The effect of word frequency on phonemic accuracy in children with cochlear implants and peers with typical levels of hearing

Jolien Faes, Joris Gillis and Steven Gillis

University of Antwerp

Abstract

The frequency of occurrence of words and sounds has a pervasive influence on typically developing children's language acquisition. For instance, highly frequent words appear earliest in a child's lexicon (Goodman, Dale & Li, 2008), and highly frequent phonemes are produced more accurately (Edwards, Beckman & Munson, 2004). This paper evaluates (1) whether word frequency influences word accuracy, and (2) whether this is also the case for children with a history of auditory deprivation. More specifically, the influence of word frequency on phonemic accuracy is examined in severe-to-profound hearing impaired children with a cochlear implant (CI), and compared to age-matched children with typical hearing, between word onset and age seven. Results show that highly frequent words are produced more accurately, except for words in the highest frequency regions, i.e., predominantly closed-class words. This effect is more pronounced in children with typical hearing as compared to children with CI are sensitive to word frequency, but to a lesser extent than peers with typical hearing.

Introduction

Examples of frequency of occurrence effects in the acquisition process of children with typical hearing appear in all linguistic domains: the acquisition of single words, inflectional morphology, syntax, etc. (Ambridge, Kidd, Rowland, & Theakston, 2015). These frequency effects concern the incidence of phonemes, sequences of phonemes, words, sequences of words, etc., and all go in the same direction: linguistic units that occur more frequently in the language addressed to children are also acquired earlier (Ambridge et al., 2015).

The present paper aims to investigate the relationship between word frequency in child-directed speech (i.e. the incidence of words) and the accuracy of children's own productions. The focus is on the development of children with CI, since little is known about frequency effects in these children. We investigate the longitudinal development of their word accuracy and compare their development with that of children with typical hearing. Children with CI are children with severe-to-profound hearing loss with partly restored hearing due to cochlear implantation. Two research questions will be addressed: (1) Does the frequency in the input affect word accuracy in children with CI? (2) Are children with CI similar to children with typical hearing? In what follows, we first discuss what can be expected from children with typical hearing, and then we contemplate if and why children with CI are expected to differ from children with typical hearing.

Frequency effects in children with typical hearing

Only a few studies consider the effect of frequency (i.e. word frequency) on word accuracy in children with typical hearing. In two-year old English-speaking children, there seems to be no relationship between frequency in the input and accuracy: Sosa and Stoel-Gammon (2012) showed that highly frequent words in the input are less variable but not more accurate in children's speech. In contrast, Ota (2006) found that Japanese two-year-olds are less likely to truncate words with a high frequency in the input. In other words, they omit syllables in highly frequent words less often and, thus, these words are more accurately produced.

These two studies are the only ones to investigate directly the effect of word frequency on word accuracy in children with typical hearing, and they leave the debate undecided: Sosa and Stoel-Gammon (2012) found no effect, whereas Ota (2006)'s results pointed in the opposite direction. However, two other frequency effects have been studied that may very well be related to word accuracy. First, frequency at the segmental level plays a role in the accurate production (and the age of acquisition) of segments and sequences of segments. Segments and segmental sequences that occur more often in the input appear earlier in children's own production and are produced more accurately (Edwards, Beckman, & Munson, 2004; Lee, Davis, & MacNeilage, 2010; Munson, 2001; Van Severen et al., 2013). Moreover, Stokes and Surendran (2005) showed that the input frequency of initial consonants accounts for approximately 40% of the variance in consonant production accuracy in English-speaking and Dutch-speaking children. Thus, frequency at the segmental level affects the accuracy of segments. In the present study we investigate whether a similar trend appears at the word level: Frequent words in the input are produced more accurately by children. Such a finding would corroborate Ota (2006) and contradict Sosa and Stoel-Gammon (2012).

Secondly, a relation has been established between word frequency and lexical acquisition: frequency at the word level influences the age of acquisition of words, which is similar to the relation of segmental frequency and the age of acquisition of segments. Children with typical hearing acquire highly frequent words earlier, except for words with the highest frequencies such as closed-class words (e.g. pronouns, articles, etc.) (Ashkenazi, Ravid, & Gillis, 2016; Goodman, Dale, & Li, 2008; Naigles & Hoff-Ginsberg, 1998). Some studies found an opposite effect (Stokes, 2010; Stokes, Kern, & Dos Santos, 2012; Storkel, 2009), but that may very well be explained by the source of their frequency counts. The studies that did not found an effect of word frequency on lexical development (Stokes, 2010; Stokes et al., 2012; Storkel, 2009) and accuracy (Sosa & Stoel-Gammon, 2012) used corpora of adult-directed speech (ADS) or the corpus of adult-directed written language of Kucera and Francis (1967).

The use of adult corpora (spoken and written) may have had an effect on the findings, since childdirected speech (CDS) differs from ADS in several respects (Bernstein Ratner, 2013; Newport, Gleitman, & Gleitman, 1977). For instance, CDS has a limited vocabulary and contains many repetitions, which affects the frequency of particular words. Moreover, it has been shown that children are sensitive to CDS but not to ADS: children's vocabulary development is only correlated with CDS and not with ADS (Weisleder & Fernald, 2013). In addition, spoken language differs from written language (Akinnaso, 1982). Goodman et al. (2008) and Van Severen et al. (2013) have shown that the effect of frequency is best captured when using a corpus of CDS as opposed to a corpus of adult-directed written or spoken language. In a similar vein, Stoel-Gammon (2011, p. 26) proposed that word frequency effects should be analysed using "a variety of measures including [...] [i.a.] general counts of input to children based on corpora from many children". The present paper takes up this suggestion.

Frequency effects in children with CI

To the best of our knowledge, there are no studies on the relation between word frequency and word accuracy in children with CI. But, they appear to be sensitive to particular aspects of the statistics of the language they hear. For instance, Guo, McGregor, and Spencer (2015) found a positive correlation between vocabulary and phonotactic probability in CDS. However this correlation only seems to hold for children with bilateral CIs and not for children with unilateral ones. Thus, children with bilateral CIs show a similar sensitivity to word statistics as children with normal hearing, while children with unilateral implants appear to be less sensitive to statistical effects at the word and the segmental level in the ambient language.

Frequency affects the composition of children's early lexicons which consist of highly frequent words in CDS (Han, Storkel, Lee, & Yoshinaga-Itano, 2015). This frequency effect decreases between four and seven years of age (Han et al., 2015). However, Guo et al. (2015) did not found a correlation between word frequency and the acquisition of monosyllabic nouns, verbs and adjectives one year post-implantation in children with unilateral and bilateral CI's.

The current paper investigates a longitudinal sample of children with CI's speech. The effect of age at implantation is included, as studies have shown benefits for earlier implantation on different domains of language development (Levine, Stother-Garcia, Golinkhoff, & Hirsh-Pasek, 2016). The

effect of word frequency on word accuracy in children with CI is also compared to that in children with typical hearing.

If there is a difference in the way frequency of occurrence affects speech production in children with CI and children with typical hearing, there are 2 options: the effect is more pronounced in children with CI as compared to children with typical hearing or the effect is less pronounced in children with CI as compared to children with typical hearing. First, it is possible that frequency effects are more pronounced in children with CI than in peers with typical hearing. Han et al. (2015) indicated that frequency effects are especially relevant early on in children's oral language development, as the effect of frequency decreases with age. As children with CI start hearing later, they have less language experience in comparison with their age-matched peers with typical hearing of the same chronological age. Following Han et al. (2015)'s suggestion, the effect of frequency in children with CI may very well be more pronounced since it is more pronounced early on in development. But it is unclear if this affects the relation between frequency and accuracy as well.

In addition, children with CI's speech perception is affected by the degraded signal they receive through the implant (Drennan & Rubinstein, 2008; Wilson, 2006). For instance, it has been shown that the current devices are unable to accurately transmit sounds at high spectral frequencies such as fricatives (Stelmachowicz, Pittman, Hoover, Lewis, & Moeller, 2004). A consequence of the degraded signal may be less fine-grained phonological representation (Nittrouer, Caldwell-Tarr, & Lowensthein, 2013). A fine-grained phonological representation is needed for accurate production. In the case of infrequent words, children have to construct a phonological representation from a degraded signal without many opportunities to adjust it. Whereas for more frequent words, children with CI may have more opportunities to adjust their phonological representations. Hence, the effect of frequency in the input may be more outspoken in children with CI.

But, the effect of frequency may also be less pronounced in children with CI. Houston, Pisoni, Kirk, Ying, and Miyamoto (2003) have shown that children with CI pay less attention to environmental speech. Therefore, they may very well be missing some aspects of the speech directed to them, such as word frequency (Saffran, Aslin, & Newport, 1996). Moreover, O'Grady (2015) has suggested that frequency effects are processing effects. As children with CI are generally found to

have problems with processing and storage of information (AuBuchon, Pisoni, & Kronenberger, 2015), it is very likely that frequency of words is not processed as efficiently as in children with typical hearing.

The present study aims to investigate the effect of word frequency in the input on word accuracy in children's productions. Two groups of children are compared, viz. children with CI and peers with typical hearing. We have three research questions: (1) Does word frequency affect production accuracy of words in children with typical hearing? (2) Is this effect also present in children with CI? And, (3), is the effect similar, more or less pronounced in children with CI?

Methods

Participants

The participants of the present study are part of the CCLC (CLiPS Child Language Corpus). This corpus consists of video recordings of the spontaneous speech of both children with typical hearing and children with CI. All children lived in Flanders, i.e. the northern part of Belgium, and are monolingual Dutch.

Nine children with CI were followed longitudinally. Monthly visits were scheduled from the moment the device was activated up to 30 months of age, and after that yearly visits were arranged up to age seven. Table 1 displays the characteristics of the CI group. All children had a congenital profound hearing loss. Before implantation, the mean unaided Pure Tone Average (PTA) was 112.56 dB HL (SD = 9.12) in the better ear. The causes of deafness were genetic (S1, S2, S4-S7, S9, mainly a mutation in the connexine-26 gene), a cytomegalovirus infection (S3) and unknown (S8). No other patent health or development problems were reported during data collection. All children received a Nucleus-24 cochlear implant, without contralateral stimulation with a hearing aid. The mean age at implantation was 11.92 months (SD = 5.25) and the mean age at implant activation was 13.11 months (SD = 5.39). Six children received a second implant during data collection (Table 1).

The children's CAP scores (Categories of Auditory Performance, Archbold, Lutman, and Marshall (1995)) improved from a mean score of 2.4 (SD = 1.43) three months after implantation to a mean score of 6.56 (SD = 0.72) 30 months after implantation. CAP scores range from 0 to 7, with 0

representing no awareness of environmental sound, and 7 stands for the use of telephone with a familiar talker (Archbold et al., 1995). At the age of five, the mean PTA had improved to 32.33 dB HL (SD = 7.11). All children were enrolled in a multidisciplinary program: they received speech and language therapy and auditory training twice a week (ca. 30 min.) and they received one 30-minute session of ergotherapy per week. All children had hearing parents who attended a sign language course during their child's rehabilitation, hence they knew some useful lexical signs, but were far from fluent in sign language. So, all children used oral language with a limited amount of lexical signs (Flemish sign language). The mean age at which the first word emerged, was 18 months (SD = 3.00, range 15 - 23 months), but there was large individual variation between the children (see Connor, Craig, Raudenbush, Heavner, and Zwolan (2006) for similar observations).

Please insert Table 1 over here.

The control group consisted of 64 children with typical hearing, for which no patent hearing, health or developmental problems were reported. The corpus of children with typical hearing was cross-sectional. The participants were 11 two-year-olds (mean = 23.53 months, SD = 0.71), 9 three-year-olds (mean = 35.88 months, SD = 1.09), 12 four-year-olds (mean = 48.35 months, SD = 1.39), 11 five-year-olds (mean = 60.41 months, SD = 1.32), 11 six-year-olds (mean = 72.43 months, SD = 2.80) and 10 seven-years-olds (mean = 83.32 months, SD = 1.74). Thus, each child in this cross-sectional part of the corpus participated only once and only one recording of each child is available.

Material

Spontaneous speech samples of unstructured interactions between the child and his/her caregiver(s) were gathered at the child's home by means of 60 to 90 minute video recordings. After each recording, a 20-minute selection of completed interactions was made (Molemans, 2011; Molemans, Van den Berg, Van Severen, & Gillis, 2012; Schauwers, 2006; van den Berg, 2012; Van Severen, 2012).

Lexical items of all speech samples were transcribed using CHILDES'CLAN program according to the CHAT conventions (MacWhinney, 2000). For the child's productions, an orthographic transcription and a phonemic transcription was made using DISC symbols. For instance, suppose a child said [bu] for the Dutch word *boek* (/buk/, Eng. 'book'). The transcription is provided in (1). In the speaker tier (*CHI), an orthographic transcription of the child's utterance is made. The phonemic tier (%pho) contains the phonemic transcription of what the child actually said. Next, a phonemic transcription of the target word, i.e. the adult equivalent of the child's production, was added, resulting in the %ohp tier in (1). The phonemic transcription of the conventional pronunciation of each target word was retrieved from the lexical database Fonilex (Mertens, 2001) and automatically added to the %ohp tier. Thus, in (1) a standard orthographic transcription of the actual child production (/bu/, incorrect production), and the %ohp tier represents the phonemic transcription of the target word, i.e. the adult equivalent of the child's actual production of a word coincides with the target or adult-like production. For utterances of the adult(s) the same procedure was followed.

(1)	*CHI:	boek
	%pho:	bu
	%ohp	buk

The reliability of the phonemic transcriptions on the %pho tiers was computed on 10% of the data. The percentage of agreement and the Kappa score were calculated. For the corpus of children with typical hearing, percentages of agreement for interrater and intrarater reliability were 63.69% and 81.51% respectively. For the CI corpus, only a percentage of agreement for interrater reliability was computed and equals 81.63%. Kappa scores were 0.60 for interrater reliability in the speech samples of children with typical hearing and 0.80 for intrarater reliability. These scores were on the edge of "moderate" to "substantial" and on the edge of "substantial" to "almost perfect" respectively (Landis & Koch, 1977). The Kappa score for intrarater reliability of the CI speech samples equals 0.87 and was interpreted as "almost perfect" (Landis & Koch, 1977).

Data analyses

As a preliminary step in the actual computation of the accuracy scores, the phonemic transcriptions on the %pho (what the speaker actually said) and %ohp (the target or standard transcription) tiers were aligned. For instance, the %pho and %ohp in (1) can be aligned as in (2):

(2) %pho: b u =
 %ohp b u k

In the example /b/ and /b/ as well as /u/ and /u/ can be easily aligned because they are identical. The /k/ on the %ohp tier is aligned with the character "=" on the %pho tier, representing an empty or missing character on the %pho tier, and hence, denotes a segment deletion. The alignment process was done by a computer program, implementing minimum edit distance dynamic programming (Elffers, Van Bael, & Strik, 2005). In essence, two strings (the %pho and %ohp tiers) are aligned using three minimal edit operations, viz. substitution, deletion and insertion of elements, and the algorithm computes the minimal number of edit operations starting from the first string to arrive at the second string. The alignments of the phonemic transcriptions of the actual productions and the target transcriptions were subsequently checked manually in order to eliminate alignment errors.

Phonemic accuracy was measured in a dynamic cost model of Levenshtein Distance (LD) as presented in Wieling, Prokic, and Nerbonne (2009) and adapted by Faes, Gillis, and Gillis (2016) to fit the purpose of computing phonemic accuracy in child speech. LD computes the distance between a child's production and its adult (target) equivalent, such as the transcriptions on the %pho and %ohp tiers in (2). LD is more fine-grained than other measures that have been applied in the literature, such as the phonological mean length of utterance (pMLU, Ingram, 2002) (for a discussion, see Faes et al., 2016).

Conceptually, LD amounts to comparing two strings of characters, i.e. the transcription of an adult target and a transcription of the child's rendition of that target. LD measures the distance between these two strings in terms of the minimal edit distance. If they are identical, the child's rendition is

adult-like and LD equals, in principle, zero. If the two strings are not identical, the deviation of the child's rendition from the adult target is computed and its "cost" or "distance" is determined. In order to compute the cost of a deviation, a model of adult spoken language is constructed by aligning a corpus of spontaneously spoken Dutch with a reference transcription provided by a pronunciation dictionary. In this way frequent deviations from the reference transcription are captured and lead to a smaller cost than infrequent deviations. For instance, in spontaneously spoken Dutch, tense vowels (/i, a, e, o, .../) are often substituted by their lax counterparts (/ , , , , , .../) or even reduced to schwa (Booij, 1995; Ernestus, 2000). Thus if the child renders /i/ as / /, this deviation is weighed less heavily than when she would render / / as /u/ or /a/. Another example: in colloquial spoken Dutch, /n/ preceded by a schwa, as in verb infinitives, nominal plurals, etc. is often deleted. Thus when a child renders the infinitive /lopen/ ('to run') as [lope], instead of the reference [lopen], this is considered a less serious error as compared to rendering it as, say, [lopel] or [loper].

LD between a child's production and its adult equivalent is calculated in two stages: (1) in a first stage, a model of adult spoken language is constructed in order to compute an initial cost model, and (2) the distance of each word production of the child is determined relative its adult target. These two stages will be briefly described. For technical details, we refer to Wieling, Margaretha, and Nerbonne (2012) and Faes et al. (2016).

In the first stage, actual adult word productions are aligned with their targets in a VC-sensitive way. This means that a vowel cannot be aligned with a consonant, and vice versa. In example (3) the adult production of the Dutch word /spelə/ (*to play*) is aligned with the target /spel@n/:

(3) Adult actual production: s p e l a =Target: s p e l a n

For each segment in the aligned transcription, the pointwise mutual information (PMI) is calculated using the formula in (4):

(4)
$$PMI(x,y) = \log_2 \frac{p(x,y)}{p(x)p(y)}$$

Where p(x, y) is the probability of encountering the pair (x, y) in the alignment, p(x) the probability of segment /x/ and p(y) the probability of segment /y/. Take the pair /s/-/s/ in example (3), the PMI of this pair is the probability of /s/ in the target lined up with /s/ in the actual production, divided by the product of the probability of /s/ in the corpus of actual adult productions and the probability of /s/ in the target corpus. The PMI of each pair of segments is calculated for the entire corpus of adult child-directed speech. Pairs of segments that co-occur frequently, receive a high PMI value.

In order to convert the PMI values into a cost, the PMI value of each pair of segments is subtracted from the maximum PMI value (see Faes et al. (2016) for an example). As a result, pairs of segments that frequently co-occur receive a smaller cost (or distance) than segments that rarely co-occur. When this final cost model is computed, the Levenshtein distance (LD) between an adult production and the target is the sum of the distances between the individual segments. Since word length affects LD, the final LD score per word is normalized by dividing it by the word length (the number of segments in the word). As such, an average LD per word is calculated.

In the second stage, the children's word productions are aligned with their targets, and the cost model constructed in the first stage is used to compute the distance between the two. The main reason for starting from that model is that otherwise errors that are highly frequent in child speech would receive a high PMI value and therefore a low LD cost. Therefore, the costs are calculated using the model of adult speech constructed in the first phase, and the eventual distance between the child's rendition and the adult target is calculated in this second phase. If a pair of segments is encountered that did not appear in the adult cost model, Katz Smoothing is applied (Chen & Goodman, 1998). Through Katz Smoothing, unobserved pairs of segments receive a small probability and thus a large cost.

Before statistical data analysis, outliers were identified by the interquartile rule and excluded from further analyses. Substandard forms (e.g. / ka/ for / k/ik) were excluded as well. Word frequency was operationalized as the token frequency of each word in the speech addressed to the children with

typical hearing and children with CI (Guo et al., 2015; Van Severen et al., 2013). The frequencies of words addressed to the two groups of children were not determined separately, as for instance, Bergeson, Miller, and McCune (2006), Tribushinina, Gillis, and De Maeyer (2013) and Vanormelingen, De Maeyer, and Gillis (2016) have shown that CDS is similar in children with CI and peers with typical hearing. The absolute frequency counts were log transformed.

Statistical analyses

Statistical analyses were carried out in R (R Core Team, 2013) by means of multilevel modelling. A multilevel model consists of two parts: A fixed part and a random one. In the fixed part of the model, the independent variables are included. The random part considers the nesting of variables and variation in the data (Baayen, 2008). The data were structured hierarchically: Different words are nested within individual children (at different ages). This variation between children and ages was considered in the random part of the model. The analyses were divided into two parts: (1) A longitudinal analysis of children with CI and (2) cross-sectional comparisons of children with CI and children with typical hearing.

The first part of the analyses included the longitudinal data of all children with CI, from word onset (median = 18 months, range 15 - 23) up to the age of seven. A total of 49,652 word tokens were examined. For the longitudinal analysis of children with CI, the random effects of the multilevel model consisted of random intercepts and slopes for each child at each age. The fixed effects of the multilevel model were age (Age, intercept at age 20 months), the age at implant activation (Clactivation) and word frequency (Frequency). Quadratic (x^2) and cubic (x^3) effects for Frequency and Age as well as the interaction between all fixed effects were included if that yielded a better model.

In the second part of the analyses, cross-sectional analyses were performed between ages two and seven. As no longitudinal data of children with typical hearing were available, it would be incorrect to include all children with typical hearing in one model. Therefore, the data of the CI corpus were split and the analyses were performed for each age separately: At age two (range 23 - 25 months), three (range 34 - 40 months), four (range 45 - 51 months), five (range 59 - 63 months), six (range 67 - 77

months) and seven (range 82 - 86 months). A total of 67,321 word tokens were examined (children with CI: 34,707; children with typical hearing: 32,614). The distribution of the word tokens per age is shown in Table 2. For each cross-sectional analysis, the random part of the multilevel model included random intercepts for each child. The fixed effects were hearing status (Hearing status) and word frequency (Frequency). Quadratic (x²) and cubic (x³) effects for Frequency and the interaction between Hearing status and Frequency were included if that yielded a better model. Only the best fitting models will be reported and a significance level of p<0.05 was set.

Please insert Table 2 over here.

Results

Longitudinal analysis of children with CI

In this section, the longitudinal analysis of Levenshtein distance (LD) in children with CI is described. The effects of age, age at implant activation and frequency were included. The fixed effect results are displayed in Table 3.

In Figure 1, the development of LD with age is presented. It is clear from Figure 1 that LD decreases with age. In other words, the phonemic accuracy of children with CI increases, as they get older. The decrease of LD is significant, as can be derived from the effect of Age (p<0.001) in Table 3. In addition, Figure 1 shows that the decrease of LD slows down from approximately 50 months of age. This effect is also significant, as indicated by a significant quadratic effect of Age (Age², p<0.001) in Table 3. Put differently, phonemic accuracy increases as children get older, but this increase flattens out from 50 months of age onwards.

Figure 2 plots the effect of word frequency on LD for different age ranges. Regardless the age range, the global effect of word frequency seems to follow a cubic trend and more precisely an inverted s-curve. LD first increases with increasing word frequency, then decreases when word frequency further increases, but eventually increases again for words that occur most frequently (Figure 2). In other words, accuracy first decreases, then increases and finally decreases (s-curve). This developmental trend of LD with word frequency is significant, as shown by significant effects of

Frequency (p<0.01), Frequency² (p<0.001) and Frequency³ (p<0.001) in Table 3. In addition, Figure 2 indicates that the global word frequency effect is dependent on age. Up to the age of four, Figure 2 shows that more frequent words have a lower LD, and hence, are more accurate. However, there is a discontinuity. Figure 2 also shows that from the age of four onwards, LD seems to increase, and thus accuracy seems to decrease, with increasing word frequency. Moreover, this developmental trend of LD is more pronounced as children grown older. Thus, word frequency affects LD, and thus phonemic accuracy, differently at different ages and this is significant as shown by a significant interaction between Frequency and Age (p<0.001) in Table 3.

With respect to age at implantation, Table 3 shows a significant effect of CI activation (p<0.01), meaning that children with CI with later implant activation have higher LD values. However, the significant interaction between CI activation and Age (p < 0.01) suggests that the effect of Clactivation becomes less pronounced as children get older. Thus, initially, the accuracy of children with CI with later implant activation is lower compared to children with earlier implant activation. But, as children grow older, the difference between children with CI with earlier and later implant activation becomes smaller. In other words, children with CI with later implant activation are catching up on their earlier implanted peers. Table 3 finally shows a significant interaction between Clactivation and Frequency (p<0.001), meaning that the effect of frequency on LD (inverted s-curve) is less pronounced in children with CI with later implant activation as compared to those with earlier implant activation. As the interaction between CI activation, Frequency and Age was not significant and did not improve the model fit, it was not included in the best fitting model reported here. However, the lack of significance of this three-way interaction suggests that the less pronounced frequency effect on LD in children with CI with later implant activation remains stable with age. In other words, children with CI with later implant activation are not catching up on their earlier implants peers: throughout development, the effect of word frequency on LD, and thus word accuracy, is smaller in children with CI with later implant activation.

> Please insert Table 3 over here. Please insert Figures 1 and 2 over here.

Cross-sectional comparisons between children with CI and children with typical hearing

Table 4 shows the fixed effect results of the cross-sectional analyses between the ages of two and seven and Figure 3 plots the effect of frequency on the LD development per age. Firstly, there is an effect of Hearing Status on LD. Table 4 and Figure 3 show that LD is significantly higher in children with CI as compared to peers with typical hearing (p<0.05) up to the age of four and that LD is similar in both groups of children from the age of five onwards (p>0.05). In other words, word accuracy is higher in children with typical hearing up to the age of four, but similar in both groups of children from the age of four, but similar in both groups of children from the age of four, but similar in both groups of children from the age of four, but similar in both groups of children from the age of four, but similar in both groups of children from the age of four, but similar in both groups of children from the age of four, but similar in both groups of children from the age of four, but similar in both groups of children from the age of four, but similar in both groups of children from the age of four, but similar in both groups of children from the age of four, but similar in both groups of children from the age of four, but similar in both groups of children from the age of four, but similar in both groups of children from the age of four, but similar in both groups of children from the age of four, but similar in both groups of children from the age of four and that both groups of children from the age of four, but similar in both groups of children from the age of four and the age of four and that both groups of children from the age of four and the age of children from the age of four and t

Secondly, Table 4 shows significant effects of Frequency on LD and significant interactions between Frequency and Hearing Status. This means that frequency has a significant effect on LD and that this effect differs in both groups of children. The precise development of LD with frequency in both groups of children at each age is visualised in Figure 3.

At the age of two and three, Table 4 points out significant cubic effects of Frequency (p<0.001) and significant interactions between Hearing status and Frequency (p<0.001). Figure 3 shows that these linear, quadratic and cubic effects follow an inverted s-curve at the age of two and three in children with typical hearing. LD increases with increasing frequency, then LD decreases, but for words in the highest frequency regions LD increases again. In other words, accuracy decreases, then increases, but decreases again in the most frequent words (s-curve). With respect to the group differences (interaction Hearing status and Frequency), Figure 3 indicates that the developmental trend is similar in children with typical hearing and children with CI at the age of two, but that the effect is more pronounced in children with typical hearing. However, at the age of three, Figure 3 further suggests that the effect of Frequency is different in children with CI: There seems to be a slight increase of LD initially, but then there is an overall decrease of LD with increasing frequency.

From the age of four onwards, there are significant quadratic effects of Frequency (p<0.001, Table 4). As Figure 3 shows the effect of frequency is similar in both groups of children, which can be inferred from the uniform curves: First LD decreases with increasing frequency, but for the most frequent words, LD increases. Put differently, accuracy increases with increased frequency, but

decreases for words with the highest frequencies (inverted u-shape). Thus, the developmental trends of Frequency and LD are similar in both groups of children from the age of four onwards (Figure 3). Nevertheless, the effect of Frequency on LD is significantly more pronounced in children with typical hearing as compared to children with CI (see interaction effects in Table 4). In other words, the changes of LD as a function of frequency are more pronounced in children with typical hearing than in children with CI. This is true for all comparisons from the age of four onwards, but it also holds at the age of two and three. So, frequency affects accuracy in both groups of children, but to a lesser extent in children with CI.

Please insert Table 4 over here. Please insert Figure 3 over here.

Relationship between frequency and the type of words

In the previous sections, it was shown that accuracy increases with increasing frequency, except for the most frequent words. It remains to be examined what types of words are represented in these highest frequency regions. We identified two word types in the entire dataset: function words and content words, as defined in Bussmann (1996). Nouns, lexical verbs, adjectives and adverbs were labelled as content words, and for instance pronouns, conjunctions, auxiliary verbs, articles were labelled as function words. Table 5 displays the results of an additional multilevel analysis considering this question. Log frequency in CDS was included as dependent variable, word type as independent variable and the word utterance itself as random effect. The intercept represents the log frequency of content words, which is 1.8581 (SE= 0.0311). The log frequency of function words is significantly higher than content words and equals 2.8639 (= 1.8581 + 1.0058), which is significantly higher (p<0.0001). Thus, the most frequent words are function words, whereas less frequent words are content words.

Please insert Table 5 over here.

Discussion

The present paper set out to evaluate the effect of word frequency on word accuracy in the spontaneous speech of Dutch-speaking children with typical hearing and children with CI. In contrast to the literature (Goodman et al., 2008; Guo et al., 2015; Han et al., 2015; Naigles & Hoff-Ginsberg, 1998; Stokes, 2010; Stokes et al., 2012; Storkel, 2009), the frequency counts in this study were based on CDS directed to the study participants. This resulted in a direct image of the effect of word frequency on children's accuracy.

Word frequency and word accuracy in children with typical hearing

In children with typical hearing, production accuracy increases with increasing frequency, except for words with the highest frequencies, which are predominantly function words. This observation agrees well with two frequency effects reported in children with typical hearing. (1) Frequency at the segmental level affects the accuracy of the production of segments and their age of acquisition (Edwards et al., 2004; Munson, 2001; Van Severen et al., 2013) Even though our results consider word frequency and word accuracy, the effect is similar: more frequent words in the input are more accurately produced. (2) Our results are in line with the effect of word frequency on lexical acquisition in children with typical hearing. Early vocabularies of children with typical hearing contain highly frequent words, except for words with the highest frequencies, i.e. function words (Goodman et al., 2008; Naigles & Hoff-Ginsberg, 1998; Stokes, 2010; Stokes et al., 2012). Similarly, in this study, highly frequent words are more accurate, except for words in the highest frequency regions, i.e. function words. The present paper revealed that this effect of frequency is not only apparent in lexical acquisition, but also in word accuracy.

However, our results are not in agreement with the findings of Sosa and Stoel-Gammon (2012), who did not find an effect of word frequency on word accuracy in children with typical hearing. This contradiction may be due to methodological differences. First of all, the frequency counts of Sosa and Stoel-Gammon (2012) are based on adult written language, which is known to differ from child-directed speech (Akinnaso, 1982; Bernstein Ratner, 2013; Newport et al., 1977). Secondly, they used a

less fine-grained measure of accuracy, i.e. proportion of word proximity (PWP), while this study used Levenshtein distance (Faes et al., 2016).

Our study reveals that the more frequent a word, the more accurate it is produced by children. However, this effect levels out, and is even reversed for the highest frequency regions. How can this be explained? We have shown that words with the highest frequency are predominantly function words. In adult spontaneous speech, function words are often reduced, because they are short forms without sentence accent. Van Bael, Baayen, and Strik (2007) have shown that function words have high rates of phoneme and syllable deletions in Dutch. For instance, the Dutch word *natuurlijk* /n tyrləək/ 'of course', 'naturally' can be found in casual speech in various different forms, including [n ty(r)lək, ntylək, ntyk, tylək, tylək] (Ernestus, 2000) and the closed-class word *dat* ('that') is more often than not produced as [d] instead of [d t]. Most likely children integrate those frequent forms in their own speech as well. This can possibly account for the lower accuracy of very highly frequent words in children's speech. Except for the most frequent words, the main conclusion is that higher frequency results in better accuracy.

Word frequency and word accuracy in children with CI

The present study revealed that word frequency in the input also affects word accuracy in children with CI's output. As for children with typical hearing, highly frequent words are produced more accurately, except for the most frequent words. Up till now only word frequency effects on lexical development have been considered in the literature. Han et al. (2015) have shown frequent words in CDS also appear earliest in vocabularies of children with CI. Those children with CI were tested at mean ages of four and seven, i.e. one and three years after implantation. Our results complement the findings of Han et al. (2015). In contrast, Guo et al. (2015) have concluded that there was no correlation between word input frequency and lexical development in children with CI. This discrepancy may be due to methodological differences. Guo et al. (2015) studied the correlation between input frequency and vocabulary size one year after implantation, while the current study traces phonemic accuracy from immediately after implant activation up to seven yeas of age, thus we were able to follow a quite extensive developmental trajectory. Moreover, the word frequency counts

in Guo et al. (2015) are based on a corpus of CDS that is not directed to study participants, while in the present study we based the frequency counts on CDS to the study's participants.

The effect of word input frequency on word accuracy was less pronounced in children with CI with later implant activation throughout the entire period studied. In other words, later implanted children's production accuracy is influenced by the frequency of words in the input, but to a lesser extent than earlier implanted children. Later implantation seems to lead to a reduced sensitivity to the statistical regularities of the ambient language, at least up to age seven. A similar advantage of earlier implant activation has been widely reported. Better outcomes are found for children with CI with earlier implantation in morphological development (Boons et al., 2013; Nicholas & Geers, 2007; Nikolopoulos, Dyar, Archbold, & O'Donoghue, 2004), speech production accuracy (Connor, Hieber, Arts, & Zwolan, 2000; Leigh, Detmman, Dowell, & Briggs, 2013; Schauwers, Taelman, Gillis, & Govaerts, 2008; van den Berg, 2012) and lexical development (Boons et al., 2013; Houston, Stewart, Moberly, Hollich, & Miyamoto, 2012; Nicholas & Geers, 2007).

The smaller frequency effect in children with CI with later implant activation may very well be explained by their more restricted language experience as compared to earlier implanted peers. It is surprising that children with later implant activation are not catching up, whereas for other domains, this "catch up" is found after a number of years of experience. For instance, for consonant cluster production, later implantation leads to an initial delay, but a catch up is found by the age of seven (Reference). Weisleder and Fernald (2013) pointed out that the amount of language experience is crucial for processing skills: Better processing skills are found in children with more language experience. This suggests that children with CI with later implant activation have less well-developed processing skills as compared to earlier implanted peers. However, O'Grady (2015) proposed that frequency effects are processing effects. In order to pick up on frequency effects, children need to process the input adequately. Thus, good processing skills are necessary for children's sensitivity to language experience as a result of later implant activation, may be less sensitive to word frequency in the input.

Differences between children with CI and children with typical hearing

In both groups of children word frequency affects phonemic accuracy. However, the analyses have shown that the effect of word input frequency on word accuracy is more pronounced in children with typical hearing as compared to children with CI. In other words, children with CI and children with typical hearing are both sensitive to language statistics (i.c. word frequency), but this sensitivity is less developed in children with CI than in peers with typical hearing.

There are various explanations for the different degree of sensitivity to word frequency in children with CI and children with typical hearing. A first explanation is linked to the amount of language experience of children with CI. They have less language experience than children with typical hearing. Spoken language input starts later due to the initial auditory deprivation and after implantation the acoustic signal is still degraded as compared to the signal available in normal hearing (Wilson, 2006). For instance, Stelmachowicz et al. (2004) have shown that the degraded speech perception in children with CI mainly affects the perception of sounds produced at high spectral frequencies such as fricatives. This also has an effect on the production of these sounds in children with CI. Fricatives occur late in children with CI's productions (Faes & Gillis, 2016; Stelmachowicz et al., 2004) and are significantly less frequent and less accurate in comparison to peers with typical hearing (Faes & Gillis, 2016). In addition, children with CI are less attentive to speech sounds in the ambient language (Houston & Bergeson, 2014; Houston et al., 2003). The degraded signal, less extensive language experience and less attention to the ambient language may influence their sensitivity to the statistical properties of the language they hear (Saffran et al., 1996). It is possible that children with CI are only starting to discover that words differ in their frequency of occurrence. In addition, the reduced language experience is related to less developed processing skills (Weisleder & Fernald, 2013). Indeed, children with CI are found to have less developed processing skills as compared to children with typical hearing. Children with CI have problems with implicit sequence learning (Conway, Pisoni, Anaya, Karpicke, & Henning, 2011) and processing and storage of information (Burkholder & Pisoni, 2003; Cleary, Pisoni, & Geers, 2001; Grieco-Calub, Saffran, & Litovsky, 2009; Kronenberger, Pisoni, Henning, & Colson, 2013; Nittrouer et al., 2013; Pisoni & Cleary, 2003, 2004; Pisoni, Kronenberger, Roman, & Geers, 2010). Thus, processing and storage skills are affected in children with CI. As O'Grady (2015) has proposed that frequency effects can be seen as processing effects, it is possible that the effect of input frequency is related to the less developed processing skills in children with CI.

Secondly, most of the children with CI in this study had unilateral implants for a long period. Even though some children in the present study received a second implant during data collection, the age at second implantation was highly variable and the duration of bilateral device use may have been too short to fully integrate bilateral input. For lexical development, Guo et al. (2015) have already shown that unilateral or bilateral implantation affects the sensitivity to language statistics: Children with bilateral implants are sensitive to segmental frequencies, whereas in children with unilateral implants this effect is significantly less pronounced. A similar bilateral advantage may also hold for the effect of word frequency on speech production accuracy. For instance Sarant, Harris, Bennet, and Bant (2014) have shown that children with bilateral implants have enhanced vocabulary, receptive language and expressive language outcomes and Baudonck, Van Lierde, D'haeseleer, and Dhooge (2011) found better speech production skills in children with bilateral implants as compared to children with unilateral implants. These beneficial effects of bilateral implantation on speech and language outcomes may have arisen from enhanced speech perception in bilateral implanted children (Dunn et al., 2010). To date, it remains quite unclear which information available in bilateral, but not in unilateral, stimulation is relevant for the effect of word frequency on word accuracy. Future studies should examine the effect of word frequency on word accuracy in both children with CI with unilateral and bilateral implants.

Finally, a last explanation for the different degree in which frequency affects accuracy in children with CI may be found in their speech rate. Children with CI are slower speakers as compared to children with typical hearing (Burkholder & Pisoni, 2003; Vanormelingen, 2016; Vanormelingen et al., 2016). This may result in more accurate production and may reduce the importance of input frequency in these children. Studies have shown that highly frequent words are articulated more quickly in adult speech (Ellis, 2002) and that words are more often reduced when the speech rate is higher (Ernestus, 2000). Hence, highly frequent words are often less accurately produced, as they are articulated at a higher speech rate, and hence, more often reduced. The eventual effect of frequency on

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accuracy in adult speech may also be found in children. But, input frequency is strongly correlated with output frequency (r = 0.79). This means that words with high frequency in the input are also those with high frequency in the output, i.e. children's own productions. Therefore, what applies to word productions in the input also applies to those in children's own production. Thus, words with the highest frequencies in the input are most likely to be those with the highest frequency in children's output. This implies that highly frequent words in the input are also articulated faster by children, which results in more reduced, and thus less accurate, speech. As the speech rate is slower in children with CI than in peers with typical hearing, they produce highly frequent words less quickly. As a result, the overall speech rate of the sentence is lower and the effect of frequency on accuracy is lower as well.

Conclusion

In children with typical hearing, sensitivity to language statistics is shown to affect the accuracy of segments (Edwards et al., 2004) and words (Goodman et al., 2008) children produce. The findings of the present study point out that it also affects the accuracy of known words in spontaneous speech productions of children. Moreover, the present study shows that children with CI are sensitive to language statistics as well, but to a lesser extent than their peers with typical hearing. The present paper adds to the body of knowledge of frequency effects in children with CI by considering the effect of word frequency on word accuracy. One of the limitations of this study is the relatively limited number of participants in both groups of children. Further research with larger groups of CI and NH participants is needed to fully understand the contribution of word frequency to phonological representations and to understand the effect of word frequency on word accuracy. Furthermore, an analysis of the effects of children's own word production frequency on accuracy might be relevant in the discussion about frequency effects as well. Moreover, Storkel (2004) has shown that early acquired words are highly frequent in children's own productions. Thus, production frequency might be another relevant frequency effect, as perception of own production fine-tunes articulation (Moreno-Torres, 2014). In addition, own production frequency is related to word learning in children with typical hearing: Children's own babbling productions guide their perception, i.e. sensitivity to and focus on similar patterns in the input, and the experience of own word productions promotes new word learning (for a discussion, see Vihman, DePaolis, & Keren-Portnoy, 2014). Next, also effects of phonotactic probability (PP), neighbourhood density (ND) and vocabulary size should be considered together with word frequency. Even though Sosa and Stoel-Gammon (2012) have shown that these factors are not correlated in child language, including the effects PP, ND and vocabulary size in addition to word frequency makes it possible to disentangle the different factors affecting word accuracy.

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TABLES

	РТА РТА	Age	٨٥٥	Age Age	Age	Age first word			
ID	D Gender unsided CI hearing 1st CI active	activation 1 ^e CI		Chronological age	Relative to implant activation				
S 1	F	120	35	9.10	13.49	14.89	75.00	20.00	5.11
S2	F	120	27	1.13	6.69	7.66	56.00	16.00	8.34
S3	F	115	25	1.59	10.00	11.66	70.00	20.00	8.34
S4	М	113	42	10.00	18.16	19.30		20.00	0.70
S5	М	93	32	4.79	16.89	17.89	76.00	18.00	0.11
S6	М	120	37	1.69	8.76	9.66		16.00	6.34
S7	F	117	23	4.00	5.16	6.13	15.00	15.00	8.87
S8	F	112	42	2.00	19.46	21.13		23.00	1.87
S9	F	103	28	5.26	8.69	9.69	23.00	15.00	5.31
Mea	n	112.56	32.33	4.40	11.92	13.11	52.50	18.00	5.00
SD		9.12	7.11	3.28	5.25	5.39	27.03	3.00	3.38
			PTA	= Pure Tone A	verage in o	dB HL (decibel h	earing leve	el)	

Table 1.	Characteristics	of	children	with	CI
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Ages are represented in months - = no second implant

Age (years;months)	Group	Age ranges (in months)	# of word tokens
	All children		67,321
All ages	CI	23 - 86	34,707
	NH		32,614
	All children		7,628
2;00	CI	23 - 25	2,752
	NH		4,876
	All children		14,807
3;00	CI	34 - 40	12,604
	NH		2,203
	All children		12,637
4;00	CI	45 - 51	6,309
	NH		6,328
	All children		11,099
5;00	CI	59 - 63	5,834
	NH		5,265
	All children		9,587
6;00	CI	67 - 77	2,698
	NH		6,889
	All children		11,563
7;00	CI	82 - 86	4,51
	NH		7,053

Table 2. Cross-sectional analyses: ages, ages ranges and the amount of word tokens

	Estimate (SE)	
(Intercept)	18.4022 (0.8753) ***	
Age	-0.1173 (0.0184) ***	
Age ²	0.0012 (0.0001) ***	
Frequency	0.0854 (0.0316) **	
Frequency ²	-0.0753 (0.0066) ***	
Frequency ³	0.0051 (0.0004) ***	
Frequency x Age	0.0059 (0.0002) ***	
Clactivation	0.1598 (0.0589) **	
Clactivation x Age	-0.0036 (0.0011) **	
Frequency x Clactivation	-0.0044 (0.0008) ***	
p≤0.05*, p≤0.01**, p≤0.001***		

Table 3. Fixed effect results of the longitudinal analysis of children with CI

Estimate (SE) 19.19 (0.31)*** -0.83 (0.37)* 0.54 (0.11)*** -0.20 (0.02)*** 0.01 (<0.01)***	Estimate (SE) 17.93 (0.19)*** -2.39 (0.28)*** 0.27 (0.06)*** -0.10 (0.01)*** 0.01 (<0.01)***
-0.83 (0.37)* 0.54 (0.11)*** -0.20 (0.02)*** 0.01 (<0.01)***	-2.39 (0.28)*** 0.27 (0.06)*** -0.10 (0.01)***
0.54 (0.11)*** -0.20 (0.02)*** 0.01 (<0.01)***	0.27 (0.06)*** -0.10 (0.01)***
-0.20 (0.02)*** 0.01 (<0.01)***	-0.10 (0.01)***
0.01 (<0.01)***	
	0.01 (<0.01)***
0.10 (0.02)***	0.22 (0.02)***
4;00	5;00
Estimate (SE)	Estimate (SE)
17.46 (0.14)***	15.87 (0.12)***
-1.21 (0.18)***	-0.09 (0.16)
-0.27 (0.02)***	-0.16 (0.02)***
0.02 (<0.01)***	0.02 (<0.01)***
0.08 (0.01)***	0.07 (0.01)***
6;00	7;00
Estimate (SE)	Estimate (SE)
15.97 (0.08)***	15.83 (0.08)***
-0.02 (0.09)	0.05 (0.09)
-0.27 (0.02)***	-0.22 (0.02)***
0.03 (<0.01)***	0.02 (<0.01)***
0.04 (0.01)***	0.02 (0.01)***
	Estimate (SE) 17.46 (0.14)*** -1.21 (0.18)*** -0.27 (0.02)*** 0.02 (<0.01)*** 0.08 (0.01)*** 6;00 Estimate (SE) 15.97 (0.08)*** -0.02 (0.09) -0.27 (0.02)*** 0.03 (<0.01)***

 Table 4. Fixed effect results of the cross-sectional analyses between ages 2;00 and 7;00

 2:00
 3:00

left out.

Table 5. Fixed effect results of the relationship between log frequency (in the input) and word type

	Estimate (SE)
(Intercept)	1.8581 (0.0311) ***
Function words	1.0058 (0.0804) ***
p≤0.05*	*, p≤0.01**, p≤0.001***