

Cochlear Implants or Hearing Aids: Speech Perception, Language, and Executive Function Outcomes

Merle Sanne Boerrigter,^{1,2} Anneke. M. Vermeulen,³ Michel Ruben Benard,³ Hans. J. E. van Dijk,³ Henri A. M. Marres,^{1,2} Emmanuel A. M. Mylanus,^{1,2} and Margreet C. Langereis^{3,4}

Objectives: We aimed to determine whether children with severe hearing loss (HL) who use hearing aids (HAs) may experience added value in the perception of speech, language development, and executive function (EF) compared to children who are hard of hearing (HH) or children who are deaf and who use cochlear implants (CIs) and would benefit from CIs over HAs. The results contribute to the ongoing debate concerning CI criteria. We addressed the following research question to achieve this aim: Do children who are HH or deaf with CIs perform better than children with severe HL with HAs with respect to auditory speech perception, and receptive vocabulary and/or EF?

Design: We compared two groups of children with severe HL, profound HL or deafness, with CIs or HAs, matched for gender, test age (range, 8 to 15 years), socioeconomic status, and nonverbal intelligence quotient. Forty-three children had CIs (pure-tone average at 2000 and 4000 Hz >85 dB HL), and 27 children had HAs (mean pure-tone average: 69 dB HL). We measured speech perception at the conversational level (65 dB SPL) and the soft speech perception level (45 dB SPL). We established receptive vocabulary using the Peabody Picture Vocabulary Test-III-NL. We tested EF using the Delis Kaplan Executive Function System battery and the Dutch Rey Auditory Verbal Learning Test. We employed the Mann-Whitney U test to compare data between the CI and HA groups. We used Chi-square goodness of fit tests to contrast the CI and HA group distributions with the norm data of children who are typically developing (TD). We harnessed Kendall's Tau-b to investigate relationships between the study variables.

Results: Both groups of children, with CIs and HAs, obtained ceiling scores for perception of speech on a conversational level. However, the HA group exhibited significantly lower perception on a soft speech level scores (68 %) than the CI group (87%). No difference was present between the receptive vocabulary distributions of the CI and HA groups. The median receptive vocabulary standard scores for both groups were well within the normal range (CI group: 93; HA group: 96). In addition, we did not find any difference in EF between the CI and HA groups. For planning and verbal memory, the distributions of observed scores for children with CIs were different from the expected distributions of children who are TD. In both groups, a large proportion of children obtained below-average scores for planning (CI: 44%; HA: 33%) and for long-term verbal memory (CI: 44%; HA: 35%). In the HA group, perception at a soft speech level was associated with receptive vocabulary and planning. In the CI group, we did not find any associations.

Conclusions: Both groups of children with severe and profound HL with HAs exhibit less favorable auditory perception on the soft speech level, but not at a conversational level, compared to children who are HH or

deaf with CIs. Both groups, children with CIs and HAs, only exhibit more problems in planning and verbal memory than the norm groups of children who are TD. The results indicate that to obtain age-appropriate levels of receptive vocabulary and EF, the perception at the soft speech level is a necessary but not sufficient prerequisite.

Key words: Cochlear implants, Executive function, Hearing aids, Indication criteria, Language.

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INTRODUCTION

Even before birth, during the development of the auditory cortex, auditory stimulation facilitates spoken language as well as social, emotional, and cognitive development (Sharma et al. 2009; Moon et al. 2013; Kral et al. 2016). Therefore, intervention should commence as early as possible to enable access to sound and the perception of speech to enhance speech and language development (Sharma et al. 2009; Kral et al. 2016). For this reason, neonatal hearing screening programs have been devised. In the Netherlands, a program has been available to all Dutch newborns since 2006 and hearing aids (HAs) are indicated for children with mild to severe hearing loss (HL), while cochlear implants (CIs) are administered to children who are hard of hearing (HH) or deaf, with a hearing threshold of 85 dB HL or higher (CI-ON 2012). For the present study, we used the hearing level classification of the World Health Organization (WHO 2020) as a reference. In babies who are HH or deaf, parents are given the option of early intervention for language/interaction and hearing via HAs or CIs (Boons et al. 2013). Early CI fitting, before the age of 12 months, provides children who are HH or deaf the ability to develop sufficient speech perception and recognition to achieve nearly age-appropriate spoken language development (Colletti et al. 2011; Leigh et al. 2013; Dettman et al. 2016). Currently, auditory access to spoken language in acoustic environments has improved due to early CI in children who are HH or deaf. This enables these children to acquire near age-appropriate language development. This level is comparable to that of children with moderate HL with HAs and, on average, is even better than that of children with severe HL with HAs (Yang et al. 2012; Leigh et al. 2016; Nekes 2016).

Children with severe HL are at risk for developmental language problems (Wake et al. 2005; Tomblin et al. 2015). Iwasaki et al. (2012) reported improved language outcomes in children who are HH or deaf with CIs compared to children with HAs. Several studies report delayed receptive and expressive language development in children with mild to severe HL with HAs (Borg et al. 2007; Koehlinger et al. 2013; Cupples et al. 2018). These language problems persist in adolescents with HL (Delage & Tuller 2007). Language and communication problems in children who are HH or deaf negatively interfere

¹Department of Otorhinolaryngology, Radboud University Medical Center, Nijmegen, The Netherlands; ²Donders Institute for Brain, Cognition and Behaviour, Radboud University, Nijmegen, The Netherlands; ³Pento Speech and Hearing Centers, Apeldoorn, The Netherlands; and ⁴Royal Dutch Kentalis, Sint-Michielsgestel, The Netherlands.

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with social and emotional development (e.g., behavior issues, mental health, social skills, and interactions) (Wake et al. 2004; Barker et al. 2009; Stevenson et al. 2010; Gentili & Holwell 2011; Netten et al. 2015). Emotional and behavioral problems are therefore frequently reported in children with severe HL, profound HL or who are deaf with or without HAs (Mitchell & Quittner 1996; van Eldik et al. 2004; van Gent et al. 2007), and are more often reported in children with severe HL with HAs than in children who are HH or deaf with CIs (Theunissen et al. 2014; Theunissen et al. 2015; Wong et al. 2017). Children who are HH or deaf who receive CIs at a young age and achieve adequate communication seem to exhibit as few emotional and behavioral problems as their normal hearing peers (Theunissen et al. 2015; Boerrigter et al. 2019).

There is an ongoing debate about whether children with severe HL could also benefit from CIs over HAs (Lovett et al. 2015; Leigh et al. 2016; de Kleijn et al. 2018) to improve their relatively poor performance in a variety of domains. Parental preferences and clinical indication criteria for the choice of device (HA or a CI) for children with prelingual severe to profound HL are mostly based on the etiology and degree of hearing level. For optimal (early) intervention in children who are HH or deaf, this choice has to be made within the first year in life. However, in babies with severe HL, the choice between HAs or CIs is difficult when the consequences for various long-term developmental areas are not yet clear. The latest research shows surprisingly promising results in children who are HH or deaf who receive CIs at an early age; they achieve speech perception scores better than those of children with severe HL with HA (Yang et al. 2012; Leigh et al. 2016; Nekes 2016). Obviously, for children who are HH or deaf, speech perception through adequately fitted HAs or CIs is key to optimizing their full developmental potential. After all, the perception of speech is an important prerequisite for language development, reading, academic performance, and social and emotional well-being. Acquiring these abilities, however, is primarily taking place in informal, incidental daily situations. In typically developing (TD) children, more than 80% of language and general knowledge is acquired in incidental learning situations (Gillis & Schaerlaekens 2000). Incidental learning (learning without direct teaching) happens in many ways (e.g., through observation, repetition, social interaction, and problem solving). Children with poor listening abilities might have problems piecing together poorly heard information and, as a result, might not profit sufficiently from incidental learning. The inability to “overhear” spoken conversations limits the access of these children to many avenues of incidental learning, and therefore restricts their acquisition of knowledge of language, social interaction and how the world works, as well as stifling their development in many areas (Sarant 2012). Incidental learning is very important in acquiring complex linguistic components such as morphosyntax, reading comprehension, narrative abilities, and verbal reasoning skills. Incidental learning requires being able to recognize speech that is spoken at a distance (often to a third party) and that is spoken in noisy environments. Discourse is known to represent the primary language medium through which academic knowledge is conveyed and acquired, and verbal cognition has been shown to be an important indicator for children’s later academic performance and socioeconomic status (SES) also in deaf children (De Raeve 2015). In addition, a recent study showed that the perception of soft speech influences

the magnitude and the timing of lexical access and competition in TD school-aged children (Hendrickson et al. 2021). This is in agreement with results in deaf children (Davidson 2011); lexical decision was poorer with stimuli at 50 dB SPL as compared to 70 dB SPL. Therefore, speech perception in adverse conditions, such as speech in noise or at soft loudness level is an important parameter. For indication purposes especially, it is proposed that speech perception measures used should reflect the listening challenges that individuals encounter in natural communication situations (Firszt et al. 2004).

Various studies have found that executive function (EF) is a good predictor of performance in everyday life. EF refers to a set of higher-order cognitive processes involved in the self-regulation of thoughts, actions, and emotions. These include mental processes such as planning, (verbal) working memory, inhibiting inappropriate responses, flexibility in adapting to changes, and decision-making, which are necessary for adaptive and goal-oriented behavior (P. Anderson 2002; V. Anderson 2002; Senn et al. 2004). EF is a good predictor of language development, academic achievement, and learning-related behaviors such as listening to instructions, following directions, and accomplishing tasks (Bull et al. 2008; Best et al. 2011; Neuenschwander et al. 2012), as well as for children’s social and emotional development (Riggs et al. 2006; Raaijmakers et al. 2008; Schoemaker et al. 2013).

In children who are HH or deaf, several EF tasks such as working memory, cognitive shifting, planning, and inhibition are poorly developed (Beer et al. 2011; Kronenberger et al. 2013; Kronenberger et al. 2014). In children who are HH or deaf, several factors can limit EF. First, this is probably due to poorer verbal communication, reduced access to social situations, and a higher cognitive load resulting from effortful listening (Lin et al. 2011; Botting et al. 2017). HL makes listening effortful, tiring, or stressful. Greater cognitive resources are needed to perceive sound and speech, with the consequence that this could be detrimental to the cognitive resources needed for other cognitive processes, such as EF (Lin et al. 2011; Pichora-Fuller et al. 2016). This makes long-term developmental outcomes for EF a relevant issue in the debate on CI indication criteria. Second, the language skills of children who are HH or deaf are associated with nonverbal EF tasks and even have predictive value (Botting et al. 2017; Jones et al. 2020). Poorer language levels thus negatively affect EF development.

Given the strong relationship among speech perception skills, language abilities, and EF, we argue that improving speech perception could lead to better language and EF development. If so, one might question whether children with severe HL with HAs are disadvantaged in terms of their developmental opportunities. It is important to investigate whether they would benefit more from CIs than from HAs.

Therefore, we aimed to determine whether children with severe HL with HAs are disadvantaged in their perception of speech, language development, and EF compared to children who are HH or deaf with CIs and if they would benefit in the long-term from CIs over HAs. Our inclusion of long-term results contributes to the debate concerning CI indication criteria. We addressed the following research question to achieve this aim: Do children who are HH or deaf with CIs perform better than children with severe HL with HAs with respect to auditory speech perception, and subsequently receptive vocabulary and/or EF?

MATERIALS AND METHODS

Participants

The participants are children with severe HL, profound HL or who are deaf with HAs or CIs. Children with HAs in this study had severe or profound HL according to the WHO classification (WHO 2020), the unaided best ear pure-tone average (PTA) of 60 dB HL or higher, averaged over the frequencies of 1000, 2000, and 4000 Hz. The mean PTA was 69 dB HL (SD = 8.7), ranging from 60 to 97 dB HL. HAs were fitted according to the latest Desired Sensation Level prescriptive formula for children (Seewald et al. 2005). Children with CIs are HH or deaf, with an unaided best ear PTA >90 dB HL. CIs were all fully inserted. We included children from ages 8 to 15 with a nonverbal intelligence quotient of 80 or higher. We recruited children who are HH or deaf at Radboud University Medical Centre and four speech and hearing centers in the Netherlands. The CI group and HA group did not differ in language, educational setting, developmental problems, test age, SES, or nonverbal intelligence quotient. We measured SES regarding parents' jobs on a 4-point scale. Children with

additional developmental problems were diagnosed with attention deficit hyperactivity disorder, autism spectrum disorder, or had motor disabilities or learning problems such as dyslexia. The ages and characteristics of children who are TD varied per test norm group for specific assessments and are reported for each test separately in the assessment section below. Table 1 provides the characteristics of children with CIs and HAs.

Ethical Considerations

The reference number of the study is 2016–2611 (July 14, 2016). The research ethics committee of the Radboud University Nijmegen Medical Centre states that the above-mentioned study does not fall within the remit of the Medical Research Involving Human Subjects Act (WMO) for children with CIs from Radboudumc. Further, for children with HAs from external audiological centers, all parents of participants and participants older than 12 provided written informed consent for the use of their data, in accordance with the Declaration of Helsinki.

TABLE 1. Descriptive statistics of the total study population

	Total study population (N = 70)		p
	CI (n = 43)	HA (n = 27)	
Demographics	n (%)	n (%)	
Sex			0.894
Male	23 (54)	14 (52)	
Female	20 (46)	13 (48)	
Age at onset of hearing loss			0.006*
<3 yr old	36 (84)	19 (70)	
≥3 yr old	1 (2)	7 (26)	
Unknown	6 (14)	1 (4)	
Etiology of hearing loss			0.018*
Genetic	28 (65)	12 (44)	
Acquired	11 (26)	5 (19)	
Idiopathic	4 (9)	10 (37)	
Fitting			0.000*
Unilateral	17 (39)	0 (0)	
Bilateral	21 (49)	27 (100)	
Bimodal	5 (12)	0 (0)	
Language			0.409
Spoken	27 (63)	21 (78)	
Sign-supported spoken language	7 (16)	3 (11)	
Dutch Sign Language	9 (21)	3 (11)	
Educational setting			0.466
Mainstream	24 (56)	19 (71)	
Special Hearing loss	15 (35)	6 (22)	
Special other	4 (9)	2 (7)	
Additional developmental problems			0.757
Yes	8 (19)	4 (15)	
No	35 (81)	23 (85)	
	M (SD)	M (SD)	p
Test age (yr)	11.85 (1.87)	11.01 (2.26)	0.096
Age at detection of hearing loss (mo)	4.70 (9.09)	19.62 (23.30)	0.000*
Age at hearing aids (mo)	11.39 (9.90)	27.96 (25.88)	0.000*
Age at implantation (mo)	34.35 (31.74)		
Duration of deafness or hearing loss before CI or HA (mo)	27.30 (23.6)	7.50 (10.07)	0.002*
Socioeconomic status†	2.32 (1.09)	2.29 (0.99)	0.898
Nonverbal IQ	102.94 (11.98)	103.10 (15.69)	0.973

*p < 0.05.

†Socioeconomic status score was measured by parental jobs, range 0–4.

CI, cochlear implant; HA, hearing aid; IQ, intelligence quotient.

and the delayed recall are transformed to percentile scores with an age-appropriate mean score of 5.5 (SD 1.5) and used as a measure of short-term verbal memory and long-term verbal memory, respectively. Normative data are based on a sample of 225 Dutch children ranging in age from 6 to 12 years (van den Burg & Kingma 1999).

Statistical Analyses

We used IBM SPSS Statistics 25 for all statistical analyses. We employed nonparametric analyses since the assumption of normality, tested with the Kolmogorov-Smirnov test, was violated for almost all study variables. First, we computed the median and interquartile range of all outcomes for the CI and HA groups separately. Second, we compared the group results for receptive vocabulary and EF of the HA and CI groups with the norm data of the TD group, and we performed a one-sample Chi-square test to examine the goodness of fit ($p < 0.05$). Differences between the distributions reflect below-average performance. Next, we contrasted the median scores for speech perception, receptive vocabulary, and EF between the CI and HA groups using the Mann-Whitney U test ($p < 0.05$). Finally, we employed Kendall's tau-b ($p < 0.05$) to investigate any relationships between speech perception, receptive vocabulary, EF, and other study variables.

RESULTS

Comparison of Outcomes Between the CI and HA Groups

Speech Perception • Children with CIs reached a median speech perception score of 87% at 45 dB SPL. The children with HAs reached a median speech perception score of 68%. The Mann-Whitney U test indicated that speech perception scores at 45 dB SPL in the CI group (mean rank = 45.14, $n = 43$) were significantly higher than those in the HA group (mean rank = 20.15, $n = 27$), $U = 995.00$, $z = 5.01$, $p < 0.01$, two tailed. This effect can be described as a large effect ($r = 0.60$) and is illustrated in Figure 1. This means that children with CIs on

average have better auditory perception at a soft speech level than children with HAs. There was no significant difference between children with CIs and HAs in speech perception performance at the conversation level. Both groups obtained ceiling scores. The median speech perception scores were 96% and 93%, respectively.

In Table 2, the nonparametric descriptive statistics for receptive vocabulary and EF are presented for the CI and HA groups, as well as the expected results from their TD peers.

Receptive Vocabulary • There was no significant difference in receptive vocabulary scores between children with CIs and HAs.

Executive Function • The Mann-Whitney U test showed no significant differences in EF domain scores between the children with CIs and HAs. Distributions of the scaled scores of both groups on the D-KEFS tests are presented in Figure 2. Figure 3 displays the distributions of the CI and HA groups for the Dutch RAVLT.

Comparison of Outcomes of CI and HA Groups With the Normative Data of the TD Group

Receptive Vocabulary • The Chi-square test for goodness of fit indicates significant differences between observed and expected distributions between the children with CIs and the norm group data of the TD children ($p = 0.033$, with a medium effect size $\omega = 0.32$); 28% of the CI group obtained below-average receptive vocabulary scores. We did not detect any significant differences in receptive vocabulary between the HA group and the norm group data of the TD children.

Executive Function • The Chi-square test for goodness of fit demonstrated, for the CI group, significant differences between observed and expected distributions based on the norm group data of the children who are TD on the Tower test (planning) ($p = 0.004$, medium effect size $\omega = 0.44$) of the D-KEFS. We did not observe a significant difference on the other tests of the D-KEFS. Short-term and long-term verbal memory of the Dutch RAVLT also revealed significantly different outcomes in the CI group. Regarding

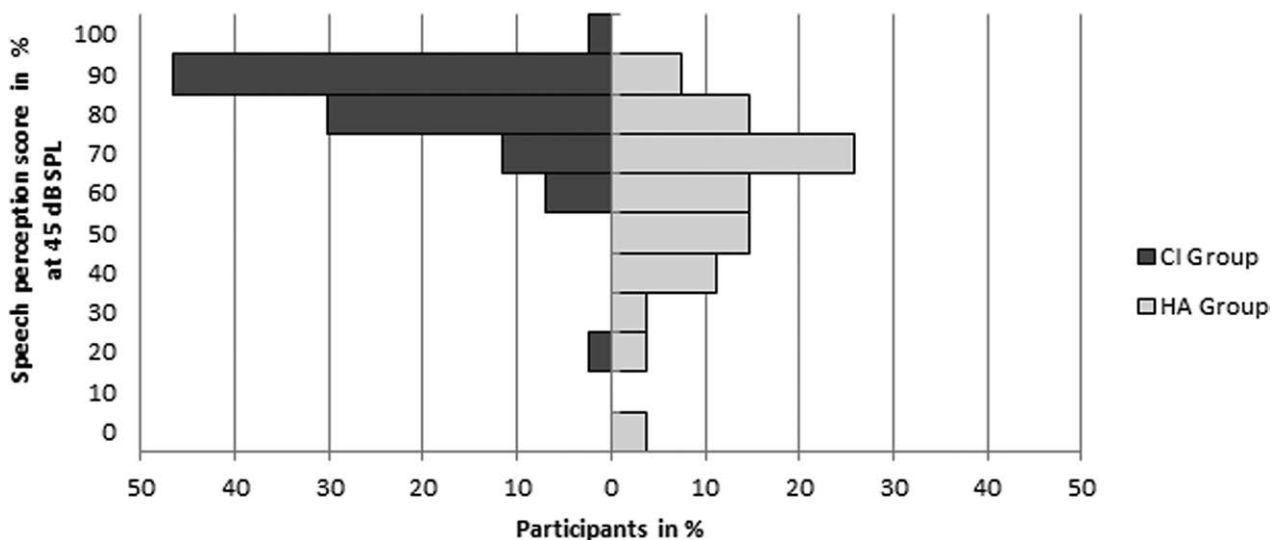


Fig. 1. Distribution of speech perception scores on the speech perception test at 45 dB SPL in the CI and HA groups. CI, cochlear implant; HA, hearing aid.

TABLE 2. The nonparametric descriptive of receptive vocabulary and EF results for the CI and HA groups and the expected results of the TD group

Materials	Distributions: mean (–1 SD to +1 SD)			% Below average		
	TD norm group	CI	HA	TD	CI	HA
PPVT						
Receptive vocabulary	100 (85–115)	93 (78–107)	94 (84–103)	16	28	22
D-KEFS						
Color-Word Interference Test						
Inhibition	10 (8–12)	10 (8–12)	10 (9–11)	16	19	7
Verbal cognitive flexibility	10 (8–12)	10 (8–12)	10 (9–12)	16	24	11
Design fluency test						
Nonverbal cognitive flexibility	10 (8–12)	11 (9–13)	12 (9–13)	16	12	19
Tower test						
Planning	10 (8–12)	8 (6–9)	9 (6–9)	16	44	33
20-questions test						
Problem solving	10 (8–12)	11 (9–12)	12 (9–14)	16	7	11
Trail-making test*						
Verbal cognitive flexibility	10 (8–12)	11 (8–12)	11 (9–13)	16	8	8
Dutch RAVLT						
Short-term verbal memory	5.5 (4–7)	4 (1–7)	5 (4–8)	16	47	23
Long-term verbal memory	5.5 (4–7)	4 (1–8)	5.5 (2–7)	16	44	35

*N = HA group: N = 25. CI group: N = 38.

CI, cochlear implant; D-KEFS, Delis Kaplan Executive Function System; EF, executive function; HA, hearing aid; PPVT, Peabody Picture Vocabulary Test; Dutch RAVLT, Ray Auditory Verbal Learning Test; TD, typically developing.

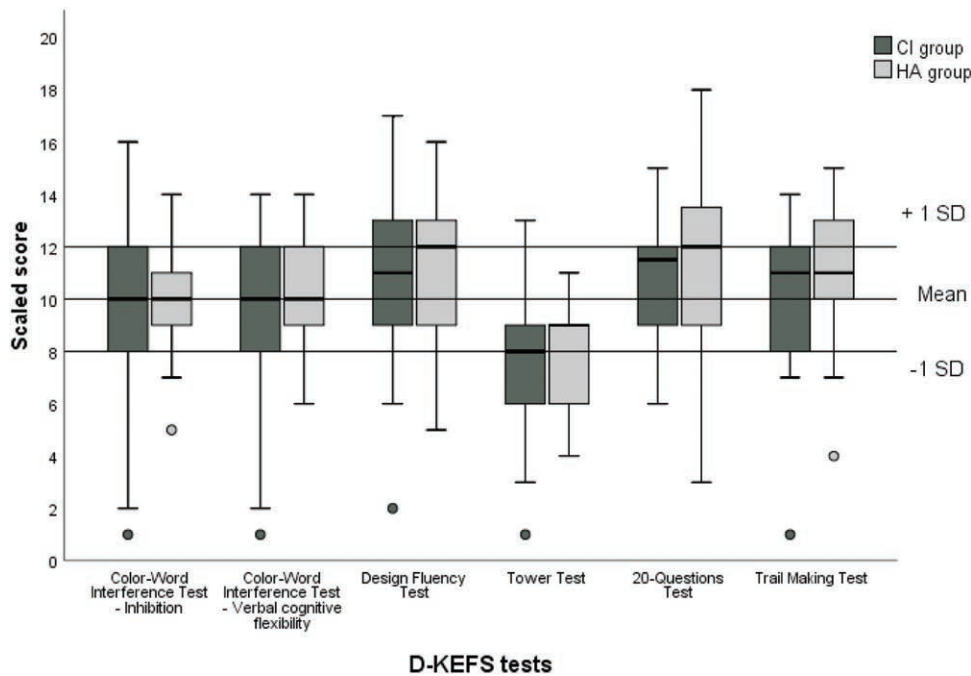


Fig. 2. Distributions of scaled scores on the D-KEFS tests in the CI and HA groups. CI, cochlear implant; D-KEFS, Delis Kaplan Executive Function System; HA, hearing aid.

differences between observed and expected distributions of short-term verbal memory, $p = 0.002$, with a large effect size, and $\omega = 0.51$; for long-term verbal memory, $p = 0.007$, with a medium effect size, $\omega = 0.45$. Forty-four percent of the CI group obtained below-average scores for planning, 47% on the short-term verbal memory tasks, and 44% on the long-term verbal memory tasks.

In the HA group, we only found a significant difference between the observed and expected distributions of the TD norm group on the Tower test (planning) ($p = 0.014$, medium effect size $\omega = 0.47$) of the D-KEFS and the long-term verbal memory tasks of the

Dutch RAVLT $p = 0.01$, with a large effect size, $\omega = 0.50$. For planning, 33% of the HA group obtained below-average scores, and 35% obtained below-average scores for long-term verbal memory.

Associations Between Speech Perception, Receptive Vocabulary, EF, and Other Study Variables for the CI and HA Groups Separately

We used Kendall's tau-b to investigate relationships among speech perception, receptive vocabulary, and EF. Looking at the relationship among speech perception, receptive vocabulary,

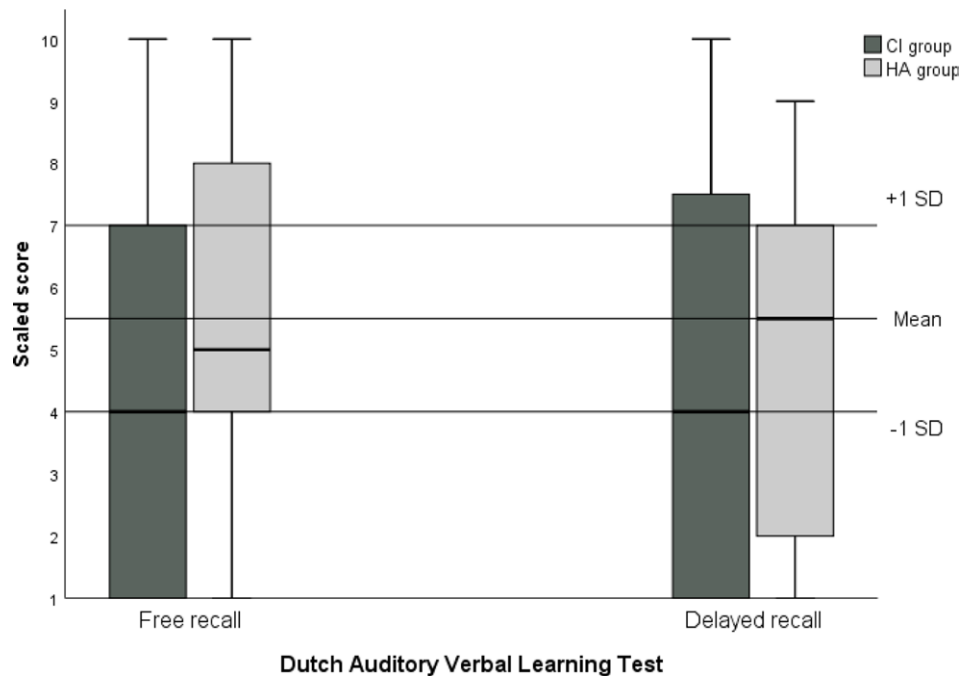


Fig. 3. Distributions of scaled scores on the Dutch RAVLT in the CI and HA groups. CI, cochlear implant; HA, hearing aid; RAVLT, Rey Auditory Verbal Learning Test.

and EF, we noted a significant, medium positive association in the HA group between speech perception at 45 dB SPL and the receptive vocabulary standard score ($\tau = 0.284$, $p = 0.042$, two-tailed, $n = 27$), and a significant, medium, positive association ($\tau = 0.307$, $p = 0.038$, two-tailed, $n = 27$) between speech perception at 45 dB SPL and the Tower test (planning). No significant relationships were found among speech perception, receptive vocabulary, and EF in the CI group. A visual representation of the significant relationships with speech perception at 45 dB SPL is presented in Figure 4. Other significant relationships are portrayed in Table 3.

DISCUSSION

We aimed to determine whether children with severe HL with HAs are disadvantaged in their perception of speech, receptive vocabulary, and EF compared to children who are HH or deaf with CIs and if they would benefit in the long-term from CIs over HAs. The research question is as follows: Do children who are HH or deaf with CIs perform better than children with severe HL with HAs with respect to auditory speech perception, and subsequently receptive vocabulary and/or EF?

First, we made a comparison between the outcomes of the CI and HA groups on speech perception, receptive vocabulary, and EF. We found no difference in the perception of speech at the conversational level (65 dB SPL) between children with CIs and HAs. This is in contrast to the results of Leigh et al. (2016), who reported that children who are HH or deaf with CIs reached higher speech perception scores than children with severe HL with HAs (PTA > 66 dB HL) for speech perception at 65 dB SPL. The difference in the outcomes of Leigh et al. (2016) compared to our study can be attributed to the age of CI. The children with CIs in the study of Leigh et al. (2016) all received their CIs before 3 years of age. The mean age at CI in our study was 2 years and 10 months, with a SD of 2 years and 8 months.

As well established, speech perception in adverse listening situations is an important factor for the linguistic, cognitive, and socioemotional development. Therefore, it is necessary to assess at lower intensity levels that reflect real-life acoustic conditions. In our study, the children with severe HL with HAs demonstrated significantly poorer perception of soft speech (45 dB SPL) compared to children who are HH or deaf with CIs, which is in line with our hypothesis. Thus, children with HAs exhibit less favorable auditory perception of soft speech. This implies that even state of the art technology does not provide a consistently optimal signal.

Yang et al. (2012) found that children with CIs had better speech in noise perception scores than children with severe HL (PTA > 70 dB HL) with HAs. This indicates that perception of speech in more challenging acoustic situations is better among children with CIs than in children with severe HL with HAs.

We compared the outcomes of the CI and HA groups to the norm group of children who are TD and computed associations between variables. Next, we discuss these findings. The receptive vocabulary of both groups was age-appropriate. This is in line with earlier reports, such as the work of Langereis & Vermeulen (2013), who indicated that phoneme scores >85% at the conversational level (65 dB SPL) enable children to develop sufficient receptive vocabulary. There was no association between the perception of speech at 65 dB SPL and receptive vocabulary probably due to the ceiling effect. Nonetheless, we found an association between the perception of soft speech at 45 dB SPL and receptive vocabulary in the HA group. Accordingly, De Raeve et al. (2015) reported a positive correlation between the perception of soft speech and verbal cognition in children with CIs. These findings suggest that increasing perception of soft speech in children with severe HL with HAs could contribute to improved language development abilities. In contrast, we did not observe a relationship between the perception of soft speech and receptive vocabulary in children who are HH or deaf with

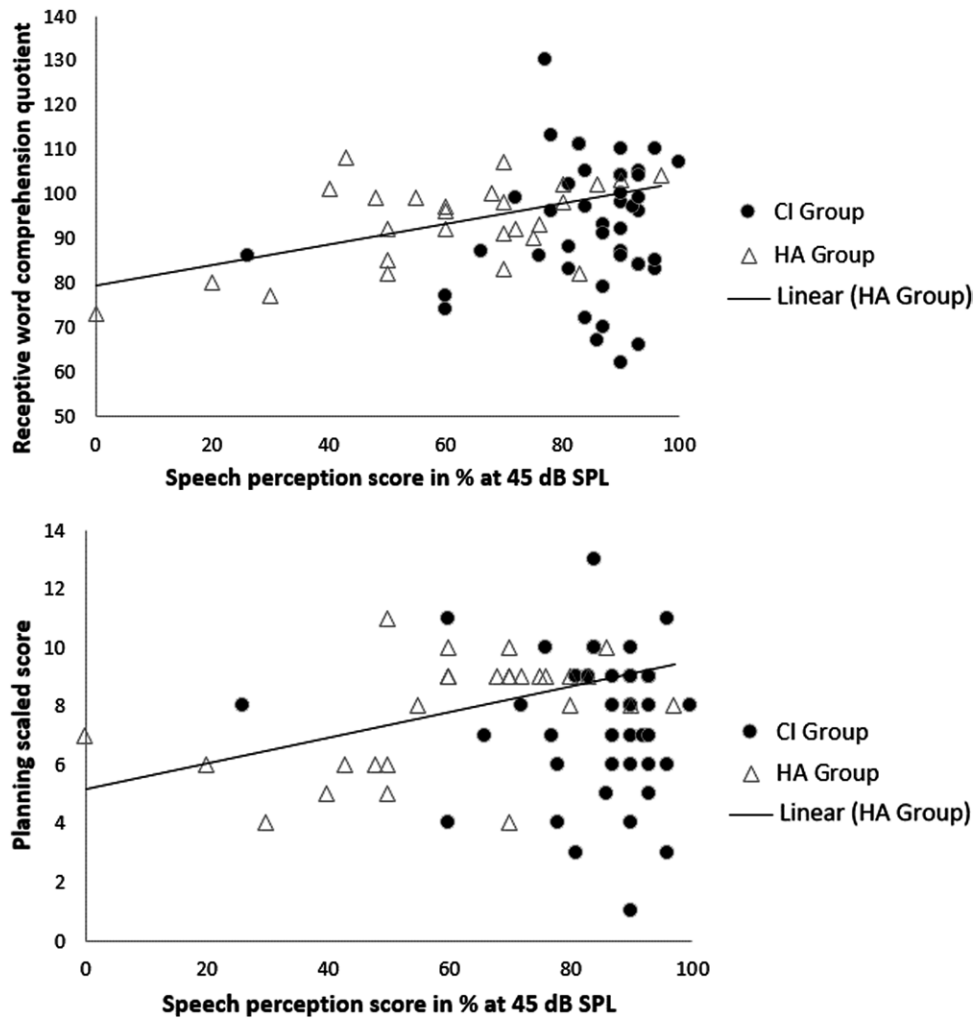


Fig. 4. Scatterplots of the Relation of Speech Perception scores with Receptive vocabulary and with planning. A and B, Scatterplot of the positive linear correlation between speech perception at 45 dB SPL and receptive vocabulary, and between speech perception at 45 dB SPL and planning in the HA group. CI, cochlear implant; HA, hearing aid.

CIs. The absence of this relationship in the CI group is likely due to the distribution of speech perception scores at 45 dB SPL of children who are HH or deaf with CIs. Approximately 80% of these children achieved soft speech perception scores of 75% or higher. The relationship between speech perception and language development has already been demonstrated (Tsao et al. 2004; Leigh et al. 2013; Dettman et al. 2016). In our study, a small group of children who are HH or deaf with CIs exhibited relatively good speech perception levels but nevertheless had poor receptive vocabulary levels. One explanation could be that 39% of the children in the CI group were fitted with a unilateral CI. Research by De Raeve et al. (2015) and Jacobs et al. (2016) implies that bilateral CI users have better language and verbal cognitive outcomes than unilateral CI users. Thirty-nine percent of the CI group in our study received unilateral implants, whereas currently, in the Netherlands, children who are HH or deaf almost always receive bilateral CIs; hence, improved developmental outcomes are expected in these children (De Raeve et al. 2015; Jacobs et al. 2016; Di Stadio et al. 2018). Another explanation for the children with CI who exhibited relatively good speech perception levels but had poor receptive vocabulary levels could be due to the relatively old age at

intervention in our CI group (our study group was implanted at a relatively old age; $M = 34$ months, $SD = 32$). As stated before, earlier implantation (before 12 months) leads to better auditory and language outcomes (Dettman et al. 2016; Kral et al. 2016; McKinney 2017). However, we did not observe a correlation between the age of implantation and receptive vocabulary. A third explanation could be related to the etiology of deafness. A frequent diagnosis in children with HL is cytomegalovirus or other etiologies of severe disabilities. Children with cytomegalovirus vary widely in their audiometric, language, and cognitive outcomes (Ciorba et al. 2009; Fletcher et al. 2018). In our study, the etiology of a large share of the HA group was unknown. We thus did not include etiology in the analyses.

Our results indicate that to obtain age-appropriate levels of receptive vocabulary and EF, the perception of soft speech is a necessary but insufficient prerequisite. For children with severe HL with HAs an association between the perception of soft speech and receptive vocabulary and planning was found. For children who are HH or deaf with CIs, poor planning and verbal memory abilities cannot be associated to deficits in the perception of soft speech. Interestingly, children with CI who have poor speech perception abilities show poor vocabulary and planning

TABLE 3. Kendall's tau-b correlations between study variables and executive function for CI (gray) and HA (white) groups

Study variables	1.	2.	3.	4.	5.	6.	7.	8.	9.	10.	11.	12.	13.	14.	15.
1. Age at onset of hearing loss (HL)	—	0.276*	—	0.120	0.222	-0.233	-0.041	0.130	0.102	-0.023	0.007	0.257	-0.210	0.095	0.058
2. Age at implantation	—	—	—	-0.169	0.037	-0.129	-0.065	0.085	0.250*	0.203	-0.025	-0.138	0.018	0.026	0.044
3. pure-tone average (PTA) best ear	-0.146	—	—	—	—	—	—	—	—	—	—	—	—	—	—
4. Socioeconomic status	0.016	—	-0.162	—	0.012	0.052	0.233	0.233	0.097	0.096	0.022	-0.093	0.273	0.259	0.067
5. Nonverbal IQ	-0.479*	—	-0.087	0.345*	—	0.417*	0.169	0.045	0.531*	0.327	0.497*	0.258	0.163	-0.025	0.163
6. Speech perception at 45 dB SPL	-0.295*	—	-0.410*	0.104	0.284	—	0.319*	0.037	0.099	0.191	0.177	-0.065	0.173	0.078	0.103
7. Speech perception at 65 dB SPL	-0.326*	—	0.088	0.211	0.387*	0.358*	—	-0.045	0.134	0.224	0.233*	-0.090	0.133	0.162	0.113
8. Receptive vocabulary	-0.188	—	0.045	0.139	0.477*	0.284*	0.191	—	0.121	0.001	0.134	0.072	0.030	0.067	0.007
9. Color-Word Interference Test—Inhibition	0.118	—	-0.088	0.291	0.119	-0.019	0.107	-0.099	—	0.564†	0.291*	-0.14	0.157	0.160	0.182
10. Color-Word Interference Test—Verbal cognitive flexibility	0.044	—	0.053	0.094	0.344*	-0.071	0.039	0.213	0.340*	—	0.256*	0.019	0.208	0.255*	0.216
11. Design fluency test—Nonverbal cognitive flexibility	-0.022	—	-0.056	0.417*	0.178	0.049	0.080	0.088	0.283	0.171	—	0.147	0.142	0.183	0.372*
12. Tower test—Planning	-0.027	—	-0.091	0.000	-0.096	0.307*	0.080	-0.019	-0.13	-0.066	0.030	—	-0.187	-0.065	-0.175
13. 20-questions test—Problem solving	-0.047	—	0.061	0.075	0.297	0.076	0.073	0.445*	-0.006	0.205	0.094	-0.013	—	0.209	0.406*
14. RAVLT short-term verbal memory	-0.024	—	0.167	0.338*	0.158	-0.039	-0.212	0.013	0.080	0.000	0.251	0.014	0.094	—	0.605†
15. RAVLT long-term verbal memory	0.020	—	0.054	0.109	-0.138	0.206	-0.038	-0.135	0.095	-0.169	0.251	0.386*	-0.48	0.319*	—

† $p < 0.05$.‡ $p < 0.001$.

CI, cochlear implant; HA, hearing aid; IQ, intelligence quotient.

skills, but children with CI who have good speech perception show a very large variability. Assessment of speech perception in challenging listening conditions, such as the perception of soft speech or speech in noise, could be relevant for CI indications. Several studies suggest broadening the auditory threshold inclusion criteria for pediatric CI to a PTA of 80 dB HL (de Kleijn et al. 2018) or even 60 dB HL (Leigh et al. 2016). In Belgium, the auditory criteria for CI were recently revised to a PTA of 70 dB HL (De Raeye et al. 2020). In line with these results, our findings suggest that it is important to determine whether auditory gain with a CI is associated with other developmental areas.

The analyses of EF outcomes, which were focused on a comparison of the CI and HA groups with the norm group, revealed a difference between the distributions of children with CIs and children who are TD in three domains: (1) planning and (2) short- and (3) long-term verbal memory tasks. Large numbers of children with CIs exhibited below-average scores compared to the TD norm group data on these tasks. The finding that children who are HH or deaf with CIs demonstrate short-term memory problems is in line with other studies. AuBuchon et al. (2015) and Watson et al. (2007) showed that children who are HH or deaf with CIs demonstrated short-term verbal memory developmental problems compared to their hearing peers. This could be attributed to the large number of children with a unilateral CI (39%) compared to the children with HAs, who all have bilateral HA fittings. Di Stadio et al. (2018) found that children with unilateral HL exhibited increased memory problems compared to children with bilateral hearing devices or their normal hearing peers. Stiles et al. (2012) indicated that children with mild or severe HL with HAs showed resilience in verbal short-term memory, with the important caveat that all children who used HAs in their study used oral communication and were enrolled in oral classrooms. It could be that the verbal/oral learning environment in the mainstream educational setting contributes to a better verbal memory system. Therefore, the educational setting could partly explain the observed differences in verbal memory between the HA and CI groups. In our study, the HA group, 71% of children were enrolled in a mainstream educational setting compared to 56% of the CI group.

Our study demonstrates that a considerable proportion of children with HL experience problems in planning. A possible explanation for this might be that inner speech is reduced in children with severe or profound HL. Inner speech has an important role in self-regulation, cognition and behavior (Alderson-Day & Fernyhough 2015), and interferes with EF. Wallace et al. (2017) investigated the role of verbal mediation in a tower test task in a hearing population, demonstrating that efficient planning relies on inner speech, which seems to be related to language development (Alarcon-Rubio et al. 2013). The fact that we did not observe a direct relationship between language and planning in our study could be because we examine receptive vocabulary only. According to Vygotsky's theory of cognitive development, inner speech comprises a transformation from external social speech to private speech to inner speech. This implies that expressive language should also be explored. Jones et al. (2020) found that expressive vocabulary affects EF development. However, inner speech is a complex, abstract level of language, containing more aspects such as thoughts, feelings, and knowledge. It is a difficult concept to measure and cannot be captured by standardized language tests (Alderson-Day & Fernyhough 2015). Further research should be undertaken to

determine the role of language and inner speech in EF in children with HL with CIs and HAs. We did identify a correlation between the perception of soft speech and planning in children with severe HL with HAs.

For children with HL who obtained deviant planning or verbal memory scores, intensive computer-based EF training could be beneficial. EF computer training seems beneficial for children with CIs children with specific memory and language problems (Kronenberger et al. 2011). Perhaps this could also be helpful for children with HAs. It remains unclear, however, how these improved skills would generalize across diverse situations and behaviors in daily life. It is therefore recommended that EF computer-based training be combined with EF training that takes place in daily life situations in which the child is typically situated, such as at school, at home, or in sports or music training (Hsu et al. 2014; Blair 2017).

CONCLUSIONS

Children with severe HL with HAs exhibit less favorable auditory perception of soft speech, but not at a conversational level, compared to children who are HH or deaf with CIs. More children with severe HL with HAs and children who are HH or deaf with CIs exhibit problems in planning and verbal memory but not in other EF compared to the norm groups of TD children. The results indicate that to obtain age-appropriate levels of receptive vocabulary and EF, the perception of soft speech is a necessary but not sufficient prerequisite.

LIMITATIONS

There are several limitations regarding the participants in our study. With respect to children who are HH or deaf with CIs, the mean age at implantation was relatively late (34 months). This may have affected language and EF results because early implantation leads to better outcomes (Dettman et al. 2016; Kral et al. 2016; McKinney 2017). Further, this group consisted of 51% unilateral CI users in contrast to the children with severe HL with HAs, who had all bilateral HAs. Research shows that bilateral CI users have better language and cognitive outcomes than unilateral CI users (De Raeve et al. 2015). Currently, children who are HH or deaf in the Netherlands almost always receive bilateral CIs and are implanted earlier in life, and improved developmental outcomes are expected in these children. With regard to children with severe HL with HAs in the study, only four children had a PTA higher than 75 dB HL. The vast majority of the group had a PTA between 60 dB and 75 dB HL. Seventy-one percent of these children were enrolled in mainstream educational settings. Children with severe HL with HAs who were enrolled in special educational settings often declined to participate. The results of children with severe HL with HAs in this study group may be an overestimation. Further research with more matching research groups is needed to answer the question of which levels of hearing threshold CI could add to speech perception ability, resulting in better receptive vocabulary and EF performance in children with severe HL.

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Address for correspondence: Merle S. Boerrigter, Radboud University Medical Center, Department of Otolaryngology (route 383) and Radboud University, Donders Institute for Brain, Cognition and Behaviour, P.O. Box 9101, 6500 HB Nijmegen, The Netherlands. E-mail: merle.boerrigter@radboudumc.nl

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