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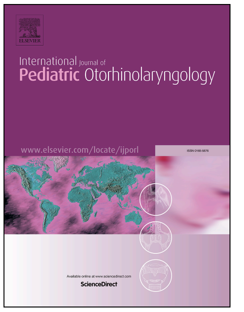

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Title

Auditory brainstem implantation in children with hearing loss: effect on speech production

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Auditory brainstem implantation in children: effect on speech production

Abstract

Auditory brainstem implantation (ABI) is a recent technique in children's hearing restoration. Up till now the focus in the literature has mainly been the perceptual outcomes after implantation, whereas the effect of ABI on spoken language is still an almost unexplored area of research. This study presents a one-year follow-up of the volubility of two children with ABI. The volubility of signed and oral productions is investigated and oral productions are examined in more detail. Results show clear developmental trends in both children, indicating a beneficial effect of ABI on spoken language development.

Keywords: auditory brainstem implantation; pediatric; oral language, volubility; sign language

1. Introduction

Hearing loss affects about 2 out of 1,000 neonates and approximately half of those children suffer from a severe-to-profound hearing loss (> 70 dB HL) [1, 2]. Congenital severe-to-profound hearing loss considerably hampers children's spoken language development. Since a couple of decades, electronic devices have been designed to partially restore severe-to-profound sensorineural hearing loss. Depending on the locus of the deficit, a cochlear implant or an auditory brainstem implant is suited. A cochlear implant bypasses damaged hair cells in the cochlea and directly stimulates the auditory nerve. An auditory brainstem implant (ABI) is warranted when the hearing loss results from a damaged cochlea, in which no cochlear implant can be implanted, or from the absence of an auditory nerve [3].

Paediatric cochlear implantation has become a fairly common means of hearing restoration in cases of inner ear malfunction. A considerable amount of research has investigated the effects on language development [e.g. 4, 5-8]. Pediatric ABI implantation is a more recent development. As a result very little is known about the effect of ABI implantation on young infants' language development. This paper is among the first linguistically motivated studies to investigate the effect of ABI implantation on oral language development.

1.1. Auditory brainstem implantation in children

Initially, an ABI was designed to restore hearing loss in adults and teenagers with postlingual deafness due to neurofibromatosis type 2 (NF2). The first prototypes of an ABI for NF2 patients were already designed in the ‘80s [3]. Since 2001, ABI surgery has been broadened in Europe to children as well as to adults and children with other pathologies than NF2, such as the absence of the auditory nerve, damaged or ossified cochlea, and cochlear nerve aplasia. Since 2013, the FDA has approved the first clinical trials of pediatric ABI implantation in the U.S. as well [3].

With respect to ABI in adults, research has already shown a clear progress in speech perception and language comprehension skills, even in the absence of lip-reading [9, 10]. With respect to ABI in children, the few studies available in the literature thus far mainly focused on infants’ perceptual progress. Children’s perception of sounds improves steadily with longer ABI use, reaching levels of mild-to-moderate hearing loss (auditory thresholds between 30 and 60 dB HL) [11]. They are aware of environmental sounds, can discriminate and identify sounds and phonetic contrasts, such as the difference between /m-s/ or /i-u/ [12, 13]. Most children are also able to understand at least simple phrases [11, 14, 15]. Better speech perception skills are found in children with earlier implantation (i.e. before the age of three) and in children who have lower hearing thresholds after surgery [11, 15].

The development of speech production after ABI implantation is an almost unexplored area of research. To the best of our knowledge, only a handful of studies investigated the speech and language outcomes after ABI surgery in children and adolescents [3, 12, 13, 16-18]. Except for Eisenberg, Hammes Gangly, Martinez, Fisher, Winter, Glater, Schrader, Loggins, Wilkinson and The Los Angeles Pediatric ABI Team [16], these studies mostly reported very general indications of the emerging language skills, such as the presence of vocalizations, words and sentences, without documenting spoken language development in more linguistic detail. Eisenberg, Hammes Gangly, Martinez, Fisher, Winter, Glater, Schrader, Loggins, Wilkinson and The Los Angeles Pediatric ABI Team [16] analyzed word patterns, vowel and consonant features at the phonemic level and phonemic accuracy thereof in 4 children with ABI. One or two years after implantation, these children started to use basic word patterns and showed development in vowel and consonant features with varying degrees of accuracy.

Eisenberg, Hammes Gangly, Martinez, Fisher, Winter, Glater, Schrader, Loggins, Wilkinson and The Los Angeles Pediatric ABI Team [16]’s results are based on children’s productions in a picture naming task (spontaneous or imitative). Yet, there is no mention of the number of sounds and words used in daily life or the linguistic characteristics of these vocalizations. Nevertheless, such information is key to determine whether the benefit of ABI implantation outweighs the surgical risk. Recently, Sennaroglu and Ziyal [19] for instance stated that "follow-up [studies] are necessary to validate the effectiveness of ABI in […] language development" (p. 441). In this perspective, Noij, Kozin, Sethi, Shah, Kaplan, Herrmann, Remenschneider and Lee [20] also highlighted that "there is [still] a lack of prospective outcome data on pediatric ABI patients" (p. 740). As a result, for instance speech and language therapy for children with ABI is entirely based on speech and language therapy of children with cochlear implants, since both groups of children have a similar background (i.e. severe-to-profound sensorineural hearing loss). However, it has not been investigated if children with ABI's language development after implantation is similar to that of children with CI after implantation. By consequence, it is unknown if children with ABI benefit from the same type of speech and language therapy as children with CI.

1.2. Aims of the present study

Children with ABI are often implanted from the age of 2 onwards [3]. So it is to be expected that they are already relatively good signers at the moment of (partial) hearing restoration. The present study investigates the effect of ABI implantation on children's spoken language development in daily life. The use of speech is investigated on three levels.

The first question addressed is: do children with ABI shift from using predominately signed to predominately oral productions? In children with cochlear implants, it was shown that oral language production vastly increases after cochlear implantation [e.g. 21]. In general, perception and language gains after ABI implantation are found to be slower than those of children with cochlear implants [16, 19, 22]. Therefore, it is expected that the children with ABI will use some oral language but will still use a large amount of sign language as well.

Second, spoken utterances are further investigated in terms of the ratio of lexical and prelexical productions and the development of this ratio with longer ABI use. In typical language development, as well as in children with cochlear implants, children first use prelexical productions predominantly and shift to lexical productions with age or longer implant use [e.g. 23, 24, 25]. In other words, their productions become more mature with development. It remains to be seen if this is the case in children with ABI as well.

Third, the development of prelexical utterances is further examined in terms of the ratio of vocalizations and canonical babbling. In typical language development [e.g. 26] and language development of children with cochlear implants [e.g. 27], the course of development is first a predominant use of vocalizations, which shifts to canonical babble with age or longer device use. It is investigated whether the prelexical utterances of children with ABI follow this typical course of language development.

2. Method

2.1. Participants

ABI implantation is a recent development in hearing restoration. In Belgium, only eight children under the age of five received an ABI since 2015. In order to be included in this study, children have to grow up in the Dutch-speaking part of Belgium. Three children with ABI implantation are currently in follow-up. All parents signed an informed written consent for participation. The study was approved by the Ethical Committee. All children have a congenital bilateral sensorineural hearing loss, without any other documented health or developmental problems. In the present study, other inclusion criteria were implantation around the second birthday and having completed at least one year of monthly follow-up. One child was excluded from the study as it was implanted at a later age and has not yet completed a one-year follow-up period.

S1 is a three-year-old female child who was implanted with a Med-El Synchrony ABI at the age of two (2;00). The sensorineural hearing loss was due to the absence of the auditory nerves. Her pure tone average hearing threshold before implantation was 120 dB HL. Two months after the surgery, the ABI was fitted and nine of the 12 electrodes were activated. Two years after surgery, the child's pure tone average hearing threshold had improved to 37.5 dB HL. We followed the child monthly for a period of one year, starting at the age of three years and two months (3;02) up to the age of four years and three months (04;03). The child and her environment used oral language (Flemish Dutch), largely supported with Flemish Sign Language.

S2 is a four-year-old female child who was implanted with a Med-El Concerto ABI at the age of two years and one month (2;01). Also in her case the sensorineural hearing loss resulted from the absence of the auditory nerves. Her pure tone average hearing threshold before implantation was 116 dB HL. Two months after surgery, the ABI was fitted and nine of the 12 electrodes were activated. Two years after surgery, the child's pure tone average hearing threshold had improved to 43 dB HL. We followed the child monthly for a period of one year, starting at the age of four years and one month (4;01) and ending at the age of five years (5;00). The child and her environment used oral language (Flemish Dutch), supported with Flemish Sign Language.

2.2. Data collection and transcription

Data collection consisted of monthly one-hour video-recordings of spontaneous, unstructured interactions between the child, her caregiver(s) and sometimes also a sibling. The video-recordings were transcribed in CHILDES' CLAN according to the CHAT conventions [28]. Each utterance was identified and the speaker was labeled (child, one of the parents, sister). Vegetative and pure (dis-)comfort sounds, such as laughing and crying, were excluded from transcription [29]. The oral and the signed productions of all participants were further transcribed. An interpreter of sign language transcribed signed productions. Oral productions were transcribed by the first author as either a lexical production or a prelexical production, following the procedure articulated by Vihman and McCune [30]. Lexical productions equal words, prelexical ones equal all precursors to words, i.e. canonical babble and other types of vocalizations. Prelexical utterances were coded following the scheme proposed by Koopmans-van Beinum and van der Stelt [29]. In that framework, canonical babbling was defined as the production of at least two sequences of a consonant and a vowel [26]. Vocalizations were defined as all other types of prelexical utterances.

Transcription reliability was checked for the transcription of oral and signed productions. The reliability of the transcription of oral productions was checked for about 10% of the data by a second transcriber. The agreement on whether there was a lexical and/or prelexical item in the utterance equaled 85.26%. The agreement on whether a prelexical utterance was a canonical babble or a vocalization equaled 84.46%. For the signed productions, about 20% of the data were retranscribed by a second and a third interpreter of sign language. The mean agreements on whether or not there was a sign in the utterance and the number of signs in the utterance equaled 89.15% (SD = 0.02) and 85.62% (SD = 0.02) respectively.

2.3. Data analyses

The development of the children's spontaneous language productions was examined in terms of their volubility with longer ABI use. Volubility was assessed by counting the number of individual signs, prelexical productions (precanonical vocalizations and canonical babbles) and lexical productions (words). Three aspects were investigated: (1) the amount of signs versus that of oral productions, (2) the amount of lexical productions versus that of prelexical productions, and (3) the amount of canonical babble versus precanonical vocalizations. In the Appendix, the raw numbers of these counts can be found. For the statistical analyses, these counts were normalized in order to account for, for instance, differences in the length of the video-recordings, etc. For the normalization a bootstrapping procedure was applied following Molemans, Van den Berg, Van Severen and Gillis [31]. For each count 10,000 random resamples were taken for each monthly data file. The sample size of the resamples was determined by the smallest number of data available for one particular file, following the procedure proposed by Molemans et al. (2012). Once set, the sample size was held constant over the different data files [31]. The mean value after this bootstrap procedure was considered as a reliable representation of the 'real' value [31].

In addition to the number of vocal and signed productions, an estimate of the children's cumulative vocabulary was made. The reason is that a similar number of word tokens and/or signed tokens can represent different numbers of distinct word/sign types, and thus vocabulary size. For instance, a child who utters 100 word tokens can in principle repeat one particular word 100 times, or these 100 utterances can be 100 different word types. Therefore, an additional analysis of cumulative vocabulary was performed. Cumulative vocabulary was determined as follows: in a first data file, i.e., the recording at the youngest age, the number of distinct word types per child was counted. In the consecutive data file, i.e., in principle one month later, each new distinct word type was added to this number, representing the cumulative vocabulary at this point, and so on [32]. In the present study, inflected forms of a particular lemma were counted as different types. For instance the noun *book* and the plural thereof, *books,* are counted as two different word types. Cumulative vocabulary was counted for word types and signed types only, since prelexical utterances were coded following a scheme with only 8 possible codes (Koopmans-van Beinum & van der Stelt 1986). The cumulative vocabulary outcomes provide an assessment of the diversity of each child's lexical richness, allowing a more qualitative interpretation of the quantitative volubility results.

2.4. Statistical analyses

Statistical analyses were performed in R [33] using generalized linear regression. The estimates and standard errors (SE) of logistic regressions are computed in logits in R. They can be converted to probabilities [34]. For each child, three analyses were performed. First, the likelihood of signs was investigated as compared to all other oral productions (i.e. both lexical and prelexical productions). Second, in the subset of oral productions, the likelihood of lexical productions (words) was compared to that of prelexical ones (i.e. both vocalizations and babble). Finally, in the subset of prelexical productions, the likelihood of canonical babble was compared to that of precanonical vocalizations. In each analysis, the length of device use, referred to as the child's hearing age, was entered as an independent variable. Hearing age was centered at the start of the data collection, i.e. at 14 months (S1) and 24 months (S2) of hearing age. All models were fitted in a stepwise manner: quadratic and cubic effects of hearing age were only retained if they yielded a better fitting model.

3. Results

Oral language volubility was assessed in two children and development with longer device use (hearing age) was examined for each child. In the tables, the results are presented in logits. For the sake of familiarity, the logits were converted into probabilities or proportions in the discussion of the results and the figures respectively.

3.1. Development of S1

The fixed effect results of the three research questions for S1 can be found in Tables 1, 2 and 3 (expressed in logits) and Figures 1, 2 and 3 (expressed as proportions). For S1, data collection started about a year after ABI surgery.

First, the likelihood of signs is compared to that of all oral productions (Table 1). Results show that the likelihood of signs is significantly lower (45.53%) than that of oral productions (54.46%) at the intercept, i.e. 14 months of hearing age ($p<.001$). However, there is a significant quadratic effect of hearing age ($p<.001$). As Figure 1 shows, the likelihood of signs first increases, and then levels off at a proportion of .50 (50%) and starts to decrease very slightly (but significantly) from 24 months of hearing age onwards.

In a second analysis, the likelihood of spoken lexical productions (i.e. words) is compared to that of prelexical ones. Results in Table 2 show that the likelihood of lexical productions is 25.18% at the intercept, which is significantly lower than the likelihood of prelexical productions (74.82%, $p<.001$). There is a significant quadratic effect of hearing age ($p<.001$, Table 2), plotted in Figure 2. As Figure 2 shows, there is first a decrease of the likelihood of lexical productions, but the likelihood of lexical productions starts to increase from 20 months of hearing age onwards.

For the third and final analyses, prelexical utterances are further examined (Table 3). At 14 months of hearing age, a prelexical utterance is significantly less likely to be a canonical babble (14.72%) than a precanonical vocalization (85.28%) ($p<.001$). Figure 3 and Table 3 show that the likelihood of canonical babbling increases with hearing age ($p<.001$), but the significant quadratic effect of hearing age indicates that this increase becomes less steep over time ($p=.005$).

Table 1. *The likelihood of signed vs. oral productions – in logits*

	Estimate	SE	Z-value	P-value
Intercept	-0.179	0.034	-5.236	<0.001
Hearing age	0.105	0.014	7.313	<0.001
Hearing age^2	-0.008	0.001	-5.329	<0.001
Hearing age^3				

[a] standard error

The intercept represents the likelihood of signs

Table 2. *Oral productions: likelihood of lexical productions vs. prelexical ones – in logits*

	Estimate	SE	Z-value	P-value
Intercept	-1.089	0.058	-18.615	<0.001
Hearing age	-0.169	0.026	-6.575	<0.001
Hearing age^2	0.011	0.003	4.153	<0.001
Hearing age^3				

[a] standard error

The intercept represents the likelihood of lexical productions

Table 3. *Prelexical productions: likelihood of canonical babble vs. vocalizations – in logits*

	Estimate	SE	Z-value	P-value
Intercept	-1.757	0.095	-18.503	<0.001
Hearing age	0.234	0.037	6.286	<0.001
Hearing age^2	-0.010	0.003	-2.838	0.005
Hearing age^3				

[a] standard error

The intercept represents the likelihood of canonical babble

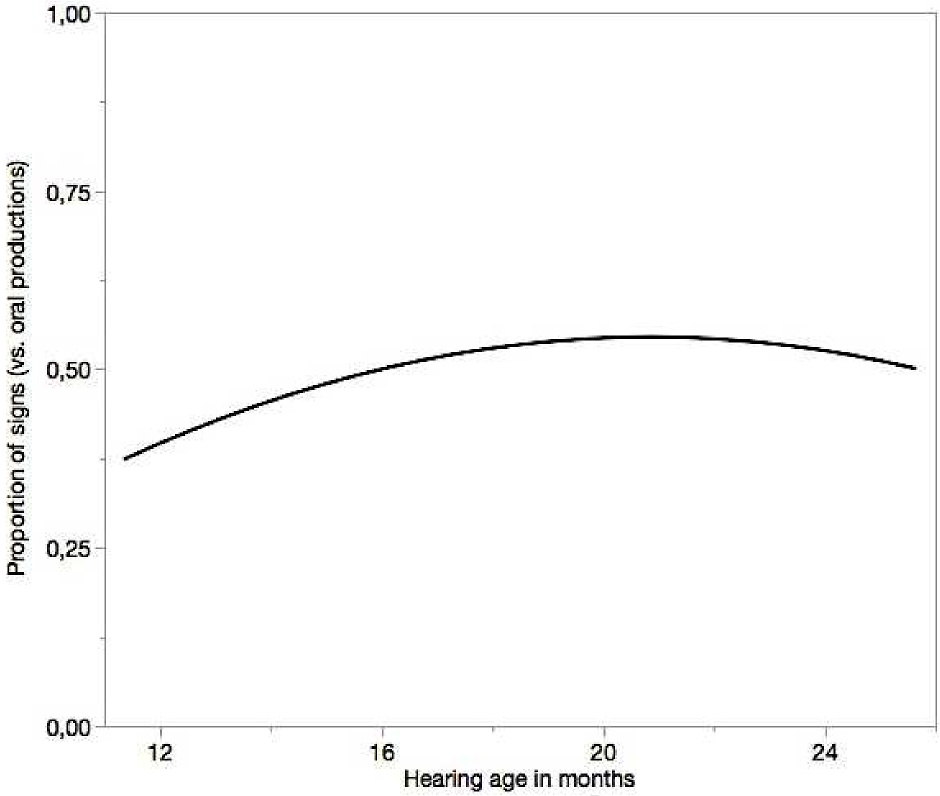

Figure 1. *Proportion of signs (vs. oral productions) with hearing age – predicted values S1*

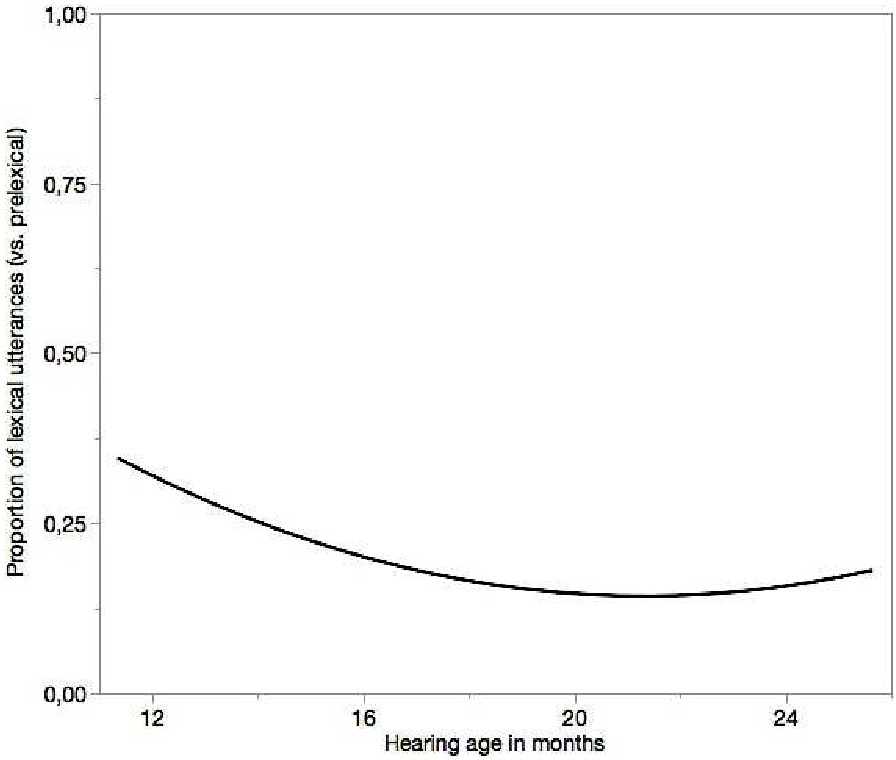

Figure 2. *Proportion of lexical productions (vs. prelexical) with hearing age – predicted values S1*

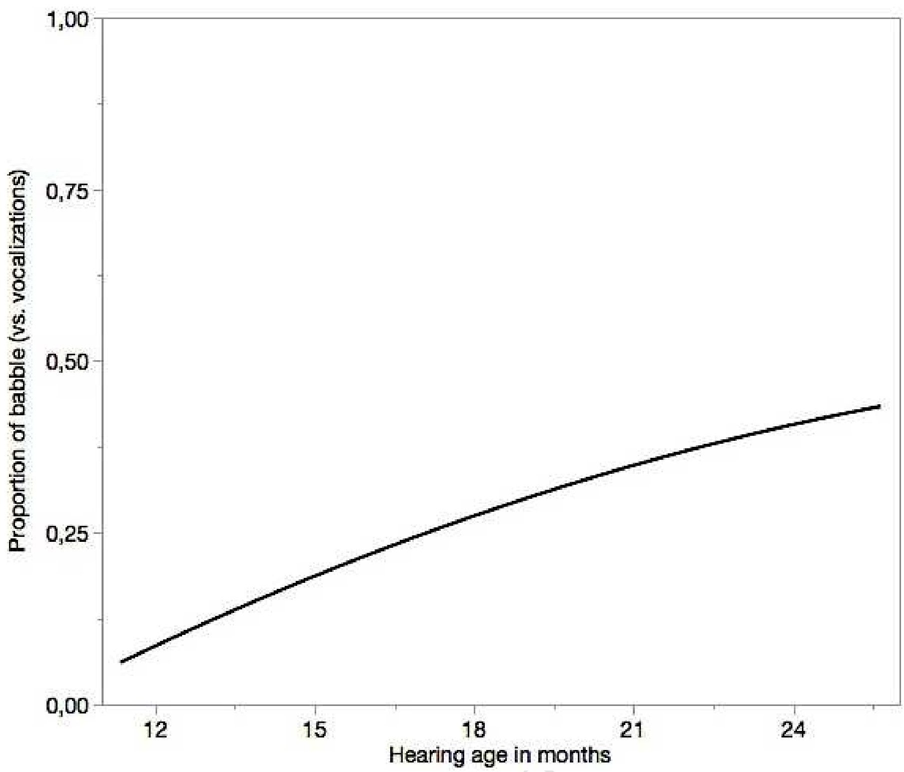

Figure 3. *Proportion of canonical babble (vs. vocalizations) with hearing age – predicted values S1*

3.2. Development of S2

The fixed effect results for the three research questions of S2 can be found in Tables 4, 5 and 6 (logits) and Figures 4, 5 and 6 (proportions). For S2, data collection started at an hearing age of 24 months.

For the first research question, the likelihood of signs is compared to that of all oral productions. The likelihood of signs is significantly lower than that of oral productions (Table 4). At the intercept, i.e. at 24 months of hearing age, the likelihood of signs is 23.92%, whereas that of oral productions is 76.08%. Even though the likelihood of signs slightly increases first (p=.016), Figure 4 indicates that it further decreases after about 27 months of hearing age (p<.001). In addition, Figure 4 clearly shows that the likelihood of signs is significantly lower than that of oral productions in the entire period and even never exceeds 25%.

For the second and third research questions, oral productions are further scrutinized. For the second research question, the likelihood of lexical productions (i.e. words) is compared to that of prelexical ones (i.e. babble and vocalizations). At the intercept, the likelihood of lexical productions is 44.40%, which is significantly lower than that of prelexical productions

(55.60%) (Table 5). This indicates that the child uses more vocalizations and/or babble than words at 24 months of hearing age. But, as Table 5 and Figure 5 show, there is an overall increase of word use: first, the likelihood increases (p<.001), then decreases slightly (p<.001) and finally increases again (p=.012). Lexical productions become more likely than prelexical ones from about 27 months of hearing age onwards (Figure 5).

For the third research question, the nature of prelexical utterances is examined by comparing the likelihood of precanonical vocalizations and canonical babble. At 24 months of hearing age, a prelexical utterance is significantly less likely to be a canonical babble (34.39%) than a vocalization (65.61%, p<.001, Table 6). However, there is an overall increase of babbling, as indicated by a significant cubic effect of hearing age (p<.001) in Table 6. In Figure 6, this increasing likelihood of canonical babble is plotted.

Table 4. *The likelihood of signed vs. oral productions – in logits*

	Estimate	SE	Z-value	P-value
Intercept	-1.157	0.062	-18.545	<0.001
Hearing age	0.087	0.033	2.409	0.016
Hearing age^2	-0.019	0.004	-5.197	<0.001
Hearing age^3				

[a] standard error

The intercept represents the likelihood of signs

Table 5. *Oral productions: likelihood of lexical productions vs. prelexical ones – in logits*

	Estimate	SE	Z-value	P-value
Intercept	-0.225	0.060	-3.784	<0.001
Hearing age	0.577	0.053	10.973	<0.001
Hearing age^2	-0.075	0.015	-4.981	<0.001
Hearing age^3	0.003	0.001	2.502	0.012

[a] standard error

The intercept represents the likelihood of lexical productions

Table 6. *Prelexical productions: likelihood of canonical babble vs. vocalizations – in logits*

	Estimate	SE	Z-value	P-value
Intercept	-0.646	0.119	-5.444	<0.001
Hearing age	0.603	0.102	5.919	<0.001

Hearing age^2	-0.12	0.028	-4.648	<0.001
Hearing age^3	0.007	0.002	3.753	<0.001

[a] standard error

The intercept represents the likelihood of canonical babble

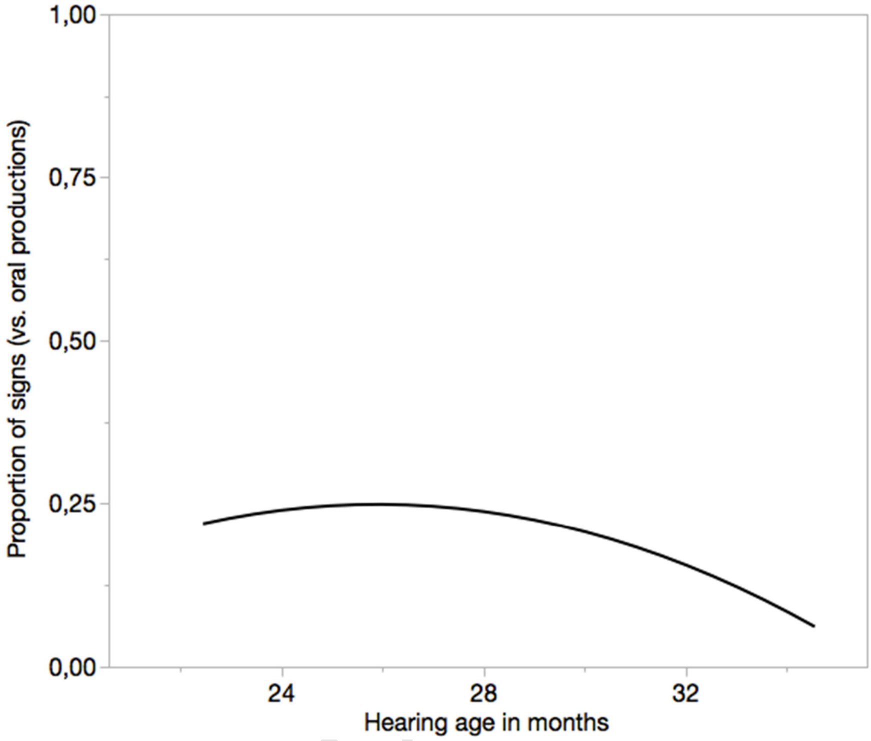

Figure 4. *Proportion of signs (vs. oral productions) with hearing age – predicted values S2*

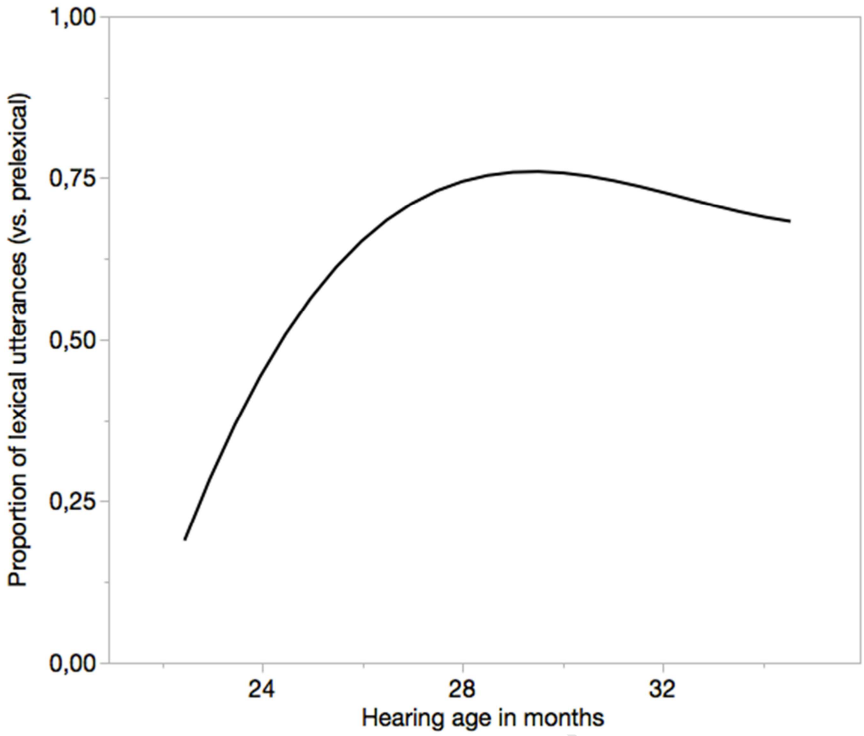

Figure 5. *Proportion of lexical productions (vs. prelexical) with hearing age – predicted values S2*

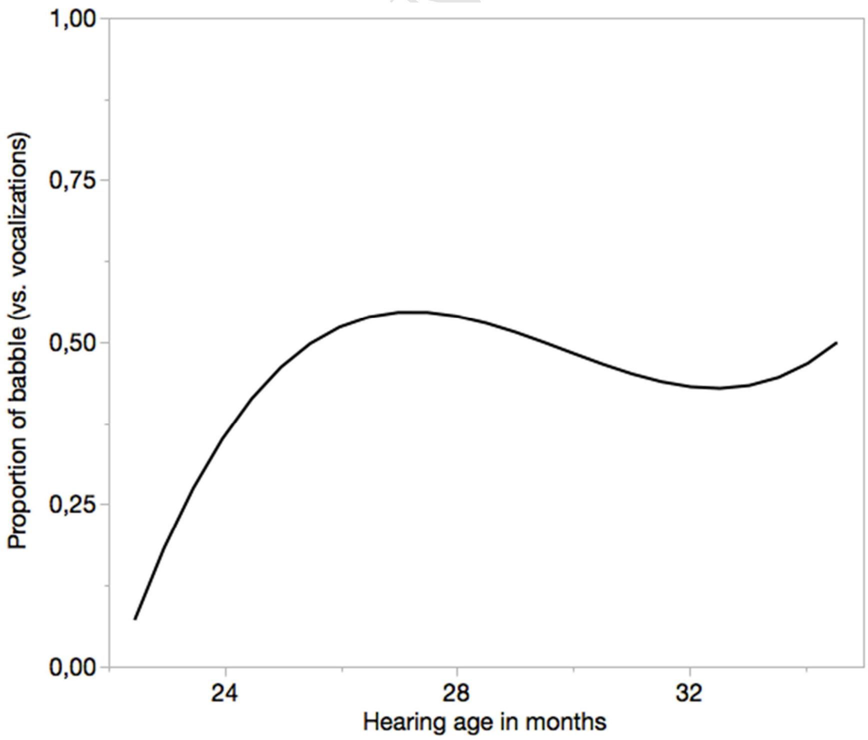

Figure 6. *Proportion of canonical babble (vs. vocalizations) with hearing age – predicted values S2*

3.3. Cumulative vocabulary of word types

Thus far, the number of oral/signed tokens was counted. But even when normalized, these quantitative counts are best interpreted against the background of word/sign types, i.e. the number of distinct words/signs in the children's vocabularies. In this way, a qualitative interpretation is offered in terms of diversity in children's productions. For instance, a similar number of word tokens (i.e. the quantitative results, e.g. 100 word tokens) can be distributed over 6 distinct word types in one child and 40 in another child. Therefore, a cumulative vocabulary count for signs and oral productions was performed for both children. Prelexical utterances cannot be counted cumulatively, since there were only 8 distinct codes in the scheme of Koopmans-van Beinum and van der Stelt [29]. In Table 7, the results of the cumulative vocabulary in words and signs of participant S1 can be found and in Table 8 those of participant S2. For S1, the cumulative spoken vocabulary increases from 4 to 77 word types and the cumulative signed vocabulary increases from 65 to 533 sign types. For S2, the cumulative spoken vocabulary increases from 77 to 343 word types and the signed one from 79 to 254 sign types. Additional statistical analyses (linear regressions) indicate a significant increase with longer device use (hearing age) in both children and for both cumulative vocabulary in words and in signs ($p<.001$ in each analysis).

Table 7. *Cumulative vocabulary counts of S1*

Age in months	Hearing age in months	Cumulative vocabulary of words	Cumulative vocabulary of signs
38	**14**	4	65
39	**15**	8	139
41	**17**	16	180
42	**18**	35	240
43	**19**	35	269
44	**20**	41	294
45	**21**	45	327
46	**22**	49	353
48	**24**	52	433
49	**25**	59	473
50	**26**	72	519
51	**27**	77	533

Table 8. *Cumulative vocabulary counts of S2*

Age in months	Hearing age in months	Cumulative vocabulary of words	Cumulative vocabulary of signs
49	**24**	77	79
50	**25**	119	106
51	**26**	160	144
52	**27**	185	168
53	**28**	231	194
54	**29**	266	210
55	**30**	279	216
56	**31**	295	225
57	**32**	308	234
58	**33**	322	240
59	**34**	331	245
60	**35**	343	254

4. Discussion

The present paper investigated the effect of ABI implantation on spoken language development in children. The oral language volubility of two children was followed monthly for a one-year period, starting 14 and 24 months after implantation respectively. As such, a period between one and three years of device use was covered. The hearing age of S1 at the end of data collection corresponds to and overlaps with that of S2 at the beginning of data collection. Moreover, the cumulative spoken vocabulary counts show continuity as well: at the end of data collection of S1, the cumulative vocabulary (in words) is about the same as that at the start of data collection of S2. This makes it possible to track a long-term development after ABI implantation, be it that the data come from only two participants. The aim of the study was threefold: (1) examine the relationship between oral and signed productions with hearing age, (2) examine the development of lexical and prelexical productions with hearing age, and (3) examine the development of prelexical productions, i.e. precanonical vocalizations and canonical babble, with hearing age. In a first part, the results pertinent to the first aim (1) will be discussed. In a second part, the results for aims 2 and 3 will be discussed together. In each part, limitations, implications and suggestions for further research are included.

4.1. Oral versus signed volubility (aim 1)

Results showed that both children use sign language in their daily communication, even though there were some individual differences as well. Whereas not more than 25% of the utterances of S2 (four years of age) consist of signs, this percentage is about 50% for S1 (three years of age). But, these results also indicate that both children's productions are above and at 50% for oral productions. In other words, even though sign language is an important part of the children's communication, oral language is as prominent or even more prominent in their communication. This result is in line with children with cochlear implants. For instance Svirsky, Stallinfs, Ying, Lento and Leonard [35] showed that cochlear implantation has a beneficial effect on the amount of oral language production as compared to oral language production before implantation. Similarly, the results of the present paper show that ABI implantation has not only an effect on perceptual abilities [as shown by e.g. 11, 13, 15], but also on spoken language production in children implanted by the age of two.

The difference in the quantity of signs and oral productions is also reflected in a more qualitative measure of the children's speech, i.e. their cumulative vocabularies of sign types. The cumulative signed vocabulary is higher for S1 than for S2, even though S1 is younger. The individual differences in amount of sign language can be attributed to different factors. First of all, there is a one-year difference in hearing age between the children. Data collection for S1 started at 14 months of hearing age, whereas data collection started at 24 months of hearing age for S2. This means that S2 has more experience with the ABI as compared to S1. It is seems likely that the length of device use affects the amount of oral and sign language in children with ABI, as it is for instance also found in children with cochlear implants [35]. Indeed, the effect of hearing age was significant in both S1 and S2, showing a quadratic, decreasing trend. In other words, after some months of device use, the number of signed productions diminished in both children's spontaneous interactions. More research should be done to disentangle the precise effect of device use on the relative amount of oral and signed production. In the present dataset, there was a small overlap between the data of the two children: the hearing age of the last three data files of S1 are similar to those of the first three of S2. Still, at this point, S1 uses more often sign language as compared to S2.

This individual difference in signed productions may also be explained by a second factor, namely the amount of sign language in the children's environment. Even though not quantified in the present paper, it might be that (child-directed) sign language has a more prominent role in S1's language environment than in S2's language environment. For oral language, studies have already shown that the quantity and quality of language input affects children's own language productions [children with cochlear implants: 36, e.g., typically developing children: 37]. It seems very plausible that a comparable effect is present in sign language: if a child gets more signed input, it is very likely the child herself uses more signs as well. Moreover, research in children with cochlear implants has shown better outcomes of oral language development for children with a predominant oral communication mode [38-41]. It may be possible that a similar effect is present in these children with ABI. Future research may examine the effect of communication mode in children with ABI on oral language use.

4.2. Oral language development (aims 2 and 3)

Oral language development was further scrutinized on two levels: the likelihood of lexical versus prelexical productions (aim 2) and, within prelexical productions, the likelihood of precanonical vocalizations versus canonical babble (aim 3). The production of canonical babble and words (lexical utterances) are generally seen as the first linguistic milestones in children's language development [42, 43]. Their presence is thus of great importance for later language development.

In both children, lexical and prelexical utterances appeared, and within prelexical utterances, vocalizations as well as canonical babble were well represented. The individual differences seem to be due to the difference in hearing age, as it was for the likelihood of signed versus oral productions. Prelexical utterances were dominant in both children's oral productions at the start of data collection (i.e. 14 and 24 months of age). S1 showed first a significant decrease of word use. But from 24 months of hearing age onwards, a slight and significant increase in lexical productions was found. In addition, the proportion of word use of S1 reaches a level at about 24 months of hearing age that is similar to that of S2 at this point. Moreover, the cumulative vocabulary of word types is similar in both children at this point as well. In other words, the characteristics of oral language development at 24 months of hearing age are comparable in both children. The ongoing development at later hearing ages of S2 shows an overall increase of lexical productions, reaching a proportion of .75 at 36 months of hearing age. Since the proportion of lexical productions is similar at 24 months of hearing age in both children, a similar development trend with continuing ABI use seems plausible for S1.

A similar picture is observed for prelexical utterances: first precanonical vocalizations are more prominent, but with hearing age, canonical babbling increases. In addition, the proportion of canonical babble at 24 months of hearing age is the same for the two children (about .35). With more extended ABI use (higher hearing age), S2 shows a further increase of canonical babbled productions, resulting in an equal likelihood of canonical babble at 36 months of hearing age.

The children in our study showed a slow, but clear progress after ABI implantation. At first, the oral productions of both children mainly consist of prelexical utterances, and more precisely precanonical vocalizations. By two years of device use, canonical babble becomes the more prominent type of prelexical utterances. Moreover, the number of lexical productions (words) increases with hearing age in both children. After 24 months of device use, 25% of the oral productions are lexical and lexical utterances represent even 75% of S2's oral productions at 36 months of hearing age. In other words, both linguistic milestones, i.e. canonical babbling and lexical production, appear in the children's development. In the period studied, their productions become more mature with hearing age: from precanonical vocalizations to canonical babble, and from prelexical to lexical. These developmental trends follow the course of development that has been observed in typically developing children and children with cochlear implants [23-27].

In-depth comparisons of the proportion of lexical and prelexical utterances, vocalizations and canonical babble with children with normal hearing of the same age as well as comparisons with children with cochlear implants of the same age and/or hearing age still leave to be desired. For Dutch, Molemans, Van Severen, Van den Berg, Govaerts and Gillis [24] have for instance shown that the proportion of lexical utterances is 80% (versus 20% prelexical utterances) at 24 months of age in children with normal hearing. This seems significantly higher than a proportion of 25% lexical productions in the children with ABI at 24 months of hearing age (48 and 49 months of chronological age). Future research is needed to investigate where children with ABI end up relative to their peers with normal hearing. Even though children with ABI's speech production seems to develop slower than that of children with cochlear implants, future studies may examine whether their volubility is similar to children with cochlear implants with the same length of device use. Thus far, we have shown that the developmental trend towards more mature oral productions is in line with typical language development and language development of children with cochlear implants.

4.3. Conclusions

The present study has shown that improved speech perception after ABI implantation results in oral language production in these children, next to the use of sign language. Even though sign language is still very important in their daily communication, both children start to use more oral language with longer ABI use. In addition, maturation in oral development was observed over the two-year period of device use: from predominantly precanonical vocalizations 12 months after implantation, to a predominant use of canonical babble at 24 months of hearing age, to a predominant use of words at 36 months of hearing age. Moreover, even within the one-year follow-up of each child, their language productions became more mature with age, which is similar to typically developing children and children with cochlear implants.

Since very little is known about the effect of ABI implantation on oral language production in children, even our limited sample of two case studies with almost non-overlapping (hearing) ages adds to the body of knowledge of language production after pediatric ABI implantation. The current results are among the first linguistically motivated ones, showing a beneficial effect of ABI on oral language production. Since ABI is a recent development in pediatric hearing restoration, the pool of participants of children with ABI is still small. Yet, more detailed analyses of (more) children with ABI's oral productions will provide more insights to the precise influence of ABI implantation.

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APPENDIX.

Table A. *Results of S1: the proportion of oral productions (vs. signs): raw numbers*

Data file	Age (months)	Hearing age (months)	Length recording (minutes)	Total number of child productions	Proportion of oral productions
1	38	14	63.97	1094	0.70
2	39	15	57.87	821	0.45
3	41	17	62.67	938	0.52
4	42	18	59.33	837	0.50
5	43	19	58.82	709	0.48
6	44	20	60.97	984	0.45
7	45	21	68.82	914	0.58
8	46	22	69.45	1132	0.44
9	48	24	65.70	1288	0.44
10	49	25	65.58	953	0.46
11	50	26	60.18	996	0.36
12	51	27	63.70	728	0.57
	Mean		*63.09*	*949.50*	*0.50*
	SD		*3.82*	*167.77*	*0.09*

Table B. *Results of S1: the proportion of lexical productions (vs. prelexical): raw numbers*

Data file	Age (months)	Hearing age (months)	Length recording (minutes)	Total number of oral productions	Proportion of lexical productions
1	38	14	63.97	762	0.16
2	39	15	57.87	367	0.40
3	41	17	62.67	485	0.46
4	42	18	59.33	418	0.19
5	43	19	58.82	342	0.09
6	44	20	60.97	443	0.12
7	45	21	68.82	530	0.12
8	46	22	69.45	498	0.12
9	48	24	65.70	571	0.10
10	49	25	65.58	439	0.22
11	50	26	60.18	360	0.23
12	51	27	63.70	416	0.13
	Mean		*63.09*	*469.25*	*0.20*
	SD		*3.82*	*115.39*	*0.12*

Table C. *Results of S1: the proportion of canonical babble (vs. vocalizations): raw numbers*

Data file	Age (months)	Hearing age (months)	Length recording (minutes)	Total number of prelexical productions	Proportion of canonical babble
1	38	14	63.97	640	0.11
2	39	15	57.87	219	0.12
3	41	17	62.67	261	0.16
4	42	18	59.33	338	0.11
5	43	19	58.82	311	0.32
6	44	20	60.97	392	0.20
7	45	21	68.82	465	0.42
8	46	22	69.45	436	0.39
9	48	24	65.70	512	0.29
10	49	25	65.58	342	0.38
11	50	26	60.18	278	0.35
12	51	27	63.70	361	0.48
	Mean		*63.09*	*379.58*	*0.28*
	SD		*3.82*	*118.45*	*0.13*

Table D. *Results of S2: the proportion of oral productions (vs. signs): raw numbers*

Data file	Age (months)	Hearing age (months)	Length recording (minutes)	Total number of child productions	Proportion of oral productions
1	49	24	69.55	1229	0.77
2	50	25	73.52	1461	0.78
3	51	26	98.05	1867	0.72
4	52	27	72.48	1460	0.76
5	53	28	74.58	2457	0.74
6	54	29	66.80	1727	0.80
7	55	30	58.45	596	0.87
8	56	31	69.28	905	0.69
9	57	32	60.13	845	0.76
10	58	33	72.45	899	0.88
11	59	34	52.62	435	0.91
12	60	35	67.68	983	0.90
	Mean		*69.63*	*1238.67*	*0.80*
	SD		*11.20*	*581.70*	*0.07*

Table E. *Results of S2: the proportion of lexical productions (vs. prelexical): raw numbers*

Data file	**Age (months)**	**Hearing age (months)**	**Length recording (minutes)**	**Total number of oral productions**	**Proportion of lexical productions**
1	49	24	69.55	944	0.33
2	50	25	73.52	1138	0.40
3	51	26	98.05	1335	0.58
4	52	27	72.48	1113	0.55
5	53	28	74.58	1820	0.79
6	54	29	66.80	1377	0.83
7	55	30	58.45	518	0.69
8	56	31	69.28	623	0.75
9	57	32	60.13	640	0.73
10	58	33	72.45	795	0.76
11	59	34	52.62	395	0.64
12	60	35	67.68	887	0.73
	Mean		*69.63*	*965.42*	*0.65*
	SD		*11.20*	*412.56*	*0.16*

Table F. *Results of S2: the proportion of canonical babble (vs. vocalizations): raw numbers*

Data file	**Age (months)**	**Hearing age (months)**	**Length recording (minutes)**	**Total number of prelexical productions**	**Proportion of canonical babble**
1	49	24	69.55	633	0.26
2	50	25	73.52	679	0.30
3	51	26	98.05	557	0.44
4	52	27	72.48	501	0.40
5	53	28	74.58	377	0.59
6	54	29	66.80	229	0.66
7	55	30	58.45	159	0.55
8	56	31	69.28	154	0.49
9	57	32	60.13	170	0.46
10	58	33	72.45	187	0.36
11	59	34	52.62	142	0.33
12	60	35	67.68	243	0.56
	Mean		*69.63*	*335.92*	*0.45*
	SD		*11.20*	*203.50*	*0.12*