Early vocabulary development in children with bilateral cochlear implants

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predictors.

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Abstract

Background: Children with unilateral cochlear implants (CIs) may have delayed vocabulary development for an extended period after implantation. Bilateral cochlear implantation is reported to be associated with improved sound localization and enhanced speech perception in noise. This study proposed that bilateral implantation might also promote early vocabulary development. Knowledge regarding vocabulary growth and composition in children with bilateral CIs and factors associated with it may lead to improvements in the content of early speech and language intervention and family counselling.

Aims: The aims were to analyse the growth of early vocabulary and its composition during the first year after CI activation and to investigate factors associated with vocabulary growth.

Methods & Procedures: The participants were 20 children with bilateral CIs (12 boys; 8 girls; mean age at CI activation 12.9 months). Vocabulary size was assessed with the Finnish version of the MacArthur Communicative Development Inventories (CDI) Infant Form and compared with normative data. Vocabulary composition was analysed in relation to vocabulary size. Growth curve modelling was implemented using a linear mixed model to analyse the effects of the following variables on early vocabulary growth: time, gender, maternal education, residual hearing with hearing aids, age at first hearing aid fitting and age at CI activation.

Outcomes & Results: Despite clear vocabulary growth over time, children with bilateral CIs lagged behind their age-norms in receptive vocabulary during the first 12 months after CI activation. In expressive vocabulary, 35% of the children were able to catch up with their age-norms, but 55% of the children lagged behind them. In receptive and expressive vocabularies of 1–20 words, analysis of different semantic categories indicated that social terms constituted the highest proportion. Nouns constituted the highest proportion in vocabularies of 101–400 words. The proportion of verbs remained below 20% and the
proportion of function words and adjectives remained below 10% in the vocabularies of 1–400 words. There was a significant main effect of time, gender, maternal education and residual hearing with hearing aids before implantation on early receptive vocabulary growth. Time and residual hearing with hearing aids had a significant main effect also on expressive vocabulary growth.

**Conclusions & Implications:** Vocabulary development of children with bilateral CIs may be delayed. Thus, early vocabulary development needs to be assessed carefully in order to provide children and families with timely and targeted early intervention for vocabulary acquisition.

**What this paper adds.**

*What is already known:* Children with unilateral CIs have delayed vocabulary development for an extended period. Overall composition of small vocabularies (≤ 100 words) may be similar to that of children with normal hearing (NH), but children with CIs may have lower proportions of nouns and grammatical word types (e.g. pronouns) than their peers with NH. Conflicting results exist regarding the effects of background factors on early vocabulary growth. Bilateral implantation has become increasingly common, but there is little research on early vocabulary growth and composition of children with bilateral CIs.

*What this study adds.* This study showed that despite early bilateral cochlear implantation, children with bilateral CIs are at risk for delayed vocabulary growth due to pre-implantation severe-profound hearing impairment (HI). Vocabulary composition in vocabularies of 1–400 words reflects noun dominance over verbs, adjectives and grammatical word types. Maternal education and residual hearing with hearing aids before implantation have main effects on early vocabulary growth. Thus, children fitted early with bilateral CIs and their families need to be provided with timely and targeted early intervention for vocabulary acquisition in order to reduce the effects of delayed vocabulary skills on later outcomes.
Introduction

Extensive knowledge on early vocabulary development of children with normal hearing (NH) with typical development (TD) exists from many languages (Bates et al. 1994, Caselli et al. 1999, Lyytinen 1999, Fenson et al. 2007, Stolt et al. 2008). Since vocabulary is closely associated with phonological skills (Storkel and Morrisette 2002, Kunnari et al. 2006), morphosyntactic skills (Silvén et al. 2004) and later reading skills (Johnson and Goswami 2010), early vocabulary development of young cochlear implant (CI) recipients may serve as an early cue for speech and language intervention needs. Thus, detailed knowledge about vocabulary growth and composition may aid speech and language therapists (SLTs) in developing the contents of early intervention and parental counselling.

In recent years, the combination of newborn hearing screening, lower age of hearing impairment (HI) diagnosis and cochlear implantation have made it possible for many children with severe to profound HI to make substantial progress in the development of receptive and expressive vocabulary (Duchesne et al. 2009, Boons et al. 2012). Despite early implantation, children with CIs may have persisting delays in vocabulary development compared to their peers with NH (Duchesne et al. 2009, Geers et al. 2009, Boons et al. 2012, Lund 2016). Today, early bilateral implantation has become increasingly common. Bilateral implant use has been shown to be associated with better sound localization acuity and speech perception ability in background noise in children (Johnston et al. 2009). Thus, the current study investigated early receptive and expressive vocabulary growth and vocabulary composition in children who were identified with severe to profound HI soon after birth and who received bilateral CIs at a mean age of one year.

Early vocabulary development in children who are typically developing

Early vocabulary development (under the age of 30 months) in children with NH who are TD has been widely documented in studies using parental diaries and/or checklist
methods. In particular, the MacArthur-(Bates) Communicative Development Inventory (CDI), offers normative data on early vocabulary growth for many languages (Bates et al. 1994, Lytinen 1999, Caselli et al. 1999, Fenson et al. 2007, Stolt et al. 2008). Mean early receptive vocabulary values vary from 5–36 words at age 0;8 (year; month), from 50–94 words at 1;0 and from 120–191 words at ages 1;3–1;4 in different languages. The respective values for expressive vocabulary are 4–10 words at age 1;0, 5–40 words at age 1;4 and usually already over 300 words at the age of 2;0. Thus, there is substantial individual variation in the age when the first words are acquired and in early vocabulary growth in general.

Children also acquire words that belong to different semantic categories of adult language (e.g., nouns and verbs). Given the large variation in vocabulary size, vocabulary composition (distribution of lexical items across various word classes) has often been investigated in relation to size rather than in relation to children’s age. Such studies show that the social-pragmatic terms (onomatopoetic words, names of people, routine words) dominate during the period of the first 50 words (Bates et al. 1994, Caselli et al. 1999, Stolt et al. 2008). When the vocabulary size is below 100 words, there seems to be an initial increase in the percentage of common nouns, followed by a slow proportional decrease. The proportion of predicates (verbs and adjectives) slowly increases only when the vocabulary size is 100–400 words. At this stage the proportion of grammatical function words (i.e., pronouns, prepositions, question words, quantifiers and connectives) is very low, but their proportion increases rapidly after 400 words. When the vocabulary size exceeds 600 words, the composition is relatively stabilized. These changes have been explained to reflect a shift in emphasis from reference, to predication and to grammar, and to reflect cognitive development, e.g. better perception abilities and/or longer memory spans (Bates et al. 1994). The early vocabulary growth and composition of children with NH who are TD and who are
acquiring Finnish follow the trends common to many languages (Lyttinen 1999, Stolt et al. 2008).

Early vocabulary development in children with cochlear implants

Children with unilateral CIs have been reported to progress in their vocabulary growth after CI activation (Connor et al. 2006, Geers et al. 2009, Koşaner et al. 2013, Rinaldi et al. 2013, Lund 2016). Studies indicate, however, that the vocabulary skills of children with CIs, or some children with CIs, remain below their age-norms with NH for an extensive period (Connor et al. 2006, Geers et al. 2009, Lund 2016). Several child-related factors such as the age at HI diagnosis, age at implantation and the duration of CI use have been associated with better linguistic skills and vocabulary growth in children with CIs (Holman et al. 2013, Rinaldi et al. 2013). A recent meta-analysis, however, showed that neither age at implantation, duration of CI use nor chronological age of the child at testing were significantly related to the magnitude of vocabulary score difference between children with CI and children with NH (Lund 2016). Indeed, higher levels of maternal education and properties of maternal input to children with CIs have also been reported to be strong predictors of progress in linguistic skills of children with CIs (Szagun and Stumper 2012).

Recent studies have reported results on early vocabulary growth (during the first two years after CI activation) in children with a relatively early age at implantation (e.g. before the age of 30 months) as reported by parental checklist methods (Koşaner et al. 2013, Rinaldi et al. 2013). Children implanted before the age of 3;0 (N=69) have been reported to acquire the first 100-word receptive vocabulary 4–6 months after CI activation and the first 100-word expressive vocabulary 7–9 months after CI activation (Koşaner et al. 2013). The vocabulary size of the children 19–24 months after CI activation has been similar to that of 19–24-month-old children with NH who were matched for hearing age (i.e., duration of CI use). Because the children with CIs were approximately 20 months older than their peers matched
for hearing age, their early vocabulary growth was still delayed compared to same-age peers with NH. Also Rinaldi et al. (2013) demonstrated that while three out of 22 children were above the range of age-peers with NH in early expressive vocabulary at the mean chronological age of 2;4 years (range 1;7–3;0), fewer than half of the CI children were within the normal range. Thus, despite early HI diagnosis (at the mean age of 5.79 months, range 2–22) and CI activation (at the mean age of 14.26 months, range 9–26) a majority of the children with CIs were delayed in their early vocabulary acquisition. It is worth noting that in the Rinaldi et al. study the children had varying listening experience with CIs at the time of assessment (mean 14.52 months, range 7–25 months), which allegedly affected the low size of expressive vocabulary in some of the children. In summary, unilateral CI activation promotes early vocabulary growth, but a majority of the children with CIs seem to lag behind their age-peers with NH. Since the delays in vocabulary growth may be difficult to recoup (Geers et al. 2009, Lund 2016), knowledge about early vocabulary growth in children with bilateral CIs may provide important information to be utilized in the development of the contents of the early speech and language intervention and parental counselling given by SLTs.

Less is known about the composition of the early vocabulary of children with CIs. Szagun (2000) found that when the size of the expressive vocabulary was ≥ 40 words, the children with CIs had a marked preference for content words (M=62%) over function words (M=38%). Le Normand et al. (2003) found that two years after CI activation, children with CIs produced less nouns, adjectives, verbs (and their different forms), pronouns and adverbs than their age-peers with NH. Three years after CI activation they still produced fewer adverbs and different verb forms than their age-peers with NH. Although they had relatively good lexical diversity (types), they had significantly reduced productivity (tokens). Nott et al. (2009), on the other hand, found that the overall composition of the expressive vocabulary in
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children with profound HI was similar to that of their peers with NH at the vocabulary sizes of 50 and 100 words. One of the children in their study used a hearing aid and 23 children had received CIs (devices fitted before 2;6 years of age). Thus, when the vocabulary size was controlled, the overall composition seemed similar between children with HI and their peers with NH. Nouns constituted the largest portion, followed by predicates and paralexicals (interjections, social words, yes/no). Despite an overall similar composition, the proportion of nouns, common nouns and grammatical word types (including pronouns) was lower and the proportion of predicates and onomatopoeic words was higher for the children with profound HI. Also Koşaner et al. (2013) found that the overall vocabulary composition was similar among children with CIs (n=17) and their normal-hearing peers, matched according to hearing age (n=11), at the vocabulary size of 100 words.

In summary, previous research on vocabulary development in children with unilateral CIs (with relatively heterogeneous ages of implantation) indicates clear delay compared to age-peers with NH. There is a scarcity of progressive follow-up studies regarding early vocabulary growth in children with bilateral CIs and associated background factors. Bilateral implant use has been shown to be associated with better sound localization acuity and speech perception in background noise in children (Johnston et al. 2009), and later receptive and expressive vocabulary (Boons et al. 2012). Thus, the current study asked whether receiving bilateral CIs at a young age might have a positive effect on early vocabulary growth. There are also only a few studies that have investigated the composition of the early vocabulary of children with CIs. Existing studies have also provided mixed findings on the difference between children with CIs and with NH and they have not provided information on vocabulary composition beyond the first 50 or 100 words.

It is also worth noting that vocabulary growth and composition of children with CIs who are acquiring Finnish language has not been studied at all. In typological terms, Finnish
is mainly characterized as an agglutinative language. The basic word formation in Finnish is the addition of endings (bond morphemes, suffixes) to stems. When certain endings are added, the basic stem (root, lexical form) is altered, i.e. there are several morphophonological alterations reflecting characteristics of fusional languages (e.g. käsi ‘hand’, käden ‘hand’s’, käteen ‘into the hand’, käsissä ‘in the hands’). Thus, Finnish words are often polysyllabic and used in alternating forms depending on the syntax. During the period of earliest words, Finnish children with TD may limit their production to two-syllable words, reducing their output of words that are longer than two syllables, but quite early on they attempt to produce polysyllabic words (Kunnari 2002). Therefore, the current study also raised the question of how children with severe to profound HI who receive bilateral CIs acquire vocabulary in an agglutinative language with alternating word forms. How do they perceive and segment these alternating word forms from continuous speech and is it reflected in early receptive and expressive vocabulary growth? Thus, the specific objectives of the current study were: 1) to analyse early vocabulary growth in children with bilateral CIs during the first year following CI activation, 2) to analyse the composition of early vocabulary in relation to vocabulary size, and 3) to investigate the factors associated with the early vocabulary growth.

Methods

Participants

The participants were 20 children (12 boys; 8 girls) with bilateral CIs implanted either simultaneously, or if sequentially (n=4), with only a 4–10-month interval between the first and the second implantation (table 1). The children came from a nationwide prospective multi-centre research project involving all five university hospitals in Finland (i.e., Helsinki, Kuopio, Oulu, Tampere and Turku). The Ethical Committee of the Northern-Savonia Hospital District, University Hospital of Kuopio, approved the study as ethically acceptable and the approval was verified in the other university hospitals. Written consent was received
from parents. The children were initially diagnosed with congenital profound HI through a combination of neonatal hearing screening and auditory brainstem responses or auditory steady-state responses. The aetiologies were GJB2 mutation (n=11) or unknown (n=9). Children diagnosed as having heart conditions, developmental brain disorders, chromosome anomalies, impairments in both hearing and vision or cochlear malformation were excluded. The children were fitted with bilateral hearing aids (HAs) at the median age of 5.5 months (range 3–14 months). All children had at least a three-month HA-trial before implantation. The children’s mean age at first CI activation was 12.9 months (SD=2.8). All children received Nucleus implants (CI24RE; CI412; CI422; CI513) and CP810 or CP910 processors. All children used the Advanced Combination Encoders (ACE) strategy. The children’s auditory thresholds were measured after CI activation and values were equivalent to mild HI.

As a typical clinical practice, the families were offered courses in signing following verification of the child’s HI. Parents of two children had also participated in a course on Cued Speech. The main communication mode of all families was speech and the parents’ use of signs (and Cued Speech) together with speech varied according to the abilities acquired from the courses. The children comprehended some signs (mean 21, SD=24) and produced some signs (mean 8, SD=15) before implantation. The families received family-centred counselling and intervention from local SLTs approximately once a week during the follow-up period of the current study. The children came from middle class families. Education level of the mothers ranged from a higher university degree (eleven of the mothers), to a polytechnic/lower university degree (four of the mothers), and to either a professional or occupational education (five of the mothers).

Instrument and data collection

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The standardized Finnish version of the CDI Infant Form: Words and Gestures (CDI: WG; Lyytinen 1999, Fenson et al. 2007) was used to collect data. The vocabulary section of the Finnish CDI: WG was comprised of 380 items presented in 19 semantic categories. Norm values (children with NH) on the receptive and expressive vocabulary size were available for ages 12 and 14 months for the CDI: WG (N=95; Lyytinen, 1999). Norm values for the expressive vocabulary were available for ages 18, 24, and 30 months for the Finnish CDI: Words and Sentences. The criteria for accepted words were explained to parents at the first data collection interval and discussed repeatedly at follow-up visits, when the questionnaires were handed to the parents. A word was accepted as understood if a child showed repeatedly a clear, immediate and correct response to it. The criteria for produced words were specified in the Finnish CDI: WG form, i.e., spontaneous (not only imitated) use of the word connected repeatedly to the same referent. If the child used a different form of the word marked on the checklist (e.g. [heppa] hevonen ‘horse’), the parents were advised to write the word form on the checklist to verify that the criteria were fulfilled. The parents completed the questionnaires during the time-point of the follow-up visit and sent them in a pre-stamped envelope. The CDI has been considered a reliable and valid method for investigating the early vocabulary development of infants and toddlers with CI (Thal et al. 2007).

**Data analyses and statistics**

**Receptive and expressive vocabulary growth.** Receptive and expressive vocabulary growth was analysed before implantation, and at 1, 3, 6, 9, and 12 months after CI activation from all the receptive and expressive words marked in the CDI: WG form lists. Data from children with CIs were compared with the normative data of the CDI: WG for receptive and expressive vocabulary at ages 12 and 14 months and for expressive vocabulary at the age of 24 months (Lyytinen 1999). This was done in order to analyse the vocabulary growth in
children with CIs both in relation to hearing age (i.e. duration of CI use) and the children’s chronological age. Since the mean chronological age of the children at first CI activation was 12.9 months (SD=2.8, range 10–22) (n=20), their mean ages during the follow-up were as follows: 13.9 months at one month after CI activation (range 11–23; n=16), 15.9 months at three months (range 13–25; n=17), 18.9 months at six months (range 16–28; n=20), 21.9 months at nine months (range 19–31; n=20) and 24.9 months at twelve months after CI activation (range 22–34; n=20). Some data was missing from pre-implantation, one month and three months after CI activation. In order to analyse whether the missing data would bias the results of the vocabulary development follow-up, ‘children with missing early data’ were compared with ‘children with no missing early data’ at six months after CI activation. An independent samples t-test showed insufficient evidence of a difference between the children for the receptive vocabulary (t=-1.158, df=18, p=0.26) or for the expressive vocabulary (t=-0.678, df=18, p=0.51). Thus, the follow-up data can be interpreted as representative.

Receptive and the expressive vocabulary composition. Vocabulary composition was analysed in relation to vocabulary size (see Bates et al. 1994, Lyytinen 1999, Stolt et al. 2008). The receptive and expressive vocabularies were grouped as follows: social terms (i.e., onomatopoetic words, names of people, routine words), common nouns, verbs, adjectives, and grammatical function words (i.e., pronouns, prepositions, question words, quantifiers and connectives). The full sample of receptive and expressive vocabulary was divided into six developmental levels: 1–20 words (receptive n=21; expressive n=35), 21–50 words (receptive n=11; expressive n=15), 51–100 words (receptive n=11; expressive n=13), 101–200 words (receptive n=20; expressive n=4), 201–300 words (receptive n=16; expressive n=6) and 301–400 words (receptive n=5; expressive n=3). The proportions of the semantic categories at each developmental level and 95% confidence intervals for proportions were calculated based on the total sum of words in these categories.
Terms associated with early vocabulary growth.

Growth curve modelling was implemented using a linear mixed model with a random intercept term for each individual and an autoregressive correlation structure for observations within each individual. Two models were fitted for receptive and expressive vocabulary growth (total number of words at each time-point): the first model included fixed-effect main effect terms for time, gender, maternal education, residual hearing, age at first HA fitting and age at first CI activation and fixed-effect interaction terms for time-by-gender and time-by-maternal education. For the second model, only those terms which were found to be statistically significant in the first model were included. Maternal education was dichotomized, given the distribution of the observations (2 levels; higher education=a higher university degree; lower education=polytechnic or lower university degree and professional or occupational education). For the purposes of statistical modelling, one imputation was performed for one child at six months after CI activation based on the mean values at three and nine months after CI activation. Before modelling, logarithmic transformation was performed on the data for expressive vocabulary, due to substantial skewness in the distribution of observations. Effect sizes reported are standardized coefficients from the mixed models (Nieminen et al. 2013) where small equals 0.2, medium equals 0.5 and large equals 0.8.

Results

Vocabulary growth

Before implantation and one month after CI activation only some children comprehended spoken words (figure 1, left panel). Six months after CI activation the children had reached the normative values of children with NH who have the same hearing age (i.e. 12 months) in receptive vocabulary (mean value of the children with CIs 111.1, SD=80.2;
normative mean 89.3, SD=63.2). After nine months of CI use, the children had reached the mean value of 14-month-old infants with NH (mean of the children with CIs 111.1, SD=80.2; normative mean 163.6, SD=82.8). Since their mean chronological age was 21.9 months (range 19–31), they had a delay of approximately seven months in the acquisition of receptive vocabulary (range 4–17). No normative data exist on receptive vocabulary in children older than 14 months as assessed with the CDI, which precluded further comparisons. Visual inspection of Figure 1 illustrates continued receptive vocabulary growth until 12 months after CI activation (mean 234.1, SD = 98.8).

The results for the expressive vocabulary showed that before implantation and during the first three months after CI activation only some children produced spoken words. The majority of the children were still at the one-word stage (i.e., ≤ 50 expressive words) (figure 1, right panel). Three months after CI activation the children had reached the norms of the children with the same hearing age (12-month-olds) in expressive vocabulary (CI children mean 4.9, SD=7.4; normative M=7.1, SD=9.4). By six months of CI use the children had exceeded the normative value of 14-month-old infants with NH (CI children mean 28.5, SD=37; normative mean 17; SD=25.2). At 12 months after CI activation, at the mean chronological age of 24.9 months (range 22–34), their mean size of expressive vocabulary was 148.6 words (SD=121.1). In contrast, 24-month-old children with NH can already have approximately 300 words in their expressive vocabulary (normative mean 277.9, SD = 162.7).

Given the large standard deviations in the early receptive and expressive vocabulary growth during the follow-up, the individual data are presented in Figure 2 (boys, left panels; girls, right panels). The individual data showed that six boys and one girl (35%) reached the norms of 12-month-old or 14-month-old infants with NH in receptive vocabulary during the
first year after CI activation. One boy and one girl (10%) reached the normative values of 12-month-old or 14-month-old infants with NH in expressive vocabulary. Individual data further showed that two boys and five girls (35% of the children with CIs) reached the norms of same-age peers with NH (24-month-olds) in expressive vocabulary during the first year after CI activation. For these seven children, the CIs were activated at the mean age of 11.9 months (range 10–13). These results demonstrated that the children with the most rapid spoken vocabulary growth may catch up to their age-peers with NH in early vocabulary acquisition. Most of the children with bilateral CIs, however, demonstrated expressive vocabulary size that was consistent with younger peers with NH.

Vocabulary composition

The analyses of the composition of the receptive vocabulary showed that the proportion of social terms (52%, 95% confidence interval 18.3–85.7) was high in small vocabularies of 1–20 words (figure 3). When the vocabulary size was 101–200 words, children had significantly more nouns in their receptive vocabulary (51.9%, 95% confidence interval 43.7–60.1) than social terms (19%, 95% confidence interval 12.5–25.4), verbs (18.2%, 95% confidence interval 11.8–24.5) or adjectives (4.1%, 95% confidence interval 0.8–7.4). The proportion of nouns peaked at 56.5% at the vocabulary size of 201–300 words (95% confidence interval 50.3–62.7) and decreased slightly thereafter. The proportion of verbs increased to the level of 17% (95% confidence interval 8.4–25.6) at the vocabulary size of 51–100 words, and remained at that level even at the vocabulary sizes of 101–400. The proportion of function words and adjectives remained below 10% at the vocabulary sizes of 1–400 words (< 8%; < 6%, respectively).

The proportion of social terms was high also in the expressive vocabularies of 1–20 words (71%; figure 4). Nouns increased rapidly at expressive vocabularies of 21–100 words
to a proportion of over 50%. When the vocabulary size was 101–200 words, nouns constituted the highest proportion (57.4%, 95% confidence interval 49.4–65.3) compared to social terms (19.5%, 95% confidence interval 13.1–25.9), verbs (13.1%, 95% confidence interval 7.7–18.5) and adjectives (4.7%, 95% confidence interval 1.3–8.1). Nouns remained as the highest proportion even at the vocabularies of 301–400 words (59%, 95% confidence interval 53.7–64.3). Verbs increased slowly, peaking at 17.7% at vocabularies of 301–400 words (95% confidence interval 13.6–21.8). The proportion of adjectives and function words remained below 10% also in the expressive vocabularies of ≤ 400 words.

Figure 3 approximately here

Figure 4 approximately here

*Terms associated with early vocabulary growth*

Growth curve modelling using a linear mixed model was used to identify main effect terms for time, gender, maternal education, residual hearing, age at first HA fitting and age at first CI activation and fixed-effect interaction terms for time-by-gender and time-by-maternal education. The model revealed a significant main effect for time, gender, maternal education (2 levels; see Methods) and residual hearing with HAs before implantation on the receptive vocabulary growth (table 2). The contribution of the individual terms is indexed by the standardized coefficients from the mixed models (first column in table 2). These standardized coefficients indicate the expected change in receptive and expressive vocabulary growth, i.e. how many standard deviations receptive/expressive vocabulary growth changes when the predictor variable changes (Nieminen et al. 2013). For example, the number of receptive words increased by 0.85 standard deviations when time changed one standard deviation. The effect sizes for time and gender were large and medium, respectively. Effect sizes for maternal education and residual hearing with HAs were small. Time and maternal education showed a statistically significant interaction. Thus, a significant advance of early receptive
vocabulary growth was seen during the first year of bilateral CI use for girls, for the children with CIs whose mothers had a higher university degree, and for children with CIs who had a higher level of residual hearing with HAs before implantation.

Growth curve modelling revealed a significant main effect of time and residual hearing with HAs before implantation also for expressive vocabulary growth (table 2). The effect size for time was large and for residual hearing, small. There were no significant interactions. In summary, a significant advance of expressive vocabulary growth was seen for children with higher levels of residual hearing with HAs before implantation. Age at first HA fitting and age at first CI activation failed to show a significant main effect on early receptive and expressive vocabulary growth during the first year of bilateral CI use.

Table 2 approximately here

Discussion

The aim of the current study was to describe the growth and the composition of the early vocabulary of children with profound HI who had received bilateral CIs and for whom the CIs were activated at the mean age of 12.9 months. The effects of time, gender, maternal education, age at first HA fitting, residual hearing with HAs, and age at first CI activation on early vocabulary growth were also analysed.

The first specific objective was to analyse early vocabulary growth before implantation and during the first year after CI activation. Vocabulary size was compared with the normative data from children with NH at ages 12, 14 and 24 months (Lyytinen 1999). Given the 12-month follow-up and the chronological ages of the children, vocabulary size could be analysed in relation to 1) the duration of CI use (i.e. 12-month hearing age) and 2) the children’s chronological age at the end of the follow-up (i.e. 24 months). The results showed that the children with bilateral CIs reached the normative value of 12-month-old infants of the
same hearing age after six months of CI use in receptive vocabulary and after three months of CI use in expressive vocabulary. Thus, during the first year of CI use, children with bilateral CIs exceeded the vocabulary size that could be expected based on mere hearing age. After one year of CI use, at the mean chronological age of 24.9 months (range 23–34), the children with bilateral CIs as a group still lagged behind the age-norms of 24-month-old toddlers with NH in expressive vocabulary. This finding was consistent with previous studies on children with unilateral CIs showing clear delay in the acquisition of vocabulary (Duchesne et al. 2009, Geers et al. 2009, Yoshinaga-Itano et al. 2010, Rinaldi et al. 2013, Guo et al. 2015).

Since the variation in early vocabulary growth was large, we also analysed the individual data (figure 2). Interpretation of only the group results might have masked the performance of individuals. In the present study, 7/20 children (35%; two boys and five girls) showed very rapid vocabulary growth during the first 12 months of CI use. The children with bilateral CIs with rapid vocabulary growth seemed to catch up to their age-peers with NH in expressive vocabulary, i.e. they reached the age-norms of 24-month-old toddlers. This was an encouraging trend. A longer follow-up would be needed to determine whether the proportion of children who catch up to their age-peers with NH in vocabulary growth actually is larger in children with bilateral CIs than in children with unilateral CIs (see Duchesne et al. 2009, Rinaldi et al. 2013). Some studies have indicated that children with bilateral CIs demonstrate significantly higher receptive and expressive vocabulary skills than children with unilateral CIs approximately three years after CI activation (Boons et al. 2012, Sarant et al. 2014). There is also some evidence indicating that especially children with bilateral CIs could be sensitive to statistical characteristics of words in the ambient spoken language akin to that reported for children with NH during the early stages of vocabulary development (Guo et al. 2015), which might enhance early vocabulary growth and contribute to vocabulary skills even later on.
In the current study, 7/20 children (35%; 7 boys and 2 girls) showed very slow vocabulary growth. They only reached the norms of 12-month-old or 14-month-old infants with NH in receptive and expressive vocabulary. Thus, the individual variation and delay in vocabulary growth in some children with bilateral CIs seemed to be very similar to that of children with unilateral CIs. Previous literature has shown that despite early diagnosis and early HA-fitting and implantation, approximately half of the children with unilateral CIs lag behind their age-level in vocabulary growth during the first years of CI use (Duchesne et al. 2009, Geers et al. 2009, Rinaldi et al. 2013, Guo et al. 2015). In the current study, some of the children with slowest vocabulary acquisition only maintained a constant rate of development after CI activation, the rate predicted by hearing age rather than accelerated rate, as also reported in some previous studies on children with unilateral CIs (Yoshinaga-Itano et al. 2010). This finding has clear theoretical and clinical relevance. We may hypothesize that slow vocabulary growth of some children with bilateral CIs could be indicative of specific difficulties in the acquisition of new words. Children with unilateral CIs have been shown to perform more poorly on word-learning tasks than their peers with NH in studies using experimental paradigms to investigate the word learning abilities (Houston et al. 2005, Houston et al. 2012). Only children who had received their CI before 12 months of age were reported to perform similar to age-matched children with NH. For the seven children with the fastest vocabulary growth in the current study, the CIs had been activated at the mean age of 11.9 months (range 10–13 months). The results of the current study emphasize that children with early bilateral implantation who only maintain a constant rate of vocabulary growth after CI activation need to be followed-up carefully. Since early linguistic development may predict later linguistic skills, such a slow rate of vocabulary growth after bilateral implantation could be indicative of word-learning difficulties and even additional disabilities, such as specific language impairment (de Hoog et al. 2015).
The current study was the first to investigate vocabulary acquisition in children with severe to profound HI who received bilateral CIs and who were acquiring Finnish. The basic word formation in Finnish, i.e. addition of endings, bond morphemes and suffixes to stems, and the morphophonological alterations induce the often polysyllabic nature of Finnish words used in alternating forms depending on syntax. We were interested in whether this was reflected in early vocabulary growth. The results of the current study showed that 35% of the children with bilateral CIs were able to catch up to the age-norms of toddlers with NH in expressive vocabulary during the first year of CI use. The results also showed that 55% of the children lagged behind the age-norms. As such, the results were in line with previous studies on children who are acquiring Germanic languages showing that children with CIs may have vocabulary acquisition delays for an extended period after implantation (Geers et al. 2009, Rinaldi et al. 2013, Lund 2016). Further studies are needed to investigate in a more detailed manner the syllable structures of the words produced and the use of the words in sentences. If children with CIs who are acquiring Finnish were to have difficulties in word acquisition, the difficulties could be reflected especially in the syllable structures of the early words and morphosyntax.

The second specific study aim was to analyse the early receptive and expressive vocabulary composition in relation to vocabulary size. In the present study, the trends in the composition of receptive and expressive vocabularies were very similar. Social terms (onomatopoetic words, names of people, routine words) constituted the largest proportion in small receptive and expressive vocabularies (≤ 50 words). Nouns constituted the largest proportion at vocabularies of 101–400 words, whereas verbs, adjectives and grammatical function words (pronouns, prepositions, question words, quantifiers and connectives) constituted the smallest proportions. These findings of noun-dominance over verbs and function words were consistent with previous studies on children with NH (Bates et al. 1994,
Caselli et al. 1999, Stolt et al. 2008) and also with recent reports on small vocabularies (≤100 words) of children with CIs (Nott et al. 2009, Koşaner et al. 2013). The present study further showed that verbs, adjectives and grammatical function words were rare and acquired at a slower rate than nouns even at vocabularies of 101–400 words. These findings were consistent with ground-breaking studies on children with NH (Bates et al. 1994, Caselli et al. 1999, Lyytinen, 1999, Stolt et al. 2008). The proportional growth of grammatical function words has typically been reported to start when the children have acquired 300–500 words. Thus, it seems that once the children received CIs a slow developmental shift from routine words and nouns to grammatical function words was evident. This developmental shift could be seen in receptive as well as in expressive vocabulary. The findings offer encouraging information on the development of vocabulary composition in children with CIs. Some reports have indicated that parents of children with HI may use, e.g. short mean length of utterances and more restricted vocabulary (Morgan et al. 2014), which in turn might have a restricting effect on early vocabulary composition. Further analyses are needed to conclude whether this encouraging trend in the vocabulary composition of children with bilateral CIs holds true also for vocabularies larger than 400 words.

The third aim of the present study was to analyse the factors associated with early vocabulary growth after bilateral implantation. In the current study, time, gender, maternal education and residual hearing with HAs before implantation had significant main effects on early vocabulary growth, while age at first HA fitting and age at CI activation did not. These findings were very consistent with some of the latest studies on children with unilateral CIs which have showed that higher socioeconomic status, higher levels of parental education and IQ are positively associated with language development in children with CIs, and not just age at implantation (Szagun and Stumper 2012, Holman et al. 2013, Lund 2016). It is recognized that the influence of socioeconomic status is mediated via maternal language input to the
Early vocabulary development in children with bilateral CIs: Authors’ accepted manuscript (AAM) published in International Journal of Language & Communication Disorders, DOI: 10.1111/1460-6984.12322.

child and, thus, the child’s experience with language (Hoff 2003). Therefore, we may hypothesize that maternal language input may have also positively affected the results of the present study. It is also worth noting that residual hearing with HAs, not age at first HA fitting, had a significant effect on early vocabulary growth. This finding was in contrast to the study by Rinaldi et al. (2013), who showed that age at HI diagnosis was a significant predictor of vocabulary development. Age at HI diagnosis and age at first HA fitting seem to predict later linguistic skills, if these procedures are severely delayed. Due to universal newborn hearing screening, however, the children in the current study were diagnosed early and thus received CIs relatively early. In summary, it seems compelling that for these children with early HI identification and bilateral implantation gender (female), higher maternal education and residual hearing with HAs before implantation are factors accounting for early vocabulary growth. The results emphasized the importance of parental counselling in early intervention regarding the role of residual hearing before implantation and the role of vocabulary input after CI activation.

In interpreting the results of the current study, some limitations must be considered. First, the 12-month follow-up period after CI activation was relatively short. We are aware that a longer follow-up would be needed in order to investigate the developmental patterns in vocabularies larger than 400 words. Second, although there were altogether 20 children with bilateral CIs in the present study, we had a small number of children with vocabulary sizes larger than 200 words, especially in expressive vocabulary. Third, we are aware that maternal education indirectly refers to diversity in maternal language input to the child, but direct analyses of maternal input would offer additional information on its quality. We are currently expanding our analyses to address the aforementioned issues.

Conclusions
Despite HI diagnosis and bilateral implantation at an early age, vocabulary growth in children with bilateral CIs may still be delayed. The children with the fastest vocabulary growth seem to catch up with their age-norms, but 55% of the children with bilateral CIs seem to lag behind the age-norms. Slow vocabulary growth after early bilateral implantation might be indicative of specific difficulties in linguistic skills. Thus, vocabulary growth of children with bilateral CIs needs to be followed-up systematically in order to provide families and children with timely and targeted early intervention for vocabulary acquisition.
References


DE HOOG, B. E., LANGEREIS, M. C., VAN WEERDENBURG, M., KNOORS, H. E. T. and VERHOEVEN, L. 2015, Linguistic profiles of children with CI as compared with children with hearing or specific language impairment. *International Journal of Language and Communication Disorders*, early online. DOI: 10.1111/1460-6984.12228


Figure captions:

Figure 1. Early receptive and expressive vocabulary growth during the first year of bilateral cochlear implant (CI) use. The lines represent mean number or words at each time interval before and after activation of the CIs. The grey area represents 95% confidence interval of the mean.

Figure 2. Individual profiles in early receptive and expressive vocabulary growth in boys (left panels) and in girls (right panels).

Figure 3. Composition of the receptive vocabulary in relation to the vocabulary size.

Figure 4. Composition of the expressive vocabulary in relation the vocabulary size.
Table 1

*Demographic characteristics of the children with cochlear implants (CIs)*

<table>
<thead>
<tr>
<th>Child number</th>
<th>Gender</th>
<th>Age at first HA fitting (months)</th>
<th>Unaided soundfield reaction threshold before CI</th>
<th>Aided soundfield reaction threshold before CI</th>
<th>Age at first CI activation (months)</th>
<th>Puretone average at 12 mos. after CI</th>
</tr>
</thead>
<tbody>
<tr>
<td>ID1</td>
<td>M</td>
<td>6</td>
<td>NR</td>
<td>NR</td>
<td>14</td>
<td>35</td>
</tr>
<tr>
<td>ID2</td>
<td>M</td>
<td>3</td>
<td>106</td>
<td>63(^2)</td>
<td>12</td>
<td>23</td>
</tr>
<tr>
<td>ID3</td>
<td>M</td>
<td>14</td>
<td>84</td>
<td>85</td>
<td>18</td>
<td>24</td>
</tr>
<tr>
<td>ID4</td>
<td>M</td>
<td>5</td>
<td>NR</td>
<td>103</td>
<td>15</td>
<td>31</td>
</tr>
<tr>
<td>ID5</td>
<td>M</td>
<td>7</td>
<td>NR</td>
<td>78</td>
<td>12</td>
<td>26</td>
</tr>
<tr>
<td>ID6</td>
<td>M</td>
<td>8</td>
<td>NR</td>
<td>54</td>
<td>12</td>
<td>35(^2)</td>
</tr>
<tr>
<td>ID7</td>
<td>M</td>
<td>6</td>
<td>NR</td>
<td>98</td>
<td>12</td>
<td>33(^3)</td>
</tr>
<tr>
<td>ID8</td>
<td>F</td>
<td>6</td>
<td>NR</td>
<td>NR</td>
<td>14</td>
<td>31</td>
</tr>
<tr>
<td>ID9</td>
<td>F</td>
<td>6</td>
<td>NT</td>
<td>80</td>
<td>11</td>
<td>49</td>
</tr>
<tr>
<td>ID10</td>
<td>M</td>
<td>3</td>
<td>NT</td>
<td>116</td>
<td>11</td>
<td>43</td>
</tr>
<tr>
<td>ID11</td>
<td>F</td>
<td>6</td>
<td>126</td>
<td>118</td>
<td>13</td>
<td>34</td>
</tr>
<tr>
<td>ID12</td>
<td>F</td>
<td>4</td>
<td>130</td>
<td>100</td>
<td>10</td>
<td>36</td>
</tr>
<tr>
<td>ID13</td>
<td>M</td>
<td>4</td>
<td>90</td>
<td>64</td>
<td>11</td>
<td>31</td>
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<tr>
<td>ID14</td>
<td>M</td>
<td>6</td>
<td>105</td>
<td>60</td>
<td>22</td>
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<tr>
<td>ID15</td>
<td>F</td>
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<td>110</td>
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<td>ID17</td>
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<td>5</td>
<td>NT</td>
<td>103</td>
<td>13</td>
<td>33(^4)</td>
</tr>
<tr>
<td>ID</td>
<td>Gender</td>
<td>Age</td>
<td>PTA 0.5–2 kHz</td>
<td>PTA 1–4 kHz</td>
<td>PTA 0.5–4 kHz</td>
<td>Mean</td>
</tr>
<tr>
<td>-----</td>
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<td>-----</td>
<td>-------------</td>
<td>-------------</td>
<td>--------------</td>
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</tr>
<tr>
<td>ID18</td>
<td>M</td>
<td>7</td>
<td>76</td>
<td>12</td>
<td>50</td>
<td></td>
</tr>
<tr>
<td>ID19</td>
<td>F</td>
<td>5</td>
<td>84</td>
<td>13</td>
<td>24</td>
<td></td>
</tr>
<tr>
<td>ID20</td>
<td>F</td>
<td>5</td>
<td>119</td>
<td>11</td>
<td>34</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Mean</td>
<td>5.7</td>
<td>120.6</td>
<td>91.5</td>
<td>12.9</td>
<td>32.4</td>
</tr>
<tr>
<td></td>
<td>SD</td>
<td>2.4</td>
<td>15.5</td>
<td>24.4</td>
<td>2.8</td>
<td>8.3</td>
</tr>
</tbody>
</table>

**Note.** 1 Pure tone average (PTA)0.5–4 kHz, (if no response obtained at a frequency, a value of 130 dB HL was applied in the calculation; British Society of Audiology, 2011); HA = hearing aids; NR = no response; NT = not tested; 2PTA1–2 kHz; 3PTA1–4 kHz; 4PTA0.5–2 kHz.
Table 2

_Growth curve modelling using a linear mixed model with a random intercept term for each individual and an autoregressive correlation structure for observations within each individual for the early receptive and expressive vocabulary growth (time-points before implantation, and 1, 3, 6, 9, and 12 months after cochlear implant [CI] activation)_

<table>
<thead>
<tr>
<th></th>
<th>Model with all terms included</th>
<th>Model with terms found significant in the first model</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>beta (SE)  t   p</td>
<td>beta (SE)  t   p</td>
</tr>
<tr>
<td><strong>Receptive vocabulary</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Within-subjects effects</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Time</td>
<td>0.848 (0.09)  9.246  &lt; 0.001</td>
<td>0.963 (0.07)  13.902  &lt; 0.001</td>
</tr>
<tr>
<td><strong>Between-subjects effects</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Gender</td>
<td>0.504 (0.16)  3.065  0.008</td>
<td>0.479 (0.17)  2.893  0.011</td>
</tr>
<tr>
<td>Maternal education</td>
<td>-0.362 (0.16) -2.243  0.041</td>
<td>-0.305 (0.16) -1.921  0.073</td>
</tr>
<tr>
<td>Residual hearing with HAs²</td>
<td>-0.234 (0.08) -2.948  0.011</td>
<td>-0.251 (0.08) -3.155  0.006</td>
</tr>
<tr>
<td>Age at first HA² fitting</td>
<td>0.073 (0.08)  0.902  0.382</td>
<td></td>
</tr>
<tr>
<td>Age at first CI activation</td>
<td>0.049 (0.08)  0.589  0.565</td>
<td></td>
</tr>
<tr>
<td><strong>Interactions</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Time*Gender</td>
<td>0.206 (0.11)  1.898  0.061</td>
<td></td>
</tr>
<tr>
<td>Time*Maternal education</td>
<td>-0.306 (0.11) -2.819  0.006</td>
<td>-0.367 (0.10) -3.502  0.001</td>
</tr>
<tr>
<td><strong>Expressive vocabulary</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Within-subjects effects</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Time</td>
<td>0.870 (0.07)  11.421  &lt; 0.001</td>
<td>0.854 (0.046)  18.7  &lt; 0.001</td>
</tr>
<tr>
<td><strong>Between-subjects effects</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Gender</td>
<td>0.219 (0.14)  1.590  0.134</td>
<td></td>
</tr>
<tr>
<td>Variable</td>
<td>Coefficient</td>
<td>Standard Error</td>
</tr>
<tr>
<td>--------------------------------</td>
<td>-------------</td>
<td>----------------</td>
</tr>
<tr>
<td>Maternal education</td>
<td>-0.268</td>
<td>0.135</td>
</tr>
<tr>
<td>Residual hearing with HAs^2</td>
<td>-0.146</td>
<td>0.070</td>
</tr>
<tr>
<td>Age at first HA^2 fitting</td>
<td>0.091</td>
<td>0.070</td>
</tr>
<tr>
<td>Age at first CI activation</td>
<td>-0.035</td>
<td>0.070</td>
</tr>
<tr>
<td>Interactions</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Time*Gender</td>
<td>0.104</td>
<td>0.090</td>
</tr>
<tr>
<td>Time*Maternal Education</td>
<td>-0.125</td>
<td>0.090</td>
</tr>
</tbody>
</table>

Note. beta = standardized coefficient; SE = standard error; t = t-value; p = p-value; (small equals 0.2, medium equals 0.5, and large equals 0.8); HA = hearing aid; Values based on logarithmic transformation due to substantial skewness in the distribution of the observations.