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CHAPTER 1

Introduction



Hearing ability is important for children to develop language, communication, and social-emotional skills as they grow up. Approximately one child in a thousand is born with permanent bilateral hearing loss (HL) (Korver et al., 2011; Zoutenbier et al., 2016). In the Netherlands, this means about 180 children each year, of whom over 50% have a moderate HL (MHL)(40-70 dB) (Zoutenbier et al.). Permanent bilateral can be caused by genetic factors (40%), acquired factors (30%), unknown causes (25%), or miscellaneous causes (5%) (Korver et al.). In this thesis the impact of MHL on young children's language and social-emotional outcomes in the context of their caregiving environment is examined.

Children with HL encounter challenges while growing up in a sound-dominated society. Auditory access is an important factor in enabling human beings to communicate with others and receive social information. Spoken conversations, for example, are usually between 40 and 65 dB and thus have limited accessibility even for children with MHL. Hearing aids offer children with HL more access to sound and speech, but in noisy environments they still encounter difficulties in perceiving auditory information. This restricted access to sound and speech puts children with MHL at risk of developing language difficulties (Tomblin et al., 2015) and social-emotional difficulties (Stevenson et al., 2015; Theunissen et al., 2014).

Children with MHL: forgotten children?

Before the introduction of newborn hearing screening (NHS) (introduced in the Netherlands in 2005), deaf children were on average about one-and-a-half years old when their hearing loss was first identified, and children with MHL were even older. Nowadays, most children with HL are diagnosed within the first few months of life, which allows for much earlier intervention. This is important, because early family-centered intervention (FCEI) has been shown to have a positive effect on the language and social-emotional outcomes of children with HL (Ching et al., 2017; Holzinger, Fellinger, & Beitel, 2011; Moeller, 2000; Yoshinaga-Itano et al., 1998).

The Netherlands has a long tradition of family-centered early intervention for deaf children and their families. Following the introduction of the NHS in 2005, early interventionists who were used to working with deaf toddlers now had to adapt their programs to the needs of deaf babies and their families. In 2008, the inclusion criteria for family-centered early intervention for children with HL were broadened, allowing children with less severe HL (MHL) to enroll. However, it remained questionable whether family-centered interventions for deaf children would also be beneficial for children with MHL. Given that children with MHL have better auditory access than deaf children, other interventions might be more effective to promote their language and social-emotional development.

At that time, limited evidence was available to answer the question of whether the existing family-centered interventions were also suitable for children with MHL. Few studies focused specifically on children with MHL: most concerned children with profound HL or

included children with a broad range of HL (i.e., from mild to profound HL). The gap in research concerning and interventions for children with moderate to severe HL was apparent as early as 1977 when, in her seminal work, Julia Davis referred to this group as “Our forgotten children” (Davis, 1977). Davis advocated a stronger focus on MHL particularly because, since these children speak relatively well and often have successful interactions with others, adults may underestimate their needs. A study by Davis and colleagues (1986) on the outcomes of 40 children with mild to severe HL showed that these children were indeed at risk for language and social-emotional difficulties. Recently there has been increasing attention in the literature for young children with MHL (e.g., Ambrose et al., 2015; Koehlinger, Horne & Moeller, 2013; Laugen et al., 2016; Moeller & Tomblin, 2015; McCreery et al., 2015; Netten et al., 2017; VanDam, Ambrose & Moeller, 2012). The results of these studies have shown repeatedly that children with MHL are at a higher risk for language and social-emotional difficulties than their normal hearing peers.

Inconsistent access to linguistic and social-emotional input

The auditory environment in which children grow up shapes their language development. Children pick up speech from the people around them and learn language by interacting with them. For example, they learn to discriminate speech sounds, understand the meaning of words, produce words, and learn the rules of their language. When access to speech is restricted, children have reduced linguistic experiences and are consequently at risk for language difficulties. Several studies have reported lower language and speech abilities for children with MHL compared with peers with normal hearing (NH) (Ambrose Vandam & Moeller, 2014; Davis et al., 1986; Hammer & Coene, 2016; Koehlinger et al., 2013; Tomblin et al., 2015). These studies point to lower language abilities (Tomblin et al.), weaker consonant production (Ambrose et al., 2014), and poorer grammatical outcomes (Hammer & Coene; Koehlinger et al.).

Moeller and Tomblin and their team (2015) propose a model of inconsistent access (MIA) (See Figure 1), in which they hypothesize that children with mild to severe HL experience limitations in their access to linguistic input. This inconsistent access results in limited potential for linguistic uptake, which in turn increases the risk that children with MHL will miss out on opportunities to learn language. Over time, these limited opportunities accumulate and reduce the children’s cumulative linguistic experiences, which affects their language development. Moeller, Tomblin and colleagues used MIA as a central hypothesis in their longitudinal research project on the speech and language outcomes of children with mild to severe HL (OCHL) (for an overview of this research project see Tomblin et al., 2015). The researchers explored three factors that might influence these children’s access to linguistic input: aided audibility, consistency of hearing aid use, and caregiver linguistic input. The results of the OCHL project supported their hypothesis: Children with MHL were shown to have poorer language outcomes than their peers with NH, and these outcomes were related to the three factors proposed.

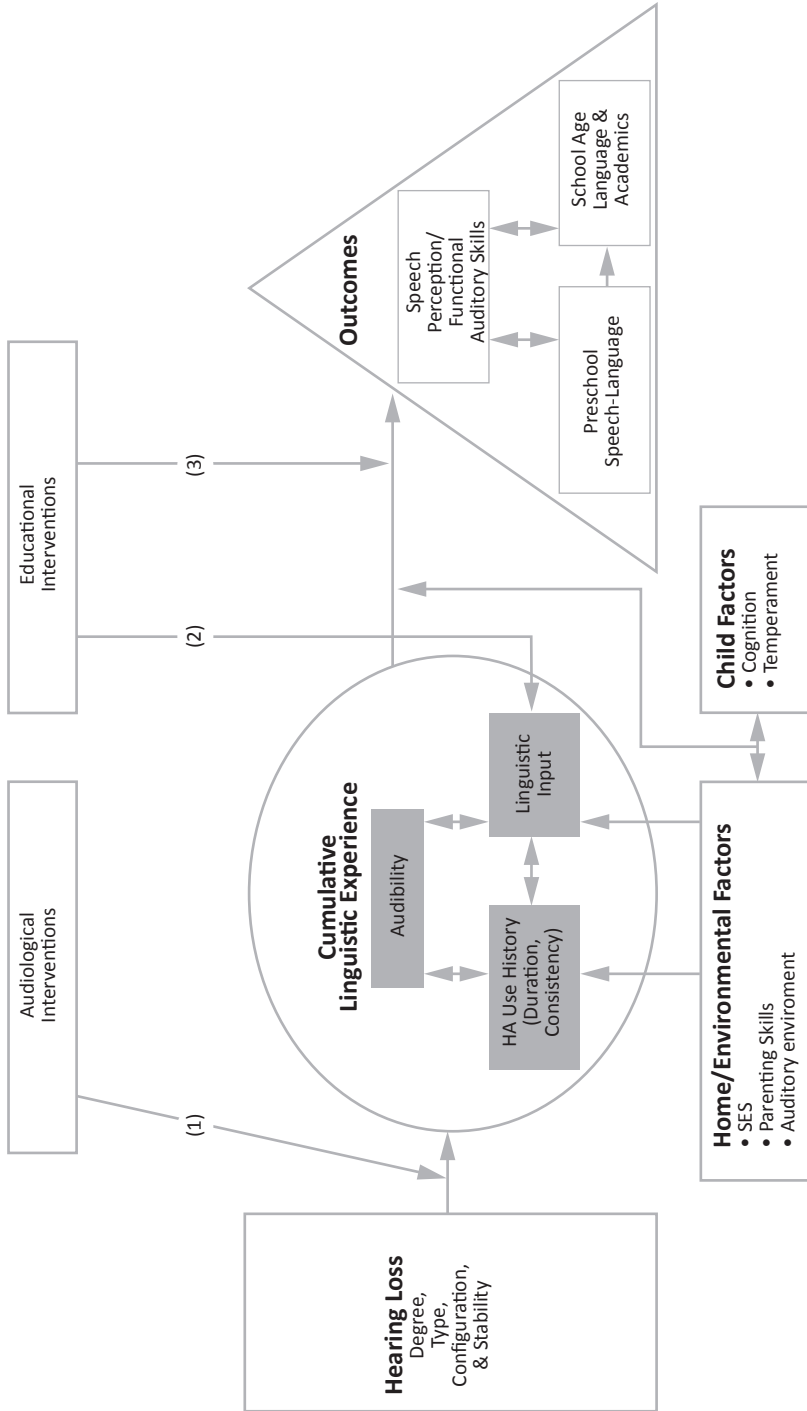


Figure 1. The model of inconsistent access (Moeller & Tomblin, 2015)

In considering the development of children with MHL, it is also important to take their social-emotional experiences and outcomes into account; this involves broadening the scope and relevance of Moeller et al.'s model. In the present research, therefore, we aim to expand the model to include social-emotional experiences and outcomes (see Figure 2). To acquire the social-emotional skills necessary to build and maintain meaningful interactions with other people, children need input from knowledgeable others in the early years. However, this type of input too is less accessible for children with MHL. Their hearing loss prevents them from overhearing conversations, for instance, so that they miss out on social information that others are privy to (incidental learning). Further, it is more difficult for them to join in interactions among peers when there is background noise, in environments such as daycare or playgrounds (Rieffe et al., 2017). However, children need opportunities to engage in these social interactions to learn about the emotions, intentions, and perspectives of others.

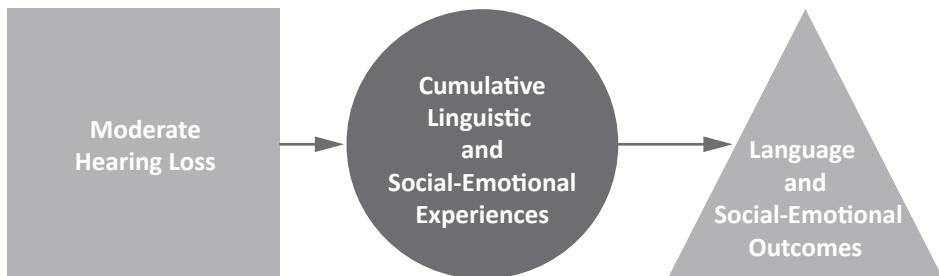


Figure 2. Reduced cumulative linguistic and social-emotional experiences.

In order to build meaningful relations with others it is essential to feel and understand the emotions of people around you. This ability, empathy, is often regarded as the social glue in relationships, because it facilitates social bonding (Hofman, 1990). The first stage of empathy, *affective empathy*, refers to feeling what the other is feeling; this stage of empathy is believed to be innate. The next stage, *cognitive empathy*, is to understand why the other is feeling that way. This capacity is stimulated by interacting with others and observing how others interact. Given their limited opportunities for incidental learning, we might expect children with MHL to have difficulties in understanding the feelings and actions of others.

Based on, and extrapolating from the model of inconsistent access, we might expect that children with MHL would experience limitations in their access to social-emotional input, which might lead to a decrease in social-emotional experiences, resulting in poorer social-emotional outcomes. Recent studies have shown that preschool children with MHL have more behavioral problems (Netten et al., 2015), problems with peers (Laugen et al., 2016), difficulties in social functioning (Netten et al., 2015), and delays in theory of mind

development (Netten et al., 2017) than their hearing peers. Further, in studies including children with MHL together with children with severe and profound HL, higher rates of psychosocial difficulties are reported among children with HL, and the outcomes were not affected by the degree of HL (e.g., Dammeyer, 2010; Kouwenberg et al., 2012; Leigh et al., 2015; Netten et al., 2016; Stevenson et al., 2010; Theunissen et al., 2014; Wong et al., 2017). Since most studies focused on the preschool or school age group, it is not clear whether these difficulties already emerge at a younger age.



Audiological interventions

The first few years of a child's life are critical for language development (Kuhl, 2010), so early auditory stimulation is very important. Audiological interventions such as the amplification of hearing aids have the potential to reduce the risks for impaired language development (Figure 3). Well-fitted hearing aids provide children with better audibility, and greater audibility allows for more linguistic experiences, which in turn is related to better language outcomes (McCreery et al., 2015).

Early hearing aid fitting (before six months) has been shown to be related to better speech and language outcomes in comparison with later fitting (Ambrose et al., 2014; Sininger, Grimes & Christensen, 2010; Tomblin et al., 2015). Also, children with more consistent daily hearing aid use have better language outcomes than children with less consistent use (Walker et al., 2015).

Not all children use their hearing aids consistently, and not all devices are fitted optimally, which means that children with MHL still have inconsistent access to sounds and speech.

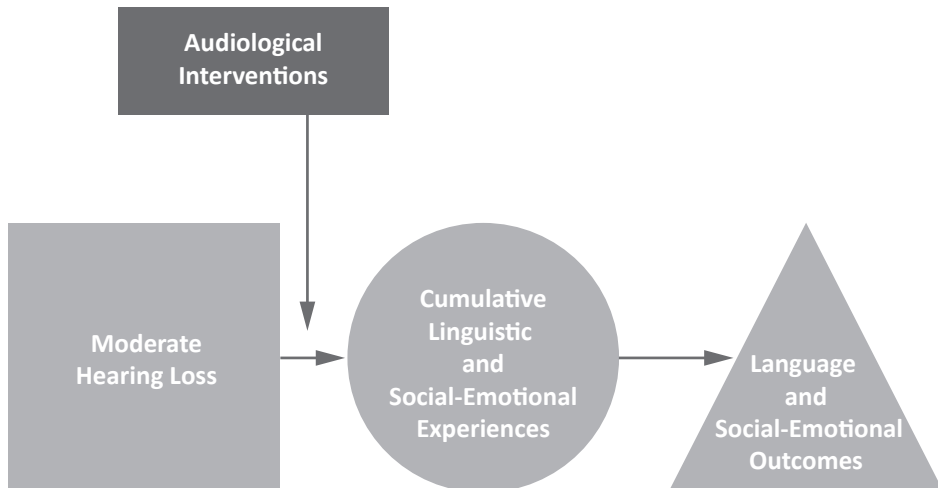


Figure 3. Audiological interventions: access to linguistic and social-emotional input

Moreover, in noisy environments it is difficult for these children to pick up all speech clearly even if they are using a hearing aid (Finitzo-Hieber & Tillman 1978; McCreery & Walker, 2017; Stelmachowic et al., 2004). This inconsistent access results in a reduced potential for linguistic uptake, which in turn increases the risk of children with MHL missing out on opportunities to learn language. Further, this inconsistent access to acoustic cues will also reduce opportunities for social-emotional learning.

The caregiving environment

In the early years of child development the caregiving environment is an important context in which learning takes place. Children acquire their knowledge and skills by interacting with family members and other caregivers. In later interactions with peers at school, in the neighborhood, or at sport clubs, children will benefit from these skills they acquired in early childhood. According to the social-ecological model, children's development is affected by their social relationships and the world around them (Bronfenbrenner, 1979).

In the model of inconsistent access home and environmental factors such as socio-economic status and parenting skills are believed to contribute to the linguistic and social-emotional experiences of children with HL (see Figure 4). Family's socio-economic status and parental educational levels have been related to a range of developmental concerns in children (e.g. Bornstein & Bradley, 2014; Hart & Risley, 1995). Children with HL who were raised in lower-income families had lower language abilities than children raised in high-income families (Ching & Dillon, 2013). Children whose mothers had higher levels of education used their hearing aids more hours a day than children whose mothers had lower levels of education (Walker et al., 2015).

Parents¹ play a crucial role in the development of their children. In the early years, when brain neuroplasticity is the greatest (Sharma, Campell & Cardon, 2015), children spend most of their time with their parents. Therefore, brain neural development may be particularly sensitive to caregiving influences during this period. Research has shown that early parent-child interactions are associated with children's language and social-emotional outcomes (Dunn, Brown & Slomkowski, 1991; Fay-Stammbach, Hawes & Meredith, 2016; Kok, Lucassen & Bakermans-Kranenburg, 2015; Moreno, Klute & Robinson, 2008; Quittner et al. 2013). For example, warm and sensitive parenting contributed to better language ability and more positive empathic child behavior (Moreno et al.).

Most children with HL have parents without HL (Mitchell & Kachmer, 2004). For these parents the HL of their child is often their first experience with HL. Parents are faced with

1 The term parents is used to refer to mothers, fathers, and other caregivers

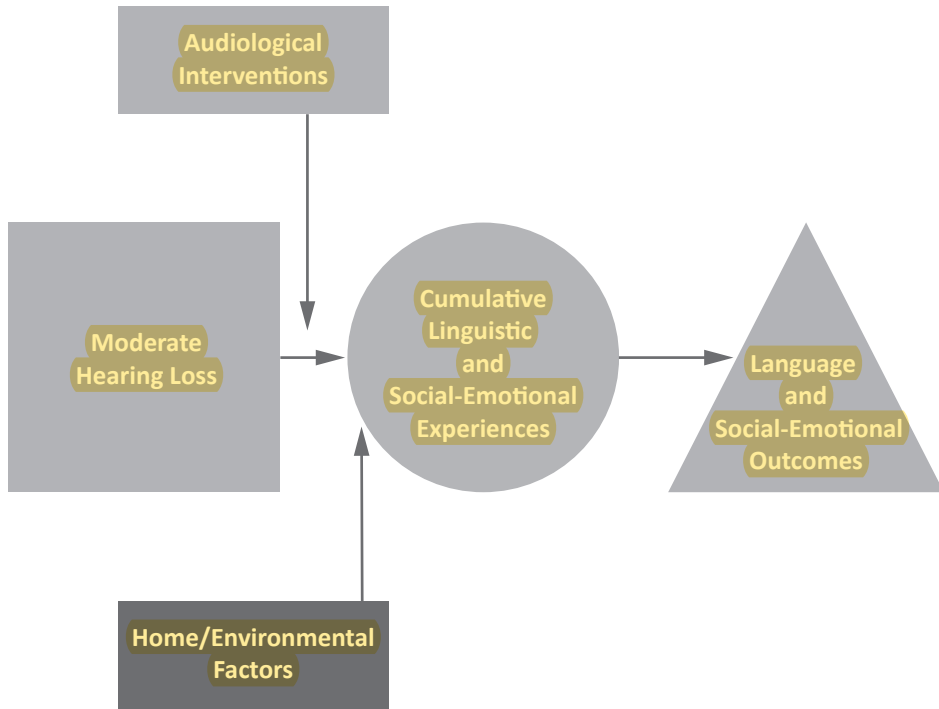


Figure 4. Home and environmental factors: access to linguistic and social-emotional input

challenges such as communication, hearing aids, and various changes in daily routines. These challenges and the associated concerns might result in higher rates of parental stress and less positive parent-child interactions. Various studies have reported associations between parental stress and various child factors such as language ability and social-emotional skills; however, in fact most found similar levels of parental stress in parents of children with and without HL (e.g. Calderon & Greenberg, 1999; Hintermair, 2000, 2006; Meadow-Orlans, 1994; Pipp-Siegel, Sedey & Yoshinaga-Itano, 2002; Stika et al., 2015; Topol et al., 2011). Nevertheless, it is important to note that none of these studies focused specifically on children with MHL.

In general, most parent-child-related studies in the population of children with HL included children with MHL together with children with more severe HL, or focused solely on children with profound HL. Research on the interactions between children with HL and their parents has generally shown more difficulties compared with their peers with NH: parents tended to be more directive and less flexible (see Pressman, Pipp-Siegel & Yoshinaga-Itano, 1999); episodes of joint engagement were briefer (Barker et al., 2009; Cejas et al. 2014; Lederberg & Mobley, 1990; Prezbindowski, Adamson & Lederberg, 1998); and parents were less sensitive (Quittner et al., 2013). These difficulties might impede children with HL in obtaining the linguistic and social-emotional input they need.

Moreover, the way parents talk to their children affects children's language ability and social-emotional skills. Both the amount of talk and the quality of parental talk have been related to children's language abilities (Hart & Risely, 1995; Rowe, Leech, & Cabrera, 2017). Children with MHL may have inconsistent access to parental talk, because they do not hear all speech well enough (because of not wearing a hearing aid and/or because of background noise). Further, they may have inconsistent access to high-quality talk because their parents do not use this kind of talk with them. High-quality talk such as asking open-ended questions, expansion and recasting is supposed to be language evoking (Desjardin et al., 2014).

Parents of children with MHL may adapt their own language level and provide less complex language in response to the lower language abilities of their children. One study showed that children with mild to severe HL were exposed to more directing language (low-quality talk) and less high-quality language by their parents than their peers with NH (Ambrose et al., 2015). In other studies, directing language was associated with lower language abilities, while high-quality language was associated with better language abilities (Ambrose et al.; DesJardin et al., 2014). In addition, parents of children with HL were shown to use less mental-state language (e.g. think, know, believe, remember, want) during interactions than parents of children with NH (Moeller & Schick, 2006; Morgan et al., 2014), which in turn was related to children's theory of mind development (Moeller & Schick). To conclude, the caregiving environment is one of the factors that is likely to contribute to the linguistic and social-emotional input to children with MHL.

Family-centered early interventions

Although the focus of their studies was on audiological interventions, Moeller and Tomblin (2015) included a key role for educational interventions in their model of inconsistent access. Because the present thesis concerns children with MHL in their early years, the focus will be on family-centered interventions (see Figure 5). FCEI programs aim to support families with a child with HL to achieve the best outcomes. Early interventionists provide parents with information about HL and support parent behaviors that promote the language and social-emotional development of children with HL.

The younger the age of children with HL when they and their parents first receive support, the better the language and social-emotional outcomes (Ching et al., 2017; Holzinger, Fellingner & Beitel, 2011; Meizen-Derr, Wiley & Choo, 2011; Moeller, 2000; Yoshinaga-Itano et al., 1998). Children who enrolled in FCEI within the first six months of life had better language outcomes than children who enrolled after six months (Yoshinaga-Itano et al.). Further, high levels of parental involvement within FCEI programs correlated with positive language outcomes (Moeller).

We are especially interested in establishing which specific elements of FCEI – for instance, interventions that enhance parents' use of language-evoking strategies – are effective in

enhancing child outcomes. Although several studies have shown the importance of linguistic input for children’s language ability, there is less evidence for specific interventions that promote parents’ communication strategies.

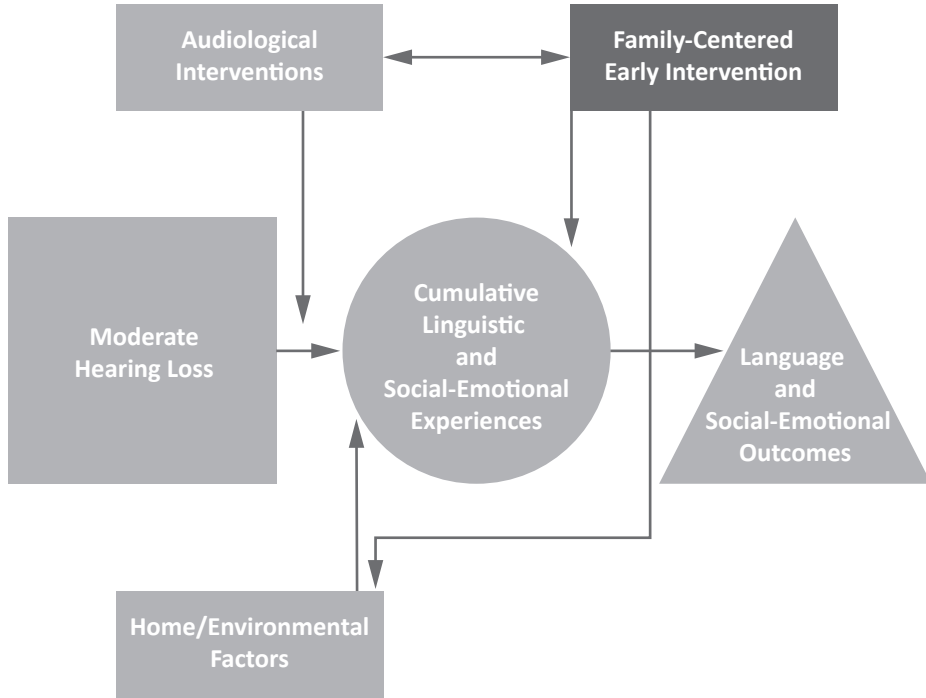


Figure 5. Family-centered early interventions: access to linguistic and social-emotional input

Rationale and outline of this thesis

Children with MHL and their families first enrolled in family-centered early intervention in the Netherlands in 2008. On the basis of the literature on children with MHL at that time, it was difficult to obtain a clear picture of their needs. Therefore, in 2009 the Dutch Foundation for the Deaf and Hard of Hearing Child (NSDSK) started a research project on the psychosocial functioning of young children with MHL and their caregiving environment. The outcomes of this project are presented in this thesis. Recently there has been more focus on this group of children, although still relatively little compared to deaf children (with or without cochlear implants). As discussed above, the few studies that have been conducted with children with MHL have shown these children to be at risk for language and social-emotional difficulties. However, most studies have focused on the preschool age, and not all participating children had benefitted from early screening and intervention. Furthermore, most studies have focused on child language outcomes. Insight into different

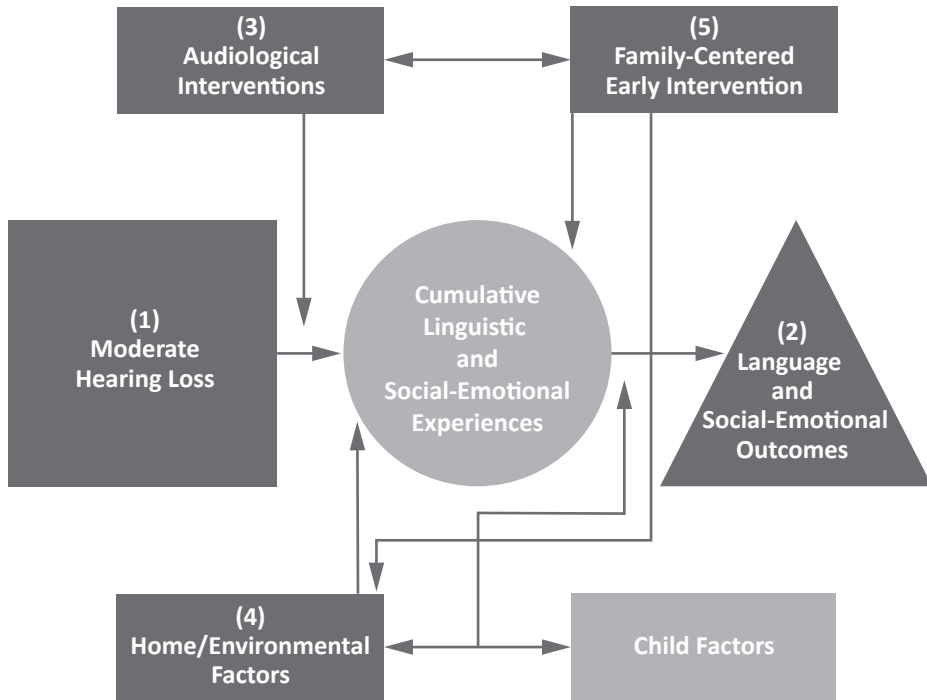


Figure 6. The expanded and adapted model of inconsistent access

developmental domains of young children with MHL who have been identified through newborn hearing screening and benefitted from early intervention can facilitate the identification of future challenges and opportunities for these children and their families.

In this thesis, the expanded and adapted model of inconsistent access (Figure 6) was used as a framework to examine the language and social-emotional outcomes of toddlers with MHL within the context of their caregiving environment. Further, we examined the effect of elements of FCEI on child outcomes and the caregiving environment. Child factors such as temperament, cognition, and additional disabilities may also create challenges for the caregiving environment to provide linguistic and social-emotional input. However, although these child factors play an important role in MIA, they were not taken into account in the current thesis.

Four chapters in this thesis concerned a study sample of children with MHL and NH with ages ranging from 17 to 45 months old. The children with MHL were recruited at three centers for FCEI in the Netherlands, and the children with NH via a well-baby clinic. The data were collected between 2009 and 2012. A fifth chapter concerned a study on the effect of an interactive reading program on parent behavior and was conducted in a

sample of parents with children with moderate to profound HL aged between 20 and 46 months old. These children and their parents were recruited at three centers for FCEI, and the data were collected between 2013 and 2014.

Chapter 2 describes a study that explored the empathy levels of toddlers with MHL compared to normally hearing peers; this was assessed by means of a parent questionnaire and observation tasks. Empathy, the ability to feel, understand, and respond affectively to the emotions of others, is an important aspect of social-emotional functioning. In addition to empathy levels, the relationship between empathy and language abilities was studied.

Chapter 3 concerns the relationship between family factors and the language ability and social-emotional functioning of young children with MHL. Specifically, the study described in this chapter examined the amount of (perceived) parental stress and social support in parents of children with MHL compared to parents of children without HL. We studied the associations between perceived parental stress and social support, children's language and social-emotional outcomes, and hearing loss-related variables.

Chapter 4 examines parent-child interaction in toddlers with and without MHL. Observations of a free-play session of parent and child were used to examine the levels of emotional availability and joint engagement in the interactions. In addition, these parent-child interaction measures were studied in relation to children's language abilities.

Chapter 5 describes a study in which the quantity and quality of parental linguistic input to toddlers with MHL was compared to that of their hearing peers. In addition, we examined the associations between the amount and quality of linguistic input, children's language abilities, and hearing-loss-related variables.

Chapter 6 concerns a study examining the effects of an interactive reading program for parents of toddlers with HL. We examined changes in parents' interactive reading behavior before and after the program and compared this behavior with that of parents who did not participate in the program.

Chapter 7 presents a summary of the main findings of this project. The outcomes of the previous chapters are then integrated and their implications are discussed. The thesis concludes with clinical implications and suggestions for future research.

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CHAPTER 1

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