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

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THE LINK BETWEEN **HEARING LOSS, LANGUAGE,** AND **SOCIAL FUNCTIONING** IN CHILDHOOD

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Vertrouwen is het grootste bewijs van liefde
(Joyce Brothers)

Voor mijn ouders

CHAPTER 1

INTRODUCTION

INTRODUCTION

Pediatric hearing loss

Hearing loss is the most common birth defect in developed countries. In The Netherlands, the incidence of hearing loss is approximately 1.7 in every 1000 live births.[1] This means that around 300 children per year are born with a hearing impairment, of which roughly 53-60% is bilateral in nature. Permanent childhood hearing loss (PCHL) is defined as a loss of at least 40 dB in the best ear.[2] Many causes for PCHL have been identified. Around 50% of all congenital hearing loss is genetic in origin. Examples of this are DFNB1 in which mutations in the GJB2 gene cause connexin deficits, and syndromes like Usher and Waardenburg syndrome that can additionally cause vision problems. Syndromes like Treacher-Collins are more related to conductive hearing loss due to aural atresia. Besides genetic causes, PCHL is often due to infections. These infections are either prenatally acquired by one of the TORCH organisms (i.e., **To**xoplasmosis, **R**ubella, **C**ytomegalovirus, and **H**erpes), or postnatal such as in cases of bacterial meningitis. Other causes of hearing loss in newborns include both prematurity on itself as well as medical interventions during the first few weeks of life (e.g., antibiotics, extracorporeal membrane oxygenation).[3] This explains the higher levels of PCHL found in children that were admitted to the Neonatal Intensive Care Unit (NICU).[1,4]

Table 1. Etiology of permanent childhood hearing loss

Hereditary (40%)	*Non-syndromic (45%)*	GJB2 homozygous, DFN mutations, mitochondrial deficits
	Syndromic (40%)	Pendred, Usher, Jervell-Lange Nielsen, Waardenburg syndrome
	Positive family history non-specified (15%)	
Acquired (30%)	*Prenatal (32%)*	Cytomegalovirus, Rubella
	Perinatal (53%)	Asphyxia, Ototoxic medication, prematurity, neonatal icterus
	Postnatal (15%)	Meningitis, ECMO therapy
Miscellaneous (5%)		Cleft palate, aural atresia
Unknown (25%)		

Consequences of hearing loss in children

Deaf and hard of hearing (DHH) children encounter many challenges growing up in a society that strongly relies on the ability to hear sounds. If children experience difficulties in capturing for instance spoken language and conversations, this will likely interfere with their capacity to learn a language and to communicate with others. Encountering a hearing loss in the first few years of life can have ongoing consequences. This is mainly due to brain plasticity. The plasticity of the brain enables young children to learn languages relatively easy. However, plasticity decreases with age, making the brain less susceptible to auditory input as children grow older.[5,6]

Besides their speech and language problems, DHH children are often confronted with problems in their social and emotional development. Two systematic reviews uniquely evaluated research on the appearance of psychopathology, emotional, and behavioral problems in DHH children and adolescents and showed higher levels of depression and somatization, and a tendency for higher levels of anxiety compared to hearing children. In addition, DHH children reported higher levels of aggression and behavioral problems as compared to hearing controls.[7] They also encountered more problems in engaging in peer relationships and friendships than hearing children.[8]

Why are DHH children at risk for all these psychosocial issues? In the last two decades, researchers attempted to identify factors that influence psychosocial development in DHH children. The first studies in this relatively new research area were only able to compare DHH children to children with normal hearing on mean scores of existing psychosocial questionnaires. These (often Quality of Life-related) questionnaires were not adapted for use in children with language difficulties. Other questionnaires were often only for parents to complete. Therefore, the next phase in research on the well-being of DHH children consisted of the use of self-reports that were adjusted to the communicative abilities of these children. This provided more insight in the social and emotional development when born with PCHL. However, this did not answer the question *why* DHH children are at risk for encountering psychosocial problems. Therefore, studies started to examine relationships between hearing-related variables, demographic characteristics and child performance. For instance, the influence of the type of hearing device has been studied widely and some researchers showed superior results for children wearing cochlear implants (CI) when compared to children wearing conventional hearing aids (HA)[9-13], whereas others did not find a difference between CI and HA users.[14,15] Up till today, the only certain conclusion we can draw from past research on the influence of type op device is that children fitted with HA never performed better than children with CI or hearing peers. [7] Others studied the relation between spoken language and social functioning, or the socioeconomic status of the families.[16-19]

Studies like the abovementioned were innovative at that time, and gained new knowledge concerning the development of DHH children's social and emotional skills. Yet, these studies suffered from several limitations. These limitations were shaped into five research objectives which formed the basis of this thesis.

LIMITATIONS IN RESEARCH IN DHH CHILDREN

1. Missing data

The first limitation in research on DHH children's development is epidemiological in nature. In clinical research (such as research in DHH children) researchers are inevitably challenged by missing data. Data may be missing for various reasons (e.g., medical files are untraceable, people moved, patients are unable to recall certain dates or situations). Yet, how a researcher deals with this problem can have ongoing consequences for the analyses, outcomes and conclusions that are drawn from studies. There might actually be valid reasons why certain data is missing, and it is possible that participants with missing data are performing different compared to participants with complete data. Hence, keeping those participants with incomplete data out of the analyses can bias the outcomes and conclusions drawn from various analyses. Another serious problem is the decrease in power that is the consequence of only analyzing complete cases, when missing data is present.[20] Especially in DHH research, study groups are often small. Leaving out participants because of missing data may prevent finding significant outcomes as a result of power issues. Therefore, there is a strong need for hands-on approaches and guidelines on how to deal with missing data-related issues in clinical research. This led to the first aim of this thesis:

Objective 1 (*chapter 2)*: To illustrate the effect of various methods to handle missing data on outcomes in clinical research.

This chapter highlights the importance of reporting missing data in clinical research. The consequences of inadequately handling missing data are explained by means of examples from the literature. A state of the art technique to handle missing data called multiple imputation is explained in this chapter.

2. The link between language, communication, and social functioning

A second challenge in research on DHH children is the ongoing innovation in this area. With the clinical implementation of otoacoustic emissions (OAE's) in the beginning of this century, hearing loss of >35 dB could be detected objectively (i.e. without active participation of the child). This technique allowed testing of the hearing capacities of newborns within a few days after birth already. The use of OAE's created a window of opportunities to start intervention of hearing loss earlier in life.[4]

Research to identify the effect of early identification and intervention of hearing loss on the development of DHH children showed improvement in their speech and language skills.[21-24] Because of early detection, young children with profound hearing loss were also implanted earlier in life. To illustrate, children nowadays preferably receive a CI before their first birthday.[25] Earlier implantation has been proven to increase language skills.[26-28] In addition, another topic that became of increasing interest was the importance of bilateral hearing. Researchers found superior results of both receptive and expressive language skills in children who were bilaterally implanted when compared to those who were unilaterally implanted.[29]

To summarize, ongoing improvements in technology (e.g., CI, digital hearing aids, and the introduction of new screening methods) kept changing the study population. Because children performed better with every step, they became incomparable to the 'traditional'

Early identification: the newborn hearing screening

Testing of the newborn's hearing abilities through OAE's was introduced in The Netherlands by means of the Newborn Hearing Screening (NHS). This nationwide screening program was gradually introduced in 2003 and nationwide spread was accomplished at the end of 2005. Newborns were tested at home or at a well-clinic. If a child fails the screening, OAE is repeated. After the second failure, hearing abilities are measured using Automated Auditory Brainstem Responses (AABR). The Joint Committee on Infant Hearing (JCIH) in their 2007 statement recommended screening within the first month of life. Children that do not pass the screening should be evaluated at an audiological center within the first three months of life.[30]
The research presented in this thesis represents all phases of the transition that took place with the introduction of the NHS in The Netherlands. Chapter 6 studies DHH children born before the introduction of the NHS, chapters 4 and 5 study children born during the implementation of the NHS and chapter 3 focusses on DHH children with cochlear implants (CI) all born after implementation of the NHS.

deaf children with severe speech and language difficulties. This raised the question of *how earlier detection, intervention and improvements in speech- and language abilities affect the social-emotional skills of early identified DHH children*? To answer this, we have to look closer at the possible effects all these innovations can have on child development in its broadest sense.

The impact of hearing loss: language development

The development of speech and language abilities is probably the most extensively studied topic when it comes to research in DHH children.[31,34,35] In order to learn a language, an essential need is to have access to this language. Because of their hearing difficulties, DHH children have diminished access or even no access at all to spoken language. Diminished input decreases the amount of opportunities to learn from. Previously, DHH children have been found to have both speech and language problems, especially children with severe to profound hearing loss who have hearing parents.[34] Nevertheless, not only children with severe hearing loss are at risk. Recent studies show that even mild hearing loss can cause language problems.[35,36]

With the introduction of early identification programs, the opportunities of DHH children have increased. Early amplification with hearing aids, early family support and innovative techniques such as CI have been proven to increase both speech and language abilities in DHH children.[35,37,38] Yet, not only the capacity to hear is what influences speech and language development in children. For successful language development, both quality and quantity of language input are important. In the first few years of life, parents or

The impact of hearing loss: the brain

The cortex in the brain consists of many functionally and histologically distinct areas (called Brodmann's areas, BA). These areas interconnect to form functional units to become able to interpret sensory input. The auditory cortex is defined by the primary auditory cortex A1 (BA41) and the secondary auditory cortex A2 (BA42) as highlighted in Figure 1.

Unlike the cochlea, the auditory cortex in the brain is not fully developed at birth. Both auditory input and interaction with the environment stimulate the development of the auditory cortex throughout childhood; a child learns to hear. In the case of congenital hearing loss, not only the auditory input is lower (or even absent), but due to language and communication problems interaction with the environment is also limited. This results in diminished auditory stimulation of the cortex.

However, input for cortical development is not only auditory in nature. Both visual and somatosensory stimuli can also serve as cues to inform a child about his or her surroundings. As a result of hearing loss, the proportion of these various types of sensory input is different. Visual and somatosensory input represent the largest part of sensory information since auditory information is lacking. Thus, the composition of sensory input differs in PCHL compared to typical cortical development. Due to this change in input, aberrant patterns develop in the brain, making the cortex less sensitive for auditory stimuli as children grow older. Furthermore, other sensory systems overtake parts of the auditory cortex and thereby decrease the auditory cortex.[5,31]

Cortical development strongly relies on so-called *sensitive periods*, in which the brain is more susceptible for alterations based on input.[32] This period of high susceptibility to environmental modification occurs in multiple areas that need to 'learn' from input, such as vision and hearing. A sensitive period in auditory development was previously demonstrated in babies by observing their ability to discriminate phonetic contrasts. In the first few months of life, children are able to discriminate between the phonetic contrasts off all languages. Yet, around the age of 8 to 10 months old, young children specialize this skill and only remain sensitive for contrasts in the mother language.[6] The brain thus learns to discriminate between useful 'sound-objects' and less useful 'noise'. In order to optimally discriminate, the cortex needs to receive high quality sounds.[33] Thus, both limited quality and quantity of auditory input affect cortical development in PCHL.

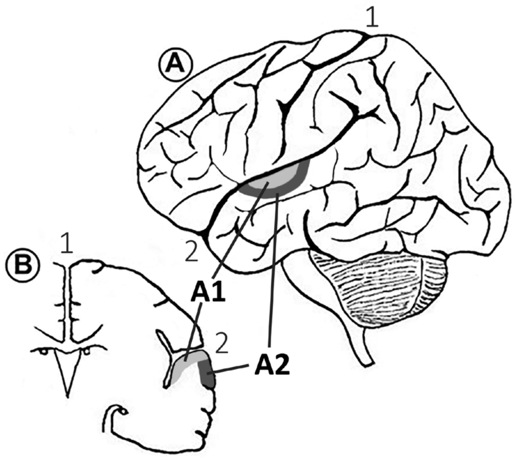

Figure 1 A. Lateral view of the brain (schematic). The primary auditory cortex (A1) is highlighted in light grey and the secondary auditory cortex (A2) is highlighted in dark grey. **B.** Coronal view.

caregivers provide the largest amount of input. Therefore, it is essential that parental input is rich in nature.[39] However, research has shown that parents of DHH children more often use shorter sentences and communicate in a more directive manner with their child to ensure comprehension and achieve daily routines (e.g., 'put on your clothes', 'finish your food').[40] In addition, they also use less mental state talk and find it more difficult to talk about abstract concepts such as emotions with their DHH child, than with their hearing children.[35] Again, diminished input decreases the chance to learn and develop a language. In this respect, better vocabulary, speech understanding, speech production and language skills were all related to lower levels of psychopathology and better social functioning.[7,8] Yet, no matter how rich a child's vocabulary is, this does not certify that the child uses this language in everyday life. Is it therefore not more reasonable to measure how well children use their language? It other words: how capable are DHH children to communicate in everyday life?

Consequences of language difficulties

Previously, deaf children encountered many challenges in daily life because their poor spoken language skills prevented them from actively participating in communication with (hearing) others. Being part of society and sharing the same mode of communication is essential to achieve high quality of language input and to learn during communication with others.[41] This is illustrated by past research examining various forms of psychopathology in DHH children and (pre)adolescents: children with CI in mainstream education reported lower levels of symptoms like anxiety or aggression than children wearing hearing aids and attended special education for the deaf.[15,42] Restoring the ability to detect sound and even to *hear* sounds (e.g., through CI) increased the opportunities of DHH children to participate in the sound-dominated society. The better children can keep up in conversations with others and join interaction with peers, the better their overall development.[7,8]

To summarize, the OAE measurements together with the systematic introduction of the NHS allow us to detect hearing loss days after birth. Early restoration of access to auditory input increases chances for adequate development of the auditory cortex. Subsequently, early intervention of hearing loss has been proven beneficial for language development of young children, with superior results in children constantly exposed to high-quality spoken language, supported by committed parents and caregivers.[31] Based on these improved chances for young children with PCHL, the JCIH recommended the amplification of hearing as soon as possible (i.e. within the first six months of life).[30] This raises the question how early identification relates to early language skills and communicative abilities of young children. And if communicative abilities increase with earlier detection, does this also increase a child's opportunities to interact with others? The above mentioned new technologies in this research field laid the basis for the third aim of this thesis:

Objective 2 *(chapter 3)*. To study the relationship between language, communication and social-emotional development in early identified DHH children.

The effect of early intervention of hearing loss on the development of language skills, communication skills and social functioning is studied in chapter 3. It aimed to identify the relation between these three variables in young DHH children.

3. The identification of causal relationships

A third limitation in past studies in DHH children is related to the type of study-design that was almost always chosen. Many studies that aimed to measure psychopathology in this group of children showed a clear relation with the child's language skills.[7,8] This is not surprising because good quality language skills allow children to actively participate in conversations with others. From these conversations, children learn about social norms and how to behave. Hence, language development stimulates social development. Yet, a language is learned through exposure. Language exposure is mostly obtained during communication with others, which stresses the need for social skills to join such conversations. Summarizing, the relation between language skills and social skills can be bidirectional. However, studies examining the level of psychopathology and its relation with other hearing-loss related factors were performed in a cross-sectional design. Cross-sectional studies evaluating these factors lack information considering the effect of time and the direction of causality. We therefore need to study child development over time in order to identify factors that cause the development of symptoms. Past research confirms this statement by clearly calling for longitudinal studies to examine the direction of causality in this relationship.[7,8,43] With the use of longitudinal studies, it is possible to identify factors that may induce or prevent the development of psychopathology later on in time. This may help to identify children who are at risk for problematic psychosocial development. Therefore the second aim of this thesis was:

Objective 3 (*chapter 4)*: To study the causal relation between language and psychosocial development in young DHH children with CI compared to hearing controls through longitudinal analyses.

The development of early signs of psychopathology over time is longitudinally examined in this chapter in both young children with a CI and hearing peers. It identifies the effect of early identification of hearing loss on the development of language skills. Next, it clarifies the relation between the development of language skills and the development of psychopathology in young children.

4. Consequences of hearing loss for social-emotional development

Sharing the same mode of communication does not only benefit language development. Being able to interact with others also allows children to learn how to behave. For instance, imagine a girl who is coloring at the kitchen table when her brother comes in. He is crying because he fell and hurt his knee. Mom immediately comes to him, cleans up the scratch on his knee and comforts her son. Just by overhearing this conversation, the girl learns about facial expressions (sadness and crying), others' emotions and feelings (empathy), comforting someone (prosocial behavior), and so on. Learning by observing your surroundings is also known as *incidental learning*. It is unplanned and unintended learning that takes place outside of educational settings. It is clear that having a hearing loss interferes with opportunities for incidental learning because not every conversation can be picked up and learnt from.[41] Because incidental learning often takes place in social situations with a lot of background noise, DHH children are less able to learn incidentally. This puts them at risk for problematic social-emotional development.

Via incidental learning, we learn to share our attention in order to engage with others. This is essential for a child's social development. Joint attention learns us that people have certain intentions in life, they want to achieve certain goals. A two-year old can only think of his own goals and believes that everyone has these same intentions. Through social learning (i.e., learning that takes place in a social context), children start to acknowledge that different people can have different thoughts, desires, and beliefs. This ability to describe mental states to others is known as Theory of Mind (ToM). The development of ToM is an essential first step to be able to understand others' feelings, and thus determines the basis of our social successes.[44-46] Participation in interaction with others is important for adequate ToM development. It is therefore of no surprise that language skills were previously found to be closely related to children's ToM abilities.[47]

Research on ToM development in DHH children compared to hearing peers found mixed results, mainly because the study groups varied greatly in age and language abilities.[44,48-56] However, the various studies on ToM in DHH children have one thing in common. They all focus on children with severe to profound hearing loss. Nevertheless, research has shown that children with moderate hearing loss (MHL) are also at risk for delays in language development.[36] Up till now, children with MHL received little attention, most likely because their (hearing) capacities are often overestimated.[40,57] Because of their challenged language development, it is likely that children with MHL are at risk for problematic ToM development. Yet, studies confirming this hypothesis are lacking and this was therefore the fourth aim of this thesis:

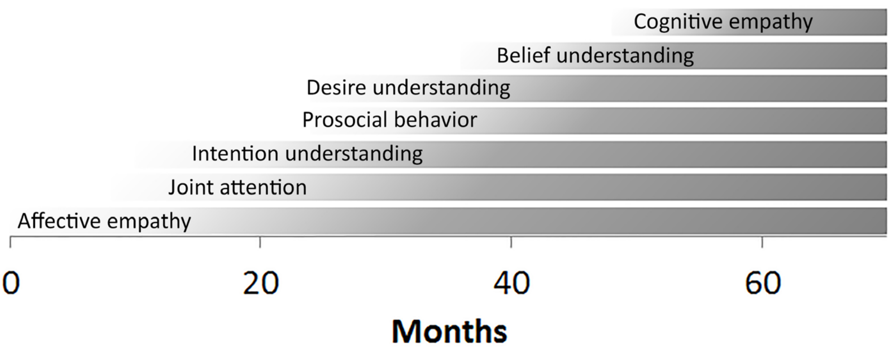

Figure 2. Schematic representation of the development of various milestones for social-emotional in childhood.

Objective 4 *(chapter 5)*: To study the developmental pattern of ToM in young children with moderate hearing loss compared to hearing controls.

This chapter examines the development of different aspects of Theory of Mind in young children with moderate hearing loss and hearing controls. The understanding of others' intentions, desires and beliefs is observed through several experiments and related to the child's language abilities.

5. Empathic abilities of DHH children

Social-emotional development strongly relies on the ability to engage in relationships with others. In order to initiate and maintain relationships, it is essential to be able recognize and understand others' feelings. How capable we are in acknowledging others' emotions and acting upon them is defined by our empathic abilities. Empathy is the capacity to feel what the other is feeling and to understand why.[58,59] Advanced empathic abilities thus distinguish between the own and the other's emotion. It defines how well we can relate to others' emotions and how we feel for the other. Empathy is regarded as the social glue in relationships because it determines our behavior towards the other and thus our relationship with the other.[60] To illustrate, if a child knows why her friend is sad, he or she can comfort him, or even help to solve the issue. In addition, children that score high on empathic skills are also better liked by their peers and teachers.[61] Empathic development is stimulated by observing how others interact (i.e., incidental learning). Therefore, DHH children are potentially more prone to develop delays or even deficits in their empathic development.

Research measuring the empathic abilities of DHH children is scarce and inconsistent. A reason for these mixed findings lies in the concept of empathy itself. Because it is such a broad term that encompasses different stages of development, it is often divided based on its development in childhood. *Affective empathy* is the first stage and is characterized by contagion with the other's emotion. As ToM develops in children they learn that different people can have different feelings. This is the basis for *cognitive empathy*: to understand what the other is feeling. Its development requires perspective taking skills. Being able to understand the other's emotions allows us to help the other. Empathy thus induces prosocial behavior.[58-60]

Because cognitive empathy is always a reaction that is based on an interaction with someone else, it can also be evaluated through multiple sources. To evaluate empathic skills as concise as possible, it would therefore be interesting to have different informants: the child itself (self-report), a close relative (parent-report) and an independent source (observation). Since this type of research has never been performed before, this was the fifth and final aim of this thesis:

Objective 5 *(chapter 6)*: To examine empathic skills in DHH (pre) adolescents compared to hearing peers.

In **chapter 6**, the empathic abilities of DHH teenagers and hearing peers are analyzed. Both self-reports, parent reports, and observations were used to define levels of affective empathy (emotion contagion), cognitive empathy (emotion understanding) and prosocial motivation. Next, the relation between empathy, school placement and language skills is studied. **Chapter 7** starts with a summary of the main outcomes of all chapters based on the aforementioned research questions. Next, these outcomes are integrated and discussed in order to draw an overall picture regarding the effect of hearing loss on language abilities and the consequences this has for a social functioning in DHH children. This chapter concludes with recommendations for future research. A summary of this thesis in Dutch can be found in **chapter 8**.

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CHAPTER 2

MISSING DATA IN THE FIELD OF **OTORHINOLARYNGOLOGY** AND **HEAD & NECK SURGERY**: NEED FOR IMPROVEMENT

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ABSTRACT

Objective

Clinical studies are often facing missing data. Data can be missing for various reasons, e.g., patients moved, certain measurements are only administered in high-risk groups, patients are unable to attend clinic because of their health status. There are various ways to handle these missing data (e.g., complete cases analyses, mean substitution). Each of these techniques potentially influences both the analyses and the results of a study. The first aim of this structured review was to analyze how often researchers in the field of otorhinolaryngology / head & neck surgery report missing data. The second aim was to systematically describe how researchers handle missing data in their analyses. The third aim was to provide a solution on how to deal with missing data by means of the multiple imputation technique. With this review we aim to contribute to a higher quality of reporting in otorhinolaryngology research.

Design

Clinical studies among the 398 most recently published research articles in three major journals in the field of otorhinolaryngology / head & neck surgery were analyzed based on how researchers reported and handled missing data.

Results

Of the 316 clinical studies, 85 studies reported some form of missing data. Of those 85, only a small number (12 studies, 3.8%) actively handled the missingness in their data. The majority of researchers exclude incomplete cases, which results in biased outcomes and a drop in statistical power.

Conclusion

Within otorhinolaryngology research, missing data are largely ignored and underreported, and consequently, handled inadequately. This has major impact on the results and conclusions drawn from this research. Based on the outcomes of this review, we provide solutions on how to deal with missing data. To illustrate, we clarify the use of multiple imputation techniques, which recently became widely available in standard statistical programs.

INTRODUCTION

"When dealing with real data, the practicing statistician should explicitly consider the process that causes missing data far more often than he does."

Rubin (p.589, 26)[1]

Missing data are almost inevitable when conducting research using patient information.[1-4] For numerous reasons, databases are incomplete and researchers have to decide how to deal with this issue. Most often in medical research, this problem is overlooked and missing data are underreported.[4,5] However, it is important for researchers to realize that standard analyzing techniques assume complete cases and consequently remove incomplete cases from the analyses. Ignoring missing data through complete case analyses introduces bias and a drop in statistical power as it insufficiently uses the available data.[2] The first aim of this structured review was to evaluate the (under)reporting of missing data in the otorhinolaryngology research field. The second aim was to analyze how researchers deal with missing data and highlight the consequences this potentially has. The third aim was to provide solutions on how to deal with missing data using modern techniques that are widely available nowadays.

The quality of medical research reports is of increasing interest to assure valid outcomes and generalizability. A growing number of journals requests authors to complete checklists such as the Consolidated Standards of Reporting Trials (CONSORT) for randomized controlled trials and the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) for observational studies.[6,7] These checklists provide a guideline for the concise report of medical research. Among other things, checklists like STROBE emphasize the importance of reporting missing data in all variables of interest and strongly recommend to give reasons for missing data where possible.

Types of missing data

What to do when confronted with missing data largely depends on under what assumption the data are incomplete. In other words, what are the characteristics of the missing data and do we know the reason why a value is missing? Epidemiologists assume three types of missing data: i.e., Missing Completely At Random (MCAR), Missing At Random (MAR), and Missing Not At Random (MNAR).[3]

Missing Completely At Random (MCAR)

The reason for missingness is completely independent of the (missing) true value, and from any other variables that are or are not included in the dataset. An example of MCAR is a questionnaire that was lost in the mail, or a broken freezer that contained frozen patient specimens. In the case of MCAR, the observed values are a random selection of the sample and thus, are representative for that population.

Missing At Random (MAR)

In the MAR condition, the reason for missingness is related to other factors that are measured within the dataset. This term can be confusing as it suggests that there is no relation between the missing values and other factors, albeit there is. For instance, in a dataset, spoken language scores are more often missing from Deaf and Hard of Hearing (DHH) children that prefer to use sign-supported language as their mode of communication. Likely, the missing scores for children that prefer to use sign language are lower than for children who prefer spoken language. In the MAR assumption, factors that are related to the missing values (e.g. communication mode) can help to reconstruct the actual level of spoken language scores.

Missing Not At Random (MNAR)

A problem arises when the reason for missing data is related to the true value, or to other unknown factors. Yet, these variables are all unknown. This is the case in data that is MNAR; data it is missing only because of its value. To illustrate, MNAR might happen when asking cancer participants about their quality of life during their out-clinic appointment. The answers might be missing because the patient was too sick to attend to clinic. Another example is patients suffering from depression that are too depressed to complete a questionnaire about their mental wellbeing. Here, the true value of the outcome measure is the reason why the specific value is missing. The difference with both MCAR and MAR is that in the MNAR condition we do not know the reason, nor can we speculate what the true value would have been, because essential information is not available.

Hypothesizing the reason for missingness and under what assumption data are missing is helpful in the process of deciding how to handle this issue. Although it is tempting to assume that data fall under either one of these three assumptions, often the pattern of missing data is a combination of more than one of the assumptions. The missing data of some patients are MCAR, others are MAR, and others are even MNAR. Reporting missing data is essential to assure valid and replicable results. Unfortunately, this is still quite unpopular in medical research. To illustrate this statement, this structured review identified how researchers in the field of otorhinolaryngology reported and handled missing data. Additionally, we explain the multiple imputation technique to adequately handle missing data.

METHODS

A literature review of the most recent articles published in three major Otorhinolaryngology/Head & Neck surgery journals was performed to identify how researchers reported and handled missing data. All articles published between September 1st 2014 and August 31st 2015 in the journals *Ear and Hearing* (159 articles), *Rhinology* (76 articles), and *Head & Neck* (679 articles) were identified. Because the third journal published over 600 articles

during that period, we decided to analyze a sub selection and included all articles published between the 1st of May and the 31st of August 2015 (163 articles). A total of 398 articles were identified. Articles were excluded if they did not describe clinical research as is the case in reviews, letters and case-reports. A total of 316 articles describing clinical research were selected for further analysis. For details on exclusion, see figure 1.
All included articles were systematically checked on terms like 'missing', 'unknown', 'remove', 'exclude', 'complete', 'absent', 'lost', and 'imputation' by the first author. The methods and results section of each article were analyzed based on two questions: i.) did the authors report missing data and if so, ii.) how did they handle the missingness in their analysis? Figures and tables were checked if numbers added up, and whether or not they reported characteristics to be 'unknown' or 'missing'. Statistical analyses were checked as to whether the degrees of freedom were consistent, if imputations were mentioned or applied, and if other likelihood-based methods were used that are able to handle missing data without excluding incomplete cases, such as linear mixed models.[8] A second researcher additionally checked 30 randomly selected articles out of the 316 articles and confirmed the findings of the first one.

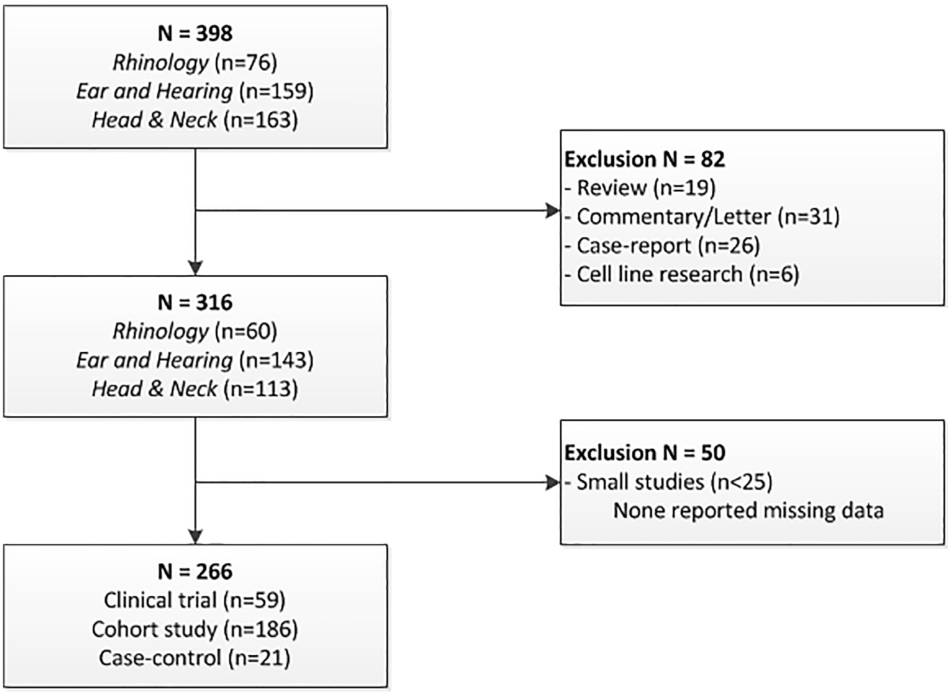

Figure 1. Flow chart of structured review

RESULTS

Of the 316 eligible articles, roughly one-fourth (85 articles) reported some kind of missing data, either in the text, or it was indirectly derived from tables, figures or analyses. In 73 of those 85 articles, complete case analyses or pairwise deletions were used. The remaining 12 articles (9 in *Ear and Hearing*, 2 in *Head & Neck*, and 1 in *Rhinology*) actively took action upon their missing data. In eight of these 12 articles, the mean substitution method was used. In two articles complete and incomplete cases were compared on several variables to illustrate that data were MCAR. In one case, a linear mixed model was used and in the remaining case, multiple imputations were performed to handle missing data, see Table 1 and Figure 2 for an overview.

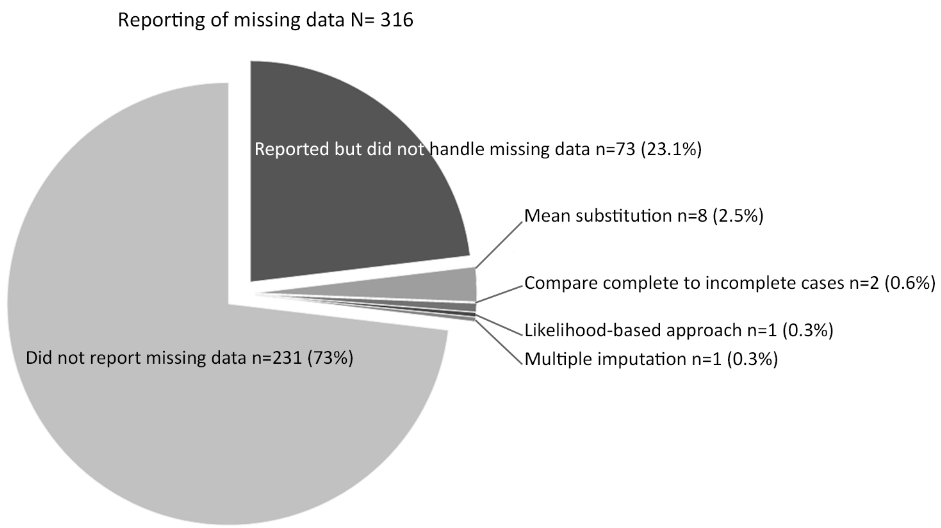

Figure 2. Proportion of papers that reported missing data

Fifty of the clinical studies in this review had a relatively small sample size (i.e., less than 25 participants). None of these small studies reported missing data. Most of these studies were experiments in the area of cochlear implantation with few participants. Because of the small sample size, these type of studies usually do not encounter missing data related issues and often only perform descriptive statistics. Therefore, we decided to perform a sensitivity analyses and excluded the 50 small studies. Excluding these studies only raised the percentage of studies that reported some kind of missing data (n=85) to nearly one-third of the total sample.

Table 1. Characteristics of selected studies that actively handled missing data

Author	Type of study	Imputation method	Detail	Journal
Aarhus et al.[9]	Longitudinal cohort	Mean substitution	Comparison of responders vs. non responders on many characteristics, report loss to follow-up and discuss the probability of selection bias	*Ear and Hearing*
Barry et al.[10]	Cross-sectional case-control	Mean substitution	Within different questionnaires, missing data were replaced by mean data	*Ear and Hearing*
Bulut et al.[11]	Cross-sectional cohort	Mean substitution	Comparison of responders vs. non responders on two characteristics, mean substitution in one questionnaire	*Rhinology*
De Kegel et al.[12]	Longitudinal case-control	Likelihood-based approach	Do not report missing data, no. of participants increases with follow-up time	*Ear and Hearing*
Hesser et al.[13]	Cross-sectional cohort	Mean substitution	Within different questionnaires, missing data were replaced by mean data if < 20% of items per scale was missing, followed by complete case analyses	*Ear and Hearing*
Hornsby & Kipp[14]	Cross-sectional cohort	Mean substitution	Missing data were replaced by mean data in one questionnaire, followed by complete case analyses	*Ear and Hearing*
Huang et al.[15]	Cross-sectional cohort	Mean substitution	Comparison of responders vs. non responders on several characteristics to account for selection bias, in one questionnaire, missing data were replaced by mean data if < 50% of items per scale was missing	*Head & Neck*
Kumar et al.[16]	Cross-sectional cohort	Mean substitution	Within one questionnaires, missing data were replaced by mean data, followed by pairwise deletions	*Ear and Hearing*
Mackersie et al.[17]	Cross-sectional case-control	Mean substitution	In ECG: artifacts were removed and missing intervals were interpolated from the adjacent interbeat interval values (<1%)	*Ear and Hearing*
Schaefer et al.[18]	Cross-sectional cohort	Mean substitution	For missing zip codes, the state average was imputed. Bootstrapping was used to obtain confidence intervals of the built model	*Head & Neck*
Sereda et al.[19]	Longitudinal cohort	Multiple Imputation	No information	*Ear and Hearing*
Stam et al.[20]	Longitudinal case-control	None	Comparison of responders vs. non responders, report selection bias because of loss to follow-up	*Ear and Hearing*

DISCUSSION

This structured review examined how often researchers in the field of Otorhinolaryngology/ Head & Neck surgery report missing data in their research. If missing data were reported, the second aim was to analyze how researchers solve missing data-related issues. The outcomes of this review underline the importance of this study. Despite the introduction of checklists (such as the STROBE) to increase the quality of reporting, the majority of researchers do not report missing data, nor step up to act adequately when confronted with missing data. This might be due to the fact that the use of such checklists is not mandatory in many journals, and their use is therefore relatively unknown. We therefore assume that this underreporting of missing data is most likely the result of unfamiliarity with the consequences of missing data assumptions rather than an unwillingness to deal with this issue.[21] To increase awareness, we will attempt to explain how several commonly used methods to handle missing data can influence results. Second, we will provide a solution on how to adequately handle missing data using modern, well-established techniques.

Complete case analyses

As can be seen in Figure 2, the majority of researchers who reported missing data did not handle this issue. Not deciding how to handle missing data results in *complete case analyses* (also called *listwise deletion*), i.e. the incomplete cases are removed from the analyses. In programs like SPSS, this is automatically done.[22] When performing a *t*-test for example, the program removes incomplete cases when conducting the test and reports the amount of cases with incomplete data. It is important to note that this method is only accurate when the cases with complete data are a random selection of the population. In other words, the incomplete cases may not differ systematically from the complete cases. Complete case analyses can thus only be used if missing data are MCAR. Strikingly, the MCAR assumption is very difficult to prove. The researcher has to be sure that there is no common reason why this specific selection of data is missing. Yet, in practice, data are most frequently MAR. Hence, the complete cases analyses technique will rarely produce the most accurate outcomes. To add, removing incomplete cases from the analyses will always result in loss of power and accuracy.

Comparison of complete and incomplete cases

In this review, four research groups attempted to prove the MCAR statement by *comparing complete and incomplete cases* on several characteristics that could potentially influence the missing variable in order to prove no differences between the two groups.[9,11,15,20] Yet, it is often impossible to test all possible related variables. As a result, assuming MCAR and removing incomplete cases from the analyses produces biased results and broadens the confidence intervals as a result of lower statistical power if data are MAR or MNAR. Unfortunately, complete case analyses are often used without hypothesizing the reason for missingness. The same goes for *pairwise deletion*. In this technique the complete cases are identified and analyzed separately. This method was identified once in this review.[16] Pairwise deletion additionally blurs the outcomes as the number of participants differs per analysis. To illustrate, if correlations are measured but the number of participants per analysis differs, this may yield biased estimates.

Mean substitution

The disadvantages of complete case analyses suggest it might be more convenient to reconstruct the missing data instead of throwing incomplete cases out. Standard techniques can then be used on the reconstructed dataset which solves the power issue. In this review, eight researchers chose to use the *mean substitution* technique, which calculates the mean of the complete cases and imputes ('fills in') this mean in all missing fields of that variable.[17] This tool was most often used when data in questionnaires was missing.[9-11,13-16] Manuals of validated questionnaires often state that a scale may be measured if n % of the items to calculate that scale is missing. For example, if a scale consists of five questions but only four are answered, the mean of these four questions is imputed in the fifth question because the questionnaire assumes a high correlation between the five items within a certain scale (i.e., the internal consistency of the scale). In one other article, zip code-specific socio-economic variables of participants with missing zip codes were replaced by the state average.[18]

However, this method has some disadvantages. Suppose there is a correlation between the outcome and the substituted value. As a result of mean substitution, the strength of this relation alters. To add, it also artificially narrows the confidence interval of the imputed variable because a higher percentage of data lies closer to the mean.

Missing data in longitudinal research

Last observation carried forward (LOCF, also known as *baseline observation carried forward*) is a method that can be used in longitudinal data. This method was not used in any of the articles in this review but is worthwhile to discuss as longitudinal data is increasingly collected, also in Otorhinolaryngology / Head & Neck surgery research. This method copies the last known observation in a row of observations and imputes it in the missing fields of that case. An advantage of this method is that it is case specific because it acknowledges the fact that every case is different and unique. However, the development over time is seriously biased by this method and special analyzing techniques should follow after LOCF. Especially if one is interested in development over time or a treatment effect, these results are biased by LOCF. An additional problem arises when the baseline measure is missing as these cases will still be excluded in complete cases analyses. In addition, cases with missing data in (one of the) confounders will be excluded when such confounders are added to the analyses.

Likelihood-based approaches

De Kegel et al. use linear mixed models in their longitudinal study to account for missing values.[12] *Likelihood-based methods* such as linear mixed models create a model based on the observed data of both complete and incomplete cases. It calculates the maximum likelihood estimate; the value of a parameter that is most likely to have resulted in the observed data. Both the likelihood estimate of the complete and incomplete cases are calculated and jointly maximized. This method does not impute values and is therefore relatively easy to use. It is a reliable method when confronted with missing data in studies with a longitudinal design. However, likelihood-based approaches are limited to linear models. Another potential pitfall when using this approach is that all the factors that are entered into the model besides the dependent variable should not have missing data. Otherwise these cases will still be excluded from the analyses.

A state of the art solution: Multiple imputation

All the above described methods to handle missing data have their limitations. We will therefore now highlight the abilities of *multiple imputations* (MI), a well-established technique that has none of the limitations described above. MI is increasingly used since popular statistical programs started to include its possibility in their interface. This technique was used in only one article in this review.[19]

Imputation means nothing more than "filling in the data". Multiple imputations indicate that the imputations were done more than once. To illustrate the mechanism behind MI, we will return to the previously mentioned fictive dataset containing language scores of DHH children in which language scores of some children were missing. In this database, we observed that children who preferred to use sign-supported language often had lower spoken language scores than children that preferred to use spoken language to communicate. If we now decide to use the preferred mode of communication of the child to predict their language scores, this would produce a more accurate result than when imputing the mean language score of the whole sample. In the same line of thinking, we also know from the complete data that children attending mainstream schools show higher language scores than those attending special education. We can therefore decide to include the type of school that the child attended into the prediction model. Additionally, the age of the child is also positively related to its language abilities, and so on. One will notice that the more variables we will put into this so-called prediction model, the more accurate the prediction of the possible language score will turn out. The MI method uses the complete data to compute a prediction model of the variable that has missing data. It then uses characteristics of the missing cases to predict the missing values in the data. Obviously, the imputation model only calculates an estimation of the unknown value. The true value lies within a certain range that was estimated by the calculated prediction model. We therefore want to insert a certain amount of uncertainty (or variance) for this value. To achieve this, instead of doing this imputation only once, we have the model predict a language score n times. This results in one large database containing n datasets in which the complete cases remain the same, but the missing values differ within the range that was estimated by the prediction model. All these complete datasets can then be analyzed simultaneously using standard techniques (e.g., *t*-tests, ANOVA's) which generates n outcomes. These outcomes are automatically pooled into one outcome with one p-value; the final result of the analysis. Pooling these n datasets will give a mean of the n imputed values together with its standard error; the uncertainty of our estimation. MI is a robust method that produces valid and unbiased outcomes.[3,23] However, its use requires some training and should always be guided by an experienced user of the MI method, especially since there is still debate about what to do when data are MNAR. Sterne and colleagues provided clear guidelines on how to report the use of MI in scientific writing to improve reproducibility and increase transparency.[5]

Without any doubt, it would be best to prevent the appearance of missing data. Although almost inevitable, this can partly be achieved by thoroughly overthinking all steps of data-collection during the design of a new study. We would therefore strongly advise researchers to contact an epidemiologist or statistician prior to the start of a new study. Studies entirely devoted to the prevention of missing data provide useful tips such as the use of user-friendly case-report forms, the conduction of a pilot-study, and teaching of research assistants prior to the start of the study.[24-26] Even if data collection has already finished, contacting an epidemiologist or statistician can be very helpful to discuss the appearance of missing data and possible methods to handle missing data related issues, in order to assure valid outcomes.

CONCLUSION

With this article we want to draw attention to the importance of reporting missing data, and urge researchers to hypothesize about why data are missing. Defining why data is missing is essential in the process of selecting the most reliable technique to solve the missing data issue and prevent researchers from drawing invalid conclusion. We strongly suggest researchers to use available guidelines for reporting research (e.g., STROBE and CONSORT). To add, we highly recommend editorial boards of scientific journals to introduce the use of such checklists to increase their familiarity and ensure high reporting standards. To improve the quality of reporting, we would also like to encourage reviewers to pay attention to missing data and its possible consequences when reviewing articles for publication. As can be seen from this review, in the Otorhinolaryngology / Head & Neck surgery research field most often missing data are not reported and they are rarely handled properly. With this review, we hope to motivate researchers to think about missing data and to use methods such as multiple imputation to maximize the use of their data in order to draw more valid conclusions in future research.

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CHAPTER 3

EARLY IDENTIFICATION: **LANGUAGE SKILLS AND SOCIAL FUNCTIONING** IN DEAF AND HARD OF HEARING PRESCHOOL CHILDREN

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ABSTRACT

Objective

Permanent childhood hearing impairment often results in speech and language problems that are already apparent in early childhood. Past studies show a clear link between language skills and the child's social-emotional functioning. The aim of this study was to examine the level of language and communication skills after the introduction of early identification services and their relation with social functioning and behavioral problems in deaf and hard of hearing children.

Study design

Nationwide cross-sectional observation of a cohort of 85 early identified deaf and hard of hearing preschool children (aged 30-66 months).

Methods

Parents reported on their child's communicative abilities (MacArthur-Bates Communicative Development Inventory III), social functioning and appearance of behavioral problems (Strengths and Difficulties Questionnaire). Receptive and expressive language skills were measured using the Reynell Developmental Language Scale and the Schlichting Expressive Language Test, derived from the child's medical records.

Results

Language and communicative abilities of early identified deaf and hard of hearing children are not on a par with hearing peers. Compared to normative scores from hearing children, parents of deaf and hard of hearing children reported lower social functioning and more behavioral problems. Higher communicative abilities were related to better social functioning and less behavioral problems. No relation was found between the degree of hearing loss, age at amplification, uni- or bilateral amplification, mode of communication and social functioning and behavioral problems.

Conclusion

These results suggest that improving the communicative abilities of deaf and hard of hearing children could improve their social-emotional functioning.

INTRODUCTION

Permanent Childhood Hearing Impairment (PCHI) is a chronic handicap that affects approximately 1 to 1.3 out of every 1000 live births.[1,2] As a result of diminished auditory input, hearing impairment causes speech and language problems.[3-6] These problems can reduce the child's ability to communicate and to understand the refinements of social language.[7]

Extensive research in young hearing children has shown a clear relation between language delays and poor acquisition of social and emotional competencies which lead to problem behavior.[5,8-11] Both impaired language development and social-emotional problems are linked to poorer social skills and academic achievement, and fewer friendships.[8,11] Others have observed this link in deaf and hard of hearing (DHH) children.[12] Besides language problems, these children have also been shown to develop more social and emotional problems than hearing peers.[13-17] For example, DHH children experience a lower quality of life and more mental health problems such as anxiety, depression, and behavioral problems than their peers without hearing loss.[18-27] However, these studies were conducted before the introduction of early identification services.

Early identification and intervention programs have improved speech and language development in DHH children.[7,28,29] It is expected that these improvements also benefit the child's ability to communicate with others as the child becomes more able to express him or herself and to interact with peers. Yet, it remains unknown if this increased ability to communicate and participate in a sound-dominated world also benefits social functioning and prevents the development of behavioral problems. In this nationwide study, we examine the level of language and communication skills after the introduction of early identification services and their relation with social functioning and behavioral problems in DHH children.

METHODS

Procedure

This study was conducted as part of the large DECIBEL-study in the Netherlands.[2] DECIBEL is an acronym for Developmental Evaluation of Children: Impact and Benefits of Early hearing screening strategies Leiden. Its purpose was to define the effect of early identification and intervention services which were introduced in the Netherlands from 2002 compared to the previously used distraction screening method. The DECIBEL collaborative study group identified and evaluated all children with a positive screening result during either the Newborn Hearing Screening (NHS) or the distraction hearing screening in whom PCHI was confirmed at an audiological center after diagnostic testing. PCHI was defined as a hearing loss of 40 dB or more in the better ear. All children were

born in the Netherlands between January 2003 and December 2005. For the present study, only DHH children who had been identified by the NHS were included since this is regarded as standard care in Western society nowadays.
Between 2008 and 2010, parents of DHH children who were born after introduction of the NHS completed several questionnaires after informed consent was obtained. With their permission, audiological and medical records were checked for background information and hearing-loss-related outcomes such as the auditory thresholds, mode of rehabilitation and speech and language development. Permission for this study was granted by the Medical Ethics Committee of the Leiden University Medical Center.

Participants
During the introduction of the NHS from January 2003 till December 2005, 279 babies were identified and confirmed to have bilateral permanent childhood hearing impairment (PCHI).[30] All these children were invited to participate in our study. Parents of 98 children granted permission to participate and 85 of these completed the questionnaires. The final study sample consisted of 85 children with bilateral hearing loss; 47 boys and 38 girls. At the time of assessment, children were between the ages of 30 and 66 months old (mean age 46 months). The degree of hearing loss varied widely. Thirty-eight children (45%) experienced moderate losses (41-60 dB), 28 children (33%) experienced severe losses (61-90 dB) and 19 children (22%) were diagnosed with profound hearing loss (> 90 dB). Most children were equipped with conventional hearing aids (n = 61; 72%), 20 children (24%) were fitted with a cochlear implant (CI) of which 4 were bilaterally implanted. Three children were amplified with a bone conduction device (BCD). In one case, the child did not wear any form of hearing amplification anymore because of poor device acceptance due to psychomotor retardation. The majority of children communicated via spoken language (n = 37; 44%) or sign-supported language (n = 35; 41%). The remaining children either used sign language (n = 9; 10%) or an individually tailored form of communication using other senses, because of additional disabilities (n = 4; 5%). In the families of nine participating children, at least one of the parents was DHH. Two children were born to families in which both parents were DHH. Background information regarding the study sample can be found in Table 1.

Materials
Receptive and expressive spoken language
The Dutch translation of the 'verbal comprehension' scale of the Reynell Developmental Language Scale (RLDS) was used to determine receptive language skills. The Schlichting Expressive Language Test (SELT) measures vocabulary by means of the subtest 'word development' and syntax by means of the 'sentence development' subtest.[31] These tests are standardized oral language tests that are part of the clinical follow–up for children with PCHI in the Netherlands and were derived from the child's medical records. As a consequence, they were conducted at a different time and age of the child than when parents completed the questionnaires. Therefore, time of assessment varies considerably

in this study (mean difference between tests 7.0 months ± 10 months *SD*). However, age-equivalent scores which represent the language development of typically developing children are available. Both language tests provide a calculation tool to convert age-equivalent scores into normally-distributed standard scores.

Communicative development

Parents completed the MacArthur-Bates Communicative Development Inventory (CDI-III) to assess communicative language development (vocabulary and syntax) and understanding.[32,33] The first part of the questionnaire contains 100 words. Parents reported whether their child currently used these words in spoken language, sign-language, or both. The second part consists of 9 nine items, each containing three sentences of increasing length and difficulty. Parents reported the degree of complexity of sentence structure that their child produced, in spoken language, sign-language, or both. They were also requested to write down three sentences that their child recently produced. The total number of utterances was counted and the mean of these three sentences was calculated and named the Mean Length of Utterance (MLU). The third part consists of 12 questions (e.g., *"Does your child ask questions starting with the word "why"?"*) that parents answered on a 3-point Likert scale (0 = *"Not Yet",*1 = *"Sometimes",* 2 = *"Often"*) to measure language understanding (comprehension, semantics, and syntaxis). Parents of 11 children reported that their child was not yet able to connect words to create short sentences. Therefore, these parents did not complete section two and three (i.e., sentence complexity and understanding). The CDI-III was originally designed to measure communicative abilities in hearing children aged 30-37 months. However, research has shown that because of their language problems, the CDI-III is a useful measurement for DHH children with a CI within the age range 32-86 months.[34] However, age-appropriate percentile-scores are not available for the 38-86 months age range. Therefore, percentile-scores from hearing children between the ages of 36-37 months old were used to calculate percentile scores for children older than 37 months.

Social functioning and behavioral problems

Behavioral problems were identified with the *Strengths and Difficulties Questionnaire* (SDQ).[35] This parent report consists of 25 statements to be answered on a 3-point Likert scale (*"Not True", "Somewhat True", "Certainly True"*) and is used to screen for mental health problems in children. From these items, two scales were calculated: social functioning and behavioral problems.[36,37] The social functioning scale consists of five items concerning 'peer problems' (e.g., *"Picked on or bullied by other children"*) that were reverse scored and five items concerning prosocial behavior (e.g., *"Often offers to help others"*). The behavioral problems scale is constructed by combining the five items from the 'behavioral problems' scale (e.g., *"Often loses temper"*) with the five items from the 'hyperactivity' scale (e.g., *"Restless, overactive, cannot stay still for long"*). The fifth scale 'Emotional symptoms' was omitted from the analyses as this scale reflects behavior and feelings that were rarely reported by parents resulting in a very low reliability (Cronbach's

Table 1. Demographic characteristics of participants

	Total study population N = 85
Age at time of assessment	
Mean - in months (*SD*)	46 (10)
Range - in months	30-66
Gender (%)	
Male	47 (55)
Preferred mode of communication (%)	
Oral language only	37 (44)
Sign-supported Dutch	35 (41)
Sign language only	9 (10)
Other	4 (5)
Type of education (%)	
Mainstream education	21 (25)
Special education for the hearing impaired	51 (60)
Special education for developmental disabilities	6 (7)
Unknown	7 (8)
Degree of hearing loss - Low Fletcher Index (%)	
Moderate 41-60 dB	38 (45)
Severe 61-90 dB	28 (33)
Profound >90 dB	19 (22)
Hearing amplification type (%)	
Hearing Aid	61 (72)
Cochlear Implant	20 (24)
BCD	3 (3)
No adjustment	1 (1)
Age at diagnosis of hearing loss - in months (*SD*)	7 (11)
Age at first amplification - in months (*SD*)	14 (13)
Duration of amplification use - in months (*SD*)	31 (13)
Additional disabilities (%)	13 (16)
CI characteristics	
Age at implantation - in months (*SD*)	25 (14)
Duration of CI use - in months (*SD*)	18 (11)
Bilateral CI (%)	4 (5)

Abbreviations: BCD Bone Conduction Device, CI Cochlear Implant, HA Hearing Aid, SD Standard Deviation alpha = 0.51). Composite scores show good psychometric properties (Cronbach's alpha = 0.78 and 0.80 for social functioning and behavioral problems, respectively). To be able to interpret the outcomes of the SDQ, scores were compared with previously published norm-scores.[38] Psychometric properties of all tests can be found in Table 2.

Table 2. Psychometric properties

	No. of items	Answer range	Mean (SD)
Strengths and Difficulties Questionnaire			
Total difficulties	20	0-2	8.7 (5.2)
Social functioning	10	0-2	15.4 (3.6)
Behavioral problems	10	0-2	5.9 (4.0)
Language skills			
RDLS - verbal comprehension quotient	67		83.2
SELT - word development quotient	62		84.6
SELT - sentence development quotient	40		85.3
Communicative development inventory			
Total words known	100	0-1	54 (32)
Total words spoken	100	0-1	50 (35)
Total words signed	100	0-1	14 (19)
Total words bimodal	100	0-1	9 (16)
Sentence complexity	9	1-3	16.7 (7)
Sentence understanding	12	1-2	13.2 (7)
Mean Length of Utterance	3	0-∞	5.4 (2.4)

Abbreviations: SD Standard Deviation, RDLS Reynell Developmental Language Scale, SELT Schlichting Expressive Language Test
Note. Language skills are displayed as standard scores with a mean of 100 and a SD of 15.
For all communication skills, raw scores are reported.

Statistical analyses

Pearson's correlations between language scores and outcomes from the CDI-III were calculated to define the relation between receptive and expressive language skills and communicative development as reported by parents. Gender differences in behavioral problems were detected using Analysis of Covariance (ANCOVA) in order to control for covariates such as age and language skills. To examine risk and protective factors influencing behavioral problems, Pearson's correlations were carried out. Because multiple correlations were computed for the relation of communication and language skills with social functioning and behavioral problems, all *p*-values were adjusted using Bonferroni correction for multiple testing. Statistical analyses were carried out using *SPSS* version 21.0 (IBM Corp., Armonk, NY).

Multiple imputation of missing data

As a result of the study design, we were confronted with missing data. Language test results were derived from the children's medical files, and these scores were absent or untraceable in 23 (receptive language) and 30 (expressive language) cases. Many statistical methods for analyzing datasets assume complete cases. Consequently, these analyses remove incomplete cases beforehand, introducing bias and a drop in statistical power.[39] Therefore, the multiple imputation technique was used to handle this problem. This technique involves filling in the missing data based on known characteristics of the participant and the relations observed in the data for other participants with complete data.[40-42]

Little MCAR's test was significant for the language scores which meant that our data was not Missing Completely at Random (MCAR) but either Missing Not at Random (MNAR) or Missing at Random (MAR). The MAR condition assumes that the underlying reason for data being missing is related to other known characteristics of the participant.[39] In clinical practice, most often language scores are missing if children are not able to complete the test session because of low verbal language skills. In our sample, it was therefore expected that language test scores were missing because of the lower spoken language abilities of these children. This assumption was underlined by the fact that children with absent language test scores more often used sign-language and more often attended special schools for the DHH than children with complete language scores. We therefore assumed the data to be MAR and multiple imputations were used to handle the missing language scores. Research on this topic has shown that five imputations are seen as sufficient to create a good estimate for each entered data point.[40] We performed five imputations and analyzed the newly formed datasets using standard techniques (i.e., ANCOVA's and Pearson's correlations).

RESULTS

Language and communicative development

Language skills

Of all participants, language scores revealed that 47% scored one standard deviation below the mean or higher (quotient ≥ 85) on receptive language (M = 82.3). On expressive language, 57% (M = 85.1) and 56% (M = 86.2) scored one standard deviation below the mean or higher for word- and sentence development, respectively.

Communication skills

Outcomes of the parent report revealed that, compared to percentile-scores, 48 children (56%) scored one standard deviation below the mean or higher on the produced words scale of which nine children (10%) scored at ceiling. Concerning language complexity and understanding, 37 (44%) and 38 children (45%) scored one standard deviation below the mean or higher. No ceiling effect was found on these scales. The MLU of 19 of the 85 children (22%) was one standard deviation below the mean or higher, without any children scoring at ceiling.

Significant correlations were found between parent-reported communicative development and language test scores. Receptive and expressive language quotients positively related to the total words spoken by the child, MLU, sentence complexity and sentence understanding (Figure 1). Negative correlations were found between the child's spoken language scores and the total number of words signed (Table 3).

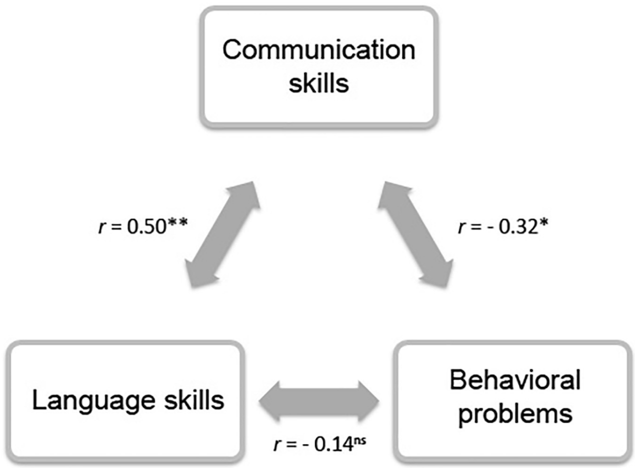

Figure 1. Pearson's correlations between language skills, communication skills and behavioral problems *$p < .01$, **$p < .001$, ns = non-significant result

Table 3. Pearson's correlations between language and communication scores

	4	5	6	7	8
1. Receptive language	.47**	-.24*	.48***	.53***	.50***
2. Expressive language sentence development	.52**	-.30**	.39**	.54**	.48***
3. Expressive language word development	.42**	-.25*	.51***	.57***	.52***
4. Words spoken		-.23*	.59***	.74***	.78***
5. Words signed			-.02	-.11	-.11
6. Mean Length of Utterance				.66***	.67***
7. Sentence complexity					.84***
8. Sentence understanding					

Note. * $p < .05$, ** $p < .01$, *** $p < .001$

Social functioning and behavioral problems

Compared to the norm-scores of the SDQ, the DHH children scored lower on social functioning $t(84) = -3.29$, $p < .001$, and higher on behavioral problems $t(84) = 2.09$, $p < .05$, regardless of gender. Pearson's correlations revealed that only the child's communicative abilities were related to the level of social functioning and behavioral problems. Higher (spoken) vocabulary was related to more social functioning and less behavioral problems. Lower sentence complexity, sentence understanding and shorter MLU were related to more behavioral problems (Table 4). In partial correlations that controlled for the age of the child, only the relation between communicative abilities and behavioral problems remained. No relation was found between the child's language skills and the level of social functioning or behavioral problems.

Table 4. Pearson's correlations between social development and communication scores

	Words spoken		Words signed		Mean Length of Utterance		Sentence complexity		Sentence understanding	
	r	Partial r	r	Partial r	r	Partial r	r	Partial r	r	Partial r
Social functioning	.26**	.25**	.08	.08	.17	.15	.14	.10	.22*	.23*
Behavioral problems	-.29**	-.27**	.00	.00	-.27*	-.23*	-.27**	-.23*	-.35**	-.32**
Total difficulties	-.29**	-.28**	.00	.00	-.21*	-.16	-.25*	-.22*	-.36**	-.34**

Note. The partial correlations were controlled for age. * p (one-tailed) < .05, ** p < .01

The influence of audiological and medical factors

Several audiological and demographic factors were entered in the correlation matrix to determine their relation to the reported levels of social functioning and behavioral problems: age at detection of hearing loss, age at first amplification, duration of amplification use, degree of hearing loss, level of maternal and paternal education, type of amplification, uni- or bilateral amplification, mode of communication, family support, speech and language therapy, and age. After Bonferroni's correction for multiple testing, no relations were found.

DISCUSSION

This nationwide study aimed to examine the level of language and communicative skills and their relation with socio-emotional functioning and the presence of behavioral problems in DHH children who received early detection services. The main findings showed that the language skills of the DHH children in this study were just within the normal range, but their communicative abilities were below average. A positive relation was found between children's communication skills in spoken language and their social functioning. Additionally, DHH children with lower communicative abilities showed behavioral problems more often.

Not surprisingly, children's level of spoken language and their communication skills were highly related in this study. Nevertheless, only communicative skills were related to children's social-emotional functioning, which emphasizes the importance of communication skills for social learning.[5,8] This can be explained by the concept of 'incidental learning': unplanned and unintended learning outside of educational settings.[43] Incidental learning is essential for social learning.[44] For example, social rules that are mostly implicit, are learned by observing and overhearing how others interact. Overhearing others can be challenging for DHH children for obvious reasons. Consequently, they miss frequent exposure to this type of social learning. It seems only reasonable that for incidental learning to succeed, this requires communication with others rather than an increase in passive vocabulary alone. Additionally, fewer communication skills could also impede children from expressing themselves, causing frustration and subsequently inducing behavioral difficulties.

It should be noted that the causal link between children's communication skills and their social-emotional functioning could be reciprocal. Good communication skills will enhance children's social functioning. In turn, lower levels of social functioning might discourage children from seeking contact with others, resulting in fewer communicative opportunities from which to benefit.

In our study, we found that parents are very capable of evaluating the speech- and language abilities of their DHH child by using the CDI-III. In line with standard language tests, parents of children with higher language skills also reported that their child was able to express longer and more complex sentences, and showed higher language understanding. These results are useful in clinical settings because language tests cannot be assessed too often due to learning and remembrance effects. Therefore, parent-reports are a useful tool to keep track of speech and language development in the meantime. Despite these promising findings we have to point out that the accuracy of parental reporting has previously been found to be influenced by the SES of the parents. In families with very low SES, the communicative abilities of the children were sometimes overestimated.[45] However, we did not find an effect of SES on the parental evaluation of the child's communication skills in this study.

In line with previous studies, the majority of children in our study sample did not show age adequate language skills although their group mean was within the normal range.[7,28,29] Despite an improvement of children's language skills after early detection and intervention services, DHH children's language levels are not yet on a par with their hearing peers, and the improvement was not sufficient to protect children from developing behavioral problems.[46] These language skills might further improve in later cohorts, because the children in our study did not always receive early intervention, despite the early detection. At the time that these children were detected, the early detection program by the NHS had just started and was still in the implementation phase during data collection.

During this period children with moderate losses received intervention relatively late due to various reasons such as lack of guidelines on reimbursement of costs.. Post-hoc analyses revealed that children with moderate losses received their first hearing aid at approximately 16 months of age whereas children with more severe losses received amplification at 12 months. Moreover, as the results of this study indicate, it might even be more favorable to focus on the development of children's communicative abilities instead of language skills only since these were related to the child's social functioning.
However, we have to note that our study sample comprised approximately one third of the total DHH cohort. It is possible that parents of children with additional handicaps or very low language skills decided not to participate, introducing a selection bias. It is also possible that parents of children who were developing well had no interest in participating in the study. For future research, it is desirable to identify reasons for non-responding.
Contrary to our hypothesis, we did not find a relation between the time of intervention and the level of social functioning or behavioral problems; neither between the age at intervention, types of support, or degree of hearing loss. Fellinger pointed this out by asking the question: "*What kind of evidence-based interventions need to follow UNHS in order to support families to actively foster the development of a strong identity and positive mental health of their child with PCHI, beyond the drive for 'normalization'?*".[47]
This question calls for longitudinal research designs with a detailed follow-up of DHH children in order to study treatment effects and causality.

CONCLUSION

The communicative abilities of early identified DHH children are not yet on a par with hearing peers. This study shows the important relationship between these skills and DHH children's social-emotional functioning. Future studies should focus on the causality of this relationship in order to improve these skills in DHH children and allow them to reach their full potential.

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CHAPTER 4

TERRIBLE TWOS OR EARLY SIGNS OF **PSYCHOPATHOLOGY**? **DEVELOPMENTAL PATTERNS** IN EARLY IDENTIFIED PRESCHOOLERS WITH **COCHLEAR IMPLANTS** COMPARED TO HEARING CONTROLS

In revision

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ABSTRACT

Objective

Cochlear implantation (CI) has dramatically improved the lives of children who are deaf or hard of hearing. However, little is known about its implications for preventing the development of psychiatric symptoms in this at risk population. This is the first longitudinal study to examine the early manifestation of emotional and behavioral disorders and their risk and protective factors in early identified preschoolers with cochlear implants compared to hearing peers.

Design

Participants were 74 children with cochlear implants and 190 hearing controls between one and five years old (mean age 3;8 years). Hearing loss was detected using the Newborn Hearing Screening in The Netherlands and Flanders. Parents completed the Early Childhood Inventory-4, a well-validated measure to evaluate the symptoms of DSM-IV-defined psychiatric disorders, during three consecutive years. Language scores were derived from the child's medical notes.

Results

Children with cochlear implants and hearing controls evidenced comparable levels of disruptive behavior and anxiety/depression (which increased with age in both groups). Greater proficiency in language skills was associated with lower levels of psychopathology. Early cochlear implantation and longer duration of cochlear implant use resulted in better language development. In turn, higher early language skills served as a protective factor against the development of disruptive behavior symptoms.

Conclusion

This longitudinal study uniquely shows that the improvement of language skills lowers the development of early signs of psychopathology. Early identification of hearing loss and cochlear implantation help children to improve their language skills.

INTRODUCTION

Extensive research has shown that children who are deaf or hard of hearing (DHH) experience higher levels of psychopathology than hearing peers.[1] Symptoms of psychopathology (e.g., anxiety, depression, aggression, and disruptive behavior) have mainly been studied in DHH school-aged children and adolescents, but research has clearly shown that DSM-IV-defined psychopathology can already be detected in preschoolers.[2] Moreover, the presence of such symptoms of social and emotional dysregulation in early childhood is a risk factor for future behavioral problems, peer rejection, and poor academic achievements.[3,4] For DHH children to reach their full potential, it is necessary to identify any signs of psychopathology as early in life as possible. Universal hearing screening programs have been introduced worldwide to identify hearing loss and start intervention as soon as possible.[5,6] Because early identification of hearing loss and cochlear implantation (CI) have especially improved the speech and language development of young DHH children, this could also benefit their mental health. This study aimed to examine the development of early signs of psychopathology in early identified DHH toddlers with CI compared to hearing controls and to identify risk and protective factors.

Early signs of psychopathology

Detecting early signs of psychopathology in toddlerhood can be challenging because parents often regard behavioral tantrums as being ‘part of the deal’ (the terrible twos). Young children experience an increased urge for autonomy; they want to do things on their own. However, their motor skills are not yet fully developed to do so, their language skills prevent them from clearly communicating their needs, and their relatively immature emotion regulation and coping skills prevent them from adequately regulating their own emotions. These skills improve with age, and (among other factors) this results in decreasing levels of disruptive behavior and lower chances of developing symptoms of anxiety and depression as children grow into school age.[7] However, if for any reason one of these skills cannot develop properly, this can result in higher levels of psychopathology.[8] Previous studies indicated that children with language problems have emotional and behavioral problems, that are not always identified because of the lack of knowledge in this area.[9]

Childhood psychopathology has major impact on society. Not only because children need extra care and support, but also because of the child’s future perspectives. Higher levels of disruptive interpersonal behavior, peer aggression and anxiety in childhood are strongly related to the development of depression and substance abuse in adulthood. The prevalence of disruptive disorders in early childhood (3-6 year) is around 11%.[10] The estimated prevalence of emotional disorders in preschool children is 3.9% for symptoms of anxiety and 1.3% for depressive symptoms.[10] Disruptive disorders in early childhood are linked to juvenile delinquency and criminal acts in adulthood, causing a serious burden for society.[8] Anxiety disorders are associated with all of the other major classes of disorders and have been proven to be a precursors for the development of depression in adulthood.[11,12] Therefore, it is of the utmost importance to detect symptoms as early as possible, in order to prevent the development of symptoms later on in life, especially in high-risk groups.

Challenges associated with pediatric hearing impairment

Hearing impairment can cause language and communication problems, which often interferes with DHH children's ability to actively participate in communication with others.[13] Diminished participation in social situations affects DHH children's opportunities for incidental learning. Incidental learning is learning by occasion, without the intention to learn. This mainly takes place outside of school settings, in everyday situations. Through observation of others, children constantly pick up different behaviors and responses and learn to replicate this at later occasions.[14] Incidental learning is one of the cornerstones for socialization. Therefore, less opportunity for incidental learning has major consequences for DHH children's social learning.[15]

The introduction of newborn hearing screening programs has enabled earlier identification and consequently earlier intervention of hearing loss in Western societies. This has been proven beneficial for the child's speech, language, and socio-emotional development[5,6], especially for children with severe to profound losses who received a CI at a young age. In fact, with early intervention and implantation, the speech and language skills of children with CI are almost comparable to those of hearing peers.[16,17] Because an improvement in language skills can benefit communicative abilities, early intervention of hearing loss can have great potential for the child's social development.[4]

Psychopathology in DHH children

An increasing interest in the development of DHH children in its broadest sense has led to numerous studies examining emotional and behavioral difficulties in DHH children. The overall results show that DHH children experience higher levels of anxiety, depression, somatic complaints, aggression, and behavioral problems than hearing children, and that they more often encounter problems in relationships with peers.[1,18] When focusing solely on DHH children with CI, the picture becomes a little brighter. School-aged children with CI are reported to show equal levels of depression, anxiety and behavioral problems as their hearing peers.[19-22] Yet, there is conflicting evidence. A large Spanish study found higher levels of behavioral problems in children with CI compared to a matched hearing group.[23] A smaller study by De Giacomo et al. found more peer problems and emotional symptoms in children with CI compared to age-matched controls.[24] The differences in these findings are likely related to whether or not the study included children who received implants relatively late and thus had less time to benefit from their CI. Yet, this is in line with the conclusions the aforementioned studies all draw; mainly children with CI who had lower language and communication skills were at risk for developing social, emotional, or behavioral problems.

Why do DHH children experience higher levels of psychopathology? One important aspect of hearing loss is that it is known to cause language delays. These language delays may prevent children from adequately communicating with others and expressing what they feel, want, and need, which can have ongoing consequences. Through conversations with others, children incidentally learn about abstract concepts such as emotions. Learning about (others') emotions helps them to understand how they feel and how to deal with their feelings; it helps children to regulate their own emotions. Emotion regulation (which includes both coping with emotions as well as emotion expression) has been proven to be problematic for DHH children with CI.[25] The young children with CI in this study used less adequate coping strategies than the hearing control group. Together with an impaired capacity for adequately communicating their needs, these lower coping skills can lead to internalization of problems resulting in withdrawal, somatic complaints, anxiety and symptoms of depression. The children with CI also expressed more negative emotions than their hearing peers which was related to more behavioral problems. Language and communication difficulties may thus hamper incidental learning which can result in higher levels of various forms of psychopathology.

Little research has been conducted regarding the effect of (early) implantation on the behavior of young children. A recent systematic review and meta-analysis stated it remained unclear what the effect of implantation is on the development of emotional and behavioral difficulties in young children with CI.[18] In addition, researchers call for longitudinal research to define causal relationships and to study the effect of age on the development of behavioral problems in children with CI.[1,4,15,18,26] To the best of our knowledge, no study has examined the effect of early intervention on the development of early signs of psychopathology in preschool children with CI.

Present study

The aim of this study was threefold. The first aim was to compare the level of disruptive behavior and anxiety/depression symptoms between early identified DHH toddlers and preschoolers with CI and age-related hearing peers. The second aim was to compare the developmental patterns of these symptoms over time in the two groups. The third aim was to identify risk and protective factors for the development of psychopathology in both young children with CI and hearing peers.

As a result of early identification and implantation and in line with previous findings, we expected to find equal levels of psychopathology in children with CI compared to hearing children.[1,6,27] Second, because higher language skills enable children to better express themselves we expected that higher language skills would serve as a protective factor against the development of psychopathology. Third, it was expected that younger age at identification and implantation would serve as a protective factor to lower the chances of developing behavioral problems.

METHODS

Participants

This study was conducted as part of a larger longitudinal research project concerning the socio-emotional development of toddlers and preschoolers with CI and with normal hearing. The sample was comprised of 190 hearing children and 74 children with bilateral severe to profound hearing loss wearing CIs.[27-29] All children were between 1 and 5 years of age (mean age 44 months) at the start of the study. Children were excluded from this study if they had any other known disability besides their hearing loss.

Hearing loss was detected using early identification programs in the Netherlands and the Dutch-speaking part of Belgium. As such, children were implanted before their 3rd birthday. Thirty-four children (46%) were implanted bilaterally. Characteristics of all participants are shown in Table 1.

Procedure

Children with CI were recruited from nine different hospitals and counseling services in the Netherlands and the Dutch-speaking part of Belgium. Hearing children were recruited from schools and daycare centers all over The Netherlands. After identification of participants, information about the study was sent to their parents or caregivers. Information regarding hearing loss and speech- and language abilities of children with CI was collected from medical notes after obtainment of informed consent. For three consecutive years, parents were annually requested to complete several questionnaires concerning the socio-emotional development of their child and a list of background variables. Permission for this study was granted by the Medical Ethics Committee of the Leiden University Medical Center.

Materials

Early signs of psychopathology

The Early Childhood Inventory-4 (ECI-4) is a parent-report questionnaire containing 108 items that is widely used to assess the symptoms of DSM-IV-defined emotional and behavioral disorders.[30] Anxiety and depression symptom severity was calculated by summing the scores for the Major Depressive Disorder (11 items), Social Phobia (3 items), and Generalized Anxiety Disorder (14 items) scales from the ECI-4. To measure disruptive behavior we summed the scores of three ECI-4 scales: Peer Conflict Scale (10 items), Oppositional Defiant Disorder (8 items), and Conduct Disorder (10 items). We combined scales because prior research with young children shows that specific symptoms within the same domain (e.g., major depressive disorder and generalized anxiety disorder) often cannot be distinguished from each other and possibly represent the same underlying disorder in toddlers.[2]

Table 1. Demographic characteristics of participants

	Total study population N = 264	
	CI	**Controls**
No. of children	74	190
Age		
Mean - in months (SD) *	39.8 (12.7)	45.6 (13.9)
Range - in months	18 - 61	18 - 66
Gender		
Male (%)	47 (63.5)	97 (51.1)
Female (%)	27 (36.5)	93 (48.9)
Socioeconomic Status† (SD)	4.4 (1.3)	4.7 (1.0)
Language Skills		
CDI - Language Comprehension (SD) **	30.0 (13.3)	40.4 (11.8)
CDI - Expressive Language (SD) **	31.2 (13.0)	43.1 (11.2)
RDLS - Receptive Language (SD)	84.8 (17.5)	
SELT - Expressive Language (SD)	82.7 (14.6)	
Preferred mode of communication		
Oral language only (%)	27 (36.5)	
Sign-supported Dutch (%)	40 (54.1)	
Sign language only (%)	7 (9.5)	
Age at first hearing aid acquisition - in months (SD)	6.9 (6.7)	
Age at implantation - in months (SD)	16.8 (7.2)	
Duration of CI use - in months (SD)	22.6 (12.0)	

†The highest level of education of each parent and their net household income were categorized on a scale ranging from zero to five. Socio-economic status (SES) was calculated by averaging these three scores.
Abbreviations: CDI Child Development Inventory, RDLS Reynell Developmental language Scales, SELT Schlichting Expressive Language Test, CI Cochlear Implant, SD Standard Deviation
* $p < 0.01$, ** $p < 0.001$

Speech and language skills

The Child Development Inventory (CDI) is an extensive 300 item parent questionnaire that creates an accurate representation of the child's development in several domains.[31] From this parent-report, two scales were used in this study. The 'Expressive Language' scale includes 50 items that define the child's expressive communication by use of vocals, gestures, and verbal behavior. Language understanding was measured by parents using 50 items that account for the 'Language Comprehension' scale. Language skills were assessed in Wave 1 and Wave 3 (Table 2).

Receptive and expressive spoken language skills were tested using the Reynell Developmental language Scales (RDLS) and the Schlichting Expressive Language Test (SELT), respectively.[32] These tests were administered as part of the post-implant rehabilitation program and therefore, scores are only available for the children with CI. Scores were derived from the child's medical notes and therefore only available at baseline. In 23 children, language scores were missing.

Table 2. Mean scores on psychopathology and language skills

	Baseline		Wave 2		Wave 3	
	CI	**Controls**	**CI**	**Controls**	**CI**	**Controls**
	n = 51	n = 159	n = 42	n = 109	n = 44	n = 77
ECI-4						
Anxiety / Depressive symptoms	0.15	0.16	0.18	0.18	0.20	0.19
Disruptive behavior	0.31	0.30	0.39	0.28	0.34	0.28
CDI						
Language Comprehension	30.0	40.4	na	na	44.0	48.7
Expressive Language	31.2	43.1	na	na	45.1	49.1

Abbreviations: CI Cochlear Implant, ECI Early Childhood Inventory, CDI Child Development Inventory, na not administered

Statistical analyses

The two groups (i.e., hearing and CI) were compared on demographic features using independent samples *t* tests for continuous variables and χ^2 test for dichotomous variables (Table 1). On average, children with CI were 5.8 months younger than the hearing children. We therefore decided to analyze our data using Linear Mixed Models (LMM) that allow us to correct for this difference in age. Assessment of model fit was evaluated using Akaike's information criterion. LMM were used to examine i.) differences in baseline levels of psychopathology, ii.) developmental changes of psychopathology over time, and between the two groups (hearing and CI), and iii.) risk and protective factors for the development of early signs of psychopathology over time. A p-value < 0.05 was considered statistically significant.

Multiple imputation of missing data

As almost inevitable in large longitudinal studies, we were confronted with missing data. Not all participants completed all three data waves and occasionally scores were missing from medical files for numerous reasons. Many statistical methods for analyzing datasets assume complete cases. Consequently, these analyses remove incomplete cases beforehand, introducing bias and a drop in statistical power.[33,34] To better deal with missing data, multiple imputations (MI) were used which involves filling in the missing data based on known characteristics of the participant and the relations observed in the data for other participants with complete data.[35-37] We were unable to find a pattern in the missing data and thus no relations between missing data and participant characteristics such as age of degree of hearing loss were found. Using the MI technique, missing scores on the CDI and ECI from 23 children with CI and 31 controls were imputed together with missing scores on the variables to calculate the SES of the family (level of parental education and net income) in 16 children with CI and 33 controls. The following variables were entered into the imputation model to estimate missing values: age, hearing status, language test scores, outcomes on the CDI and ECI, SES of the family and gender. We performed ten imputations and analyzed the newly formed datasets using standard analyzing techniques. Pooled results are reported. Only missing data at baseline were imputed because the LMM technique is robust enough to correct for missing follow-up data in a longitudinal design.[38]

RESULTS

Language development

To confirm the extensively studied positive effect of early identification and implantation on the language development of young DHH children [5,6,16,17] we were interested in the development of expressive language and language comprehension over time in our sample. A multilevel LMM with Language Comprehension as the dependent variables, Time as the determinant for repeated measurement and Age, Age at implantation, and Time as the fixed effects revealed that younger age at implantation and longer duration of implant use increased Language Comprehension over time ($t = -3.35$, $p < 0.001$ and $t = 3.80$, $p < 0.001$, respectively). A comparable effect was found for Expressive Language development: ($t = -3.58$, $p < 0.001$ and $t = 3.96$, $p < 0.001$ for Age at implantation and Duration of implant use, respectively). Children with bilateral CIs had higher receptive language skills at baseline than unilaterally implanted children, $t = -2.41$, $p < 0.05$. This difference was not significant for expressive language skills ($t = -1.99$, $p = 0.053$ and , $t = -1.91$, $p = 0.063$ for word development and sentence development, respectively). On average, bilateral users were implanted five months earlier than children with only one CI, $t = 3.15$, $p < 0.01$.

Development of symptoms over time

In order to evaluate the development of anxiety/depression symptoms over time, a multilevel LMM with Time as the determinant for repeated measurements and Age, SES, Group, Language Comprehension, and Time as fixed effects was performed. A main effect was found for Time ($t = -2.58$, $p < 0.01$), Language Comprehension ($t = -2.11$, $p < 0.05$) and Age ($t = 4.24$, $p < 0.001$). No difference was found between the two groups. A better model fit was accomplished by adding an Age*Time interaction term to the model ($t = -2.46$, $p < 0.05$). Figure 1 shows the raw data for all three waves without the imputed values. The outcome of the LMM described above was plotted in this same figure. This line (which is based on the imputed dataset) shows that the relation between age and the development of anxiety/depression symptoms can best be described by means of a parabola with a peak in symptoms at the age of 80 months. An increase in symptoms over time was seen in the younger children whereas a decrease of symptoms over time was found in children older than four at baseline.

A comparable model was run for the development of disruptive behavior symptoms and revealed an increase of symptoms with Age (t = 3.30, p < 0.001), which was qualified by an Age*Time interaction (t = -2.00, p < 0.05). No influence was found for Language Comprehension, Language Expression, Group or SES. The relation between age and the development of disruptive behavior symptoms can also be described by means of a parabola with a peak in symptoms at the age of 67 months. The raw data collected in all three waves is plotted in Figure 2. The plotted line is the function that can be derived from the LMM that is based on the imputed dataset.

Risk and protective factors for the development of symptoms

To identify risk and protective factors for the development of both anxiety/depression and disruptive behavior symptoms, the format of the database was changed into a long format with two time points instead of three (i.e., symptoms after one and two years). This way,

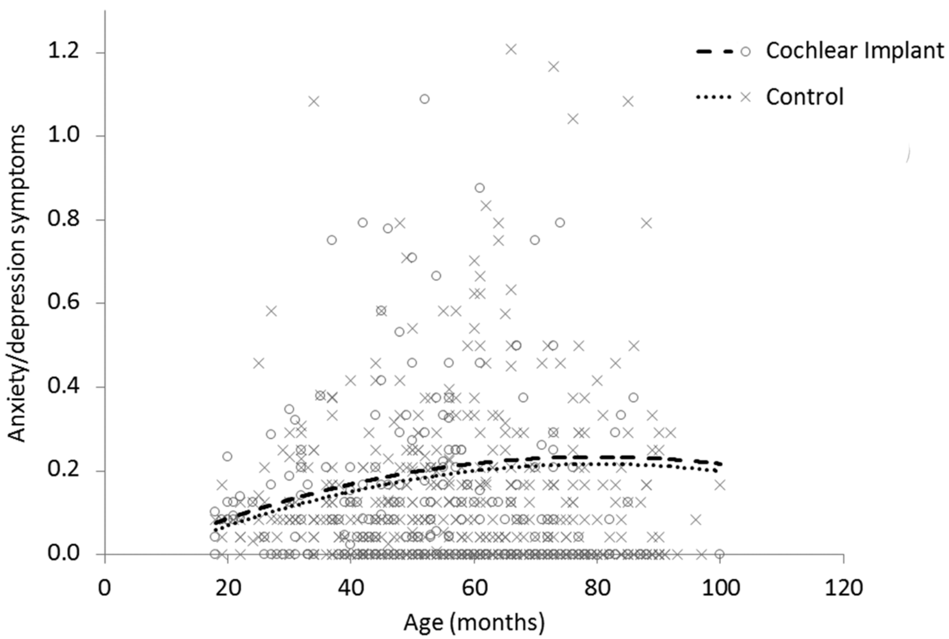

Figure 1. Development of anxiety/depression symptoms with age.
Outcomes on the anxiety/depression scale from the Early Childhood Inventory on all three waves (Y-axis) are plotted against the age of the child (X-axis) for children with CI (dots) and the control group (crosses). The lines describe the mean development of symptoms as age increases. No significant difference between the two groups was found.

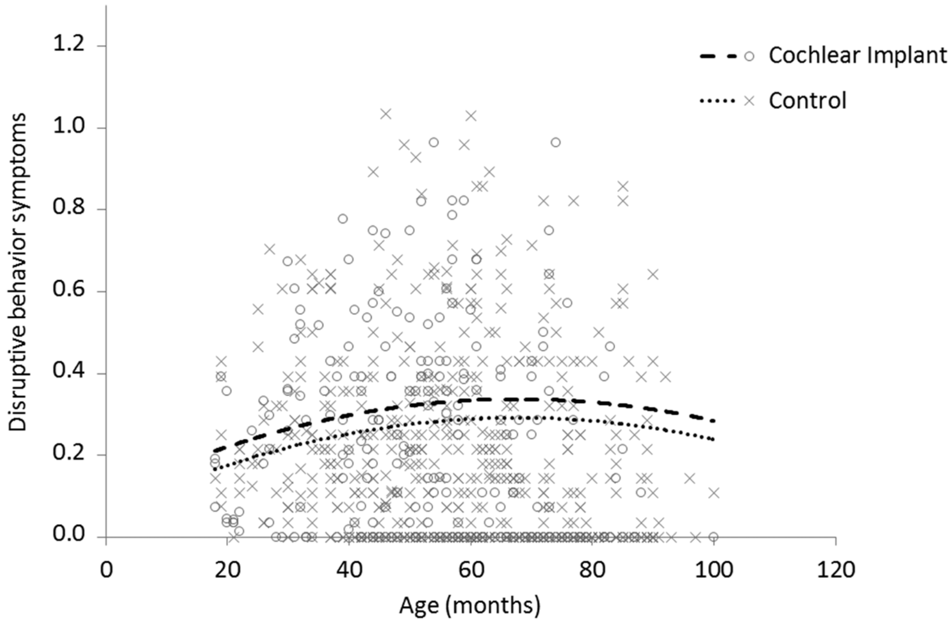

Figure 2. Development of disruptive behavior symptoms with age.
Outcomes on the disruptive behavior scale from the Early Childhood Inventory on all three waves (Y-axis) are plotted against the age of the child (X-axis) for children with CI (dots) and the control group (crosses). The lines describe the mean development of symptoms as age increases. No significant difference between the two groups was found.

the baseline levels of symptoms (measured at Wave 1) could be entered as covariates into the model to identify other unique predictors of psychopathology besides the level of symptoms at baseline. A LMM with anxiety/depression symptoms as the dependent variable and Group, Gender, Age, SES, Language Comprehension, Language Expression, and baseline level of anxiety/depression symptoms as fixed effects revealed a highly significant positive effect for baseline level of anxiety/depression symptoms only ($t = 9.14$, $p < 0.001$). Children that scored high at baseline remained to score high after two years.

A similar LMM with disruptive behavior revealed a positive effect for baseline level of disruptive behavior symptoms ($t = 7.53$, $p < 0.001$) and Age ($t = 1.97$, $p < 0.05$). For Language Comprehension and Expressive Language, a highly significant negative effect was found ($t = -2.87$, $p < 0.01$ and $t = -3.24$, $p < 0.001$, respectively). Higher levels of Language Comprehension and Expressive Language contributed to the prevention of the development of disruptive behavior, regardless of their hearing status.

The influence of CI on the development of symptoms

To identify the effect of several CI-related variables on the development of early signs of psychopathology, factors such as Age at implantation and Duration of implant use were entered into a LMM with Age and Time as fixed effects. No effect was found for uni- or bilateral implantation or communication mode. No direct influence was found for the Age at first amplification, Age at implantation, or the Duration of CI usage. From the implanted children, spoken language test scores were available at baseline. Baseline spoken language scores had no influence on the development of symptoms.

DISCUSSION

This longitudinal study is the first to compare the developmental pattern of early signs of psychopathology between preschool children with CI and hearing controls. In line with our hypotheses and previous studies, preschoolers with CI showed levels of psychiatric symptoms equal to hearing peers.[1,6,27,39] Moreover, the developmental patterns of these symptoms were comparable for both groups. Symptoms of psychopathology increased with age, and higher language skills contributed to the prevention of disruptive behavior symptoms but not anxiety/depression symptoms in both groups. Within the group of children with CI, a younger age at implantation and longer duration of implant use led to an increased language development.

An increase in language skills over time led to a decrease of symptoms of depression and anxiety. This finding is in line with previous studies in typically developing children.[40] As children grow older, the levels of their language and communication skills are of increasing importance in determining the amount of social interaction they have access to. From these social interactions, children may learn new words and meanings and so the relationship between language and symptoms of anxiety and depression is reciprocal. Unfortunately, this reciprocity can also work in a negative way. When children encounter language delays, this may result in withdrawal from interactions. This in turn lowers their social interaction time which may result in loneliness, lower self-esteem and feelings of depression and social anxiety.[4]

Higher language skills at a young age help to protect against the development of early signs of disruptive behavior in our study. This can be explained by the fact that early language skills help children to communicate their needs and wishes. When children are less able to make themselves understandable to others, this causes frustration, resulting in higher levels of aggressive and disruptive behavior.[40]

This study shows that early identification and intervention of hearing loss through CI results in better language skills, as previously established in other studies.[6,16,17] The longitudinal design of this study uniquely adds insight into the protective effect of language skills on the development of early signs of psychopathology. A younger age at implantation increased the language skills of children with CI over time. Through improving language skills, an indirect effect of early intervention on the development of psychopathology was shown. These findings support our hypothesis on the importance of language development for children's social-emotional development.

Possibly, the limited spread in both the age at implantation and the severity of symptoms prevents us from finding a direct relationship between age at implantation and levels of psychopathology. Furthermore, it is plausible that other factors that we did not measure in this study influenced the level of symptoms such as cognitive development, maternal sensitivity or parenting styles.[41] However, the absence of a direct causal relationship between age at intervention and the level of psychosocial development is consistent with previous literature.[39,42]

A factor that could have biased our results is the etiology of hearing loss in this group. Hearing loss due to meningitis for instance requires quick action from professionals as it is known to cause rapid ossification of the cochlea.[43] Children suffering from severe hearing loss after meningitis are therefore often implanted as soon as possible and also bilateral when possible, since waiting may decrease the chances for successful implantation and subsequent functioning of the implant. This is reflected in our data. The cause of hearing loss was more often found to be due to meningitis in the bilaterally implanted children than in the group of unilaterally implanted children. All nine bilaterally implanted children with a history of meningitis received their implants simultaneously. In addition, the bilaterally implanted children received their implants earlier than unilaterally implanted children. The question now is, how did this affect our data? Children who suffered from meningitis most likely had normal hearing prior to infection. This implies that the auditory cortex in the brain has been susceptible to auditory stimulation. Restoring auditory stimulation by CI may thus yield very good results in these children. On the other hand, it is also known that meningitis does not only affect the cochlea. It may also damage the auditory nerve and cause other neurological deficits which may result in lower hearing abilities and subsequent language development.[44] From this study, it therefore remains unclear what the role of bilateral implantation is on the development of language and psychosocial skills, which is a limitation of this study. Yet, the superior language skills that were found in the bilaterally implanted children when compared to unilaterally implanted children in this study are in line with previous research.[45,46] Future studies should address this important aspect. In addition, such studies should also include information regarding the type of amplification of the contralateral ear in unilaterally implanted children when comparing unilaterally and bilaterally implanted children.

Regardless of the child's hearing status, early signs of psychopathology increased with age with a peak in disruptive behavior symptoms around the age of five years and for anxiety/depression symptoms at approximately six years of age. In line with previous studies, both symptoms decreased in intensity afterwards.[47] This can be explained by the fact that by the age of six, children go to school, participate in sports and more frequently take part in social situations. Through incidental learning and by trial and error, they learn how to respond in different social environments. Among other things, the development of language skills allows young children to increasingly express their selves and engage in communication with others.

CONCLUSION

To the best of our knowledge, this is the first longitudinal study that examines the development of early signs of psychopathology in DHH preschoolers with CI compared to hearing age-related peers. The results of this study shed new light on the development of early implanted children. The longitudinal design shows the clinical importance of early intervention on the development of language skills. It underlines the important effect of language development on the psychosocial functioning of DHH children.

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CHAPTER 5

CAN YOU HEAR WHAT I THINK? **THEORY OF MIND** IN YOUNG CHILDREN WITH **MODERATE HEARING LOSS**

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ABSTRACT

Objectives

The first aim of this study was to examine various aspects of Theory of Mind (ToM) development in young children with moderate hearing loss (MHL) compared to hearing peers. The second aim was to examine the relation between language abilities and ToM in both groups. The third aim was to compare the sequence of ToM development between children with moderate hearing loss and hearing peers.

Design

Forty-four children between 3 and 5 years old with moderate hearing loss (35-70 dB HL) who preferred to use spoken language were identified from a nationwide study on hearing loss in young children. These children were compared to 101 hearing peers. Children were observed during several tasks to measure intention understanding, the acknowledgement of the other's desires and belief understanding. Parents completed two scales of the Child Development Inventory (CDI) to assess expressive language and language comprehension in all participants. Objective language test scores were available from the medical files of children with MHL.

Results

Children with moderate hearing loss showed comparable levels of intention understanding but lower levels of both desire and belief understanding than hearing peers. Parents reported lower language abilities in children with MHL compared to hearing peers. Yet, the language levels of children with MHL were within the average range compared to test normative samples. A stronger relation between language and ToM was found in the hearing children than in children with MHL. The expected developmental sequence of Theory of Mind skills was divergent in approximately one fourth of children with moderate hearing loss, when compared to hearing children.

Conclusion

Children with moderate hearing loss have more difficulty in their ToM reasoning than hearing peers, despite the fact that their language abilities lie within the average range compared to test normative samples.

INTRODUCTION

Engagement in social interactions is essential for the social-emotional development of children. In order to induce and maintain relationships, children need to learn that different people have different intentions, desires, and beliefs. The ability to apply such mental states to others is known as 'Theory of Mind' (ToM). Through ToM development, children will start to understand that our mental states explain our actions (e.g., dad chooses coffee for dessert because *he* prefers coffee over ice-cream). ToM development has been studied extensively over the last two decades.[1,2] These studies revealed that both language and communicative abilities are very important for an adequate ToM development (see Stanzione & Schick[3] for a review). The importance of this relation has been illustrated previously by many studies in deaf children of hearing parents. Outcomes show severe delays in the ToM development of deaf children of hearing parents[4-7] that may continue to be problematic during adolescence.[8,9] One explanation offered in the literature for these findings lies in the reduced abilities of parents (especially hearing parents who sign) to discuss abstract concepts such as thoughts and emotions compared to hearing-haring dyads.[10] Children with moderate hearing loss (MHL) share the same mode of communication as their hearing parents. However, these children often still encounter language difficulties.[11-13] Therefore, children with MHL are also potentially at risk for inadequate ToM development. Nevertheless, until now, no research has focused on the development of ToM in children with moderate hearing loss, which is the aim of this study.

Children with moderate hearing loss

A substantial number of children have hearing loss thresholds falling in the moderate range (40-70 dB HL). When wearing their hearing aids, children with MHL can function reasonably well in quiet areas and in one-on-one conversations. They can hear what is said when they are not disturbed by background noise that interferes with their hearing aids, their ability to recognize consonants, and directional hearing.[14,15] However, the hearing capacities of these children are frequently overestimated. Children with MHL frequently encounter difficulties in fully understanding what is said in daily interactions, especially in noisy environments such as daycare centers and classrooms.[16] Children with hearing loss encounter difficulties in speech perception when listening to speech in noise.[17] Furthermore, the children's hearing aids (HAs) are often not fit optimally, which may negatively impact their hearing potential.[11,18] For the child's surroundings, it is often difficult to understand what a child with MHL does hear and what input is missed. Diminished access to social conversations could potentially diminish their opportunities for social learning, which has ongoing consequences for their social-emotional development.

ToM development and hearing loss

In studies on ToM development, the majority of research has focused on only one aspect of ToM development, that is, the understanding of (false) beliefs. Yet, Wellman and others emphasize on the importance of studying ToM in its broadest sense. Thereby, it is important to be aware of the fact that the acknowledgement of others' intentions and desires precedes the understanding of others' (false) beliefs.[19] This was previously demonstrated in large studies examining the developmental sequence of ToM development in deaf children and children with an autism spectrum disorder. These studies show that deaf children generally show the same sequential pattern of ToM development as hearing peers, albeit slower.[20,21] This delayed ToM development can have ongoing consequences for a child's social development.[22,23]

Intention understanding

An essential precursor for the development of ToM is the ability to acknowledge others' intentions.[24] Growing consciousness of the fact that others' actions are guided by their intentions teaches children to separate human beings from objects. Only by knowing someone else's intentions, one can understand the person's actions. To illustrate, the physical movement of an object from one person to the other can be interpreted as giving, sharing, loaning, returning, or trading something. Yet, without intention understanding, we do not know why actions happen. In typically developing children, intention understanding begins to emerge in the second year of life.[25] An important aspect of intention understanding is joint attention; the ability to share attention with someone else concerning an object or situation. Drawing someone's attention to a certain situation increases language development and strengthens relationships. Studies in young children show equal levels of joint attention in deaf children with CI compared to age-related peers, whereas less engagement in joint attention was seen in deaf children without a CI.[26-28]

Desire understanding

The next important step in ToM development is the ability to acknowledge others' desires and to be able to distinguish between one's own and the other's desires. Desire understanding gradually takes place after a child's third birthday.[29] Abstract concepts such as taste allow children to understand the subjectivity of desires. For example, a child needs to learn to understand that dad does not like to eat cheese whereas the child herself really likes a cheese sandwich. Research on desire understanding in deaf children can be extracted from the work by Peterson[20,21,30,31] and Remmel[32,33] who found no difference in desire understanding when comparing school-aged deaf children (with and without CI) to hearing preschoolers. Only one study compared preschoolers with CI to age-related hearing peers. When focusing on children with sufficient language comprehension, children with CI were able to appreciate the protagonist's desire when it matched their own desire. Yet, they were outperformed by their hearing peers when the protagonist in the vignette had a dissimilar desire.[26]

Belief understanding

Classic false belief tasks include the change-of-location and the unexpected-content task. In both tasks, the child is questioned about the behavior of a story character. In the story, this character holds a belief that opposes the actual truth. Around the age of four, children start to appreciate other's beliefs. Research shows equal levels of belief understanding in deaf children born to deaf parents compared to hearing peers born to hearing parents (see Stanzione and Schick for an overview).[3,6] However, deaf children of hearing parents performed lower on false belief understanding than hearing children, with so-called late signers showing the least favorable results.[4,21] This difference can be explained by the quality and quantity of communication. Both deaf children who acquire oral communication and deaf children who acquire sign language relatively late (because it is their second language) may encounter limited participation in high-quality social interactions involving mental state talk, be it in school or with their family at home.[34-36]

A limited number of studies on false belief understanding in deaf and hard of hearing children compared to hearing controls found no differences in ToM abilities. However, in these studies children were much older than the control group, making the groups difficult to compare.[20,21,32,33,37] Since the introduction of early identification of hearing loss and early cochlear implantation, results have changed. Because of early implantation, young children with CI had relatively better language skills. These improved language skills enabled them to join in conversations more often which could potentially stimulate their ToM skills. Consequently, studies started to compare children with CI to age-related peers. Yet, these studies in young children still found lower levels of belief understanding in preschoolers with CI as compared to hearing peers.[26,38]

ToM and language

The relation between ToM and language abilities has been studied extensively. A meta-analysis examining this relation reported a strong relation between the two indices.[39] Since there has been an ongoing debate regarding the direction of causality between language and ToM development, this was one of the aims of this meta-analysis. Even though a bidirectional relationship was found in longitudinal studies (i.e., early language predicted later ToM development and early ToM skills predicted later language development), the relation reporting early language skills to be beneficial for later ToM development was significantly stronger than vice versa. However, this review only included studies that examined this relation in typically developing children.

In DHH children, the relation between language and ToM skills seems complex. False belief tasks for instance contain 'mental state verbs' and 'if/then statements'. In order to understand such complex ToM tasks, a certain level of language and communication skills is needed to succeed. As a result it is often unclear what it is exactly that such tasks are measuring: the child's ToM skills or their language capacities. Schick et al. therefore used ToM tasks that required minimal language skills to measure ToM abilities in deaf children of hearing parents. Results showed that the deaf children in their study also performed lower on the low-verbal tasks compared to hearing children and deaf children of deaf parents, indicating the importance of access to communication with others. This statement was underlined by the fact that complement processing skills (i.e., the ability to give meaning to a sentence or statement) were found to predict performance on low-verbal ToM tasks, yet vocabulary comprehension skills did not.[6]

The language skills of young children with MHL have recently been studied thoroughly by Tomblin and colleagues. Their study showed that the language skills of children with MHL were, on average, approximately 1 standard deviation lower than the language skills of hearing children. This may have been caused by their reduced ability to fully capture what is said in daily conversations. Missing out on the subtleties and nuances of communication may interfere with their capacity to understand what people mean to achieve when communicating to others. Subsequently, these difficulties can interfere with the development of adequate ToM skills.

Present study

The first aim of this study was to examine ToM abilities and its precursors in children with MHL compared to hearing children. Although children with MHL and their hearing caregivers share the same mode of communication (i.e. spoken language), it is also known that parents of children with hearing loss use less mental state talk in their conversations with their child.[40] Additionally, due to various reasons children with MHL often still encounter (mild) language and communication problems.[11] These difficulties could prevent them from fully benefiting from social interaction and incidental learning about others' intentions, desires, and beliefs. We therefore hypothesized that children with MHL of hearing parents would have lower ToM skills than hearing children. The second aim of this study was to define the relation between language skills and the development of ToM in children with MHL and in hearing controls separately. We expected language skills to be positively related to both desire and belief understanding because a certain level of language is needed to develop these skills. We expected no difference in the strength of this relation between the two groups. The third aim of this study was to evaluate the developmental sequence of various ToM concepts both in children with and without MHL. Because of language difficulties, we expected a delayed but not qualitatively different development of ToM in children with MHL compared to peers with normal hearing.

METHODS

Procedure

The children with MHL in this study were identified through the DECIBEL-study. DECIBEL stands for Developmental Evaluation of Children: Impact and Benefits of Early hearing screening strategies Leiden. The DECIBEL-study was conducted in The Netherlands between 2008 and 2010 to define the influence of early detection of hearing loss on the development of young DHH children. This nationwide study identified all children who were born with hearing loss between January 2003 and December 2005. Hearing loss was detected using Ototacoustic Emissions (OAEs) which enables identification of hearing loss of 35 dB HL or more. The database consisted of 210 children with permanent bilateral hearing loss. Ethical approval for the DECIBEL-study was obtained through the Medical Ethics Committee of the Leiden University Medical Center.[41,42]

For participation in the social-emotional assessments of the DECIBEL-study, children needed to fulfill additional inclusion criteria. Children needed to be at least 36 months old, their unaided hearing loss in the better ear should not exceed 70 dB HL, children had to use conventional hearing aids or bone conduction devices (BCD) and it was requested that their preferred mode of communication was either spoken, or sign-supported Dutch. This resulted in 74 children who were eligible for participation, and their parents were invited to participate. Finally, parents of 44 children gave informed consent (response rate 59.5%). Children were visited at home. A researcher sat with the child in a quiet room and conducted several tasks which will be explained in more detail below. The session was video-recorded in order to score the child's behavior afterwards. The camera was positioned so that both the experimenter and the child were recorded. This allowed for both observation of the child's behavior as well as to check if all tasks were correctly performed by the experimenter. The sequence of the tasks and observations that were performed was as follows: 1) Intention understanding, 2) False belief task, 3) Similar desire task, 4) Imperative pointing, 5) Dissimilar desire task, 6) *other tasks and observations not mentioned in this study*, 7) Dissimilar desire task, 8) Declarative pointing, 9) *other tasks and observations not mentioned in this study*, 10) Similar desire task. Completion of the whole set of tasks and observations took approximately 35 to 45 minutes per child.
Parents were requested to complete several questionnaires to gain background information. Medical history and language scores were derived from the child's medical files. A control group of hearing children was collected as part of another nationwide study. These children were previously described by Ketelaar et al. and were recruited from all over the Netherlands through mainstream primary schools and daycare centers.[26] From this large control sample, we were able to compose a subsample of 101 hearing children with a comparable age and sex distribution. Parents of children in the control group reported no history of hearing loss in their child.

Participants
All children were between 40 and 70 months old during home observations (mean age 57 months). Of the 44 children with MHL, 27 were boys (61.4%). Their hearing loss varied with a pure-tone-average between 35 and 70 dB HL in the better ear (mean loss 50 dB HL). Residual hearing was calculated by averaging unaided hearing thresholds at 500, 1000 and 2000 Hz. Six children had a hearing loss between 35 and 40 dB. One child used a BCD, all others used hearing aids. All but one were aided bilaterally. All children understood spoken language, yet five of them (11.4%) preferred to use sign-supported Dutch. Parents of seven children with MHL reported having hearing loss themselves. Three children with MHL had an additional handicap. One child was diagnosed with Turner

syndrome, one child suffered from muscle-tone dysregulation and the third child had a mild hypotonic hemiparesis. These three children did not differ from the rest of the MHL group in age, language skills, or on any of the ToM tasks. The control group consisted of 101 children with normal hearing, 55 were boys (54.5%). Demographic characteristics of both groups are listed in Table 1.

Table 1. Demographic characteristics of participants

Total sample N= 145	**MHL**	**Controls**
	n = 44	**n = 101**
Age - in years (SD)	4.8 (0.8)	4.8 (0.8)
Range - in months	40-69	40-70
Gender, No (%)		
Male	27 (61.4%)	55 (54.5%)
Female	17 (38.6%)	46 (45.5%)
Maternal Education (SD) †	3.2 (0.7)	3.4 (1.0)
Language (SD)		
CDI - Expressive language ‡	45.2 (5.7)	48.9 (1.7)**
CDI - Language Comprehension ‡	43.5 (6.5)	46.8 (2.8)**
Reynell Developmental Language Scales	n = 37	
Language Comprehension Quotient (SD)	92.49 (13.12)	
Schlichting Expressive Language Test	n = 34	
Word Quotient (SD)	94.85 (16.31)	
Sentence Quotient (SD)	94.35 (11.24)	
Age at detection - in months (SD)	17.1 (17.4)	
Range - in months	0-54	
Degree of hearing loss - in dB HL (SD)	50 (9)	
Range - in dB HL	35-70	
Age at first amplification - in months (SD)	26.4 (18.2)	
Device, No (%)		
Hearing Aid	43 (97.7%)	
BCD	1 (2.3%)	
Preferred mode of communication, No. (%)		
Oral language only	39 (88.6%)	
Sign-supported Dutch	5 (11.4%)	

Abbreviations: MHL Moderate Hearing Loss, SD Standard Deviation, CDI Child Development Inventory, BCD Bone Conduction Device. * $p< 0.01$, ** $p< 0.001$
† Categories: 0 = don't know, 1 =no education/primary education, 2 = lower general secondary education, 3 = higher general secondary education, 4 = college/university ‡ Raw scores

MATERIALS

Intention understanding

Comprehension of other people's intentions was measured using three tasks. The 'Intention Understanding task' based on the design of Meltzoff[43] and adapted by Ketelaar et al.[26] was used to define whether children understand others' intentions when trying to achieve a certain goal, even if the person is unable to succeed. To illustrate this, in one of these tasks the researcher attempts to put a string of beads in a cup. After failing to get the string in the cup, she hands it over to the child. Children succeed if they put the string of beads in the cup. With each task (trying to stack two cups and fitting a tube in a slightly bigger one) the researcher makes three attempts before handing the task to the child. This results in a maximum score of three if all intentions are understood correctly.

The 'Declarative Comprehension task' measures joint attention.[26,44] During this task, the researcher acts surprised and points to an object out of sight of the child. The researcher then looks back and forth between the object and the child. The subsequent behavior of the child was observed and children could receive up to three points when they looked at the object, looked at the researcher and, attempted to communicate about the object.

The third task to measure intention understanding was the 'Imperative Comprehension task'.[26,44] This task starts with the researcher pointing towards an object that is within reach of the child but not of the researcher. After pointing towards the object, the researcher holds up her hand with the palm facing up to request the object. The child succeeds if he or she actively responds to this gesture either by handing over the object or refusing to do so (e.g., saying no, shaking his/her head). Three points were awarded if the child succeeded the first time. If not, up to two additional attempts were performed between the other tasks and the score decreased by one point each time until a score of zero was attained after three unsuccessful attempts.

Desire understanding

The acknowledgement of others' desires was assessed using the 'desire task'.[26] This task uses vignettes to measures two types of desires: similar and dissimilar desires. In the similar desire condition, the child is presented with a picture showing two types of food (e.g., tomato and ice-cream). The child is asked what he or she prefers to eat. The researcher then tells a story about a boy who also likes the food that the child just chose. Then the child is asked: "Now the boy can choose a snack. What will the boy choose to eat?" This question is followed by two control questions: "Does the boy like [Snack 1]?" and "Does the boy like [Snack 2]?" The child is awarded one point if he or she answers all three questions correctly. In the dissimilar desire task, the only difference is that the protagonist in the story does not like the snack that the child preferred but instead likes the opposite snack.

Belief understanding

Belief understanding was measured using an adapted version of the traditional false-belief Sally-Anne task by Baron-Cohen.[26,45] In this task, the child sees a drawing of a boy playing with his model airplane. The boy hides his plane and leaves the scene. When the boy is away, a girl grabs the plane and hides it in a different location. On the next drawing the boy returns and the child is asked: "Where will the boy look for his plane?". This question is followed by two additional questions to check comprehension: "Where did the boy hide his plane before he went away?" and "Where is the plane now?". One point was awarded only if the child was able to answer all three questions correctly. All tasks mentioned above have previously been used in different clinical groups with good reliability.[26,46]

Language

In order not to interfere with the regular evaluations of the child's speech- and language therapists, test scores were derived from the child's medical files. Therefore, language scores were not available from the hearing children. Receptive language abilities were assessed with the *verbal comprehension* scale of the Dutch version of the Reynell Developmental Language Scale (RLDS).[47] The *word development* and *sentence development* scales of the Dutch version of the Schlichting Expressive Language Test (SELT) were used to assess expressive language abilities. These language tests are used throughout The Netherlands to assess language development, especially in high-risk groups. Raw scores are standardized according to age using quotients in which the population mean in hearing children is 100 with a standard deviation of 15. Language quotients within one standard deviation from the mean are considered to be in the normal range (85-115).

Parent-reported language skills

Two scales of the Child Development Inventory (CDI) were used to assess language skills in all participants.[48] Parents completed 50 items that together represent the *Expressive Language* scale and measures expressive communication ranging from simple gestures and words to complex language (e.g., *Asks questions beginning with "what" or "where"*). The *Language Comprehension* scale also consists of 50 items and relates to the understanding of simple instructions to the understanding of complex concepts (e.g., *Understands the meaning of at least six location words, such as "in, on, under, beside, top, bottom, above, below"*).

Statistical analyses

To assess differences between the two groups on ToM abilities and precursors (mixed design) analyses of covariance (ANCOVA's) were used to test both between-group and repeated-measures variables. Because the outcome on the False Belief task was dichotomous (i.e., pass or not) logistic regression was used to predict the effect of group and age on belief understanding. Pearsons' correlations and partial correlations were used to identify the relation between ToM skills and language abilities, taking the age of the child into account. Fisher *r*-to-z transformations were used to compare if the correlation coefficients differed between children with MHL and hearing controls.

To define whether ToM development evolved in the same manner in both children with MHL and in hearing children, participants were grouped into four stages of increasing ability to successfully complete the desire and belief tasks. [20,21,49] Because the Desire tasks each consisted of two vignettes, children needed to pass both tasks successfully in order to pass for this stage. ToM-Stage 1 was assigned when the child was unable to successfully complete any of the desire or belief tasks. Successful acknowledgement of similar desires resulted in assignment of the child to Stage 2. Stage 3 was assigned when a child also managed to acknowledge dissimilar desires. If a child mastered all ToM skills he or she was assigned to Stage 4. When other patterns were shown by the children, these were categorized as divergent. Categories were compared using the likelihood ratio test because some categories contained fewer than 5 participants.

Missing data

In the group of children with MHL, verbal comprehension scores were missing from 7 participants, word development scores were missing from 11 participants and sentence development scores were missing from 10 children. When conducting standard analyses such as ANCOVA's and Pearson's correlations, incomplete cases are automatically excluded from the analyses. Excluding these participants might give bias and would lower the power of our results. Therefore, missing language scores on the RLDS and the SELT were reconstructed using multiple imputations. This technique estimates a prediction model based on the complete cases and uses this model to predict outcomes of missing scores.[50-54] Language scores were predicted using the child's age, language skills as reported by their parents (CDI), and observations during the ToM tasks. Ten imputations were performed because research has shown that this is a sufficient number to make a robust estimation of each unique data point.[53,54] Statistical analyses were carried out using the program *SPSS* version 23.0.[55] One child with MHL refused to answer the dissimilar desire task. In analyses concerning desire understanding, this participant was excluded. Because of low language skills, one child was not able to perform the desire and false belief understanding task. This child was excluded in analysis that included these variables.

RESULTS

Intention understanding

The mean scores on outcomes of all observations are shown in Table 2. To assess if children with MHL differed from hearing children in their ability to acknowledge others' intentions, a mixed-design ANCOVA was performed with Intention understanding (Intention understanding, Declarative pointing, and Imperative pointing) as the within-subject variable, Group (MHL vs. hearing) as the between-subjects variable and Age as the covariate. No main effects were found. An interaction effect was found for Intention understanding × Group F_{HF} (1.936, 267.225) = 3.063, $p < 0.05$, $\eta^2 = 0.02$. Age significantly influenced intention understanding (F (1, 138) = 3.971, $p < 0.05$). Subsequent paired t-tests in both groups separately revealed that children with MHL showed relatively lower Intention understanding compared to Declarative and Imperative pointing (as indicated by the number superscripts in Table 2). In the hearing group, children scored relatively higher on Imperative pointing as compared to Declarative pointing and Intention understanding. Intention understanding abilities increased with age.

Table 2. Mean scores on different aspects of ToM observations in both groups.

	Mean (SD)		
Observation	**MHL**	**Control**	**range**
Intention understanding	2.05 (1.03)^{a1}	2.32 (0.88)^{a1}	0-3
Joint attention			
Imperative Comprehension	2.62 (0.87)^{a2}	2.70 (0.72)^{a2}	0-3
Declarative Comprehension	2.57 (0.67)^{a2}	2.37 (0.58)^{a1}	0-3
Desires			
Similar	0.67 (0.38)^{a1}	0.93 (0.23)^{b1*}	0-1
Dissimilar	0.62 (0.42)^{a1}	0.89 (0.28)^{b1*}	0-1
False belief	0.44 (0.50)^{a}	0.63 (0.48)^{b†}	0-1

Abbreviations: MHL Moderate Hearing Loss, SD Standard Deviation. Letter-superscripts indicate differences at $p < 0.05$ on rows (between groups), number-superscripts indicate differences at $p < 0.05$ on columns (between tasks within groups). * Groups differed on both desire tasks at $p < 0.001$. † Groups differed on the false belief task at $p < 0.01$

Desire understanding

The ability to acknowledge others' desires was assessed using a mixed ANCOVA with Desires (Similar and Dissimilar) as the within-subject variable, Group (MHL vs. hearing) as the between-subject variable and Age as the covariate. This analysis revealed a main effect for Group (F (1,141) = 30.967, $p < 0.001$, $\eta^2 = 0.18$) and Age (F (1,141) = 12.714, $p < 0.001$, $\eta^2 = 0.08$). On both Similar and Dissimilar desires, children with MHL scored lower than the hearing group (as indicated by the letter superscripts in Table 2). Older children were better in acknowledging others' desires than younger children.

Belief understanding

The understanding of false beliefs was analyzed by logistic regression with Group (MHL vs. hearing) and Age as predictors. The Odds Ratio (OR) of 0.41 in Table 3 shows that the chance of successfully completing the false belief task was lower in children with MHL. The understanding of false beliefs increased with age, as indicated by the OR of >1.

Table 3. Logistic regression predicting False belief understanding

	B (SE)	Wald	Odds Ratio	p-value
Constant	-4.273 (1.36)	9.91	0.14	0.24
Group	-0.887 (0.39)	5.11	0.41	0.002
Age	0.084 (0.02)	12.70	1.09	0.000

Note. Model χ^2 (2) = 18.50, p< 0.001, Group was dummy coded: 0=control group, 1= children with moderate hearing loss

Language and ToM

Children with MHL were found to have language quotients within the normal range compared to test normative samples (*M*= 92.5, *M*=94.9, and *M*=94.4 for receptive language, word development, and sentence development, respectively. Parent-reported language skills were lower in the MHL group compared to the hearing control group (t (46.422)= -4.276, $p < 0.001$, and t (50.419)= -3.326, $p < 0.01$ for expressive language and language comprehension, respectively).

The relation between age and the ToM tasks was assessed first because age was thought to be a possible confounder of the relation between ToM and language abilities, as shown in Table 4. Pearson's correlations revealed a positive relationship between age and all tasks in both groups. Partial correlations corrected for Age revealed a positive relation between both Expressive language and Language comprehension as reported by parents, and all ToM tasks. However, the relation between both parent-reported language indices and Similar desire was absent in the MHL group and significantly different from the hearing group (z = 2.12, $p < 0.05$, and z = 2.69, $p < 0.01$ for Expressive language and Language comprehension, respectively). This same pattern was seen in the relation between the Dissimilar desire task and Expressive language (z = 2.11, $p < 0.05$).

Table 4. (Partial) correlations between different aspects of ToM, parent-reported language skills, and age.

	Age	Language Comprehension (CDI)		Expressive Language (CDI)	
	r	partial r		partial r	
		MHL	Control	MHL	Control
Similar desire	.23*	.09	.53**	.22	.55**
Dissimilar desire	.24*	.26*		.13	.48**
False belief	.30**	.24*		.29**	

Note: Partial correlations are corrected for age. Only when correlations between the two groups significantly differed (calculated using Fisher r-to-z), both coefficients are given separately. *Abbreviations*: MHL Moderate Hearing Loss, CDI Child Development Inventory. *$p < 0.01$, **$p < 0.001$

The six children with a PTA between 35 and 40 dB HL were compared with the 38 remaining children with a hearing loss between 40-70 dB. The parents of these six children with mild hearing loss reported higher Expressive language scores (*t* (39.10)= -3.715, *p* < 0.01 than the parents of children with MHL. No difference was found in their Language comprehension scores. We also observed better understanding of similar desires in the group of children with mild hearing loss compared to the children with MHL (*t* (11.87)= -2.691, *p* < 0.05. No differences were found in Intention understanding, Dissimilar desires or False belief understanding between the two groups.

Objectively measured language scores were available for the children with MHL. Correlation coefficients are shown in Table 5. When solely focusing on this group, a positive relation was found between both Receptive and Expressive language and Similar desire and False belief, but not with Dissimilar desire. The Degree of hearing loss was negatively related to Similar desires. No relations were found between the Age at first amplification and the three ToM abilities.

Table 5. Partial correlations in participants with MHL between different aspects of ToM observations, language test scores, and hearing loss related factors, corrected for age *(N=43)*.

	Receptive language	Expressive language			
	RDLS	SELT			
	Verbal comprehension	Word development	Sentence development	Age at first amplification	Degree of HL
Similar desire	.36*	.31*	.32*	.19	-.41**
Dissimilar desire	.24	.03	-.01	.04	-.14
False belief	.56***	.44**	.35*	-.05	-.30

Abbreviations: HL Hearing Loss, RDLS Reynell Developmental Language Scales, SELT Schlichting Expressive Language Test. * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$

Different stages of ToM development

Children with MHL were more often in the lower ToM stages than their hearing peers (χ^2 (4) = 25.632, *p* < 0.001). The various ToM stages can be found in Table 6. More than half of all hearing children (54.4%) mastered all ToM skills compared to 25% of children with MHL. A 4 (ToM stages) x 2 (Group) mixed ANOVA with Age as the dependent variable revealed no differences in age between the two groups in any of the ToM stages, although the overall mean age per ToM stage was different (*F* (3, 114) = 7.462, *p* < 0.001. With increasing age, children more often succeeded in the higher ToM stages. Figure 1 illustrates the relation between the different ToM stages and age. Despite the fact that we did not find a difference in age per ToM stage between the two groups, a tendency of hearing children reaching the higher ToM stages earlier in life can be seen.

Table 6. Different stages of ToM development.

ToM Stage	Similar desire	Dissimilar desire	False belief	MHL No. (%)	MHL Age (range)	Control No. (%)	Control Age (range)
1	-	-	-	12 (27.3)	52.9 (42-67)	5 (5.0)	54.8 (48-65)
2	+	-	-	3 (6.8)	50.3 (43-58)	4 (4.0)	48.0 (40-54)
3	+	+	-	6 (13.6)	59.8 (43-66)	26 (25.7)	56.9 (41-70)
4	+	+	+	11 (25.0)	63.0 (56-68)	55 (54.4)	59.9 (46-70)
			Total (%)	32 (72.7)		90 (89.1)	
Divergent				12 (27.3)	57.9 (40-69)	11 (10.9)*	54.2 (43-67)

Abbreviations: ToM Theory of Mind, MHL Moderate Hearing Loss. - : Participant was not able to successfully complete this task. + : Participant successfully completed this task. * $p < 0.05$

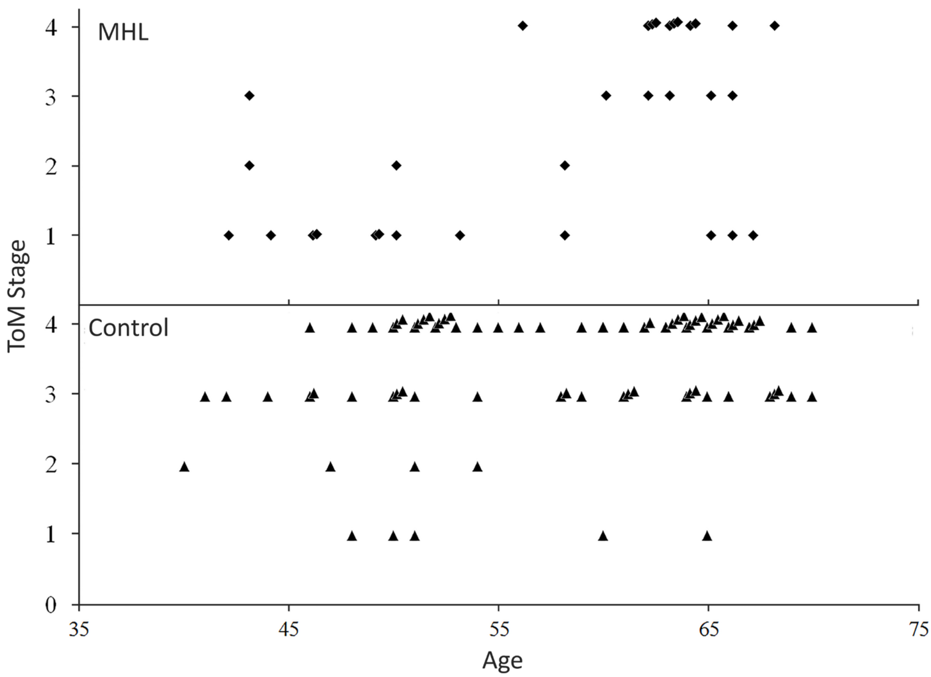

Figure 1. Spread of ToM Stages by age across participants, separated for children with MHL and hearing controls. Abbreviations: ToM Theory of Mind, MHL Moderate Hearing Loss. Note: Data points are jittered for children of the same age assigned to the same ToM Stage.

Approximately one-fourth (12; 27.3%) of children with MHL showed a divergent sequence compared to 11 (10.9%) in the hearing group (χ^2 (1)= 6.163, $p < 0.05$). The divergent sequences were so idiosyncratic that each appeared in only one or two children. For reasons of clarity, these sequences were not visualized here. Compared to children with normal developmental sequences, the children showing divergent sequences did not differ on characteristics such as age and language capacities. When focusing only on the group of children with MHL, no differences were found in age at detection, age at amplification of first hearing device, degree of hearing loss, and language capacities when comparing children with divergent sequences to those with the most common ToM development sequences.

DISCUSSION

The current study aimed to examine various aspects of Theory of Mind in children with moderate hearing loss compared to hearing peers. As far as we are aware, this is the first study to show that even moderate hearing loss can have detrimental effects on ToM abilities. In turn, these diminished ToM skills can have ongoing consequences for the social development of children with MHL. In line with our hypothesis, children with MHL had more difficulty with the acknowledgement of others' desires and beliefs than children without hearing difficulties. Furthermore, children with higher language skills were more able to acknowledge the other's perspective than those with lower language skills.

Both groups were equally able to understand others' intentions. However, children with MHL had relatively more difficulties than hearing controls with interpreting others' intentions when the other's goal was not achieved compared to more directive intention understanding tasks. Perhaps the nature of the hand gestures in the joint attention tasks was much more explicit than in the intention understanding tasks. It has previously been found that parents of children with MHL show more directive communication towards their child than parents of hearing children.[56] Possibly, children with MHL are better used to this direct form of non-verbal communication using gestures to focus attention than to more indirect forms of communication where they need to interpret the situation before they understand what is going on. The hearing children on the other hand are relatively good in joint attention compared to the MHL group, this task only asks for a shared focus of attention, without having to participate actively.

Albeit most children with MHL showed sequences of ToM development similar to hearing children, one in four children showed a divergent pattern compared to one in ten in the hearing group. Children with MHL who showed such divergent sequences did not differ in their language abilities or in other hearing loss related factors such as age at detection of hearing loss or age at start of hearing amplification compared to those with normal sequences. However, we should interpret these results with care as these analyses were done in rather small groups. A lack of power could have prevented us from finding significant results. Because we were not able to identify factors that influenced such divergent development, we can only speculate about causes for divergent development. Possibly, the duration of testing was more exhausting for children with hearing loss. Since the belief understanding task was administered at the beginning of the test session, it may be that the children paid more attention than when administering the desire task at the end. In addition, beliefs were measured by a single task whereas to pass the (dis)similar desire tasks, children needed to succeed on the test twice resulting in a higher chance to fail one of them and obtaining a negative score. Yet, all tasks have previously been used successfully in different clinical groups (i.e., preschoolers with a CI and preschoolers with an autism spectrum disorder) with reliable results.[26,46]

Despite their relatively good intention understanding skills, children with MHL fall behind compared to hearing peers on more language dependent skills such as desire and belief acknowledgement. In line with previous studies in children with more severe hearing loss wearing a CI, it is likely that a hearing loss may act as a barrier that prevents sufficient access to social communication in our sound-dominated world. This reduced ability to adequately receive social cues may cause a delay in ToM development.[26,38] The relationship between ToM and hearing loss can be explained by several challenges that children with hearing loss and their families have to face. One aspect is the input children with hearing loss receive from their parents. In the first few years of life, parents provide the largest proportion of verbal input to the child. When parents talk about how others feel, what they want or wish for, they stimulate ToM understanding in their children.[57,58] However, research has shown that the quality of input that parents present to their child with MHL is frequently lower than in hearing children.[40] As a result, children with MHL may encounter more difficulties increasing their language capacities. This in turn may prevent them from higher quality interactions that are essential in order to discuss abstract concepts such as other's mental states and emotions.

However, *what* is said is not only important, but also *how* it is said. Both diversity in syntactic structures and the introduction of various speakers can positively influence ToM development.[5,58,59] Yet, parents of children with hearing loss often choose more simple and clear formulations when talking to their child. A relatively larger proportion of communication is also more directive in nature, aiming to instruct the child instead of discussing or explaining the child's surroundings. Parents adjust the complexity of their language to the child's language abilities.[40] Although simple and clear communication can benefit language understanding in children with MHL, limited diversity of input may also hamper more complex language development in the long run. Again, diminished opportunities to learn about others' perspectives may lead to less experience in ToM usage in children with MHL.

With the introduction of cochlear implantation, the focus of research on hearing loss has shifted. Improving and understanding the effects of this highly innovative technique became the goal of many funders and commercial companies for obvious reasons.[60] But how about the children with moderate hearing loss? A recent special issue of Ear and Hearing discussing the Outcomes of Children with Hearing Loss (OCHL) Study by Moeller and colleagues addressed the challenges that children with MHL have to face. Among other things, this large longitudinal study revealed that children with MHL are still at risk for the development of language delays. The outcomes of the present study in which the language skills of children with MHL are in the low-normal range compared to test normative samples are in line with these findings. Despite their relatively normal language skills, the parent-reported language skills of children in the MHL group were below the average range. These scores possibly better reflect children's communication skills in daily life, because parents do not base their judgment on one particular moment but on the child's average skills over a longer period in time. Because communicative abilities determine how well a child is able to join conversations with others, this may also better reflect their opportunities for incidental learning, which subsequently determines their social development. This is in line with the outcomes of the OCHL study in which qualitative aspects of conversations were important for a child's language output.[11,40] Our study is unique in providing insight into the relation between language skills and different aspects of ToM.

Parent-reported language skills were strongly related to ToM in the hearing controls. Yet, the relation between desire understanding and parent-reported language skills in children with MHL was almost absent. On the other hand, we found a relation between objective test-scores and desire understanding. It is possible that parents rate their child's language skills in daily life, and take account for their lower communication skills in interactions with others and in noisy environments. They acknowledge the difficulties their child with MHL has in communication with others. This obviously differed from the quiet language-test settings in clinical surroundings. During the ToM observations in this study there was no time limit so children could take their time which might have benefitted their ToM outcomes compared to how they would have responded in hectic daily life. Still, this does not explain the absent relation between objective language tests and the dissimilar desire task. This absence could be the result of our study design. Children completed the false belief tasks relatively early and the dissimilar desire tasks relatively late during the test session. In addition, the dissimilar desire task was preceded by a rather difficult task that is not described in this study. Possibly, the children became tired and lost their concentration. Concentration difficulties are well known in children with various degrees of hearing loss.[61] Either way, this finding highlights the importance of this study. It aims to trigger both parents and professionals to be alert when it comes to ToM development in young children with MHL. It shows that although parents are well able to understand their child and professionals rate their language abilities to be within the average range, these children are at risk for delays in their social development. In addition, the outcomes of this study suggest it might be better to also focus on the child's communicative abilities than to solely rely on language test scores.[11]

Future research

We would like to point out that this study is a first attempt to address ToM-related difficulties in children with MHL. Some of the analyses were done in rather small groups and using a cross-sectional design. A second limitation of the current study concerns the absent language scores in the control group. Although norm-scores were available for typically developing (hearing) children, it would be more convenient to directly compare the two groups. Although a clear difference in ToM skills was found between the two groups, we feel that we are only able to hypothesize about a possible delay when focusing on the developmental patterns of ToM in young children with MHL. To confirm our findings, there is a strong need for longitudinal research that is able to link age, language and ToM abilities of increasing difficulty to confirm causality and to focus on different developmental patterns in this specific group of young children. In addition, future research should also include participant and family-related factors that may influence social development like the cognitive abilities of the child (e.g., phonological working memory, executive functioning) and the socioeconomic status of the family as these factors are known to influence language skills and general development. This study was unable to show a direct link between hearing loss-related factors such as the age at detection or the age at first HA amplification and ToM. However, factors like audibility and early access to HA's have been proven to influence language skills in MHL children and should therefore be integrated in future studies when studying social functioning in this group of children.[11]

CONCLUSION

The present study shows that children with MHL often encounter problems in developing age-appropriate ToM skills, even though their language capacities are within the normal range. These difficulties can seriously hamper social learning since ToM skills are essential for inducing and maintaining relationships. Early intervention programs should emphasize the importance of developing skills to acknowledge the other's perspective in this specific group of children.

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CHAPTER 6

LOW **EMPATHY** IN DEAF AND HARD OF HEARING **(PRE)ADOLESCENTS** COMPARED TO NORMAL HEARING CONTROLS

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ABSTRACT

Objective
The purpose of this study was to examine the level of empathy in deaf and hard of hearing (pre)adolescents compared to normal hearing controls and to define the influence of language and various hearing loss characteristics on the development of empathy.

Methods
The study group (mean age 11.9 years) consisted of 122 deaf and hard of hearing children (52 children with cochlear implants and 70 children with conventional hearing aids) and 162 normal hearing children. The two groups were compared using self-reports, a parent-report and observation tasks to rate the children's level of empathy, their attendance to others' emotions, emotion recognition, and supportive behavior.

Results
Deaf and hard of hearing children reported lower levels of cognitive empathy and prosocial motivation than normal hearing children, regardless of their type of hearing device. The level of emotion recognition was equal in both groups. During observations, deaf and hard of hearing children showed more attention to the emotion evoking events but less supportive behavior compared to their normal hearing peers. Deaf and hard of hearing children attending mainstream education or using oral language show higher levels of cognitive empathy and prosocial motivation than deaf and hard of hearing children who use sign (supported) language or attend special education. However, they are still outperformed by normal hearing children.

Conclusion
Deaf and hard of hearing children, especially those in special education, show lower levels of empathy than normal hearing children, which can have consequences for initiating and maintaining relationships.

INTRODUCTION

Hearing impairment poses many challenges to the developing child. Deaf and hard of hearing (DHH) children for instance frequently encounter language and communication problems. These difficulties in communication may result in reduced opportunities for incidental learning. Especially abstract concepts such as emotions are therefore more difficult to understand for children with hearing loss.[1] Regulating and understanding one's own emotions is essential for the development of adequate empathic abilities. Consequently, DHH children are prone to develop lower empathic skills than normal hearing (NH) peers. Because empathy is of major importance in initiating and maintaining social relationships, this could have ongoing consequences in the development of DHH children.

Empathy

Empathy is defined as the ability to perceive and understand another person's emotional state and the competence to appropriately respond to others' emotions.[2,3] It is needed to induce prosocial behavior: free-willing behavior to benefit others.[4] Therefore, empathy is often referred to as 'social glue' in relationships.[4-6]

From a developmental perspective, empathy has been divided into different layers: affective empathy, cognitive empathy and prosocial motivation. Affective empathy, also known as emotional contagion, is the process in which the emotional states of others cause a level of arousal in the observer. It consists of non-conscious behavioral mimicry of others' facial, vocal, and bodily expressions.[7] This 'mirroring of emotions' is thought to be present at birth and originates from the Mirror Neuron System (MNS) in the brain.

Through functional Magnetic Resonance Imaging (fMRI) studies, neuroscientific research has shown these neural networks. For instance, making a sad face or observing a sad face both activate the MNS via the amygdala and the anterior insula of the brain. This motor activation is then associated with an emotion representation; the person acknowledges a sad feeling.[8-10] These patterns strongly suggest that the formation of cortical representations about one's own feelings is a necessary condition to engage in vicarious predictions about the emotions of others[5] (see Lamm & Majdandžić for an in-depth discussion of the plausibility of this assumption).[11]

Whereas young children become upset and need comforting themselves through affective empathy, also referred to as 'contagious crying' or 'emotional sharing', around the age of two children change from a self-focused perspective towards another-focused perspective. Consequently, children gradually become to understand that their sad feelings are caused by another person in distress. This evokes an urge to support or comfort that person, as to relief their distress.[3,5,12]

Cognitive empathy develops as children grow older and involves a more sophisticated comprehension of the other person's emotional state.[13] The child starts to understand why the other is upset. Understanding emotions in others serves different goals. First, the observer is capable to distinguish between its own and the other's emotions and thereby decreasing their own feelings of distress. Second, understanding the other leads to an increased tendency to support the other, and to care for the other.[11] This intrinsic prosocial motivation is essential because it signals to the person in distress that the observer pays attention to and highly values their emotions. He or she understands what is happening and wants to support. Moreover, a stronger level of cognitive empathy can also help to overcome in-group preferences.[11] The long-term purpose of cognitive empathy and prosocial motivation is to induce and maintain good social relationships.[12] As such, the development of cognitive empathy is largely dependent on social learning. fMRI studies have indirectly shown this as the relation between the MNS and self-reported cognitive empathy is less clear than for affective empathy.[7,11] For social learning to develop, this requires incidental learning skills; unplanned and unorganized learning abilities, with no educational intentions. Social learning takes place while interacting with others, and by trial-and-error.

A lack of empathy is associated with violence, aggression, criminality, and insensitive and unemotional behavior.[14] Empathic dysfunction has been associated with several psychiatric disorders such as psychopathy, autism spectrum disorders[15], conduct disorder, acquired sociopathy[16], and schizophrenia[17]. Children and adolescents who show little or no empathy are deemed to fail in our social world, and are put aside as having antisocial behavior. These behavioral problems may lead to the development of an antisocial personality disorder later in life.[18] Hence, it is of major importance for children to adequately develop empathic skills.

Empathy in deaf and hard of hearing children

Little is known about the development of empathy in DHH children. However, certain prerequisites for successful empathic maturation, such as emotion recognition and regulation together with development of a Theory of Mind (ToM), have recently been addressed in this population. Studies show lower levels of emotion recognition and labelling of emotions in deaf preadolescents than in NH peers. In this population the onset of deafness was related to the ability to recognize emotions. Prelingually deaf preadolescents were more vulnerable than their postlingually deaf peers.[19] Regarding emotion awareness, DHH children were found to be less able to address multiple emotions in the negative domain simultaneously (e.g., anger and sadness) than NH peers during several emotion tasks. In the same study, children had to focus on approaching strategies towards an emotion-evoking situation. The results show less effective emotion regulation in DHH children than in NH peers.[20] ToM has been measured in toddlers with cochlear implants (CIs). Initially children with CIs were found to perform as well as NH children. However, at an older age they fell behind on more advanced ToM abilities such as false belief tasks.[21] Regarding their empathic behavior, no differences were found between young children with a CI and NH peers.[5] Yet, because of the young age of these children (1-5 years), only the affective domain of empathy could be taken into account in this study.

Present study

Because of the continuous development of cognitive empathy in childhood and preadolescence we are interested whether empathic abilities in DHH children and adolescents differ from those of their NH peers. Therefore, the aim of this study was to determine the differences in the levels of self-reported and observed empathy between DHH on the one hand, and NH children and adolescents on the other. To identify those factors that may be most influential for the levels of empathy in DHH children we also investigated the influence of several audiological factors on empathic abilities, such as language development, intelligence, degree of hearing loss, age at intervention of hearing loss, type of device, mode of communication, and educational setting.

On the basis of the research mentioned above we expected to find equal levels of affective empathy in DHH and NH children. However, regarding the development of cognitive empathy and prosocial motivation we expected DHH children to fall behind as a consequence of, among other things, their impaired ToM development. Concerning several audiological variables such as type of hearing amplification, it has been reported that DHH children wearing CIs experience lower levels of behavioral problems than children wearing Hearing Aids (HAs).[22] Therefore, we expected to find differences in empathic ability between these two groups.

Gender differences have been described frequently in the literature. Girls consistently report higher levels of affective empathy and prosocial behavior than boys. Some researchers doubt these conclusions. They hypothesize that the reported differences are a result of differences in social desirability between boys and girls.[8,23] If true, we would find higher levels of self-reported affective empathy and prosocial motivation in girls, regardless of their hearing status but equal levels of empathy and supportive behavior during observations.

Due to the improved developmental outcomes after early intervention programs as reported by Yoshinaga-Itano et al.[24], we expected a relation between age at detection and intervention of hearing loss, and empathic abilities. Educational placement (mainstream or special schools) and mode of communication (spoken or sign language) have been reported to be related to levels of psychopathology in DHH children.[25-28] We therefore expected that children attending mainstream education and using spoken language as their preferred mode of communication show higher levels of empathy.

METHODS

Participants

We recruited 122 DHH children and a control group consisting of 162 NH children from all over The Netherlands and the Dutch-speaking part of Belgium to participate in this study. All children were between 9 and 16 years of age at time of assessment. The age of 9 as a cut-off point was chosen because the children needed to be able to reflect on their own emotions and behavior.[29] All children had an IQ of 80 or higher and no other known disabilities besides their hearing loss. Of all DHH children, 52 were fitted with a CI and 70 children wore conventional HAs. Hearing impairment was defined as experiencing a loss of ≥40 dB in the best ear that was detected pre- or perilingually. Children with postlingual onset or detection of hearing loss were excluded. The NH group was matched with the DHH group on sex and mean age. As can be seen in Table 1, gender, intelligence, socio-economic status (SES), and age did not differ between the groups. No differences were found in type of school and mode of communication when comparing children wearing a CI with children using HAs. The onset of hearing impairment differed between the two groups; χ^2 (1, n = 115) = 3.92, $p < .05$. The HA-group presented more perilingual onset of hearing impairment than the CI group. As expected the degree of hearing loss differed between the two groups χ^2 (2, n = 114) = 73.62, $p < .001$. Children with a CI mainly experienced profound losses whereas children with HAs showed more moderate to severe hearing losses. Permission for this study was granted by the Medical Ethics Committee of the Leiden University Medical Center under number P10.137.

Procedure

To increase the external validity of our findings, we tried to ensure diversity in our study population and recruited children all over The Netherlands and the Dutch-speaking part of Belgium via hospitals, speech- and hearing centers, primary and secondary schools, and special schools for the deaf. Written parental informed consent was obtained for all participating children. The assessment was carried out in a quiet room. Before starting the tests, children were assured that their answers would remain anonymous. Questions appeared one by one on a laptop. Depending on their preferred mode of communication, DHH children could choose between two versions of the questionnaires: a written text, or a version in which this text was simultaneously accompanied by sign language. The questionnaires were assessed as part of a larger study on the socio-emotional development of DHH and NH children. In between several tests, the experimenter acted live emotions to observe empathic reactions and supportive behavior during the test session. Parents completed questionnaires at home, they were also asked to complete a list of background variables such as net income and level of education. A socioeconomic status (SES) score was calculated using the net income of the family, job and level of education of both parents. Audiological variables were extracted from the child’s medical and/or audiological notes.

Table 1. Demographic characteristics of participants

	Total study population N = 284		HI study population N= 122	
	HI	**Controls**	**CI**	**HA**
No. of children	122	162	52	70
Age				
Mean - in years (SD)	11.9 (1.8)	11.9 (1.3)	11.8 (2.0)	12.0 (1.7)
Range - in months	100 - 194	99 - 176	100 - 194	110 - 188
Gender				
Male (%)	60 (49)	73 (45)	24 (46)	36 (51)
Socioeconomic Status (SD)	11.5 (2.3)	11.7 (2.3)	11.7 (2.3)	11.3 (2.4)
Nonverbal intelligence (SD)	10.3 (2.8)	10.7 (2.5)	10.0 (2.7)	10.5 (3)
Language Skills (SD)	6.5 (2.7)	7.0 (1.9)	6.1 (2.8)	6.7 (2.6)
Preferred mode of communication (%)				
Oral language only	94 (77)		39 (75)	55 (78)
Sign-supported Dutch	26 (21)		13 (25)	13 (19)
Sign language only	2 (2)		0 (0)	2 (3)
Type of education				
Regular education (%)	74 (61)	162 (100) *	32 (62)	42 (60)
Onset of hearing loss (%)				
Prelingual	103 (84)		48 (92)	55 (78) *
Perilingual	12 (10)		2 (4)	10 (14) *
Unknown	7 (6)		2 (4)	5 (7)
Degree of hearing loss (%)				
Moderate - 40-60 dB	29 (24)		0 (0)	29 (41) **
Severe - 61-90 dB	25 (21)		1 (2)	24 (34) **
Profound - >90 dB	60 (49)		49 (94)	11 (16) **
Unknown	8 (6)		2 (4)	6 (9)
Age at detection of hearing loss - in months (SD)	19.1 (15.7)		14.6 (10.4)	22.9 (18.3) **
Age at 1st hearing aid acquisition - in months (SD)	24.8 (17.0)		17.3 (10.2)	31.2 (18.9) **
CI characteristics				
Age at implantation (CI) - in months (SD)			44.5 (32.6)	
Duration of CI use - in months (SD)			99 (33)	
Bilateral CI (%)			13 (25)	

Abbreviations: HI Hearing Impaired, CI Cochlear Implant, HA Hearing Aid, SD Standard Deviation
* $p < .05$, ** $p < .01$

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MATERIALS

The instruments used in this study are described here. Psychometric characteristics of all questionnaires are shown in Table 2.

Self-reported empathy

The Empathy Questionnaire for Children and Adolescents (EmQue-CA) consists of a total number of 18 items, scored by children on a 3-point Likert scale (1= not true, 2= somewhat true and 3= true). The items measure the different levels of empathy: affective empathy, cognitive empathy and the urge to support the other. The 'affective empathy' scale defines to what extent emotions in others cause isomorphic feelings in the observer (e.g., "If a friend is sad, I also feel sad"). The scale measuring cognitive empathy defines to what level children understand the emotions they observe in others (e.g., "When a friend is angry, I tend to know why"). The third scale prosocial motivation' defines the tendency to support a distressed other (e.g., "If a friend is sad, I like to comfort him"). The Questionnaire was validated for NH children of 9 years and older.[30,31] The internal consistency of the scales is acceptable to good; and the questionnaire shows a good three-factor structure[30], which warrants that the questionnaire is suitable to make group comparisons.[11]

From the Emotion Awareness Questionnaire (EAQ), the 'attendance to others' emotions' scale was used (e.g., If a friend is upset, I try to understand why). Children rated how valuable they found other children's emotions on a 3-point Likert scale (1= not true, 2= sometimes true, 3= often true).[32] The internal consistency of the scale is acceptable.

The 'emotion recognition' scale from the Emotion Expression Questionnaire (EEQ) was scored by parents (e.g., Does your child know when you are angry?). The questions were rated on a 5-point Likert scale (1= (almost) never, 2= rarely, 3= sometimes, 4= often, 5= (almost) always).[2] The internal consistency of the scales is good.

Measurement invariance was not assessed for the above described questionnaires. However, the questionnaires were specifically designed to use in different clinical groups (Children with specific language impairments, autism spectrum disorders, and DHH children). Therefore, items were formulated with short sentences to increase understanding. Previous studies have shown consistent and positive outcomes in these groups.[30,31]

Observation of empathy

Participating children were faced with 'live' emotions from the experimenter to observe to what extent they would show empathic reactions. Multiple situations were acted out, which aimed to evoke attention for the situation and/or the experimenter's emotion and prosocial responses directed at the experimenter. Before data collection started, experimenters were instructed on how to simulate emotions. Emotions were modeled by a psychologist experienced in simulating emotions for behavioral assessment purposes. Additionally, experimenters watched multiple video clips of emotion simulations, which

Table 2. Psychometric properties of empathy questionnaire, EAQ, EEQ and observations

			Total study population N= 284				HI study population n= 122	
	No. of items	Answer range	Crohnbach's alpha		M (SD)		M (SD)	
			HI	Controls	HI	Controls	CI	HA
Self report								
Empathy Questionnaire	18	1-3	.83	.81	2.2 (0.3)	2.4 (0.3)**	2.2 (0.3)	2.3 (0.3)
Affective empathy	7		.66	.68	1.9 (0.4)	2.1 (0.4)	1.9 (0.4)	2.0 (0.4)
Cognitive empathy	5		.68	.66	2.3 (0.4)	2.5 (0.4)**	2.3 (0.4)	2.3 (0.4)
Prosocial motivation	6		.76	.72	2.6 (0.4)	2.7 (0.3)**	2.6 (0.4)	2.5 (0.4)
EAQ - Attendance to others' emotions	5	1-3	.64	.60	2.3 (0.4)	2.5 (0.4)**	2.2 (0.4)	2.3 (0.4)
Parent report								
EEQ - Emotion recognition	6	1-5	.78	.74	2.6 (0.3)	2.6 (0.3)	2.6 (0.3)	2.5 (0.3)
Experimenters observation								
Attention to emotion	8	1-3			2.5 (0.4)	2.2 (0.4)**	2.4 (0.5)	2.5 (0.4)
Suportive behavior	1	1-3			2.6 (0.6)	2.9 (0.4)**	2.6 (0.6)	2.6 (0.5)
WISC non-verbal intelligence˜	26	0-7			10.3 (2.8)	10.7 (2.5)	10.0 (2.7)	10.5 (3)
CELF-IV language development˜	35	0-1			6.5 (2.7)	7.0 (1.9)	6.1 (2.8)	6.7 (2.6)

Abbreviations: HI Hearing Impaired, CI Cochlear Implant, HA Hearing Aid, SD Standard Deviation, EAQ Emotion Awareness Questionnaire, EEQ Emotional Expressivity Questionnaire.
Note. * $p < .05$, ** $p < .01$, ˜ normscores

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were obtained during a pilot study. Specific instructions were provided regarding the duration and intensity of the emotions displayed, as well as regarding the verbal and non-verbal cues that accompanied these. Experimenters then practiced and video-recorded multiple emotion simulations themselves, and received feedback on their performance from the trainer. Training continued until all experimenters could simulate the emotions in a natural way, as judged by the trainer.

In the first situation, the experimenter pretended to receive text messages from a friend. The experimenter reached for her phone and pretended to read the first message, after which she shared with the participant that it contained an invitation from her friend to go to the movies that night. The experimenter had an excited, happy facial expression and said that she was looking forward to it. After that, she put away the phone and continued the test session. Approximately 30 minutes later the experimenter pretended to have received another text message. This time, she shared with the participant that her friend had to cancel the appointment, meanwhile showing a disappointed, sad facial expression. After five seconds, the experimenter stored her phone and carried on with the session. During and after revealing the second message, the experimenter observed the behavioral and verbal responses of the participant.

In the second situation, the experimenter pretended she could not find her pen. Earlier, the pen was placed outside the direct line of sight of the experimenter (i.e., behind a binder), but in full view of the participant. For a duration of ten seconds, the experimenter looked around and searched her bag, stating that she could not find her pen. Meanwhile, children's responses were observed.

In the third situation, the experimenter collected testing materials and dropped one item on the floor. The experimenter looked at the item and said 'oops', but continued to gather the rest of the materials. Children's behaviors in response to the situation were observed.

Children's reactions across all three situations were scored on a checklist (1= no, 2= slightly, 3= yes) and were grouped into 'attention to emotions' (e.g., looking at the experimenter) and 'supportive behavior' (e.g., returning the lost pencil). Unfortunately, due to time restraints scores from 9 CI children, 9 HA children and 1 NH child are missing.

Language skills and intelligence

Nonverbal intelligence of participants was assessed using two components of the Wechsler Intelligence Scale for Children-Third Edition (WISC): block design (duplicating geometric designs with cubes) and picture concepts (arranging pictures to create logical stories).[33] These scores were compared with scores of earlier completed intelligence tests (either the Snijders-Oomen or the WISC).[34] A high correlation was found previously by Theunissen et al.[22] making the shorter subtest a good reflection of the child's intelligence level. The WISC has been proven to show excellent test-retest abilities and long-term stability.[35]

Sufficient language abilities are regarded essential to ensure comprehension of the different questionnaires. This was tested using a sentence comprehension and a story comprehension task. Children using oral language as their preferred mode of communication completed the Dutch version of the Clinical Evaluation of Language

Fundamentals – Fourth Edition (CELF-IV).[36,37] The CELF has been proven to show high stability coefficients. Studies were conducted in several clinical groups including children with language disorder, and hearing impairment.[10] DHH children who preferred communicating by sign (supported) language completed subtests from the Assessment Instrument for Sign Language of The Netherlands.[38]

Statistical Analyses

Group demographics were compared using independent t-tests. To compare the levels of empathy (affective empathy, cognitive empathy and prosocial motivation) between the different subgroups repeated measures Multivariate Analysis of Variance (MANOVA) and Multivariate Analysis of Covariance (MANCOVA) were used. In case of differences between subgroups within the DHH children, when sample sizes were small (< 40 children per group) the assumption of normality was violated. Therefore, to compare levels of empathy between these subgroups (e.g., uni- versus bilateral CI, pre- versus perilingual onset of hearing loss) a non-parametric test was chosen (i.e., Mann-Whitney U test). Correlations between the empathy subscales and audiological factors were calculated using Pearson's correlations. These correlations were compared between the different groups using Fisher's r-to-z transformations to be able to show significant differences between correlations. Statistical analyses were carried out using the program SPSS version 21.0 (IBM Corp., Armonk, NY).

RESULTS

Self-reported empathy in DHH and NH children

To analyze the differences in self-reported empathy levels between children with a CI, those with HAs, and hearing children, we carried out a repeated measures MANOVA with Group (CI, HA, NH) as the between-subjects variable and self-reported empathy (affective empathy, cognitive empathy, prosocial motivation) as the within-subjects variable. The analysis showed a main effect for empathy ($F_{HF}(1.97, 553.96) = 303.81$, $p < .001$, $\eta_p^2 = .52$) and for group ($F(2, 281) = 11.44$, $p < .001$, $\eta_p^2 = .08$), which was qualified by an empathy x group interaction ($F_{HF}(3.92, 553.96) = 2.46$, $p < .05$, $\eta_p^2 = .02$). Post-hoc t-tests showed that on affective empathy children with CIs scored lower than the NH group. Scores on affective empathy by children with HAs did not differ from NH children. DHH children overall scored lower on cognitive empathy and prosocial motivation than NH peers, regardless of their type of hearing amplification.

Because of the known influence of language development and intelligence on the socio-emotional development of DHH children, these variables were added as covariates in the analyses. In a MANCOVA that corrected for language development and intelligence, the main effect for group remained ($F(2, 236) = 6.30$, $p = .002$, $\eta_p^2 = .05$), but the interaction effect was no longer significant ($F_{HF}(3.95, 465.57) = 1.55$, $p = .19$, $\eta_p^2 = .01$). Language development was significantly related to the levels of empathy ($F(1, 236) = 5.25$, $p = .02$) whereas intelligence was not (Figure 1).

A gender x self-reported empathy repeated measures MANOVA was conducted to define differences in self-reported empathic abilities between boys and girls, regardless of their hearing status. Results showed a main effect for empathy (F(1.97, 555.75) = 393.96, p < .001, η_p^2 = .58) and for gender (F(1, 281) = 11.10, p = .001, η_p^2 = .04), which was qualified by an empathy x gender interaction (F_{HF}(1.97, 555.75) = 4.33, p < .05, η_p^2 = .02). Post-hoc analysis revealed that girls scored higher on affective empathy and prosocial motivation than boys. Equal levels of cognitive empathy were reported. The aforementioned results were combined in a 3 (hearing group) x 2 (gender) x 3 (self-reported empathy) repeated measured MANCOVA with language development and intelligence as covariates. The main effect for group remained (F(2, 233) = 5.75, p = .004, η_p^2 = .05) whereas the results no longer showed a main effect for gender (F(1, 233) = 3.54, p = .06, η_p^2 = .02).

Concerning attendance to others' emotions, a 2 (DHH, NH) x 2 (boys, girls) one-way ANCOVA that corrected for language skills and intelligence revealed an effect for hearing group (F (1, 235) = 8.52, p < .01) and gender (F (2, 235) = 18.04, p < .001). NH children reported higher scores than DHH children and girls scored higher than boys. Language development was significantly related to the attendance towards others' emotions (F(1, 240) = 4.80, p < .05). A one-way ANCOVA to compare the effect of hearing group and gender on emotion recognition as scored by parents corrected for language development and intelligence showed no differences between the hearing groups or gender (F (1, 182) = 0.03, p = .87 and (F (1, 182) = 0.065, p = .80, respectively).

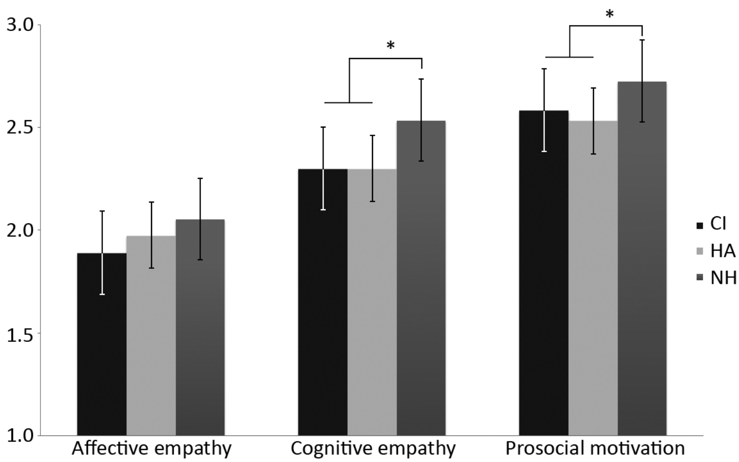

Figure 1. Mean empathy scores per group. * p < .01

Observation of empathy and supportive behavior

Differences between gender and hearing status in observed empathic behavior during the live emotions tasks were assessed with language and intelligence as covariates. A 2 (DHH, NH) x 2 (boys, girls) mixed ANCOVA revealed an effect for hearing status and for gender; DHH children scored higher than their NH peers on emotion attention(F (1, 220) = 28.80, $p < .001$); regardless of their type of hearing amplification. Girls scored higher than boys (F (1, 220) = 10.94, $p = .001$). To compare DHH and NH boys and girls on their observed supportive behavior, a 2 (DHH, NH) x 2 (boys, girls) mixed ANCOVA was performed showing an effect for hearing status but not for gender (F (1, 220) = 16.03, $p < .001$ and F (1, 220) = .66, $p = .42$, respectively). Conversely to their 'emotion attention', NH children more often showed supportive behavior than DHH children.

Audiological and socio-demographic factors influencing empathy

In order to properly examine levels of empathy between DHH children at special education (for the deaf and hard of hearing child) and at mainstream education, a MANCOVA was performed with school-type (special or mainstream) as the between-subjects variable, the self-reported levels of empathy as the within-subjects variables and language development as a covariate. The two groups did not differ in background and audiological characteristics (e.g., age at detection of hearing loss, age at intervention, intelligence, SES). The analysis showed a main effect for empathy (F_{HF}(2, 204) = 9.16, $p < .001$, $\eta_p^2 = .08$) and for school-type (F(1, 102) = 4.38, $p < .05$, $\eta_p^2 = .04$). Post-hoc ANCOVA's revealed higher levels of cognitive empathy in DHH children attending mainstream schools than in DHH children attending special schools, (F_{HF}(1, 102) = 7.89, $p < .01$), whereas for affective empathy and prosocial motivation no significant differences were found (F_{HF}(1, 102) = 1.61, $p = .21$. and F_{HF}(1, 102) =.91, $p = .34$, respectively). No significant differences were found in observed empathic reactions nor in parent reported emotion recognition or attendance to others' emotions comparing DHH children in mainstream and special education when corrected for their language skills.

When comparing the child's preferred mode of communication DHH children using sign (supported) language scored lower on self-reported prosocial motivation and on observed attention to emotions than DHH children who preferred to use spoken language (U = 986.5, z = -2.95, $p = .003$ and U = 802.5, z = -2.32, $p = .021$, respectively). Two participants solely communicated by sign-language. All analyses were rerun without these two participants. The results did not differ.

No significant differences were found between the levels of empathy in children regarding the moment of detection of their hearing loss (i.e., pre- or perilingual). When comparing within the CI group, parents reported higher levels of emotion recognition in unilaterally implanted children compared to bilaterally implanted children (U = 82, z = -2.54 $p = .01$).

In the DHH group, the relation between several continuous audiological variables (degree of hearing loss, age at detection of hearing loss, age at intervention of hearing loss, age at implantation) and the levels of empathy (self-report, parent-report and observed) were analyzed by means of Pearson's correlations. No relations were found between these variables and the different levels of empathy.

DISCUSSION

Empathy is an important capacity which helps to build and maintain positive social relationships.[39] It has been argued that affective empathy (i.e., feeling what the other person feels) is neurologically hard-wired, i.e., present in children despite their social learning experiences.[35] Yet, the level of cognitive empathy (i.e., understanding the other's emotions) depends for instance on the extent to which children can participate in a social environment.[39] We hypothesized that DHH children would be seriously disadvantaged in this respect. The outcomes of this study support our hypothesis: DHH children report equal levels of affective empathy as NH peers. Even higher levels of attention to others' emotions in DHH children than in NH children were found during an observation task. Yet, DHH children reported lower levels of cognitive empathy, and valued emotional information about other people as less important. Moreover, both a self-report and an observation task show less supportive behavior in the DHH group compared to NH peers. In other words, DHH children might feel what the other person feels, and also attend to those emotions, but they have less understanding of their causes; they value others' emotions as less important, and also react less adaptively to supporting the person in distress. Yet, especially the capacity for cognitive empathy, whereby one is more inclined not only to feel for the other, but also take the perspective of the other person, is essential in overcoming in-group preferences and avoiding parochialism.[11]

Consistent with other research in the domain of empathy, girls scored higher than boys on affective empathy and prosocial motivation. Only during the observation tasks, no differences were found between boys' and girls' tendency to behave supportive. Within the DHH group we see that children in mainstream schools, or those who used spoken language as their primary mode for communication, did better on cognitive empathy than their DHH peers in special schools or using sign or sign(-supported) language, respectively. Unfortunately, they are still outperformed on these abilities by NH children.

Although the level of affective empathy was equal in both groups, this was only after we controlled for the children's language capacity. Language abilities were taken into account since previous studies have shown communication skills and interaction with others are improved by sufficient language skills, resulting in better socio-emotional development and fewer symptoms of psychopathology in DHH children.[6,22,27,40] However, impaired language skills only partly explain the lower empathic abilities we found in DHH children. Even when we control for language skills, we still find that DHH children are outperformed by their NH peers on empathic abilities that are more dependent on social learning such as cognitive empathy and prosocial motivation. This indicates that for fully-fledged empathic functioning sufficient language skills alone are not enough.

By observing how others interact we learn how to deal with our own and others emotions and to place them in a social context. This so-called incidental learning (i.e., learning by experience and with no educational intentions) is essential in order to develop empathic behavior [41]. Observing how a mother comforts her son after he lost his favorite football not only helps to understand how the boy feels (i.e., cognitive empathy) but also shows an adequate response (i.e., prosocial behavior). Since incidental learning often implies overhearing conversations between others with quick and snappy dialogues, missing the opportunity for this kind of learning will disadvantage DHH children.

For adequate cognitive empathy to develop a child needs to be able to recognize emotions in others.[42] Previously, lower levels of emotion recognition were reported in DHH toddlers[43] and school-aged children compared to NH peers.[44] This could explain the impaired level of cognitive empathy in the DHH group in our study. However, our study also indicates that DHH children are just as capable as their NH peers when it comes to recognizing emotions in others. It may be that with increasing age DHH children are able to catch up on this ability, and identification of emotions in others no longer seems to be the problem. It is the more complex interpretation of the whole emotion-evoking situation that causes confusion: why is my friend angry, what has happened?

The DHH population is often characterized by its heterogeneity (e.g., differences in degree of hearing loss, type and duration of hearing amplification, educational setting, mode of communication). In our study DHH children attending mainstream schools reported higher levels of cognitive empathy than DHH children in special schools for the Deaf and Hard of Hearing. Yet, we have to note that reasons for professionals to advise children to attend special education are diverse. Language skills and intelligence are factors influencing school placement in DHH children. Because these abilities can also influence empathic functioning, we considered them to be confounding factors. However, our study indicates that even if the levels of language skills and intelligence are equal, DHH children attending special education still have difficulties understanding others' emotions. Despite these difficulties, DHH children in special education do not differ in their tendency to behave prosocial when compared to DHH children that attend mainstream education.
Children in special schools more often use sign language as their preferred mode of communication. In our study we found that children who use sign (supported) language showed less prosocial motivation. However, when comparing signers in special and mainstream education we found no differences in any of their empathic abilities. Previous studies reported differences in socio-emotional development between children with CIs and those wearing HAs in favor of the children wearing CIs. Our study indicates that when the child's focus needs to shift to 'the other' instead of 'the self', these differences no longer appear and both groups show equal levels of empathy. Yet, these results have to be interpreted with caution as the groups used for these analyses were rather small.
It is important to note that the children in this study were born before the start of early detection and intervention programs in the Netherlands and Belgium. Therefore, these children were rehabilitated at a relatively late age (e.g., mean age at first hearing amplification 24.8 months, mean age at implantation 44.5 months). With the introduction

of newborn-hearing screening programs, intervention and rehabilitation now preferably starts before the child is six months old.[4] As early intervention programs have been shown to improve speech and language skills, these improvements will hopefully lead to better communication skills, resulting in more effective incidental learning and higher empathic functioning. Future research is needed to define the impact of early intervention on these aspects of social-emotional development.

In conclusion, with this study we hope to have created awareness of the impaired empathic abilities of the DHH child. This will severely affect their social relationships, because there is a strong positive association between empathy and friendship quality in both NH and DHH children.[45-48] Lower empathic abilities influence a child's social interaction, for example during play. For cooperative play with peers children need to share one another's goals, desires, and beliefs.[49] Not being able to empathize with the other may result in less participation in play with others, causing isolation in the DHH child.[50] For their socio-emotional development DHH children benefit from achieving sufficient language skills. Yet, it takes more to obtain sufficient empathic abilities. If these abilities are to improve more attention could be paid to these issues in rehabilitation programs and family support. Professionals should create awareness concerning empathic abilities in the child's surrounding . Parents and teachers can contribute to the development of empathic skills by actively involving the DHH child in emotion-evoking situations, or by talking about emotions more often. Future research should focus on the development of rehabilitation programs for DHH children that actively support the development of empathic abilities.

Future studies

The psychometric properties of the empathy questionnaire were satisfying with good reliability in both DHH children and their NH peers. However, to assure that DHH children are as capable as hearing children in understanding the items well, further psychometric properties will be useful to examine. Item response theory models can shed further light on issues such as measurement invariance, which includes differential item functioning. Because of power issues we were not able to perform this type of analyses. Future studies with a larger cohort of DHH children are needed to address these issues. Regarding the design of this study, we have to point out that cross-sectional data were used, which prevents us from drawing conclusions about causality. Therefore, we started longitudinal data collection to confirm the assumptions made here.

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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 positively related to social functioning and negatively related to behavioral problems. Nevertheless, there was no direct relation between language skills and the two social-emotional 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 language-related 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 field[13] 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 CI’s or 2 HA’s) and bimodal (1 CI 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

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 radio-frequency 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 social-emotional 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 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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CHAPTER 8

NEDERLANDSE SAMENVATTING

In Nederland komt gehoorverlies voor bij 1 tot 2 op de 1000 kinderen. In ongeveer een kwart van de gevallen is de oorzaak niet bekend. De mate van gehoorverlies kent een grote variatie. In dit proefschrift worden kinderen besproken met een gehoorverlies variërend van matig (40 dB, te vergelijken met het niet meer kunnen horen van een radio die zacht aan staat), ernstig (70-90 dB; te vergelijken met het niet meer horen van een vrachtwagen die met hoge snelheid passeert) tot doof (het kind kan geen geluid waarnemen). Afhankelijk van de mate van gehoorverlies komt een kind in aanmerking voor het dragen van hoortoestellen, dan wel voor implantatie van een cochleair implantaat (CI). Uit eerder onderzoek is bekend dat slechthorende kinderen vaker spraak- en taalproblemen ondervinden. Dit kan vele consequenties hebben voor hun sociaal-emotionele ontwikkeling. In dit proefschrift wordt getracht een link te leggen tussen het gehoorverlies van kinderen, de gevolgen hiervan op hun taal- en communicatievaardigheden, en het effect hiervan op hun sociaal-emotionele ontwikkeling.

In **hoofdstuk 1** wordt een inleiding gegeven over gehoorverlies bij kinderen. Mogelijke oorzaken maar vooral de gevolgen van gehoorverlies op de kinderleeftijd worden besproken. Onderzoek heeft uitgewezen dat de spraak- en taalproblemen die slechthorende kinderen vaak ondervinden vele gevolgen hebben voor hun sociaal-emotionele ontwikkeling. Deze gevolgen (onder andere communicatieproblemen en minder mogelijkheden tot sociaal leren) worden in dit eerste hoofdstuk besproken. Verder wordt in hoofdstuk 1 ingegaan op de invoering van de neonatale gehoorscreening, waardoor een eventueel gehoorverlies steeds eerder in het leven van een kind kon worden geïdentificeerd. Aan de hand van uitleg over de invloed van auditieve stimuli op het brein wordt het belang van vroege revalidatie van het gehoor duidelijk gemaakt.

Aandacht voor de sociaal-emotionele ontwikkeling van slechthorende kinderen is essentieel voor het toekomstperspectief van deze groep. Immers, de sociale vaardigheden van een kind bepalen hoe succesvol een kind is in het aangaan van vriendschappen en het onderhouden van relaties. Onderzoek op dit gebied wint steeds meer terrein. Echter, de studies die reeds verricht werden kennen beperkingen. Deze worden besproken in het eerste hoofdstuk. Deze beperkingen vormen de basis voor de diverse deelonderzoeken beschreven in hoofdstukken twee tot en met zes.

De eerste beperking die beschreven wordt in dit proefschrift gaat over hoe onderzoekers omgaan met missende waarden in hun (klinische) onderzoek; zogenaamde ‘missing data’. **Hoofdstuk 2** is een review van de meest recente klinische studies in drie grote KNO-tijdschriften waarbij gekeken werd of onderzoekers missing data rapporteerden en hoe ze hier vervolgens mee omgegaan zijn tijdens het analyseren van hun database. Hieruit bleek dat het overgrote deel van de onderzoekers geen melding deed van (zeker aanwezige) missing data. De overgebleven groep onderzoekers rapporteerden missing data wel, maar negeerden deze bevinding vervolgens veelal. Slechts een klein percentage van de onderzoekers paste zijn analyses aan na het vinden van missing data. In hoofdstuk

2 wordt uitgelegd wat de gevolgen hiervan kunnen zijn. Tevens wordt uitgelegd hoe een onderzoeker om kan gaan met missende waarden door gebruik te maken van multipele imputaties, een relatief nieuwe statistische techniek.

Het doel van **hoofdstuk 3** was het onderzoeken van de relatie tussen de taalvaardigheid, de communicatievaardigheden en het sociaal functioneren van slechthorende kinderen van wie het gehoorverlies op jonge leeftijd ontdekt is. De taal- en communicatievaardigheden van deze groep kinderen lagen gemiddeld meer dan 1 standaard deviatie onder het gemiddelde. Ook werd een lager niveau van sociaal functioneren en werden er meer gedragsproblemen gerapporteerd in deze groep kinderen. Verder werd er geen relatie gevonden tussen de leeftijd waarop het gehoorverlies ontdekt werd en het sociaal functioneren. Taal en communicatievaardigheden waren sterk aan elkaar gerelateerd. Tevens toont deze studie aan dat niet de taalvaardigheid, maar juist de communicatievaardigheden van deze slechthorende kinderen van belang zijn voor hun sociaal-emotionele ontwikkeling. Goede communicatievaardigheden hingen samen met beter sociaal functioneren en minder gedragsproblemen.

Hoofdstuk 4 bouwt voort op eerder onderzoek uit onze groep waaruit gebleken is dat kinderen met gehoorverlies meer kans hebben op het ontwikkelen van psychopathologie zoals symptomen van angst, depressie en gedragsproblemen. In hetzelfde onderzoek werden ook meer problemen gezien bij kinderen met conventionele hoortoestellen dan bij kinderen met een CI. In hoofdstuk 4 werd bij peuters en kleuters (1-5 jaar oud) met een CI het niveau van psychopathologie in kaart gebracht door middel van oudervragenlijsten. Aangetoond werd dat jonge kinderen met een CI evenveel psychopathologie vertoonden als normaalhorende leeftijdsgenootjes. Het unieke aan het onderzoek beschreven in hoofdstuk 4 is het feit dat de kinderen 3 jaar lang gevolgd zijn. Hierdoor kon de ontwikkeling over tijd beschreven en geanalyseerd worden. Ook de ontwikkeling van symptomen over tijd verschilde niet tussen de twee groepen. Wel kon door het longitudinale karakter van de studie het positieve effect van taal op de ontwikkeling van symptomen van psychopathologie aangetoond worden. In tegenstelling tot in hoofdstuk 3 waren we in dit hoofdstuk dan ook in staat om de causaliteit aan te tonen tussen communicatievaardigheden en de psychosociale ontwikkeling van kinderen. Hoe beter de ontwikkeling van taal- en communicatievaardigheden, hoe minder symptomen van angst en depressie gezien werden. Een goede taalontwikkeling op jonge leeftijd droeg bij aan het voorkómen van symptomen van agressie en hyperactiviteit.

In **hoofdstuk 5** werd een meer specifieke groep slechthorende kinderen bestudeerd: kinderen met matig gehoorverlies (35-70 dB). Dit is vrij uniek aangezien het meeste onderzoek wereldwijd gaat over kinderen met ernstig gehoorverlies. In dit hoofdstuk werden verschillende aspecten van Theory of Mind (ToM) geobserveerd en geanalyseerd. ToM is de mogelijkheid om je te kunnen verplaatsen in de gedachtewereld van een ander. Deze capaciteiten ontwikkelen zich met name rond de peuter- en kleuterleeftijd, onder andere door het observeren van anderen. Uit de studie beschreven in hoofdstuk 5 bleek dat het intentiebegrip van kinderen met matig gehoorverlies gelijk was aan dat van normaalhorende kinderen. Kinderen met matig gehoorverlies vonden het echter moeilijker om andermans wensen en gedachten (zogenaamde desires en beliefs) te begrijpen, helemaal als deze anders waren dan hun eigen wensen, of als deze gedachten niet klopten met de werkelijkheid. Verder laat deze studie zien dat de taalvaardigheden van kinderen met matig gehoorverlies binnen de normaalwaarden lagen. Hun communicatievaardigheden lagen echter verhoudingsgewijs veel lager. Hoge taal- en communicatievaardigheden waren in deze studie gerelateerd aan betere ToM vaardigheden. Ook werd aangetoond dat het patroon waarop ToM zich ontwikkelde in kinderen met matig gehoorverlies vaker afwijkend was dan dat van de controlegroep (kinderen zonder gehoorverlies).

De studie in **hoofdstuk 6** beschrijft het empathisch vermogen van slechthorende kinderen en tieners (9-16 jaar oud) vergeleken met normaalhorende leeftijdsgenoten. Zowel uit zelfrapportage, als uit observaties bleek dat de affectieve component van empathie gelijk was in beide groepen. Hiermee wordt bedoelt dat zowel kinderen met als zonder gehoorverlies ‘ besmet’ raakten door andermans emoties. Wanneer het echter gaat over cognitieve empathie, het begrijpen van andermans emoties, dan werd gezien dat slechthorende kinderen hier meer moeite mee hadden. Ze toonden dan ook minder prosociaal gedrag dan horende kinderen. Hierbij kan gedacht worden aan iemand anders helpen of troosten. Ook werd in deze studie gekeken naar de invloed van de omgeving op het empathisch vermogen van kinderen. Sociaal leren is immers de motor achter het aanleren van empathische vaardigheden. Hierin werd gezien dat slechthorende kinderen op het speciaal onderwijs meer moeite hadden met het begrijpen van andermans emoties (cognitieve empathie) dan slechthorende kinderen op het regulier onderwijs. Er werd echter geen verschil gezien in de mate van prosociaal gedrag.

De bevindingen uit de hoofdstukken twee tot en met zes komen samen in **hoofdstuk 7** waaruit drie hoofduitkomsten te herleiden zijn. Als eerste blijkt dat de taalontwikkeling bij veel slechthorende kinderen achterblijft. Slechts bij de kinderen met matig gehoorverlies in hoofdstuk 5 en de oudere kinderen in hoofdstuk 6 werden scores binnen de normaalwaarden gezien. Echter wanneer we kijken naar de communicatievaardigheden van slechthorende kinderen, dan zien we niet alleen dat deze lager zijn dan de gestandaardiseerde normscores, maar ook dat deze in verhouding overal lager zijn dan de taalscores van het kind. Dit leidt ons naar de tweede conclusie van dit proefschrift. Juist de communicatievaardigheden van een kind zijn belangrijk om conversaties met anderen aan te gaan. De communicatievaardigheden bepalen de mogelijkheden die een kind krijgt om sociaal gedrag te observeren en te leren. Dit verklaart waarom herhaaldelijk een sterke relatie gevonden wordt tussen de communicatievaardigheden en het sociaal functioneren van slechthorende kinderen. Ten derde wordt in hoofdstuk 7 ingegaan op het effect van vroege identificatie van en interventie bij gehoorverlies. Vroege interventie in de vorm van CI lijdt tot het verbeteren van de taal- en communicatievaardigheden van slechthorende kinderen. Er werd echter geen directe relatie gevonden tussen vroege identificatie/interventie van gehoorverlies en het sociaal-emotioneel functioneren van slechthorende kinderen. In dit afsluitende hoofdstuk worden mogelijke verklaringen gegeven waarom deze directe relatie vooralsnog niet aangetoond kon worden. Hoofdstuk 7 sluit af met nieuwe vragen die ontstaan zijn naar aanleiding van de beschreven onderzoeken en doet naar aanleiding van deze vragen enkele suggesties voor toekomstig onderzoek.

APPENDICES

ABBREVIATIONS

AN(C)OVA	Analysis of (Co-)Variance
BCD/BAHA	Bone Conduction Device *or* Bone-Anchored Hearing Aid
CDI	Child Development Inventory
CELF	Clinical Evaluation of Language Fundamentals
CI	Cochlear Implant
CONSORT	Consolidated Standards of Reporting Trials
dB	Decibel
DECIBEL	Developmental Evaluation of Children: Impact and Benefits of Early hearing screening strategies Leiden
DHH	Deaf and Hard of Hearing
DHS	Distraction Hearing Screening
EAQ	Emotion Awareness Questionnaire
ECI	Early Childhood Inventory
EEQ	Emotional Expressivity Questionnaire
EmQue-CA	Empathy Questionnaire for Children and Adolescents
fMRI	functional Magnetic Resonance Imaging
HA	Hearing Aid
HI	Hearing Impaired
LMM	Linear Mixed Model
LOCF	Last Observation Carried Forward
MAR	Missing At Random
MAN(C)OVA	Multivariate Analysis of (Co-)Variance
MCAR	Missing Completely At Random
(M)CDI	MacArthur-Bates Communicative Development Inventory
MHL	Moderate Hearing Loss
MI	Multiple Imputation
MLU	Mean Length of Utterance
MNAR	Missing Not At Random
MNS	Mirror Neuron System
NH	Normal Hearing
NHS	Newborn Hearing Screening
PCHL	Permanent Childhood Hearing Loss

RDLS	Reynell Developmental Language Scale
SD	Standard Deviation
SDQ	Strengths and Difficulties Questionnaire
SELT	Schlichting Expressive Language Test
SES	Socioeconomic Status
SPSS	Statistical Package for the Social Sciences
STROBE	Strengthening the Reporting of Observational Studies in Epidemiology
ToM	Theory of Mind
WISC	Wechsler Intelligence Scale for Children

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"I get by with a little help from my friends.."
John Lennon and Paul McCartney

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