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

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CHAPTER 1

INTRODUCTION

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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.

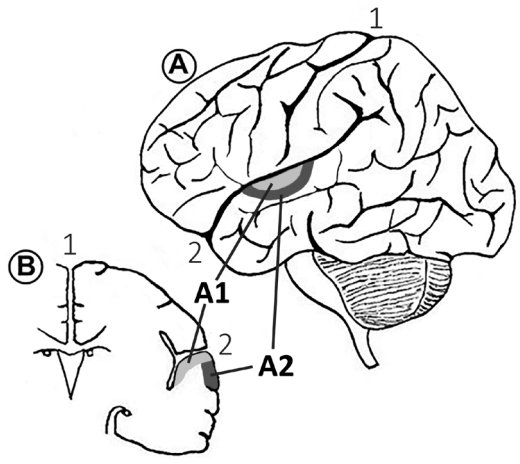

Figure 1 A. Lateral view of the brain (schematic). The primary auditory cortex (A1) is highlighted in light grey and the secondary auditory cortex (A2) is highlighted in dark grey. **B.** Coronal view.

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:

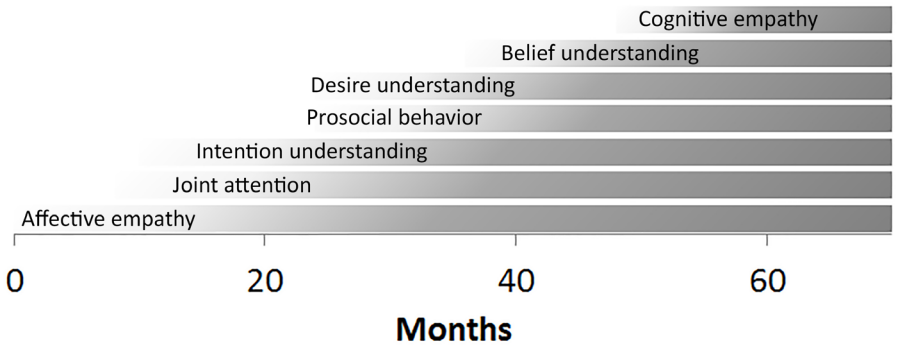

Figure 2. Schematic representation of the development of various milestones for social-emotional in childhood.

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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