Word characteristics and speech production accuracy in children with auditory brainstem implants : a longitudinal triple case report

**Reference:**
Faes Jolien, Gillis Steven.- Word characteristics and speech production accuracy in children with auditory brainstem implants : a longitudinal triple case report
Clinical linguistics and phonetics - ISSN 0269-9206 - Philadelphia, Taylor & francis inc, 35:9(2021), p. 874-890
Full text (Publisher's DOI): https://doi.org/10.1080/02699206.2020.1838613
To cite this reference: https://hdl.handle.net/10067/1723570151162165141

## Abstract

Auditory brainstem implants (ABI) in children in the first years of life is a recent innovation. Analyses of their speech and language development on the basis of spontaneous language samples are still largely lacking. The aim was to investigate the phonological complexity of the words children with ABI use in their spontaneous speech, and to compare their accuracy with that of children with cochlear implants (CI) and children with normal hearing (NH). Longitudinal recordings of spontaneous speech were collected of three children with ABI. Children with ABI target mainly words of low phonological complexity in their spontaneous speech, just as children with NH and children with CI do. The complexity of the words they attempt increases over time, but this development is less outspoken in comparison to children with CI and NH at the same hearing ages. The accuracy of the ABI children's word productions is situated in the lower ranges of the 95% confidence intervals of the NH and the CI groups, and – depending on the specific measure – even fall below the 95% border. The ABI intervention appears to be beneficial in the three cases studied, although their development is slow compared to children with CI and NH.

**Keywords:** auditory brainstem implant ABI, paediatric, speech production, accuracy, phonology

## Introduction

Cochlear implants (CIs) have improved the auditory perception of children with severe-to-profound sensorineural hearing deficits. This improved access to ambient speech has also led to vast improvements of their spoken language development (e.g. Faes and Gillis, 2016; Geers et al., 2017; Von Mentzer et al., 2015). When a severe-to-profound hearing loss results from anatomical malformations of the cochlea, from cochlear nerve deficiencies, or from the absence of the auditory nerves, children cannot be aided with a CI. An auditory brainstem implant (ABI) is an alternative option, which has been used since the beginning of this century for pediatric hearing restoration (Puram et al., 2016). Instead of inserting electrodes into the cochlea, an ABI is an array of surface electrodes placed on the cochlear nucleus in the auditory brainstem, bypassing the cochlea and the auditory nerve. Except for the locus of the stimulation, there is another important difference between hearing restoration with a CI versus an ABI. The spiral ganglion, which is stimulated by the CI, is highly tonotopically organized, whereas the neural pathways in the cochlear of the brainstem are characterized as unpredictable (Wong et al., 2019). This appears to make the auditory outcomes of the ABI intervention less predictable and even inferior to that of CI interventions (Wong, et al., 2019).

ABIs were initially developed and designed to restore hearing in adults with neurofibromatosis type 2 (NF2) (Edgerton, House, & Hitselberger, 1982). Gradually, also adults with other non-tumor inner ear pathologies were implanted with ABIs (Puram and Lee, 2015). Around the turn of the century, children with prelingual deafness who were not eligible for cochlear implantation were treated with ABI in Europe (V. Colletti et al., 2002), and since 2013 the first pediatric clinical trials were approved in the US (Puram and Lee, 2015).

Since pediatric ABI implantation is a recent evolution, basis and clinical research in this population is limited, especially on speech production. Yet, different studies pointed to the need of such research to evaluate to long-term effectiveness of pediatric ABI implantation beyond speech perception (e.g. Asfour, Friedman, Shapiro, Roland, & Waltzman, 2018; Puram and Lee, 2015) and the need of evidence based clinical practice (Hammes Ganguly, Schrader, & Martinez, 2019). To date, speech and language therapists have no other option than base their therapy of children with ABI on their knowledge of working with children with hearing loss and CI (Hammes Ganguly, et al., 2019). Thus, more research is needed to unravel the peculiarities of language production in children with ABI in order to adapt intervention strategies to their specific needs.

***Children with ABI***

According to a consensus statement (Sennaroglu, Colletti, et al., 2016), a pure tone average with ABI may be expected between 30 and 60 dB HL. Sound detection and environmental sound awareness occurs during the first year of device use (Sung et al., 2018), but the time required to obtain sound detection can vary greatly from 2 weeks to up to 18 months of ABI use (Teagle, Henderson, He, Ewend, & Buchman, 2018). Better auditory performance and speech production development appear with earlier age at ABI implantation (Aslan et al., 2020), the absence of additional disabilities (L. Colletti, Shannon, & Colletti, 2014; Sennaroglu, Colletti, et al., 2016; van der Straaten et al., 2019) and lower hearing thresholds (Sennaroglu, Colletti, et al., 2016).

In general, only the best performing children with ABI appear to be are able to understand (at least) common phrases without the support of lip-reading (e.g. Aslan, et al., 2020; L. Colletti, et al., 2014; van der Straaten, et al., 2019), which amounts to open set speech recognition. Hence, it can be inferred that spoken language understanding and production is challenging for children with ABI. For instance, it takes the best performing children a very long period of time (60 to 72 months) to achieve their highest level of intelligibility, i.e. intelligible speech for an experienced listener with or without lip-reading (Speech Intelligibility Rate (SIR) 3 to 4, on a five-point scale) (Sennaroglu, Sennaroglu, et al., 2016; van der Straaten, et al., 2019).

Currently, detailed linguistic studies of children with ABI's speech production development are starting to appear. Children with ABI are shown to pass along the consecutive milestones of spontaneous language development, namely vocalizing, babbling and lexical productions (Faes, Boonen, & Gillis, 2019; Faes and Gillis, 2019a). Even though they expand their word use, their vocabulary sizes remain outside the 95% confidence intervals of children with CI and typically hearing (TH), even after four years of hearing experience (Faes and Gillis, 2019b). For expressive language, van der Straaten, et al. (2019) showed that children with ABI's average performance, as a group, is comparable to that of children with CI with additional disabilities. The two best performing children with ABI with no additional disabilities reported by van der Straaten, et al. (2019) have expressive language scores between the means of children with CI with and without disabilities.

Eisenberg et al. (2018) reported in more detail on four children with ABI's speech productions. After two years of device use, all children produced words with one or two syllables, matching the target with 40% to 100% accuracy. Despite being often incorrect, children also started to produce the required word-initial consonants most of the time and some children did also include word-final consonants. After two to three years of ABI use, all children produced some full vowels. In a similar vein, Teagle, et al. (2018) showed that two out of five children are producing consonant and vowel features with 50% to 90% accuracy by three years of device use.

Even though there was considerable individual variation between the children in Eisenberg, et al. (2018) and Teagle, et al. (2018), most of them used vowels and consonants and produced basic word patterns. In the present paper, spontaneous word productions of three children with ABI are analyzed in comparison to children with CI and TH. Teagle et al. (2018) indicated that their 6-month interval language testing was unable to capture the children's subtle progress. In the present study, a monthly follow-up design was implemented in order to capture more sensitively smaller changes in children's speech development.

### *The goal of this study*

This study is a triple-case study of ABI children's spontaneous speech production. Children's word use is analyzed from two perspectives: (1) the adult words that the children target or attempt to produce, henceforth, *target words*, and (2) the children's actual productions of those words, henceforth, children's *renditions*. For instance, if a child utters /bu/ for the Dutch adult word /buk/ (Eng. *book*), the latter is the adult target and the former the child's rendition. Two research questions are pursued:

(1) What kind of words do children with ABI use in spontaneous production?

This part investigates the characteristics of the adult words that children attempt to produce. More specifically, three aspects are studied: the length of the target words in segments and syllables, and the phonological complexity (see below for further elaboration) of the target words.

(2) How far are children with ABI in their phonological development?

The second research question focuses on children's own productions, i.e., the actual renditions of the target words. Two aspects are considered: the phonological complexity of the children's renditions and their production accuracy.

The phonological complexity of the adult targets and the children's renditions are measured using Ingram (2002)'s phonological Mean Length of Utterance (pMLU). pMLU is a measure of the phonological complexity of whole-word productions, which takes into account the number of (correct) consonants and the word's length. Computed on the target words children attempt to produce, it provides a measure of the complexity of those words, and computed on the children's renditions it provides a measure of the phonological complexity of the children's own actual productions. The ratio of both measures constitutes the Proportion of Whole-Word Proximity (PWP), a measure of the accuracy of the children's renditions relative to the target words.

The children with ABI’s speech production is systematically compared to that of children with CI and TH. The children with CI and children with ABI studied here are characterized by a sensorineural congenital hearing loss. Hence, it may be hypothesized that they follow similar developmental trajectories after implantation. However, a different type of hearing restoration as well as a later age at implantation in children with ABI may just as well result in – entirely or partially – different patterns of development. Since detailed analyses of their phonological development are still lacking, the present study will be beneficial in gaining insight into the process, and may eventually fine-tune speech and language therapy for these children (Hammes Ganguly, et al., 2019). Moreover, a better understanding of the possible outcomes of these children’s speech and language outcomes may be of some guidance when evaluating the benefits of ABI against the surgical risks.

## Method

### *Participants*

This study was approved by the Ethical Committee for the Social Sciences and Humanities of the University of Antwerp (SHW_17_16).

#### *Children with auditory brainstem implants (ABI)*

ABI implantation is a far more recent development as compared to CI implantation. As a result, the pool of participants for the children with ABI was very limited: since 2015, only eight children received an ABI under the age of five in Belgium. Restricting the criteria for inclusion in the study group to children who are raised in Dutch, which includes only the northern part of Belgium (Flanders), and to children with no other reported developmental or health problems, led to only three participants with ABI.

ABI1 has a congenital sensorineural hearing loss resulting from the absence of the auditory nerves. The pure tone average (PTA) hearing threshold before implantation was 120 dB HL (decibel hearing level). At two years of age, ABI1 was implanted with an ABI (Med-El). The implant fitting started two months later. Nine of the 12 electrodes could be activated. At four years and nine months of age, ABI1 received a second ABI. Two years after implantation, the child's PTA hearing threshold improved to 37.5 dB HL. ABI1 was raised in oral Dutch, supported with Flemish Sign Language. Monthly data collected for ABI1 started about a year after implantation (age 3;02, years;months) and lasted two and a half years (until age 5;07).

ABI2 was born with a sensorineural hearing loss, resulting from the absence of the auditory nerves, with a PTA threshold of 116 dB HL according to the medical records. ABI2 received her ABI (Med-El) at age 2;01. Two months later, the implant was fitted and nine out of 12 electrodes could be activated. Two years after implantation, the PTA threshold had improved to 43 dB HL. ABI2 was raised in oral Dutch, partly supported with Flemish Sign Language. Monthly data collection started two years after implantation (age 4;01) and lasted for a period of two years (until age 6;03).

ABI3 has a congenital sensorineural hearing loss, diagnosed as an auditory neuropathy with PTA thresholds of 90 to 95 dB HL in the better ear before implantation (no further test). ABI3 received first a CI at 8 months of age. After cochlear implantation, the child's PTA threshold improved to 33 dB HL. Nevertheless, the child seemed to have very limited effect of the CI and was consequently implanted contralateral with an ABI (Med-El) at four years of age[i]. After two months, the ABI was activated and all electrodes could be fitted. The child was raised in oral Dutch, with support of Flemish Sign Language. Monthly data collection started two months before ABI implantation (age 3;10) and ended a year and a half later (until 5;04). Between age 4;10 and age 5;00, there was no data collection due to personal reasons.

*Children with cochlear implant (CI)*

Nine children with CI (CI1 – CI9) participated in this study as a control group. Table 1 presents individual data of these children. All children had a sensorineural hearing loss due to a cochlear deficit. Their mean PTA was 112.56 dB HL before implantation (SD = 9.12). All children received a Nucleus-24 cochlear implant and fitting started one month after the surgery. The mean age of implantation was one year (SD = 5 months). After the implantation, the mean PTA improved to 32.22 dB HL at two years of age (SD = 7.11). Six children also received a second implant (see Table 1). All children were raised orally in Dutch, with only a limited support of lexical signs. Data collection started immediately after the initial fitting of the implant. There was a monthly follow-up up to 30 months after implantation and yearly follow-up sessions were included up to the children's seventh birthday. The CI corpus was collected as part of a past research project on CI children's language acquisition and development.

*Insert Table 1 over here.*

*Children with typical hearing (TH)*

A total of 81 children with typical hearing (TH) participated as a second control group. This group of children with TH comprises two subgroups. The first subgroup consists of 30 children with TH, followed longitudinally between six and 24 months of age (longitudinal TH corpus). The second subgroups of children with TH is a cross-sectional one (cross-sectional TH corpus), with 9 three-year-olds (mean = 3;00, SD = 0;01), 12 four-year-olds (mean = 4;00, SD = 0;01), 10 five-years-olds (mean = 5;00, SD = 0;01), 10 six-year-olds (mean = 6;00, SD = 0;03) and 10 seven-years-olds (mean = 7;00, SD = 0;02). The TH corpora were collected as part of different past research projects designed to investigate various aspects of typically developing children's speech and language (Faes, 2017; Molemans, 2011; Molemans, Van den Berg, Van Severen, & Gillis, 2012; van den Berg, 2012).

***Data collection and transcription***

A schematic overview of the data collection and transcription of the different corpora is presented in Table 2. For the ABI corpus, CI corpus and longitudinal TH corpus (up to two years of age), monthly one-hour video recordings of spontaneous, unstructured interactions between child and caregiver(s) were collected. For the cross-sectional TH corpus (between ages three and seven), only one recording per child was made, and the mean recording time was shorter: 11 minutes (SD = 3 minutes) for the three-year-olds, 15 minutes (SD = 6) for the four-year-olds, 14 minutes (SD = 4) for the five-years-olds, 23 minutes (SD = 9) for the six-year-olds and 19 minutes (SD = 6) for the seven-year-olds. For the CI corpus and the longitudinal TH corpus, twenty-minute selections were made from the full one-hour recordings, thereby excluding silent and noisy passages (Schauwers, 2006). This was done to keep transcription time within reasonable limits. For the ABI corpus, the full one-hour recordings were used, since there were far less useable data in the recordings. Also for the cross-sectional TH corpus, the full recordings were used for the same reason. All data were bootstrapped in order to manage these sample duration differences (see section data analyses).

*Insert Table 2 here.*

All video recordings were imported in CHILDES' CLAN and transcribed following the CHAT conventions (MacWhinney, 2000). Pure (dis)comfort and vegetative sounds, such as crying or coughing, were excluded. The orally produced child utterances were identified as either lexical or prelexical. Each lexical child utterance was orthographically transcribed, and phonemically annotated. Both a phonemic transcription of the actual child production and the adult target word were added. The target represents the model and was retrieved from Fonilex (Mertens, 2001), a lexicon of Dutch words and their standard Flemish pronunciation variants. Afterwards, the phonemic transcription of the child production and the phonemic transcription of the target were automatically divided into syllables. Finally, both transcriptions were aligned at the phoneme level, using a dynamic alignment implementation based on Algorithm for Dynamic Alignment of Phonetic Transcriptions (ADAPT) (Elfers, Van Bael, & Strik, 2005). The alignments were verified manually and corrected if needed.

The interrater reliability of the phonemic transcriptions of the corpora was checked for 10% of the data. For the ABI corpus the agreement in a phoneme-to-phoneme comparison equaled 80.05%. For the CI and TH corpora, interrater reliability of the phonemic transcriptions was checked for 10% of the data: the agreement on consonant (manner and place) and vowels features (place and height) equaled 81.63% for the CI corpus and 78.77% for the TH corpus.

Detailed information about data collection, transcription and additional reliability metrics can be found in Schauwers (2006) for the CI corpus and in Faes (2017), Molemans (2011), van den Berg (2012) and Van Severen (2012) for the TH corpus.

### *Data analyses*

The goal of the present study is twofold: (1) investigate the characteristics of children's target words in spontaneous production, and (2) study the phonological development and accuracy of production. Five different measures were used:

| *Goal* | *Measure* | *Description* |
| --- | --- | --- |

| | | |
|---|---|---|
| (1) | • Target word length in syllables | The number of syllables in the target word |
| | • Target word length in phonemes | The number of phonemes in the target word |
| | • $pMLU^{Target}$ | The phonological Mean Length of Utterance of the target word (Ingram, 2002). pMLU is a measure of the segmental complexity of the target words. It is computed by adding for each word the number of segments and the number of consonants. By averaging over the number of different words attempted by the child, the measure approximates the complexity of the target words at the segmental level. For instance, the Dutch word /banan/ (*banaan*, Eng. *banana*) has five segments and three consonants, resulting in eight pMLU points. These eight points are eventually summed up with the pMLU points of all the target words uttered by the child and divided by the number of target words to achieve the $pMLU^{target}$. |
| (2) | • $pMLU^{Child}$ | The phonological Mean Length of Utterance of the child's actual productions (Ingram, 2002). $pMLU^{Child}$ is calculated in a similar way as the $pMLU^{Target}$, but considers the actual child production instead of the target word. More specifically, the length of the child's production and the number of correctly replicated consonants are added, and eventually averaged. For instance, if the child utters /nan/ for /banan/ (*banaan*, Eng. *banana*), the result |

equals 3 pMLU points for the number of segments, plus 2 pMLU points for the number of correctly produced segments (twice /n/). Ingram (2002) defined several additional rules for data selection, which will not be touched upon here. Moreover, he proposed six consecutive stages of phonological development, according to their characteristic $pMLU^{Child}$ value: Stage I pMLU range 2.5 – 3.5, Stage II pMLU range 3.5 – 4.5, Stage III pMLU range 4.5 – 5.5, stage IV pMLU range 5.5 – 6.5, Stage V pMLU range 6.5 – 7.5, and beyond V.

- PWP — The phonological Whole-Word Proximity. PWP is a measure of accuracy and is calculated by dividing the $pMLU^{Child}$ by the $pMLU^{Target}$ (Ingram, 2002). This implies that PWP equals 1 if the child's rendition completely coincides with the adult target and approaches zero as the child's target is further away from the target.

Spontaneous speech samples (transcribed recordings) were used to analyze children's productions. The recordings were of different lengths. Hence, a procedure was implemented to take into account the differences in sample size: in order to normalize the sample sizes, a bootstrapping procedure was implemented (Efron, 1979), according to the practical guidelines delineated in Molemans, et al. (2012). Each data file was resampled 1,000 times. In each of these samples, 100 child utterances (i.e. word productions) were randomly chosen. All measures were calculated for each word production and a mean was determined for each of the measures. These values are considered as a reliable mean after the bootstrapping normalization (Molemans, et al., 2012).

Comparisons between children with ABI and children with CI and TH are made relative to hearing age. Hearing age is defined as the time after device switch on. For ABI3, hearing age is defined as the time after ABI device switch on. For children with TH, hearing age equals their chronological age.

## Results

### *Target word characteristics*

The first goal of this study was to investigate children's target words in spontaneous speech production. Figures 1 – 3 represent the values of ABI1, ABI2 and ABI3, in comparison with children with CI and children with TH. For the children with CI and TH, 95% confidence intervals are shown.

*ABI1*

ABI1's target words have fewer syllables, fewer phonemes and are phonologically less complex (pMLU$^{target}$) in comparison to the words targeted by the children with CI as well as the children with TH throughout the period studied. With increasing hearing age, the number of syllables in the target words remains relatively stable: ABI1 targets monosyllabic words over the entire period (range: 0.79 – 1.46 syllables). Sometimes, during the video recordings, the child was exercising sound production in combination with fingerspelling, so that the child for instance produced the consonant [p] in combination with the correct sign for it in fingerspelling. Moreover, sometimes, it occurred that the child signed for instance *book* and produced for instance the first consonant of it, [b] in this case. Therefore, syllable length was sometimes below 1.

The number of syllables in the target words of children with CI and TH reaches levels between one and two. The mean number of phonemes increases from two to two and a half in

ABI1, whereas children with CI and TH have mean values well above three at the same hearing ages. For both measures, ABI1 reaches the extreme lower part of the 95% confidence intervals of children with TH. In comparison to children with CI, ABI1 falls out or reaches the lowest part of the 95% confidence intervals.

Phonological complexity of ABI1's target words increases from approximately three to four. However, the $pMLU^{target}$ is considerably lower as compared to children with TH (values between five and six) and children with CI (values approximate five) at the same hearing ages. In comparison to children with TH, ABI1 reaches the lower range of the confidence intervals. In comparison to children with CI, ABI1 even falls out of the 95% confidence intervals for almost the entire period.

*ABI2*

ABI2 targets words with a similar syllable length as the mean child with CI and TH. The mean value for all these children lies between one and one and a half. The number of phonemes in the target words of ABI2 reaches values between two and a half and three. In this sense, ABI2 stays below the mean values of three to four of children with CI and TH. But, ABI2 falls within the lower ranges of the 95% confidence intervals of both groups of children in the entire study period.

The phonological complexity of the target words ($pMLU^{target}$) of ABI2 reaches values of approximately four and slightly increases to approximately four and a half by the end of the period studied. Similarly to the other measures, ABI2's values stay below the mean of the children with CI (values approximate five) and children with TH (values between five and six), but within the lower range of the 95% confidence intervals for these children.

*ABI3*

The mean number of syllables of the target words of ABI3 approximates that of children with CI and TH, with values between one and one and a half. Also with respect to the number of phonemes in his target words, ABI3 reaches values similar to those of children with CI with the same hearing age (values around three). Children with TH have slightly more phonemes in their target words (three and a half), but ABI3 falls within the lower range of the 95% confidence intervals of these children.

For the phonological complexity of the target words (pMLU$^{target}$) ABI3 seems to reach similar mean values as children with CI, between four and a half and five. The phonological complexity of the target words is slightly higher in children with TH (between five and a half and six), but ABI3 falls well in the lower range of the 95% confidence interval of these children.

*Insert Figures 1 – 3 here.*

***Children's phonological development***

The second goal of this study was to investigate two aspects of children's own productions: the phonological complexity of children's actual productions (pMLU$^{Child}$, Figures 4) and the accuracy of production (PWP, Figure 5).

*ABI1*

The phonological complexity of ABI1's actual productions (pMLU$^{Child}$) remains quite stable over the entire period studied. In contrast, the pMLU$^{Child}$ is considerably higher in children with CI and TH, with values of at least four in both groups of children. With a value of two, ABI1 did not yet reach the first stage identified by Ingram (2002), whereas children with TH and CI reach stages II and III within the same period of hearing age. Unsurprisingly, ABI1 falls completely out of the 95% confidence intervals of both children with CI and children with TH.

With respect to production accuracy, the PWP of ABI1 decreases from approximately 0.80 to approximately 0.65 by the end of the period studied. Within that same time period, the PWP increases for both children with CI and TH from just below 0.80 to accuracy scores of approximately 0.90. Initially, ABI1 still falls within the 95% confidence intervals of both groups of children, but not as the hearing age increases.

*ABI2*

The phonological complexity of ABI2's spontaneous productions ($pMLU^{Child}$) approximates a value of three and a half over the entire period studied. According to Ingram (2002), ABI2 can be situated at the beginning of stage II. At the same hearing ages, the phonological complexity of children with CI's productions increase from four to five (stages II and III) and the phonological complexity of children with TH's productions increase from four to four and a half (stage II). Over the entire studied period, however, ABI2 falls out of the 95% confidence intervals of both groups of children.

The production accuracy (PWP) of ABI2 lies between 0.75 and 0.80 over the entire period. At the same hearing ages, however, production accuracy increases from 0.80 to approximately 0.90 in children with CI and children with TH. Most of the time, ABI2 does not reach the lowest part of the 95% confidence intervals.

*ABI3*

The phonological complexity of ABI3's actual productions remains stable with increasing hearing age, with a $pMLU^{Child}$ of approximately three and a half. This situates ABI3 at the beginning of Ingram (2002)'s stage II. At the same hearing ages, the $pMLU^{Child}$ of children with CI increases from two to four. So, initially, the $pMLU^{Child}$ of ABI3 is higher than the mean of children with CI ($pMLU^{Child}$ of two, stage I), but within the higher range of the 95% confidence interval of these children. By the end of the data collection of ABI3, the mean values of ABI3 and children with CI are similar. Both ABI3 and children with CI are in stage II. In comparison to children with TH, the mean values of ABI3 are slightly lower than those of children with TH ($pMLU^{Child}$ of four), but these children can be situated in stage II as well. In addition, ABI3 falls well in the lower range of the 95% confidence intervals of the children with TH.

ABI3's production accuracy (PWP) decreases from approximately 0.83 to 0.73. This decreasing trend is similar as the one observed in children with CI at the same hearing ages. After the period studied for ABI3, children with CI start to increase their production accuracy. We do not have data for ABI3 at the older hearing ages. Also in comparison to children with TH, ABI3 has similar mean PWP values by the end of the period studied in this child. In a similar vein, children with TH increase their production accuracy from that point onwards. Unfortunately, we do not have data for ABI3 at these later hearing ages.

*Insert Figures 4 and 5 here.*

## Discussion

### *Phonological development in children with ABI*

Results revealed that the three children with ABI spontaneously attempt and use mainly monosyllabic words with two up to three phonemes with longer device use, thus presumably words without consonant clusters. This is in line with Eisenberg, et al. (2018), who reported that children with ABI started to use basic word patterns with consonants and vowels in a naming task. Compared to children with CI and TH, the children with ABI in this study fall mostly out of the 95% confidence intervals of the reference groups for target word complexity and phonological complexity of their own productions ($pMLU^{Child}$).

Production accuracy is lower in ABI1 and ABI2 as compared to children with CI and TH and falls out of these reference groups' 95% confidence intervals. For ABI3, PWP seems similar to that of the reference groups. Unfortunately, when children with CI and TH increase their accuracy rate, the data collection for ABI3 stopped, so that we cannot see if the child is following the same curve or not. Even though the two other children with ABI target phonologically less complex words, which could possibly result in higher accuracy rates, their production accuracy is lower. Given the fact that both ABI1 and ABI2 target words of the same complexity ($pMLU^{Target}$), the higher $pMLU^{Child}$ rates of ABI2 are reflected in the accuracy scores as well: ABI2 keeps up with children with CI in the lower 95% confidence interval range for a longer period and has a higher accuracy score as compared to ABI1. Still, after about 33 months of hearing age, ABI2 started to lag behind on children with CI as well.

It should be noted that the hearing ages of the three children with ABI only partially overlap. Immediately after implantation ABI3's results seem to be similar to that of children with CI and within the (lower) ranges of the 95% confidence intervals of TH group. A detailed inspection of the figures indicates that this is probably explained by the child's performance with CI alone. When looking at ABI2's results, for instance, who did not wear a CI and was implanted with her ABI by age two already, the difference with the reference groups is still present even after three to four years of device use.

Even though they are not catching up on their peers with CI and TH, children with ABI do progress over time and are beginning to target more complex words when they are using their device for a longer period of time. But, they do not seem to increase their phonological complexity in their own production, whereas the children with CI and TH show a clear increase with hearing age. The phonological complexity of the three children with ABI's productions remained in stage I or stage II. In contrast, children with CI and TH increased their phonological complexity in production from stage I up to stage III-IV within the same period.

Similar observations have been made for children with CI (Faes and Gillis, 2018; Schauwers, 2006). Initially, up to two and a half years of age, children with CI tend to attempt words that are shorter and have a lower phonological complexity as compared to age-matched children with TH. Possible explanations were the later onset of word production in children with CI and the relation between lexical and phonological development. Early words are typically phonologically simple and are predominantly also produced correctly. With the expansion of the lexicon, more complex words appear, and these bring about a drop in articulatory precision. Only later the production accuracy increases again, so that the entire curve shows a u-shape (Ferguson and Farwell, 1975; Sosa and Stoel-Gammon, 2006). Due to the later onset of word production, children with CI are in an earlier phase of the u-shaped learning as compared to TH age-matched peers initially. The three children with ABI seem to be in an early phase of this u-shaped learning curve. This hypothesis is strengthened by the observation that the (spoken) lexicon sizes of the same children with ABI (ABI1 and ABI2) are much smaller as compared to those of children with CI (Faes and Gillis, 2019b). So, it seems that the children with ABI are just starting to use their first words in this first step of u-shaped learning, which is reflected in the low phonological complexity of the target words and in a relatively high accuracy rate that is even similar to that of the average child with CI and TH initially (see ABI3).

Production accuracy decreased with increasing device use for the children with ABI. The u-shaped learning process may be involved here as well (Ferguson and Farwell, 1975; Sosa and Stoel-Gammon, 2006). Their first words have a low phonological complexity and seem to be relatively accurately produced. As they expand their lexicon size and therefore also attempt to produce more complex target words, accuracy rates are dropping. If children with ABI are indeed in this u-shaped learning curve, the accuracy is expected to increase again after some time (Ferguson and Farwell, 1975; Sosa and Stoel-Gammon, 2006). In the current sample of three children up to four years of device use, this was not the case yet.

Also in children with CI, phonological skills and accuracy in production have been found to lag behind those of children with TH (e.g. Ertmer and Goffman, 2011; Faes, Gillis, & Gillis, 2016; Schauwers, 2006; van den Berg, 2012). In contrast to the three children with ABI however, several studies also showed that children with CI were able to catch up on their age-matched TH peers after about four or more years of device use (Faes, et al., 2016; Faes and Gillis, 2018; Nicholas and Geers, 2007). In other words, after about four years of device use, some children with CI increased their accuracy in production again, whereas the children with ABI in this study did not yet reach this point. This seems also in line with the observation that speech intelligibility, closely related to production accuracy, develops very slowly even in the better performing children with ABI (Sennaroglu, Sennaroglu, et al., 2016; van der Straaten, et al., 2019).

***Limitations, clinical implications and future research***

The number of participants with ABI is a serious limitation of the present study. Because ABI implantation is a recent innovation in children, only 8 children were implanted in Belgium thus far. But, since Belgium is divided in three different language areas, only those children raised in the Flemish-speaking part could participate in this study. An extra factor that narrowed down the pool of participants is the exclusion criterion regarding children with additional developmental or health disabilities. Only children with ABI with no other developmental or health disabilities were included. This made the participating children with ABI as homogenous as possible. Eisenberg, et al. (2018, p. 258) already indicated that there is large amount of variation between the children with ABI and that 'typically developing [children with ABI are] more likely to demonstrate progress'. Moreover, van der Straaten, et al. (2019) highlighted that impact of additional disabilities on children with ABI's expressive language development, with better outcomes for those children without additional disabilities. Thus, the children studied here represent the better performing ABI implantees.

The phonological complexity of ABI children's word productions as well as the phonological complexity of their target words increased, but they are by no means approaching levels of phonological development of children with CI and TH with similar hearing ages. ABI1 and ABI2 in this study were, on average, one year older than the children with CI. This means that even though their motor control and cognitive skills are presumably further developed than the children with CI, they are lagging behind on phonological development when compared to children with CI with the same hearing age (but one year younger). Presumably, the later onset of hearing (on average one year) has such an important impact on the slower development and later onset of word production in children with ABI that it is even present when matching the groups on hearing age. For ABI3, this difference in hearing age and chronological age is even more outspoken, since this child was implanted later, at four years of age. Another possible explanation may be that the different types of implants (ABI/CI), and thus the difference in electric stimulation of the brain, are a key factor in the slower development progress in children with ABI. These hypotheses are still open for future research.

From a clinical perspective, the present results suggest that speech and language therapists need to be cautious in their expectations of working with children with ABI, as also indicated by Hammes Ganguly, et al. (2019). Copying clinical practices common with CI users may be too challenging for children with ABI, and therapists may need to introduce their therapy based on phonologically less complex words and/or spend more time to those type of structures. More research on the children with ABI's consonant and vowel production and production accuracy seems necessary in order to disentangle which sounds seem to hinder increasing their phonological production complexity and phonemic accuracy.

### *Conclusion*

In this triple case study, children with ABI spontaneously target monosyllabic words and exhibit low phonological complexity in their spontaneous productions. The phonological complexity of their own productions as well as the adult words they attempt to produce increases over time, resulting in a lower accuracy rate at older hearing ages. If they follow a similar developmental pattern of u-shaped learning as children with CI and TH, an increase of their production accuracy still lies ahead. After four years of device use, however, this increasing trend was not yet observed in the present cohort.

Our results suggest that is important to maintain also visual input in communication. Our results point out that they use oral communication and even show a clear, but slow development of their oral phonology. But, results also show that they are considerably lagging behind their peers with TH and CI. Therefore, we expect the children to not be able to rely on only oral communication, but still need sign language for their daily communication as well.


## Acknowledgements

We would like to thank the children and their parents for participation in this study. We would like to thank Molemans I., Van Severen L., Van den Berg R. and Schauwers K. for their work on the data collection and transcription of the NH and CI corpus.


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## Tables

*Table 1*. Characteristics of the children with CI

| ID | Gender | PTA unaided (dB HL) | PTA CI (dB HL) (age 2;00) | Age CI implantation | Age second CI |
|---|---|---|---|---|---|
| CI1 | F | 120 | 48 | 13.49 | 75.00 |
| CI2 | F | 120 | 30 | 6.69 | 56.00 |
| CI3 | F | 115 | 33 | 10.00 | 70.00 |
| CI4 | M | 113 | 48 | 18.16 | - |
| CI5 | M | 93 | 38 | 16.89 | 76.00 |
| CI6 | M | 120 | 53 | 8.76 | - |
| CI7 | F | 117 | 42 | 5.16 | 15.00 |
| CI8 | F | 112 | 38 | 19.46 | - |
| CI9 | F | 103 | 28 | 8.69 | 23.00 |
| *Mean* | | *113.00* | *40.10* | *12.05* | *52.50* |
| *SD* | | *8.72* | *8.24* | *4.96* | *27.03* |

dB HL = decibels Hearing Level, PTA Pure Tone Average
Ages are presented in months

- = no second CI

*Table 2*. Overview of the different corpora

| | | | TH corpus | |
|---|---|---|---|---|
| | **ABI corpus** | **CI corpus** | **TH longitudinal (6 - 24 months)** | **TH cross-sectional (3 - 7 years)** |
| Number of participants | 3 | 9 | 30 | 51 |
| Recording length | One hour | One hour | One hour | Means per age range between 11 and 23 minutes |
| Video used for transcription | Full recording | 20-minute selection | 20-minute selection | Full recording |

Figures.

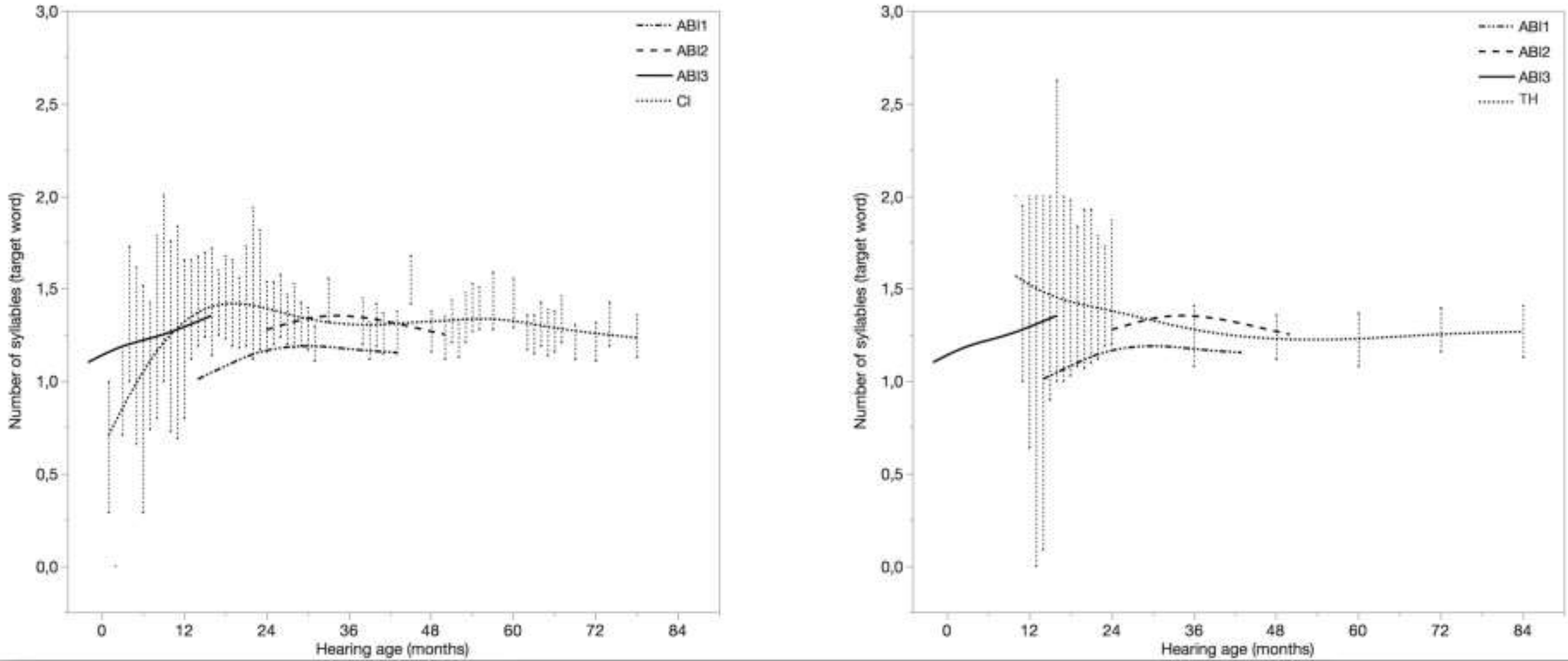
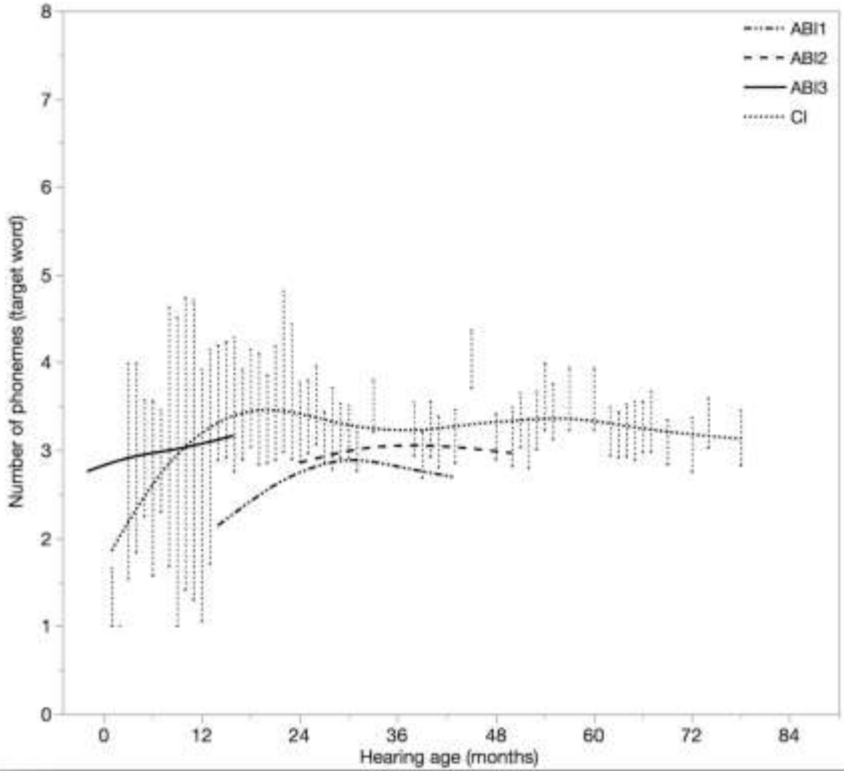



*Figure 1.* Development of the number of syllables in the target words



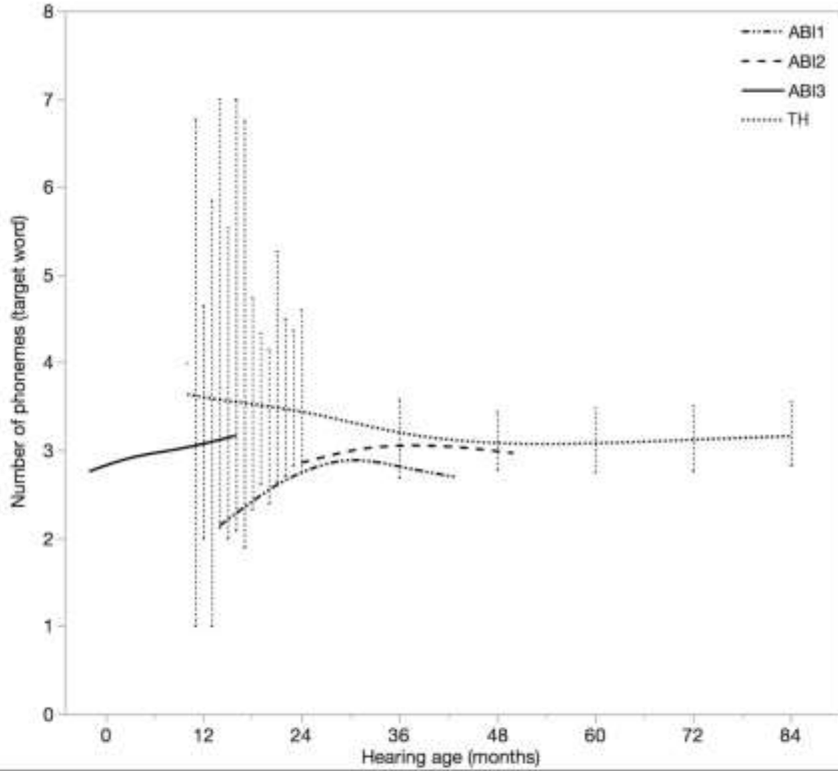




*Figure 2.* Development of the number of phonemes in the target words

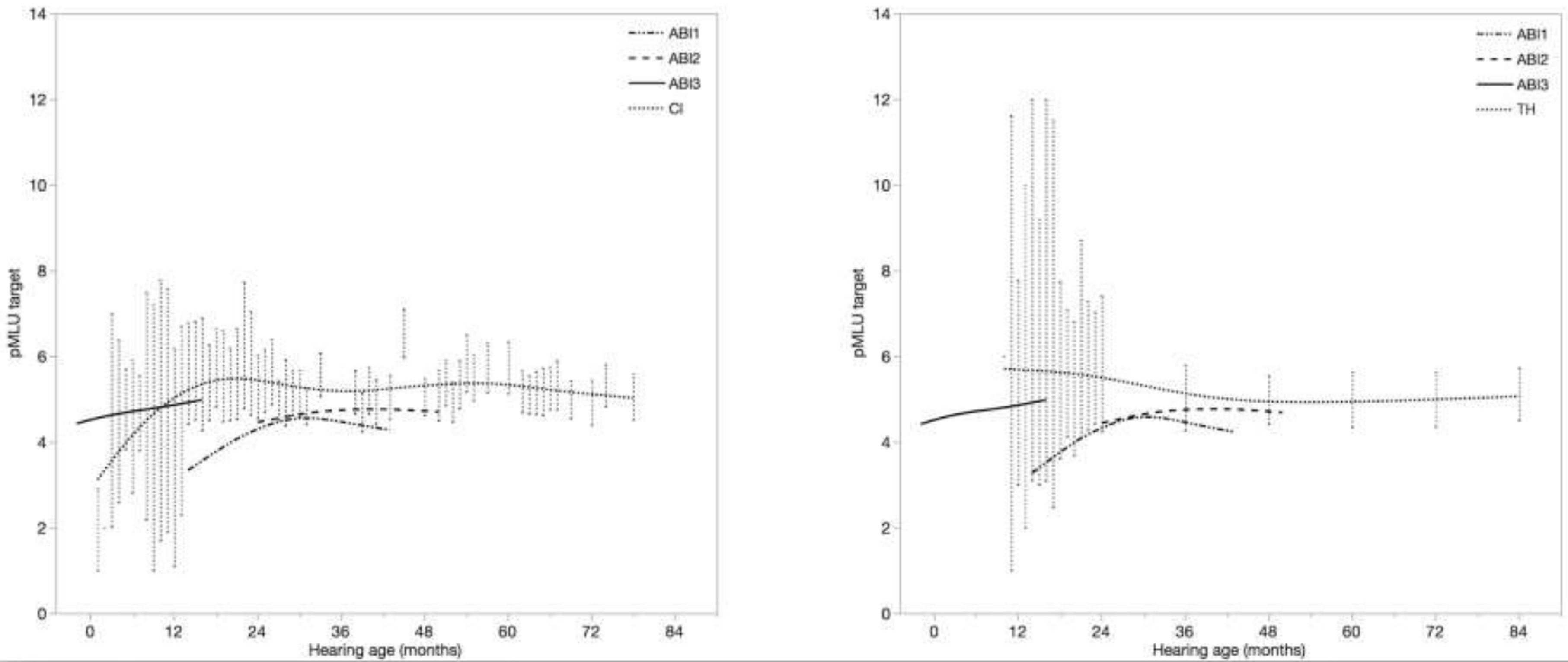


*Figure 3.* Development of pMLU of the target words

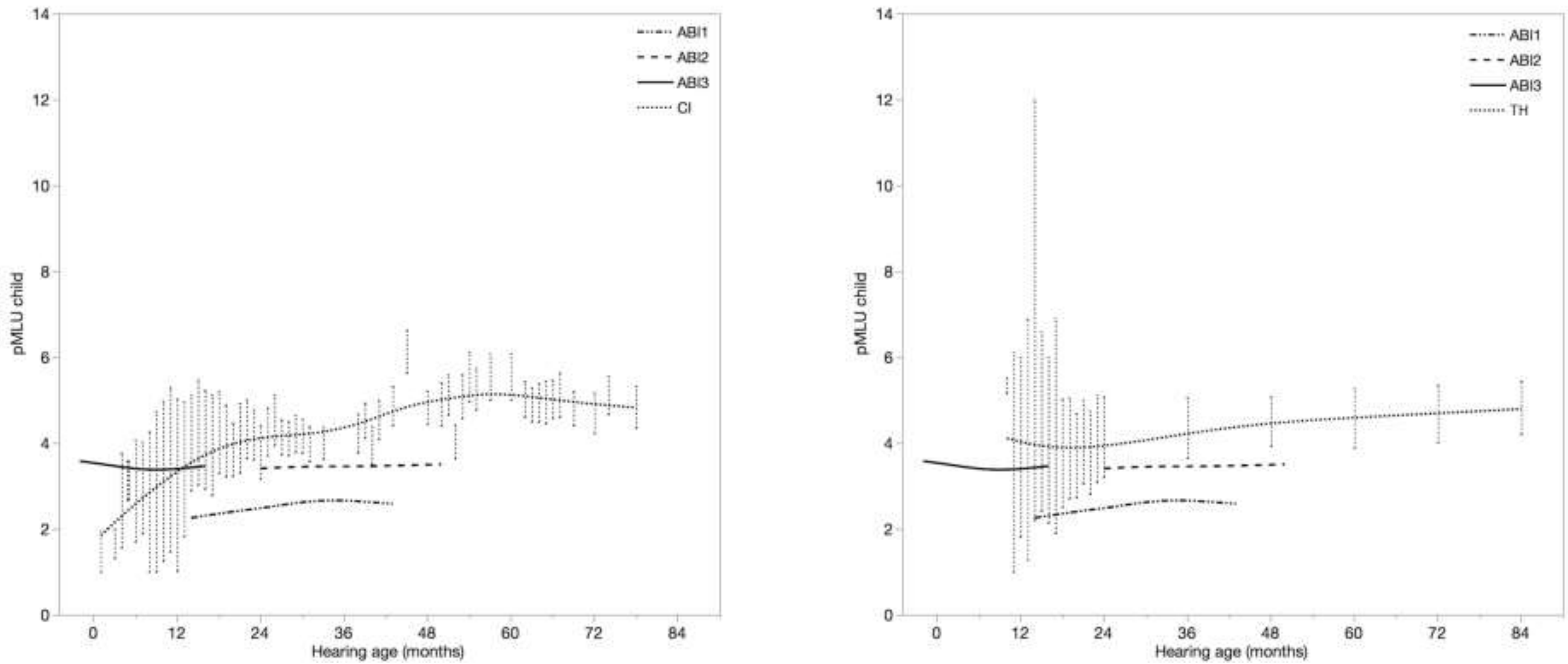



*Figure 4.* Development of pMLU of children's actual produced words

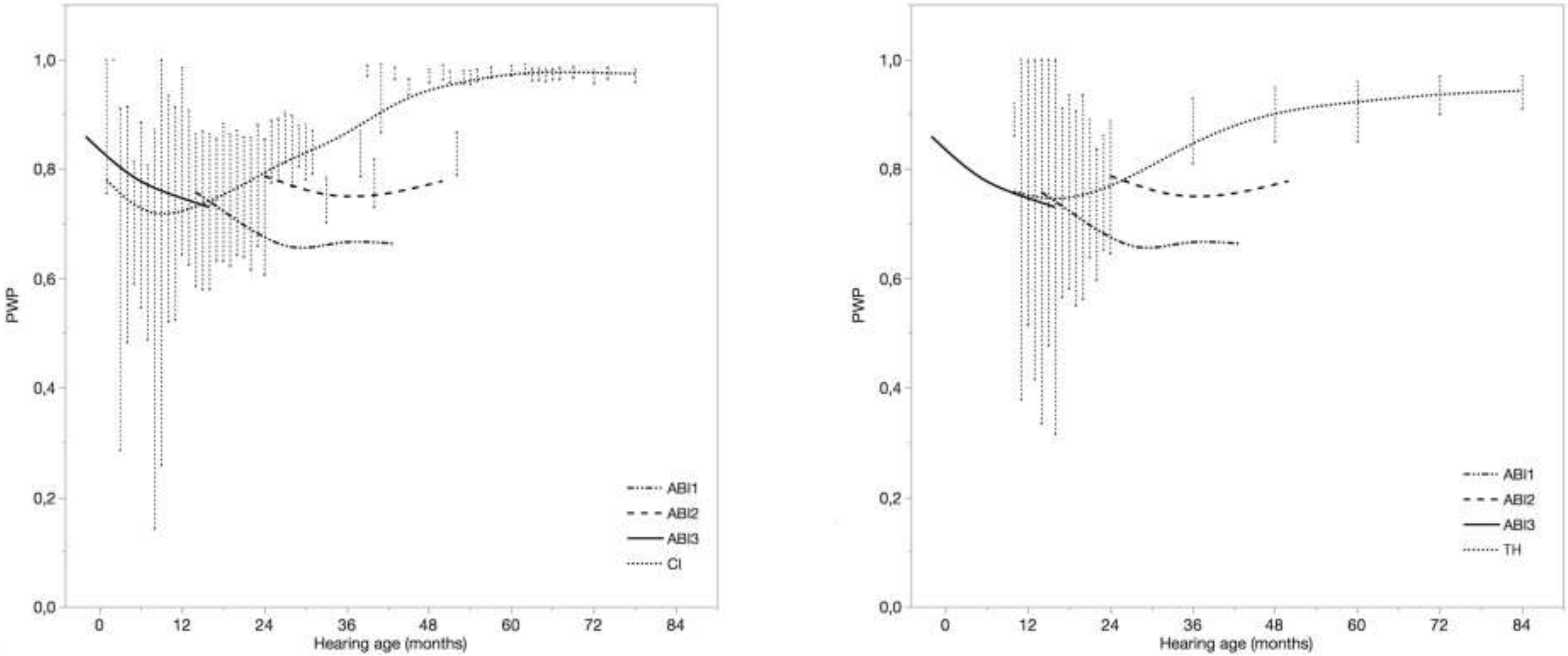



*Figure 5.* Development of PWP

**Figure captions.**

*Figure 1.* Development of the number of syllables in the target words

*Figure 2.* Development of the number of phonemes in the target words

*Figure 3.* Development of pMLU of the target words

*Figure 4.* Development of pMLU of children's actual produced words

*Figure 5.* Development of PWP

**Endnotes**

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[i] Similar cases of children with hypoplastic cochlear nerve who demonstrate sound detection with a CI at around 30–40 dB HL, who nevertheless display very limited speech and language development were previously reported in the literature. These children benefit from an ABI (Sennaroglu et al., 2016: 168). In many cases, CI is an option to be pursued before ABI surgery: a trial period with a CI prior to evaluation for an ABI is advisable whenever possible (Buchman et al., 2011; Farhood et al., 2017).