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Receptive Vocabulary of Children With Bilateral Cochlear Implants From 3 to 16 Years of Age

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Objectives: The vocabulary of children with cochlear implants is often smaller than that of their peers with typical hearing, but there is uncertainty regarding the extent of the differences and potential risks and protective factors. Some studies indicate that their receptive vocabulary develops well at first, but that they fail to keep up with their typical hearing peers, causing many CI users to enter school with a receptive vocabulary that is not age-appropriate. To better understand the receptive vocabulary abilities of children with cochlear implants this study explored age-related differences to matched children with typical hearing and associations between vocabulary skills and child-level characteristics.

Design: A retrospective cross-sectional study with matched controls was conducted at the Norwegian national cochlear implant center at Oslo University Hospital. Eighty-eight children (mean age 8.7 years; range 3.2 to 15.9; 43 girls, 45 boys) who had received bilateral cochlear implants before 3 years of age were compared with two groups of children with typical hearing. One group was matched for maternal education, sex, and chronological age, the other group was matched for maternal education, sex, and hearing age. Receptive vocabulary performance was measured with the British Picture Vocabulary Scale.

Results: Cochlear implant users' receptive vocabulary was poorer than that of age-matched children with typical hearing ($M = 84.6$ standard points, $SD = 21.1$; children with typical hearing: $M = 102.1$ standard points, $SD = 15.8$; mean difference −17.5 standard points, 95% CI [−23.0 to −12.0], $p < 0.001$; Hedges's $g = -0.94$, 95% CI [−1.24 to −0.62]), and children with cochlear implants were significantly more likely to perform below the normative range (risk ratio = 2.2, 95% CI [1.42 to 3.83]). However, there was a significant nonlinear U-shaped effect of age on the scores of cochlear implant users, with the difference to the matched typical hearing children being largest (23.9 standard points, on average) around 8.7 years of age and smaller toward the beginning and end of the age range. There was no significant difference compared with children with typical hearing when differences in auditory experience were accounted for. Variability was not significantly different between the groups. Further analysis with a random forest revealed that, in addition to chronological age and hearing age, simultaneous versus sequential implantation, communication mode at school, and social integration were predictors of cochlear implant users' receptive vocabulary.

Conclusions: On average, the receptive vocabulary of children with cochlear implants was smaller than that of their typical hearing peers. The magnitude of the difference was changing with age and was the largest for children in early primary school. The nonlinear effect of age might explain some of the ambiguity in previous research findings and could indicate that better intervention is required around school entry. The results emphasize that continuous monitoring and support are crucial to avoid far-reaching negative effects on the children's development and well-being.

Key words: Children, Cochlear implant, Language development, Vocabulary.

Abbreviations: BPVS-II = British Picture Vocabulary Scale, 2nd edition; CI = cochlear implant; 95% CI = 95% confidence interval; dB HL = decibels hearing level; dB SPL = sound pressure level in decibel; df = degrees of freedom; SD = standard deviation; TH = typical hearing.

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Cochlear implants (CI) allow profoundly deaf children to access their sound environment and acquire spoken language, but the language skills of children with CI are often not on par with those of their peers with typical hearing (TH; Ruben 2018; van Wieringen & Wouters 2015). One area in which such deficits exist is receptive vocabulary (Lund 2016; Nittrouer & Caldwell-Tarr 2016): In a meta-analysis of 16 studies that have compared the receptive vocabulary of children with CI to that of age-peers with TH, Lund (2016) found that children with CI in all studies had significantly poorer receptive vocabulary skills, with effect sizes (*d*) between 0.46 and 2.00. Across the studies, there was wide variation in the CI users' mean chronological age (range, 4.1 to 10.1 years) and mean age of CI activation (range, 16 to 46.5 months). However, a meta-regression found no significant associations between either of these variables and the size of the vocabulary deficit, as well as no effect of time since implantation. Lund (2016) concluded that children with CI enter school with vocabulary knowledge that is poorer than that of their peers and that even children who receive CI early cannot be expected to catch up quickly. If this is true, it might indicate that current rehabilitation programs do not sufficiently support children throughout this period of their language development. To further investigate this issue, the present cross-sectional study assesses the vocabulary performance of children with CI who are between 3 and 16 years of age—covering the period from preschool to the end of compulsory education.

IMPACT OF CHILD-LEVEL CHARACTERISTICS ON RECEPTIVE VOCABULARY OF CHILDREN WITH CI

Although Lund (2016)'s meta-regression did not find an effect of implantation age on vocabulary development, individual studies have shown that earlier age at implantation facilitates spoken language development. Especially children who receive a CI in the first year of their life often perform comparably to TH children on measures of receptive vocabulary at school entry (Dettman et al. 2016; Karltorp et al. 2020; Wenrich et al. 2019).

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A beneficial effect of implantation in the first year of life is also in line with neurodevelopmental theories that emphasize the importance of early auditory experience for the development of the brain networks that underly language learning. Thus, the benefit of cochlear implantation for congenitally deaf children is largest during early development, when the brain is most sensitive to spoken language input (Bruijnzeel et al. 2016; Kral et al. 2019; Werker & Hensch 2015). However, the positive effect of early implantation may later be obscured to some extent by variation in developmental trajectories: as children become older, some early-implanted children fall behind, while some late-implanted children catch up (Dunn et al. 2014). Thus, the effect of age at implantation might depend on the range of age and implantation age under investigation (Geers et al. 2007).

Vocabulary development is also associated with age and time since implantation, most likely because these are proxies for auditory experience. Compared with their TH peers, children with CI have a deficit in auditory experience and more auditory experience gives them more time to catch up with their TH peers. However, to catch up, their vocabulary needs to grow at a faster-than-normal rate, and it is unclear whether this is the case. Some studies found that the vocabulary of children with CIs grew slower than that of their TH peers, causing the gap to widen over time. For example, Blamey et al. (2001) found that the receptive vocabulary of children with CI was increasingly falling behind the norm between 4 and 13 years of age. At 13 years of age, it was predicted to be roughly equivalent to that of 8-year-old children with TH. However, the onset of hearing loss for the children in this study was up to 3.4 years of age and they were implanted, on average, at 3.5 years of age. Connor et al. (2000) found an age-equivalent improvement in receptive vocabulary scores of only 3.3 years in the 8 years after implantation. Again, the age at implantation in this study was high, with an average of 5.6 years. Similarly, El-Hakim et al. (2001) found that the receptive vocabulary of children who were implanted at an average of 5.8 years of age was growing at half the rate of that of TH children over the 9 years after implantation. Moreover, the growth rate was decreasing over time. In all of these studies, children were implanted relatively late. Nowadays, children typically receive their implants substantially earlier. In more recent samples, one might therefore expect higher vocabulary growth rates. Indeed, Hayes et al. (2009) found faster than normal vocabulary growth in the 6 years following the implantation for children who were implanted before 2 years of age, indicating that these CI users caught up with their TH peers as their implant experience increased. However, toward the end of the age range, the vocabulary growth decelerated. In a study of children who had received bilateral CI no later than 18 months of age, Wie et al. (2020) found that the gap in receptive vocabulary was closing at first but then widened again 4 to 5 years after the implantation, around the time that children entered school. Thus, it appears that at different ages, vocabulary grows at different rates and that promising vocabulary growth in the first years after the implantation is no guarantee that the children will maintain age-appropriate vocabulary performance as they grow older. Wie et al. (2020) speculated that the widening gap might be the consequence of an increasingly difficult-to-acquire vocabulary or that it might be caused by changes in the educational environment: mainstream primary school classrooms could be less conducive to vocabulary acquisition of children with CI than to that of children with TH.

Many other factors influence language development after cochlear implantation and their interplay is complex (Cosetti & Waltzman 2012). It is often difficult to determine whether something is a cause or an effect of differences in developmental trajectories. For example, better cognitive abilities, such as working memory and inhibition-concentration, benefit vocabulary acquisition (Dettman et al. 2016; Wenrich et al. 2019), possibly because they can facilitate speech understanding in acoustically or cognitively demanding situations such as classrooms and other noisy everyday listening situations (Pichora-Fuller et al. 2016). This might be particularly useful for vocabulary acquisition, as better speech perception promotes incidental learning of new vocabulary (Davidson et al. 2014). At the same time, cognitive development can be hampered by auditory deprivation and language delays (Kral et al. 2016). Similarly, language development and social development are intertwined in such a way that if a hearing impairment disrupts social interactions this can set into motion a vicious circle of psychosocial and language problems (Boerrigter et al. 2019; Hoffman et al. 2015; Wong et al. 2018). In line with this explanation, Haukedal et al. (2018) report small to moderate associations between the language skills of children with CI and their quality of life.

Many child-related characteristics that are associated with language outcomes concern the child's daily environment. For example, children in mainstream educational settings tend to perform better on measures of spoken language (Boons et al. 2013a; Busch et al. 2020; Geers et al. 2007). Likely, this is because the educational setting is chosen based on the children's language performance. However, it is also conceivable that mainstream classrooms provide a richer spoken language learning environment. On a related note, differences in language outcomes are associated with the communication mode that children use at home and school: In a sample of 97 children who had received a CI before 22 months of age (mean age at CI activation 10.6 months), Geers et al. (2017) found that exposure to sign language or sign-based communication systems (e.g., Cued Speech) in the first three years after implantation was negatively associated with measures of spoken language at late elementary school age, even after controlling for other characteristics. Children who were exposed to signing were less likely to achieve age-appropriate spoken language outcomes compared to children who were not exposed to signing. Thomas and Zwolan (2019) found that children who had received their CI before 5 years of age and were in auditory-verbal intervention programs (which focus on listening) outperformed their peers in oral communication programs (which encourages the use of lipreading to supplement listening) and total communication programs (which encourages the use of gestures in communication) on measures of spoken language up to 7 years after the implantation.

Other beneficial properties of the home environment include more language input, higher parental sensitivity and more parental engagement in the rehabilitation (Boons et al. (2012a); Cosetti & Waltzman 2012; Niparko et al. 2010; Quittner et al. 2013; Sarant et al. 2015). A higher family socioeconomic status is also associated with better language outcomes after cochlear implantation (Niparko et al. 2010; Szagun & Stumper 2012), perhaps because of its impact on many characteristics of the child's daily language learning environment.

In their meta-analysis, Lund (2016) found that the difference in receptive vocabulary was larger in studies where children

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with CI were compared with children with TH who were matched for child-level characteristics like socioeconomic status and cognitive abilities. Such matched comparisons arguably paint a clearer picture of the impact of hearing loss and show the importance of child-level characteristics for vocabulary development (Lund 2016).

THE NORWEGIAN CONTEXT

Norway provides an interesting context for developmental research: It is a strong redistributive welfare state with a relatively egalitarian society, low income inequality, and high rates of inter-generational mobility. High-quality health care is provided to all residents and education is publicly financed, free, and highly standardized, with very little variation in school quality (Hermansen et al. 2019; Isungset et al. 2021). As a result, the relation between maternal education and child vocabulary is less evident in Norway than in other countries (Frank et al. 2021). Compared with, for instance, the United States, where social inequalities are higher and education and health care are less standardized, children in Norway receive more equal opportunities, and those opportunities are to a lesser degree determined by their family background (Isungset et al. 2021). This also applies to access to early bilateral implantation and high-quality clinical care: Deaf children in Norway are eligible to receive bilateral cochlear implants in the first year of life, free of charge. There is also only one pediatric cochlear implant center in the country so that all children undergo the same clinical procedures. Consequently, one might expect differences between Norwegian samples of children with CI and samples from other more heterogeneous contexts. In particular, children with CI in Norway may be less affected by their family's socioeconomic status and similar background characteristics. Yet, despite universal access to early bilateral implantation, a good standard of clinical care, and high-quality education, Wie et al. (2020) found a decline in vocabulary performance of Norwegian children with CIs some years after the implantation, which poses the question of which other factors are responsible.

Is the Variation in the Receptive Vocabulary Abilities of Children With CI Larger Than Normal?

Variation in language outcomes after cochlear implantation is notoriously large (e.g., van Wieringen & Wouters 2015). Even in relatively homogenous samples of children with bilateral early implantation, language performance tends to vary more widely than that of their TH peers (Wie et al. 2020). However, many studies do not explicitly test whether the difference in variability is significant. Higher-than-normal variability would mean that the group's average performance is less representative of individual children's experiences, even if it is within the normative range. Moreover, wide variation might be caused by children in the fringes of the distribution who require special attention and perhaps different kinds of intervention for successful rehabilitation. Therefore, a closer look at the extent of variation in the vocabulary skills of CI children is called for.

Current Study

In the literature, there is uncertainty regarding the size of the gap in receptive vocabulary between children with CI and children with TH as well as its trajectory. There is evidence for a non-linear effect of age on vocabulary growth as well as an influence of child-level characteristics such as age at implantation and contextual factors like educational setting. Thus, the primary goal of this study was to compare the vocabulary skills of children with CI to those of their TH peers across a wide age range, covering the period from preschool to the end of compulsory education. As much as possible, the sample was selected to be representative of current clinical practices and optimal developmental conditions, only containing children with relatively early bilateral implantation and no additional disabilities affecting language. All participants were treated by the same clinical team to a high standard of care. Moreover, the egalitarian educational system and strong social welfare system in Norway provided a context, in which the contributions of socioeconomic inequalities and similar factors on child development were expected to be relatively small.

The control groups of children with TH in this study were matched for maternal education (as an indicator of socioeconomic status), sex, and age to more clearly determine the impact of the hearing impairment on vocabulary development. In a second analysis, we controlled for the CI user's hearing age (i.e., implant duration) to test whether differences in vocabulary can be explained by differences in auditory experience, that is, whether children with CI perform similar to children with TH who are younger but have had a similar amount of auditory experience.

Furthermore, we explored inter-individual variation in the receptive vocabulary skills of children with CI. First, we assessed whether the variation in the receptive vocabulary of CI users is larger than the variation among their TH peers. As discussed above, this provides additional context for interpreting the group's performance and might have implications for clinical practice. Finally, we investigated which child-level characteristics explain inter-individual variation in CI users' vocabulary development, including aspects of the home and educational environment, age at implantation, speech perception, and nonverbal abilities.

Similar to previous studies, we expected to find a deficit in CI users' vocabulary scores and a potentially non-linear effect of age on the size of the vocabulary gap. We also expected that child-level characteristics such as age at implantation and communication mode would explain some of the inter-individual variability in vocabulary skills. Regarding the effect of age on vocabulary, we were interested in whether the difference between CI users and children with TH would be larger around the time that the children enter school, and what would happen in the years after that. If it is true that children with CI fall behind their TH peers when they enter school, this would be an important consideration for language intervention around this age. If vocabulary growth looks promising in the years leading up to school entry, this might lead clinicians or caregivers to believe that the child is well on track and prematurely reduce support. Finally, understanding the amount of inter-individual variation and how it's related to child-level characteristics can help to understand the scope of the problem, identify children who are at risk, and highlight possible avenues for intervention.

METHODS

In this cross-sectional retrospective study, we compared the receptive vocabulary of 88 young CI users to matched children

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with TH and explored the influence of child-level characteristics on between-group and interindividual variation.

Group of Children With CI

The sample of children with CI was part of a larger cross-sectional data collection to which all Norwegian CI users were invited who had received a CI between 1988 and 2015 and before 18 years of age. Of the eligible Norwegian CI users, 82% (496/606) had participated. For the present study, we selected all 88 participants who were 3 to 16 years old, had severe to profound bilateral hearing loss (i.e., a pure tone average above 80 dB HL) at birth or before 6 months of age, received a CI before age 3, had at least 75 standard points on a measure of nonverbal abilities and no diagnosed developmental disability, and had a primary caregiver using Norwegian spoken or sign language as their first language. The data were collected between 2013 and 2016. Tests were administered at Oslo University Hospital by trained research assistants and additional information was gathered from questionnaires and medical records. Permission for the data collection was granted by the parents and the Regional Committee for Medical and Health Research Ethics.

Child-Level Characteristics of CI Users

Background Information • We collected information about whether children were born prematurely, age at cochlear implantation, type of implantation (simultaneous bilateral or sequential bilateral), etiology of deafness, whether their mother, father, or siblings had a hearing loss, and educational placement.

Maternal education was registered as *less than high school*, *high school*, or *higher education,* based on the mother's self-reported highest level of completed education. Because there was only one mother in each group that had the lowest level of education (no high school education), we used two categories of maternal education for all analyses except the matching, that is, high school or less ("low") and higher education ("high").

Furthermore, hearing age (time since implantation) was calculated for each participant as an indicator of their auditory experience. We used the date of the children's first cochlear implantation as the reference point because information on audibility and amplification before the CI was often not available and for most participants, the benefit that they would have had from using a hearing aid before cochlear implantation was unclear and likely minimal.

Mode of Communication and Speech Intelligibility • Based on parent reports, we coded the children's communication mode at home and school into four categories: *only spoken language, spoken with sign support, spoken language and sign language,* and *only sign language*. We also asked the parents to rate the intelligibility of their child's speech using the Speech Intelligibility Rating scale from Allen et al. (2001).

Social Integration • Parents answered two questions about social interactions on a scale from 0 (*never*) to 10 (*always*): 'How often does your child avoid social settings due to difficulties with hearing?' and 'How often is your child excluded from social settings because of difficulties with hearing?'.

Speech Perception • As a direct measure of speech perception, children were asked to repeat 50 pre-recorded phonetically balanced monosyllabic Norwegian words (Øygarden 2009) presented in an anechoic chamber at 65 dB SPL, and the percentage of correct repetitions was recorded. This test was not done with 13 children. All of these children were younger than 5 years of age (mean age 3.9 years, *SD* = 0.54, range 3.2 to 4.8 years), and the test was likely considered too difficult for them by the test administrator. Notably, 9 children in this age range (range 3.5 to 4.8 years) did complete the speech perception test.

As an indirect measure of speech perception, parents answered four questions from the Speech, Spatial, and Qualities of Hearing Scale (SSQ; Gatehouse & Noble 2004), rating their children's ability to understand speech in individual and group conversation in silent and noisy rooms. Answers were provided on a 10-point scale and averaged. Other questions from the SSQ were not included, because they were not applicable (e.g., "Can the child understand speech when it is driving a car?") or considered too difficult to answer (e.g., "Does the child perceive music and voice as separate objects?").

Nonverbal Abilities • Depending on their age, the children's nonverbal cognitive abilities were assessed with the Block Design subtest of the Wechsler Preschool and Primary Scale of Intelligence-III (3 to 4 years; Wechsler 2002), Raven's Coloured Progressive Matrices (4 to 9 years; Raven 2004), or Raven's Standard Progressive Matrices Plus (9 to 16 years; Raven & Court 2003). Two children were instead tested with the Leiter International Performance Scale-Revised (Roid & Miller 1997) and the Wechsler Intelligence Scale for Children, 4th edition (Wechsler 2003).

Matched Groups of Children With TH

Two groups of children with TH were selected from the Norwegian norm sample of the British Picture Vocabulary Scale, 2nd edition (BPVS-II; Dunn et al. 1997; Lyster et al. 2010) and matched to the CI group with propensity score matching (Ho et al. 2007). The BPVS-II norm sample contains 1,012 typically developing Norwegian children (age 2.3 to 16.3 years; see Table 1). The first TH group was matched on sex, maternal education, and chronological age. The second TH group was matched on sex, maternal education, and hearing age (i.e., they were younger than the CI users but had a similar amount of auditory experience; Table 1). Children with TH that were included in the first matched group were excluded from the matching procedure for the second group. For two children with CI, information about maternal education was missing and was imputed with predictive mean matching based on their age and BPVS-II standard score (they were assigned to the high school and higher education categories, respectively). Children with TH for which information about maternal education was missing (261/1,011, 26%) and one child with TH who had an extremely low BPVS-II score were not used in the matching.

Outcome Measure: Receptive Vocabulary Knowledge (BPVS-II)

As a measure of receptive vocabulary, we used the Norwegian version of the BPVS-II. The test was conducted in spoken Norwegian. For the presentation of the instructions, sign language support was offered to the CI users but none of them requested it. We obtained age-referenced standard scores for all children. In addition, we obtained hearing-age-referenced standard scores for the CI users, that is, scores that take into account the children's lack of auditory experience. These were based on the years since the implantation of the first CI instead of the children's chronological age.

TABLE 1. Age, sex, parental education and receptive vocabulary scores measured with the BPVS-II by group

	CI	TH, Matched by Age	TH, Matched by Hearing Age	BPVS-II Norm Sample*
n	88	88	88	1,011
Age				
Mean (SD), mo	104.8 (45.8)	105.0 (47.4)	96.0 (37.8)	104.9 (43.8)
Range, mo	38–191	40–192	31–158	27–196
Sex, n (%)				
Female	43 (49%)	43 (49%)	38 (43%)	510 (50%)
Male	45 (51%)	54 (51%)	50 (57%)	501 (50%)
Education mother, n (%)				
Missing	2	—	—	261
Less than high school	1 (1%)	1 (1%)	1 (1%)	31 (4%)
High school	26 (30%)	27 (31%)	20 (23%)	442 (59%)
Higher education	59 (69%)	60 (68%)	67 (76%)	277 (37%)
BPVS-II standard scores				
Mean (SD)	84.6 (21.1)	102.1 (15.8)	—	100.4 (15.1)
Range	0–125.8	67.1–140.1	—	58.6–152.4
≥1 SD below norm, n (%)	40 (45%)	18 (20%)	—	177 (17%)
≥2 SD below norm, n (%)	23 (26%)	2 (2%)	—	22 (2%)
Residual SD model 1	20.1	15.4	—	—
BPVS-II standard scores, hearing age referenced				
Mean (SD)	102.8 (22.2)	—	100.8 (16.7)	—
Range	29.6–174.6	—	63.6–142.2	—
≥1 SD below norm, n (%)	20 (23%)	—	17 (19%)	—
≥2 SD below norm, n (%)	5 (6%)	—	4 (5%)	—
Residual SD model 2	19.3	—	16.8	—

Note. Percentages exclude incomplete data and may not add up to 100% due to rounding.
**Raw data from Lyster et al. (2010).*
BPVS-II, British Picture Vocabulary Scale, 2nd edition.

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Recalculation of BPVS-II Standard Scores • The official Norwegian norm tables of the BPVS-II (Lyster et al. 2010) only provide scaled scores that are age-corrected but not normalized, and there were no corresponding scores for the extremely low raw scores of 16 children with CI (18%; mean age 10.3 years; range 3.6 to 14.8 years). To avoid working with left-censored data and norm scores with an unknown distribution, we used the raw data from the original Norwegian BPVS-II norm sample (n = 1,011; one outlier was excluded) to estimate age-related quantiles of the BPVS-II raw scores and calculate standard scores for all participants of our study. We used the method for centile estimation described in Stasinopoulos et al. (2020, ch. 13) to select and fit a model with age-dependent parameters for the location, scale, and shape of the distribution of raw scores (specifically, a Box-Cox power exponential distribution; Rigby & Stasinopoulos 2004). The quantiles from this model are displayed as gray lines in Figure 1. This is a rather flexible model but to obtain unbiased quantiles and calculate valid standard scores we found it crucial to accurately capture the distribution of raw scores. Model comparisons based on the Generalized

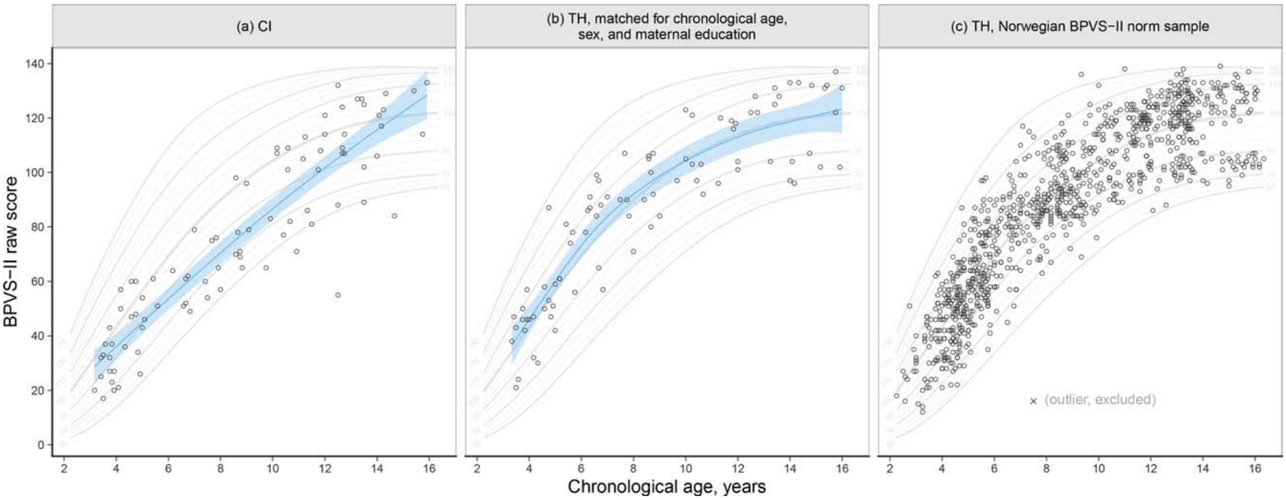

Fig. 1. Raw scores of CI users (n = 88), children with TH, matched for age, sex, and maternal education (n = 88), and the Norwegian norm sample (n = 1,012) on the British Picture Vocabulary Scale, 2nd edition (BPVS-II) receptive vocabulary test (Lyster et al. 2010). The blue line and ribbon show predicted raw scores and 95% confidence intervals from a general additive model (Table 2 Model 3). Gray lines in the background indicate the corresponding modeled standard scores based on the norm sample of children with TH (i.e., mean 100, SD 15) from 55 (−3 SD) to 145 (+3 SD) standard points in 5-point intervals. One outlier in the norm sample was excluded from the analyses. CI indicates cochlear implant; TH, typical hearing.

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TABLE 2. Generalized additive models predicting scores on the BPVS-II receptive vocabulary test

Parametric Coefficients	Estimate	Standard Error	t	*p*
Model 1. BPVS-II standard scores, CI vs TH matched for sex, parental education, and age				
(Intercept)	102.03	3.04	33.53	<0.001
Group, CI vs. TH	–17.33	2.74	–6.33	<0.001
Sex, female vs. male	–5.20	2.76	–1.88	0.06
Maternal education, high vs. low	3.83	2.96	–1.29	0.20
Smooth terms	Estimated df	Reference df	F	*p*
Age in group CI	2.17	29	0.28	0.015
Age in group TH	0.003	29	<0.001	0.41
R^2_{adj}	22.2%			
Model 2. BPVS-II standard scores, CI vs TH matched for sex, parental education, and hearing age				
(Intercept)	97.36	3.24	33.01	<0.001
Group, CI vs TH	1.10	2.79	0.39	0.70
Sex, female vs. male	–1.17	2.82	–0.42	0.68
Maternal education, high vs. low	5.21	3.14	1.66	0.10
Smooth terms	Estimated df	Reference df	F	*p*
Hearing age in CI group	2.69	29	0.93	<0.001
Hearing age in TH group	<0.001	29	<0.001	0.70
R^2_{adj}	12.8%			
Model 3. BPVS-II raw scores, CI vs. TH matched for sex, parental education, and age				
(Intercept)	87.43	1.41	62.18	<0.001
Group, CI vs. TH	–12.5	1.98	–6.32	<0.001
Smooth terms	Estimated df	Reference df	F	*p*
Age in CI group	1.80	9	52.7	<0.001
Age in TH group	3.17	9	44.2	<0.001
R^2_{adj}	83.9%			

Note. Models 1 and 4 predict BPVS-II standard scores referenced by age; Model 2 predicts scores referenced by hearing age. Age and hearing age were mean centered. Models were fitted using restricted maximum likelihood. Smooth terms use thin plate regression splines (tprs) with additional penalization of the null space, nonlinear interactions use tprs tensor products. Nonlinear age effects were modeled independently per group. df = degrees of freedom. Estimated df reflect the complexity of the smooth terms. Roughly, 0 indicates horizontal line, that is, no association, 1 indicates a linear trend, 2 a quadratic trend, 3 a cubic trend.

BPVS-II, British Picture Vocabulary Scale, 2nd edition; CI, cochlear implant; TH, typical hearing.

Akaike Information Criterion and visual diagnostics indicated that this model did so better than simpler models. The recalculated standard scores are on an age-referenced normalized scale that has a mean of 100 and a standard deviation of 15 in the norm group of Norwegian children with TH.

Group Differences in Receptive Vocabulary

We performed two group comparisons of receptive vocabulary performance: CI users were compared to children with TH using matching and standard scores based on (1) their chronological age and (2) their hearing age. The group's mean BPVS-II standard scores were compared with two-sided Welch *t*-tests for unequal variances. Effect sizes were assessed with Hedges's *g*. Risk ratios were used to compare proportions of children performing below the normative range, that is, children with a standard score below 85, which corresponds to 1 *SD* below the age-referenced normative mean. Group differences in variability were assessed with bootstrapped 95% confidence intervals (95% CIs) of *SD* ratios.

To control for the effects of the matching variables (age/hearing age, sex, and maternal education) on receptive vocabulary and analyze age trends in the BPVS-II raw scores, we used generalized additive models. This type of model was chosen because, based on the previous literature, we expected a nonlinear effect of age on the group difference. For the two CI users for which maternal education was missing, the imputed values from the matching procedure were used. Our predetermined significance level for all of these statistical analyses was 0.05.

Effect of Child-Level Characteristics on CI Users' Vocabulary

In an additional analysis that included only the CI users, we investigated associations between child-level characteristics and receptive vocabulary with random forests (Strobl et al. 2009). This nonparametric model does not predetermine relations between variables. It can capture nonlinear and unexpected associations and is robust against multicollinearity. Random forests group similar observations using decision trees, which recursively split the sample along one of the predictor variables into two increasingly homogenous branches. The outcome for a given observation can be predicted by averaging the outcomes of all observations in the same branch (see Fig. 4 for an example). To make the results more robust, the random forest averages predictions from multiple trees, whereby each tree is based on slightly perturbed data and a random selection of predictors at each split. The importance of predictors can be determined by how often they act as a decisive splitting variable across all trees in the random forest. When a random permutation of a predictor's values improves the predictions, the predictor's importance can become negative, meaning that it is not associated with the outcome.

We used a random forest with 3,000 trees, a random selection of 6 predictors to be considered at each split, a minimum sum of weights of 5 required for a node to be split further, and no restrictions on the depth of the tree. These hyperparameters were chosen through 5-fold bootstrap cross-validation with resamples stratified for age. However, the final model was trained

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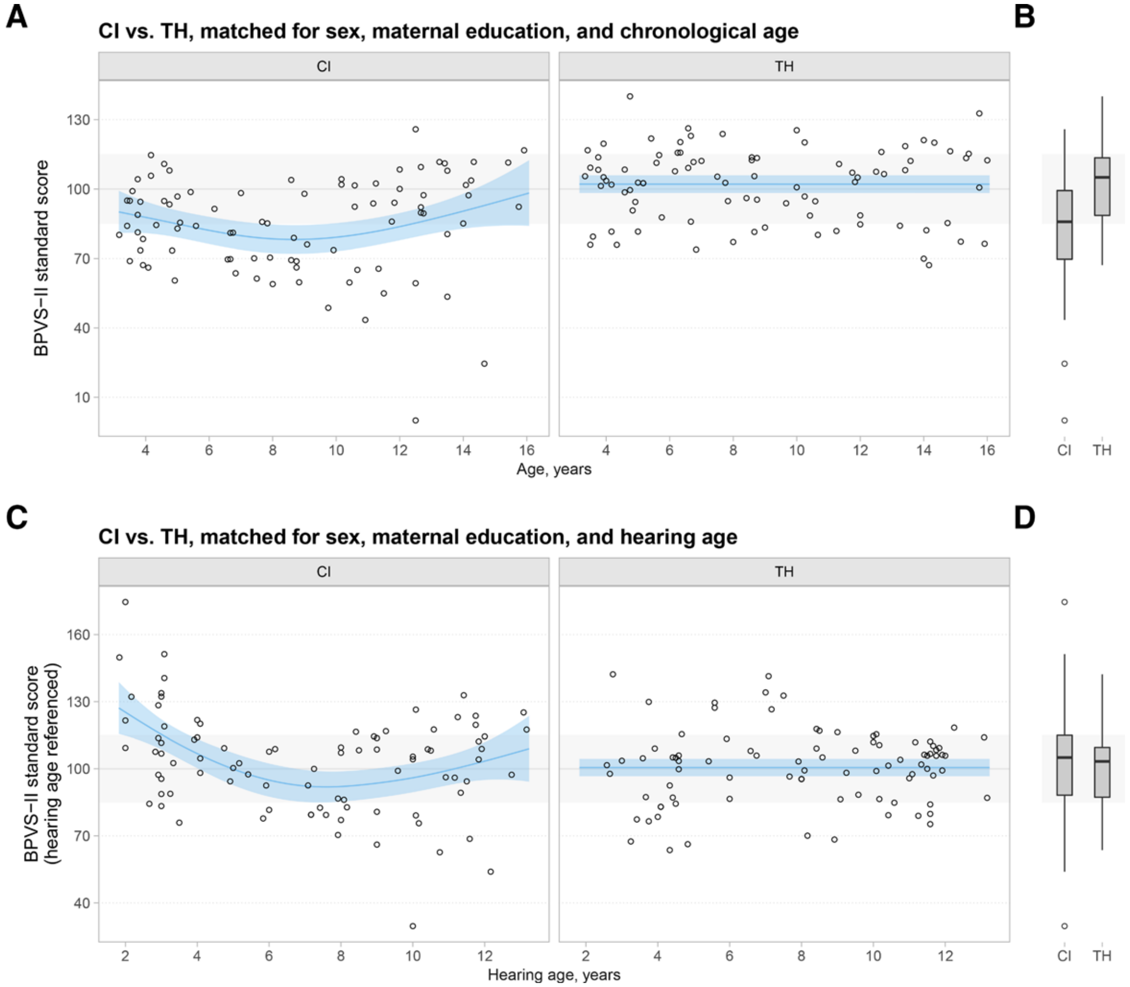

Fig. 2. Standard scores of children with CIs and age-matched children with TH on the British Picture Vocabulary Scale, 2nd edition (BPVS-II) receptive vocabulary test. The blue line and ribbon show the predicted marginal mean and 95% confidence interval (Table 2 Models 1 and 2). The gray ribbon shows the normative mean ± 1 *SD*. BPVS-II scores by sex and maternal education were not significantly different and are not shown. Hearing age = time since implantation for CI group, chronological age for TH group. CI indicates cochlear implant; TH, typical hearing.

and evaluated on the entire sample because the small dataset did not allow train-test-validation of the random forest model.

The predictors that were used in the random forest were: chronological age, hearing age, age at implantation, sex, premature birth (yes/no), family history of hearing loss (yes/no), implantation type (sequential or simultaneous), speech perception (word repetition and parent-reported), nonverbal cognitive abilities, social integration (two questions), maternal education (low versus high), and communication mode at home and school (both with 4 levels). If a participants' data for a predictor was missing at any given split, they were assigned to the majority branch.

Statistical Software

All analyses were performed with R 4.0.3 (R Core Team, 2020), using the package *mice* (van Buuren & Groothuis-Oudshoorn 2011) for imputations with predictive mean matching, *matchit* (Ho et al. 2011) for propensity score matching of children with TH to the CI users, *gamlss* (Rigby & Stasinopoulos 2005) to estimate quantiles of the BPVS-II raw scores in the TH norm sample and calculate standard scores, *mgcv* (Wood 2017) to fit generalized additive models for the group comparisons, and *partykit* (Hothorn & Zeileis 2015) for the random forest analysis.

RESULTS

Sample Characteristics

The sample included 88 children with CIs and 2 matched groups of children with TH. Of the 88 CI users, 82 (93%) had bilateral severe to profound hearing loss at birth, and 6 (7%) had become deaf before 6 months of age—three due to Meningitis, one due to auditory neuropathy disorder, and for two the cause of deafness was unknown. For the largest group of the CI users (*n* = 32, 37%), the etiology of deafness was unclear. The other children had a hearing loss due to Connexin 26 mutation (*n* = 27, 30%), CMV infection (*n* = 6, 7%), meningitis (*n* = 3, 3%), auditory neuropathy (*n* = 2, 2%), birth injury (*n* = 5, 6%), Jervell-Lange-Nielsen-Syndrome (*n* = 5, 6%), or other syndromes such as Pendred and Waardenburg (*n* = 8, 9%). Fourteen of the CI users were born prematurely – two of them very prematurely, before 28 and 32 weeks of gestational age, respectively.

All children with CIs had bilateral implants, but one child used only one CI because they did not get any speech perception benefit from their second CI, which was implanted relatively late, at 7 years of age. The average age at implantation of the first CI was 17.8 months (*SD* = 8.57, range 5 to 36 months) and the average hearing age was 7.1 years (*SD* = 3.5;

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Age at implantation
mean 17.8, SD 8.6; median 17, IQR 14.7; range 5–36

Type of implantation
simultaneous: 58/88 (66%), sequential: 30/88 (34%)

Premature birth
no: 74/88 (84%), yes: 14/88 (16%)

Nonverbal abilities
mean 98.1, SD 12.4; median 95, IQR 15.8; range 75–126

Speech perception (word repetition)
mean 88.3, SD 7.9; median 90, IQR 10.0; range 66–100

Speech perception (parent report)
mean 7.2, SD 1.5; median 7, IQR 1.8; range 3–10

Hearing loss in family
no: 68/87 (78%), yes: 19/87 (22%)

Communication mode at home
a: 66/87 (76%), b: 15/87 (17%), c: 4/87 (5%), d: 2/87 (2%)

Communication mode at school
a: 60/86 (70%), b: 18/86 (21%), c: 7/86 (8%), d: 1/86 (1%)

Social integration (Excluded...)
mean 1.7, SD 2.1; median 1, IQR 3.0; range 0–8

Social integration (Avoids...)
mean 2.7, SD 2.4; median 2, IQR 4.0; range 0–9

Sex
male: 45/88 (51%), female: 43/88 (49%)

Fig. 3. Standard scores on the British Picture Vocabulary Scale, 2nd edition (BPVS-II) receptive vocabulary test as a function of child-level characteristics of children with CIs. For associations with other characteristics see also Figure 1 and Table 1. In panels of communication mode and social integration, points were jittered horizontally to reduce overplotting. The gray ribbon shows the normative mean ± 1 *SD*. The number of missing data points for each child-level characteristic (if any) are indicated in the respective panel. CI indicates cochlear implant.

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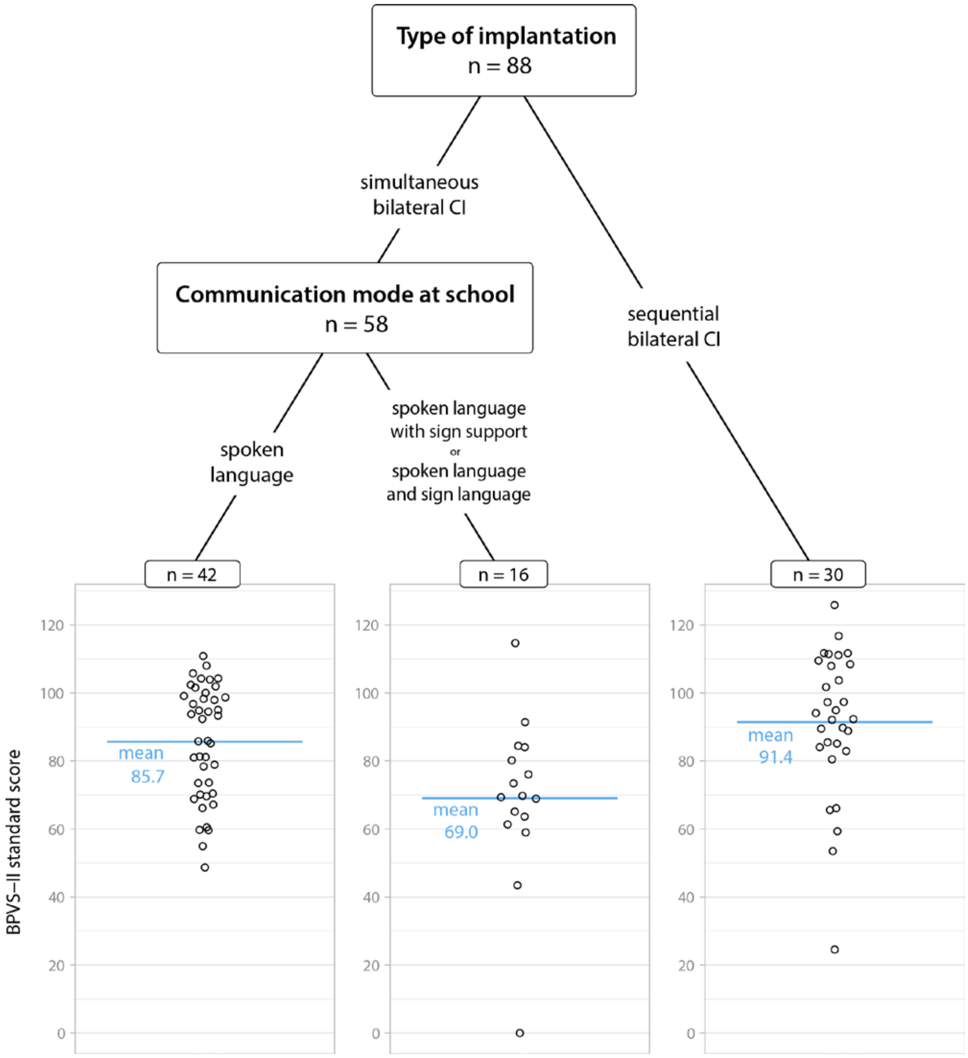

Fig. 4. Example of a decision tree from the random forest. The nodes represent predictor variables in the tree and the number of participants in the respective branch of the tree. The labels on the branches indicate the value of the predictor variable that a participant must have to be sorted into the respective branch. Participants with missing data for a given predictor are sorted into the majority branch. CI indicates cochlear implant.

median = 7,5, *IQR* = 6.6; range 1.8 to 13.2 years). Thirty children (34%) had sequential CI surgeries, with an average inter-implant interval of 32 months (*SD* = 18, range 2 to 61 months). Due to this study's age range and cross-sectional design, all selected participants had received their CI between 2000 and 2014. Fifty-eight children were born before universal neonatal hearing screening was introduced in Norway in 2008. Their median age at diagnosis of deafness was 10 months (IQR = 11.3; range 0 to 30 months; 4 missing).

According to their parents, 85 children (97%) used their CIs during all waking hours and 2 (2%) used them for most waking hours with some breaks (1 missing). On the Speech Intelligibility Rating (Allen et al. 2001), 77 children (88%) were rated by their parents as easily intelligible, 4 (5%) were rated as intelligible to concentrated listeners who lip read, and 5 (6%) as unintelligible (2 missing). Seventy-nine children (90%) were in mainstream education, with 4 (5%) of them additionally attending special schools or groups for children with hearing loss. Seven children (8%) exclusively attended special education, and two 3-year olds were not in any educational program.

The CI users and the two groups of matched children with TH were very similar concerning the matching variables, that is, age (respectively hearing age), sex, and maternal education (see Table 1). For additional details on child-level characteristics, see Table 1 and Figures 2, 3.

Comparisons to Children With TH Matched by Chronological Age

We compared differences in receptive vocabulary using the children's age-referenced standard scores of the BPVS-II. Overall, the CI users' receptive vocabulary was significantly poorer than that of children with TH who were matched for chronological age, sex, and maternal education (CI users: $M = 84.6$ standard points, $SD = 21.1$; TH: $M = 102.1$ standard points, $SD = 15.8$; mean difference −17.5 standard points, 95% CI [−23.0 to −12.0], $p < 0.001$; Hedges's $g = 0.94$, 95% CI [−0.62 to 1.24]), and their risk of performing more than 1 *SD* below the normative mean was 2.2 times higher (95% CI [1.42 to 3.83]; Table 1, Fig. 2A).

However, when modeling the effects of chronological age, sex, and maternal education on receptive vocabulary with a generalized

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additive model, we found a significant nonlinear effect of age in the CI group. This effect was U-shaped, such that the CI users' average score was relatively high and within the normative range at the start and end of the age range (3 and 16 years, respectively) but more than 1 *SD* below the normative mean between 5.0 and 12.3 years of age. The lowest scores were predicted around 8.7 years of age, with a predicted average of 78.2 standard points and a difference to the TH group of −23.9 standard points. In the TH group, there was no significant age effect and the average score was near the normative mean across the age range. As a result, the groups had different mean BPVS-II standard scores at some ages but not at other ages (Fig. 2A, Table 2 Model 1).

Notably, from around 13 years of age until the end of the age range (i.e., 16 years; Fig. 1C), the average BPVS-II raw score in the TH group was plateauing. Sixteen is also the end of the age range for which the BPVS-II has been adapted to Norwegian, thus, this plateau might be a ceiling effect caused by a lack of more difficult items. In contrast, the average raw scores of the CI group increased linearly, with no clear evidence of a flattening of the slope (Fig. 1A, Table 2 Model 3).

The variability of receptive vocabulary performance was not significantly different between the groups, neither for the unconditional scores (*SD* ratio 1.34, 95% CI [0.98 to 1.63]), nor when age, sex, and maternal education were accounted for by the generalized additive model (residual *SD* ratio 1.29 (95% CI [0.91 to 1.61]).

Comparison to Children With TH Matched by Hearing Age

Next, we compared receptive vocabulary performance taking into account the CI group's limited auditory experience. That is, the BPVS-II standard scores were based on hearing age (i.e., time since implantation) instead of chronological age. Moreover, the matched group of children with TH were younger but had similar amounts of auditory experience.

Overall, CI users did not perform significantly different from hearing-age matched children with TH (CI users: M = 102.8 standard points, SD = 22.2; TH: M = 100.8 standard points, SD = 16.7; mean difference 2.0 standard points, 95% CI [−7.8 to 3.9], $p = 0.505$) and did not have a significantly higher risk of performing 1 *SD* or more below the normative mean (risk ratio 1.18, 95% CI [0.64 to 2.18]; Table 1, Fig. 2C). As with the age-referenced scores, the generalized additive model indicated a significant U-shaped effect of hearing age in the CI group. The predicted mean hearing-age-referenced standard scores of the CI group were lowest around 7.6 years of hearing age, with a predicted mean score of 91.9 standard points. Even at the lowest point, the predicted mean of the CI group was within the normative range and it was above the normative mean at the low end of the age range. Again, the TH group performed close to the normative mean across the age range. As a result, the groups had different means at some ages but not at others (Fig. 2C, Table 2 Model 2).

The variability in BPVS-II scores was not significantly different between groups, neither for the unconditional scores (*SD* ratio 1.33, 95% CI [0.97 to 1.62]) nor when the three covariates were accounted for by the model (residual *SD* ratio 1.15, 95% CI [0.85 to 1.40]).

Effects of Child-Level Characteristics on CI Users' Receptive Vocabulary Scores

We used a non-parametric random forest model to detect associations between child-level characteristics and receptive vocabulary of children with CI. Figure 3 shows the 12 predictors that were entered into this analysis in addition to chronological age and hearing age (see Fig. 2).

Overall, the random forest explained 26% of the variance in CI users' BPVS-II standard scores. However, it should be noted that the random forest model is relatively flexible and was trained and tested on the full sample, thus it may be prone to overfitting. That is, the variance that this model would explain in unseen data would certainly be smaller.

Five variables were found to be important predictors of the children's receptive vocabulary skills, that is, they consistently helped to improve predictions of the children's receptive vocabulary performance across the trees in the random forest. Ordered from most to least important, these were type of implantation (i.e., whether the child had simultaneous or sequential bilateral implantation), communication mode at school, the parent-reported frequency with which the child avoids social situations because of their hearing loss, chronological age, and hearing age. Other child-level characteristics were not found to be important predictors in the random forest, that is, they did not consistently help to improve predictions. Notably, a sensitivity analysis with different hyperparameters (i.e., different rules for the constructions of the trees in the random forest) showed that there were differences in the consistency with which the predictors were found to be important. Specifically, type of implantation, communication mode at school, and social integration were usually among the important predictors, while the others were not and some of the other predictors (e.g., nonverbal cognitive abilities and age at implantation) did appear to be important when other model configurations were used, which may be a sign of collinearity between them or indicate that these child-level characteristics affect receptive vocabulary mainly through interactions with other factors. The predictions from the random forest are robust against collinearity like this, but it makes it difficult to interpret the roles of the individual predictors.

Descriptively, children in our sample who had received CI sequentially scored higher (M = 91.4, SD = 21.4) than children who had received their implants simultaneously (M = 81.1, SD = 20.2). Children who, according to their parents, exclusively used spoken language at school performed better on the BPVS-II (M = 86.3, SD = 19.2) than those who used spoken language with sign support (M = 82.3, SD = 16.1) or spoken language and sign language (M = 66.1, SD = 36.1). There was only one child who exclusively used sign language. Interestingly, they scored exceptionally well (BPVS-II standard score 126). The parent-reported frequency with which the children avoid social situations because of their hearing loss was negatively associated with their vocabulary scores. That is, children who more often avoided social situations tended to have lower scores (Kendall's τ −0.10). The nonlinear effects of chronological age and hearing age on receptive vocabulary scores have already been described above. Notably, these cannot be disentangled from the effects of implantation type and age at implantation because, perhaps because of changes in clinical practice, younger children had received their CIs at a significantly earlier age (Kendall's τ 0.40, 95% CI [.28, 51]) and children with simultaneous CIs were significantly younger (mean age, 7.3 years, SD = 2.77) than children with sequential CIs (mean age 11.6 years, SD = 4.0), Welch *t*-test, mean difference 4.4 years, 95% CI [2.7 to 6.0], $p < 0.001$.

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DISCUSSION

This study investigated the receptive vocabulary of children with bilateral CI who were 3 to 16 years old. First, the children were compared with matched children with TH. Then, the influence of child-level characteristics on interindividual variability was explored.

Age-Related Differences in Receptive Vocabulary

In line with previous reports (Lund 2016; Välimaa et al. 2018), the receptive vocabulary of children with CIs was overall poorer than that of matched children with TH, and the majority scored below the normative range. However, the size of the gap between children with CI and children with TH varied strongly with age. This was caused by a nonlinear effect of age on vocabulary scores of CI users. Specifically, the gap in vocabulary knowledge increased from 12.0 standard points at 3 years of age, to 23.9 standard points at 8.7 years of age and then narrowed again to 4.1 standard points at 16 years of age. The strong nonlinear effect of age on the vocabulary gap might explain the discord in the literature regarding age-appropriateness, trajectory, and variability of CI users' vocabulary skills because it suggests that these depend on the age range under investigation.

Interestingly, CI users performed comparably to TH children matched by hearing age, showing the effect of delayed auditory input on the language development of children with CIs. Although hearing age might be a fairer grounds for comparison, large developmental delays (in our sample, the average age at implantation was 17.8 months) put children at risk for long-term problems with, for instance, psychosocial development and educational attainment (Forrest et al., 2018; Johnson et al., 2010; Wong et al., 2018). It should be noted that the effect of auditory experience is also mediated by the children's daily CI use, which varies substantially between children (Busch et al., 2017; Busch et al., 2020). All families in this study reported full-time CI use. However, parents often over-report their children's CI use. A more reliable measure, such as data logs, might provide further insights (Walker et al., 2013).

Why Was the Age-Effect U-Shaped?

It is unclear why the gap between children with CI and children with TH was larger at 8.7 years of age than at the start and end of the age range (3 and 16 years, respectively). Perhaps, the gap grows in the years after implantation because it becomes more difficult for CI users to acquire new vocabulary. This might be due to increasing abstractness and decreasing frequency of advanced vocabulary (Hansen, 2017). For example, when words occur less frequently, a stable phonemic representation must be acquired through fewer encounters. As a result, the CI users' poor access to acoustical information becomes increasingly disadvantageous. It is also possible that the school environment—which children enter around six years of age in Norway—is more challenging for children with hearing impairments than for children with TH (Chute & Nevins 2003; Vermeulen et al. 2012). Settling in new educational environments is challenging for children with disabilities (Ravenscroft et al. 2017). Children with hearing impairments struggle with classroom acoustics (Iglehart 2016; Neuman et al. 2012), social integration (Bat-Chava & Deignan 2001; Fitzpatrick & Olds 2015), and literacy development (Johnson & Goswami 2010). Moreover, CI users who are placed in special education classrooms might receive different, potentially poorer, spoken language stimulation than their mainstreamed peers.

Our cross-sectional sample cannot reveal the causal origins of the increasing gap between children with CI and children with TH. However, our results match those of a longitudinal study from our group, in which the receptive vocabulary of early-implanted children with CIs equaled that of their TH peers at first, but began to fall behind 4 to 6 years after implantation (Wie et al. 2020), which is around the same time where we observed the largest vocabulary deficit in the current study.

Notably, the vocabulary gap in the current study narrowed again toward 16 years of age. Perhaps, CI users eventually gain a foothold when they settle in appropriate educational environments, or their language problems are noticed and addressed—both would underline the importance and potential benefit of early identification and intervention. U-shaped effects are a common phenomenon in child development, and it has been hypothesized that they indicate a shift toward more systematic processing strategies that cause a temporary loss in processing efficiency (Pauls et al. 2013). Age-related differences in lexical processing strategies between children with CI and children with TH have been reported before (Löfkvist et al. 2012). Thus, it is conceivable that the increasing vocabulary gap we observed is the result of the CI users going through such a shift at a later time than the TH children and that the successful acquisition of the new lexical processing strategies eventually allows the CI users to catch up with their TH peers, thus closing the gap. It is also conceivable that between-group differences in nonverbal abilities contributed to the differences in language development. While all children were within the normative range on a standard test of nonverbal cognitive abilities, detailed information on nonverbal abilities was not available for the children with TH so that it was not possible to match the groups on this variable.

A very different explanation for the closing vocabulary gap is that the Norwegian BPVS-II evaluates the receptive vocabulary skills of teenagers with CI too optimistically because children with TH reach ceiling-level performance toward the end of the age range. A comparison of the raw scores suggests this: The modeled mean of TH children's raw scores begins to plateau around 13 years of age and deceleration of the increase in raw scores is visible even before that, in both the matched group and the full TH norm sample (Figs. 1B, C). In contrast, the average raw score of the children with CI increases linearly, with no sign of a plateau (Fig. 1A).

Perhaps, a vocabulary test that better discriminates between high-performing children in that age range would reveal that children with CIs keep lagging behind their TH peers. Furthermore, a test that covers different aspects of lexical development, such as lexical breadth or lexical processing strategies (Löfkvist et al. 2012; Nation 2014), might show differences that the BPVS-II is not sensitive enough to detect. The language problems of older children with CI might also be more prominent in tests of higher-level language abilities (Boons et al. 2013b). More generally, one language measure alone may not represent the language abilities of children with CI well. Although children's development in different aspects of language is usually highly correlated (Tomblin 2019; Tomblin & Zhang 2006), using multiple measures of a hypothetical construct is always preferable. Moreover, there is evidence for subgroups of children with CI whose performance shows discrepancies across language domains (de Hoog et al. 2016; Duchesne et al. 2009).

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Regardless of the aspect of language that they assess, standardized language tests might overestimate CI users' performance under realistic listening conditions, which often pose additional acoustical, cognitive, and social challenges (Chute & Nevins 2003; Punch & Hyde 2011; Zaltz et al. 2020). The uncertainty around the interpretation of the BPVS-II scores shows the importance of choosing the right assessment material in research and practice and highlights a limitation of our study, namely the use of just a single language measure. Unfortunately, the BPVS-II was the only test where a large sample of matched TH controls was available to us.

Impact of Child-Level Characteristics on Interindividual Variation

The interindividual variability in CI users' language abilities is often said to be wide (e.g., van Wieringen & Wouters 2015). In our sample, the variability in the receptive vocabulary skills of CI users exceeded the variability in the TH group by about a third, but this difference was not statistically significant. Notably, this was despite the relatively wide range of implantation ages in our sample, which is known to contribute substantially to variability in outcomes (Dettman et al. 2016; Karltorp et al. 2020).

Some child-level characteristics explained inter-individual variation in CI users' receptive vocabulary performance. Besides the effects of age and hearing age discussed above, we found associations with implantation type (simultaneous versus sequential), communication mode at school, and one parent-reported measure of social integration ("How often does the child avoid social situations due to their hearing loss?").

Surprisingly, children in our sample who had received their CIs sequentially performed better than those who had received them simultaneously. In previous studies, longer inter-implant periods have been found to affect outcomes negatively (Boons et al. 2012b; Gordon & Papsin 2009). Thus, one might expect a negative effect of sequential implantation. However, likely due to changes in clinical practice, sequentially implanted children in our sample were significantly older than simultaneously implanted children and might therefore have benefitted from the ceiling effect in the TH group's test scores or the environmental changes discussed above. Unfortunately, our cross-sectional sample does not allow us to disentangle the effects of age and implantation type. Another explanation for the better performance of sequentially implanted children could be that children with severe bilateral hearing loss typically receive CIs simultaneously, whereas children with sequential implantations might have had a relatively good residual hearing in the other ear and thus more access to auditory input even before the implantation. The negative effect of longer inter-implant intervals could have been further mitigated through bimodal stimulation, that is, by using a hearing aid with the non-implanted ear between implantations. Under some circumstances, this might even improve outcomes (Davidson et al. 2019; Wenrich et al. 2019). Unfortunately, reliable information on residual hearing and amplification before or between implantations was not available.

Communication mode at school was associated with receptive vocabulary while communication mode at home was not. Children in our sample who exclusively used spoken language at school performed better than children who used spoken language with sign support or spoken language and sign language in conjunction. One explanation for the association between communication mode at school and receptive vocabulary is that it reflects the type of educational setting that the child is in, which is often chosen based on the children's language abilities. Thus, increased use of sign support or sign language in a child's school might indicate that they receive some form of special needs education and might therefore reflect existing problems with spoken language development rather than be a cause of such problems. The lack of an effect of communication mode at home might be related to the relatively small number of children who used non-verbal communication modes: Most children were exclusively using spoken language at home (n = 66, 74%) and school (n = 60, 67%), and no child exclusively used sign language in both contexts. Overall, around 90% of the sample used spoken language or spoken language with sign support. In addition, communication mode was reported by the parents and may therefore not be completely accurate.

We also found an association of vocabulary skills with social integration, particularly, with the frequency with which children avoid social situations due to their hearing loss. In our sample, children who did so less had a larger receptive vocabulary. It is unclear whether the children avoided social situations because they struggled with language or whether their avoidance of social situations caused language problems to aggravate. However, previous studies have argued that language and psychosocial problems in children with hearing loss are interconnected (Boerrigter et al. 2019; Hoffman et al. 2015; Wong et al. 2018). Notably, social integration was assessed through parent reports. Depending on the parents' relationship with the child and their own expectations and worries, they may not always evaluate the child's social integration realistically. Previous studies have found differences between the perspectives of children with CIs and their parents regarding aspects of the children's quality of life (Haukedal et al. 2020). Nevertheless, the question appears to reflect something about the children's social behavior that is associated with their vocabulary development.

Interestingly, we found no evidence for an effect of nonverbal abilities or speech perception on the receptive vocabulary of CI users. These factors were associated with vocabulary skills in other studies (Geers et al. 2017; Niparko et al. 2010; Thomas & Zwolan 2019). There is also no doubt that nonverbal abilities and speech perception play a role in vocabulary development (Cejas et al. 2018; Davidson et al. 2014; Zaltz et al. 2020). Better speech perception increases opportunities to pick up language (Boderé & Jaspaert 2017; Davidson et al. 2014) and, conversely, a robust vocabulary and good cognitive abilities facilitate speech perception (Klein et al. 2017; McCreery et al. 2019). An explanation for the lack of an effect in our study might be that all children had nonverbal abilities within the normative range and overall speech perception performance was high. Moreover, speech perception was only measured through parent reports, which may not be accurate enough, and with a monosyllable speech perception test in quiet, which was not performed with most of the younger children. This might have further masked an association between speech perception and vocabulary development. It is also possible that these factors influence vocabulary development mainly in interactions with other variables so that their direct effect is less prominent.

Maternal education (as a proxy for socioeconomic status) was also not predictive of vocabulary skills, perhaps because of the low variability in this variable, or because in the Norwegian context, children's access to high-quality education and

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healthcare is relatively independent of their family background (Isungset et al. 2021).

Limitations

It seems likely that the relevance of some child-level characteristics for vocabulary development was obscured by the homogeneity and small size of our sample. To some extent, this was inevitable given the population under study—children with CIs in Norway. With a participation rate of 82% in the original data collection, our sample arguably represents this population well. However, some selection effects might be present and information about the CI users who refused to participate was not available. Generalizability is also limited by the cross-sectional nature of the study, which resulted in strong associations between many of the predictors and limits the interpretability of the age-related differences in vocabulary performance between children with CI and children with TH.

While our findings might not generalize to countries with very different health care and educational systems, it provides an insight into children's vocabulary development in a context where socioeconomic factors are less relevant. Furthermore, children who receive early bilateral CIs, a focus on oral communication, and mainstream education – like the ones in our sample – make up an ever-growing part of the population of children with CI worldwide.

CONCLUSIONS

We found that deficits in receptive vocabulary were common among children with CIs. The deficits were particularly large in the first years of primary school. Therefore, supporting and monitoring the language development of children with CI is vital, especially around the time when they enter school, which is when they seem to fall behind their peers with TH. Age-appropriate vocabulary in the first years after implantation does not guarantee continuously good performance. Fortunately, some predictors of vocabulary skills might be susceptible to intervention, including communication mode (Costa et al. 2019; Kaipa & Danser 2016) and social integration (Nicastri et al., 2021). Understanding the vocabulary development of CI users, risks and protective factors can help decide when and which support is needed.

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