a critical period during which the access to spoken language and thus auditory stimulation is essential in order for language development to proceed appropriately [2,5,24].

It is thought that the year old child has a neural mechanism engaged from as early as the 6th month of fetal life that can separate out sound patterns which are auditorily relevant in utero to lay out the foundation of language [25]. Research indicates that deprivation of hearing interferes with this process [25]. Language can broadly be divided into 3 fundamental elements or language processing classes: (1) phonology; (2) semantics; and (3) syntax, which encompass part of the structural components of language. Phonology refers to the physical structure of the sound in an auditory-based language; semantic is the meaning assigned to specific stimuli and syntax is the organizational structure of stimuli which produces complex meanings [25]. It is established that complex language can only develop once phonology has been established during the first 2 years of life [25]. It is possible that children implanted later than 2 years of age are 'held-back' in this respect. Indeed, it has been shown that children who have been deprived of appropriate sensory stimulation do not develop language successfully, whereas children who have acquired severe deafness after the phonological period demonstrate superior semantic and syntactic development when compared to children deafened throughout the whole phonological period [26]. However, unlike phonological development the potential for semantic and syntactic development persists [25]. Apparently, the window of semantic development can last up to the late teenage years and the sensitive period for semantic development is in the first 4 years [25].

Specific time periods relevant to the success of cochlear implantation have yet to be identified [2,8,21] and it has been suggested that language acquisition may not be a single sensitive or critical period [2]. Rather several sensitive periods exist specific to various aspects of language development [25,27,28]. However, it is apparent that to determine the validity of cochlear implantation, outcomes in young children need several years of follow-up for the effects to become evident [29].

This study aimed to assess the impact of cochlear implantation under 2 years of age on speech and language development in children with a hearing impairment. The children were followed-up for an extended period, throughout the course of their normal

rehabilitation routine, to allow for accurate interpretation of the outcomes. We hypothesized that an implantation age of less than 2 years would promote language development in deaf children. Retrospective data of early cochlear implanted children, evaluated in terms of grammatical ability and speech development was used to determine the benefits of early implantation in children with a hearing impairment compared to their hearing peers.

2. Materials and methods

2.1. Patients

This was a retrospective analyses of data obtained from 28 children implanted with a cochlear implant prior to 2 years of age and attending regular therapy at the Cochlea-Implantat-Centrum Süd, Würzburg, Germany. The mean age at implantation was 14.3 ± 5.5 months. The mean age at follow-up ranged from 33.3 ± 7.3 to 72.6 ± 16.3 months. The mean age at implantation is featured in Table 1.

Written informed consent was collected from parents or caregivers, on behalf of children for their participation and evaluation in the habilitation program, and to the use of their data.

2.2. Experimental design

2.2.1. Grammatical development

TROG-D is a test adapted to German to determine the reception of grammar in children aged 3–11 years [30]. The TROG-D assesses grammatical comprehension by measuring the understanding of 18 different sentence constructions given 4 times each using different test stimuli.

The German speaking AWST-R [31], "Aktiver Wortschatztest" or active vocabulary test, is a test for children between 3–5½ years old. It is a test of vocabulary expression in which participants verbalize one-word responses to pictures shown. The AWST-R contains graphical depictions of 51 nouns and 24 verbs. The quantitative analysis of word expression is appraised by a qualitative score [31].

2.2.2. Speech development

To investigate the speech development of children the "Sprachentwicklungstest für Kinder" (SETK) was used [32]. The SETK is an age specific speech development test designed for children aged 2-5 years. The SETK-2 test, specifically for 2 year old children, is composed of 4 sub-tests (Word Comprehension, Sentence Comprehension, Word Production and Sentence Production). The SETK-3-5 developed for the evaluation of the stage of speech development and correlation with auditive memory in children aged 3-5 years comprises 4 sub-tests for children up to 4 years and 5 sub-tests for children 4–5 years old. Children up to 4 years are tested in Sentence Comprehension, the Encoding of Semantic Relations, Morphological Syntax and Phonological Working Memory for Nonsense Words. Similarly, children aged 4–5 are tested in Sentence Comprehension, Morphological Syntax and Phonological Working Memory for Nonsense Words, however in addition sentence and word Memory Span.

Table 1Mean time in years of cochlear implant use.

Mean (years) ± SD
4.45 ± 1.10
$\boldsymbol{3.38 \pm 0.62}$
1.77 ± 0.43
2.65 ± 0.54
$\boldsymbol{3.92 \pm 0.49}$

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2.3. Statistical analyses

Demographic features and basic characteristics of the test materials are shown as mean, standard deviation (SD) and median. The interpretation of referenced-test results was performed via comparison to a normative group of children. Normative T-values and percentage range for a given test were determined by the test developer. To detect differences between the results of children with CIs under two years of age and mean normative data of hearing children, one-sample *t*-tests were performed. The *T*-mean values of the CI children were compared to the mean normative Tvalue of hearing children. To look at the effects of age at implantation on grammatical and speech development more closely the children were split up into three groups: implanted before 12 months, between 12-18 months, and 18-24 months of age. The difference between the three age groups was tested using Mann-Whitney *U*-test. Individual and group results of TROG-D, AWST-R, SETK-2 'Word Comprehension', SETK-3-5 'Sentence Comprehension' and SETK-4-5 'Sentence Comprehension' are depicted graphically. Missing data were not replaced, but treated as "missing" values. A p-value of <0.05 was determined as statistically significant. IBM SPSS Statistics 19 was used for all analyses. Graphs were created in Microsoft Office Excel 2010.

3. Results

Overall the grammatical and speech development of children implanted early (<2 years) was within the same range as hearing children (Fig. 1), when compared in *T*-values. However, it is evident that a significant degree of variation exists between children, both in the range of hearing and in the performance of the CI group. Grammar development scores on TROG-D determined that 10 out of 19 children were within the normative range of their peers (Fig. 2; Table 2). Children implanted before 12 months showed a trend toward better grammar development than hearing children;

with 6 children scoring better than the average hearing child. In contrast, all children except one, performing below the normative range of hearing children in grammatical development were implanted after 12 months of age.

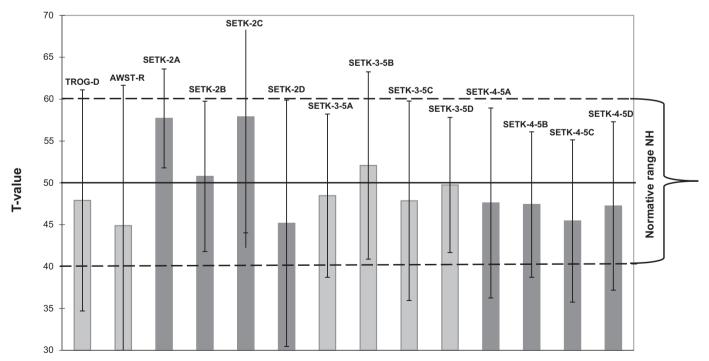
The AWST-R values of 3 out of 9 cochlear implant recipients were within the normative range of hearing children (Fig. 3; Table 2). All the cochlear implanted children within the normative range of hearing children were implanted before 12 months of age. Two CI children implanted after 18 months reached even higher values than the average. Four CI children were below the normative range of their hearing peers. The child showing the poorest performance of this group had been implanted between 12–18 months of age.

Results of one-sample t-test showed a significant mean difference between CI children and the normative group for SETK-2 in 'Word Comprehension' (p = 0.003). The CI group performed better than the normative group in this test. The mean T-value of the subscale SETK-4–5 'Phonological Working Memory for Nonsense Words' was also significantly different to the mean T-value of the normative group (p = 0.031), but in this instance the CI group scored lower values. All other subscales were not significantly different from the mean T-value of the normative group. However, for SETK-2 'Sentence Comprehension', SETK-2 'Word Production', and SETK-3–5 'Encoding Semantic Relations' the CI group scored higher values than their hearing peers (Table 3; Fig. 1).

Speech development data for children aged 2–5 years are shown in Table 3. The mean age at SETK-2 (n = 10) was 33.3 \pm 7.3 months. Speech development in children tested with SETK-2 determined that all 10 children were within or above the normative range of the 'Word Comprehension' of hearing children (Fig. 4). Six of the cochlear implanted children performed better than their hearing peers in terms of Word Comprehension. Of the children performing better 4 were implanted before 12 months and the remaining 2 after 18 months of age.

The mean age of SETK-3-5 (used for children aged 3-4 years) (n = 15) was 43.7 ± 3.0 months. The 'Sentence Comprehension' of 10

TROG-D, AWST-R, SETK-2, -3-5, and -4-5



Note: Error bars are representing standard deviations.

Fig. 1. Mean *T*-values (±1 SD) of children cochlear implanted under two years of age compared to the *T*-norm of normal hearing children for reception of grammar (TROG-D), active vocabulary test (AWST-R), and speech development tests at 2 (SETK-2), 3 (SETK-3-5) and 4-5 (SETK-4-5) years of age.

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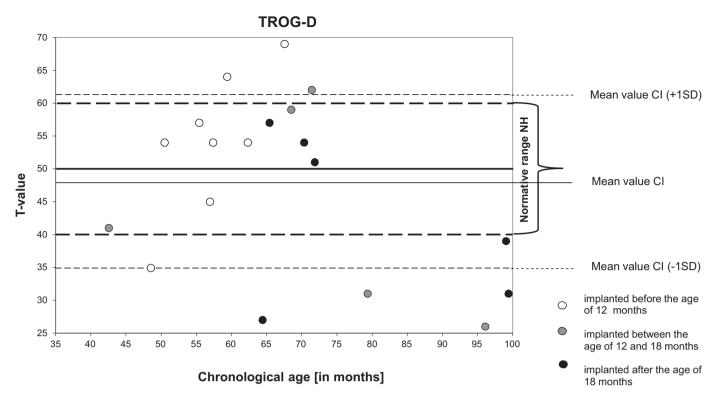


Fig. 2. Mean *T*-value (±1 SD) and individual *T*-values of reception of grammar (TROG-D) in children cochlear implanted under two years of age compared to the *T*-norm of normal hearing children. Cochlear implanted children are sub-divided according to age at implantation: ≤12 months (white marks), 12–18 months (gray marks), ≥18 months (black mark). The normative range of hearing children is denoted within brackets.

of these children was within the same range as their hearing peers (Fig. 5; Table 3). One child implanted before the age of 12 months scored above the normative range. Of 4 children, below normative values, 3 were implanted between 12 and 18 months of age and one child was implanted before the age of 12 months; however the Sentence Comprehension of this child neared the normative range of hearing children.

The mean age at SETK-4–5 (n = 21) was 58.3 \pm 7.0 months. Of 21 patients tested 13 fell within the normative range of hearing children (Fig. 6; Table 3). Seven out of a total of 11 children implanted under the age of 12 months were within the normative range, and 3 scored above the normative range. The remaining implanted children within the normative range were 3 between the age of 12 and 18 months and 3 implanted after 18 months of age. Of the 5 children not scoring within the normative range 1 was implanted before 12 months while the remaining were all implanted after 12 months.

An effect of age on grammatical and speech development could be found for younger implanted children (<12 months) who reached higher scores than children implanted after 12 months of age. Except for the TROG-D, children implanted between 18 and 24 months reached higher scores than children implanted between 12

Table 2 Outcomes (mean \pm standard deviation (SD), and median *T*-values), numbers (*n*) and *p*-values of one-sample *T*-tests comparing *T*-values of children cochlear implanted under two years of age to the mean normative *T*-value of hearing children (test value = 50) of: (A) test for the reception of grammar (TROG-D) and; (B) active vocabulary test (AWST-R), in cochlear implanted children under two years of age.

	N	$Mean \pm SD$	Median	Mean difference	p-Value (2-sided)
Reception of grammar: (A) TROG-D	19	47.9 ± 13.2	54	-2.11	0.496
Active vocabulary: (B) AWST-R	9	44.9 ± 16.8	45	-5.11	0.387

and 18 months. The comparison was statistically significant between the CI group implanted younger than 12 months and those implanted between 12 months and 18 months for SETK-3–5 'Sentence Comprehension' (Mann–Whitney U-test: p = 0.076). No significant difference on scoring could be found for the other age comparisons (p = 0.130–1.000).

4. Discussion

Cochlear implantation in children below 2 years of chronological age serves to provide children with a hearing impairment access to auditory stimuli through hearing [17]. Auditory stimulation aims to promote an increase in speech understanding and oral linguistic development [9,14,15].

The early diagnosis and intervention of hearing loss has led to a younger age at cochlear implantation in children with severe-toprofound hearing loss. The follow-up of early implanted children suggests that age at implantation determines expressive language development, auditory skills, speech understanding and oral linguistic development [9,14,15,21]. Consequently, young age at implantation is deemed an important determinant of outcome. The data presented herein suggest that cumulatively children implanted under 2 years of chronological age perform as well as their hearing peers in terms of speech and grammar development. Furthermore, data suggest that there are several benefits to cochlear implantation in suitable recipients under the age of 2. Word Comprehension is better in children implanted with a CI under 2 years of age and there is a tendency toward better Sentence Comprehension, word production and the encoding of Semantic Relations in these children compared to the average hearing child as illustrated by SETK in this study. More specifically, the present data illustrates that, in general, children receiving a cochlear implant under the age of 1 year perform as well as or better than their hearing peers at follow-up in terms of

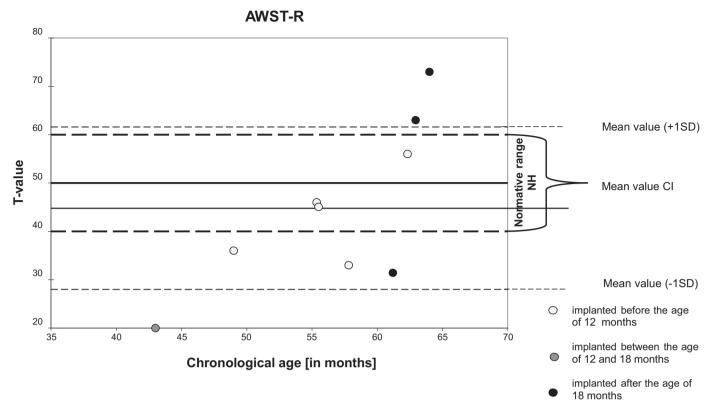


Fig. 3. Mean *T*-value (±1 SD) and individual *T*-values of active vocabulary (AWST-R) in children cochlear implanted under two years of age compared to the *T*-norm of normal hearing children. Cochlear implanted children are sub-divided according to age at implantation: ≤12 months (white marks), 12–18 months (gray marks), ≥18 months (black marks). The normative range of hearing children is denoted within brackets.

grammatical and speech development. In contrast, children implanted after 12 months of age in the present study showed a comparative tendency toward poorer performance.

TROG-D scores highlighted a trend toward better grammar development in children implanted before 12 months. Similarly, Coletti et al. demonstrated that grammar development scores on the TROG after five years of activation showed that no child of the 12–23 or 24–35 months implanted group were above the 75th percentile, whereas 77% of children of the 2–11 month group were above the 75th percentile of their hearing peers [33]. Furthermore, the proportion of children falling below the normative range in the reception of grammar as determined by TROG-D was greater in children implanted after 12 months compared to those implanted before. At follow-up, of the children who fell below the normal range of hearing children in SETK-3–5, 75% were implanted after 12 months and 80% of SETK-4–5 children falling below the norm were implanted within this period.

This is consistent with the hypothesis that a sensitive or critical period exists for the development of speech, in which auditory stimulation is essential [34]. Consistently, Dettman et al. showed that language comprehension of infants implanted under 1 year is better [3]. The average growth for language development in children implanted under 1 year was better than their older implanted peers and implanted children demonstrated language comprehension and expression comparable to hearing children [3]. Likewise, Schauwers et al. demonstrates that the auditory performance of children implanted under 1 year follows normal development and that only implantation under 1 year sustains the babbling onset of infants within the normal range [35]. Babbling, a state of language acquisition in infants is an important prognostic indicator for speech and language development in children [36].

Consequently, it appears that addressing hearing loss in the sensitive period would benefit the long-term development of the child [21], thus limiting adverse consequences in terms of speech

Outcomes (mean \pm standard deviation (SD), median *T*-values), numbers (*n*) and *p*-values of one-sample *T*-tests comparing *T*-values of children cochlear implanted under two years of age to the mean normative *T*-value of hearing children (test value = 50) the speech development tests SETK-2, SETK-3 and SETK-4–5.

Test	N	$Mean \pm SD$	Mean difference	p-Value (2-sided)
SETK-2 (A) Word Comprehension	10	57.7 ± 5.9	7.70	0.003*
SETK-2 (B) Sentence Comprehension	9	$\textbf{50.8} \pm \textbf{9.0}$	0.78	0.802
SETK-2 (C) Word Production	9	$\textbf{57.9} \pm \textbf{13.9}$	7.89	0.126
SETK-2 (D) Sentence Production	6	$\textbf{45.2} \pm \textbf{14.7}$	-4.83	0.458
SETK-3-5 (A) Sentence Comprehension	15	48.5 ± 9.8	-1.53	0.552
SETK-3-5 (B) Encoding Semantic Relations	14	$\textbf{52.1} \pm \textbf{11.2}$	2.07	0.500
SETK-3-5 (C) Morphological Syntax	14	47.9 ± 11.9	-2.14	0.513
SETK-3-5 (D) Phonological Working Memory	12	$\textbf{49.8} \pm \textbf{8.1}$	-0.25	0.916
SETK-4–5 (A) Sentence Comprehension	21	47.6 ± 11.3	-3.00	0.237
SETK-4-5 (B) Morphological Syntax	22	47.4 ± 8.7	-2.86	0.153
SETK-4-5 (C) Phonological Working Memory	16	$\textbf{45.4} \pm \textbf{9.7}$	-5.60	0.031*
SETK-4-5 (D) Memory Span (word)	23	$\textbf{47.2} \pm \textbf{10.1}$	-2.85	0.232

Significant difference between CI children and hearing children.

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SETK-2: Word Comprehension

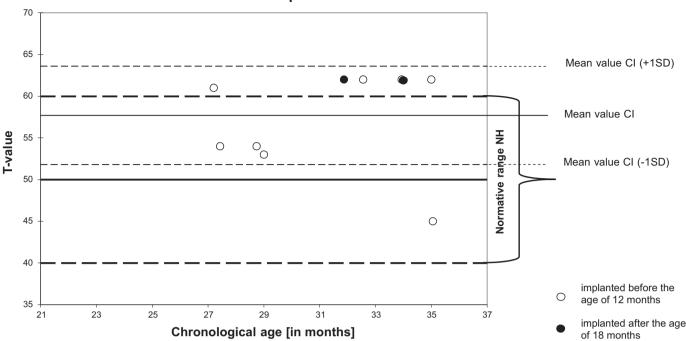


Fig. 4. Mean T-value (± 1 SD) and individual T-values of 'Word Comprehension' testing at 2 years of age (SETK-2), in children cochlear implanted under two years of age compared to the T-norm of normal hearing children. Cochlear implanted children are sub-divided according to age at implantation: ≤ 12 months (white marks), 12-18 months (gray marks), ≥ 18 months (black marks). The normative range of hearing children is denoted within brackets.

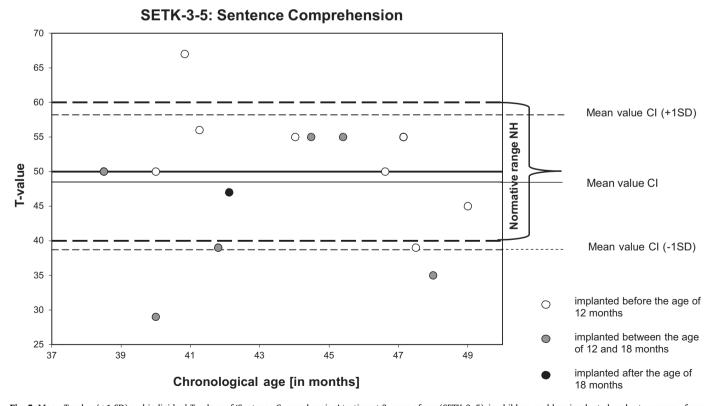


Fig. 5. Mean *T*-value (±1 SD) and individual *T*-values of 'Sentence Comprehension' testing at 3 years of age (SETK-3–5), in children cochlear implanted under two years of age compared to the *T*-norm of normal hearing children. Cochlear implanted children are sub-divided according to age at implantation: ≤12 months (white marks), 12–18 months (gray marks), ≥18 months (black mark). The normative range of hearing children is denoted within brackets.

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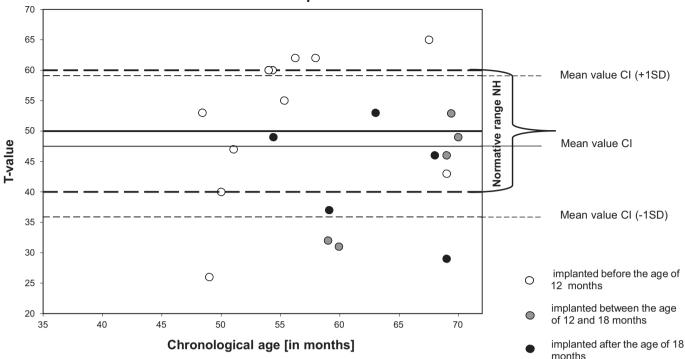


Fig. 6. Mean *T*-value (±1 SD) and individual *T*-values of 'Sentence Comprehension' testing at 4 to 5 years of age (SETK-4–5), in children cochlear implanted under two years of age compared to the *T*-norm of normal hearing children. Cochlear implanted children are sub-divided according to age at implantation: ≤12 months (white marks), 12–18 months (gray marks), ≥18 months (black mark). The normative range of hearing children is denoted within brackets.

and language development that may stigmatize children with hearing loss permanently [1]. The effects of early intervention are most evident in Word Comprehension as determined by SETK-2, which is significantly greater than that of hearing children. We suggest that this effect is illustrative of the proportion of children in this group implanted below 12 months which exceeds the proportion of children implanted in all other SETK-2 comparisons. Thus, the present data indicates that the critical period of auditory pathway development during which pre-lingually deaf children should be implanted may end much earlier than previously reported in the literature.

It is possible that children implanted later than 12 months of age are 'held-back' due to the deprivation of early auditory stimulation. Furthermore, it is established that complex language can only develop once phonology has been established during the first 2 years of life [25]. This suggests that delayed implantation would limit phonological development in the first 2 years of life. Indicating that improvements in language development after cochlear implantation at less than 2 years of age are largely due to phonology. However, unexpectedly the present study illustrates that the average Phonological Working Memory of nonsense words of the children in the present study was poorer than the average of the hearing children at follow-up between 4 and 5 years of age. Despite this the children are still within the normative range of their hearing peers. To determine the significance of phonological learning on language development long-term follow-up of early implanted children before 2 years of age and children implanted after 2 years is needed.

It is important to note that despite indications that there could be a sensitive period within the 1st year of life in this study and abundant evidence that age at implantation is a strong predictor of speech and language outcome, wide variation in individual outcomes exists [8,18,22]. This is a common problem and is likely to occur due to patient specific differences [37], independent of

their hearing loss. Thus, the composition of the normative group may be relevant [38]. Furthermore, a child's capacity to learn language is determined greatly by their intellectual ability and certain characteristics related to a child's behavior and character may also be involved [39]. Another confounding factor in several retrospective studies is the lack of robust and reliable outcome measures of monitoring children with a CI [8].

In the present study 28 children reported reliable outcome measures after receiving a cochlear implant before 2 years of age, even though several more were enrolled in the initial study and implanted. Of interest the root cause of failure to collect robust and reliable data often lies with the age at the time of testing, differences in ability, problems with articulation (important for some subtests), additional needs [40], unwillingness of children to participate/concentrate, or multilingual ability (authors own unpublished observations). The net effect of unreliability in outcome measures is a tendency to select patients, which quite often introduces bias to the analyses. In support of the present study, patients have not been selected and children have been assessed for up to 5 years of age after cochlear implantation. It is established that the outcomes of cochlear implantation in young children needs several years to become evident [29].

In conclusion, the data presented herein indicates that children implanted before 2 years of age perform overall better post-implantation. The data illustrating the effects after 5 years of age after cochlear implantation is a strong indicator of the effects of early auditory stimulation on speech and grammar development. Moreover, this study underscores the concept of a sensitive period in which auditory stimulation is critical for neuroplasticity in infants. The generally better performance of children implanted with a CI before 1 year of age, as determined by SETK, is supportive of the recent trend toward cochlear implantation under 1 year of age in profoundly deaf children.

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