

The link between hearing loss, language, and social functioning in childhood

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CHAPTER 7

GENERAL DISCUSSION

The aim of this thesis was to study the link between hearing loss, language skills, and social functioning in deaf and hard of hearing (DHH) children. A real challenge when conducting research in DHH children is related to the heterogeneity of the study population. Factors such as the degree of hearing loss, the preferred mode of communication, and the presence of other comorbidities can have significant influence on the development of children with hearing loss. Therefore, the studies included in this thesis were conducted in various populations of DHH children, in order to increase the generalizability of our outcomes. To illustrate, studies were conducted based on DHH children within specific age groups (e.g., preschoolers, schoolchildren), with various degrees of hearing loss (e.g., moderate hearing loss, profound hearing loss wearing cochlear implants (CI)), at different age of detection (i.e., early detection via the Newborn Hearing Screening (NHS), relatively late detection via the Distraction Hearing Screening (DHS)), and also children with additional handicaps were included in two of the studies (i.e., **chapter 3 and 5**).

The main outcomes per research objective will be summarized in the first paragraph. These outcomes will be discussed in the second paragraph to draw a general overview of the relation between hearing loss, language skills and social functioning in DHH children. The third paragraph will contain limitations of the studies conducted in this thesis and the last paragraph concludes with directions for future research.

MAIN OUTCOMES

OBJECTIVE 1. TO ILLUSTRATE THE EFFECT OF VARIOUS METHODS TO HANDLE MISSING DATA ON OUTCOMES IN CLINICAL RESEARCH.

A structured review was performed to illustrate the consequences that missing data can potentially have on outcomes in clinical research. The aim of this study was to create awareness among the importance of reporting missing data and to provide solutions for handling this issue. As can be seen in **chapter 2**, reporting and handling missing data is still quite unpopular in otolaryngology research. However, stimulating researchers to report missing data naturally forces them to think about solutions to handle missing data in their analyses. When confronted with missing data in this thesis, the multiple imputation technique was used to ensure optimal use of all available information from participants (**chapter 3**, **4**, and **5**).

OBJECTIVE 2. TO STUDY THE RELATIONSHIP BETWEEN LANGUAGE, COMMUNICATION AND SOCIAL-EMOTIONAL DEVELOPMENT IN EARLY IDENTIFIED DHH CHILDREN.

The relation between language skills, communicative abilities, and social functioning in DHH toddlers was closely looked at in the study described in **chapter 3**. Language skills were closely related to communicative skills. These communicative skills in turn, were

GENERAL DISCUSSION

positively related to social functioning and negatively related to behavioral problems. Nevertheless, there was no direct relation between language skills and the two socialemotional indices. We were also unable to find a link between the age at detection of hearing loss and the level of social functioning in this group of DHH preschoolers.

OBJECTIVE 3. TO STUDY THE CAUSAL RELATION BETWEEN LANGUAGE AND PSYCHOSOCIAL DEVELOPMENT IN YOUNG DHH CHILDREN WITH CI COMPARED TO HEARING CONTROLS THROUGH LONGITUDINAL ANALYSES.

The longitudinal study described in **chapter 4** showed comparable levels of psychopathology in both children with CI and hearing peers. Second, it confirmed the findings of previous research by showing that early cochlear implantation resulted in higher language and communication skills in children with profound hearing loss.¹⁻³ In turn, this study uniquely added insight into the beneficial consequences of improving communication skills in these young children. Better communication skills prevented the development of symptoms of disruptive behavior such as aggression and hyperactivity over time. In addition, increasing the communication skills decreased the level of depression and anxiety in young children.

OBJECTIVE 4: TO STUDY THE DEVELOPMENTAL PATTERN OF TOM IN YOUNG CHILDREN WITH MODERATE HEARING LOSS COMPARED TO HEARING CONTROLS.

The understanding of others' intentions, desires, and beliefs was the focus of the study described in **chapter 5**. The unique aspect of this study lay in the fact that we only analyzed children with MHL (35-70 dB) and compared them to hearing peers. This specific group of children was chosen because their capacities are often overestimated and the consequences of their moderate hearing loss have rarely been studied. Through observations of both children with MHL and hearing peers we were able to show that both groups were equally able to understand the other's intentions. However, more advanced ToM tasks such as the acknowledgement of others' desires and beliefs were more difficult for children with MHL. These latter two tasks put a higher demand on the child's language skills and were therefore thought to be more challenging for children with MHL. However, we found that children with MHL had language skills within the normal range. Yet, when parents rated their child's communication skills, these were lower than those of hearing peers. ToM skills are learned both through observing others, and during conversations with others. This emphasizes the need for properly developed communication skills, in order to stimulate such opportunities for incidental learning in children with MHL.

OBJECTIVE 5: TO EXAMINE EMPATHIC SKILLS IN DHH (PRE) ADOLESCENTS COMPARED TO HEARING PEERS.

The consequences of diminished chances for incidental learning are also shown in the study described in **chapter 6**. Through incidental learning we learn to reflect on someone else's emotions, in order to strengthen our relationship with the other. These so-called empathic skills were studied by means of self-reports, observations, and parental input. Wiefferink et al. previously showed that young children with a CI encounter problems with recognizing facial expressions.⁴ In line with this finding, the DHH children described in **chapter 6** showed to have more difficulty with the understanding of others emotions. They were also less able to behave prosocial when others were in need. This study also aimed to identify the role of the environment on the empathic abilities of DHH children and showed that children in special education had more difficulty with understanding the other's emotions than DHH peers in mainstream education. Yet, there was no difference between their tendencies to behave prosocial.

GENERAL DISCUSSION

Through studying various aspects of social-emotional functioning in a great variety of DHH children we were able to link several factors that continuously seem to influence social development. The outcomes of the studies performed in this thesis will therefore be combined and discussed in the light of these factors.

Language abilities

In line with many studies in the past, the DHH children examined in this thesis often encountered language problems,⁵⁻⁷ although there was one exception. As recently reported by Tomblin and colleagues, children with moderate hearing loss (described in **chapter 5**) were shown to have language scores within the normal range.⁸ Yet, these scores can only be achieved if children are amplified with hearing aids (HAs) as early in life as possible and if they wear their HAs regularly. These essential prerequisites for developing appropriate language skills stress the thin line that this group of children is walking on.

Surprisingly, language skills were sparsely related to the social-emotional development of DHH children. No relationship was found between language test scores and early signs of psychopathology such as signs of anxiety, depression or behavioral problems. Neither did we find a relation between expressive and receptive language scores and the child's level of social functioning. The absence of a clear relation between language skills and social functioning was endorsed by a comparable study that examined the relation between language skills and social inclusion in DHH children.⁹ An explanation for not finding this link most likely lies in the way DHH children learn vocabulary, which is most of the time by professionals, and in school. This is in contrast with how we learn about social rules and emotions, which is indirect, by observing others, and outside of school. As explained in the introduction of this thesis, DHH children have more difficulty with incidental learning, which explains the lower levels of social functioning and more behavioral problems that were found in **chapter 4**.

However, the studies described in **chapter 5** and **6** focusing on the child's ability to acknowledge the other's thoughts or feelings (i.e., ToM skills and empathy) did show a positive relation with language skills. Both self-reported as well as parent-reported empathic skills and also observations of the child's abilities to acknowledge others' desires and beliefs were better in children with higher language scores. Only the presence of high quality language skills thus likely increased chances for participation in social situations. It seems that scores need to be above a certain threshold for DHH children to be able to join conversations. This is illustrated by the fact that language scores of the DHH children in **chapter 5** and **6** were within the normal range. However, this is not the case in most DHH children. This thesis shows that the relation between language skills and social-emotional functioning is mostly indirect, and that it is even more important that a child is able to use its language capacities in the right way. We therefore have to focus on the communication skills of DHH children.

Communication skills

Parents provide the largest amount of interaction time with their child during the first years of life.^{10,11} Therefore, parents were asked to rate their child's communication skills in all studies concerning young DHH children (chapters 3, 4, and 5). This included complexity and length of produced sentences but also the understanding of languagerelated concepts with increasing complexity. The communication skills of the DHH toddlers in the different studies conducted in this thesis were proportionally lower than their language skills. To illustrate, of all children in the DECIBEL-study analyzed in chapter 3, 43% had expressive language scores that were more than 1 standard deviation (SD) below the mean whereas 55% had communicative abilities that were more than 1SD below the mean. Even more striking was the mean length of produced utterances, which was below age-appropriate norm-scores in 78% of all DHH children in this same study. The discrepancy between language and communicative abilities was the largest in children with MHL. Albeit their language scores were within the normal range, their mean communicative abilities were far below that of hearing peers. Thus, focusing on how a child uses its 'language knowledge' seems more reasonable in order to study a child's functioning in daily life.9

Needless to say, language abilities and communicative abilities are strongly related. Good quality language skills increase the opportunity to engage in communication with others.^{7,11} During these interactions with others, children learn the refinements of social language and how to respond adequately. Vice versa, children can expand their vocabulary and learn new concepts if communication skills are good. This in turn stimulates their language skills. Language and communication skills were indeed found to be highly related to each other in **chapter 3**.

Children's communication skills were also highly related to social functioning, the appearance of behavioral problems (**chapter 3**), and the ability to acknowledge others' desires and beliefs (**chapter 5**). Moreover, early communication skills were shown to be a protective factor against the development of aggression and disruptive behavior. An increase in communicative skills over time also decreased levels of depression and anxiety in preschoolers with a CI (**chapter 4**). Hence, in order to stimulate social-emotional development, we have to increase the changes for incidental learning by actively involving DHH children in conversations, to stimulate communicative abilities and increase exposure to social norms.

Age at detection and intervention

Almost all young children described in this thesis were identified by the NHS which means that their hearing loss was detected at an early age. So, how come that these early identified children did not seemed to have benefitted from this service when it came to their language skills? As pointed out in **chapter** 3 and **5**, and by Korver et al., the children that were part of the DECIBEL-study and were identified through the NHS did not all benefit from early intervention and adaptation after detection.¹² This was due to the fact that the children evaluated in the DECIBEL-study were all born during the implementation phase of the NHS. As a result, intervention services simply were not ready to handle all these young DHH babies. In addition, the government did not reimburse the costs for support in children with MHL yet, so these children were still left in the cold when it came to early support. In addition, both parents and caregivers did not yet have the skills or the knowledge to adequately handle problems in this group of children.

The persistence of language problems in early detected children highlights the fact that early identification on its own is not sufficient. For adequate language development, it needs to be followed by early intervention. This will allow restoring the ability to perceive sounds as soon as possible in order to develop adequate language skills. The outcomes discussed in **chapter 4** support this by showing that earlier implantation of CI resulted in higher language scores in young toddlers. This so-called 'the earlier, the better' statement is widely accepted within this research field¹³ with some researcher even recommending implantation as soon as six months after birth.^{14,15}

Through restoring early auditory input, language skills can be improved and this in turn improves communicative abilities of DHH children. Higher levels of communication skills allow children to actively participate in conversations and interactions with others and stimulate the development of social behavior. Early rehabilitation may thus also improve a child's functioning in social environments. This was shown in **chapter 4** in which earlier implantation led to higher communicative abilities in children with CI. These abilities prevented the development of early signs of psychopathology. Yet, none of the studies described in this thesis were able to show a direct relationship between early intervention and social-emotional development in DHH children. This might be due to the definition

of the term 'intervention'. Early support comes in many forms (e.g., early adaptation of hearing device, family support programs, speech and language therapists), which can all have various impact in various families. In the studies discussed in this thesis, the age at first adaptation or implantation was used to determine the age at intervention. Yet, it could be that other factors of the early intervention program are more important for social development (e.g., advice from a family counselor on how to communicate with your baby) and are thus missed if we only measure when a child wore his or her first HAs. A second explanation for not finding a direct relation perhaps lies in the fact that so many other factors are known to influence social learning such as parental input, maternal sensitivity, cognitive development, and parenting styles.¹⁶ Unfortunately, these factors were not measured in this thesis and we can therefore only speculate about their influence on the development of DHH children. However, as also pointed out by Ketelaar et al. the absence of a direct causal relationship between age at intervention and the level of psychosocial development is consistent with previous literature.¹⁷⁻¹⁹

Hearing loss-related factors

Other factors that are closely related to the child's hearing loss were also analyzed to define their relationship with social functioning in DHH children. Overall, hardly any relationships were found between social functioning and factors such as the degree of hearing loss, mode of communication, and additional speech and language therapy. Only in **chapter 5** a higher degree of hearing loss was related to lower ToM skills (desire and belief understanding). No other studies examined ToM in children with MHL and therefore no comparison with other studies could be made.

In the last decade, the influence of bilateral (2 Cl's or 2 HA's) and bimodal (1 Cl and 1 HA) stimulation on speech and language skills of young DHH children is of increasing interest. Whether compared to bimodal or bilateral stimulation, unilateral amplification shows inferior results when it comes to the speech recognition and subsequent language development. ²⁰⁻²² In **chapter** 4 this finding was confirmed by showing that the communication skills of DHH toddlers with bilateral CI were higher than those of unilateral implanted children despite the fact that the children with bilateral devices were younger. In addition, the children with bilateral implants were - on average - implanted 5 months earlier than the children that received only one CI. So not only early amplification, but also bilateral amplification may increase a child's chances to participate in society and increase opportunities for social learning.

Environmental factors

As pointed out previously, the ability to interact with the environment determines how well a child is able to participate in society and engage in relationships. One way to measure this influence is to compare children that are exposed to different types of environments such as is determined by school placement. In special schools for the deaf, children are allocated to smaller classes and receive more intensive support from teachers

and staff. Mainstream education on the other hand has the advantage of being close to home and being a good reflection of the (predominantly hearing) society. Based on the results found by Theunissen et al. we therefore focused on the relation between school placement and empathic abilities in **chapter 6**.²³ When taking account for the child's degree of hearing loss and language abilities, DHH children in mainstream education were better able to understand the other's feelings. This suggests that DHH children in mainstream education receive quantitatively more chances to observe others and learn incidentally. However, these results need to be interpreted with care because we do not know the reason for school placement. It may be that other factors that we did not measure but that did influence school placement may also determine a child's social learning capacities.

In past research, the socioeconomic status (SES) of a child and his or her family have been proven to influence a child's development. To illustrate, the earliest CIs were often implanted in children of parents with a high level of education, because they had heard of the opportunities of such new techniques and were very motivated to get their child implanted. We therefore examined the relation between the SES of the child and different developmental outcomes in **chapters 3**, **4**, and **5**. No effect of the SES of the child on any of the social-emotional, or on the language and communicative outcomes were found. Most likely, the chances of receiving care are equal for all families in The Netherlands and Belgium and other factors have been proven to be more important for the child's development besides their socio-economic background.

LIMITATIONS

"Once we accept our limits, we go beyond them." (Albert Einstein)

As in every research, the studies discussed in this thesis have their limitations. One of these concerns the reporting and handling of missing data. Although suitable techniques were used in **chapter 3**, **4**, and **5**, unfortunately this was not done in **chapter 6**. Mainly the lack of knowledge at the time of analyzing and writing down the results reported in **chapter 6** let to this inconvenience. By analyzing how others report missing data, and by explaining possible pitfalls in **chapter 2**, we aimed to increase insight in how to handle missing data.

A second limitation of this thesis is related to the effect of different interventions on child development. To study the effect of early intervention on social-emotional development, we mainly used the age at first adaptation or implantation. Unfortunately, the effect of other types of intervention, or their combination is still relatively unknown. Part of this problem is due to differences in various CI-rehabilitation programs that various CI-centers offer. Most of them are designed by the CI-centers their selves and not based on scientific

GENERAL DISCUSSION

proof. Yet, how do we determine the effect of certain types of support, speech- and language support, duration, and intensity of these programs? Ideally, the various CI-centers should join forces and reveal their 'rehabilitation-secrets' in order to compare programs and come to evidence-based therapeutic programs. This will allow for tailor-made rehabilitation programs specified to each individual in order to optimize results. A third limitation concerns the duration of the follow-up of DHH children after early intervention services. The outcomes reported in this thesis were all still quite short-termed. If we want to define the long-term effects of improvements in language and communication skills on children's social-emotional development we have to follow children into their teens. This will allow us to compare early identified children with improved language and communication skills to the later identified children that were previously studied.²⁴

The fourth limitation is related to the potential effect of selection-bias on our results. As in many cohort studies like those described in this thesis, we were confronted with selection-bias. Especially in clinical groups, parents or children may decide not to participate because of various reasons. It is possible that children with more (health-related) problems tend not to take part in research. Outcomes can therefore look brighter than they actually might have been if we had studied the entire population. This was particularly a problem in **chapter 5**. Selection bias was first introduced the moment parents were invited to participate in the study in which they were asked to complete several online questionnaires. However, the second part of the DECIBEL-study involved home visits. Because this part also demanded active participation of their child, not all parents agreed to pursue with this part of the study. It is very likely that this again resulted in selection bias. Future studies should document reasons for not participating in order to grade the level of selection bias in their studies.

DIRECTIONS FOR FUTURE RESEARCH

It is a well-known phenomenon in research: conducting research generates more questions than it answers. This paragraph will therefore highlight some directions for future research to increase our understanding of the relation between childhood hearing loss and the child's social-emotional development.

As described in the introduction of this thesis, language learning is dependent on the quality and quantity of input. When we focus on language and communication development in DHH children, a future step would include analyses of such language input. To what kind of sounds is a child exposed during the day? How do parents approach their child? To what extent do they vary in the language they use in communications with their child? A first step in conducting such kind of research was initiated by means of the Language ENvironment Analysis (LENA) system [LENA Foundation, Boulder, CO, USA].

This system consists of a small recording device that can be attached to the child's clothes and records all auditory input. This input is automatically transcribed and can be used for research purposes but also as a feedback system for parents on their language involvement. The LENA system is also able to detect challenges generated by background noise. As a result, parents can for instance be taught to turn off the TV during the day as this interferes with a child's opportunities to hear language and benefit from incidental learning by observing others.

In the light of technology helping researchers to study interactions, there is currently one other innovation that is worth mentioning. Veiga et al. introduced the use of radiofrequency identification devices (RFID). This device (in the form of a badge) was attached to children's clothes and when in close proximity to each other, the badges connect and this is recorded. The children were also video-taped while playing, to be able to observe the type of interactions. With the use of the RFID badges, young children's play behavior during recess time was recorded and analyzed and afterwards linked to their level of social competence.²⁵ By adding a microphone to the RFID, this would create an even more valuable tool for studying quality and quantity of input of children with hearing loss. Studying their interactions will gain more insight into how DHH children learn incidentally, how often, and with who. By combining this with new analyzing techniques such as big data analyses, this creates unique information regarding social interactions of DHH children. At this very moment, an international collaboration is set up to start using the RFID badges in a school with both DHH and normal hearing children. Nowadays DHH children are more often allocated to inclusive educational settings. It is therefore of increasing importance to study how well DHH children can develop their social skills in an environment that is predominantly designed for hearing children.

Chapter 5 in this thesis shows that children with MHL are at risk for problematic socialemotional development. However, research in this specific group is scarce. Future studies should extent our knowledge on the development of this group of children, who are often overlooked. How well this group of children can function in daily life is possibly of increasing importance. This has to do with evolution in the area of cochlear implantation. Due to early implantation and support, children with CI function increasingly well. They more frequently reach aided hearing thresholds between 25 and 35 dB SPL. It may be that early implanted DHH children are more and more alike children with MHL wearing HAs. This becomes visible when we compare the results of chapter 5 to a study by Ketelaar et al. who studied ToM skills in young children with CI. This study showed that if language skills were high enough to understand the ToM tasks, children with CI scored comparable to our children with MHL.¹⁹ Creating insight into the consequences that partly restored hearing abilities have on child functioning is essential for providing support and training. In addition, this type of research also calls for a different approach to how we measure hearing loss, or hearing thresholds. This has several implications. First, it might be more realistic to test hearing thresholds in noise, as this simulates hearing capacities in daily

GENERAL DISCUSSION

life better than when tested in a quiet room. Second, to assess hearing capacities in daily life we might have to consider measuring aided hearing thresholds instead of unaided thresholds as this is more in line with reality. Using these types of measurements better simulates a child's functioning in everyday situations.

As pointed out in the limitations-section, there is a remaining urge for increasing our understanding regarding the effect of the content of the rehabilitation program on the development of implanted children. What kind of support do families need? With children identified and amplified at a very young age, the focus of support given in the first year has changed gradually. Professionals have to shift their attention to the family instead of only focusing on the child. Research needs to identify which factors influence child performance when children grow older. This not only calls for research concerning different types of interventions, but also for studies with a long-term follow-up to be able to identify causal relationships. This knowledge can be of use when designing a more extensive support program for children with all kinds of hearing losses.

Since this thesis supports the finding that DHH children encounter many challenges in social learning with ongoing consequences that can persist into adulthood, it is time for action. Recognizing early symptoms of problematic social-emotional development allows us to design methods that can help DHH children in their social learning experiences. Both educating parents and teaching the children can stimulate talking about abstract concepts such as emotions as well as increasing exposure to social situations and communication. At this moment, researchers in different fields are exploring the possibilities of for instance virtual reality games to increase exposure and teach children how to behave socially in certain situations. Because of new technologies, smartphones can be used in many ways to stimulate social learning (e.g., apps, interactive games, and social media). Creative new ideas such as these examples will hopefully give a positive impulse to social learning experiences in DHH children.

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