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Morphosyntactic development after auditory brainstem implantation in three Dutch-speaking children with profound hearing loss

Jolien Faes, Joris Gillis & Steven Gillis

Abstract

The present study is the first to investigate morphosyntactic development in congenitally profound hearing-impaired children without additional disabilities who received an auditory brainstem implant (ABI) in Flanders (Belgium). Auditory brainstem implantation (ABI) is a relatively recent development in paediatric hearing restoration. Very early implanted children's spontaneous language production has hardly been studied and reported on in the international literature. Our study is the first longitudinal investigation of ABI children's syntagmatic (syntactic) development, as indexed by mean length of utterance (MLU) and their paradigmatic (morphological) development, as measured by mean size of paradigm (MSP). The development of children with ABI is compared to that of children with cochlear implants (CI) and children with typical hearing (NH). These groups were matched to the ABI group in two ways: based on their chronological age, and based on their hearing age (i.e., the length of their hearing experience). The grammatical development of three-to-six children with ABI is considerably lagging behind their age-matched peers with CI and NH. But group differences decreased when the children were matched on hearing age instead of chronological age. However, the differences were still significant: children with ABI produce significantly shorter sentences (MLU) and fewer different verb forms per lemma (MSP). In addition, considerable variation was found between the children with ABI, but even the best performing child with ABI was not able to close the gap with hearing age-matched peers with CI and NH. To conclude, our results show that grammatical development is fairly limited in

children with ABI, even in a group of children with ABI with a very advantageous profile in the ABI population.

Keywords: auditory brainstem implantation; morphosyntactic development, lexical development, language

Introduction

With cochlear implantation, the auditory and speech perception of children born with a severe-to-profound sensorineural hearing loss improves considerably, ameliorating the hearing loss up to hearing levels between 20 and 40 dB HL (decibels hearing level). Even though the electrical signal provided by the cochlear implant (CI) is still degraded and underspecified as compared to unprocessed signals received by normally hearing listeners (Castellanos et al., 2020), this improved access to sound and ambient speech has also led to considerable improvements of children with CI's spoken language development (e.g. Faes et al., 2016; S. Gillis, 2018; Niparko et al., 2010; Toe & Paatsch, 2013; Warner-Czyz & Davis, 2008; Watson et al., 2006). For instance, soon after implantation, children with CI start to babble (Kishon-Rabin et al., 2005; Schauwers et al., 2008). Moreover, they seem to catch up with their normally hearing peers after several years of device use on some aspects of their linguistic development, such as morphological and syntactic complexity (Duchesne & Marschark, 2019; Faes et al., 2015). Still, there are large individual differences in children's performance after cochlear implantation, which have been attributed to factors such as the age at implantation, additional non-auditory disabilities, etc. (Boons et al., 2012; Pisoni et al., 2017; Ruffin et al., 2013).

Notwithstanding the relative success of cochlear implantation, some children receive little or no benefit from a CI. 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, i.e., cases in which a cochlear implant is impossible or does not lead to satisfactory outcomes, an auditory brainstem implant (ABI) may be a viable alternative option. Instead of inserting electrodes into the cochlea, an ABI is an array of surface electrodes placed on the cochlear nucleus in the auditory brainstem, thus bypassing the cochlea and the auditory nerve. ABIs have been used since the beginning of this century for paediatric hearing restoration (Puram et al., 2016).

Auditory brainstem implantation: history and incidence

Auditory brainstem implants (ABI) were developed in the early 1980ies to restore hearing in adults with neurofibromatosis type 2 (NF2) (Edgerton et al., 1982). NF2 causes inter alia tumours in the area of the auditory nerves and surgical removal of these tumours often causes damage resulting in hearing loss. Even though the ABI was designed for adults with hearing loss related to NF2, its use was rapidly extended to adults with other inner ear pathologies, such as cochlear (nerve) aplasia, cochlear ossification, cochlear malformation, and the absence of the auditory nerves (V. Colletti et al., 2009; Puram & Lee, 2015). Since 2001, the ABI is also used in paediatric populations in Europe (V. Colletti et al., 2001), and a good decade later, the first clinical trials were set up in the US (Puram & Lee, 2015).

A cochlear implant (CI) and an auditory brainstem implant (ABI) have an external and internal part. The external part of both devices consists of a microphone and a processor, which capture environmental sounds and convert them into a digital code. In the internal part, the digital code is sent to a number of electrodes, which transform it into an electric signal. In case of a CI, these electrodes are placed within the cochlea and stimulate the auditory nerve. As such, the CI bypasses absent or damaged hair cells in the cochlea itself. The electrodes of an ABI are placed directly on the cochlear nucleus of the brainstem, thus stimulating the brainstem directly. An ABI bypasses absent auditory nerves or a damaged cochlea in which no electrodes could be inserted (Puram & Lee, 2015).

In the case of absent auditory nerves and/or absent or damaged cochlea(s), a CI is not applicable and an ABI is the only option (V. Colletti et al., 2002). Yet, a CI trial period has been recommended whenever possible, before turning to an ABI (Buchman et al., 2011; Farhood et al., 2017). With little benefit of the CI in such a case, an ABI is an alternative option (Hammes Ganguly et al., 2019). In practice, many children receive first a CI and, at a later age, a contralateral ABI (Batuk et al., 2020; Sennaroglu, Colletti, et al., 2016). Recent studies indicate that children with cochlear nerve deficiency seem to benefit from the combination of CI and ABI in speech perception, as compared to a condition with only a CI or only an ABI (Batuk et al., 2020; Friedman et al., 2018).

The number of cases of ABI surgery is relatively low. For instance, in Belgium (with approximately 11.5 million registered inhabitants in 2019), only eight children under the age of five received an ABI between 2015 and 2018. This figure is drawn from the statistics of RIZIV (National Institute for Health and Disability Insurance, NIHDI) of officially registered and reimbursed cases. To the best of our knowledge, there are no data available about the incidence of ABI candidacy relative to the birth figures in Belgium. However, for Flanders, the Dutch speaking part of Belgium, the region where the participants of the current study are living, an estimate can be extrapolated from the figures provided by the Flemish agency *Child and Family* (*Kind & Gezin*). In the period 1999-2018 there were on average 65,616 (SD=3,785) newborns per year in Flanders. Over the same period, the average number of newborns with a bilateral hearing loss of more than 70 dB HL was 37.26 (SD = 8.99), or on average 0.56 per 1,000 births. Assuming that these newborns constitute the group of potential CI recipients, and that 2.1% of those are potential recipients of ABI (according to Kaplan et al., 2015), an estimated 0.012 (SD = 0.002) per 1,000 births are eligible for ABI.

Paediatric auditory brainstem implantation

After implantation, children with ABI can reach hearing thresholds between about 30 and 60 dB HL (decibels hearing level) (e.g. Choi et al., 2011; Eisenberg et al., 2018; Sennaroglu, Colletti, et al., 2016; Teagle et al., 2018; Wilkinson et al., 2017). With continued hearing experience, they can identify and discriminate sounds, phonetic contrasts and understand simple sentences with or without lip-reading (L. Colletti et al., 2014; V. Colletti et al., 2002; V. Colletti et al., 2004; da Costa Monsanto et al., 2014). The better performing children can even reach open set speech perception (without lip-reading) and some of them can even telephone with familiar adults after at least five years of device use (L. Colletti et al., 2014; V. Colletti et al., 2002; V. Colletti et al., 2004; da Costa Monsanto et al., 2014; van der Straaten et al., 2019; Yucel et al., 2015). These well-developed speech perception skills are only reachable for children with ABI that are early implanted (Aslan et al., 2020), have low hearing thresholds after implantation (Sennaroglu, Sennaroglu, et al., 2016) and have no additional disabilities (L. Colletti et al., 2014; Sennaroglu, Colletti, et al., 2016; van der Straaten et al., 2019). These are also the children with the better speech production skills (van der Straaten et al., 2019).

According to early research on children with ABI's speech production skills, the best performing children appeared to be vocalizing, babbling and some were also producing words and sentences (e.g. Bayazit et al., 2014; V. Colletti et al., 2002; V. Colletti et al., 2004; Eisenberg et al., 2008; Puram & Lee, 2015). More recently, the speech production development of children with ABI has been scrutinized in more detail. For instance, vocalizations, babbles and words were quantified using normalized measures, showing that children with ABI reach the babbling and word onset after several years of device use (Faes et al., 2019; Faes & Gillis, 2019a, 2019b). As to lexical and phonological development, children with ABI produce ambient language phoneme(s) (features), syllables and basic word patterns, though often incorrectly (Eisenberg et al., 2018; Faes & Gillis, 2020, 2021; Teagle et al., 2018) and have a growing number of words in their lexicon (Faes & Gillis, 2019b). However, it should be reiterated that these developments pertain to the better performers, i.e. children without additional disabilities, with early implantation and with low hearing thresholds after implantation.

But even the better performing children with ABI are lagging behind when compared to children with normal hearing (NH) and children with cochlear implants (CI) with a similar length of hearing experience. For lexical as well as phonological development, children with ABI mostly perform lower than the 95% confidence intervals of the children with NH and CI without additional disabilities (Faes & Gillis, 2019b, 2020). According to van der Straaten et al. (2019), the expressive language skills of children with ABI without additional disabilities can be situated between the means of children with CI with and those without additional disabilities.

In addition, considerable interindividual variation between children with ABI has been reported. Even after controlling for factors impacting the speech perception and production outcomes such as additional disabilities, age of implantation and hearing thresholds after implantation (Aslan et al., 2020; Sennaroglu, Colletti, et al., 2016; van der Straaten et al., 2019), there still remain considerable differences between the (pace of) development of children with ABI (Eisenberg et al., 2018; Faes & Gillis, 2020, 2021). To date, it is unclear which (additional) factors cause and determine these differences.

The present study

In the present study, morphological and syntactic aspects of children with ABI's spontaneous speech were analysed. Three children with ABI (one child with a CI and a contralateral ABI) were followed longitudinally. A monthly follow-up design was set up, in order to capture small changes in the children's development, as for instance Teagle et al. (2018) indicated that children with ABI's slow and slight progress could not be captured by their 6-month interval design. The children in this study can be considered to be *good* performers, given their absence of additional disabilities, their early implantation and their low hearing thresholds after implantation. Their development was compared to that of children with CI and children with typical, normal hearing (NH).

To date, little is known about the children's longitudinal speech and language development. Some studies already investigated phonological and lexical aspects of their speech and language production, but – to the best of our knowledge – no information is available yet with respect to their morphosyntactic development. Morphosyntactic development will be measured in two ways: (1) Mean Length of Utterance (MLU) as a proxy for syntagmatic development, and (2) Mean Size of Paradigm (MSP) of verbs as a proxy for paradigmatic, inflectional development.

MLU, as presented by Brown (1973), is a measure of general grammatical development, which gives an indication of sentence complexity (Hammer, 2010) and morphosyntactic complexity (Mimeau et al., 2015) and is used as an indication of potential issues in linguistic development (Klee & Fitzgerald, 1985). MLU is measured by dividing the number of morphemes (or words or syllables) by the number of utterances in a speech sample (Brown, 1973; Flipsen & Kangas, 2014; Hickey, 1991; Parker & Brorson, 2005; Rice et al., 2010). It is shown to increase with age in different populations, such as typically developing children (Blake et al., 1993; Faes et al., 2015; Rice et al., 2010), children with SLI (Hewitt et al., 2005; Rice et al., 2010) and children with CI (e.g. Blamey et al., 2001; Faes et al., 2015; Hammer, 2010; Moreno-Torres & Torres, 2008; Nicholas & Geers, 2007; Nittrouer, Caldwell-Tarr, et al., 2014; Nittrouer, Sansom, et al., 2014; Schauwers, 2006). For the CI-NH comparison, research has pointed out that early implanted children with CI appear to catch up for MLU with their NH peers after five to seven years of device use (Faes et al., 2015; Hammer, 2010; Nicholas & Geers, 2007). Yet, no information about MLU in children with ABI has been reported in the literature.

Mean Size of Paradigm (MSP) is a measure of paradigmatic, inflectional richness in speech production (Xanthos & Gillis, 2010; Xanthos et al., 2011). It is calculated as the number of different inflected forms per root (or lemma) (Xanthos & Gillis, 2010). The richer the paradigm, or in other words, the more inflected word forms per lemma, the higher the MSP. In the present study the development of the MSP of Dutch verbs in children's language is investigated. The reason for this restriction is that Xanthos et al. (2011) showed that in weakly inflected languages such as Dutch (Laaha et al. 2007), MSP of nouns hardly surpasses one, i.e., one word form per lemma, while the morphological richness of the verbal paradigm is relatively much higher in typically developing children's speech. For children with ABI, no information about their inflectional development is available in the literature thus far. For children with CI, MSP and inflectional development is shown to lag behind that of children NH initially, with fewer inflectional diversity, and, in addition, more errors in case and gender marking, avoidance of plural marking, etc. (Faes et al., 2015; Guo et al., 2013; Hammer, 2010; Laaha et al., 2015; Szagun, 2002). However, with extended device use, early implanted children with CI are able to catch up with their NH peers by five to seven years of age (Faes et al., 2015; Hammer, 2010).

Method

Participants

Three groups of children participated in this study: children with auditory brainstem implants (ABI, N = 3), children with cochlear implants (CI, N = 9) and children with typical, normal hearing (NH, N = 15). None of the children was reported with other health, developmental, motor or cognitive problems, except for their hearing loss in the ABI and CI groups. All children were raised in Dutch by parents with no reported hearing issues, and they belonged to the mid-to-high SES strata of the population.

Children with ABI

Three children with ABI and their families participated in this study. In Belgium, only eight children were implanted with auditory brainstem implants between 2015 and 2019. Inclusion criteria for the present study were (a) Dutch-speaking, excluding children from the French- and German-speaking part of Belgium, and (b) children with no additional disabilities. These criteria reduced the available cohort to the three children participating in this study.

ABI1 was a female child who was born with a sensorineural profound hearing loss with a Pure Tone Average (PTA) hearing loss of 120 dB HL (decibel hearing level). The hearing loss resulted from the absence of the auditory nerves. The child received a first ABI at two years of age. Nine electrodes were activated. A contralateral ABI was implanted later, at age 4;09 (years;months). Two years after the first ABI implantation, the child's PTA had improved to 37.5 dB HL. The child was raised in oral Dutch, with support of Flemish sign language. Data collection for this child started about a year after the first implantation (i.e., at age 3;02) and ended more than two years later, at age 5;07.

ABI2 was a female child, also born with a sensorineural profound hearing loss as a result of the absence of the auditory nerves. The child's PTA before implantation was 116 dB HL. At age 2;01, she was implanted with an ABI and nine electrodes were activated. Two years after implantation, the child's PTA had improved to 43 dB HL. The child was raised in oral Dutch, supported with Flemish Sign Language, but to a lesser extent as compared to ABI1 and ABI3. Data collection of ABI2 started two years after implantation, at age 4;01 and ended two years later at age 6;02.

ABI3 was a male child diagnosed with auditory neuropathy with a PTA of 95 dB HL in the better ear. The child was first implanted with a cochlear implant (CI) at age 0;08. After cochlear implantation, the PTA had improved to 33 dB HL. Nevertheless, little effect on speech and language development was observed over the years. So, the child received a contralateral ABI at age 4;00. At implant fitting, all electrodes were activated. ABI3 was raised in oral Dutch, with support of Flemish Sign Language. Data collection started two months before ABI implantation and went on up to age 5;04. Between ages 4;10 and 5;00, no data were collected due to personal reasons.

Control groups

Two control groups were included into this study: a group of children with CI and a group of children with NH.

Nine children with cochlear implants (CI) participated in this study as a first control group (Table 1). All children were born with a sensorineural hearing loss, with an average PTA of 112.50 dB HL (SD = 9.75) before implantation. The mean age at implantation was 11.14 months (SD = 5 months). Six out of eight children received a second CI at an older age (range 15 months to 75 months). After implantation, the mean PTA improved to 38.75 dB HL (SD = 8.66) at two years of age. All children were raised in oral Dutch, with a limited amount of lexical signs in support. Data collection started immediately after implantation and went on monthly up to 30 months after first implantation, and yearly at the older ages up to the children's seventh birthday.

Longitudinal data of 15 children with normal hearing were drawn from the Dutch section of the CHILDES corpora (https://childes.talkbank.org/access/DutchAfrikaans/). The transcriptions of the following corpora were used, with the age ranges between brackets: from the Groningen Corpus Abel (1;10.30-3;04.01), Daan (1;08.21-3;03.30), Iris (2;01.01-3;06.15), Matthijs (1;10.13-3;03.05) and Peter (1;05.09-2;08.22), from the Utrecht Corpus Hein (2;04.11-3;01.24), from the Van Kampen Corpus Laura (1;09.04-3;06.09) and Sarah (1;06.16-3;05.30), two triplets from the Schaerlaekens Corpus: Gijs, Joost and Katelijne (1;8.29-2;10.23) and Arnold, Diederik and Maria (1;10.18-3;01.07). In addition, the corpus of the child Jolien (1;05.09-2;05.00) was selected from the CLiPS child language corpora.

In addition to these longitudinal data, a cross-sectional NH corpus was added, with 10 two-year-olds, 9 three-year-olds, 10 four-year-olds, 12 five-year-olds, 10 six-year-olds and 10 seven-year-olds. More information about these children can be found in Faes (2017).

Table 1. Individual data of children with cochlear implants.

ID	Gender	PTA unaided (dB HL)	PTA with CI (dB HL) (at age 2;00)	Age at CI implantation	Age at second CI
CI1	F	120	48	1;01	6;03
CI2	F	120	30	0;07	4;08
CI3	F	115	33	0;10	5;10
CI4	M	113	48	1;06	-
CI5	M	93	38	1;05	6;04
CI6	M	120	53	0;09	-
CI7	F	117	42	0;05	1;03
CI8	F	112	38	1;07	-
CI9	F	103	28	0;08	1;11
Mean		*112.50*	*38.75*	*11.14*	*52.50*
SD		*9.75*	*8.66*	*5.02*	*27.03*

dB HL = decibels Hearing Level, PTA = Pure Tone Average

Ages are presented in years;months
- = no second CI

Data collection and transcription

For all children, monthly audio and video recordings of approximately one hour were made at the child's home. These recordings involved spontaneous, unstructured interactions between the child and the caregiver(s). Sometimes, siblings were present as well. In the present study the data of 608 recording sessions were analyzed. The children's utterances were transcribed orthographically in CHILDES' CLAN according to the CHAT conventions (MacWhinney, 2000). All verbs were tagged automatically with the CLAN software tool minMOR for Dutch and disambiguated manually. Each verb was lemmatized, decomposed morphologically and assigned a part-of-speech tag.

Data analyses

Morphosyntactic development was investigated by means of two measures: (1) Mean Length of Utterance and (2) Mean Size of Paradigm. Mean Length of Utterance (MLU) was calculated by dividing the number of words per utterance by the total numbers of utterances. This calculation was done with CLAN's MLU tool on the dependent %mor tier. Mean Size of Paradigm (MSP) was calculated by dividing the number of distinct word forms per verb lemma. For MSP only verbs were included in the present study. The software *MSP Meter* (J. Gillis, 2013) was used, which was run cumulatively over the consecutive files (ordered by increasing age of the child) automatically taking into account the entropy of each verb's paradigm as well as the frequency distribution of the various verb forms in a verb paradigm (i.e. the weighted entropy-based MSP). The MSP calculations were done cumulatively over time without resampling.

Children with ABI were matched with the control groups relative to their chronological age (in the descriptive part of the results section) and relative to their hearing age, that is, the length of their device use expressed in months. For children with CI and ABI, hearing age equals their length of device use, in months. For children with NH, their hearing age is identical to their chronological age. For ABI1, monthly data started at a hearing age of 14 months up to a hearing age of 43 months. For ABI2, monthly data varied between a hearing age of 24 months and 50 months. For ABI3, hearing age was expressed as a function of ABI use. Data of this child started at a hearing age of -2 months, i.e. two months before ABI implantation, but with already three years of CI use at that time, and went on till 16 months of hearing age.

Statistical analyses

Statistical analyses were performed in R using multilevel models. Multilevel models are constructed with 2 parts: a random part and a fixed part. The random part of the model takes into account the variance and nesting of variables. In the present study, this nesting and variance between children and different ages is captured by adding a random effect of Hearing age and a random effect of child ID. In the fixed part of the model, either MLU or MSP is added as the dependent or predicted variable. Independent or explanatory variables were hearing age (in months), hearing status (CI or NH) and the interaction between both variables. Only utterances containing lexical items were considered in the analyses and, hence, MLU and MSP values equaling zero were excluded from the analyses.

For the statistical analyses, only the longitudinal datasets were included, the cross-sectional part of the data was not considered. For each child with ABI, two analyses were performed: one for MLU and one for MSP, with matching data of children with NH and children with CI for the hearing ages available of each child with ABI. For ABI1, data with hearing ages between 12 and 41 months were selected, for ABI2 between 22 and 48 months and for ABI3 between -2 (2 months before ABI implantation, but with CI) and 16 months. The intercept for each analysis was set at the beginning of the ABI data. That is, at 12 months of hearing age for ABI1, at 22 months of hearing age for ABI2 and at 2 months of hearing age (with ABI) for ABI3. For ABI3, no data with children with NH could be matched.

Results

Descriptive comparison between groups

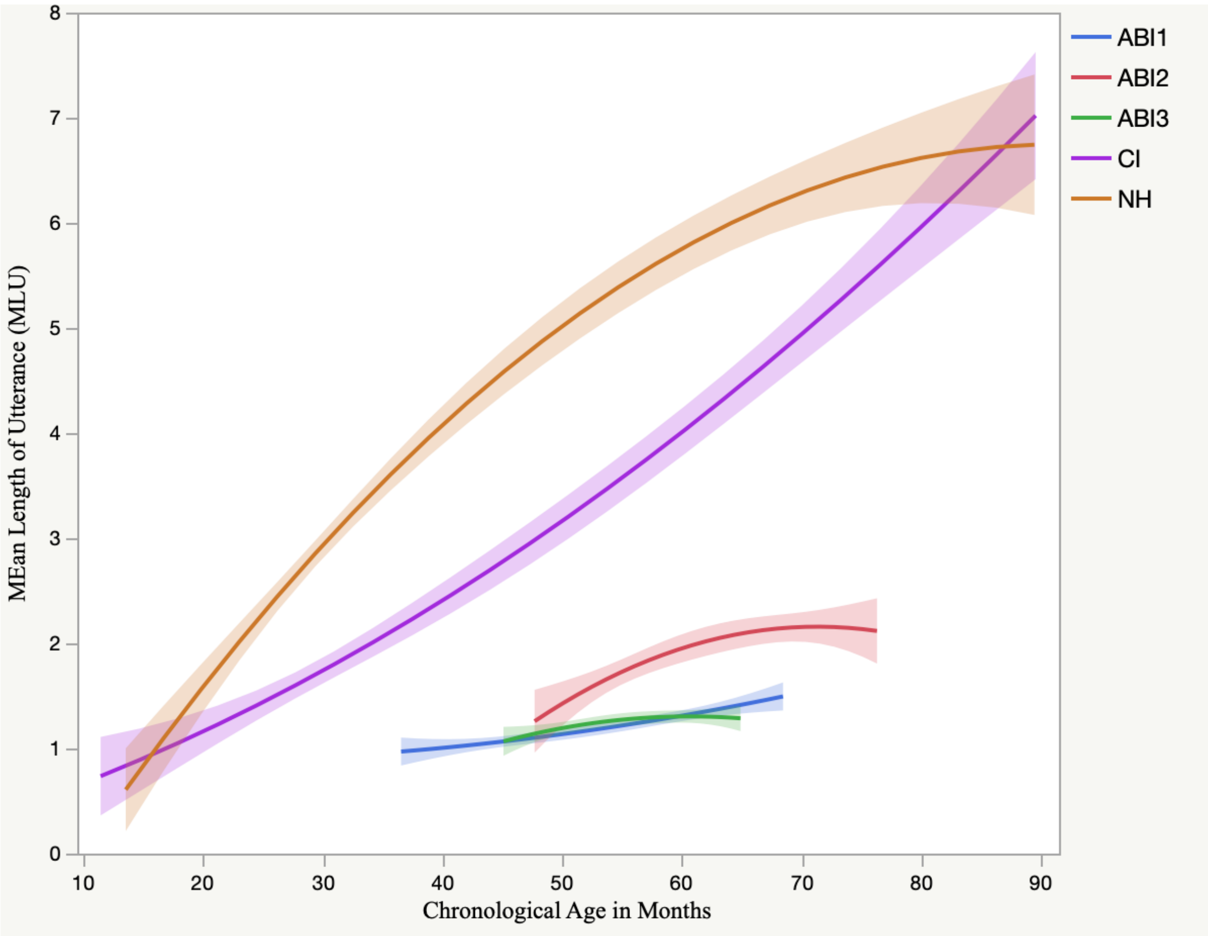
In Figure 1, the development of MLU is plotted for all children (ABI1, ABI2, ABI3, the CI control group and the NH control group) as a function of chronological age and hearing age. A similar figure is displayed in Figure 2 for MSP. All longitudinal data for children with CI and children with NH were included into the graphs, in addition to the cross-sectional data of children with NH. Since the fitted data for the NH children are not only from longitudinal data, they should be interpreted with the necessary caution.

For both measures (MLU and MSP), the three children with ABI score considerably lower than the children with CI and children with NH when matched on chronological age. Only ABI2 seems to approach age-matched CI and NH levels of MSP development. In other words, the three-to-six-year-old children with ABI are lagging considerably behind their three-to-six-year-old peers with CI and their three-to-six-year-old peers with NH.

But given the later onset of hearing for children with ABI in comparison to children with NH and even in comparison to children with CI, a comparison based on chronological age may seem a bit off. The same holds for the CI-NH comparison since also children with CI have a later hearing onset as compared to children with NH. Even though a comparison on chronological age is an intuitive point of departure (for instance parents want to know if their child with implant will reach age-appropriate language levels), the difference in hearing experience skews the comparison. Therefore, hearing age is often suggested as a more appropriate alternative. That is, children’s development is traced based on the length of their hearing experience, expressed as hearing age in months.

For instance, for MLU, the difference between children with CI and children with NH found in the comparison on chronological age disappears when their development is paralleled on hearing age (Figure 1). The same effect is obtained for MSP, but only partially for hearing experience (Figure 2). In other words, the differences between the two groups diminish when hearing age is used as an alternative yardstick. Thus, the comparison of children with NH and those with CI shows that the outspoken difference in their MLUs disappears when the children are compared as a function of their hearing age instead of their chronological age. A similar observation can be made for MSP, though here it takes much longer for the two developmental curves to meet at some point.

The difference in MLU and MSP between each child with ABI and the children with NH and CI – matched on hearing age – is less outspoken when compared to the same comparison on chronological age. Especially for MSP, the values appear to approximate these of children with CI and children with NH. But these are the observed values. The statistical assessment of the differences will be presented in the next section.



MEan Length of Utterance (MLU)
Chronological Age in Months
ABI1
ABI2
ABI3
CI
NH
0
1
2
3
4
5
6
7
8
10
20
30
40
50
60
70
80
90

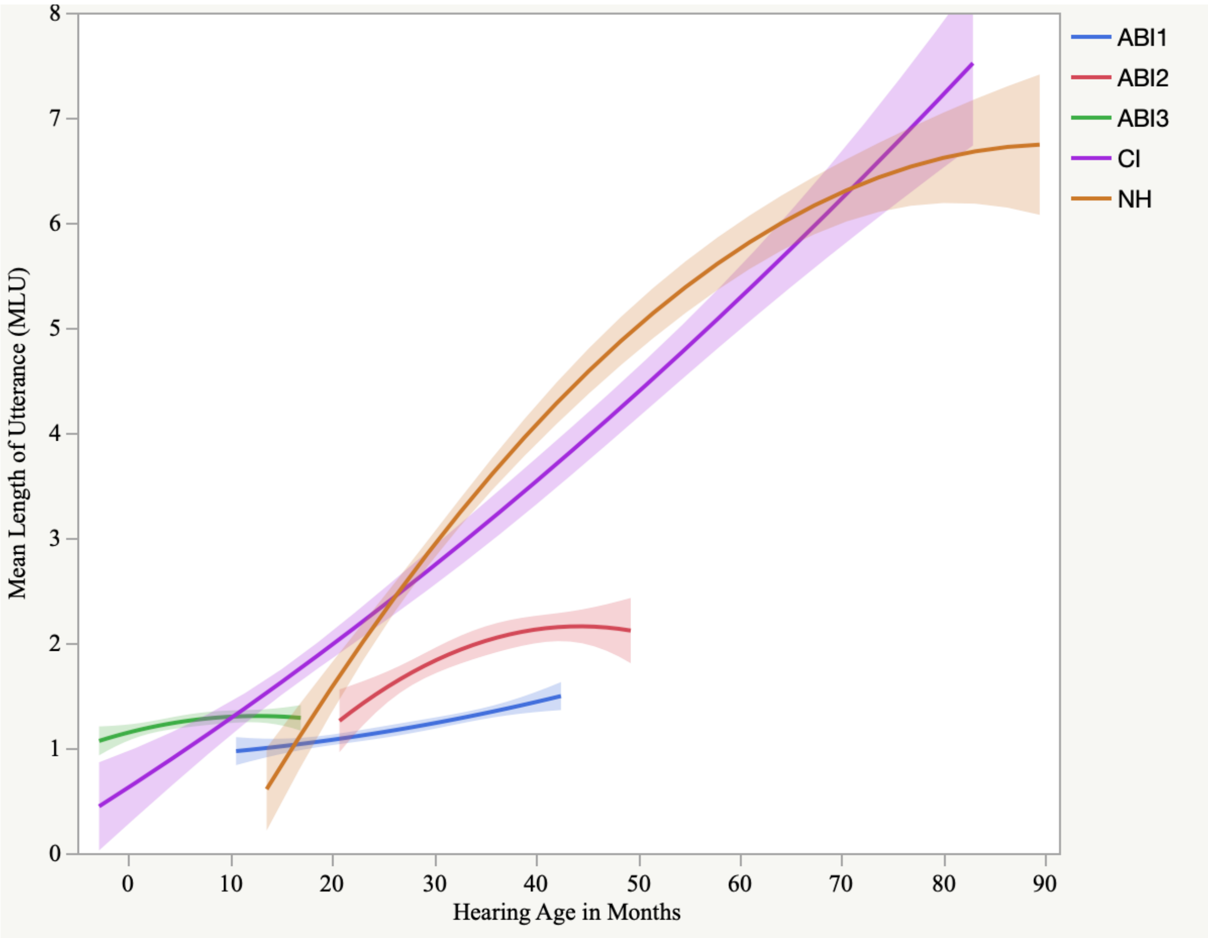
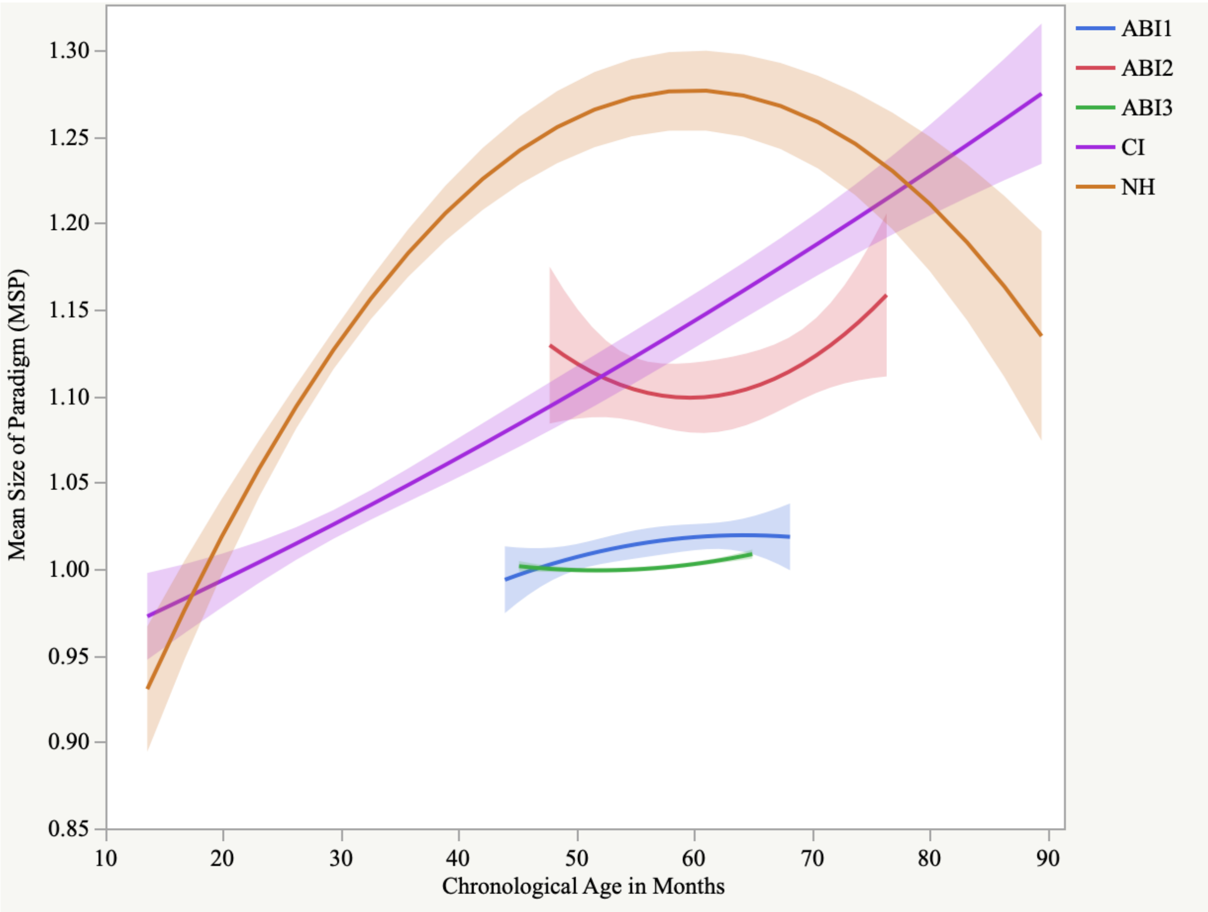


Figure 1. Quadratic fit of observed MLU of all children: comparison on chronological age and hearing age



ABI1
ABI2
ABI3
CI
NH
1.30
1.25
1.20
1.15
1.10
1.05
1.00
0.95
0.90
0.85
Mean Size of Paradigm (MSP)
10
20
30
40
50
60
70
80
90
Chronological Age in Months

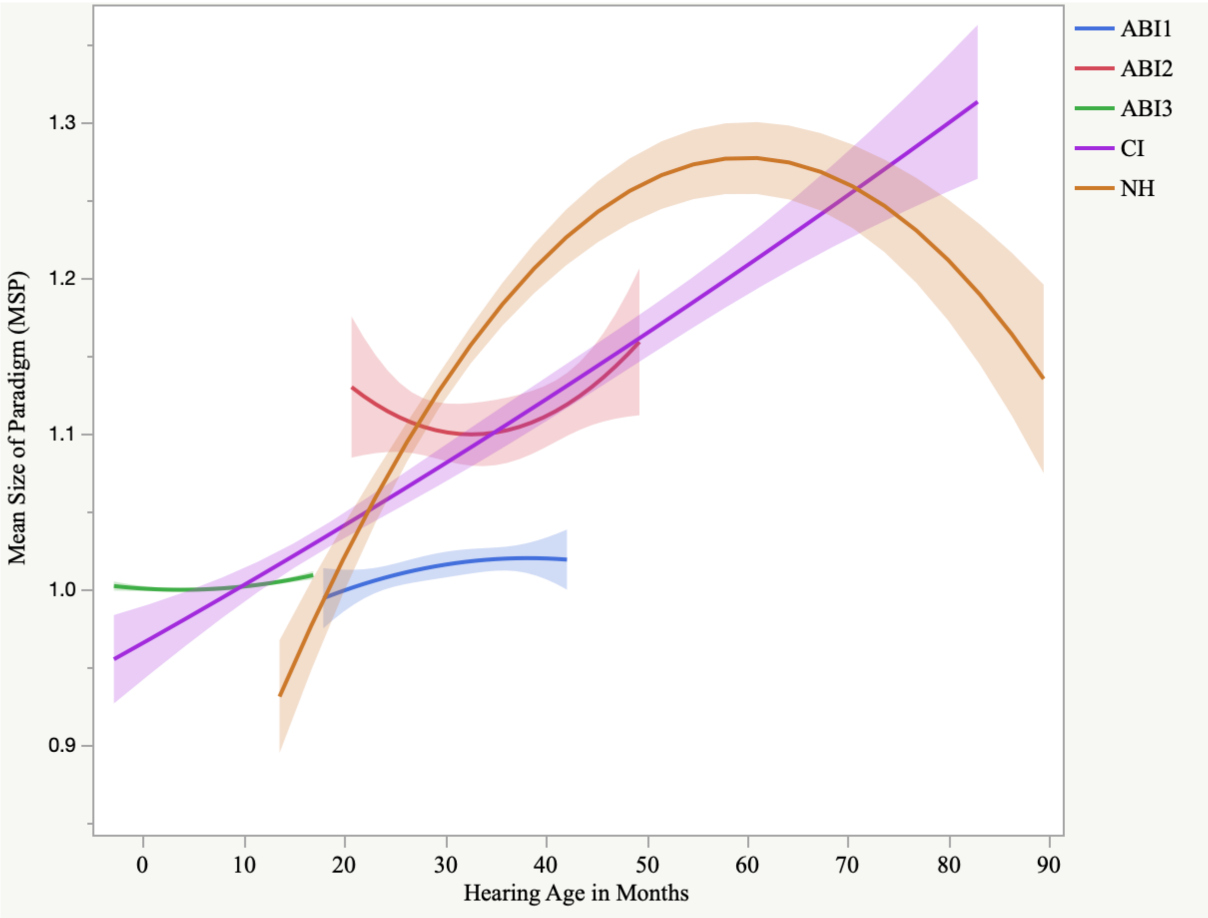


Figure 2. Quadratic fit of observed MSP of all children: comparison on chronological age and hearing age

Statistical analyses: comparisons on hearing age

In Table 2, the fixed effect results of ABI1 are shown, both for MLU and MSP. Longitudinal data of children with CI and children with NH are matched for hearing ages between 12 and 41 months, as these were the data available for ABI1.

At the intercept, i.e. 12 months of hearing age, MLU is estimated at 0.97 and MSP at 1.00 for ABI1. So, on average, an utterance comprises one word and the child uses only one form per verb. There is no significant effect of hearing age in the MLU and MSP values ($p>0.05$ in both analyses). In other words, there is no significant increase of MLU and MSP between 12 and 41 months of hearing age in ABI1. For both measures MLU and MSP, there is no significant difference between ABI1 and children with CI nor between ABI1 and children with NH at the intercept. Both the main effect of hearing status [CI] and the main effect of hearing status [NH] are not significant ($p>0.05$) in the analyses for MLU and MSP. But, there are significant interaction effects between hearing age and hearing status [CI] and [NH] in the MLU analysis ($p<0.0001$ for the two analyses shown in Table 2) and in the MSP analysis (resp. $p<0.01$ and $p<0.0001$ as shown in Table 2). These interaction effects suggest that there is a significant increase of MLU and MSP in both control groups as hearing age increases. In that sense, the difference between ABI1 – who does not show an increase of MLU and MSP with hearing age – and the other control groups becomes significant at the older hearing ages.

Table 2. Fixed effect results of ABI1 for MLU and MSP.

	Estimate	**SE**	**t-value**	**p-value**
	MLU			
Intercept	0.97	0.73	1.33	>0.05
Hearing age	0.02	0.01	1.43	>0.05
Hearing status [CI]	0.31	0.77	0.41	>0.05
Hearing status [NH]	-0.92	0.76	-1.22	>0.05
Hearing status [CI] x Hearing age	0.08	0.01	6.42	<0.0001
Hearing status [NH] x Hearing age	0.15	0.01	12.54	<0.0001
	MSP			
Intercept	1.00	0.08	13.93	<0.0001
Hearing age	0.00	0.00	0.70	>0.05
Hearing status [CI]	0.00	0.07	0.03	>0.05
Hearing status [NH]	-0.07	0.07	-0.96	>0.05
Hearing status [CI] x Hearing age	0.00	0.00	2.62	<0.01

Hearing status [NH] x Hearing age	0.01	0.00	7.06	<0.0001

An estimate and/or SE of 0.00 indicates a value smaller than 0.01

In Table 3, the fixed effect results of ABI2 are shown for MLU and MSP. Data of children with CI and children with NH are matched between hearing ages 22 and 48 months, i.e. the available data range for ABI2.

At the intercept, i.e. 22 months of hearing age, MLU is estimated at 1.54 and MSP at 1.10 for ABI2. The average sentence comprises one and a half word and the child uses approximately one form per verb. For MLU, there is a significant effect of hearing age ($p<0.05$), showing that ABI2's utterances become significantly longer with prolonged hearing experience. For MSP, there was not such an effect ($p>0.05$), indicating that the child did not incrrease the number of word forms per verb. At the intercept, i.e. 22 months of hearing age, there were no significant effects of hearing status [CI] and hearing status [NH], suggesting similar MLU and MSP values in all children ($p>0.05$ in all analyses). However, there were significant interactions between hearing age and hearing status [CI] and hearing age and hearing status [NH] for MLU ($p<0.001$ and $p<0.0001$) as well as MSP ($p<0.01$ and $p<0.0001$). These effects point out that the increase of MLU and MSP values with more hearing experience is more outspoken in the CI and NH groups than in ABI2. In other words, the differences between ABI2 and the two control groups significantly increase with hearing experience.

Table 3. Fixed effect results of ABI2 for MLU and MSP.

	Estimate	**SE**	**t-value**	**p-value**
	MLU			
Intercept	1.54	0.80	1.92	>0.05

Hearing age	0.03	0.01	2.15	<0.05
Hearing status [CI]	0.72	0.84	0.86	>0.05
Hearing status [NH]	0.39	0.83	0.47	>0.05
Hearing status [CI] x Hearing age	0.05	0.02	3.05	<0.001
Hearing status [NH] x Hearing age	0.11	0.01	7.42	<0.0001
MSP				
Intercept	1.10	0.08	14.63	<0.0001
Hearing age	0.00	0.00	0.89	>0.05
Hearing status [CI]	-0.05	0.08	-0.65	>0.05
Hearing status [NH]	-0.06	0.08	-0.77	>0.05
Hearing status [CI] x Hearing age	0.00	0.00	2.96	<0.01
Hearing status [NH] x Hearing age	0.01	0.00	8.84	<0.0001

An estimate and/or SE of 0.00 indicates a value smaller than 0.01

In Table 4, the fixed effect results for MLU and MSP of ABI3 are displayed. Only longitudinal data of children with CI were available, matched between hearing ages -2 and 16 months, i.e. ABI3's hearing ages with the ABI device. The intercept was set at the beginning of the ABI data, i.e. 2 months of hearing age.

At the intercept, ABI3's MLU is estimated at 1.19 and ABI3's MSP at 1.00. In other words, the child's utterances comprise on average 1.19 words and the child uses one word form per verb. The lack of a significant effect of hearing age (p>0.05 in Table 4), suggests that MLU and MSP do not change significantly with longer hearing experience. At the intercept, children with CI's MLU and MSP values estimated at slightly lower values than those of ABI3, but these differences were not significant (p>0.05 in both analyses). But whereas ABI3 did not show an increase with hearing age, children with CI did show an increase. The significant interactions between hearing age and hearing status [CI] for both measures ($p<0.0001$ for MLU and $p<0.05$ for MSP) indicate that the difference between ABI3 and children with CI enlarges with hearing experience, with children with CI outperforming ABI3 when they have used their implants for a longer period.

Table 4. Fixed effect results of ABI3 for MLU and MSP.

	Estimate	SE	t-value	p-value
	MLU			
Intercept	1.19	0.30	4.03	<0.0001
Hearing age	0.01	0.01	0.97	>0.05
Hearing status [CI]	-0.56	0.32	-1.77	>0.05
Hearing status [CI] x Hearing age	0.06	0.01	4.83	<0.0001
	MSP			
Intercept	1.00	0.01	82.51	<0.0001
Hearing age	0.00	0.00	0.64	>0.05
Hearing status [CI]	-0.01	0.01	-1.00	>0.05
Hearing status [CI] x Hearing age	0.00	0.00	2.23	<0.05

An estimate and/or SE of 0.00 indicates a value smaller than 0.01

Discussion

The present study investigated the grammatical development of three children with an auditory brainstem implant and compared it with the grammatical development of congenitally hearing-impaired children with a cochlear implant and children with typical hearing. Two aspects were analyzed: mean length of utterance (MLU), as a proxy for syntagmatic development, and mean size of paradigm (MSP) of verbs, as a proxy for paradigmatic development. The children's longitudinal development was cast against their chronological ages, meaning that the ABI, CI and NH children's development was compared in the same chronological age time window. In addition, their development was aligned relative to the amount of hearing experience, i.e., their hearing age. Three conclusions can be drawn from the results: (1) as expected, the three children with ABI lag considerably behind their peers with NH and peers with CI when compared on chronological age. (2) When matching the groups instead on hearing age (i.e. length of hearing experience), the difference between the children with ABI and children with CI and NH diminished, but was still significant. And (3), ABI2 outperformed the other two children with ABI with respect to MLU and MSP, even at similar hearing ages, suggesting considerable interindividual variation among the users of ABI.

Group comparisons

Compared to children with NH of the same chronological age, the hearing-impaired children have significantly lower MLU and MSP values. Their utterances are on average shorter and they use less different word forms per verb lemma. However, the development over time of the hearing-impaired children is quite different. Whereas the children with CI's MLU values show a considerable increase, the increase is far less outspoken in the case of the children with ABI. Moreover, notwithstanding the initial delay of children with CI in comparison with their peers with NH, they seem to be closing the gap over time. This development has been reported in the literature: early implanted children with CI appear to catch up with their hearing age-mates and seem to have closed the gap around the age of five (Faes et al., 2015; Hammer, 2010; Nicholas & Geers, 2007). A similar development is not apparent in children with ABI: although their MLU and MSP increases very slightly over time, the increase is insufficient to even start closing the gap with their age-matched peers with CI and NH.

However, the group differences are smaller when hearing age is used as the basis for comparison. For instance, for children with CI, the differences with children with NH disappeared almost completely for MLU and partially for MSP (Figures 1 and 2). For the three children with ABI, the differences were reduced as well, especially for ABI2, but they still caught the eye. In other words, the grammatical development of children with ABI is considerably lagging behind that of peers with NH and CI when matched on chronological age. Three-to-six-year old children with ABI are far from approaching the syntagmatic and paradigmatic skills of three-to-six-year old children with CI and children with NH. In turn, children with CI are also lagging behind their age-matched peers with NH, but are catching up (Faes et al., 2015). When the groups were matched on hearing age, the differences between children with NH and those with CI became much smaller and disappeared almost completely. Children with ABI developed in the direction of those with CI and NH but their slower development and less pronounced progress was striking.

Even though children with ABI were matched on hearing age with children with CI and NH, their grammatical measures MLU and MSP significantly lower. The children with ABI produce shorter sentences and use fewer different forms per verb lemma. For all children with ABI, their average utterances were one-word utterances, and they produced one verb form per lemma. With the available longitudinal data, possible progress could be traced over a period of about two years of hearing experience in each child with ABI. However, children did not seem to increase their sentence length or use more different forms of a particular verb. Neither of the three participants with ABI showed a significant effect of hearing age on their MLU and MSP values. The only exception was ABI2, whose utterance length (MLU) slightly increased with longer hearing experience. Children with CI and children with NH also started with one-word utterances and only one verb form per verb paradigm. But, in contrast to the three children with ABI, they expanded their utterances and verb paradigms with increased hearing experience. Consequently, the difference between children with ABI and children with CI and NH enlarged with prolonged hearing experience.

At the beginning of each child with ABI's data, there were no apparent differences with the control groups. This is not surprising given the chronological age – hearing age dichotomy discussed earlier. At the start of the data collection for ABI1, for instance, children with NH had a chronological (and thus also hearing age) of one year. At this age, children with NH are just starting to use basic word forms in one-word utterances. Also, the average child with CI had a chronological age of two years (with one year of hearing age) at the start of ABI1's data. At two years of age, multiword utterances are, on average, relatively rare in children with CI and NH's production. However, with increasing hearing experience utterance length increases and verb paradigms expand in the children with CI and NH. However, this development was not noticed in the three children with ABI, not even in the most advanced, ABI2.

Individual variation in children with ABI

Although our study group of children with ABI is still fairly limited – only three children participated – there are some striking patterns: on the one hand, their development is fairly similar (e.g., hardly any significant effect of hearing age on MLU and MSP), but, on the other hand, there is a considerable amount of interindividual variation in the ABI group. First of all, the grammatical development observed in ABI3 seems largely due to the child's CI experience. A closer look at the graphs suggests that ABI3 reaches a MLU value of one already at earlier hearing ages, even before offset of ABI data (depicted by a negative hearing age), i.e., before the activation of his ABI. In a similar vein, ABI3 has a paradigm of one word per verb form already at the start of his ABI experience. This suggests that ABI3 is benefitting from the CI to reach these levels of grammatical development, rather than from the ABI already at these early hearing ages. For the period studied here, the child did not show any substantial increase of syntagmatic or paradigmatic richness that seems to result from the use of the ABI device. For speech perception, Batuk et al. (2020) and Friedman et al. (2018) already suggested that children seem to benefit from the CI-ABI combination rather than a CI-only or ABI-only condition. Therefore, it may be that the effect of the ABI on ABI3's grammatical performance is yet to come: the perceptual gain could generate some benefit to the child's productive speech skills with ABI experience.

Secondly, ABI2 is outperforming the other children and ABI1 is lagging behind compared to the other two. With respect to syntagmatic development (MLU), ABI1's values are lower than those of ABI2 and ABI3 at similar hearing ages, and ABI3 is probably benefitting from his CI experience resulting in markedly higher values. Also for paradigmatic development (MSP), the first verbs only appeared at 19 months of hearing age for ABI1 (see Figure 2), even though the data collection started already at 12 months of hearing age. In contrast, ABI3 had already an MSP of one at earlier hearing ages, likely due to the CI use. But even with the benefit of the CI, the syntagmatic and paradigmatic development of ABI3 is less advanced than that of ABI2. In addition, ABI2 is also outperforming ABI1 on both measures. The difference between ABI2 and the other children with ABI can clearly be derived from the figures. But the statistical analyses also pointed into the same direction: the intercept of ABI2 was set at 22 months of hearing age, whereas these for ABI1 and ABI3 were much earlier. Yet, the MLU and MSP values of ABI2 were not shown to differ significantly from the CI and NH groups of children at the intercept, but only at the older hearing ages. For ABI1 and ABI3, instead, the non-significant differences with CI and NH groups were only present at much earlier hearing ages of 2 months and 12 months for ABI3 and ABI1 respectively.

So, overall, ABI2 is outperforming ABI1 and ABI3 on grammatical development, even though this last child could benefit from the CI. Also for lexical and phonological development, considerable variation between children with ABI was found in the literature (Eisenberg et al., 2018; e.g. Faes & Gillis, 2019b; Faes & Gillis, 2020, 2021). It is unclear which factors add to ABI2's more developed grammatical performance. Factors such as age at implantation and hearing thresholds with ABI – known to impact children with ABI's speech perception and speech production (Aslan et al., 2020; Sennaroglu, Colletti, et al., 2016) – are similar in ABI1 and ABI2 and can therefore not crucially explain the differences between the two children. In a similar vein, none of the three children were reported to have additional disabilities, another factor often shown to impact children with ABI's speech and language development (Sennaroglu, Colletti, et al., 2016; van der Straaten et al., 2019). More research is needed to disentangle other factors that contribute to these individual differences.

Concluding remarks

Children with ABI are lagging behind their hearing age-matched peers with CI and NH on grammatical development (MLU and MSP). This delay is also apparent, though less pronounced, when the children are matched on hearing age. Initially, a similar observation holds for children with CI compared to children with NH but they appear to close gap, which is definitely not the case for children with ABI. We can only speculate as to what causes these differences in the language development of children who are equipped with the two devices. Hence, the following observations are tentative and underline the urgent need for further research.

First of all, the speech signal provided by the CI and the ABI is poorer as compared to that available in normal hearing (Drennan & Rubinstein, 2008). Even though to date little is known about the precise difference in speech signal provided by the ABI versus the CI, it may be assumed from the literature that the signal provided by the ABI is even poorer as compared to the CI (Wong et al., 2019). Especially for MSP, this degraded speech perception is likely to impact the results. In Dutch, verbs are predominantly inflected by adding a suffix (e.g., /t/ or /ə/) to the stem. Such unstressed suffixes are of low salience even for children with NH. Therefore, it is likely that children with ABI – and to a lesser extent also children with CI – are missing these low salient unstressed items in speech perception. As speech production relies on speech perception, this inevitably affects hearing impaired children's production of low salient suffixes as well. MSP takes into account the different forms of a verb that are produced. But these verb forms often only differ by a low salient unstressed suffix. For instance, the difference between first and the second and third person of most regular verbs is the suffix -*t*, similar to the third person -*s* in English (*ik werk 'I work' vs. jij werkt 'you work', hij werkt 'he works',* similar to *I work, vs. he works)*. The more limited perception of such suffixes most probably affects their production. In addition, children with CI are shown to be less attentive to speech as compared to children with NH (Houston & Bergeson, 2014) and have low executive functioning skills (e.g., attention deficits) (Kronenberger et al., 2014). Therefore, they are assumed to focus more on salient items in the speech signal (Svirsky et al., 2002) at the expense of the low salient grammatical morphemes. In the future, research is needed to find out if similar effects play a role in children with ABI.

Secondly, different aspects of children's working memory skills are related to measures such as MLU and MSP. For the production of sentences (sentence planning, MLU) as well as for the production of different verb forms per lemma (MSP), children need to store information in their mental lexicon and working memory. Since little information is available for children with ABI, we will first explain the CI-NH difference and then come back to the ABI group.

For children with CI, it has already been shown that storage of information is problematic (Nittrouer et al., 2013). For MSP, this storage problem in children with CI is linked to the mental lexicon. In order to produce different word forms per lemma – which results in higher

MSP counts –, these word forms need to be stored in the lexicon first. If fewer word forms are stored, as in the CI group as compared to the NH group, fewer can be produced as well and, consequently, MSP values will be lower. Turning to MLU, problematic storage in working memory affects sentence length (MLU). When producing a sentence, two parts of working memory are especially active: phonological short-term memory for storing information, and the general executive (Baddeley, 2003). The longer the sentence, the more active the general executive will become, reducing the capacity of the phonological short-term memory. So, longer sentences are increasing the cognitive load, since more information must be stored and handled in a phonological short-term memory with reduced activity (Willis & Gathercole, 2001). In children with NH, auxiliaries are for instance omitted in longer sentences due to processing limitations (Valian, 1991). In other words, increased cognitive load will reduce sentence length (Charest et al., 2015). In children with CI, phonological short-term memory is poorer as compared to children with NH (Burkholder & Pisoni, 2003; Cleary et al., 2001; Kronenberger et al., 2013; Pisoni & Cleary, 2003, 2004; Pisoni et al., 2010). Hence, when sentences become longer, phonological short-term memory is reduced even more. Therefore, it is likely that the performance on sentence length lags behind in the children with CI as compared to children with NH.

For children with ABI, it remains unknown if similar effects of reduced storage, phonological short-term memory and working memory are playing a role in their even poorer performance on grammatical measures such as MLU and MSP. However, delayed onset of language experience is often linked to poorer working memory skills in children (Holmes et al., 2010; Marshall et al., 2015). Given the fact that children with ABI had a prolonged time of auditory deprivation as compared to children with CI, the effects found in children with CI (such as poor phonological working memory skills, poor storage of information, poor executive functioning, poor attention to speech) are expected to be even more prominent in children with ABI. In that case, the effects of these working memory and storage problems will be even more outspoken in the ABI group, resulting in poorer language scores – as witnessed by the literature on children with ABI in comparison to children with CI (and NH). Yet, more research is needed to test these hypotheses.

Thirdly, the poor performance of children with ABI on MLU and MSP can be linked to their lexical development. Research has shown that the children with ABI studied here, expand their lexicon sizes continuously, but that they are lagging behind when compared to children with CI and children with NH (Faes & Gillis, 2019b - the same children with CI were included in both studies). But to produce longer sentences and more verb forms per lemma, these words must be acquired. For children with CI, poor attention to speech and reduced speech perception skills were already linked to their poor phonological representation of words and consequently to their poorer word learning skills. It may be the case that a similar effect is present in the children with ABI. Their speech perception is even more reduced, which may impact their novel word learning. For children with CI, lexical development is one of the better developed aspects of language (e.g. Duchesne et al., 2009). Moreover, in Faes et al. (2015), MSP is shown to be more closely related to lexical development than MLU: inflected word forms are unique words to be incorporated in the mental lexicon (e.g. Lukatela et al., 1987; Lukatela et al., 1980). Therefore, children with CI seem to catch up on their peers with NH earlier for MSP than for MLU. For children with ABI, it is as yet unknown if lexical development is one of their strengths in language development. In addition, even though they were expanding their lexicon size, this expansion was very slow (Faes & Gillis, 2019b). It is unclear if this slight increase in novel word learning could affect sentence length and verb paradigms. Therefore, more information is needed in the types of words learned by the children. To increase sentence length, for instance, children with ABI must be learning function words in addition to content words. To date, no information is available on development of open-class and closed-class words. In more general terms: the nature of children with ABI's lexical development is uncharted territory that still needs to be explored in more depth.

To conclude, children with ABI's morphosyntactic development is extremely slow. Even four years after implantation, the mean length of utterance of the best performing child in this study (ABI2) did not surpass two words and the average number of verb forms per lemma ranged between one and one and a half. Currently, there is little other information available in the literature with respect to the morphosyntactic development of children with ABI. Whereas some studies already looked into lexical and phonological development (e.g. Eisenberg et al., 2018; Faes & Gillis, 2019b, 2020, 2021; Teagle et al., 2018; Wilkinson et al., 2017), other aspects of language development remain unresearched. Our results show that grammatical development is fairly limited, even in a group of children with ABI that can be considered as the better performers in the ABI population.

Declaration of interest

There is no conflict of interest to be reported.

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