Running title: Intelligibility in CI and NH children

Title: Long-term effects of cochlear implantation on the intelligibility of speech in French-speaking children.

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Keywords: intelligibility, cochlear implant, spontaneous speech.

Abstract

In this study, we compare the intelligibility of speech in children with a cochlear implant and children with a normal hearing. Two groups of thirteen children matched in chronological age were recorded in a narrative task. Selected excerpts of their productions were graded by naïve and expert listeners. The results show that 1) cochlear-implanted children have a lower intelligibility than their normally-hearing peers, 2) early implantation before two years of age is a predictor of good intelligibility, and 3) late implantation after two years of age does not prevent the children from eventually reaching a good level of intelligibility.

INTRODUCTION

Intelligibility can be defined as "the accuracy with which a listener correctly understands another person's spoken message as it was intended" (Svirsky et al., 2007) and is therefore crucial to successful human communication. In recent years, there has been a growing interest in using intelligibility scores as assessment tools for speech therapists (i.a. McLeod et al., 2012), to help them evaluate delays in phonological development or speech pathologies and track the progress of children undergoing speech therapy (i.e. the evolution of their speech production). Since the 1980s, intelligibility has been seen as one of the markers of a successful cochlear implantation and has been used to understand which factors can predict positive outcomes of a cochlear implantation.

Cochlear implantation is proposed to severely-to-profoundly hearing-impaired children and adults, allowing them to access audio information, and leading to a better perception of sounds of their environment in general, and speech sounds in particular. This improved perception helps them develop their speech production, and therefore their ability to communicate orally with others (Niparko et al., 2010). However, the signal provided through the cochlear implant is degraded in comparison with the audio information available with a normal hearing. Perception with an implant remains partial and for pre- and perilingually deaf CI children, it can lead to delayed phonological development and language acquisition at several linguistic levels, when compared to normally-hearing peers of the same age. Indeed, CI children produce speech sounds with less accuracy than NH peers (i.a. Chin & Pisoni, 2000; Gaul-Bouchard et al., 2007; Faes & Gillis, 2016), they have more difficulties with morphological and syntactic characteristics of speech or with narrative skills than NH peers (i.a. Le Normand, 2004; Boons et al., 2013; Geers & Nicholas, 2013) and they have a smaller lexicon and are less active in oral communication than NH peers (Briec et al., 2012). Several studies in speech and language acquisition in CI children have emphasized the role of early cochlear implantation before two years of age (i.a. Fryauf-Bertschy et al., 1997; Govaerts et al., 2002; Geers, 2004; Artières et al., 2009) as a predictor to a phonological development and language acquisition with a similar trajectory to that of NH children after several years of implant use, also confirming the existence of a critical period in language development.

The few studies on speech production presented here (among others) focus on CI children's abilities as speakers, and their language and speech developments are assessed though objective characterizations of their productions, often compared to normative data. However, studying intelligibility is a different challenge: evaluating how one's production is understood by listeners necessarily implies subjective assessment of the speakers' production by listeners.

Intelligibility has been found to be correlated with chronological age in 3;9- to 6;2-year-old children in the study by Flipsen & Colvard (2006), and in 2;5- to 18-year-olds in the study by Habib et al. (2010), but not in 4;8- to 11;1-year-olds in the study by Khwaileh & Flipsen (2010); it has been found to be correlated with age at implantation in Calmels et al. (2003), Habib et al. (2010), Svirsky et al. (2007), and Montag et al. (2014) but not in the studies of Blamey et al (2001), Flipsen & Colvard (2006), Khwaileh & Flipsen (2010); and it is correlated with hearing age in the studies by Blamey et al (2001), Flipsen & Colvard (2006), Khwaileh & Flipsen (2010), and Miyamoto et al. (1997). These studies have varying outcomes that might be explained by different speech materials considered (most studies use repeated words and utterances or even read sentences but seldom use spontaneous speech to assess the children's intelligibility), by different measurements used to judge the CI children's intelligibility (either transcriptions of speech samples or perceptual judgements of the reception of the children's production are used indifferently to assess intelligibility) and by the listeners' familiarity with pathological speech in children (speech therapists, phoneticians, students, naïve listeners).

In particular, the task used in these intelligibility studies might be the cause of variation in results, as they test different speaking abilities in children that don't always fit with Svirsky et al.'s (2007) definition of intelligibility. For example, a number of studies in CI children consider intelligibility as a level of accuracy in orthographic or phonetic transcriptions of segments, words or utterances by adult listeners (Blamey et al., 2001; Flipsen & Colvard, 2006; Montag et al., 2014), as a level of understanding of the children's speech production by other speakers (e.g. Calmels et al., 2003; Van Lierde et al., 2005), or a distance of the children's speech productions to a target (e.g. Chuang et al., 2012; Perrin et al., 1999; Poissant et al., 2006): in these studies, intelligibility is found to be lower in CI children, when compared to normative data or to NH children.

Further studies use standardized tests comprising both annotations and grades to assess intelligibility. Miyamoto et al. (1997), Habib et al. (2010), Khwaileh & Flipsen (2010), Hassanzadeh (2012) use the Beginners's Intelligibility Rating (BIT, Osberger et al., 1994): listeners (speech therapists, phoneticians, students…) are asked to rate the intelligibility of the child's speech as their perceived accuracy of sentences or segments in a repetition task, and to transcribe excerpts of the child's speech, from which a percentage of segmental accuracy is computed. Calmels et al. (2003) use the Speech Intelligibility Rating (SIR, Allen et al., 1989): the children's own speech therapists assign a grade reflecting how well other listeners (e.g. parents, speech therapists) might understand the child's connected speech, but this study does not provide direct judgements by those listeners.

The observation of the different methodologies used to assess CI children's intelligibility reveals a confusion between intelligibility and accuracy. Most of them do not assess how much of a child's speech is understandable in everyday life. This is what was targeted in the present article through listeners' subjective judgements of children's speech production. In this study, we aim at comparing intelligibility of spontaneous speech in hearing-impaired children using cochlear-implants (CI) after several years of implant use, and children with normal hearing (NH) matched in chronological age, and at understanding which factors influence the children's intelligibility. We are also interested in understanding if the listener's familiarity with pathological speech in children influences the perceived intelligibility of speech: we compare intelligibility ratings by naïve listeners vs. expert listeners (Speech and Language therapists). We use spontaneous speech as test material since it reflects the children's speaking abilities in as much an ecological setting as possible. This choice also helps avoiding an interference of other cognitive processes at play in repetition or reading tasks.

The methodology we chose is motivated by 1) the lack of studies in French-speaking CI children (only Perrin et al., 1999 and Calmels et al., 2003 provide assessments of the intelligibility of French-speaking CI children, with small groups of children and no NH control group), and 2) the need to assess intelligibility as the reception of the children's message by listeners in an ecological setting.

We question 1) the role of hearing abilities in producing intelligible speech (i.e. comparing intelligibility in NH and CI children), 2) the age factors influencing intelligibility in CI and NH children, 3) the existence of a critical period in language acquisition and the predictors in a successful cochlear implantation, and 4) the influence of expertise in judging intelligibility of children's speech (i.e. comparing judgements by expert and naïve listeners).

METHOD

The protocol consists in two stages. First, we collect spontaneous speech from children with CI and children with NH, from which sample sentences are selected for the evaluation. Then, we submit these speech samples to adult listeners, who are asked to rate their intelligibility on a 7-point scale.

Participants: children

Participants in this study are 13 CI children (six girls and seven boys) and 13 NH children (seven girls and six boys), matched in chronological age (t(23.954) = 0.039, p = .9692). The NH children were 6;5 to 10;6 years old (mean: 8;2; sd: 1;3) and the CI children were 6;6 to 10;7 years old (mean: 8;2; sd: 1;3) at the time of the recordings. For the CI children, age at implantation ranged from 1;1 to 6;6 years (mean: 3;2; sd: 1;9) and hearing age ranged from 2;2 to 9;1 years (mean: 5;3; sd: 2;3). All children were monolingual speakers of standard French, raised in the Lyon-Grenoble area. All NH children were screened for language and hearing impairments. Detailed ages of the children are given in Table 1 below.

Participants: listeners

Two groups of adult listeners participated in the study: 9 expert listeners (9 women, speech therapists/specialized school teachers) and 17 naïve listeners (8 women, 9 men, with no training in phonetics). Both groups were matched in age (t(22.345) = 0.89699, p = .3793): the expert listeners were 25;11 to 38;7 years old (mean: 30;3; sd: 4;0) and the naïve listeners were 21;3 to 43 years old (mean: 28;6; sd: 5;11) at the time of the experiment. They were all native speakers of French and were living in various regions of France at the time of the experiment. All declared that they had not been diagnosed with hearing impairments.

Speech samples

We recorded CI and NH children in a narrative task: after seeing sequences of a cartoon, the children were asked to describe to the experimenter what they had just seen, and were encouraged to give as much detail as they could. The recordings took place in quiet rooms. We used a digital Marantz PMD-670 recorder (mono, sampling frequency 44 100 Hz, 16 bits), and an external microphone placed on a tripod, approximately 40 cm from the children’s mouths.

Five independent utterances were extracted from each child’s corpus (mid part of the corpus): all minimally included a subject, a verb and a complement (word, phrase or short clause) and were preceded and followed by a pause. All excerpts were then normalized in intensity (60 dB) on Praat (Boersma & Weenink, 2015), to ensure a constant volume for all stimuli. We used a total of 130 stimuli (26 speakers*5 sentences).

Evaluation procedure

The experiment was presented to each listener in quiet rooms, using a laptop and headphones. We used a script for perception experiments on Praat (Boersma & Weenink, 2015). Participants were asked to rate the intelligibility of each utterance on a 1-to-7 point scale (Likert, 1932): with 1 for "not intelligible/understandable at all" and 7 for "fully intelligible/understandable". When needed, participants could listen to each utterance two additional times. Utterances were presented in random order. There was no time constraint. A training phase was always preceding the actual experiment: we built eight stimuli from a similar corpus recorded with the same procedure (Scarbel, 2012). The children in the training phase were different from those in the actual test phase.

Statistical analyses

Intelligibility scores on a 7-point scale can be viewed as ordered multicategorical data. Ordinal data can be modeled by several multinomial regression models (Agresti, 2002), among which the cumulative logit model. The dependency between the scores of a single child and a single listener is taken into account through random effects. The (logit transformed) cumulative probability, for child i and listener j, to have a score at most equal to c ($c = 1, \dots, 7$) is modeled through a linear regression with random effects:

$$\text{logit}\left[P(Y_{ijk} \leq c | X_{ij}, \xi_i, \xi_j)\right] = \alpha_c - X_{ij}\beta - \xi_i - \xi_j \quad (c = 1, \dots, C)$$

where Y_{ijk} is the kth score of child i given by listener j ($i = 1, \dots, I$, $j = 1, \dots, J$ and $k = 1, \dots, K$), α_c is the cutpoint-specific intercept associated to category c, X_{ij} is the design matrix,

β is a vector of fixed effects, ξ_i is the normal random effect of child i and ξ_j the normal random effect of listener j.

Estimation was obtained with the clmm function of the ordinal R library. The following factors were considered in the initial model: group: NH vs. CI, chronological age, expertise of the listener (expert vs naïve). On the basis of the studies by Fryauf-Bertschy et al. (1997), Govaerts et al. (2002), Geers (2004), and Artières et al. (2009) mentioned above, we decided to use age at implementation as a categorical variable, and to differentiate between CI children implanted before vs. after 24 months of age. The effect of hearing age could not be explored in this study due to the high correlation to age at implantation: indeed, the early-implanted children in this study are also the children with the highest hearing ages. A selection of the significant covariates based on AIC comparisons (Akaike, 1974) and ANOVA tests between embedded models leads to the following final model:

$$\text{logit}\left[P(Y_{ijk} \leq c|X_{ij}, \xi_i, \xi_j)\right] = \alpha_c - \beta_1 1_{CI\_after24,i} - \beta_2 1_{NH,i} - \beta_3 x_i - \xi_i - \xi_j$$

where β_1 and β_2 are the fixed effects of the CI children implanted after the age of 24 months (β_1) and of the NH children (β_2) with respect to the CI children implanted before the age of 24 months group, $1_{CI\_after24,i}$ and $1_{NH,i}$ are the dummy variables indicating whether child i is a CI child implanted after the age of 24 months or not, and a NH child or not, β_3 is the chronological age effect for the CI children implanted after the age of 24 months and x_i is equal to the chronological age whether child i is a CI child implanted after the age of 24 months and 0 otherwise. In summary, we have significant effects of group (NH, early-implanted CI, late-implanted CI) and of chronological age for late-implanted CI children. No significant effect is found for expertise of the listener.

The probability for any typical child to obtain a score at most c can be computed. For example, for a typical child in the NH children group, the probability of a score at most c is

$$P(Y_i \leq c|NH) = \text{logit}^{-1}(\alpha_c - \beta_2)$$

The probability of a score equal to c can also be computed from

$$P(Y_i = c|NH) = P(Y_i \leq c|NH) - P(Y_i \leq c - 1|NH)$$

.

RESULTS

Table 1 below present the distribution of intelligibility scores, given to each group of children (CI children implanted after the age of 24 months, CI children implanted before the age of 24 months and NH children).

Table 1
Distribution of intelligibility scores (percentage of all responses), given to each group of children by all listeners

	Intelligibility scores							
	1	2	3	4	5	6	7	Total
CI children, implantation after 24 months	11.06	16.25	13.85	13.27	12.12	12.98	20.48	100
CI children, implantation before 24 months	1.38	7.08	12.31	10.00	12.46	18.00	38.77	100
NH children	0.47	1.12	2.19	4.08	9.17	17.87	65.09	100

In this study, three levels of intelligibility were set: high intelligibility (scores 5 to 7), average intelligibility (score 4) and low intelligibility (scores 1 to 3). As shown in Table 1, NH children have a high level of intelligibility, as they mostly receive scores 5 (9.17 % of all scores given to them), 6 (17.87 %) and 7 (65.09 %). Similarly, early-implanted CI children have a high level of intelligibility, as they receive scores 5 (12.48 % of all scores given to them), 6 (18.00 %) and 7 (38.77 %). However, late-implanted CI children receive a more balanced distribution of scores, as they all range from 11.06 % (score 1) to 20.48 % (score 7), preventing from clearly characterizing their level of intelligibility.

The statistical analysis of the data indicates that NH children have a significantly higher intelligibility than early-implanted CI children (early-implanted CI children vs. NH children: Estimate = 1.77448, SE = 0.59372, z = 2.989, $p < .01$), and that early-implanted CI children have a significantly higher intelligibility than late-implanted CI children (early-implanted CI children vs. late-implanted CI children: Estimate = -11.26358, SE = 3.01872, z = -3.731, p <.001).

Figure 1 presents the evolution of intelligibility with chronological age in all three groups of children (late-implanted CI children with an implantation after 24 months, early-implanted CI children with an implantation before 24 months, and NH children).

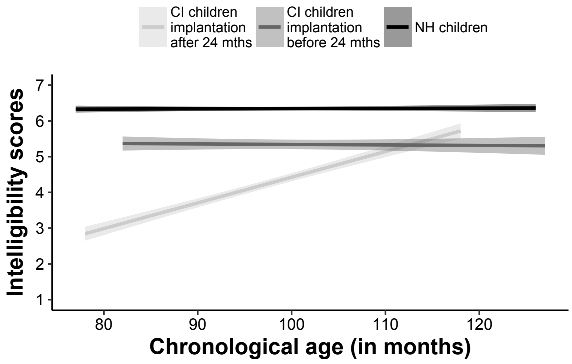

Figure 1:
Effect of chronological age (in months) on the intelligibility scores for each group of children

As shown on Figure 1, both NH children and early-implanted CI children have a stable intelligibility (from 6;5 to 10;7 years of age): even though it is lower than that of NH children as shown by the statistical analysis, intelligibility in early-implanted CI children does not evolve during the age span of our study. However, for late-implanted CI children, intelligibility is low in younger children, whereas it reaches the level of early-implanted CI children in older children. This is confirmed by the statistical analyses, as detailed in the method section: there is a significant effect of chronological age in late-implanted CI children only (Estimate = 0.10008, SE = 0.03044, z = 3.288, p <.01).

Table 2 and Figure 2 present the predicted probability for a group of children to have a low, average or high intelligibility at various ages covering the age span of our study, computed from the statistical modeling of the data, as previously detailed in the method section.

Table 2:
Predicted probabilities of intelligible speech in children (Probability higher than 0.50 in bold font)

		Predicted probability of intelligible speech		
Children's group	Chronological age (yrs)	Low intelligibility (scores 1-3)	Average intelligibility (score 4)	High intelligibility (scores 5-7)
NH children	any	0.02	0.02	**0.96**
CI children (implantation before 24 months)	any	0.09	0.11	**0.80**
CI children (implantation after 24 months)	6	**0.86**	0.08	0.06
	7	**0.64**	0.17	0.19
	8	0.35	0.22	0.43
	9	0.14	0.14	**0.72**

Table 2 allows to make predictions as to the expected level of intelligibility of the children at different ages.

As shown on Table 2, both the NH children and the early implanted CI children have a very high probability of being highly intelligible at any age (probability of high intelligibility: 0.96 for the NH children and 0.80 for the early-implanted CI children). For the late-implanted CI children however, age affects the predictions: it is not possible to predict the intelligibility for this group at the age of eight years (probability of low intelligibility: 0.35, of average intelligibility: 0.22, of high intelligibility: 0.43). On the contrary, it is possible to predict that younger children of this group will most likely have a low level of intelligibility (probability of low intelligibility: 0.86 at 6 years and 0.64 at 7 years), and older children a high level of intelligibility (probability of high intelligibility: 0.72 at 9 years), approaching that of the group of early-implanted CI children.

The last goal of our study was to understand if listeners' expertise influences the perception of intelligibility. Figure 2 shows that both groups (Experts and Naïve listeners) give overall similar scores. This is confirmed statistically, since listeners' expertise was not selected as a pertinent covariate (i.e. with a potentially significant effect) to model the data as previously explained in the method section.

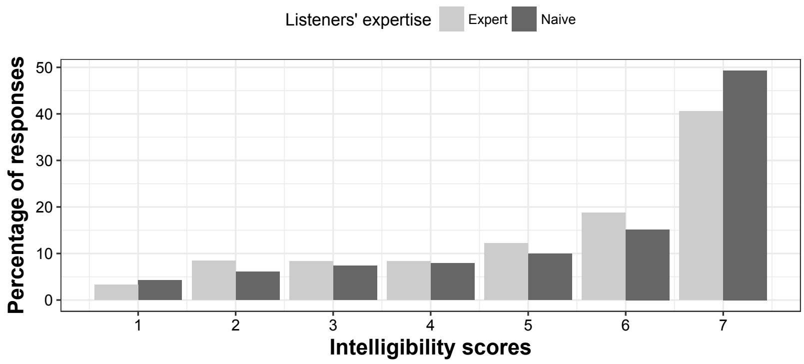

Figure 2:
Scores (in percent) for each group of listeners (Expert vs. naïve listeners)

The familiarity of expert listeners with various types of pathological speech does neither lead to a harsher judgement of intelligibility nor does it help them having a better understanding of variation in the less intelligible children's speech production than naïve listeners.

DISCUSSION

This study investigated the long-term outcomes of pediatric cochlear implantation in the intelligibility of speech. It focused on the factors at play when producing (i.e. hearing abilities, age factors) or perceiving (i.e. listeners' expertise) intelligible speech.

Our results show that even though children with a cochlear implant have gained in intelligibility several years after the implantation, their intelligibility is not as high as that of NH children. Indeed, from six to eleven years of age, their connected speech is perceived as intelligible, but even the level of the most intelligible CI children remains below that of NH children.

Age at implantation plays a part in achieving intelligibility: CI children with an implantation before the age of two years are significantly more intelligible than children with a later age at implantation. This confirms the results of several studies: Calmels et al. (2013)'s study using conversation and spontaneous production as materials in CI children with an age at implantation ranging from one to ten years; Habib et al. (2010)'s study using sentence repetitions in 2;5- to 18-year-old children with an age at implantation ranging from eight to 36 months; Svirsky et al. (2007)'s study using sentence-repetitions in 2;5 to 5-year old children; and Montag et al. (2014)'s study using sentence-repetitions in children over nine years of age. Our results for the late-implanted CI children are comparable to Blamey et al.'s (2001) study comparing intelligibility before and following implantation in which no evidence of an effect of age at implantation in late-implanted children (between 2 and 5 years) could be found. However, they differ from other studies, e.g. Flipsen & Colvard (2006) studying intelligibility in younger CI children (i.e. no effect of age at implantation in children aged 4- to 7-years) or Khwaileh & Flipsen (2010) using transcription in word- and sentence-repetition tasks as indices of intelligibility (i.e. no benefit of early implantation in 4- to 11-year old children). All these studies and our own are focused on intelligibility at different age ranges or stages following the implantation (i.e. short-term or long-term) and they all contribute to the overall description of the development of intelligible speech in CI children.

Our results on the effect of chronological age on CI children's intelligibility are in line with earlier studies: effects of chronological age were found in 3;9- to 6;2-year-old children (Flipsen & Colvard, 2006), and in 2;5- to 18-year-old children (Habib et al., 2010), but not in 4;8- to 11;1-year old children (Khwaileh & Flipsen, 2010). Indeed, comparing our results to these studies shows that intelligibility is evolving with chronological age in younger CI children, or children in a greater age range, but not in CI children of comparable age. Our results however show distinct results in early- and late-implanted children, which is a different result from the study by Khwaileh & Flipsen (2010).

The different effects of chronological age on intelligibility for the three groups of children are questioning the existence of a critical period in language acquisition and speech development, since it is not affecting the intelligibility of the NH children and the early-implanted CI children but it is affecting that of CI children with a late implantation. It could be argued that young NH children and early-implanted CI children have developed language and speech abilities early on, which helps them be intelligible at an early age (before six years), but that late-implanted CI children have a similar but longer acquisition trajectory, due to a later access to audio information and oral communication. Further explorations in younger children (before the age of five) could help us understand when the speech of typically-developing and early-implanted CI children becomes intelligible (i.e. when their probability to receive grades 5 to 7 is higher than 0.50). Similarly, studying intelligibility in older CI children would give more details on the evolution of intelligibility in children: have early-implanted CI children reached their highest level of intelligibility or did they reach a plateau in a still-evolving process of language acquisition? We were not able to study the effects of hearing age on intelligibility due to the intra-group homogenous profiles of early- and late-implanted CI children. Comparing intelligibility in two groups of early- vs. late-implanted CI children with variable lengths of device use could help us understand to what extent hearing age has an effect on intelligibility in both early- and late-implanted CI children.

Our study provides indications about the long-term outcomes of cochlear implantation for speech intelligibility, and about which factors can predict a good level of intelligibility in CI children: 1) a good intelligibility is expected more than two years after the implantation, yet remaining lower to that of NH children, 2) early implantation (i.e. before two years of age) is a predictor of a good level of intelligibility 4) late implantation (i.e. after two years of age) will most likely lead to a delayed pattern in speech intelligibility when compared to early implantation and 3) after the age of eight years, late-implanted children begin to catch up with their early-implanted peers.

Finally, our study shows that the listeners' expertise does not impact their perception of the children's intelligibility, since judgements by expert and naïve listeners were not significantly different.

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