Speech and language in congenitally deaf children with a cochlear implant

Steven Gillis, University of Antwerp

To appear as:

Gillis, S. (2017). Speech and language in congenitally deaf children with a cochlear implant. In A. Bar-On & D. Ravid (Eds.), *Handbook of Communications Disorders: Theoretical, Empirical, and Applied Linguistic Perspectives*. Berlin: Mouton De Gruyter.

Content

1. Introduction

1.1. Congenital hearing loss and cochlear implants

Approximately 3 out of 1,000 neonates suffer from hearing loss. A severe-to-profound (> 70 dBHL) bilateral hearing loss is detected in approximately 1 neonate per 1,000 births. Thus, the incidence of hearing loss is relatively high as a congenital deficit. In addition, hearing loss can be progressive after birth, can have a delayed onset or can be caused by exogenous factors early in life, which means that the estimated prevalence of hearing deficits in the early years will be higher than the 1/1000 of babies with congenital hearing impairment (Kral & O'Donoghue 2010). It is estimated that two-thirds of the children with hearing loss show the deficit at birth (Mahdieh et al. 2012). In any case, until the 80ies of the previous century, babies with a severe-to-profound sensorineural hearing loss could be fitted an acoustic hearing aid -- which did not address their cochlear malfunctioning -- in order to assist their residual hearing as well as possible. But it was not until the advent of cochlear implants (CI) that their hearing loss could be reduced to areas between 20 and 40 dBHL, which represents a “mild” hearing loss.

A cochlear implant is an electronic device that functions as a sensory aid, converting mechanical energy into coded electrical pulses that directly stimulate the auditory nerve fibers, bypassing damaged or missing hair cells in the cochlea. Part of the CI is surgically inserted into the cochlea and the mastoid, and the remaining part is worn externally. The external components consist of a microphone, a signal processor, and a transmitter coil. The microphone receives acoustic signals and converts them into an analog electrical signal that is sent to the processor, which converts the signal into an electrical or digital pattern that is transmitted to the internal part by means of the two coils (the external transmitter coil and the internal receiver coil) through the skin. The internal part then stimulates the electrodes in the cochlea. The electrodes are thus able to deliver electrical stimulation to excite the cochlear neurons of the auditory nerve.

This description pertains to “conventional” cochlear implants: direct electrical stimulation of the auditory nerve. However even in cases of severe hearing deficit, residual hearing may remain, most of the time in the lower frequencies, which may still be amply stimulated by an acoustic hearing aid. That is why a new generation of devices combines a cochlear implant for high frequency hearing loss and acoustic hearing aid in the same ear for sustaining low frequency hearing. This type of cochlear implant is referred to as a hybrid cochlear implant or as combined electric and acoustic stimulation (EAS, James et al. 2006). Also other types of implantable hearing devices are marketed now, such as several kinds of implantable middle ear devices and brainstem implants for those cases in which conventional hearing aids or cochlear implants are not suited: at present a cochlear implant is the best solution for individuals with a well functioning auditory nerve but with a malfunctioning cochlea (Möller 2006).

1.2. Early detection of hearing loss

Early auditory experience is quintessential for developing speech and spoken language skills (Jusczyk 1997, Oller 2000). For this reason impaired hearing should be detected as early as possible so that rehabilitation can start at a very young age (Yoshinago-Itano 2004, Yoshinago-Itano et al. 1998). The purpose of newborn hearing screening is to test babies' hearing in the first few days after birth, or at least within the first months of life. There are objective methods for screening neural responses to sound stimuli. These electrophysiological measures detect automated auditory brainstem responses (AABR) or otoacoustic emissions (OAE) invoked by sound stimuli (usually clicks). Nowadays OAE and AABR can routinely be measured and are absolutely non-invasive: babies can easily sleep through the procedure of the hearing test.

Screening programs which aim to reach every single neonate using OAE and AABR have been implemented in various countries. For instance, in the USA the *EHDI* program[1] (*Early Hearing Detection and Intervention*) aims to identify every newborn with a permanent hearing loss before 3 months of age and to provide "timely and appropriate intervention services before 6 months of age". In Flanders, the northern part of Belgium, the governmental child well-being organisation *Kind & Gezin* launched a UNHSP (Universal Neonatal Hearing Screening Program) in 1998 (De Raeve 2006, Desloovere et al. 2013). The impact of this program was impressive: the age at which babies were diagnosed with a hearing loss decreased from 12 months -- before the program had started -- to 1 month. Consequently the age at first intervention decreased from 13 to 2 months, the first hearing aids were fitted at 3 months, and a growing number of cochlear implantations took place during the first and second year of life (De Raeve & Wouters 2013). As such, early detection is but a first, though indispensible, step in a long chain of rehabilitation steps. The Flemish example shows, for instance, that each year approximately 96% of all neonates are screened. Moreover, by integrating screening, diagnosis, early intervention and

[1] http://www.infanthearing.org/states_home/index.html, accessed 15/12/2014.

rehabilitation in one program (via a well-defined cooperation protocol between different caregivers and health services), it became a unique project with long-term effects: referral for a cochlear implant assessment can be accomplished before the age of nine months (Philips et al. 2009). Children eligible for it receive a CI on average between 14 and 16 months of age, and even before their first birthday. In the preschool population (i.e., between approximately two and six years of age) 94% of the children with a profound hearing loss had received a cochlear implant in 2010, and 25% of them were wearing bilateral implants (De Raeve & Lichtert 2012). Of the children with cochlear implants (and no additional disabilities), 85%-90% eventually enter mainstream education (De Raeve 2014, De Raeve & Lichtert 2012, Desloovere et al. 2013, Van Kerschaver et al. 2007).

This description of UNHSP pertains to the region of Flanders, the Dutch speaking part of Belgium. Elsewhere the situation may not be identical, for instance, in the French speaking part of Belgium, UNHSP started only in 2007 (Vos et al. 2014), but most developed countries have started screening programs (Nikolopoulos 2015).

Thus, in the end, early detection and proper rehabilitation of hearing impairment has a positive effect on speech perception and production, and language development (Geers 2006, Yoshinago-Itano et al. 1998), but also on a further broad spectrum of aspects of an individual’s personal, social, educational, and professional life (Hoffman et al. 2014, Kochkin 2010, Quittner et al. 2004).

1.3. Cochlear implant candidacy and audiological outcome

In the early days of pediatric cochlear implantation, candidacy requirements included an unaided pure-tone average of 90 dBHL or more, and aided thresholds of

60 dBHL or higher, and in addition absence of speech discrimination and word-recognition with well-fitted hearings aids. But the guidelines for CI candidacy have developed over the years (Geers 2006). For instance, currently in the USA FDA guidelines[2] permit implantation in children above two years of age with severe hearing loss, i.e., a hearing loss of 70 dBHL or higher. For children in their second year of life, profound deafness, i.e., a hearing loss of at least 90dBHL, is the criterion. Moreover several centers in the USA provide cochlear implants to children in their first year of life "'off-label' when there is strong evidence that an infant is profoundly deaf and not progressing in his or her speech and hearing development with hearing aids." (Houston et al. 2012: 459). In this respect there is variation in the selection criteria that hold in different countries[3].

Most implant users' thresholds lower to 20 to 40 dBHL across all frequencies, i.e. a mild hearing loss. This implies that the implant enables detection of virtually all speech sounds and provides a hearing sensitivity and functionality superior to that obtained with conventional acoustic hearing aids. A sensorineural hearing loss is characterized by an elevated threshold on pure-tone audiometry and by a lower frequency resolution. A good frequency resolving power of the cochlea is essential for normal speech and language development: hearing impaired individuals not only fail to hear many sounds, if they do hear them, they often fail to discriminate them. Conventional hearing aids amplify sounds, but they do not improve the frequency discrimination. That is, a hearing impaired individual will perceive sound better with

[2] http://www.nidcd.nih.gov/health/hearing/pages/coch.aspx

[3] For instance, in Belgium the official reimbursement criteria (by social security) since 2006 are (1) pure tone average thresholds of 85 dB HL or greater at 500, 1000, and 2000 Hz; (2) threshold of peak V in brainstem auditory evoked potentials at 90 dB HL or higher; (3) little or no benefit from hearing aids (De Raeve & Wouters 2013).

a hearing aid, without necessarily understanding better what is said. Cochlear implants by contrast not only amplify sound but they also aim at a (partial) restoration of the frequency resolution of the cochlea. This is the major advantage of a cochlear implant over a hearing aid in cases where the hearing loss is severe-to-profound and the cochlear tuning is deficient. It is, however, wise to keep in mind that given this remarkable advantage, cochlear implants still have their limitations: they do not restore normal hearing, outcomes vary among patients, and are very dependent on the actual fitting of the device, and performance is still considerably degraded by ambient noise (O'Donoghue 2013).

2. Deaf children with a cochlear implant: a "moving target"

Studies of spoken language developmental in deaf children with a cochlear implant started appearing in the last decades of the previous century and since then the population has received a growing interest judging from the sheer number of scientific writings. Trying to grasp the gist of children's speech and language development is not trivial since the population can be characterized as "a moving target", to use the words of Geers (2006). Moreover studies employ different methodologies so that research findings are not always easily comparable.

In this section we will review some of the intricacies of this population: on the one hand, cochlear implantation is a fairly recent technological innovation, and new developments are announced regularly. These developments are manifold, for instance, new hardware for the device or innovations in its speech processor. Innovations also concern the age at which children are eligible for implantation: the age when the intervention is allowed to occur has lowered substantially over the past decades. In the USA cochlear implants have been FDA-approved for use in eligible children beginning at 12 months of age since 2000. But in the same year the first child under the age of 6 months received her implant in Belgium. Thus a number of changes have occurred – technological innovations, changes in the eligibility of the pediatric cochlear implant users, etc. -- which makes the sample of CI children reported on in the literature “an ever moving target”. On the other hand, these changes and developments have occurred simultaneously, which constitutes another aspect of this “moving target”: it is difficult to determine the impact of each one of them separately on young children’s speech and language development.

Cochlear implant technology has evolved tremendously in the last three decades. For instance, the algorithms that process the incoming speech signals have provided much more details in consecutive generations of devices. The speech extraction schemes in the early implants presented only limited spectral information (fundamental frequency, first two formants) providing ample support for lip reading but were only a very restricted aid to speech perception (Clark et al. 1983, Dowell et al. 1985). More sophisticated strategies developed later provided more information, especially in the higher frequencies, leading to better speech perception (Clark 1989). In the mid-90ies, the introduction of the bandpass filtering principles led to even further enhancements in representing speech features and in a higher rate. These improvements in the speech processing algorithms each led to improvements in speech perception (Sarant 2012). A development in the last decade aims to make use of the (low-frequency) residual hearing of the implantees. For this purpose electrical stimulation is combined with acoustic stimulation, leading again to improved speech perception (Campbell et al. 2013).

Another telling example concerns the number of electrodes in the CI device. The first children were implanted in Melbourne in 1980 with the single channel House cochlear implant (Eisenberg & House 1982). In 1985 the first children received a multichannel CI, and nowadays implants are used with up to 24 channels.

Hence, much more information can be analyzed by the speech processor, and increasingly more information can be transmitted. As these developments enhance speech perception, they also have a vast impact on children's language and speech production (Geers 2006, Peng et al. 2004, Sarant 2012). But this implies a "moving target": research findings that were obtained in children equipped with devices with older generation technology should be cautiously compared with findings in children using newer generation technology. Due to i.a. improved technology the latter have access to more and higher quality information from the device, and hence their understanding and production of speech can be expected to be superior (Geers & Nicholas 2013).

But there is more to it than only technological advances. In two recent studies, the speech intelligibility of 63 CI users was investigated (Montag et al. 2014, Ruffin et al. 2013). Their experience with the device varied from 8 to 18 years, with a mean of 12 years, and they had all received their implant before the age of 7. Systematic differences were found in the group of participants: CI users who used their implant longer appeared to be less intelligible than those with a shorter period of device experience. More specifically, they observed age cohort effects: participants with more than 15 years of CI use were less intelligible than participants with 7 to 9 years of experience. At first sight, this is a surprising finding, since it could be expected that the more experience a CI user has the better the performance would be. Looking for an explanation of this seemingly counterintuitive finding, Montag et al. (2014) found that the duration of CI use strongly correlated with the chronological year in which the implant took place. Over the past decades the medical and audiological criteria for CI candidacy and age of implantation have changed significantly. It appeared that, indeed, the participants with more years of device experience were also implanted at an older age. Hence, the age cohort effect apparently reflects the decrease in the age at which hearing impaired children have been implanted. In other words, the participants' age at implantation was a significant predictor of their long-term speech intelligibility scores, and the higher age at implantation in the older cohort accounted for the relationship between age cohort and language intelligibility outcome. Thus, changes in the clinical practice over the years – a demographic variable -- have an impact on the language outcomes in CI users.

In addition to this "moving target" there is also a "vanishing target". The ultimate aim of auditory restoration is for children with a cochlear implant to (eventually) reach a level of speech and language comparable to that of normally hearing peers. This implies that researchers compare spoken language of implanted children with a matched group of normally hearing children. But from another perspective, children with a cochlear implant have a severe-to-profound hearing loss, and, hence, researchers want to find out if their speech and language performance is comparable or better than that of a matched group of children with comparable auditory characteristics (aided and unaided thresholds) wearing acoustic hearing aids. However testing comparable groups of cochlear implant and hearing aid users has become increasingly difficult because the number of available profoundly deaf hearing aid users who do not elect to receive a cochlear implant is becoming smaller and smaller, and thus they constitute a “vanishing target” (Geers 2006). However from previous research it can be concluded that on average children with a CI have more effective speech and language skills than children with similar hearing loss wearing acoustic (or tactile) hearing aids (Svirsky et al. 2000).

3. Characteristics of spoken language development

3.1. Development assessed with standardized tests

Bearing in mind that children with a cochlear implant constitute “a moving target”, the published results of language and speech assessments should be evaluated with care (Montag et al. 2014). The studies reported in the literature take – broadly speaking -- two different methodological strands: administrating standardized tests and analyzing (spontaneous) speech samples. Standardized tests such as the *Reynell Developmental Language Scales (RDLS)* or *the Peabody Picture Vocabulary Test (PPVT)* are used to evaluate children’s expressive and/or receptive language abilities. One of the advantages of such standardized tests is that they allow to determine the *language age* of children. For instance, a language age or age-equivalent of 3 years means that the child has the language skills equivalent with those of typically developing three-year-olds. If the language age differs from the chronological age, then the *language quotient* (i.e., language age divided by chronological age) provides an estimate of the delay or the advance of the child’s linguistic abilities. In order to estimate the relative progress over time, the rate of development is computed by dividing the change in the language age by the change in chronological age over a particular period of time.

In a large-scale study of 188 children with a cochlear implant, Niparko et al. (2010) investigated children’s progress in spoken language comprehension and production with the RDLS. The children were implanted before the age of five, and were tested every six months for a period of three years starting when their device was activated. Three age groups participated: (1) a group implanted before 18 months (N=72), (2) a group implanted between 18 and 36 months (N = 64), and (3) a group implanted after 36 months (N=52). Several important findings are reported. In comprehension as well as in production, children with a CI make considerable progress, but on average the developmental trajectories were significantly slower compared with normally hearing peers. But this *average* picture hides an important characteristic of the population of implanted children that has been remarked by many other investigators: the trajectories are markedly more variable in the CI group in comparison with normally hearing children. This means that there is more variation between CI children than between NH children. Part of the variation comes from the different ages at which the children received their CI. In the youngest group (implanted before 18 months) the developmental trajectories for comprehension and production were significantly steeper than in the two other groups. The increase of the children’s abilities in the youngest group was even comparable to that of the normally hearing control group, but the increase in the two other groups was slower than that of normally hearing children. This means that for the youngest implanted children the gap with the normally hearing ones did not widen, while for the later implanted ones, the gap did not become narrower. But since the hearing impaired children already started at a much lower level, even the youngest implanted children still had a delay at the endpoint of the study, i.e., three years after implantation. In order to illustrate what this entails: the expressive level of normally hearing children age 2.3 years is attained by the youngest group of CI children at the age of 3.4 years, by the children implanted between 18 and 36 months at 4.5 years, and by the even later implanted children at 5.2 years.

Variation between children's language abilities at a particular age are a well-documented phenomenon: also in the group of normally hearing children, the RDLS scores differed. But a second main finding of Niparko et al. (2010), also reported in many other studies, is that the variation within a group of CI children is significantly larger. Language and speech outcomes differ more considerably from child to child. This variation has an important implication: some CI children do exhibit scores comparable to age matched hearing peers, while others still show a marked language delay even after may years of device use. Boons et al. (2012a) report on a large scale study of 288 children who were tested 1, 2, and 3 years after they received a CI (mean age at CI: 2;02, SD = 1;11). Two standardized tests were administered, viz. the RLDS for receptive language skills and the SELT (Schlichting Developmental Language Test) assessing expressive language skills at the word and sentence level. Huge variation in performance was attested: some children scored age appropriately, even beyond what could be expected given their chronological age. On the other side approximately one out of four children "[…] failed to develop a language level comparable with half of their chronological age […]" (Boons et al. 2012a: 632). Thus, after three years of device use the gaps in language development between implanted children and their NH peers were not eliminated yet (Boons et al. 2012a, Niparko et al. 2010).

What language levels can be expected after an extended period of device use? Do children with CI eventually catch up with their NH peers? Geers & Nicholas (2013) report on a longitudinal study involving 60 children implanted at a mean age of 22.7 months (SD=7.7). These children were tested at the age of 4.5 years (Nicholas & Geers 2006) and again when they were approximately 10.5 years of age and had on average used their devices for 8.6 years. A number of tests were administered assessing both receptive and productive language development at the word and sentence level. Over half of the sample achieved scores within the average range for NH peers, and 73% of the children implanted before 18 months scored within that range. There were differences between tests. Measured as the percentage of children ranging within or above 1SD from the normative mean, the following percentages are reported: 82% for expressive vocabulary, 72% for receptive vocabulary, 77% for expressive language, 52% for receptive language, and 68% for a overall language score. This suggests that a majority of the implanted children have caught up with their hearing age mates for both expressive and receptive vocabulary and expressive language (including syntax), but that this is the case in a far lesser extend for receptive language measures. Listening to language, processing it, and comprehending it, appears to be much harder for CI children than producing words and sentences.

A finer assessment of the language development profiles after a prolonged use of early CI was targeted by Duchesne et al. (2009). They used the RDLS and showed that after six years of implant use, more than half of the CI children had receptive and expressive age-appropriate language skills at the word level, while less than 50% of the same group of children had receptive and expressive age-appropriate language skills at sentence level. Even though other standardized tests were used, similar outcomes were found in for instance Caselli et al. (2012), Geerset al. (2003), Schorret al. (2008) and Young & Killen (2002). Moreover Duchesne et al. (2009) identified four developmental profiles: (1) lexical and grammatical language components are within normal limits in comprehension as well as in production; (2) delayed development across domains; (3) Normal lexical abilities but a receptive grammatical delay (productive grammar was not assessed), and (4) idiosyncratic discrepancies across domains.

3.2. Development assessed by analyses of spontaneous speech

Studies of children's spontaneous speech or elicited speech in well-targeted experiments, provide an opportunity to analyze in more detail the language and speech of children with a cochlear implant and to unravel non typical phenomena and error patterns.

After implantation a burst in children's speech and language development is witnessed: growth curve analyses show a steep increase of the acquisition curve. In early implanted children the slope is even comparable to that of normally hearing children (Connor et al. 2006, Tomblin et al. 2005, 2008). This profile is independent of the age at implantation, but with later ages at implantation the slope of the curve becomes less and less steep, and, hence, the gap between development in NH and CI children becomes larger.

The onset of canonical babbling is a well-documented example. In NH children, the onset of canonical babbling is expected to occur between approximately 6 and 11 months of age (Molemans et al. 2012, Oller 2000). Children with severe-to-profound hearing loss show marked delays for babbling onset (Koopmans-van Beinum et al. 2001, Nathani et al. 2007), some do not even babble at all (Oller & Eilers 1988). After cochlear implantation children typically start babbling within a few months after the activation of their device. They need on average 4 months of auditory exposure for babbling to take off (Colletti et al. 2005, Ertmer & Mellon 2001, Moeller et al. 2007, Moore & Bass-Ringdahl 2002, Schauwers et al. 2008, Schramm et al. 2009, Wright et al. 2002). Hence CI children need less time to reach this milestone in vocal development than NH children. However, even when implanted very early in life, babbling onset in CI children is delayed in terms of chronological age, and with later ages at implantation, the delay becomes even more important.

Except for being delayed in babbling onset, does speech and language development proceed as in normally hearing infants? The general picture that emerges is that, at a very general level, development in both groups runs parallel but looking in more detail at particular phenomena reveals discrepancies. The latter will be exemplified by looking at studies of (1) children's prelexical babbling development, (2) the development of production accuracy, and (3) morpho-syntactic development.

3.2.1. Prelexical vocal development

Most published reports assessing the prelexical vocal repertoire in young, early implanted CI children indicate substantial progress after a limited amount of aided hearing experience and even close-to-normal patterns of speech (Anderson et al. 2004, Colletti et al. 2005, Ertmer & Mellon 2001, Schauwers et al. 2004). But detailed analyses of children's babbling show that at the segmental as well as at the suprasegmental level there are marked differences between children with CI and NH children. Looking at which consonants are used in canonical babbles (e.g., [baba], [mama]) it appears that children with CI use markedly more stops than NH children (Schauwers et al. 2008). They seem to make the difference in sonority between the syllable initial consonant and the vowel as large as possible (stops are the least sonorous segments, vowels the most sonorous). At the suprasegmental level, although the length of the babbles is not significantly different (Molemans 2011), CI children's babbles show significantly less variation: reduplicated babbles in which a syllable is repeated (e.g., [baba]) predominate in CI children, while in NH variegated babbles with non-identical syllables (e.g., [bama], [papu]) are significantly more frequent viewed over the entire babbling period. Thus the babbles of CI children are more repetitive as a majority of them consist of a mere repetition of the same syllable.

Thus, soon after they receive their CI, very early implanted children show a burst in their vocal development: they start to produce canonical babbles and their development seems to follow that of normally hearing children. But when homing in on specific details of those babbled utterances, particular discrepancies can be remarked, such as a predominant use of stops and an overall more repetitive structure.

3.2.2. Phonetic and phonemic accuracy of word productions

As a second example, the segmental accuracy of words is discussed. Segmental accuracy refers to the overall accuracy of children's pronunciation of words: how many phonemes are produced correctly (i.e., phonemic accuracy, which is to be distinguished from phonetic accuracy). For English-speaking children with CI, accuracy is reported to increase with longer implant use (Blamey et al. 2001, Eriks-Brophy et al. 2013, Tobey et al. 2003, Tomblin et al. 2008). After four years of implant use, overall phonemic accuracy is 62.9% (Tomblin et al. 2008). Accuracy increases after six years of implant use to 76.28% according to Tomblin et al. (2008) and approximately 86% according to Blamey et al. (2001). In Blamey et al. (2001) mean age at implantation was 3;9 (SD = 0;11) and in Tomblin et al. (2008) mean age at implantation was 4;6 (SD = 2;1).

In children implanted at an earlier age, viz. on average 1;5, segmental accuracy is shown to be significantly higher in NH children than in children with CI. At age 3;6 and after on average two years of device use, the percentage of phonemes correct is 83% for NH children and 53% for children with CI (Ertmer et al. 2012). At age 4;0, the same trend emerges in the accuracy of word initial consonants in a short sentence repetition task. For NH children of that age, all initial consonants except fricatives and affricates (86% accuracy) reach ceiling accuracy, while for children with CI initial consonant accuracy is only 62% (Ertmer & Goffman 2011). 72% of the hearing impaired children reached average scores at age 5;0 (Eriks-Brophy et al. 2013) and thus seem to have caught up with their NH peers.

In a recent study, Faes, Gillis & Gillis (in press) investigated phonemic accuracy in children acquiring Dutch, implanted at a median age of 1;0. The material consisted of spontaneous speech samples drawn from mother-child conversations.

The children were studied up till age 5 (with up to 4;6 years of device use at that age). They found that the accuracy of children with CI is lower than the accuracy of NH age-matched peers during the first year after CI activation. The delay remained significant when assessed at the age of 3 and 4, but the distance between the accuracy scores of the NH and CI children became smaller. Eventually at age 5, the children with CI reached a phonemic accuracy score that was similar to that of their NH peers.

Thus for phonemic accuracy, children with CI seem to catch up gradually with NH peers (Faes et al. in press). But looking at the fine phonetic detail of the children’s consonant and vowel productions indicates that there still remain important differences with the speech of NH children. For instance, Verhoeven et al. (in press) assessed the vowel productions of the CI children also studied by Faes et al. (in press). They found that at the age of 6, after approximately 5 years of device use, the vowel space of the CI children was significantly smaller than that of the NH age-matched peers. This suggests that CI children pronounce their vowels much more centralized and less differentiated than NH children. Comparable results are reported for vowel productions in Croatian (Liker et al. 2007), Greek (Nicolaidis et al. 2007), and German (Neumeyer et al. 2010), although results are not always equivocal (see Baudonck et al. 2011).

3.2.3. Morpho-synatctic development

Analyses of morphological and syntactic development in children’s (spontaneous) speech are rather scarce. In general, the development of CI children’s inflection is found to lag behind compared to NH peers. For instance for German, Szagun (2001) showed that inflectional morphology of CI children is less advanced compared to NH children. More precisely, case and gender marking of articles and noun plurals are less accurate in CI children. With respect to nominal plurals, NH children are found to make errors, children with CI make very similar errors, but in addition CI children frequently do not mark plurals, and therefore avoid error making (Szagun 2001). Likewise, Laaha et al. (2015) showed that Dutch and German CI children produce significantly more singular nouns compared to age-matched NH peers in an plural elicitation task. Whereas CI children have difficulties with the inflection of nouns and articles, no differences with respect to verbal morphology in German are found (Szagun 2001). For English, Guo et al. (2013) showed that tense marking is less accurate in CI children as compared to NH peers up to five years of implant use. Hammer (2010) replicated these findings for Dutch speaking children: CI children are delayed with respect to verb morphology, subject-verb agreement and past tense marking. Nevertheless, they appear to have caught up for nominal and verbal morphology by age seven (Hammer 2010).

The richness of the inflectional paradigms of nouns and verbs, i.e., how many different forms of a stem or root occur in the children's language, shows a development from a delay to age appropriate richness. Early implanted children with CI acquiring Dutch start out with significantly poorer verbal and nominal paradigms, but after approximately three years of device use, at a chronological age of 5 years, they appear to have caught up with NH peers (Faes et al. 2015).

In studies of children's spontaneous speech, a similar relative developmental pace of lexical and grammatical development is found as in studies using standardized tests: the delay of CI children covers a shorter period for lexical development as compared to grammatical development, as measured by MLU (Moreno-Torres & Torres 2008). But early implanted CI children are found to catch up with their NH peers when studying their spontaneous speech (Faes et al. 2015, Nicholas & Geers, 2007, Tribushinina et al., 2013).

4. Factors affecting spoken language development

What are the factors affecting spoken language development in severe-to-profound congenitally hearing-impaired children? What are the predictors of successful language acquisition and development after cochlear implantation? In the literature several factors have been identified that appear to affect the eventual success of a cochlear implant intervention in severe-to-profound hearing impaired children (Boons et al. 2012a, Cosetti & Waltzman 2012, Geers 2006). Three sets of factors will be described in this section: (1) factors related to the children's audiological condition, such as their pre-operative hearing, (2) factors related to a child's individual condition, such as the presence of additional disabilities or the child's nonverbal cognitive functioning, and (3) environmental factors, such as the socioeconomic background of the child's family.

Given the important inter-individual variation in the spoken language outcomes of CI device users, a central question in research concerns why particular children perform well while others perform less well? This question pertains to short-term effects (e.g., what makes a baby implanted at 12 months of age babble after one month of exposure to sound and another one only after six months?) as well as long-term effects (e.g., why do particular children communicate at age-appropriate levels much earlier than others, and why do some children with a CI remain language delayed?). Essentially the answers to these questions are similar to the answers that are provided for individual variation in language acquisition and development in typically developing children: characteristics of the language learning child and characteristics of the child's environment determine the path of spoken language development. But in comparison to typically developing children there are additional factors, viz. factors related to their auditory condition, such as the causes of their hearing impairment, the various steps in the intervention, the follow-up history after the intervention.

4.1. Audiology related factors

4.1.1. Age at identification of hearing impairment

Undoubtedly one of the main predictors of successful spoken language acquisition is the age at which a hearing loss is detected and the severity of the hearing deficit is determined. Very early identification is a crucial milestone in rehabilitation since it triggers – under optimal conditions – a number of further steps: once a baby is diagnosed with a hearing deficit, further steps can be taken such as fitting acoustic hearing aids, monitoring the gain in hearing these may bring about, and may be eventually cochlear implantation. But the surgical procedure is only one step in a chain of events that should be instigated if necessary by early detection of a hearing deficit. Newborn hearing screening programs are the ultimate tool in this respect, and hence, the success of language acquisition in hearing-impaired children depends on them (Yoshinaga-Itano 2004, 2006).

4.1.2. Preoperative hearing levels

Most studies agree that the degree of hearing loss established upon identification of a hearing deficit has an important impact on speech perception, speech intelligibility, receptive and productive language development after cochlear implantation. Less hearing loss is a predictor of better outcomes (Artières et al. 2009, Holt & Svirsky 2008, Szagun 2001, but see Nicholas & Geers 2006).

4.1.3. Age at fitting of hearing aids

Early identification of hearing loss has been identified as a factor that contributes to better language and speech outcomes after cochlear implantation. But quite a few studies found that an earlier age of hearing aids fitting – the first step after identification of a hearing loss in the rehabilitation process in most audiological centers -- was also a significant predictor of better outcomes: children whose hearing loss was identified within the first six months and who received hearing aids, showed superior language skills when compared to children who were identified later (Artières et al. 2009) and showed better overall speech intelligibility at later ages (Holt & Svirsky 2008, Nicholas & Geers 2006).

4.1.4. Age at Implantation

The age at implantation is an important predictor of language acquisition and development. Children implanted at an earlier age fare better than children implanted later. This appears as a robust and undisputed finding in the literature on CI children's receptive and expressive language development (i.a. Artieres et al. 2009, Connor et al. 2006, Dettman et al. 2007, Geers et al. 2009, Geers & Nicholas 2013, Holt & Svirsky 2008, Nicholas & Geers 2008, Niparko et al. 2010,).

Several reasons motivate this finding. First of all, during the first year of life children's perceptual abilities develop enormously: they "home in" on the ambient language, adapting their discrimination and categorization of speech sounds to the language they hear, use the suprasegmentals of the language to segment words and utterances from the speech stream (Jusczyk 1997). Moreover, their speech production attains a number of important milestones during the first few years of life, such as the onset of canonical babbling and the production of the first words. These are not only important milestones in speech production and perception, in the early years also the relationship between the auditory and the articulatory world are established, allowing children to connect their speech-motor programs with the sounds they hear (Redford 2015). Given their poor auditory abilities, severe-to-profound hearing impaired children do not have sufficient auditory stimulation to go through all these important developments during the first years of life, so that early implantation is called for.

A second important reason that motivates early implantation is that the absence of sound input during the first few years of life can result in irreversible changes to the auditory cortex. The developing auditory system is maximally plastic at birth, and this plasticity decreases with age. There appears to be a sensitive period for neural development that is crucial for spoken language development. This limited window of opportunity is maximal in the first 3.5 years, decreases dramatically after 7 years and may be completely closed by 12 years (Gilley et al. 2008, Kral & Sharma

2011, Sharma et al. 2004, 2005). Hence, it is of prime importance that this window of opportunity is not missed.

What is the optimal age window for implantation? The benefits of fitting a CI under the age of 2 have been documented extensively (Anderson et al. 2004, Boons et al. 2012a, Hammes et al. 2002, Svirsky et al. 2004). Currently the question whether implantation in the first year of life has any beneficial effects on children's language and speech, and if so, whether these effects are lasting, are under investigation. At present, the evidence is mixed. Some studies report no advantage or just a limited or a non-lasting advantage of implantation during the first year of life (Colletti et al. 2011, Holt & Svirsky 2008, Lesinski-Schiedat et al. 2004, Vlastarakos et al. 2010). The most elaborate study up till now investigated 35 children implanted before 12 months of age and 85 children implanted before 24 months, three years post implantation (Leigh et al. 2013). The results are mixed: on some measures significant differences between the two groups are reported, while on other measures the difference did not reach significance. For instance, on speech production (viz. percentage phonemes correct) there was no significant difference between the two groups. Both perform significantly poorer than normally hearing peers. But on receptive vocabulary (measured with the PPVT), the youngest implanted children score significantly better than the older implanted ones. Implantation in the first year of life leads to age appropriate receptive vocabulary skills, while later implantation leads to a significant lasting delay after three years of device experience.

4.1.5. Duration of device use

In addition to the age at implantation, the length of device use appears to have a crucial impact on children's linguistic functioning. For instance, Geers & Nicholas (2013) studied children who had received a CI between their first and third birthdays. At 4.5 years of age they found a significant effect of length of device use: everything else being equal, longer hearing experience resulted in more advanced language production and comprehension. However, this effect appeared to have faded out by 10.5 years of age.

Whether age at implantation or length of device use are the most decisive factors in predicting language outcomes in children who received a CI is still a matter of debate. Geers & Nicholas (2013) found that children with a more advanced level of language use at 4.5 were also more advanced at 10.5 years, and these were the children who were implanted at the earliest ages. Thus, Geers & Nicholas (2013) report a lasting effect of the age of implantation: the age at which children gain access to spoken language through their first implant still has an effect on their linguistic functioning at 10.5 years of age, with younger implanted children still outperforming later implanted ones.

However, Szagun & Stumper (2012) note that whereas age at implantation has been treated as a major influence on the language development of children with CIs, the amount of variance that it actually explains is small (see also Geers et al. 2009; Tomblin et al. 2005). Moreover, their research indicates that instead of age at implantation, length of device use is a more determining factor: a lasting effect of the length of children's robust auditory experience was detected, while the effect of age at implantation was not significant.

4.1.6. Bilateral (or contralateral) cochlear implant

In the early days of cochlear implantation, children received a single implant: unilateral implantation. Nowadays bilateral implantation – a device in both ears – seems to have become standard practice in many implant centers around the world.

Children with a unilateral implant exhibit excellent speech perception abilities in optimal conditions, such as a quiet room, but their listening abilities become far less accurate under more natural conditions: a noisy background is often reported to hamper interaction, soft or whispered speech are not accurately captured, and locating a speaker in a conversational setting is often difficult. Hence, on the one hand children with a unilateral CI exhibit levels of speech and language development that they would never have obtained with a (acoustic) hearing aid, and many studies in the past have shown that they can reach age appropriate linguistic functioning. But, on the other hand, many children with a unilateral CI have been reported to show delays in language and speech development (Geers 2002, Tobey et al. 2003).

Bilateral implantation has significant positive effects over the unilateral condition on speech perception in quiet and in noise and on sound-source localization (Vincent et al. 2012). The impact on children's receptive and expressive language development is less clear. Some studies report no beneficial effects of bilateral implantation (Niparko et al. 2010, Nittrouer & Chapman 2009). Others found a marked improvement of particular aspects of language comprehension and/or production (Boons et al. 2012a, 2012b, Sarant et al. 2014, Sparreboom et al. 2015). For instance, children with bilateral implants show significantly faster rates of receptive vocabulary development (measured by standardized tests such as the

Peabody Picture Vocabulary Test - PPVT) than children in the unilateral condition (the magnitude of the effect was moderated by the child's age at activation, Boons et al. 2012b, Sarant et al. 2014). This faster vocabulary acquisition may be explained by the fact that because bilateral children's hearing is more robust to noise, their ability to learn incidentally (by overhearing) is superior to that of unilaterally implanted children.

Bilateral implantation brings up the question whether the devices should be implanted *simultaneously* or *sequentially*, and whether this has an effect, and more importantly for the present chapter, which mode of operation results in better spoken language performance. The research results are not equivocal. For instance, Sparrenboom et al. (2015) did not find a significant effect of the duration of first or only implant use on receptive vocabulary in children who received their first implant on average at 1;08 and their second on average at 5;01, with a mean inter-implant delay of 3;4 and tested when they were 10 years of age (average 10;8). But Boons et al. (2012b) report that a shorter interval between both implantations was related to higher standard scores: children who underwent a simultaneous cochlear implantation performed better than children who underwent two sequential cochlear implantations. This issue certainly needs further scrutiny.

4.1.7. Unimodal versus bimodal stimulation

The issue of unimodal versus bimodal stimulation is fairly complicated. On the one hand, in the case of unilateral implantation, the issue is: should auditory stimulation with an acoustic hearing aid in the non-implanted ear, be continued after a cochlear implant, i.e. an implant on one ear and a hearing aid on the other? Does it result in beneficial effects? The answer provided by Nittrouer & Chapman (2009), based on a review of the literature and on their own empirical research, can be briefly summarized as follows: children who had bimodal stimulation at any point in their lives fare better than children who never had bimodal stimulation.

With the introduction of hybrid cochlear implants, bimodal stimulation can also mean bimodal implantation (or combined electric and acoustic stimulation). The impact of this type of device on children's language and speech development is still unclear due to the fact that bimodal implants are a recent innovation. But it may be expected that due to better perception of the low frequencies, children's speech may considerably improve: their vowel production quality may improve and their production of intonation at the word and sentence level is also expected to ameliorate. However, to the best of our knowledge, studies investigating these aspects of speech production contrasting children with a unimodal and a bimodal implant are lacking at present.

4.1.8. Device placement, fitting and audiological rehabilitation

Several factors related to the actual implantation, the consecutive fitting of the device and the audiological rehabilitation program after device switch on, have only been sparsely dealt with in the literature, while at the same time they may have an important impact on later language and speech outcomes (Nicholas & Geers 2006). In a large scale study involving 188 participants implanted in 6 large implant centers situated in different regions of the United States, the RLDS were administered to all children. In reporting the results of the study, implant center was mentioned as one of the predicting variables: "Center was found to be significantly associated with different rates of increase in comprehension scores." (Niparko et al. 2010: 1504) This means that the language outcomes of children implanted in one place differ from children treated in another one. The reasons for this disparity can be manyfold, but are hardly touched upon in the relevant literature. One factor is the device placement and fitting: the electrodes can be completely inserted or only partially, thus, insertion depth is a relevant parameter. In addition, fitting of the device after it was brought in place, amounts to programming the speech processor for optimal (speech) perception. This process results in a unique program, or "map" for each individual cochlear implant user. Even in adults this "cochlear parametrisation is a difficult and long task, with results ranging from perfect blind speech recognition to patients who cannot make anything out of their implant and just turn it off." (Bourgeois-République et al. 2004: 296) It can readily be inferred that in the case of small children fitting is an even more challenging task with highly variable success, a task that largely depends on "the skill of the audiologist" (Nicholas & Geers 2006: 276).

After device switch on, most children enter into an intensive speech and language training program. Since different audiological centers may follow different rehabilitation schemes, also this factor may contribute to the outcomes found in children implanted in different centers.

4.2. Child related factors

4.2.1. Nonverbal cognitive abilities

Nonverbal cognitive ability is a significant predictor of language development in typically developing children as well as in children with an atypical profile, such as SLI, Specific Language Impairment (Botting 2005). Also in the population of severe-to-profound hearing-impaired children with a cochlear implant, nonverbal cognitive abilities are a strong predictor of language (Boons et al. 2012a, b, Geers et al. 2008, Sarant et al. 2014).

4.2.1. Cause of deafness

The cause of congenital hearing impairment can be classified into three broad categories: (1) a genetic cause (e.g., mutation of the connexin 26 gene) represents ca. 40% of the cases, (2) infectious causes, such as cytomegalovirus or other viral infections, represent 30% of the cases, and (3) an unknown cause is concluded in the remaining 30%. In the genetic category, a further distinction can be made between syndromic deafness, such as Usher syndrome, Alport syndrome, Waardenburg syndrome, etc. About 400 syndromes have an associated hearing loss (Krall & O'Donoghue 2010). Syndromic deafness accounts for approximately 30% of the cases, the remaining 70% is nonsyndromic.

The exact impact of the cause of hearing impairment on the speech and language outcomes of cochlear implantation is not well understood. In a number of studies the outcome of an implantation is compared in children with cytomegalovirus (CMV) infection and children with presumed genetically determined deafness (connexin) as the presumed cause of their hearing loss. Ramirez Inscoe & Nikolopoulos (2004) report on 16 children with CMV: in comparison with connexin children, they tended to perform more poorly. However, in a more recent study, Philips et al. (2014) show that when the CMV group is further subdivided into those with a normal MRI scan and those with an abnormal MRI scan, the former perform equally well as the connexin children, or even slightly better, while the latter seem to catch up for speech perception, but not for speech production. More research in this area is definitely required.

4.2.2. Gender

For receptive and productive language measures, boys score lower than girls at 5 years of age, but this effect fades out and was no longer evident when children are tested at 8 years of age (Geers et al. 2009, Sarant et al. 2014). This result is in agreement with other research showing that the gap in language ability between boys and girls in early life closes with increasing age (Ely 2005).

4.2.3. Additional disabilities

Approximately 30 to 50% of the children with severe-to-profound hearing loss suffer from additional disabilities, such as autistic spectrum disorders, behavioral difficulties, cognitive difficulties, oro-facial disorders, visual impairment, vocal tract anomalies, or a combination of these additional difficulties. A direct comparison of 67 CI children with and 104 CI children without additional disabilities revealed that five years after implantation, additional disabilities have a negative impact on the children's overall speech intelligibility (Nikolopoulos et al. 2008). More specifically, 70% of the children with additional disabilities had developed connected speech intelligibility, while this was 96% for the children without additional disabilities. Moreover, only a small portion of the former group was intelligible for unfamiliar adult listeners. This implies that this group of hearing impaired children need special attention after implantation since additional disabilities appear to be negative predictive factors for the eventual outcome (Beer et al. 2012, Gérard et al. 2010).

4.3. Environmentally related factors

4.3.1. Communication modality

Most infants who are deaf or hard of hearing are born into hearing families (an estimated 96%, Mitchel & Karchmer 2004). This poses a problem of communication modality in the family: oral communication is the obvious mode for hearing parents, but with a deaf child the need for signing in combination with oral language (so called total communication) is also an option. The influence of communication modality on speech and language outcomes is difficult to assess (Kirk et al. 2002) but evidence points out that children living in environments that strongly emphasize oral language tend to have better speech and language outcomes (Boons et al. 2012a, Geers et al. 2003, Johnson & Goswami 2010, Kirk et al. 2002, but see McDonald Conner et al. 2000). In a similar vein, children who attend mainstream education (from earlier on) have a better spoken language development than children in special schools (Geers et al. 2003).

4.3.2. Family related factors

A number of family related factors have been shown to play a role in the success of CI children's rehabilitation: socioeconomic status (SES), maternal education level, the involvement of the family in therapy, parenting style, family size. These factors are related to a certain extend, as for instance SES is determined by education level.

Not surprisingly, high levels of parental involvement have a positive impact on children's language development. For instance, joint picture book reading is a well-known source of vocabulary development (Fletcher & Reese 2005, Vernon-Feagans et al. 2008). Sarrant et al. (2014) found a similar effect in children with CI: the time parents spent reading to their children significantly affected their vocabulary and language scores. Interestingly, the amount of time children spent watching a screen had a negative effect on those scores.

The influence of SES is well known from studies of NH children (Hart & Risley 1995): higher SES families tend to provide their children a qualitatively and quantitatively richer linguistic environment. They talk more, provide more vocabulary, more complex language structures, and in so doing present children more and better opportunities to pick up language. Children's language development tends to benefit from this richer input as judged, for instance, by their richer receptive and expressive vocabularies (Hoff 2003, Rowe 2008). A similar facilitative role is played by SES in the case of CI children (Niparko et al. 2010, Sarant et al. 2014).

The precise effect of these family related factors on CI children's linguistic development is not straightforward since there may well be many mediating factors (Frush Holt et al. 2012). One factor that plays a role is maternal language input: how much input does the child receive? And how finely tuned is the input to the language level of the child? Mothers of young CI children from mid-to-high SES provide equal amounts of speech and they appear to be even more responsive contingently upon their children’s utterances in comparison with mid-to-high SES mothers of NH peers (Vanormelingen et al. 2015). In a follow-up study, Vanormelingen and colleagues compared interactions of the same group of mid-to-high SES NH and CI children with the interaction behaviour of mothers of low SES during the first two years of life. They found that mothers in the latter group were not only significantly less talkative, they were also less responsive to their children’s vocal efforts. Even more dramatically: these mothers became even less talkative and less responsive over time. The effects on the children’s vocal development was noticed early in life: their onset of babbling was seriously delayed in almost half of the participating children (Vanormelingen & Gillis in prep). Szagun & Stumper (2012) investigated mother infant dyads with different educational backgrounds (classified according to the number of years of schooling) and established that educational level correlated significantly with measures such as MLU (mean length of utterance) and number of expansions (expanding an incorrect child utterances “that boat” to “yes, that’s a boat”). Higher educational level meant higher MLU and more expansions, and these characteristics of maternal language input implied faster linguistic growth in their implanted children: a richer vocabulary (more word types), more complex language (higher MLU), and higher scores for inflectional morphology 30 months after implantation. Hence, these authors conclude that the CI children’s “home linguistic environment”, or their experience with language, largely determines their progress in language acquisition.

5. Conclusion

Since the introduction of cochlear implants, severe-to-profound hearing-impaired children are given access to auditory information. The device permits them to develop speech and language skills that surpass those of children with comparable hearing deficits equipped with acoustic hearing aids. In this sense cochlear implantation is a successful innovation. Although implanted children start with an initial delay in spoken language, a quite significant group eventually reaches age appropriate levels of linguistic functioning. But the individual variation is also quite significant: while some children do catch up with their normally hearing peers, others do not achieve much language comprehension and production even after five years of device use (Barnard et al. 2015).

At present the individual variation in linguistic outcomes of cochlear implant recipients remains poorly explained. In the literature various factors have been proposed as determinants of the success of the intervention, but there are many factors that have been identified as predictors, and it proves to be very difficult to control them all in a single study. Moreover, cochlear implant users are a constantly “moving target”, which complicates the identification of factors to which successful language development can be attributed and that account for individual variation in the outcomes.

Finally not until very recently the underlying consequences of early auditory deprivation and the consequences of “electrical hearing” for speech perception and production and language comprehension and production are under scrutiny (Houston et al. 2012). Much more research is needed to substantiate recent claims that children with CI pay less attention to speech than their normally hearing peers, have significantly reduced working memory capacity, and hence seem less well equipped for acquiring new words and developing grammar. Consequently our understanding of the neurocognitive underpinnings of these psycholinguistic processes are even less well developed and are in need of further investigation.

References

Anderson, I., Weichbold, V., D'Haese, P., Szuchnik, J., Quevedo, M., Martin, J., . . . Phillips, L. (2004). Cochlear implantation in children under the age of two - what do the outcomes show us? *International Journal of Pediatric Otorhinolaryngology, 68*, 425-431.

Artières, F., Vieu, A., Mondain, M., Uziel, A., & Venail, F. (2009). Impact of early cochlear implantation on the linguistic development of the deaf child. *Otology & Neurotology, 30*(6), 736-742.

Barnard, J., Fisher, L., Johnson, K., Eisenberg, L., Wang, N.-Y., Quittner, A., . . . Niparko, J. (2015). A prospective longitudinal study of U.S. children unable to achieve open-set speech recognition 5 years after cochlear implantation. *Otology & Neurotology*.

Baudonck, N., Van Lierde, K., Dhooge, I., & Corthals, P. (2011). A comparison of vowel productions in prelingually deaf children using cochlear implants, severe hearing-impaired children using conventional hearing aids and normal-hearing children. *Folia Phoniatrica et Logopaedica, 63*, 154-160.

Beer, J., Harris, M., Kronenberger, W., Frush Holt, R., & Pisoni, D. (2012). Auditory skills, language development, and adaptive behavior of children with cochlear implants and additional disabilities. *International Journal of Audiology, 51*, 491-498.

Blamey, P., Sarant, J., Paatsch, L., Barry, J., Bow, C., Wales, R., . . . Tooher, R. (2001). Relationships among speech perception, production, language, hearing loss, and age in children with impaired hearing. *Journal of Speech, Language, and Hearing Research, 44*, 264-285.

Boons, T., Brokx, J., Dhooge, I., Frijns, J., Peeraer, L., Vermeulen, A., . . . van Wieringen, A. (2012a). Predictors of spoken language development following pediatric cochlear implantation. *Ear & Hearing, 33*(5), 627-639.

Boons, T., Brokx, J., Frijns, J., Peeraer, L., Philips, B., Vermeulen, A., . . . van Wieringen, A. (2012b). Effect of pediatric bilateral cochlear implantation on language development. *Archives of Pediatric and Adolescence Medicine, 166*(1), 28-34.

Botting, N. (2005). Non-verbal cognitive development and language impairment. *Journal of Child Psychology and Psychiatry, 46*(3), 317-326.

Bourgeois-République, C., Chabrier, J.-J., & Collet, P. (2004). Automatic fitting of cochlear implants with evolutionary algorithms *Proceedings of the 2004 ACM symposium on Applied computing* (pp. 296-300).

Campbell, A., Dillon, M., Buchman, C., & Adunka, O. (2013). Hearing preservation cochlear implantation. *Current Otorhinolaryngology Reports, 1*, 69-79.

Caselli, M., Rinaldi, P., Varuzza, C., Giuliani, A., & Burdo, S. (2012). Cochlear implant in the second year of life: Lexical and grammatical outcomes. *Journal of Speech, Language, and Hearing Research, 55*, 382-394.

Clark, G. (1989). The bionic ear and beyond. *Journal of the Otolaryngological Society of Australia, 6*(4), 244-249.

Clark, G., Tong, Y., & Dowell, R. (1983). Clinical results with a multichannel pseudobipolar system. *Annals of the New York Academy of Science, 404*, 370-

377.
Colletti, L., Mandalà, M., Zoccante, L., Shannon, R., & Colletti, V. (2011). Infants versus older children fitted with cochlear implants: Performance over 10 years. *International Journal of Pediatric Otorhinolaryngology, 75*, 504-509.
Colletti, V., Carner, M., Miorelli, V., Guida, M., Colletti, L., & Fiorino, F. (2005). Cochlear implantation at under 12 months: Report on 10 patients. *The Laryngoscope, 115*, 445-449.
Connor, C., Craig, H., Raudenbush, S., Heavner, K., & Zwolan, T. (2006). The age at which young deaf children receive cochlear implants and their vocabulary and speech-production growth: Is there an added value for early implantation? *Ear & Hearing, 27*, 628-644.
Cosetti, M., & Waltzman, S. (2012). Outcomes in cochlear implantation: Variables affecting performance in adults and children. *Otolaryngology Clinics North America, 45*(1), 155-171.
De Raeve, L. (2006). Making the case for early hearing detection and intervention in Flanders, Belgium. *Volta Voices*(14-17).
De Raeve, L. (2014). *Paediatric cochlear implantation: Outcomes and current trends in education and rehabilitation.* (PhD), Radboud Universiteit Nijmegen, Nijmegen.
De Raeve, L., & Lichtert, G. (2012). Changing trends within the population of children who are deafor hard of hearing in Flanders (Belgium): Effects of 12 years of universal newborn hearing screening, early intervention, and early cochlear implantation. *The Volta Review, 112*(2), 131-148.
De Raeve, L., & Wouters, A. (2013). Accessibility to cochlear implants in Belgium: State of the art on selection, reimbursement, habilitation, and outcomes in children and adults. *Cochlear Implants International, 14*(S1), 18-25.
Desloovere, C., Verhaert, N., Van Kerschaver, E., & Debruyne, F. (2013). Fifteen years of early hearing screening in Flanders: Impact on language and education. *B-ENT, 9*, 81-90.
Dettman, S., Pinder, D., Bruggs, R., Dowell, R., & Leigh, J. (2007). Communication development in children who receive the cochlear implant younger than 12 months: Risks versus benefits. *Ear & Hearing, 28*, 11S-18S.
Dowell, R., Brown, A., Seligman, P., & Clark, G. (1985). Patient results for a multiple-channel cochlear prosthesis. In R. Schindler & M. Merzenich (Eds.), *Cochlear implants (10th Anniversary Conference on Cochlear Implants)* (pp. 421-431). New York: Raven Press.
Duchesne, L., Sutton, A., & Bergeron, F. (2009). Language achievement in children who received cochlear implants between 1 and 2 years of age: Group trends and individual patterns. *Journal of Deaf Studies and Deaf Education, 14*(4), 465-485.
Eisenberg, L., & House, W. (1982). Initial experience with the cochlear implant in children. *Annals of Otology, Rhinology & Laryngology, 91*(2), S67-S73.
Ely, R. (2005). Language and literacy in the school years. In J. Berko Gleason (Ed.), *The development of language* (pp. 395-443). Boston: Allyn & Bacon.
Eriks-Brophy, A., Gibson, S., & Tucker, S. (2013). Articulatory error patterns and phonological process use of preschool children with and without hearing loss. *The Volta Review, 113*(2), 87-125.
Ertmer, D., & Goffman, L. (2011). Speech production accuracy and variability in young

cochlear implant recipients: Comparisons with typically developing age-peers. *Journal of Speech, Language and Hearing Research, 54*(1), 177-189.
Ertmer, D., Kloiber, D., Jung, J., Connell Kirleis, K., & Bradford, D. (2012). Consonant production accuracy in young cochlear implant recipients: Developmental sound classes and word position effects. *American Journal of Speech-Language Pathology, 21*, 342-353.
Ertmer, D., & Mellon, J. (2001). Beginning to talk at 20 months: Early vocal development in a young cochlear implant recipient. *Journal of Speech, Language, and Hearing Research, 44*, 192-206.
Faes, J., Gillis, J., & Gillis, S. (2015). Syntagmatic and paradigmatic development of cochlear implanted children in comparison with normally hearing peers up to age 7. *International Journal of Pediatric Otorhinolaryngology, 79*, 1533-1540.
Faes, J., Gillis, J., & Gillis, S. (In press). Phonemic accuracy development in children with cochlear implants up to five years of age by using Levenshtein Distance. *Journal of Communication Disorders*.
Fink, N., Wang, N.-Y., Visaya, J., Niparko, J., Quittner, A., Eisenberg, L., & Tobey, E. (2007). Childhood development after cochlear implantation (CDaCI) study: Design and baseline characteristics. *Cochlear Implants International, 8*(2), 92-116.
Fletcher, K., & Reese, E. (2005). Picture book reading with young children: A conceptual framework. *Developmental Review, 25*, 64-103.
Frush Holt, R., Beer, J., Kronenberger, W., Pisoni, D., & Lalonde, K. (2012). Contribution of family environment to pediatric cochlear implant users' speech and language outcomes: Some preliminary findings. *Journal of Speech, Language, and Hearing Research, 55*, 848-864.
Geers, A. (2002). Factors affecting the development of speech, language, and literacy in children with early cochlear implantation. *Language, Speech, and Hearing Services in Schools, 33*, 172-183.
Geers, A. (2006). Spoken language in children with cochlear implants. In P. Spencer & M. Marschark (Eds.), *Advances in spoken language development of deaf and hard-of-hearing children* (pp. 244-270). Oxford: Oxford UP.
Geers, A., Brenner, C., & Davidson, L. (2003). Factors associated with development of speech perception skills in children implanted by age five. *Ear & Hearing, 24*(18), 24S-35S.
Geers, A., Moog, J., Biedenstein, C., Brenner, C., & Hayes, H. (2009). Spoken language scores of children using cochlear implants compared to hearing age-mates at school entry. *Journal of Deaf Studies and Deaf Education, 14*(3), 371-385.
Geers, A., & Nicholas, J. (2013). Enduring advantages of early cochiear implantation for spoken language development. *Journal of Speech, Language, and Hearing Research, 56*, 643-653.
Geers, A., Nicholas, J., & Sedey, A. (2003). Language skills of children with early cochlear implantation. *Ear & Hearing, 24*(1S), 46S-58S.
Geers, A., Tobey, E., Moog, J., & Brenner, C. (2008). Long-term outcomes of cochlear implantation in the preschool years: From elementary grades to high school. *International Journal of Adiology, 47 (Suppl. 2)*, S21-S30.
Gérard, J.-M., Deggouj, N., Hupin, C., Buisson, A.-L., Monteyne, V., Lavis, C., . . . Gersdorff, M. (2010). Evolution of communication abilities after cochlear

implantation in prelingually deaf children. *International Journal of Pediatric Otorhinolaryngology, 74*, 642-648.

Gilley, P., Sharma, A., & Dorman, M. (2008). Cortical reorganization in children with cochlear implants. *Brain Research, 1239*, 56-65.

Guo, L., Spencer, L., & Tomblin, B. (2013). Acquisition of tense marking in English-speaking children with cochlear implants: A longitudinal study. *Journal of Deaf Studies and Deaf Education, 18*(2), 187-205.

Hammer, A. (2010). *The acquisition of verbal morphology in cochlear-implanted and specific language impaired children.* (PhD), Universiteit Leiden, Leiden.

Hammes, D., Novak, M., Rotz, L., Willis, M., Edmondson, D., & Thomas, J. (2002). Early identification and cochlear implantation: Critical factors for spoken language development. *Annals of Otology, Rhinology & Laryngology, 111*, 74-78.

Hart, B., & Risley, T. (1995). *Meaningful differences in the everyday experience of young American children*. Baltimore: Brookes.

Hoff, E. (2003). The specificity of environmental influence: Socioeconomic status affects early vocabulary development via maternal speech. *Child Development, 74*, 1368-1378.

Hoffman, M., Quittner, A., & Cejas, I. (2014). Comparisons of social competence in young children with and without hearing loss: A dynamic systems framework. *Journal of Deaf Studies and Deaf Education*.

Holt, R., & Svirsky, M. (2008). An exploratory look at pediatric cochlear implantation: Is earliest always best? *Ear & Hearing, 29*(4), 492-511.

Houston, D., Beer, J., Bergeson, T., Chin, S., Pisoni, D., & Miyamoto, R. (2012). The ear is connected to the brain: Some new directions in the study of children with cochlear implants at Indiana University. *Journal of the American Academy of Audiology, 23*, 446-463.

James, C., Fraysse, B., Deguine, O., Lenarz, T., Mawman, D., Ramos, A., . . . Sterkers, O. (2006). Combined electroacoustic stimulation in conventional candidates for cochlear implantation. *Audiology & Neurotology, 11*(S1), 57-62.

Johnson, C., & Goswami, U. (2010). Phonological awareness, vocabulry, and reading in deaf children with cochlear implants. *Journal of Speech, Language, and Hearing Research, 53*, 237-261.

Jusczyk, P. (1997). *The discovery of spoken language*. Cambridge: MIT Press.

Kirk, K., Miyamoto, R., Lento, C., Ying, E., O'Neill, T., & Fears, B. (2002). Effects of age at implantation in young children. *Annals of Otology, Rhinology & Laryngology, 111*(69-73).

Kochkin, S. (2010). The efficacy of hearing aids in achieving compensation equity in the workplace. *The Hearing Journal, 63*(10), 19-28.

Koopmans-van Beinum, F., Clement, C., & van den Dikkenberg-Pot, I. (2001). Babbling and the lack of auditory speech perception: A matter of coordination? *Developmental Science, 4:1*, 61-70.

Kral, A., & O'Donoghue, G. (2010). Profound deafness in childhood. *The New England Journal of Medicine, 363*(15), 1438-1450.

Kral, A., & Sharma, A. (2011). Developmental neuroplasticity after cochlear implantation. *Trends in Neurosciences, 35*, 111-122.

Laaha, S., Blineder, M., & Gillis, S. (2015). Noun plural production in preschoolers with early cochlear implantation: An experimental study of Dutch and German. *International Journal of Pediatric Otorhinolaryngology, 79*, 561-569.

Leigh, J., Dettman, S., Dowell, R., & Briggs, R. (2013). Communication development in children who receive a cochlear implant by 12 months of age. *Otology & Neurotology, 34*, 443-450.

Lesinski, -. S., A., Illg, A., Heermann, R., Bertram, B., & Lenarz, M. (2004). Pediatric cochlear implantation in the first and the second year of life: A comparative study. *Cochlear Implant International, 5*(4), 146-159.

Liker, M., Mildner, V., & Sindija, B. (2007). Acoustic analysis of the speech of children with cochlear implants: A longitudinal study. *Clinical Linguistics & Phonetics, 21*(1), 1-11.

Mahdieh, N., Rabbani, B., & Inoue, I. (2012). Genetics of hearing loss. In S. Naz (Ed.), *Hearing loss* (pp. 211-246). Rijeka: InTech Europe.

McDonald Connor, C., Hiebr, S., Arts, H. A., & Zwolan, T. (2000). Speech, vocabulary, and the education of children using cochlear implants: Oral or total communication? *Journal of Speech, Language, and Hearing Research, 43*, 1185-1204.

Mitchell, R., & Karchmer, M. (2004). Chasing the mythical ten percent: Parental hearing status of deaf and hard of hearing students in the United States. *Sign Language Studies, 4*, 138-163.

Moeller, M., Hoover, B., Putman, C., Arbataitis, K., Bohnenkamp, G., Peterson, B., . . . Stelmachowicz, P. (2007). Vocalizations of infants with hearing loss compared with infants with normal hearing: Part I – Phonetic development. *Ear & Hearing, 28*, 605-627.

Molemans, I. (2011). *Sounds like babbling. A longitudinal investigation of aspects of the prelexical speech repertoire in young children acquiring Dutch: Normally hearing and hearing-impaired children with a cochlear implant.* (PhD), University of Antwerp, Antwerp.

Molemans, I., van den Berg, R., Van Severen, L., & Gillis, S. (2012). How to measure the onset of babbling reliably? *Journal of Child Language, 39*, 523-552.

Möller, A. (2006). History of cochlear implants and auditory brainstem implants. *Advances in Otorhinolaryngology, 64*, 1-10.

Montag, J., AuBuchon, A., Pisoni, D., & Kronenberger, W. (2014). Speech intelligibility in deaf children after long-term cochlear implant use. *Journal of Speech, Language, and Hearing Research, 57*, 2332-2343.

Moore, J., & Bass-Ringdahl, S. (2002). Role of infant vocal development in candidacy for and efficacy of cochlear implantation. *Annals of Otology, Rhinology & Laryngology, 111*, 52-55.

Moreno-Torres, I., & Torres, S. (2008). From 1-word to 2-words with cochlear implant and cued speech: A case study. *Clinical Linguistics & Phonetics, 22*, 491-508.

Nathani, S., Oller, D., & Neal, A. (2007). On the robustness of vocal development: An examination of infants with moderate-to-severe hearing loss and additional risk factors. *Journal of Speech, Language, and Hearing Research, 50*, 1425-1444.

Neumeyer, V., Harrington, J., & Draxler, C. (2010). An acoustic analysis of the vowel space in young and old cochlear-implant speakers. *Clinical Linguistics & Phonetics, 24*(9), 734-741.

Nicholas, J., & Geers, A. (2006). Effects of early auditory experience on the spoken language of deaf children at 3 years of age. *Ear & Hearing, 27*, 286-298.
Nicholas, J., & Geers, A. (2007). Will they catch up? The role of age at cochlear implantation in the spoken language development of children with severe to profound hearing loss. *Journal of Speech, Language, and Hearing Research, 50*, 1048-1062.
Nicholas, J., & Geers, A. (2008). Expected test scores for preschoolers with a cochlear implant who use spoken language. *American Journal of Speech-Language Pathology, 17*, 121-138.
Nicolaidis, K., & Sfakiannaki, A. (2007). An acoustic analysis of vowels produced by Greek speakers with hearing impairment *Proceedings of 16th International Congress of Phonetic Sciences* (pp. 1969-1972). Saarbrücken.
Nikolopoulos, T. (2015). Neonatal hearing screening: What we have achieved and what needs to be improved. *International Journal of Pediatric Otorhinolaryngology, 79*(5), 635-637.
Nikolopoulos, T., Archbold, S., Wever, C., & Lloyd, H. (2008). Speech production in deaf implanted children with additional disabilities and comparison with age-equivalent implanted children without such disorders. *International Journal of Pediatric Otorhinolaryngology, 72*, 1823-1828.
Niparko, J., Tobey, E., Thal, D., Eisenberg, L., Wang, N.-Y., Quittner, A., & Fink, N. (2010). Spoken language development in children following cochlear implantation. *JAMA - Journal of the American Medical Association, 303*(15), 1498-1506.
Nittrouer, S., & Chapman, C. (2009). The effects of bilateral electric and bimodal electric–acoustic stimulation on language development. *Trends in Amplification, 13*(3), 190-205.
O'Donoghue, G. (2013). Cochlear implants — science, serendipity, and success. *New England Journal of Medicine, 369*(13), 1190-1193.
Oller, D., & Eilers, R. (1988). The role of audition in infant babbling. *Child Development, 59*(22), 441-449.
Oller, K. (2000). *The emergence of the speech capacity*. Mahwah: Lawrence Erlbaum.
Peng, S., Spencer, L., & Tomblin, J. (2004). Speech intelligibility of pediatric cochlear implant recipients with 7 years of device experience. *Journal of Speech, Language, and Hearing Research, 47*, 1227-1236.
Philips, B., Corthals, P., De Raeve, L., D'haenens, W., Maes, L., Bockstael, A., . . . Dhooge, I. (2009). Impact of newborn hearing screening: Comparing outcomes in pediatric cochlear implant users. *The Laryngoscope, 119*, 974-979.
Philips, B., Maes, L., Keppler, H., & Dhooge, I. (2014). Cochlear implants in children deafened by congenital cytomegalovirus and matched Connexin 26 peers. *International Journal of Pediatric Otorhinolaryngology, 78*(3), 410-415.
Quittner, A., Leibach, P., & Marciel, K. (2004). The impact of cochlear implants on young deaf children. *Archives of Otolaryngology - Head & Neck Surgery, 130*, 547-554.
Ramirez Inscoe, J., & Nikolopoulos, T. (2004). Cochlear implantation in children deafened by cytomegalovirus: Speech perception and speech intelligibility outcomes. *Otology & Neurotology, 25*(4), 479-482.
Redford, M. (2015). The acquisition of temporal patterns. In M. Redford (Ed.), *The*

handbooook of speech production (pp. 379-403). Hoboken, NJ: John Wiley & Sons.

Rowe, M. (2008). Child-directed speech: Relation to socioeconomic status, knowledge of child development and child vocabulary skill. *Journal of Child Language, 35*, 185-205.

Ruffin, C., Kronenberger, W., Colson, B., Henning, S., & Pisoni, D. (2013). Long-term speech and language outcomes in prelingually deaf children, adolescents and young adults who received cochlear implants in childhood. *Audiology and Neurotology, 18*, 289-296.

Sarant, J. (2012). Cochlear implants in children: A review. In S. Naz (Ed.), *Hearing loss* (pp. 331-382). Rijeka: InTech.

Sarant, J., Harris, D., Bennet, L., & Bant, S. (2014). Bilateral versus unilateral cochlear implants in children: A study of spoken language outcomes. *Ear & Hearing, 35*(4), 396-409.

Schauwers, K., Gillis, S., Daemers, K., De Beukelaer, C., & Govaerts, P. (2004). The onset of babbling and the audiological outcome in cochlear implantation between 5 and 20 months of age. *Otology & Neurotology, 25*, 263-270.

Schauwers, K., Gillis, S., & Govaerts, P. (2008). The characteristics of prelexical babbling after cochlear implantation between 5 and 20 months of age. *Ear and Hearing, 29*, 627-637.

Schorr, E., Roth, F., & Fox, N. (2008). A comparison of the speech and language skills of children with cochlear implants and children with normal hearing. *Communications Disorders Quarterly, 29*, 195-210.

Schramm, B., Bohnert, A., & Keilmann, A. (2009). The prelexical development in children implanted by 16 months compared with normal hearing children. *International Journal of Pediatric Otorhinolaryngology, 73*, 1673-1681.

Sharma, A., Dorman, M., & Kral, A. (2005). The influence of a sensitive period on central auditory development in children with unilateral and bilateral cochlear implants. *Hearing Research, 203*, 134-143.

Sharma, A., Tobey, E., Dorman, M., Bharadwaj, S., Martin, K., Gilley, P., & Kunkel, F. (2004). Central auditory maturation and babbling development in infants with cochlear implants. *Archives of Otolaryngology - Head and Neck Surgery, 130*, 511-516.

Sparreboom, M., Langereis, M., Snik, A., & Mylanus, E. (2015). Long-term outcomes on spatial hearing, speech recognition and receptive vocabulary after sequential bilateral cochlear implantation in children. *Research in Developmental Disabilities, 36*, 328-337.

Spencer, P., Marschark, M., & Spencer, L. (2011). Cochlear implants: Advances, issues, and implications. In M. Marschark & P. Spencer (Eds.), *The Oxford handbook of deaf studies, language, and education* (pp. 452-470). New York: Oxford University Press.

Svirsky, M., Robbins, A., Kirk, K., Pisoni, D., & Miyamoto, R. (2000). Language development in profoundly deaf children with cochlear implants. *Psychological Science, 11*, 153-158.

Svirsky, M., Teoh, S., & Neuburger, H. (2004). Development of language and speech perception in congenitally, profoundly deaf children as a function of age at cochlear implantation. *Audiology and Neuro-otology, 9*, 224-233.

Szagun, G. (2001). Language acquisition in young German-speaking children with cochlear implants: Individual differences and implications for conceptions of a 'sensitive phase'. *Audiology and Neuro-otology, 6*, 288-297.
Szagun, G., & Stumper, B. (2012). Age or experience? The influence of age at implantation and social and linguistic environment on language development in children with cochlear implants. *Journal of Speech, Language, and Hearing Research, 55*, 1640-1654.
Tobey, E., Geers, A., Brenner, C., Altuna, D., & Gabbert, G. (2003). Factors associated with development of speech production skills in children implanted by age five. *Ear & Hearing, 24*, 36S-45S.
Tomblin, B., Barker, B., Spencer, L., Zhang, X., & Gantz, B. (2005). The effect of age at cochlear implant initial stimulation on expressive language growth in infants and toddlers. *Journal of Speech, Language, and Hearing Research, 48*, 853-867.
Tomblin, B., Peng, S.-C., Spencer, L., & Lu, N. (2008). Long-term trajectories of the development of speech sound production in pediatric cochlear implant recipients. *Ear & Hearing, 51*, 1353-1368.
Tribushinina, E., Gillis, S., & De Maeyer, S. (2013). Infrequent word classes in the speech of two- to seven-year-old children with cochlear implants and their normally hearing peers: A longitudinal study of adjective use. *International Journal of Pediatric Otorhinolaryngology, 77*, 356-361.
Van Kerschaver, E., Boudewijns, A., Stappaerts, L., Wuyts, F., & Van den Heyning, P. (2007). Organisation of a universal newborn hearing screening programme in Flanders. *B-ENT, 3*, 185-190.
Vanormelingen, L., De Maeyer, S., & Gillis, S. (2015). Interaction patterns of mothers of children with different degrees of hearing: Normally hearing children and congenitally hearing-impaired children with a cochlear implant. *International Journal of Pediatric Otorhinolaryngology, 79*, 520-526.
Vanormelingen, L., De Maeyer, S., & Gillis, S. (in press). A comparison of maternal and child language in normally hearing and children with cochlear implants. *Language, Interaction and Acquisition*.
Vanormelingen, L., & Gillis, S. (in preparation). Babbling onset and consonant characteristics in babbles of children with different SES.
Verhoeven, J., Hide, O., De Maeyer, S., Gillis, S., & Gillis, S. (in press). Hearing impairment and vowel production. A comparison between typically developing, hearing-aided and cochlear implanted Dutch children. *Journal of Communication Disorders*.
Vernon-Feagans, L., Pancsofar, N., Willoughby, M., Odom, M., Quade, E., & Cox, M. (2008). Predictors of maternal language to infants during a picture book task in the home: Family SES, child characteristics and the parentig environment. *Journal of Applied Developmental Psychology, 29*, 213-226.
Vincent, C., Bebear, J., Radafy, E., Vaneecloo, F., Ruzza, I., Lautissier, S., & Bordure, P. (2012). Bilateral cochlear implantation in children: Localization and hearing in noise benefits. *International Journal of Pediatric Otorhinolaryngology, 76*, 858-864.
Vlastarakos, P., Proikas, K., Papacharalampous, G., Exadaktylou, I., Mochloulis, G., & Nikolopoulos, T. (2010). Cochlear implantation under the first year of age — The outcomes. A critical systematic review and meta-analysis. *International*

Journal of Pediatric Otorhinolaryngology, 74, 119-126.
Vos, B., Lagasse, R., & Leveque, A. (2014). Main outcomes of a newborn hearing screening program in Belgium over six years. *International Journal of Pediatric Otorhinolaryngology, 78*(9), 1496-1502.
Wright, M., Purcell, A., & Reed, V. (2002). Cochlear implants and infants: Expectations and outcomes. *Annals of Otology, Rhinology & Laryngology, 111*, 131-137.
Yoshinaga-Itano, C. (2004). Levels of evidence: Universal newborn hearing screening (UNHS) and early hearing detection and intervention systems (EHDI). *Journal of Communication Disorders, 37*, 451-465.
Yoshinaga-Itano, C. (2006). Early identification, communication modality, and the development of speech and spoken language skills: Patterns and considerations. In P. Spencer & M. Marschark (Eds.), *Advances in spoken language development of deaf and hard-of-hearing children* (pp. 298-327). Oxford: Oxford UP.
Yoshinaga-Itano, C., Sedey, A., Coulter, D., & Mehl, A. (1998). Language of early- and later-identified children with hearing loss. *Pediatrics, 102*, 1161-1171.
Young, G., & Killen, D. (2002). Receptive and expressive language skills of children with five years of experience using a cochlear implant. *Annals of Otology, Rhinology & Laryngology, 111*, 802-811.