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Beyond hearing : social-emotional outcomes following cochlear implantation in young children

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Citation

Ketelaar, L. (2014, June 4). *Beyond hearing : social-emotional outcomes following cochlear implantation in young children*. Retrieved from <https://hdl.handle.net/1887/25886>

Version: Corrected Publisher's Version

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Downloaded from: <https://hdl.handle.net/1887/25886>

Note: To cite this publication please use the final published version (if applicable).

Cover Page



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The handle <http://hdl.handle.net/1887/25886> holds various files of this Leiden University dissertation.

Author: Ketelaar, Lizet

Title: Beyond hearing : social-emotional outcomes following cochlear implantation in young children

Issue Date: 2014-06-04

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**Beyond hearing:
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Lizet Ketelaar

ISBN: 978-90-6464-778-9

Cover design: Paul Ketelaar / Ferdinand van Nispen

Layout: Ferdinand van Nispen, Citroenvlinder-DTP.nl, Bilthoven, The Netherlands

Printed: GVO drukkers & vormgevers B.V. | Ponsen & Looijen, Ede, The Netherlands

The studies reported in this thesis were conducted at the department of Developmental and Educational Psychology, Leiden University, The Netherlands; in collaboration with NSDSK (Dutch Foundation for the Deaf and Hard of Hearing Child), Amsterdam, The Netherlands; department of Otorhinolaryngology, Leiden University Medical Center, Leiden, The Netherlands; and Center for Autism, Oegstgeest, The Netherlands.

This research was financially supported by ZonMw.

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**Beyond hearing:
Social-emotional outcomes
following cochlear implantation in young children**

PROEFSCHRIFT

ter verkrijging van
de graad van Doctor aan de Universiteit Leiden,
op gezag van Rector Magnificus prof.mr. C.J.J.M. Stolker,
volgens besluit van het College voor Promoties
te verdedigen op woensdag 4 juni 2014
klokke 15.00 uur

door

Lizet Ketelaar
geboren te Alphen aan den Rijn
in 1980

Promotiecommissie

- Promotores:** Prof.dr. C. Rieffe (Universiteit Leiden; NSDSK)
Prof.dr.ir. J.H.M. Frijns (Leids Universitair Medisch Centrum)
- Co-promotor:** Dr. C.H. Wiefferink (NSDSK)
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Prof.dr. L. Surian (University of Trento)

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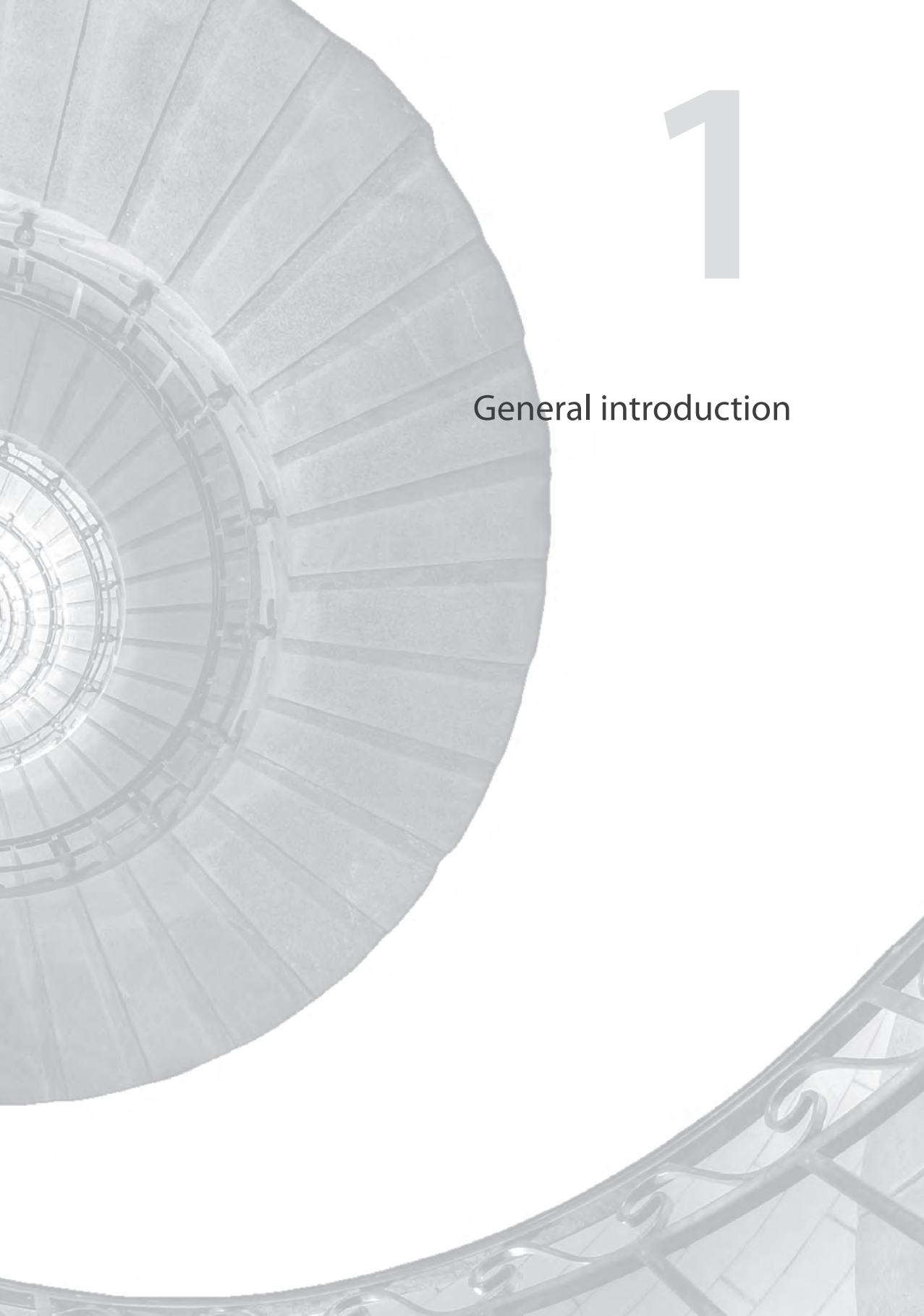




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General introduction



With a prevalence of around 1 per 1000 newborns (Korver, 2010; Watkin & Baldwin, 2011), each year approximately 150 to 200 children in the Netherlands are born with a hearing impairment, half of whom have a severe (61-90 dB) or profound (>90 dB) hearing loss (Korver, 2010). The neonatal hearing screening program, implemented in the Netherlands between 2002 and 2006, enables early diagnosis of congenital deafness and offers the possibility for early intervention. Today's intervention strategies, at least for young children with severe to profound hearing loss born in Western countries, usually involve implanting an electronic device called a cochlear implant (CI) in the cochlea (De Raeve & Lichtert, 2011; Hyde & Power, 2006). Instead of merely amplifying sounds like a traditional electro-acoustic hearing aid, a CI converts sounds into electrical pulses that directly stimulate the auditory nerve via an electrode array inserted in the cochlea, bypassing the damaged part of the ear and enabling individuals with hearing impairments (HI) to perceive sounds.

To date, the majority of research has been dedicated to the 'primary' outcomes of cochlear implantation, such as sound perception and speech and language development. Results from these studies generally indicate considerable improvements in these areas following cochlear implantation, particularly for children implanted before the age of 2 (Boons et al., 2012a; Connor, Craig, Raudenbush, Heavner, & Zwolan, 2006; De Raeve, 2010; Ganek, Robbins, & Niparko, 2012; Nikolopoulos, Archbold, & O'Donoghue, 1999; Niparko et al., 2010; Svirsky, Robbins, Kirk, Pisoni, & Miyamoto, 2000; Svirsky, Teoh, & Neuburger, 2004). However, it has also become apparent that providing HI children with a CI does not fully restore normal hearing, and therefore these children are still faced with difficulties in the auditory domain. Notably, in spite of improvements in language skills following cochlear implantation, many children still have significant language delays compared to normal-hearing (NH) peers (cf. Ganek et al., 2012). Moreover, children with CI have difficulties perceiving speech in noisy situations (Schafer & Thibodeau, 2006), and complex sounds such as music (Stabej et al., 2012). Children with CI also tend to be less sensitive to prosodic aspects of language like intonation (Most & Michaelis, 2012), which inform the listener about the correct interpretation of the message and about the speaker's emotional state.

In comparison to the overwhelming body of literature concerning auditory- and language-related outcomes of cochlear implantation, relatively little is known about the 'secondary' outcomes, that is, how having a CI affects



children's social-emotional functioning in daily life. It is well known that HI children (without CI) more often experience social-emotional difficulties than NH children (for a review, see Theunissen et al., 2014), yet to date few studies have examined whether these difficulties are also present in HI children who have received a CI. Moreover, the studies that have been conducted vary a lot with respect to important aspects such as sample size, comparison group, informant, age at implantation, and so on. Consequently, these studies have produced mixed findings, with some claiming substantial improvements in certain aspects of social-emotional functioning following implantation (e.g., Hopyan-Misakyan, Gordon, Dennis, & Papsin, 2009; Huttunen et al., 2009; Remmel & Peters, 2009; Tasker, Nowakowski, & Schmidt, 2010), and others reporting persisting delays in comparison to NH peers (e.g., Macaulay & Ford, 2006; Peterson, 2004; Wang, Su, Fang, & Zhou, 2011; Wiefferink, Rieffe, Ketelaar, De Raeve, & Frijns, 2013; Wiefferink, Rieffe, Ketelaar, & Frijns, 2012a).

Technology keeps evolving and as a result of that HI children are implanted at ever-younger ages¹ with more sophisticated devices, which should increase their chances for optimal development. In order to establish whether these advances in technology indeed make a healthy social-emotional development possible we have to monitor these children's development. Only then will the professionals who counsel these children be able to adequately target those areas of development that require attention. Therefore, the studies included in this thesis all aim to shed light on aspects of social and emotional functioning in a group of young HI children who have received their implant(s) relatively early in life, in order to give an overview of the status quo in the Dutch population of children with CI.

This chapter will first introduce social-emotional development from a functionalist perspective. Subsequently, an account of what is known about the social-emotional development of HI children with and without CI will be provided, followed by a discussion of two factors which play a central role in children's social-emotional development: parents and communication. Next, the heterogeneity of the population of HI children will be highlighted before the chapter is concluded with the main research questions and an outline of the studies included in this thesis.

¹ Currently, the standard in the Netherlands is to implant children before their first birthday.

A Functionalist Perspective on Social-Emotional Development

Emotions signal that something in the environment needs our attention, and they induce changes in bodily systems such as the muscular and nervous system. For example, facial muscles contract, heart rate increases and blood flows to extremities in order to prepare us to quickly deal with the situation at hand (Levenson, 1999; Scherer, 2000). The 'fight-or-flight' response, aimed at survival of the individual, may be the best known example of an action tendency that can be triggered, but there is a host of other goals (personal as well as social) that may be evoked by a situation (Gross & Thompson, 2007).

Besides alerting us that something is at stake, emotions also have a strong communicative function (Parkinson, 1996; Scherer, 2000). In the first few months of life, infants start to express a range of emotions, including happiness, anger, sadness, and fear. These emotions, along with surprise and disgust, are called basic emotions. They develop at an early age and are assumed to be innate and universal (Izard, 1991). At first, the expression of an emotion is a mere reflection of the infant's internal state, but infants soon learn to appreciate the communicative value of emotions, and will start to express emotions intentionally. They quickly learn that smiling will lead to positive responses from caregivers, and that crying will lead to comforting and attention to their needs.

Emotions are often caused by a social event, and in turn exert an influence on the social situation at hand. The expression of emotions informs our surroundings of our intentions, needs, goals, and so on (Keltner & Haidt, 1999). These emotional messages do not stand alone, but are part of a chain reaction (Gross & Thompson, 2007; Scherer, 2000); the communication partner is affected by the perceived emotion (for example, sadness) of the sender, which may trigger an emotion (empathy) and corresponding action tendency (comforting) in the partner. This chain reaction in turn can have an effect on the social relationship between the communication partners (strengthening of the friendship). From this example, it becomes apparent that emotional functioning and social functioning are difficult to distinguish from one another. Emotional functioning refers mainly to the extent to which one's own and other people's emotions are understood and to the ability to regulate one's emotions, whereas social functioning involves the extent to which one is able to adaptively interact with others.



Children's emotional competence increases with age (Pons, Harris, & de Rosnay, 2004). Although infants are already able to experience and express emotions such as happiness upon seeing a familiar person and sadness or anger in case of physical discomfort, they do not fully comprehend these emotions yet. With increasing age, children gradually learn to label emotions, to link them to the situations that evoked them, and to consciously regulate (e.g., mask or tone down) the expression of emotions according to the demands of the (social) situation (Denham et al., 2003; Pons et al., 2004; Saarni, 1999). Besides the challenge of dealing with their own emotions, there is the additional challenge of attending to other people's emotions and learning to interpret these correctly. Together, these emotion skills provide a foundation for children's social functioning across different settings, such as home, neighborhood and school (Calkins, Gill, Johnson, & Smith, 1999; Denham et al., 2003).

The most important developmental task that young children face is how to successfully interact with people in their surroundings, and especially with their peers (Denham et al., 2003). In other words, children need to become socially competent. Social competence is associated with success in other areas of development, such as school readiness and achievement (Guay, Boivin, & Hodges, 1999; Welsh, Parke, Widaman, & O'Neil, 2001; Wentzel, 1991; Ziv, 2013), and is a key contributor to mental health and well-being later in life (Burt, Obradovic, Long, & Masten, 2008; Denham et al., 2003). Children who are able to regulate their own emotions and, equally important, who are able to acknowledge other people's emotions are more likely to show adequate behavioral responses in social situations. Conversely, children with underdeveloped emotion skills are more likely to bluntly express their emotions, without regard for the other person's feelings. Consequently, emotionally competent children are often better liked by other people and regarded as more socially competent than children with less sophisticated emotion skills (Denham et al., 2003; Diener & Kim, 2004).

Social-Emotional Development of Hearing-Impaired Children

In general, typically developing children seem to acquire social-emotional skills effortlessly over time. However, it takes a well-oiled machine for all of these skills to develop age-appropriately. This becomes clear when turning to the atypically developing group of HI children, who have been found to experience considerable social-emotional difficulties. For example, in comparison to NH children, HI children are reported to show more behavior problems (Barker et al., 2009; Theunissen et al., in press; Van Eldik, Treffers, Veerman, & Verhulst, 2004), to more often experience symptoms of depression and/or anxiety (Kouwenberg, Rieffe, & Theunissen, 2011; Theunissen et al., 2011; Theunissen et al., 2012; Van Eldik et al., 2004; Van Gent, Goedhart, Hindley, & Treffers, 2007), and to have difficulties with peer relations (Cappelli, Daniels, DurieuxSmith, McGrath, & Neuss, 1995; Kouwenberg, Rieffe, Theunissen, & de Rooij, 2012; Wolters, Knoors, Cillessen, & Verhoeven, 2011). Most of these studies have been conducted with school-aged HI children who were not fitted with a CI, but these findings may nonetheless be indicative of the social-emotional functioning of children with CI. As children with CI were sound-deprived prior to implantation and a CI does not fully restore normal hearing, their social-emotional development could still be impaired.

Now let us turn to some abilities that may lie at the root of these poor outcomes in HI children. A core ability which motivates people to behave well towards others is *empathy*: the ability to sympathize with other people and to respond to their needs. Empathy is often referred to as the 'social glue' in human relationships, in the sense that it prompts people to help and not harm each other, and thus promotes social relationships (Baron-Cohen & Wheelwright, 2004; Eisenberg & Miller, 1987; Jolliffe & Farrington, 2006). Obviously, a blatant disregard of a friend's distress would not do the friendship any good, whereas an empathic response shows that you care, which will strengthen the bond between you. Newborns already show signs of empathy in their reflexive crying in response to another infant's crying sounds (McDonald & Messenger, 2011). These early behavioral responses have led researchers to suggest that the predisposition to experience empathy is hardwired in the human brain (Decety & Meyer, 2008). Witnessing another person's emotion automatically triggers the so-called mirror neuron system, which makes the neurons fire as if the onlooker



is experiencing the emotion oneself (Preston & de Waal, 2002). In children, but also in adults, this mirroring of emotions can, for example, be seen when the onlooker also flinches as if in pain in response to seeing someone stumble and fall. This affective catching of the other person's emotion allows the onlooker to experience what the person in distress is experiencing, and ought to make it easier for the onlooker to respond appropriately.

Assuming that a neurological basis for empathy indeed exists, no differences in the ability to experience empathy should be expected to emerge between HI children and NH children. Yet, empathy is supposed to involve not only an affective but also a cognitive component (Baron-Cohen & Wheelwright, 2004), which makes the story of HI children's empathy development a bit more complex. The cognitive component of empathy denotes the ability to understand the other person's distress (Jolliffe & Farrington, 2006). In other words, it reflects the capacity to consider the other person's perspective. In typically developing children, this cognitive component usually kicks in during the second year of life with the ability to differentiate between self and other. Children realize that it is the other person who is experiencing the distress, which is a starting point to make inferences about the origin of the distress (Hoffman, 1987). Identifying its cause allows children to select the appropriate prosocial response (e.g., comforting, distracting, sharing) to alleviate the distress. Although prosocial responses can already be observed in 1-year-olds, these responses increase both in frequency and sophistication over time (Zahn-Waxler, Radke-Yarrow, Wagner, & Chapman, 1992). For example, a 1-year-old child might understand that a crying playmate needs comforting and will call his own mother. A 3-year-old child, on the other hand, might fetch the playmate's mother because he understands that this will be much more comforting to his playmate.

The cognitive component of empathy is closely related to, and sometimes even used interchangeably with *Theory of Mind* (ToM), as both involve perspective-taking skills. Whereas there is a paucity of studies concerning empathy in HI children, a by comparison almost overwhelming amount of studies describes ToM in this population. These studies (e.g., Peterson, 2009; Peterson & Siegal, 2000; Russell et al., 1998; Terwogt & Rieffe, 2004; Woolfe, Want, & Siegal, 2002) convincingly point to a significantly delayed development of perspective-taking skills in HI children compared to NH children. This could imply that, even though the predisposition for empathy is inborn, impaired perspective-taking skills might nonetheless hamper HI children's empathy development.

Beyond the capacity to understand the origins of other people's distress, ToM refers to the wider notion that other people's actions and emotions are the result of their inner thought processes, i.e., their intentions, desires and beliefs, which are not necessarily consistent with reality or with the onlooker's preferences or goals (Wellman, 1990). The ability to ascribe mental states to other people makes it possible to predict and make sense of other people's behavior, and to coordinate one's own responses accordingly. ToM is considered to be essential in order to attain social competence, which is illustrated by studies showing that well-developed ToM skills are associated with higher levels of peer acceptance and positive social interactions (Slaughter, Dennis, & Pritchard, 2002; Watson, Nixon, Wilson, & Capage, 1999), whereas deficits in ToM are associated with peer problems and behavior problems (Caputi, Lecce, Pagnin, & Banerjee, 2012; Olson, Lopez-Duran, Lunkenheimer, Chang, & Sameroff, 2011).

While typically developing children show a dramatic increase in ToM understanding between the ages of 2 and 5 (Wellman, Cross, & Watson, 2001), HI children generally reach a fully-fledged ToM at a much later age, lagging several years behind their NH peers (for an overview, see Peterson & Siegal, 2000). Albeit still limited, ToM development in children with CI has received a fair amount of attention over the last couple of years. Whereas studies conducted with HI children without CI have consistently found a delay in ToM development, findings from studies conducted with children with CI are not as straightforward. Some studies report that ToM skills of children with CI are on a par with those of NH children (Rommel & Peters, 2009; Tasker et al., 2010; Ziv, Most, & Cohen, 2013). Yet, others report impairments in comparison to NH peers (Macaulay & Ford, 2006; Peterson, 2004). The studies by Peterson and by Ziv and colleagues were the only ones comparing HI children with CI to HI children with conventional hearing aids, in addition to NH children. Both studies found no differences in ToM skills between the HI group with CI and the HI group without CI. Yet, in the Peterson study both HI groups were outperformed by the NH group, whereas this was not the case in the study by Ziv and colleagues. Although these studies provide a first insight into the ToM skills of children with CI, a consensus as to these children's ToM development has clearly not yet been reached. It should be noted that the samples in some of the studies were quite small, and some children were implanted relatively late compared to current standards. Moreover, in some of the studies children were up to 12 years old, which makes it hard to identify any delays in skills that would typically develop between the ages of 2 and 5.



As described above, people rely on ToM skills during social interactions to infer how to respond to other people. Yet, on a broader scale, it is people's moral sense which guides their behavior. This moral sense denotes a basic understanding of what is right and wrong. It is the corresponding experience of so-called *moral emotions* such as shame, guilt and pride which alert us when we have violated a norm or when we have done something exceptionally well. Moral emotions urge people to behave appropriately, i.e., according to the norms and values of the group or society at large, and thereby aid children to become socially competent (Barrett, 1995; Stearns & Parrott, 2012). The importance of moral emotions is stressed by the finding that psychopaths and criminal offenders often report a lack of these emotions (Holmqvist, 2008; Mealey, 1995). It seems that feeling guilty when harming another person or feeling ashamed when violating a norm prevents one from doing this again. Quite like empathy, moral emotions make us want to do the right thing and avoid doing harm to others. In other words, they help to regulate our behavior in ways that are consistent with the demands of our surroundings.

Moral emotions are acquired in interaction with the social environment. In order to experience a moral emotion, children need to be aware of the prevailing standards and to be able to judge their behavior according to these standards (Lewis, Alessandri, & Sullivan, 1992). This puts a lot of strain on young children's cognitive capacities. It was therefore once believed that children would not be able to experience moral emotions before the age of about 7 years old because they would not have reached the required level of cognitive maturity (Piaget, 1932/1965). Yet, ensuing studies have shown that infants and preschoolers already show behaviors that are interpreted as signs of moral emotions (Barrett, 2005; Draghi-Lorenz, Reddy, & Costall, 2001; Lewis et al., 1992). This seemingly incompatible finding can be explained in terms of social scaffolding, a notion which stems from Vygotsky's (1978) sociocultural theory. Social scaffolding refers to the role of parents as guides or mentors who instruct, model, and assist their children as needed during their learning process. Young typically developing children learn from their parents how they should behave, and when they have violated a norm and consequently should feel ashamed or guilty. Or, in the case of pride, when they have exceeded expectations (Mills, 2005). The amount of scaffolding needed will gradually decrease until children have fully internalized a set of moral standards and are able to regulate their behavior accordingly.

Moral emotions in HI children (with or without CI) have not been examined to date. Taking into account the requirements for moral emotions, we might assume that HI children will lag behind their NH counterparts. First, to experience moral emotions without any external guidance, once again perspective-taking skills come into play. As discussed earlier, these are known to be impaired in HI children (Peterson & Siegal, 2000), and might still be impaired in children with CI (Macaulay & Ford, 2006; Peterson, 2004). For example, in order to feel guilty, you need to understand that the other person was harmed and that it is your behavior which caused this harm. Second, HI children, also after they have received their CI, do not have access to the social world on a comparable level to NH children. Persisting language difficulties (cf. Ganek et al., 2012) could hamper communication with role models, making it plausible that HI children (with or without CI) do not receive the same amount of social scaffolding as their NH peers. Since moral emotions are learned in interaction with the social environment (Barrett, 1995; Mills, 2005), opportunities to learn about these might be limited for children with CI.

Underlying Factors: Parenting Practices and Communication

The learning processes involved in attaining emotional and social competence are subject to input from knowledgeable others. In early childhood this input is mainly provided by parents. As discussed before, parents can actively try to teach their children new things through the process of social scaffolding. Yet, a lot of teaching takes place implicitly. Infants for example observe their parents' behavioral response in an ambiguous situation in order to know how to react themselves, a process which is known as social referencing (Feinman, 1982). Inconsistent responses or a lack of response from parents may leave children confused as to how they should interpret a situation or to what is expected from them. A harsh and punitive parenting style, with little room for children to explore boundaries, and little or no social scaffolding, hampers children's social-emotional development. Sensitive and responsive parenting, on the other hand, may help children acquire social-emotional skills (Kawabata, Alink, Tseng, Van Ijzendoorn, & Crick, 2011; Stack, Serbin, Enns, Ruttle, & Barrieau, 2010).



Raising a HI child undoubtedly is challenging for parents. While the neonatal hearing screening enables early intervention, the diagnosis of deafness shortly after birth usually comes as a shock for parents. Since 90% of HI children are born into hearing families (Mitchell & Karchmer, 2004), parents have to learn to adapt to the special needs of their HI child. This may mean that parents have to familiarize themselves with sign language in order to effectively communicate with their children, a language which is grammatically and syntactically very different from spoken language and therefore very hard to master. If parents opt to have their HI child fitted with a CI, this implies enrollment in a rehabilitation program, frequent hospital visits, doing speech exercises at home, taking their child to a specialized play group for HI children which might not be located around the corner, and so on. In sum, parenting a HI child (with or without CI) is demanding and stressful (Hintermair, 2006; Quittner et al., 2010; Zaidman-Zait, 2008). High experienced levels of stress, in turn, could negatively impact the way these parents raise their children. Although to date no research is available on parenting styles of parents with children with CI, parents of HI children without CI are reported to lean more towards a punitive and less towards a sensitive parenting style than parents of NH children (Knutson, Johnson, & Sullivan, 2004; Meadow-Orlans, 1997).

Spoken language is the primary tool for parents to teach their NH children new things, and this becomes even more important as children grow older. Many skills, including those in the social-emotional domain, are learned or refined through discourse, either through direct instructions or explanations, or by overhearing conversations between other people (Vaccari & Marschark, 1997). Language and communication issues in HI children are often reported to underpin these children's social-emotional difficulties (Peterson & Siegal, 2000; Vaccari & Marschark, 1997). Parents of HI children experience difficulties when interacting with their children, and interact with their children less frequently compared to parents of NH children (Barker et al., 2009; Gale & Schick, 2009; Prezbindowski, Adamson, & Lederberg, 1998). Having conversations about abstract topics such as emotions is particularly difficult if children's primary mode of communication is sign language as most parents are not fluent signers (Peterson & Siegal, 2000; Vaccari & Marschark, 1997). Consequently, it is deemed more difficult for HI children than for NH children to gain emotion knowledge and to put this knowledge to use during social interactions.

Providing HI children with a CI restores sound perception to some degree and could therefore benefit these children's spoken language acquisition, and in turn their communication skills. Multiple studies have indeed shown the positive effects of cochlear implantation on language development (e.g., Boons et al., 2012a; Ganek et al., 2012; Niparko et al., 2010; Svirsky et al., 2000; Tomblin, Spencer, Flock, Tyler, & Gantz, 1999). Yet, many children (in particular those implanted after two years of age) do not reach age-equivalent spoken language skills even after years of CI use (Boons et al., 2012a; Ganek et al., 2012; Nicholas & Geers, 2007). This could imply that communication between parents and their children with CI is also still impaired, which could negatively impact children's ability to learn from their parents.

Heterogeneity of the Population

Thus far in this introduction, findings have been described as being applicable to the whole population of HI children (with and/or without CI). However, it should be noted that the population of HI children with CI is inherently heterogeneous. Although the population of typically developing children by no means is a homogeneous one, for example with regard to socio-economic status, upbringing, or intelligence, the population of children with CI varies on a number of additional aspects. To name just a few: the age at which children acquired their hearing loss, the degree of hearing loss pre-implantation, the extent to which children benefit from their implant, uni- or bilateral implantation, contralateral stimulation, age at implantation and duration of implant use, parental hearing status, preferred language mode, presence of additional disabilities, characteristics of the rehabilitation program children are enrolled in, and so on. Although the sample of children with CI involved in the studies which form the content of this thesis was not nearly large enough to control for each of these factors, efforts were made to keep a few of them constant and to examine the influence of others. All CI participants were born to hearing parents, had no apparent additional disabilities, had severe to profound hearing losses which were either congenital or acquired prelingually (i.e., before the age of 2 years), and had received their (first) implant at the mean age of 1.5 years and at least before the age of 3 years. Note that this sample was selected to reflect the Dutch population of young children with CI at the present time.

However, generalizations to the whole population of young children with CI, especially to those with concomitant disabilities, should be made with caution.

Rationale and Outline of this Thesis

Numerous reasons could be provided that underline the importance of studying social-emotional functioning in this particular population of children with CI. First and foremost, knowledge concerning this area of development in children with CI is lacking, while there is ample evidence that social-emotional skills of deaf children without CI do not always develop age-appropriately or along the same developmental trajectories as in the NH population (Barker et al., 2009; Gray, Hosie, Russell, Scott, & Hunter, 2007; Kouwenberg et al., 2012; Ludlow, Heaton, Rosset, Hills, & Deruelle, 2010; Peterson & Siegal, 2000; Theunissen et al., 2011; Van Gent et al., 2007; Woolfe et al., 2002). Ideally, we would like to compare a sample of children with CI to a control group of NH children as well as to a control group of HI children without CI, in order to pry out the effect of a CI on HI children's social-emotional development. However, this is rendered impossible by the mere fact that nowadays almost all parents of children with severe to profound hearing losses opt for their child to receive a CI (De Raeve & Lichert, 2011; Fortnum, Marshall, & Summerfield, 2002; Johnston, 2004; Preisler, Tvingstedt, & Ahlstrom, 2002). The few children that do not receive a CI are often not comparable to the majority that does. For example, these children have additional disabilities or they have HI parents who more often choose not to have their child implanted. HI children from HI parents share a common language and for example have well-developed ToM skills, as opposed to HI children from NH parents (Peterson & Siegal, 1999).

A second reason worth mentioning here is the potential for timely interventions that the outcomes of the studies reported in this thesis may offer. Assuming that children with CI will show impairments or delays in at least some areas of social-emotional development, outcomes of these studies will enable professionals to target interventions at specific areas of social-emotional development that require attention. Not only will this be more efficient than interventions directed at the whole spectrum of social-emotional development, but detecting risk factors and impairments early in life offers the possibility to remediate these before they progress or negatively affect other



areas of development, which is both cost effective for society and beneficial for children's well-being.

The main question that underlies all studies included in this thesis is whether young (i.e., 1-to-5-year-old) HI children with a CI are comparable to NH peers regarding their social-emotional functioning. Part of this question is answered by examining group differences with respect to levels of social-emotional functioning. Additionally, in several studies the relations between various aspects of social-emotional functioning are examined in both groups of children separately. And finally, possible factors underlying the social-emotional development of children are studied, such as language skills, parenting practices, or CI-related variables.

In *Chapter 2*, the development of a parent questionnaire aimed at measuring empathy in young (1-to-5-year-old) children is described. This questionnaire is used in the study described in *Chapter 3*, which examines the ability of children with CI and NH to empathize with other people, and how this ability is related to social competence. *Chapter 4* focuses on the theory-of-mind abilities of children with CI compared to NH children. *Chapter 5* concerns the extent to which moral emotions are expressed by children with CI and their NH peers, and the associations within each group with various aspects of social functioning. *Chapter 6* examines the role of parents with regard to their children's social-emotional development. In the concluding *Chapter 7*, findings from the previous chapters are integrated and their implications are discussed.



2

Assessing empathy in young children: Construction and validation of an Empathy Questionnaire (EmQue)

The contents of this chapter are published in:
Rieffe, C., Ketelaar, L., & Wiefferink, C.H. (2010).
Personality and Individual Differences.

Abstract

Empathy is a basic human capacity that supposedly serves to regulate relationships, supporting collaboration and group cohesion. The capacity to automatically imitate and level with affective expressions in others is supposed to be innate, but already during infancy children need to learn how to regulate their own affective arousal and direct their attention towards the affective state of the first person. In this study, the EmQue, a newly developed parent questionnaire regarding empathy-related behaviors in young children was examined. Responses of 109 parents confirmed the hypothetically assumed levels of empathy in infants and toddlers (mean age = 30 months): (a) Contagion, (b) Attention to Others' Feelings and (c) Prosocial Actions. Associations with other parent questionnaires (e.g., emotion recognition, understanding and regulation) and child behaviors (Empathy, Frustration, Theory of Mind) further confirmed the EmQue's validity. The EmQue could be of clinical relevance during early childhood, particularly for specific clinical groups that are expected to be at risk in this respect, such as children with an Autism Spectrum Disorder or deaf children.

Acknowledgement. The authors want to thank Anna Brett for correcting our English.

Introduction

Empathy is a basic human capacity that is important in daily social life. It refers to the ability to respond affectively to emotions in others, aiming at reacting adaptively to another's needs, e.g. to console, support or spare the other person (Decety & Jackson, 2004; Hoffman, 1987). In fact, it is supposed to be the important motor behind many prosocial behaviors or behavior that strengthens group cohesion and cooperation (Jolliffe & Farrington, 2006; Zahn-Waxler et al., 1992). Deficits in empathy can play a critical role in the development of externalizing behavior and other behavioral problems (Jolliffe & Farrington, 2006; LeSure-Lester, 2000; Miller & Eisenberg, 1988). Empathic behaviors can be observed from infancy onwards (Hoffman, 1987). However, besides the obviously important observational studies, no questionnaires are yet developed to systematically operationalize empathy in young children. In this study we constructed and validated a parent questionnaire to examine empathy in infancy and toddler hood.

Levels of Empathy

Hoffman (1987) distinguished four levels of empathy. Although these levels are assumed to develop sequentially, they are not mutually exclusive (Hoffman, 1990). De Waal (2008) supposed that the different levels of empathy represent a Russian doll model, suggesting that each following level builds onto the former levels.

The first level that Hoffman identified is 'Global Empathy', which others labeled 'Emotion Contagion' (Hatfield, Cacioppo, & Rapson, 1993), and which manifests itself in the first year of life. At this level, infants attend to others' emotions, albeit inadaptively because witnessing someone in distress may result in a similar affective response. For example, the crying of one infant may trigger equal responses in other babies. It is assumed that people are 'hardwired' to automatically imitate and synchronize affective expressions (Decety & Jackson, 2004), but infants this young cannot yet differentiate between self and other, which causes them to act as though what happened to the other person happened to them (Vreeke & van der Mark, 2003). Alternatively, infants this young might still have difficulties to control their level of arousal. If true, the ability for self-regulation should be associated negatively with symptoms of emotion contagion.



The second level, which we will label 'Attention to Others' Feelings' is assumed to start at about one year of age. At this level, Hoffman argued, infants become aware that although they feel distressed, it is not oneself but someone else who is in actual danger or pain. In other words, infants become more aware of other people's emotions, which can be observed because they direct their attention to affective displays of others. Moreover, infants develop the capacity to attend to others' emotions with less personal distress. The own response to the distress of another child may now be transformed into concern for the victim.

At the third level, which we will label 'Prosocial Actions', Hoffman argued that children become more responsive to others' emotional displays, and start to react prosocially. A longitudinal study by Zahn-Waxler and colleagues (1992) showed that children develop this capacity to intervene on behalf of others during the second year of life, which can take a variety of forms, including helping, sharing, and comforting.

The fourth level in Hoffman's theory, 'Empathy for Another's Life Condition' develops during late childhood and refers to empathic responses not only confined to the situation but also with another's general level of distress or deprivation. This empathic level may motivate to feel empathy for people who live in more unfavorable circumstances, and support them by, for example, donating money to charity funds.

Current Study

Although these different levels of empathy that Hoffman initially described are widely acknowledged theoretically, to date it is unclear if they indeed exist and can be distinguished empirically. Additionally, it is unclear if these levels are uniquely related to other constructs. The current study examines the psychometric properties of a newly developed questionnaire for parents aimed at observing the first three levels of empathy in infants' and young children's behaviors: the Empathy Questionnaire (EmQue). Because the fourth level develops in late childhood, this level is not incorporated in the questionnaire. The following issues were examined: (1) the assumed three-factor structure of the EmQue, (2) the internal consistency of the three subsequent EmQue scales (Emotion Contagion, Attention to Others Feelings and Prosocial Actions), and (3) the criterion and concurrent validity of these three EmQue scales through their associations with related measures.

The criterion validity of the EmQue scale Attention to Others' Feelings was assessed through examination of its associations with the scale Attention of the Empathy Task. Similarly, the associations of the scale Prosocial Behavior with the scale Consolation of the Empathy Task and with Prosocial Behavior of the SDQ parent questionnaire were examined. The concurrent validity of the three EmQue scales was assessed through examining their associations with another parent questionnaire that taps into social and emotion regulation skills of children between one and five years old; and with additional observations obtained through offering children tasks, including an emotion regulation task and a Theory of Mind task.

The EmQue scale Emotion Contagion was expected to correlate positively with measures that indicate poor emotion regulation. The EmQue scale Attention to Others Feelings was expected to correlate positively with other measures indicating attention to other people's emotions or emotion recognition. The EmQue scale Prosocial Actions was expected to correlate positively with other measures indicating prosocial behavior, but also emotion understanding and attention to others' emotions. Additionally, a positive correlation was expected for this scale with a measure for understanding the subjective nature of other people's mental states, a so-called Theory of Mind task.

In order to examine the frequently assumed sequential nature of the first three levels that were identified in empathy, correlations of the EmQue scales with age were calculated. If contagion indeed precedes children's ability to attend to others' affective states, which in turn precedes children's ability for empathically-driven prosocial behavior, correlations with age should increase over these three levels. In other words, regarding the three levels of empathy, contagion should correlate least strongly with age; whereas prosocial behavior should correlate most strongly.

Method

Participants and Procedure

In this study, parents of 109 one- to five-year-old children (mean age = 30 months, $SD = 13$ months; 57 girls, 52 boys) filled out questionnaires (EmQue, Strengths and Difficulties Questionnaire (SDQ), Emotion Expression Questionnaire (EEQ)).



Parents (87 mothers, 11 fathers, 11 unknown) completed the questionnaires at home through the internet or a paper-and-pencil version.

Children and their parents were recruited via day-care centers, schools and acquaintances of the researchers. The Empathy Task and the Frustration Task were conducted with 31 children from the younger half of the sample (age 12–30 months old; 15 boys, 16 girls), whereas the Theory of Mind task was conducted with 42 children from the older half of the sample (age 31–60 months old; 26 boys, 16 girls).

Compared with national data from Statistics Netherlands (CBS) our sample corresponded in many ways with the Dutch population of parents from one- to five-year-old children (mean age mothers = 34 years; mean age fathers = 37 years). However, the current sample had relatively more parents of Dutch origin (94%), and with higher educational level (80% of the mothers and 73% of the fathers had a BA degree or higher). Most children (94%) lived with 2 parents (of which most children lived with a father and a mother, and 2 children lived with 2 mothers), 25% of the children had no siblings, and almost all children attended some kind of day-care or school for at least one day a week (95%).

Materials

The EmQue consists of 20 items (Table 1), representing three facets of empathy that should be observable in young children: (a) Emotion Contagion, (b) Attention to Others' Feelings, and (c) Prosocial Actions. Parents can rate the degree to which each item, reflecting a type of behavior, applied to their child over the past two months on a 3-point scale (0 = never, 1 = sometimes, 2 = often).

An initial version of the EmQue consisted of 60 items (formulated by a team of developmental psychologists, school teachers and a child psychiatrist who is working with children with an Autism Spectrum Disorder; and based on and inspired by the many observational studies that are available, e.g., by Eisenberg and colleagues), and was tested in a pilot study. A total of 83 parents (mean age of children = 35 months, $SD = 13$ months; 37 boys, 46 girls) completed the questionnaire for this pilot. However, 33 items were removed due to an insufficient fit with the selection criteria (i.e., 30 items were deleted because the particular item had a missing value rate higher than 75%; 3 items were deleted because they differentiated between boys and girls) and 7 items were deleted due to an insufficient fit on their intended factor. Additionally, 4 items were

reformulated in order to represent the three levels of empathy. This resulted in the current 20-item version.

The SDQ is a brief behavioral screening questionnaire (Goodman, 1997), consisting of 25 items. The scale Prosocial Actions (5 items) is used for this study. Parents can rate each item on a 3-point scale (0 = not true, 1 = somewhat true, 2 = certainly true). The internal consistency of the scale Prosocial Actions is usually good (Van Widenfelt, Goedhart, Treffers, & Goodman, 2003). This was confirmed in the present study (Table 2).

The EEQ is a 35-item parent-report questionnaire, designed for this study, and measures emotion expression of the child. Three scales are used for the validation of the EmQue: (a) Emotion Regulation (8 items), which indicates the extent to which children can calm themselves or be calmed by their parents when angry or sad; (b) Others' Emotion Recognition (6 items) which indicates the extent to which the child can recognize the parents' and others' emotions; and (c) Emotion Understanding (6 items), which indicates the extent to which children can evaluate an emotional episode. Parents can rate to what degree each item is true on a 5-point response scale (1 = (almost) never, 2 = rarely, 3 = sometimes, 4 = often, 5 = (almost) always). The internal consistencies of the scales are good (Table 2).

Tasks with Children

The Empathy Task examines children's responses to emotional displays by the experimenter, based on a task displaying pain designed by Zahn-Waxler et al. (1992). The task consists of three separate acting-out performances by the experimenter (happiness with a pen, anger with a pen that fails to write, and pain/sadness while burning one's fingers on a cup of tea). Following each acting-out performance, the experimenter scores the child's behavioral reactions on a checklist that was designed for this study. The checklist consists of two scales: (a) Attention to Others' Emotions (6 items) and (b) Consolation Behavior (5 items, not including the happy event). The internal consistencies of the scales are good (Table 2).



Chapter 2

Table 1 Items of the Empathy Questionnaire for Infants and Toddlers, EmQue

<i>Emotion Contagion</i>	
1	When another child cries, my child gets upset too
4	My child also needs to be comforted when another child is in pain
7	When another child makes a bad fall, shortly after my child pretends to fall too
10	When another child is upset, my child needs to be comforted too
13	When another child gets frightened, my child freezes or starts to cry
16	When other children argue, my child gets upset
19	When another child cries, my child looks away
 <i>Attention to Others' Feelings</i>	
3	When my child sees other children laughing, he/she starts laughing too
6	When an adult gets angry with another child, my child watches attentively
9	My child looks up when another child laughs
12	When adults laugh, my child tries to get near them
15	My child looks up when another child cries
18	When another child is angry, my child stops his own play to watch
20	When other children quarrel, my child wants to see what is going on
 <i>Prosocial Actions</i>	
2	When I make clear that I want some peace and quiet, my child tries not to bother me
5	When another child starts to cry, my child tries to comfort him/her
8	When another child gets upset, my child tries to cheer him/her up
11	When I make clear that I want to do something by myself (e.g. read), my child leaves me alone for a while
14	When two children are quarrelling, my child tries to stop them
17	When another child gets frightened, my child tries to help him/her

The Frustration Task was designed for this study and examines children's responses to a frustrating event. The experimenter opens a bottle in front of the child, closes it again and then asks the child to open the bottle. The child is unaware of the fact that the bottle contains a safety lock that makes it impossible for children to open. The experimenter waits a minimum of 30 s, a maximum of 60 s and then opens the bottle a little bit so the child can now finish the task successfully. During this waiting period, the experimenter scores the child's behavioral reactions on a checklist, consisting of three scales. The scale Diversion (5 items) that is used for this study denotes the extent to which children can divert their attention from the negative stimulus. An example item is 'the child starts doing something else'. One item is formulated contradictory ('the child keeps trying') and recoded. The internal consistency of the scale is good (Table 2).

The Theory of Mind task consisted of the Uncommon Desire Task (Rieffe, Terwogt, Koops, Stegge, & Oomen, 2001), in which children are presented twice with a drawing of a more and a less desirable food item (broccoli and cake in one vignette; tomato and ice-cream in another vignette) and asked which item they prefer. Following this response, a drawing is shown of a child behind a table, with the same 2 items lying in front of him. Children are told that the protagonist's preference is opposite to theirs; and asked what item the protagonist will take from the table. Two control questions ("Does the boy like item 1?" and "Does the boy like item 2?") are also asked per vignette. The total of 6 questions form a scale (minimum score = 0, maximum score = 6) indicating children's ability to understand the subjective nature of mental states, showing good psychometric properties (Table 2).

Table 2 Internal Consistencies of the EmQue, SDQ, EEQ, and Tasks for Children (Frustration Task, Empathy Task, Theory of Mind Task)

	Number of items	Number of participants	Mean (SD)	Cronbach's Alpha	Inter-item correlation
<i>Parent questionnaires</i>					
EmQue (0–2)					
Emotion Contagion	6	109	.42 (.31)	.58	.19
Attention to Others' Feelings	7	109	1.34 (.36)	.71	.26
Prosocial Actions	6	109	.67 (.44)	.80	.41
SDQ, Prosocial Behavior (0–2)	5	109	1.17 (.55)	.82	.48
EEQ, Emotion Regulation (1–5)	8	107	2.73 (.49)	.72	.26
EEQ, Others' Emotion Recognition (1–5)	6	107	3.73 (.64)	.77	.38
EEQ, Emotion Understanding (1–5)	5	107	3.49 (1.22)	.96	.82
<i>Tasks for children (0–1)</i>					
Empathy Task (12–30 months)					
Attention	6	31	.55 (.32)	.82	.43
Consolation	5	31	.10 (.19)	.68	.37
Frustration Task, Diversion (12–30 months)	5	31	.20 (.27)	.71	.39
Desire Task (31–60 months)	6	40	.76 (.29)	.77	.36

Analyses

The internal structure and homogeneity of the EmQue scales was established with Principal Component Analysis (PCA) and reliability analyses. Oblimin rotation with Kaiser normalization was used, as the restriction of scale independence was considered too stringent. The factor count was limited to the assumed 3 factors. The criterion validity was examined by means of correlations of the EmQue scales with the scales of the Empathy Task (only children aged 12–30 months old) and the parent scale Prosocial Behavior of the SDQ. For the SDQ scale, also a regression analysis was performed. The concurrent validity was similarly investigated by means of correlations (and additional regression analyses with the parent questionnaire) of the EmQue scales on the Frustration Task (only children aged 12–30 months old), the Theory of Mind task (only for children aged 31–60 months old), and the scales of the EEQ (Emotion Regulation, Recognition of Others' Emotions, Emotion Understanding).

Table 3 EmQue Items (20 items) and PCA Factor Loadings

	Emotion Contagion	Attention to Others' Feelings	Prosocial Actions
Ep1	.37		
Ep4	.74		
Ep7	.45		
Ep10	.79		
Ep13	.52		
Ep16	.35		
Ep19	–		
Ep3		.45	
Ep6		.62	
Ep9		.67	
Ep12		.41	
Ep15		.70	
Ep18		.56	
Ep20		.42	
Ep2			.73
Ep5			.63
Ep8			.63
Ep11			.76
Ep14			.60
Ep17			.72

Note. Only factor loadings >.35 are displayed.

Results

Factor Structure and Internal Consistencies

A PCA on all 20 items in the EmQue, with the factor count limited to the assumed 3 factors (Table 3), shows that all items loaded $> .35$ on their keyed factor (explaining 40% of the variance), with the exception of item 19 (scale Emotion Contagion). This item was removed from the scale in further analyses. The interfactor correlations between the 3 scales were significant for Emotion Contagion \times Attention to Others' Feelings ($r = .29; p < .01$); Emotion Contagion \times Prosocial Actions ($r = .24; p < .01$); and Prosocial Actions \times Attention to Others' Feelings ($r = .40; p < .01$), as should be the case for a multifaceted construct.

Coefficients for the internal consistency regarding the EmQue scales are presented in Table 2. The Cronbach's Alphas of two EmQue scales (Attention to Others' Feelings and Prosocial Actions) meet the expected minimum of .70, respectively. The scale Emotion Contagion shows lower, but still acceptable, psychometric properties.

Criterion and Concurrent Validity

The Pearson correlation coefficients and the regression coefficients (Table 4) show that some scales have substantial and significant relations to one or more scales of the EmQue. The correlations with the EmQue scale Emotion Contagion are positive for Prosocial Behavior (SDQ), Others' Emotion Recognition (EEQ) and Emotion Understanding (EEQ). However, the correlation with the scale Consolation (Empathy Task) is negative. Regarding the two measures for emotion regulation, Contagion correlates negatively with Emotion Regulation (EEQ) and Diversion (Frustration Task). The regression analyses of the EmQue scales on the prediction of the parent questionnaires, confirms this negative association, and shows a unique contribution of Emotion Contagion to the prediction of Emotion Regulation.

The EmQue scale Attention to Others' Feelings is positively correlated with Prosocial Behavior (SDQ), Others' Emotion Recognition (EEQ), Emotion Understanding (EEQ), Attention (Empathy Task), and the Uncommon Desire Task (but not when corrected for Age). The regression analyses of the EmQue scales on the prediction of the parent questionnaires show that this scale only contributes uniquely to the prediction of Others' Emotion Recognition(EEQ).



The correlations with EmQue scale Prosocial Actions are positive for Prosocial Behavior (SDQ), Others’ Emotion Recognition (EEQ), Emotion Understanding (EEQ), Attention (Empathy Task), Consolation (Empathy Task) and the Uncommon Desire Task. When corrected for Age, the correlation with Attention is no longer significant. The regression analyses show that the scale Prosocial Actions only has a unique contribution to the prediction of Emotion Understanding.

Table 4 Correlation Coefficients (Partial Correlations Corrected for Age) of the EmQue Scales with Parent Questionnaires and with Tasks for Children; and Regression Coefficients of the EmQue Scales with Parent Questionnaires and Age

	Empathy Questionnaire		
	Emotion Contagion	Attention to Others’ Feelings	Prosocial Actions
	<i>Correlations</i>		
SDQ, Prosocial Behavior	.23**	.40**	.67**
Emotion Regulation	-.16*	.02	-.01
Others’ Emotion Recognition	.19*	.48**	.43**
Emotion Understanding	.18*	.29**	.62**
Frustration Task, Diversion	-.37* (-.38*)	-.19 (-.11)	-.25 (-.12)
Empathy Task, Attention	-.01 (-.03)	.35* (.33*)	.32** (.25)
Empathy Task, Consolation	-.39* (-.42*)	.25 (.12)	.41* (.31*)
Desire Task	.20 (.24)	.32* (.27)	.34* (.30*)
	<i>Standardized regression coefficients</i>		
	Age		
SDQ, Prosocial Behavior	.01	.05	.15
Emotion Regulation	-.23	-.20*	.07
Others’ Emotion Recognition	.27**	.04	.37**
Emotion Understanding	.74**	.13	.02

Note. Interactions with Age in the regression analyses were not significant and are not shown in this table
* $p < .05$; ** $p < .01$

Relation with Age

Pearson correlations were calculated in order to verify whether the three EmQue scales were related to Age. The age of the participating children varied from 12 to 60 months. Significant correlations were found for Age x Prosocial Actions ($r = .54$; $p < .01$) and Age x Attention to Others’ Feelings ($r = .17$; $p < .05$), but not for Age x Emotion Contagion ($r = .04$; $p > .05$). Additionally, Age had a unique contribution to the prediction of Others’ Emotion Recognition and Emotion Understanding.

Discussion

The outcomes of this study suggest that core features of empathy can be identified in early childhood through parent reports. The three levels of empathy that were identified based on the literature indeed appeared as separate, albeit related, facets of empathy when examining the questionnaire's factor structure, but also when examining the criterion and concurrent validity of the separate levels with other related measures. The expected correlations of the separate scales with other related measures remained when correcting for the age of the participants. Consistent with the literature, Prosocial Actions and Attention to Others' Feelings were positively related to the age of the participants (Zahn-Waxler et al., 1992), but Emotion Contagion was not.

First, the three-factor model appeared a useful representation of the Empathy Questionnaire. One item (item 19) that failed to load on the intended factor, was removed from the scale Contagion. The remaining 19 items represented three one-dimensional factors. Second, the scales Attention to Others' Feelings and Prosocial Actions showed good psychometric properties with good internal consistencies, whereas the internal consistency for Emotion Contagion was lower, but still acceptable. Third, the criterion and concurrent validity of the three scales were good and largely in accordance with the expectations. Some measures correlated to all three empathy levels, whereas other measures differentiated between the levels of empathy. However, additional regression analyses showed that only the intended scales of the EmQue contributed uniquely to the prediction of the criterion variables. These outcomes will be discussed in more detail.

Two scales of the EmQue, Attention to Others' Feelings and Prosocial Actions, were identified in the explorative factor analysis as two distinguishable constructs. Moreover, the two scales also showed associations with the corresponding scale in the empathy observation task but not with the other scale (when corrected for age). In other words, not only could parents distinguish between these two levels of empathy, but children's behavioral responses showed the same configuration as observed by an experimenter.

Yet, the parent questionnaires that correlated with Attention to Others' Feelings and Prosocial Actions did so significantly to both scales, thus did not differentiate between these two levels of empathy. This might support the Russian doll idea by De Waal (2008), implying that Attention to Others' Feelings



is a precondition for children's prosocial behavior. Additional regression analyses however did show unique contributions of these two EmQue scales to the intended scales of the parent questionnaires: Prosocial Actions contributed uniquely to the prediction of prosocial behavior and emotion understanding, whereas only Attention to Others' Feelings contributed to the prediction of recognizing other people's emotional states.

Adequate social skills, emotion understanding and recognition of others' emotions were also abilities related to a stronger tendency for signs of contagion (becoming affectively aroused when another person is in distress). Yet, these associations were less strong than for the other two EmQue scales. Moreover, there was an unanticipated negative correlation with consolation behavior in response to the experimenter's emotional displays (Empathy Task). Despite their adequate social skills as reported by their parents, children who score higher on contagion might function less adequately regarding their social relationships.

Emotion Contagion was also shown to be distinctive from the other two scales with respect to its significant correlation with poor emotion regulation (negative correlations with Emotion Regulation and with Diversion in the Frustration Task). This supports the idea that the tendency to become affectively aroused due to distress in another person can be intensified or prolonged by an inability to regulate the own emotion experience (Decety & Meyer, 2008). Considering also that contagion in this study seems unrelated to age, it does suggest that it involves a trait which is not very beneficial to children's social functioning: these children seem to have difficulties dealing with their own emotional arousal when observing others' distress.

Overall, the outcomes of this study confirmed the three levels of empathy that are frequently noted in the literature to be apparent in very young children. Empathy is considered to be a keystone in children's social development. With the present instrument, future studies could be used to more closely examine the unique predictive value of the different levels of empathy to children's emotional and social functioning; and examine how these levels contribute to different aspects of children's development.

Distinguishing between these levels empirically might also bear important clinical relevance. It could be relevant to know how these levels develop differently in clinical groups that are known for their impaired empathic reactions, such as children with an Autism Spectrum Disorder (ASD)

or deaf children (Dyck, Farrugia, Shochet, & Holmes-Brown, 2004). Possibly, children with ASD attend to others' feelings to an extent that is compatible to typically developing children (Begeer, Rieffe, Terwogt, & Stockmann, 2006). Yet, their ability to act prosocially could be impaired. Alternatively, a discussion is currently going on suggesting a dysfunctional mirror neuron system in people with ASD, although the outcomes are not always supporting this assumption (Raymaekers, Wiersema, & Roeyers, 2009). If true however, children with ASD would also show impairments on the first level of empathy, contagion.

Answers to these questions might give new insights into how to decrease the chances of developing different forms of psychopathology. Note however that this study is only a first attempt in applying this questionnaire. A limitation is that only observable behaviors were taken into account in this study, whereas empathy also concerns one's intentions, which are not necessarily acted out. Research on older children could also employ self-reports and include aspects of empathy that are not always visible, such as the intention for consoling behavior, or the fourth level 'Empathy for Another's Life Condition', as suggested by Hoffman.





3

Social competence and empathy in young children with cochlear implants and with normal hearing

The contents of this chapter are published in:
Ketelaar, L., Rieffe, C., Wiefferink, C.H., & Frijns, J.H.M. (2013).
The Laryngoscope.

Abstract

This study set out to examine the levels of social competence and empathic behaviors in children with cochlear implants in comparison with normal-hearing children, and to determine whether empathy predicts social competence to the same extent in both groups of children. A total of 150 children (mean age 39 months) participated in the study; 61 with cochlear implants and 89 without hearing loss. Parent reports and observation measures were employed to measure empathy and social competence. Results showed that levels of empathy and social competence in children with cochlear implants and normal-hearing children were similar. Empathic behaviors were predictive of social competence in both groups alike. Emotion acknowledgment was more predictive of social competence for children with cochlear implants than for normal-hearing children. Language skills were unrelated to social competence or empathic behaviors in children with cochlear implants. In conclusion, children with cochlear implants showed no delay concerning social competence or empathic behaviors. The factors contributing to social competence, however, differed between the groups. This should be kept in mind when developing rehabilitation programs for children with cochlear implants.

Acknowledgments. The authors thank the parents and children who participated in this study. Additionally, we thank Ineke Smit for correcting our English.

Introduction

Social competence, i.e., the ability to engage in social interactions skillfully, is an important predictor of social outcomes such as peer relationships and popularity, but also of academic outcomes such as school readiness and achievement (Denham et al., 2003). Yet, attaining social competence is a challenge for many hearing-impaired (HI) children (Ita & Friedman, 1999). Disruptions in communication prevent these children from optimally benefiting from social learning (Moeller, 2007). Cochlear implantation improves children's language and communicative abilities (Ganek et al., 2012), and is therefore expected to also boost their social skills. Some studies support this claim (Huttunen et al., 2009; Huttunen & Valimaa, 2010; Martin, Bat-Chava, Lalwani, & Waltzman, 2011; Nicholas & Geers, 2003) but the body of literature is small and researchers have paid little or no attention to factors that may contribute to social competence in children with cochlear implants (CI). One important factor that influences social competence of normal-hearing (NH) children is empathy (Eisenberg, Spinrad, & Sadovsky, 2006; Saarni, 1999), which strengthens cooperation and group cohesion (Zahn-Waxler et al., 1992). Children who show higher levels of empathy are better liked by their peers and teachers, and are judged to be more socially competent in general (Eisenberg et al., 2006). Although infants have an inborn propensity for empathy, this ability further develops through social interactions (Decety & Jackson, 2004). Newborns are not yet able to differentiate between themselves and others and they therefore tend to be overwhelmed when witnessing other people in distress. However, it is this affective 'catching' or mirroring of others' emotions that enables the development of concern for others during the second year of life, once the ability to distinguish between self and other has developed, and that will ultimately induce prosocial behaviors such as helping or comforting (Hoffman, 1987; Rieffe, Ketelaar, & Wiefferink, 2010; Zahn-Waxler et al., 1992).

Little is known about the development of empathy in HI individuals. In fact, in only one study seven- to ten-year-old HI children were found to display less prosocial behaviors than their NH classmates (Wauters & Knoors, 2008). To our knowledge no studies have been conducted with children with CI. However, from research into aspects related to empathy we may infer that the development of empathy in HI individuals is impaired. Studies have shown that HI children are less inclined to conceal their own emotions in order to



protect other people's feelings (Hosie et al., 2000), and less often expect their peers to behave empathically (Rieffe & Terwogt, 2006). Furthermore, although HI children are able to attribute basic emotions (i.e., happiness, anger, sadness, fear) correctly in a social context (Rieffe & Terwogt, 2000), they are more prone to errors in the identification and attribution of complex or ambiguous emotions (Dyck et al., 2004; Ludlow et al., 2010; Most & Aviner, 2009). All of these studies were conducted with older children (ages 6 to 17). Studies with younger children with CI (ages 2 to 5) also report impaired emotion recognition skills (Wang et al., 2011; Wiefferink et al., 2013). The current study set out to measure emotional and social skills in very young children, which is challenging because they are not yet able to reflect on their feelings or intentions. Therefore, different instruments were employed to gain an insight into their empathic behaviors and level of social competence. Experimenters as well as parents reported on empathic behaviors displayed by the children. Additionally, parents were asked to report the extent to which the children were able to acknowledge, i.e., correctly interpret, emotions displayed by their parents, and the extent to which children were socially competent. This study aimed to 1) compare young, early-implanted children with CI with a matched group of NH children on social competence and empathic behaviors, 2) examine whether the different instruments for measuring empathic behaviors contribute to the prediction of social competence to the same extent in both groups of children, and 3) assess whether language aspects were related to empathic behaviors or social competence in children with CI.

Method

Participants

A total of 150 children from the Netherlands and the Dutch speaking part of Belgium participated in this study. All children were born to hearing parents and had no apparent (additional) disabilities. Age, gender and socioeconomic status did not differ between the groups. Characteristics of the samples are reported in Table 1. All children with CI were prelingually, profoundly deaf and had been implanted before the age of three years. Sixty-eight % had been using their CI for 12 months or more. Parents indicated that practically all children were wearing their CI full-time, only two children were wearing the device

often but not always. Approximately half the group was bilaterally implanted (simultaneously or sequentially), the other half was unilaterally implanted (either with or without a conventional hearing aid in the other ear). All children with CI entered a tailored rehabilitation program after implantation, which includes specialized playgroups, technical support for the device and speech therapy.

Table 1 Demographic and Medical Profile of Participants

	CI (n = 61)	NH (n = 89)
Age, mean (SD), mo	39 (15.1)	39 (16.7)
Age, range, mo	14 – 77	14 – 76
Male, No. (%)	37 (58%)	52 (61%)
Socioeconomic status		
Maternal education, mean (SD) ^a	3.45 (0.82)	3.60 (0.58)
Net household income, mean (SD) ^b	3.61 (1.18)	3.66 (0.84)
Age at implantation, mean (SD), mo	16 (7.3)	
Age at implantation, range, mo	6 – 35	
Time with (first) CI, mean (SD), mo	21 (13.1)	
Time with (first) CI, range, mo	1 – 44	
Preferred mode of communication, No. (%)		
Oral language only	23 (38%)	
Sign-supported Dutch	28 (46%)	
Sign Language of the Netherlands	3 (5%)	
Combination of communication modes	7 (11%)	
Reynell Developmental Language Scales		
Language Comprehension Quotient, mean (SD)	0.86 (0.18)	
Schlichting Expressive Language Test		
Sentence Quotient, mean (SD)	0.84 (0.14)	
Word Quotient, mean (SD)	0.89 (0.19)	

^a (1 = no / primary education, 2 = lower general secondary education, 3 = higher general secondary education, 4 = college / university).

^b (1 = less than €15,000, 2 = €15,000 – €30,000, 3 = €30,000 – €45,000, 4 = €45,000 – €60,000, 5 = More than €60,000).

Materials

The Empathy Questionnaire for infants and toddlers (19 items; Rieffe et al., 2010) measures empathic behavior displayed in daily situations (Table 2). Parents rated the degree to which each item represented their child's behavior over the past two months (0 = *never*, 1 = *sometimes*, 2 = *often*). The questionnaire shows good internal consistency (Table 3).

The Empathy Observation (Rieffe et al., 2010) examines children's empathic responses to three emotions that are acted out nonverbally by the



experimenter: happiness when clicking with a pen, anger with a pen that fails to write, and pain/sadness upon hurting one's finger. Children's responses were scored on a 20-item checklist (0 = *not at all*, 1 = *a little*, 2 = *a lot*) for the three events combined (Table 2). The internal consistency is good (Table 3).

Children's ability to acknowledge emotions in their parents was measured by the scale Others' Emotion Recognition (6 items) of the Emotion Expression Questionnaire (Rieffe et al., 2010) (Table 2). Parents rated on a five-point scale to what extent each item was true for their children (1 = *(almost) never*, 2 = *rarely*, 3 = *sometimes*, 4 = *often*, 5 = *(almost) always*). The internal consistency of the scale is good (Table 3).

Social Competence (9 items) was assessed with the Dutch version of the Strengths and Difficulties Questionnaire (Goodman, 1997; Muris, Meesters, & van den Berg, 2003). The scale Social Competence comprised the two original scales Prosocial (5 items) and Peer Problems (5 items). Three items of the Peer Problems scale were reversed so that higher scores reflected lack of peer problems. One item of the Prosocial scale ("Helpful if someone is hurt, upset or feeling ill") was removed because of conceptual overlap with items of the Empathy Questionnaire. Parents rated each item on a three-point scale (0 = *not true*, 1 = *somewhat true*, 2 = *certainly true*). The internal consistency of the compound scale is acceptable (Table 3).

Oral language comprehension and word and sentence production of children with CI were measured using the Dutch version of the Reynell Developmental Language Scales (RDLS) and the Schlichting Expressive Language Test (SELT) respectively (Van Eldik, 1998). An extensive study with 288 children with CI reported an average score of 0.78 ($SD = 0.18$) for children implanted before the age of two and an average of 0.66 ($SD = 0.20$) for children implanted before the age of five (Boons et al., 2012a).

Table 2 Items of the Empathy Measures

<i>Empathy Questionnaire</i>	
1	When another child cries, my child gets upset too
2	When I make clear that I want some peace and quiet, my child tries not to bother me
3	When my child sees other children laughing, he/she starts laughing too
4	My child also needs to be comforted when another child is in pain
5	When another child starts to cry, my child tries to comfort him/her
6	When an adult gets angry with another child, my child watches attentively
7	When another child takes a bad fall, shortly after my child pretends to fall too
8	When another child gets upset, my child tries to cheer him/her up
9	My child looks up when another child laughs
10	When another child is upset, my child needs to be comforted too
11	When I make clear that I want to something by myself (e.g., read), my child leaves me alone for a while
12	When adults laugh, my child tries to get near them
13	When another child gets frightened, my child freezes or starts to cry
14	When two children are quarrelling, my child tries to stop them
15	My child looks up when another child cries
16	When other children argue, my child gets upset
17	When another child gets frightened, my child tries to help him/her
18	When another child is angry, my child stops his own play to watch
19	When other children quarrel, my child wants to see what is going on
<i>Emotion Acknowledgment</i>	
1	Can your child adequately assess other people's emotions?
2	Does your child know when you are angry?
3	Does your child know when you are happy?
4	Does your child know when you are afraid?
5	Does your child know when you are sad?
6	Does your child know when you are having fun?
<i>Empathy Observation</i> ^a	
1	Child responds to experimenter's emotion
2	Child stops playing and looks at adult
3	Child tries to follow what is happening
4	Child physically approaches experimenter
5	Child mimics experimenter's facial expression
6	Child re-enacts/imitates event
7	Child tries to comfort experimenter ^b
8	Child tries to help experimenter ^c

^aItems were scored during each of the emotion-evoking events (happiness, anger, pain/sadness), except for item 7 and 8

^bAssessed during pain/sadness event only

^cAssessed during anger event only



Table 3 Internal Consistencies, Means, and SDs for Measures of Emotional and Social Functioning

	No. of items	Min-Max	Cronbach's Alpha	Inter-item correlation	CI (n = 61)	NH (n = 89)
					M (SD)	M (SD)
<i>Parent questionnaires</i>						
Empathy Questionnaire	19	0-2	.78	.16	0.84 (0.29)	0.85 (0.25)
Emotion Acknowledgment	6	1-5	.74	.35	3.73 (0.60)	3.75 (0.57)
Social Competence	9	0-2	.60	.13	1.42 (0.34)	1.42 (0.32)
<i>Experimenter observation</i>						
Empathy Observation*	20	0-2	.85	.23	1.04 (0.29)	0.81 (0.37)

* $p \leq .001$.

Procedure

Children with CI were recruited through hospitals and family counseling services all over the Netherlands and Dutch-speaking part of Belgium. NH children were recruited through day-care centers and elementary schools in the Netherlands. All children were tested individually in a quiet room at home, school or hospital. Three empathy-evoking events were alternated with other tasks (not presented in this manuscript) during a videotaped test session. Parents filled in questionnaires. Additional information, such as household income and age at implantation, was obtained from parents and/or medical records. Informed consent was obtained for all children and the study was approved by the university's medical ethics committee.

The tasks were administered to children in the CI group by one of two hearing experimenters who were fluent in sign language. Although in both groups the empathy tasks were administered nonverbally and did not require a verbal response, children with CI were addressed in their preferred mode of communication during the test session.

Statistical Analyses

The first research question was addressed by carrying out independent sample *t*-tests in order to compare children with CI and NH children on their empathic behavior and social competence. In order to answer the second research question, relations between the different measures of empathic behavior and social competence were examined by means of Pearson's correlations. Additionally, a stepwise regression analysis was carried out to examine the effect of measures of empathic behavior on social competence, and to examine

whether these effects differed for children with CI or NH. For that reason, group status was included as a dummy variable (NH = 0; CI = 1) and the independent variables were centered. Repeating the regression analysis excluding children who had less than one year experience with their CI did not produce different outcomes. Furthermore, the analysis was also carried out including age as an independent variable, but age did not contribute to the regression model. For reasons of clarity, the outcomes of these analyses are not reported. The third research question was addressed through Pearson's correlations in order to examine the relation of language-comprehension and production skills with measures of empathic behavior and social competence in children with CI.

Results

Group Differences

Mean scores in Table 3 reveal that parents of children with CI scored their children just as high as parents of NH children on Social Competence ($t(148) = -0.08, p = .933, \eta^2 = .00$), Emotion Acknowledgment ($t(148) = 0.20, p = .843, \eta^2 = .00$), and the Empathy Questionnaire ($t(148) = 0.10, p = .925, \eta^2 = .00$). Surprisingly, children with CI were rated higher than their NH peers on empathic behaviors as observed by experimenters ($t(145.57) = -4.29, p \leq .001, \eta^2 = .11$). Post-hoc t -tests with Bonferroni correction on the individual items of the Empathy Observation demonstrated that the difference was largely attributable to children with CI receiving higher scores than NH children on items that involved looking at the event or the experimenter.

Relations between Empathy Measures and Social Competence

A marginally significant correlation between the Empathy Questionnaire and the Empathy Observation was found for the whole group ($r = .16, p = .047$), but not for each group separately (CI: $r = .22, p = .086$; NH: $r = .15, p = .159$), which indicates that these instruments measure different aspects of empathic behavior. Table 4 shows Pearson's correlations of Age, Empathy Questionnaire, Empathy Observation, and Emotion Acknowledgment with Social Competence for both groups separately. Age did not correlate with Social Competence in either group (CI: $r = .02, p = .874$; NH: $r = .20, p = .061$). The Empathy Questionnaire as well as the Empathy Observation correlated with Social Competence in both



groups. A marginally stronger correlation was found in the CI group for the Empathy Questionnaire with Social Competence than in the NH group ($Z = 1.93$, $p = .054$), no difference in strength of the correlation was found for the Empathy Observation. Emotion Acknowledgment was only significantly associated with Social Competence in the CI group, the correlation was significantly stronger in the CI than in the NH group ($Z = 2.19$, $p = .029$).

Table 4 Pearson's Correlations and Hierarchical Regression Analysis for Measures of Empathy on Social Competence (N = 150)

	CI (n = 61)	NH (n = 89)	R^2_{adj}	β	p
	r	r			
<i>Step 1</i>			.24**		
Group				-.09	.234
Empathy Questionnaire	.51**	.23*		.25	.001
Empathy Observation	.32*	.41**		.32	≤ .001
Emotion Acknowledgment	.48**	.15		.19	.014
<i>Step 2</i>			.26**		
Group				-.09	.268
Empathy Questionnaire				.16	.118
Empathy Observation				.36	≤ .001
Emotion Acknowledgment				.05	.596
Group x Empathy Questionnaire				.13	.223
Group x Empathy Observation				-.05	.619
Group x Emotion Acknowledgment				.21	.034

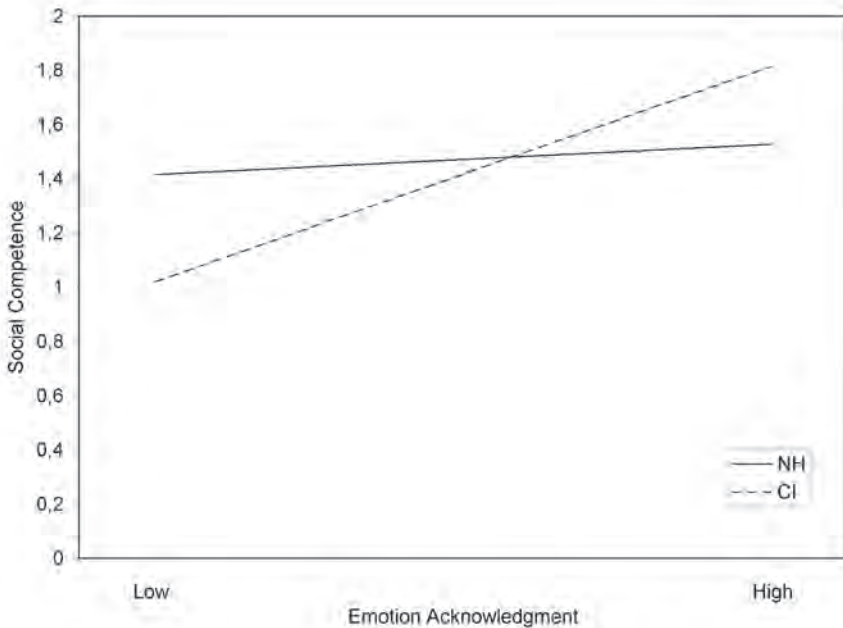
Note. $\Delta R^2 = .04$ for Step 2 ($p = .050$).

* $p < .05$. ** $p \leq .001$.

In order to examine whether the different measures of empathic behavior contributed to the prediction of social competence, and to assess whether these contributions differed between the groups, a hierarchical regression analysis with Group, Empathy Questionnaire, Empathy Observation, Emotion Acknowledgment (Step 1), and the interaction terms Group x Empathy Questionnaire, Group x Empathy Observation, and Group x Emotion Acknowledgment (Step 2) as predictor variables and Social Competence as dependent variable was carried out. Because of the low correlation between the Empathy Questionnaire and the Empathy Observation, both were included in the analysis in order to examine whether they uniquely contributed to the prediction of Social Competence. Results of the analysis in Table 4 show that all three measures of empathic behavior uniquely contributed to the

prediction of Social Competence. Adding the interactions with Group in Step 2 resulted in an increase in model fit. Group only interacted with Emotion Acknowledgment, which implies that Group status moderates the effect of Emotion Acknowledgment on Social Competence. As shown in Figure 1 correlations in Table 4 are confirmed, demonstrating a stronger association in children with CI than in children with NH between Emotion Acknowledgment and Social Competence when controlled for the other two measures of empathic behavior.

Figure 1. Group status moderates the effect of Emotion Acknowledgment on Social Competence.



Role of Language Skills in Children with CI

Mean language scores in Table 1 show that the CI group in this study exceeded the average language score of children implanted by the age of two as reported in the study by Boons and colleagues (2012a). No significant correlations between the language indices and any of the measures for empathic behavior and social competence were found (Table 5).



Table 5 *Pearson's Correlations of Language Skills with Empathy and Social Competence Measures for Children with CI*

	Empathy Questionnaire	Empathy Observation	Emotion Acknowledgment	Social Competence
RDLS	-.01	-.09	.24	-.11
SELT-W	.15	.17	.11	-.02
SELT-S	.02	.10	.13	.01

Note. RDLS = Reynell Developmental Language Scales, SELT-W = Schlichting Expressive Language Test – Word Development, SELT-S = Schlichting Expressive Language Test – Sentence Development

Discussion

This study aimed to assess the level of social competence in children with a CI as compared to NH children, and to examine whether empathic behavior contributed to the development of social competence to the same extent in both groups. Our results show equal levels of social competence in children with CI and NH children, which is in contrast with a study by Wiefferink, Rieffe, Ketelaar, and Frijns (2012a) that did note a difference on the same measure of social competence in favor of NH children. The sample of NH children included in the Wiefferink study was slightly older than in the current study, which could explain this different outcome as NH children become increasingly socially skilled with age (Rubin, Bukowski, & Parker, 1998).

Contrary to our expectations, the empathic behavior of children with CI was not impaired. In fact, experimenters judged children with CI to show more empathic behavior than their NH peers, although it is unclear whether the frequent eye contact by children with CI that caused this difference actually implies more attention to the emotional impact of the situation, or whether it simply reflects a greater reliance on visual cues conveyed by their communication partner. This finding should be examined further in follow-up studies. Yet, our study indicates that in both groups alike empathic behavior is an important factor regarding social competence. The ability to acknowledge emotions, however, was related to better social competence only in the CI group. This may denote a more other-focused orientation in these children, which is an important quality for gaining an understanding of the causes of others' emotions (Denham, 1998).

The finding that the empathic behaviors of children with CI were on a par with those of NH children seems to contradict findings from other

studies showing an impaired understanding of other people's mental states and subsequent emotions in children with CI (Ketelaar, Rieffe, Wiefferink, & Frijns, 2012a; Peterson, 2004), even when this was measured nonverbally (Wiefferink et al., 2013). Yet, empathy consists of two complementary aspects: affective and cognitive empathy (Baron-Cohen & Wheelwright, 2004). Affective empathy comes first in development, and refers to the supposedly innate ability to attend to others' emotions and become aroused as if feeling the same emotion, which does not have to be learned through language. This was the main aspect measured in our study, and it appears to be intact in children with CI. The presumed independence between affective empathy and language was indeed confirmed in this study by the fact that no correlations between the empathy indices and language skills were found.

The other aspect, cognitive empathy, develops during the preschool years and beyond, and refers to the ability to understand the cause of others' distress. This requires insight into others' mental states, which is dependent upon language (Astington & Jenkins, 1999; Ketelaar et al., 2012a; Peterson, 2004). Because this insight appears to be impaired in children with CI, cognitive empathy, which was not measured in our study, could be expected to be less well-developed in this particular group. Future studies should address this.

Although this study was restricted to the relation between affective empathy and children's social competence, these outcomes show that all indices for empathic behavior make a valuable contribution to the prediction of social competence, which also implies that these measures are complementary. Given the fact that the questionnaire is an instrument measuring children's empathic displays toward peers or familiar adults as judged by parents, whereas the observation instrument measures children's empathic responses toward an unfamiliar adult as judged by a trained observer, it is not surprising that these instruments produced different results and in fact measured different manifestations of empathic behavior.

Our study paints a positive picture of the social skills of children with CI, demonstrating that young, early-implanted children with CI do not show delays in social competence or of an important factor associated with social competence: empathic behavior. Early implantation combined with counseling appears to prevent the development of impairments in social functioning that are common in hearing-impaired children without CI (Ita & Friedman, 1999). This study has also indicated that the factors contributing to social functioning



Chapter 3

in children with CI are not necessarily the same as for NH children. In order to optimize the rehabilitation process it is therefore important to identify which factors contribute to the development of which skills in these children.



4

Does hearing lead to understanding?
Theory of mind in toddlers and preschoolers
with cochlear implants

The contents of this chapter are published in:
Ketelaar, L., Rieffe, C., Wiefferink, C.H., & Frijns, J.H.M. (2012).
Journal of Pediatric Psychology.

Abstract

Theory of Mind (ToM) refers to the ability to understand the subjectivity of people's intentions, desires, and beliefs. Research shows that ToM in deaf children is delayed, yet the few studies that examined ToM in deaf children with a cochlear implant (CI) report contradictory results. This study examined multiple aspects of ToM in early-implanted children. Three intention tasks were administered to 72 children with CI and 69 normal-hearing (NH) children (12 – 60 months old). Furthermore, three desire and belief tasks were administered to a subsample of children aged 30 months or over. Children with CI showed intention-understanding skills equal to NH children, but lagged behind on desire and belief understanding, even after excluding children with language delays. Children with CI appear to master the initial stages of ToM development, but fall behind on more advanced ToM abilities. Yet, both groups showed similar patterns of development.

Acknowledgments. The authors thank the parents and children who took part in this study. The authors also thank Ineke Smit and Makoto M. Tsutsui for correcting our English.

Introduction

The ability to understand that people's actions and emotions are governed by their mental states, i.e., their subjective experience of reality rather than an objective reality, is essential for adequate social functioning. This skill – called a Theory of Mind (ToM) – usually develops during early childhood (Premack & Woodruff, 1978; Wellman, 1990) and is clearly linked to language abilities (Astington & Jenkins, 1999). Research has shown that the development of a ToM in children with communicative disorders, such as children with an autism spectrum disorder, specific language impairment, or deaf children, is significantly delayed (Farrant, Fletcher, & Maybery, 2006; Peterson & Siegal, 2000). With respect to this latter group of deaf children, exposure to language is often diminished. Over 90% of deaf children are born to hearing parents (Mitchell & Karchmer, 2004) who are not skilled in sign language, which could account for language and, in turn, ToM delays in these children (Peterson & Siegal, 2000). However, nowadays over 80% of young, profoundly deaf children growing up in Western countries receive a cochlear implant (CI) (De Raeve & Lichtert, 2011; Hyde & Power, 2006). This device bypasses the damaged part of the ear by directly stimulating the auditory nerve, and enables these children to perceive sounds, although not to the same extent as people without hearing impairments. How this affects ToM development is as yet unclear, and is therefore the focus of this study.

Rehabilitation programs following cochlear implantation provide insight into the CI's efficacy in individual children by monitoring their sound perception, language acquisition, and social skills. Scientific studies have shown that cochlear implantation improves children's language and communicative skills, especially when children are implanted early, yet many children still fall behind their normal-hearing (NH) peers (Ganek et al., 2012). Other studies have shown improvements in quality of life (Loy, Warner-Czyz, Tong, Tobey, & Roland, 2010) and social competence (Ketelaar, Rieffe, Wiefferink, & Frijns, 2013; Martin et al., 2011). Improved language skills and social competence as a consequence of (early) cochlear implantation could be indicative of corresponding improvements in ToM in these children. Yet, to date, little research has been dedicated to ToM in children with CIs. Moreover, previous studies (Macaulay & Ford, 2006; Peters, Rimmel, & Richards, 2009; Peterson, 2004; Rimmel & Peters, 2009; Tasker et al., 2010) have reported mixed findings, and most have solely focused on false belief understanding as indicator of ToM. Therefore, this study



aims to contribute to the existing body of literature by a) assessing different aspects of ToM functioning of early-implanted children with CI compared to NH peers by employing a broad range of tasks, and b) examining the relation between language aspects and ToM in children with CI.

Theory of Mind

A ToM is grounded on three fundamental mental concepts that develop in a fixed order in typically-developing children (Colonnaesi, Rieffe, Koops, & Perucchini, 2008; Wellman & Liu, 2004): intentions, desires, and beliefs. Intentions refer to what we are directed at or are trying to accomplish. Desires refer to wishes, hopes, and needs. Beliefs refer to thoughts, expectations, convictions, and ideas. Together, these provide a framework for understanding complex concepts like figurative speech, white lies, jokes, mistakes, and so on (Wellman, 1990). Deficits in ToM have been related to a wide variety of negative outcomes in early childhood and beyond, including, but not limited to, peer problems (Caputi et al., 2012), externalizing behavior problems (Olson et al., 2011; Sharp, 2008), and internalizing behavior problems (Wolkenstein, Schonenberg, Schirm, & Hautzinger, 2011). Although a fully-fledged ToM is not present until children understand the subjective character of all three states, the development of children's ToM understanding varies per construct and will be discussed separately.

Intentions

At around age one, typically-developing children start to understand that people's actions are intentional. Meltzoff (1995) demonstrated that 18-month-old toddlers already make the distinction between human and inanimate agents. Toddlers who saw a mechanical device trying but failing to perform an action on an object produced the intended act less often than toddlers who saw the same behavior performed by a human agent. Several studies using Meltzoff's paradigm for intention understanding have shown that toddlers become increasingly able to comprehend other people's intentions as they grow older (Bellagamba, Camaioni, & Colonnaesi, 2006; Meltzoff, 1995).

Another indicator of children's understanding of intentions is joint attention: the process of coordinated visual attention between two people and an object or event. Infants start to reliably track an adult's gaze or pointing gesture to an object late in their first year of life, which is interpreted as the

ability to understand other people's intent to share something (e.g., Tomasello, Carpenter, & Liszkowski, 2007). Within the joint-attention paradigm, declarative comprehension refers to a child's ability to understand that another person wants to show them something, whereas imperative comprehension refers to the child's ability to understand that someone is directing their attention in order to request an object.

In children with hearing impairments, the development of intention understanding is relatively understudied. From the few studies available, it appears this ability develops normally in deaf children without implants until the age of 18 months (Spencer, 2000), but delays have been noted in older deaf children (Prezbindowski et al., 1998). To our knowledge, intention understanding in children with CI has only been examined in one study with nine children with a mean age of 30 months, which indicated that children with CI showed joint-attention skills to the same extent as NH children (Tasker et al., 2010).

Desires

In one of his ground-breaking studies, Wellman (1990) presented typically-developing two-year-olds with a story in which the protagonist likes to swim. The protagonist can either go to the park or to the swimming pool. When children were asked where they thought the protagonist would go, most were able to answer correctly. However, although these results show that two-year-olds acknowledge that desires motivate people to act or feel in a certain way, it is more difficult to assess whether children also appreciate the subjectivity of desires. It is possible that children correctly interpret the causal relationship between desires and actions or emotions, but still perceive desires as objective features (Perner, 1991). Therefore, some researchers investigated desires that were expected to be judged undesirable by children (Moore et al., 1995; Rieffe et al., 2001). These studies demonstrated that a majority of three- and four-year-old children neglected the protagonist's desire in their predictions of the protagonist's behavior or emotions. Instead, children's own desires formed the basis for their predictions of others' (emotional) behavior.

To this day, little research has examined desire understanding in children with CI. To the best of our knowledge, only one study included a measure of desire understanding amongst a range of ToM tests (Rommel & Peters, 2009). Results from this study indicated that children with CI performed equal to NH children. However, this study included only 15 children who were age-matched



to the NH control group, with ages ranging from 3 to 7 years old, and ages at implantation from 1 to 5 years old.

Beliefs

Although children grasp the subjectivity of desires before they acknowledge the subjectivity of beliefs (Wellman, 1990; Wellman & Liu, 2004), the developmental pattern of belief understanding resembles that of desire understanding. Whereas almost all typically-developing three-year-olds can correctly predict that the protagonist will look under the porch and not in the kitchen if the protagonist is looking for his dog and thinks the dog is under the porch (Wellman, 1990), this does not necessarily reflect children's understanding of the subjectivity of beliefs. The ultimate test in this regard is the classical false-belief task, originally developed by Wimmer and Perner (1983). In this task, participants have to understand that the protagonist holds a belief about reality that contradicts their own belief. Although participants know that mother moved Maxi's chocolate from the blue to the green cupboard in his absence, they should understand that Maxi does not know this, and should consequently predict Maxi's actions on the basis of his (false) belief instead of their own when Maxi returns to get his chocolate. Around the age of four children are able to correctly predict that Maxi will go to the blue cupboard. Alternative belief tasks, such as the so-called 'changed-belief task' which examines children's capacity to acknowledge their own changed beliefs about real features of the world (i.e., a rock that turns out to be a sponge, Flavell, Flavell, & Green, 1983), show the same age pattern.

What little research has been conducted on ToM in children with CI has focused mostly on false-belief understanding. Conclusions from these studies have not been uniform, with some (Macaulay & Ford, 2006; Peterson, 2004) reporting impairments, whereas others report that children with CI do understand false beliefs (Peters et al., 2009) and even find no delays compared to NH children (Rommel & Peters, 2009). These studies were conducted with small groups of children with a wide age range, both chronologically (3–12 years) and at implantation (1–6 years). Given the fact that deaf children today are implanted at ever-younger ages – which could positively affect their ToM development because of earlier access to spoken language – it is important to replicate previous research with a large group of early-implanted children.

Current Study

The main goal of this study was to compare ToM skills of young children with CI to NH children, and to examine the relations between these skills and age, timing of implantation, and language comprehension. Ideally, we would have liked to include a group of deaf children never fitted with a CI in order to determine its effects. However, nowadays the vast majority of young, profoundly deaf children are fitted with a CI (De Raeve & Lichtert, 2011; Hyde & Power, 2006). The few that do not receive an implant are often not comparable to the ones that do because they have additional handicaps or have one or two deaf parents.

Compared to previous studies, this study included a large sample of young children with CI who were implanted at an early age. Additionally, a variety of ToM measures were employed to ensure construct validity, as well as to identify group differences on each of the concepts separately. Three nonverbal intention tasks, as well as two desire tasks and one false belief task that placed minimal demand on language comprehension and production were administered. We expected that the intention-understanding skills of children with CI would be comparable to those of their NH peers because these skills in typically-developing children are first seen at a preverbal age (around the first birthday) (Bellagamba et al., 2006; Tomasello et al., 2007). Moreover, findings from the Tasker and colleagues (2010) study indicated no differences between children with CI and NH children with regard to an aspect of intention understanding: joint attention. In contrast, we expected children with CI to fall behind their NH peers on desire and belief understanding because language is required as a tool for mental reasoning, and language skills are often still impaired in children with CI (Ganek et al., 2012). We expected to find a relation between language skills and performance on ToM tasks and, because previous studies have demonstrated a relation between language and timing of implantation (cf. Ganek et al., 2012), we also expected early implantation to benefit ToM skills. Finally, we assumed that children with CI who used spoken language as opposed to sign language would have more opportunities to communicate with others, which, in turn, would benefit their ToM development.



Method

Participants and Procedure

This study included 72 children with CI from nine different counseling services and hospitals all over the Netherlands and from one counseling service in the Dutch-speaking part of Belgium. Seventy-two percent of the sample was recruited directly by health care professionals who were involved in the rehabilitation of these children. A positive response rate of 84% (16% chose not to participate) implies that this part of the sample is representative of the population of children with CI in the Netherlands. The remaining 29% of the sample was recruited via letters dispersed by the counseling services that participated in the study. The response rate was much lower; 26% chose to participate, 5% refused to participate and 69% did not respond at all. Since no information is available on the non-respondents, it is unknown whether this part of the sample is representative of the population. Additionally, 69 NH children from schools and day-care centers all over the Netherlands were included. All children were between 1 and 6 years old, were born to hearing parents, had no apparent (additional) disabilities, and children in both groups came from similar middle to high socio-economic backgrounds (judging from household income and maternal education). Table 1 shows descriptive statistics. No age differences were found between the CI and NH group. Additionally, both groups showed comparable gross and fine motor skills, which was taken as an indication of comparable cognitive functioning (Piek, Dawson, Smith, & Gasson, 2008) because it is impossible to obtain reliable IQ scores at this young age.

The majority of children with CI were born deaf. The remaining children became deaf before their second birthday, creating a homogeneous sample of children with prelingual (i.e., before language starts developing) and profound (i.e., over 90 dB in the better ear) hearing loss. Including only children with prelingual hearing loss avoided the potentially beneficial influence of language skills that developed before the onset of deafness. In addition, children had received their (first) implant at an early age, i.e., before age three, except for one child who was implanted at 3;3 years old. Two thirds of the CI group was unilaterally implanted, the others were bilaterally implanted. Parents indicated that their children wore their CI (almost) always, except for two children who wore it often. As is customary in the Netherlands and the Dutch-speaking part of Belgium, all implanted children in the sample entered a rehabilitation

Table 1 Demographic and Medical Profile of Participants

	CI (n = 72)	NH (n = 69)
Age, mean (SD), mo	37.39 (13.49)	39.51 (12.87)
Age, range, mo	14 – 60	12 – 60
Male, No. (%)	42 (58%)	39 (57%)
Socioeconomic status		
Maternal education, mean (SD) ^a	3.37 (0.89)	3.59 (0.64)
Net household income, mean (SD) ^b	3.72 (1.12)	3.54 (0.85)
Age at implantation, mean (SD), mo	16.94 (7.66)	
Age at implantation, range, mo	6 – 39	
Time with (first) CI, mean (SD), mo	19.04 (12.67)	
Time with (first) CI, range, mo	1 – 44	
Etiology of hearing loss, No. (%)		
Unknown congenital	33 (46%)	
Infection (meningitis, cytomegalovirus)	14 (19%)	
Genetic (connexin mutation, Waardenburg syndrome, large vestibular aqueduct syndrome)	6 (9%)	
Prematurity	3 (4%)	
Information unavailable	16 (22%)	
Preferred mode of communication, No. (%)		
Spoken language only	26 (36%)	
Sign or sign-supported language	46 (64%)	

^a(1 = no / primary education, 2 = lower general secondary education, 3 = higher general secondary education, 4 = college / university)

^b(1 = less than €15,000, 2 = €15,000 – €30,000, 3 = €30,000 – €45,000, 4 = €45,000 – €60,000, 5 = More than €60,000)

program. Such programs are tailored to the individual child's needs and include specialized playgroups for the deaf, monitoring (and adjustment if necessary) of the CI by an audiologist, and speech therapy.

The study was approved by the university's medical ethics committee. NH children were recruited through day-care centers, playgroups, and primary schools in the Netherlands. Children with CI were recruited through hospitals and family council services in the Netherlands and the Dutch speaking part of Belgium. Parents received information on the study in writing and were required to sign an informed consent form.

The tasks were administered to children in the CI group by one of two experimenters who were fluent in sign language and experienced in assessing language and cognitive skills of young deaf children. Depending on their preferred mode of communication, Children with CI were administered the tasks in SLN, SSD, or spoken Dutch.



The intention-understanding tasks were administered to all children regardless of age or language skills, whereas the desire and belief tasks were administered only to children aged 30 months or over, providing they met a preset level of language comprehension.

Materials

Motor and language development. The Child Development Inventory (CDI; Ireton & Glascoe, 1995) assesses general development. The scales Language Comprehension (spoken and/or signed, 50 items), Gross Motor (30 items), and Fine Motor (30 items) were used in this study. Since deaf children tend to have balance issues (Gheysen, Loots, & Van Waelvelde, 2008), seven items referring to balance were removed from the Gross Motor scale. Parents indicated for each item whether it described their child's behavior (0 = no, 1 = yes). The scales showed very high reliability (Cronbach's Alphas between .91 and .98).

To assess whether or not children had language-comprehension skills that were deemed sufficient to be administered the ToM tasks, language comprehension of children aged 30 months or over was further determined by the experimenter in two ways. First, the experimenter asked parents whether their children understood certain simple sentences (e.g., "The boy walks to his bike"). Second, children's passive vocabulary was assessed with a picture task. The experimenter named or signed 13 objects from the tasks (e.g., carrot, plane) and asked children to identify the corresponding pictures. Children were considered to have insufficient language-comprehension skills if parents reported that their children did not understand the sentences, or if children misidentified more than four pictures. ToM tasks were not administered to children younger than 30 months, or to children with insufficient language comprehension, because we assumed that they would not understand the stories and therefore, would not be able to succeed.

Intention tasks. The Intention-Understanding Task (Meltzoff, 1995) measures children's understanding of other people's intentions with regard to objects. The experimenter showed three separate intentions by repeatedly trying to perform an action but failing to succeed (e.g., dropping a string of beads into a cup). The materials were then handed to the children, who could earn a maximum of three points if they completed the intended actions.

The Imperative-Comprehension Task (Colonnesi et al., 2008) assesses joint attention by examining children's responses to the experimenter's pointing

gesture towards an object on the table that was beyond the experimenter's but within the children's reach. Children passed when they handed the object or placed it near the experimenter, or refused to give it by saying "no" or shaking their head. The task was administered three times, or until the children passed. Children earned three points if they produced the target behavior the first time, one point was deducted for each additional trial needed, down to a score of zero if the target behavior was not produced after three trials.

During the Declarative-Comprehension Task (Colonnesi et al., 2008), which measures joint attention, the experimenter pointed towards an object in the room (e.g., a poster on the wall) outside children's direct field of vision. Children could earn three points, one for each of the following behaviors: (a) looking at the object, (b) eye contact with the experimenter after looking, and (c) smiling or vocalizing toward the object.

Desire tasks. In the Common- and Uncommon-Desire Tasks (Rieffe et al., 2001), children were presented with four vignettes. Children were shown a picture of a more and a less desirable food item (e.g., a carrot and a piece of cake) and asked which item they preferred to eat. In the next picture a boy was introduced. In the two vignettes used in the Common-Desire Task, the protagonist's preference was in accordance with the child's preference. In the two vignettes used in the Uncommon-Desire Task, the protagonist's preference conflicted with the child's preference. A test question: "Which food will the boy pick?" and two control questions: "Does the boy like [food A]?" and "Does the boy like [food B]?" were asked. Children could earn one point per vignette if all three questions were answered correctly. Mean scores were calculated for the two common- and the two uncommon-desire vignettes separately.

Belief task. The False-Belief Task was adapted from Baron-Cohen, Leslie, and Frith's (1985) Sally-Anne Task. A short picture story was presented about a boy that put his toy airplane in one location and while he was away, a girl placed it somewhere else. Next, the boy returned and the children were asked the test question: "Where will the boy look for his plane?" and two control questions: "Where is the plane really?" and "Where did the boy put the plane when he went away?". Children were awarded one point if they answered all three questions correctly.



Results

Intention Tasks

A2(Group:CI,NH)x3(Task:IntentionUnderstanding,ImperativeComprehension,Declarative Comprehension) multivariate analysis of variance produced no main effect for Group, $F(1, 138) = 0.10, p = .752, \eta^2_p < .01$. In accordance with our hypothesis, the CI group performed as well as the NH group on the intention tasks. A main effect was found for Task, $F(2, 276) = 12.60, p < .001, \eta^2_p = .08$, indicating that the Intention-Understanding Task yielded lower scores than the two joint-attention tasks (Table 2).

To rule out that differences were masked by performances of the older children in the sample, independent t-tests were performed for each of the intention tasks with children under the age of 30 months only (21 CI, 14 NH). Results showed no differences between CI and NH children.

Table 2 Mean Scores on Intention and Desire and Belief Tasks as a Function of Group by Task

Instrument (min-max)	CI	NH	Between-group difference (95% CI)	Total
	Mean (SD)	Mean (SD)		Mean (SD)
<i>Intention tasks all children</i>				
	<i>n = 72</i>	<i>n = 68</i>		<i>n = 140</i>
Intention-Understanding (0-3)	1.93 (1.17)	1.97 (1.15)	-.04 (-.43, .35)	1.95 ² (1.15)
Imperative-Comprehension (0-3)	2.33 (1.04)	2.59 (0.85)	-.26 (-.57, .06)	2.46 ¹ (0.96)
Declarative-Comprehension (0-3)	2.40 (0.62)	2.21 (0.64)	.20 (-.01, .41)	2.31 ¹ (0.63)
<i>ToM tasks all children ≥ 30 months old</i>				
	<i>n = 51</i>	<i>n = 52</i>		<i>n = 103</i>
Common-Desire (0-1)	0.31 ^b (0.42)	0.67 ^a (0.44)	-.36 (-.53, -.19)	0.37 ¹ (0.46)
Uncommon-Desire (0-1)	0.20 ^b (0.36)	0.62 ^a (0.43)	-.42 (-.57, -.26)	0.30 ² (0.42)
False-Belief (0-1)	0 ^b	0.31 ^a (0.47)	-.31 (-.44, -.18)	0.12 ³ (0.32)
<i>ToM tasks children with sufficient language comprehension</i>				
	<i>n = 22</i>	<i>n = 47</i>		
Common-Desire (0-1)	0.73 ^{a1} (0.34)	0.75 ^{a1} (0.40)	-.02 (-.22, .18)	
Uncommon-Desire (0-1)	0.43 ^{b2} (0.44)	0.69 ^{a1} (0.40)	-.26 (-.47, -.05)	
False-Belief (0-1)	0 ^{b3}	0.34 ^{a2} (0.48)	-.34 (-.55, -.14)	

Note. Different letter-superscripts indicate differences at $p < .05$ on rows. Different number-superscripts indicate differences at $p < .05$ on columns.

Desire and Belief Tasks

The desire and belief tasks were administered to children aged 30 months or over, provided they had shown sufficient language comprehension as per the criteria described in the materials section. For the subsequent analysis, children with insufficient language comprehension according to these criteria were assumed to have failed these tasks and were consequently given the score 0. Two NH children refused to perform the False-Belief Task and were left out of all further analyses, leaving a total of 103 children (51 CI, 52 NH) that were included in a 2 (Group: CI, NH) \times 3 (Task: Common Desire, Uncommon Desire, and False Belief) multivariate analysis of variance. The analysis showed main effects for Group, $F(1, 101) = 35.78, p < .001, \eta^2_p = .26$ and Task, $F(2, 202) = 37.46, p < .001, \eta^2_p = .27$. In line with our expectation, the NH group outperformed the CI group. Furthermore, children performed better on the Common- than on the Uncommon-Desire Task, while the False-Belief Task yielded the lowest scores (Table 2).

Table 3 Mean Scores on Age, Language Comprehension and Timing of Implantation as a Function of Group by Language-Comprehension Skills

	Insufficient language comprehension			Sufficient language comprehension		
	CI (<i>n</i> = 29)	NH (<i>n</i> = 5)	Between-group difference (95% CI)	CI (<i>n</i> = 22)	NH (<i>n</i> = 47)	Between-group difference (95% CI)
	Mean (SD)	Mean (SD)		Mean (SD)	Mean (SD)	
Age, mo	41.03 ^{bc} (8.18)	31.20 ^c (1.79)	9.83 (2.26, 17.41)	49.45 ^a (5.28)	45.83 ^{ab} (8.55)	3.63 (.27, 6.98)
CDI, LC (0-1)	0.61 ^b (0.23)	0.81 ^{ab} (0.06)	-.21 (-.50, .08)	0.79 ^a (0.11)	0.87 ^a (0.13)	-.08 (-.15, -.01)
Implant age, mo	16.13 ^a (7.57)			19.00 ^a (8.17)		
Implant use, mo	16.33 ^b (11.69)			29.61 ^a (8.05)		

Note. Different letter-superscripts indicate differences on rows at $p < .05$.

Note. CDI, LC: Child Development Inventory, Language Comprehension.

Desire and Belief Understanding of Children with Sufficient Language-Comprehension Skills

According to the formulated criteria described in the materials section, 29 out of 51 children with CI and 5 out of 52 NH children aged 30 months or over had insufficient language-comprehension skills. Oneway ANOVAs with Bonferroni



correction showed that CI and NH children with insufficient language-comprehension skills were younger than their respective counterparts with sufficient skills, $F(3, 99) = 10.50, p < .001, \eta^2 = .24$ (Table 3). Moreover, in the CI group children judged to have insufficient comprehension skills were also scored lower by their parents on the CDI scale Language Comprehension than those with sufficient skills. Furthermore, this scale showed no differences between CI and NH children who were judged to have sufficient comprehension skills, $F(3, 73) = 11.99, p < .001, \eta^2 = .53$ (Table 3). Additionally, children with CI who had sufficient language-comprehension skills on average were implanted at the same age as children with CI who had insufficient skills, $t(45) = -.33, p = .746, \eta^2 < .01$, but they had been using their implant for a longer time, $t(45) = -2.61, p < .012, \eta^2 = .12$ (Table 3).

A 2 (Group: CI, NH) x 3 (Task: Common Desire, Uncommon Desire, and False Belief) multivariate analysis of variance for children exclusively with sufficient language-comprehension skills showed a main effect for Group, $F(1, 67) = 7.08, p = .010, \eta^2_p = .10$ and Task, $F(2, 134) = 49.06, p < .001, \eta^2_p = .42$, which was qualified by a Group x Task interaction, $F(2, 134) = 4.13, p = .018, \eta^2_p = .06$. When only children with sufficient language-comprehension skills were included, children with CI were still outperformed by their NH peers on the Uncommon-Desire Task and the False-Belief Task, but not on the Common-Desire Task. Furthermore, children with CI performed better on the Common than on the Uncommon-Desire Task, whereas this difference was not present in the NH group. Both groups had higher scores on the desire tasks than on the belief task (Table 2).

Influence of Language and Implantation Timing on Task Performance

Relations of language and implantation timing with the belief task could not be assessed for the CI group because all children with CI failed this task.

To examine whether children who used spoken language performed better on the intention and the desire and belief tasks than children who used sign language, children with CI were divided into two groups: one group ($n = 26$) that relied solely on spoken Dutch and one group ($n = 46$) that relied on some form of sign language (i.e., Sign Language of the Netherlands, Sign-Supported Dutch, or a combination of modes). Contrary to our expectations, multivariate analyses of variance revealed no differences in scores on the intention tasks, $F(1, 70) = 0.04, p = .834, \eta^2_p < .01$ or the desire and belief tasks, $F(1, 49) = 0.83, p = .367, \eta^2_p = .02$ between children who did or did not use signs.

Table 4 shows correlations of Age, the CDI scale Language Comprehension, and implantation timing with the desire and belief tasks for each group separately. Correlations corrected for age were also calculated because age was assumed to be a confounding variable; older children tend to have better ToM skills, but also better language skills and, in the case of the CI group, probably more experience with their CI. Age correlated with all desire and belief tasks in both groups. Language Comprehension correlated with all tasks for both groups. Unexpectedly, however, after correcting for Age only the correlation with the Common-Desire Task for the NH group remained significant, although we did see a trend ($p = .052$) for the CI group. Contrary to findings from studies on language development (Ganek et al., 2012), and to our own expectations, age at implantation and duration of implant use were not related to performance on the desire and belief tasks. Because of the strong correlation between age and duration of implant use ($r = .63, p < .001$), correlations when controlled for duration of implant use are not reported.

Table 4 Correlation Coefficients (Partial Correlations Corrected for Age) of ToM Tasks with Age, Language, and Timing of Implantation per Group

	CI			NH		
	Age	CDI, LC	Implant Age	Implant Use	Age	CDI, LC
Common-Desire	.54**	.42* (.33)	.15 (.05)	.28 (-.09)	.60**	.64** (.41*)
Uncommon-Desire	.36*	.31 (.23)	.04 (-.04)	.22 (-.01)	.56**	.51** (.23)
False-Belief	^a	^a	^a	^a	.66**	.45* (.01)

* $p < .01$; ** $p < .001$ (2-tailed).

^aNo correlations were computed because all children with CI failed the False-Belief Task.

Note. CDI, LC: Child Development Inventory, Language Comprehension.



Discussion

Theory of Mind (ToM) is a very important skill for social functioning which starts to develop in early childhood, and is known to be impaired in deaf children (Peterson & Siegal, 2000). Early cochlear implantation is thought to help deaf children develop a ToM, but the effect of a CI on young children's ToM functioning has been relatively underexamined. The current study therefore focused on ToM in a large group of early-implanted children. To date, the few studies that have been conducted with children with CI almost all focused on false-belief tasks as sole index for ToM (Macaulay & Ford, 2006; Peters et al., 2009; Peterson, 2004), whereas two other important aspects of ToM, intention understanding and desire understanding, have received very little attention (Rommel & Peters, 2009; Tasker et al., 2010). The current study aimed to measure children's performance on all three aspects of ToM in order to identify if, and where on the spectrum, children with CI show deficits.

Even though children in the current study were implanted at a considerably earlier age than children in previous studies, their ToM was still affected. As expected, outcomes of this study show that children with CI can understand other people's intentions to the same extent as NH children. Yet, in contrast to some studies (Peters et al., 2009; Rommel & Peters, 2009), but in line with others (Macaulay & Ford, 2006; Peterson, 2004), we found that children with CI have difficulties understanding other people's desires and (false) beliefs in comparison to NH peers, even after correcting for verbal skills. Possibly, children with CI show a developmental delay with respect to ToM functioning. Alternatively, these children might follow a qualitatively different developmental path that allows them to master the initial stage of ToM (i.e., intention understanding), but does not allow them to grasp the more complex concepts of diverse desires and beliefs. The performance of the CI group on the various tasks resembles the pattern of development found in NH children, with understanding of intentions developing before understanding of desires and beliefs (Wellman, 1990), which supports the assumption of a delayed rather than a qualitatively different development. However, only longitudinal research can confirm this assumption.

Another question that remains to be answered is why children with CI have difficulties developing a fully functioning ToM. Although research with NH children has demonstrated a close link between language and ToM

(Astington & Jenkins, 1999), and language is often still impaired in children with CI (Ganek et al., 2012), our study demonstrated that ToM was also impaired in a subsample of children with CI with adequate language skills. Therefore, even though cochlear implantation is known to benefit language abilities (Ganek et al., 2012), we cannot expect interventions targeted at promoting language in children with CI to also improve their ToM. We suggest that attention be directed at the specific content of conversations between children with CI and their parents as it is unclear whether quality of conversation is comparable for children with CI and NH children. Mental-state conversations, for example during picture-book reading, are of particular interest because of the link to ToM development found in both NH children (Ruffman, Slade, & Crowe, 2002) and deaf children without CI (Moeller & Schick, 2006), and the fact that parents often report problems engaging in mentalistic conversations with their deaf children (Peterson & Siegal, 2000). If this assumed link is also present in children with CI, intervention and rehabilitation programs should try to boost parents' skills to communicate with their children about mental states.

This study significantly contributes to existing literature in this area because of our larger sample sizes, and thus greater statistical power. Therefore, we are able to draw firm conclusions regarding the ToM abilities of these children. However, generalizations to other children with CI should be made with caution because of the heterogeneity of this population. First, not all children benefit from their CI to the same extent, even when they are implanted at the same age and with the same device. Second, although some factors that could have influenced ToM development, such as age at implantation and duration of implant use, have been taken into account in this study, numerous other potential factors come to mind such as unilateral versus bilateral implantation, speech perception after implantation, and type of device implanted. A far larger sample would have been needed for a thorough study of the influence of each of these factors. Finally, research into speech development after implantation has shown a gradual improvement of abilities until five years post-implantation (O'Donoghue, Nikolopoulos, Archbold, & Tait, 1998). This might also apply to ToM development. Given the limited amount of experience the children had with their CI (19 months on average), it is possible that the ToM delay will gradually vanish when they have had more opportunity to take advantage of their implants. Consequently, replication of these findings is desirable, as well as collecting longitudinal data for several years after implantation.



Future studies should also address whether the early ToM impairments that we found in these children with CI affect other areas of functioning (e.g., peer relations), and contribute to the development of later psychopathology (e.g., aggression, depression), as has been observed in typically-developing children (Caputi et al., 2012; Olson et al., 2011; Sharp, 2008; Wolkenstein et al., 2011), or whether these negative outcomes can be prevented if children with CI indeed catch up to NH children on ToM skills. Nonetheless, this study is an initial effort to gain an understanding of the development of this growing group of children that seem to fall between two stools: not deaf, but not quite like NH children either. There are important practical implications for parents and professionals (e.g., teachers, counselors). They should be made aware of the developmental delays and be given tools to assist the children in building a ToM, especially since more and more children with CI attend mainstream schools and are treated as NH children.



5

The interplay between moral emotions and social behavior in young children with normal hearing and with cochlear implants

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Abstract

Moral emotions such as shame, guilt and pride are the result of an evaluation of the own behavior as (morally) right or wrong. The capacity to experience moral emotions is thought to be an important driving force behind socially appropriate behavior. Nonetheless, the relation between moral emotions and social behavior in young children has not been studied extensively, let alone in those with a hearing impairment. This study compared young children with hearing impairments who have a cochlear implant (CI) to normally hearing (NH) peers regarding the extent to which they display moral emotions, and how this relates to their social functioning and language skills. Responses of 184 NH children and 60 children with CI (14 to 61 months old) to shame/guilt and pride inducing events were observed. Parents reported on their children's social competence and externalizing behavior, and experimenters observed children's cooperative behavior. Additionally, children's language skills were assessed. Results show that children with CI displayed moral emotions to a lesser degree than NH children. A relation between moral emotions and social functioning was found in the NH group, but not in the CI group. Language skills were unrelated to moral emotions, yet emotion vocabulary was related to social functioning. Facilitating emotion language skills in addition to general language skills could promote children's social functioning. Future studies should examine which factors are associated with moral emotions, particularly in children with CI.

Acknowledgments. We thank the children and their parents for participating in this study. We also thank the students who have helped collecting data with the NH children, and Anke Otten and Bernadette Vermeij for collecting data with the children with CI.

Introduction

Research in the normal-hearing (NH) population has demonstrated that moral emotions such as shame, guilt, and pride are important determinants of social competence, reflected in being liked by others, for example (Barrett, 1995; Stearns & Parrott, 2012). Conversely, an impaired ability to experience these emotions is associated with a range of undesirable behaviors in preadolescents, including bullying and aggression (Menesini & Camodeca, 2008; Olthof, 2012) and, at the far end of the spectrum, even criminal behavior and psychopathy in adolescents and adults (Holmqvist, 2008; Mealey, 1995). These studies indicate that moral emotions play a significant role in regulating social behavior. Research examining this relation in younger children is largely lacking, but is warranted given these striking findings in older children and adolescents.

The ability of children with hearing impairments to experience and express moral emotions, and how this ability relates to their social functioning, has received little or no attention to date even though social problems are known to exist in this population. Children and adolescents with prelingual, severe to profound hearing loss often experience social difficulties, for example manifested in problematic peer relations (Wolters et al., 2011), a higher incidence of behavior problems (Barker et al., 2009), and more symptoms of psychopathy (i.e., cold-blooded manipulative behavior, lack of empathy) (Theunissen et al., in press) compared to NH peers. These days, the vast majority of young children with severe to profound hearing loss born in Western countries receive a cochlear implant (CI), often before their second birthday (De Raeve & Lichtert, 2011; Hyde & Power, 2006). This electronic device bypasses the damaged part of the ear by directly stimulating the auditory nerve which, combined with extensive rehabilitation, enables sound perception and benefits spoken language skills (De Raeve, 2010; Ganek et al., 2012). Yet, how cochlear implantation affects these young children's social functioning, and which factors underlie their social development, is largely unknown. Determining risk or protective factors is important in light of optimizing rehabilitation programs for children with CI. This study is the first to explore the moral development of children with CI, and to examine its relation to these children's social functioning.



Function and Development of Moral Emotions

Emotions have a social function, motivating a person to find a balance between the own interest and certain social requirements, in order to optimize interpersonal relations (Keltner & Haidt, 1999; Rieffe & De Rooij, 2012). Yet, a number of theorists have argued that moral emotions take a special position in the spectrum of emotions (e.g., Barrett, 1995; Eisenberg, 2000; Lewis et al., 1992; Shariff & Tracy, 2009; Tangney, Stuewig, & Mashek, 2007; Tracy & Robins, 2004a). Crucially, moral emotions include a self-evaluative component, and occur when people judge their own behavior as (morally) right or wrong. This self-awareness or self-reflection causes the individual to correct the own behavior or even better, to prevent oneself from making moral transgressions in the future (Barrett, 1995; Tangney et al., 2007). Each moral emotion has its own function, and a corresponding pattern of behavior. Although the same event may cause shame in one person and guilt in the next (Tangney et al., 2007), a distinction between these two emotions can be made based on appraisals concerning stability and globality (Tracy & Robins, 2004a). Shame arises when a failure or transgression is attributed to a global and stable cause (e.g., 'I broke the vase because I am clumsy'). Guilt, on the other hand, is caused by specific and unstable attributions (e.g., 'I broke the vase because I did not look where I was going'). Both emotions communicate to others that you are aware of your transgression and feel bad. Yet, shame is associated with escape-related behavior (e.g., making yourself smaller, avoiding eye contact), whereas guilt is associated with reparative behavior (e.g., apologizing, trying to undo the consequences) (Barrett, 1995; Tangney et al., 2007). Pride can be felt when you have accomplished something notable, which makes you want to repeat or sustain the behavior that led up to this emotion. Its expression is aimed at drawing attention to this accomplishment (e.g., expanded posture, making eye contact), and signals to others that you (temporarily) deserve a higher status within the group (Shariff & Tracy, 2009; Tracy & Robins, 2004b). In sum, moral emotions discourage inappropriate, i.e., morally incorrect, and reinforce appropriate, i.e., morally correct, behavior. Therefore, it is not surprising that emotionally competent NH children - those who know when and how to express (moral) emotions - are generally perceived to be more socially competent as well (Barrett, 1995; Denham et al., 2003; Stearns & Parrott, 2012).

Moral emotions require certain insights and capacities, which develop over time. Children need to be aware of the dominant moral standards, and

have to be able to evaluate their own behavior in this context (Lewis et al., 1992; Tangney et al., 2007; Tracy & Robins, 2004a). Yet, in a review of the empirical literature, Draghi-Lorenz, Reddy, and Costall (Draghi-Lorenz et al., 2001) argued that signs of pride, guilt or shame can already be observed in infancy, before children have internalized moral standards. At this early age these emotions are a result of external evaluations and not of children's own judgment of their behavior (Mills, 2005). Whether or not children with CI will express moral emotions to the same extent as NH children when their behavior is evaluated by others is unclear. However, we do know that children with CI have difficulties recognizing other people's emotions (Wang et al., 2011; Wiefferink et al., 2013), and are less sensitive to intonation (Most & Michaelis, 2012). Therefore, these children might not pick up on more subtle forms of feedback, which for example is relayed by someone's facial expression or tone of voice. If children with CI are indeed less aware of other people's evaluations of their behavior, this could prevent them from displaying the appropriate moral emotion.

During the toddler period, most NH children become increasingly able to evaluate their own behavior based on what they have learned from previous feedback, and will start to generalize this knowledge to other situations. From three years up, NH children start to internalize a personal set of moral standards which will eventually channel their (emotional) behavior, independent from outside guidance (Lewis et al., 1992; Mills, 2005; Tracy & Robins, 2004a). In order for this personal set of moral standards to develop, children first need to be able to judge their own behavior through the eyes of other people, which requires certain socio-cognitive abilities. The best-known example of these is the so-called Theory of Mind (ToM), which entails the capacity to take other people's perspective into account (Premack & Woodruff, 1978). NH children show a major development in their ToM understanding between the ages of 2 and 5 years old (Wellman, 1990), but ToM skills of children with CI are known to fall behind during this crucial period. In early and middle childhood, children with CI are less able than their NH peers to predict other people's behavior based on these people's desires and expectations, but tend to use their own frame of reference instead (Ketelaar et al., 2012a; Peterson, 2004). A limited understanding of other people's perspectives also implies a limited understanding of other people's judgments about their behavior. Consequently, children with CI may be less inclined to express moral emotions because they do not realize that they have done something that would be judged as reprehensible or admirable by others.



The Role of Communication and Socialization

Emotions are subjective experiences in response to meaningful events. Yet, how these emotions are interpreted and displayed is modulated by the social environment (Barrett, 1995; Keltner & Haidt, 1999). Children learn to regulate their emotions and behavior through interactions with role models; by observing parents' responses to events, but also through verbal communication. Children with CI have experienced a period of time before receiving their CI during which they were deprived of access to spoken language, and even with a CI a large proportion of these children still faces language delays (Ganek et al., 2012). In addition, parents of children with hearing impairments more often have difficulties conversing with their children, particularly about abstract topics (Peterson & Siegal, 2000). This includes having conversations about emotions; helping children to label their emotions and discussing how to communicate emotions appropriately. Moreover, this could also prevent parents from discussing moral matters with their children. Language delays and restricted communication could seriously impair the quality of interactions between hearing parents and their children with CI and, in turn, these children's ability to understand their own and other people's emotions (Morris, Silk, Steinberg, Myers, & Robinson, 2007). Moreover, children with CI are likely to miss out on so-called incidental learning, i.e., picking up conversations in which they are not directly addressed, as has been shown for children with hearing impairments who did not have a CI (Vaccari & Marschark, 1997). An impoverished quality of interactions with NH people in their immediate surroundings combined with a limited capacity for incidental learning could negatively impact these children's ability to become independent moral agents.

Current Study

This study's first aim is to examine the extent to which young children with NH or with CI display moral emotions in an experimental setting. Because of communication difficulties and limited opportunities for incidental learning (Peterson & Siegal, 2000; Vaccari & Marschark, 1997), children with CI presumably have had less opportunity to learn and internalize moral standards, and subsequent moral emotions. Moreover, these children have an impaired insight into other people's emotions and perspective (Ketelaar et al., 2012a; Peterson, 2004; Wang et al., 2011; Wiefferink et al., 2013), which further hampers their ability to make inferences about their own behavior from cues in their

environment. Therefore, we expect children with CI to display moral emotions to a lesser degree than NH peers.

Second, the interplay between moral emotions and social behavior is examined. Because moral emotions play such a crucial role in social functioning of the NH population at later ages (Holmqvist, 2008; Menesini & Camodeca, 2008; Stearns & Parrott, 2012), we expect to observe this relation already early in life, both in the NH group and in the CI group. Given that we expect lower levels of moral emotions in children with CI in comparison to their NH peers, we expect that this will also lead to lower levels of social functioning in these children.

Third, we wish to verify whether communication indeed plays an important role in the development of moral emotions. We examine children's language skills as a determinant of communication. Particularly emotion-related language might be important for children's social-emotional development. Regardless of children's hearing status, we expect to find a positive relation between their ability to understand and use emotion language and the extent to which they express moral emotions. Within the CI group, we also assess whether general spoken language abilities are related to moral emotions. Fourth, we explore whether earlier implantation promotes children's social and emotional functioning similar to what has been found for their spoken language skills (Ganek et al., 2012).

Table 1 Sample Characteristics

	CI (n = 60)	NH (n = 184)
Age, mean (SD), mo	38 (14.3)	38 (12.1)
Age, range, mo	14 - 61	14 - 61
Male, No. (%)	36 (60%)	110 (60%)
Socioeconomic status		
Maternal education, mean (SD) ^a	3.46 (0.83)	3.63 (0.62)
Net household income, mean (SD) ^b	3.68 (1.12)	3.65 (0.96)
Age at implantation, mean (SD), mo	16 (7.3)	
Age at implantation, range, mo	6 - 35	
Time with (first) CI, mean (SD), mo	21 (12.8)	
Time with (first) CI, range, mo	1 - 44	

^a(1 = no / primary education, 2 = lower general secondary education, 3 = higher general secondary education, 4 = college / university).

^b(1 = < €15,000, 2 = €15,000 - €30,000, 3 = €30,000 - €45,000, 4 = €45,000 - €60,000, 5 = > €60,000).



Methods

Participants

This study is part of a larger research project which focuses on various areas of the social-emotional development of young children with CI and with NH. A total of 244 children (60 with CI and 184 with NH) from the Netherlands and the Dutch-speaking part of Belgium participated in this study. All children were born to hearing parents and had no apparent mental health disorders such as ADHD or autism spectrum disorders. Characteristics of the samples are reported in Table 1. Age, gender and socioeconomic status (based on maternal education and net household income) did not differ between the groups. All children with CI had prelingual, severe to profound hearing loss and had received their (first) implant before the age of three years. Sixty-eight percent had been using their CI for 12 months or more at the time of data collection. Parents indicated that practically all children were wearing their CI full-time. Approximately half the group had two implants, the other half had one. All children with CI entered a tailored rehabilitation program after implantation, which includes joining specialized playgroups, receiving technical support for the device, speech therapy, and visits with a psychologist. At the time of data collection, 22 children (37%) preferred to solely use spoken language, the remaining 38 children (63%) preferred to use some form of signed language, mostly Sign-Supported Dutch (i.e., spoken Dutch supported by signs).

Materials

Indices for moral emotions

Shame/guilt. Three tasks were designed to evoke feelings of shame and/or guilt. In the Broken Car Task, children were led to believe they had broken the experimenter's toy car (i.e., the wheels would come off when children played with it). The other two tasks involved failure on an assignment that appeared to be easy, which supposedly evokes shame and/or guilt (Lewis et al., 1992). In the Copy Task, children were asked to copy a drawing made by the experimenter. Children always received negative feedback upon completion of their drawing. In the Bottle Task, the experimenter asked children to open a bottle that, unknowingly to the child, was equipped with a child-proof safety cap. The experimenter opened and closed the bottle before handing it to the child, demonstrating it could be opened easily. Based on previous research

(Alessandri & Lewis, 1993; Barrett, 1995; Lewis et al., 1992), the occurrence of the following four behaviors was coded on a three-point scale (0 = *not at all*, 1 = *a little*, 2 = *a lot*) for each of the tasks: 1) negative response to the situation, 2) gaze aversion / turning away from situation, 3) collapsed body, 4) corners of the mouth turned down / lower lip pushed outward (pouting). Hiding one's face was also scored but showed a floor effect for all tasks and was removed from the scale. A single, overall score (ranging between 0 and 2) for shame/guilt was computed by averaging the ratings of the four items across the three tasks. The reliability of the scale meets the expected minimum of Cronbach's Alpha $> .70$ (Nunally, 1978) (Table 2).

Pride. Two pride-evoking tasks were administered, both of which involved mastery. These two tasks directly followed their shame/guilt-evoking counterparts, which would have set the stage for children to believe that these tasks were hard. Mastering them this time around was assumed to evoke pride (Lewis et al., 1992). In the Copy Task, children again were asked to copy a drawing but this time were given positive feedback. In the Bottle Task, the experimenter looked at the bottle that the child had been unable to open and 'discovered' that the cap was screwed on wrong. She then (without the child noticing) released the safety lock before handing the bottle back and encouraging the child to try again. The experimenter made sure children succeeded to open the bottle this time. Based on previous studies (Alessandri & Lewis, 1993; Lewis et al., 1992; Tracy & Robins, 2004b), three separate cues for pride were scored on a three-point scale (0 = *not at all*, 1 = *a little*, 2 = *a lot*): 1) positive response to situation, 2) smiling / laughing, 3) eye contact and erect posture. Pointing to the outcome or applauding was also scored but showed a floor effect for both pride tasks and was removed from the scales. A single, overall score (ranging between 0 and 2) for pride was computed by averaging the ratings of the three items across the two tasks. The reliability of the scale meets the expected minimum of Cronbach's Alpha $> .70$ (Nunally, 1978) (Table 2).

Indices for social functioning

Social Competence was assessed by calculating mean scores for the items of the Prosocial and Peer Problems scales (5 items each) from the Dutch parent-report version of the Strengths and Difficulties Questionnaire (SDQ) (Goodman, 1997; Muris et al., 2003). Parents rated each item on a three-point scale (0 = *not true*, 1 = *somewhat true*, 2 = *certainly true*). Negatively formulated



items on the Peer Problems scale were reversed so that higher scores were indicative of less peer problems. The reliability of the scale meets the expected minimum of Cronbach's Alpha $> .70$ (Nunally, 1978) (Table 2).

Cooperation. Following each test session, experimenters completed a questionnaire that was designed for the purpose of this study concerning the child's behavior during the test session. The scale Cooperation (9 items) reflects the extent to which children were motivated to complete the tasks, and how responsive they were to the experimenter's instructions. Items were rated on a three-point scale (0 = *not*, 1 = *sometimes*, 2 = *often*), mean scores across the items were calculated. The reliability of the scale meets the expected minimum of Cronbach's Alpha $> .70$ (Nunally, 1978) (Table 2). Data were missing for one NH child.

Externalizing Behavior was assessed by calculating mean scores for the Hyperactivity and Behavioral Problems scales (5 items each) from the Dutch parent-report version of the SDQ (Goodman, 1997; Muris et al., 2003). Parents rated each item on a three-point scale (0 = *not true*, 1 = *somewhat true*, 2 = *certainly true*). The reliability of the scale meets the expected minimum of Cronbach's Alpha $> .70$ (Nunally, 1978) (Table 2).

Indices for language

Emotion Vocabulary. Children's emotion language was measured with the Emotion Vocabulary Questionnaire, a parent-report measure that was designed for the purpose of this study. Parents rated whether their children knew and used (either in spoken or sign language) each of 20 emotion and/or mental state words (0 = *no*, 1 = *yes*). Basic emotions such as happy or angry, more complex emotions such as jealous or disappointed, and mental states such as dreaming or thinking were represented in the questionnaire. A mean score across the items was calculated to indicate children's emotion vocabulary. The reliability of the scale meets the expected minimum of Cronbach's Alpha $> .70$ (Nunally, 1978) (Table 2). Data were missing for one child with CI.

Spoken language understanding and production scores of children with CI were obtained via records from hospitals and counseling services. Part of the rehabilitation process after implantation involves monitoring children's language development, most commonly by administering the Dutch versions of the Reynell Developmental Language Scales for language understanding and the Schlichting Expressive Language Test for word and sentence production (Van

Eldik, 1998). Hospitals and counseling services were asked to provide children's most recent scores. Recent language scores were unavailable for 11 children, and the language skills of 5 additional children were assessed with alternative, incomparable instruments, leaving us with language scores of 43 children with CI (Table 2). Using independent sample *t* tests, we compared children in the CI group with and without language scores and found no differences regarding age or any of the indices for social-emotional functioning.

Procedure

Children with NH were recruited through day-care centers, preschools, and elementary schools in the Netherlands. Children with CI were recruited through hospitals and family counseling services all over the Netherlands and the Dutch-speaking part of Belgium. All children were tested individually in a quiet room at home, school or hospital. The emotion-evoking events were interspersed among other tasks not presented in this manuscript. Parents filled in questionnaires. Additional information, such as household income and age at implantation, was obtained from parents and/or medical records. Informed consent was obtained for all children and the study was approved by the university's medical ethics committee.

The tasks were nonverbal in nature (common gestures made clear what was expected) and did not require a verbal response from the children. The tasks were administered to children in the CI group by one of two hearing experimenters who were fluent in Dutch sign language and sign-supported Dutch. Children with CI were addressed in their preferred mode of communication (spoken or signed language) during the test session.



Table 2 Internal Consistencies, Means, and SDs for Measures of Social and Emotional Functioning

	No. of items	Min-Max	Cronbach's Alpha	Inter-item correlation	CI (n = 60)	NH (n = 184)
					M(SD)	M(SD)
<i>Moral emotions</i>						
Shame***	12	0-2	.79	.24	0.19 (0.19)	0.41 (0.33)
Pride*	6	0-2	.81	.41	0.70 (0.49)	0.89 (0.54)
<i>Language</i>						
Emotion Vocabulary***	20	0-1	.92	.37	0.46 (0.26)	0.57 (0.28)
Language Understanding					86.49 (17.59)	
Word Production					89.08 (18.67)	
Sentence Production					84.09 (14.47)	
<i>Social functioning</i>						
Social Competence	10	0-2	.70	.17	1.42 (0.35)	1.48 (0.33)
Externalizing Behavior	10	0-2	.71	.20	0.61 (0.38)	0.53 (0.31)
Cooperation	9	0-2	.87	.43	1.62 (0.41)	1.62 (0.43)

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

Results

Group Differences on Moral Emotions and Social Functioning

To examine group differences for moral emotion expressions, social functioning and language, ANCOVAs controlling for age were performed. The ANCOVA for Shame/guilt revealed a main effect for Group (NH, CI), $F(1, 241) = 27.38$, $p < .001$, Cohen's $d = 0.84$. Similarly, the ANCOVA for Pride revealed a main effect for Group (NH, CI), $F(1, 241) = 6.47$, $p = .012$, Cohen's $d = 0.39$. Mean scores per group (Table 2) indicate that children with CI expressed moral emotions to a lesser extent than NH children. No main effects for Group (NH, CI) were found for any of the measures of social functioning (Social Competence: $F(1, 241) = 1.70$, $p = .194$, Cohen's $d = 0.18$; Cooperation: $F(1, 240) = 0.00$, $p = .990$, Cohen's $d = 0.00$; Externalizing Behavior: $F(1, 241) = 3.11$, $p = .079$, Cohen's $d = 0.23$). Additionally, the ANCOVA for Emotion Vocabulary showed a main effect for Group (NH, CI), $F(1, 240) = 16.48$, $p < .001$, Cohen's $d = 0.41$, indicating that NH children knew and used more emotion words than children with CI. Spoken language skills were only obtained for children with CI. On average, children with CI scored 1 SD below the normative mean ($M = 100$, $SD = 15$) (Table 2).

Relation of Moral Emotions with Social Functioning, and the Role of Language

Shame/guilt and Pride increased with age in both groups of children. Cooperation also increased with age in both groups, whereas Social Competence only increased with age in the NH group. Externalizing Behavior was unrelated to age in the NH group, but increased with age in the CI group (Table 3). Because age was a confounding variable, correlations between moral emotions and social functioning were calculated corrected for age (Table 4). Results for the NH group show that Shame/guilt was related to Social Competence and that Pride was related to both Social Competence and Cooperation. Moral emotions were unrelated to Externalizing Behavior in the NH group. Results for the CI group show that moral emotions were unrelated to social functioning.

Next, we explored the association between various indices of language and the expression of moral emotions, as well as social functioning (Table 5). Emotion language, as indexed by Emotion Vocabulary, was unrelated to moral emotions in both groups. It was however positively related to Social Competence in both groups, and negatively related to Externalizing Behavior in the CI group only. None of the indices for spoken language skills in the CI group were related to moral emotions or social functioning.

Finally, we examined the effect of implantation timing on indices of moral emotions and social functioning in the CI group. As can be seen in Table 3, younger age at implantation and longer use of the implant were positively related to Pride but not to Shame/guilt or any of the indices for social functioning. Although not an initial focus point of this study, we also examined whether implantation timing affected children's language skills, like previous research indicated (Ganek et al., 2012). In accordance with previous studies, we found that younger age at implantation and longer time with CI (both corrected for chronological age) were related to better spoken language skills. However, implantation timing was unrelated to emotion language, as indicated by children's Emotion Vocabulary (Table 3).



Table 3 Pearson Correlations of Moral emotions, Social Functioning, and Language with Age and Implantation Timing

	Chronological age		Age at implantation ^a	Time with CI ^a
	NH	CI	CI	CI
<i>Moral emotions</i>				
Shame/guilt	.33***	.40**	.00	-.01
Pride	.25**	.40**	-.33*	.35**
<i>Social functioning</i>				
Social Competence	.34***	.09	-.18	.17
Cooperation	.51***	.55***	-.18	.17
Externalizing Behavior	-.09	.34**	.21	-.21
<i>Language</i>				
Emotion Vocabulary	.76***	.73***	-.26	.25
Language Understanding		.17	-.48**	.47**
Word Production		.14	-.50**	.48**
Sentence Production		.04	-.58***	.57***

^aCorrected for chronological age

* $p < .05$. ** $p < .01$. *** $p < .001$ (two-tailed).

Table 4 Pearson Correlations of Moral emotions with Social Functioning per Group, Corrected for Age

	Shame/guilt	Pride	Social Competence	Cooperation	Externalizing Behavior
Shame/guilt	-	.19	.10	.23	-.10
Pride	.27***	-	-.04	.12	.06
Social Competence	.29***	.18*	-	.12	-.41***
Cooperation	.03	.16*	.19*	-	-.12
Externalizing Behavior	-.10	.00	-.41***	-.18	-

Note. Right upper corner shows correlations for CI group, left lower corner shows correlations for NH group.

* $p < .05$. ** $p < .01$. *** $p < .001$ (two-tailed).

Table 5 Pearson Correlations of Language Indices with Moral emotions and Social Functioning, Corrected for Age

	Shame/guilt	Pride	Social Competence	Cooperation	Externalizing Behavior
Emotion Vocabulary	.11/-.05	-.08/-.15	.35***/.39**	.11/.05	-.05 / -.56***
Language Understanding	-.08	.00	.12	.08	-.13
Word Production	-.11	.08	.19	.14	-.09
Sentence Production	-.02	.17	.19	.06	-.15

Note. Correlations with Emotion Vocabulary are provided separately for NH and CI respectively. Correlations with other language indices are provided for CI group only.

* $p < .05$. ** $p < .01$. *** $p < .001$ (two-tailed).

Discussion

Research shows that moral emotions such as shame, guilt and pride promote positive social behavior and protect against negative social behavior in the typically developing population (Menesini & Camodeca, 2008; Stearns & Parrott, 2012; Tangney et al., 2007). Yet, to our knowledge, this study is the first to examine whether the link between moral emotions and social behavior can already be observed in young children (age 1 to 5). We examined this in a group of typically developing children and in a group of atypically developing children, that is children with hearing impairments who had received a CI. The latter group of children is assumed to have a limited capacity for acquiring social-emotional skills because of restricted communication with their surroundings (Peterson & Siegal, 2000; Vaccari & Marschark, 1997). We aimed to examine how this would affect their ability for moral and social functioning.

Our study confirms previous studies (Barrett, 2005; Lewis et al., 1992) which demonstrated that young, typically developing children already display moral emotions, and that this ability increases with age. As expected, children with CI expressed shame/guilt to a lesser extent than their NH peers in response to staged emotion-evoking events (i.e., failing on a mastery task and breaking a toy). In addition, children with CI also showed less pride than NH children when they succeeded on a mastery task. General feedback on children's performance or behavior was provided by the experimenter, which should have focused children's attention on their failure or transgression (or in the case of pride, on their success). Nonetheless, children with CI seemed to be less aware than their NH peers of what was expected of them in terms of moral behavior in these situations. On a positive note, as in the NH group, a relation was found between age and moral emotions in children with CI. This could imply that moral skills develop along the same lines as in NH children, but at a different pace. However, longitudinal studies should of course confirm this.

Expectations concerning the interplay between moral emotions and social behavior were not met. First, we expected a relation between the extent to which NH children expressed moral emotions and their level of social behavior. Yet, we only found a relation between moral emotions and positive social behavior, but not with negative social behavior. In other words, a better-developed moral sense did promote positive behavior, but did not seem to prevent NH children from displaying negative social (i.e., externalizing) behavior.



It should however be noted that parents of NH children reported quite low levels of behavior problems, which could have masked an association between moral expressiveness and negative behavior. Future studies could try to examine this association in children who show high levels of behavior problems. Second, we expected that the interplay between moral and social behavior would be similar in the NH and the CI group. In contrast to this expectation, we found no associations between moral emotions and social behavior in the CI group. As in the NH group, parents of children with CI reported low levels of behavior problems in their children, which could explain the lack of an association between moral emotions and negative social behavior. Yet, the absence of a relation between moral emotions and positive social behavior requires some additional attention. Levels of positive as well as negative social functioning were equal in both groups of children, which, in combination with the absence of a relation between moral emotions and social functioning in children with CI, leads us to question the importance of a delayed development of moral emotions for these children.

The absence of a relation between moral emotions and social behavior in children with CI could be explained by differences in the ways children acquire social-emotional competence. Instilling a moral sense in children is not about teaching them a repertoire of socially appropriate behaviors, yet it is about providing children with the resources to judge their own (intended) behavior as right or wrong. Children are likely to receive explicit feedback on the behavior they display, particularly if this behavior stands out in a positive or a negative way. Yet, the feedback provided will not always include an evaluation of the morality of children's behavior. Consequently, children might have to rely on more implicit ways of learning in order to develop a moral sense, one of these being incidental learning. Children with CI have less opportunities than their NH peers for incidental learning. They have difficulties overhearing other people's conversations, particularly in noisy environments (Boons et al., 2012b). This could explain why the social skills of children with CI are comparable to those of their NH peers while their moral development is delayed. Moreover, factors related to the rehabilitation program that children in the Netherlands enter after receiving their CI could also play a role. Children with CI receive a lot of attention from adults during their frequent visits to the hospital and in their specialized playgroups. This could provide them with ample models for appropriate social behavior. Yet, it is unlikely that these role models also explain

how and when to express moral emotions.

Although the social skills of children with CI seem to develop well, it remains to be seen whether these will continue to develop at the same pace as NH children's social skills, in the absence of equally well-developing emotional skills. As children grow older, more sophisticated social skills are expected, which could draw more heavily on children's emotional skills. Children with CI are found to have an impaired ToM (Ketelaar et al., 2012a; Peterson, 2004), which hampers their ability to judge their own behavior from another person's perspective, and thus could prevent them from experiencing and displaying moral emotions when these are called for. A lack of expressing moral emotions following a transgression does not seem to damage the social relationships of children with CI at this early age. Yet, children's social competence was evaluated by parents and parents of children with CI might be more protective and consequently more forgiving towards their children than NH parents. Down the line, peer relations become more important and peers may be less forgiving when children cross the line without showing remorse. There is no doubt that everyone will violate the social norms from time to time. Yet, individuals who display or report feelings of shame or guilt following a transgression are less likely to be socially rejected than those who seem to be indifferent to their wrong-doing (Martens, Tracy, & Shariff, 2012; Stearns & Parrott, 2012).

To verify whether communication is important for children's moral and social development, we examined one of its key components: language. Language did not turn out to be the important determinant of children's moral emotion expressions we had hypothesized. Children who used more emotion language did not express more moral emotions. Emotion language was, however, associated with more positive social behavior in both groups, and with less negative behavior in the CI group. Important to note is that general spoken language skills of children with CI did not influence their emotional or their social functioning. These outcomes support the assumption previously made by other researchers (Peterson & Slaughter, 2003) that it is not just the ability to understand and produce general language that is important for social-emotional functioning; it is the ability to understand and use emotion language in daily conversations that is critical for adequate social functioning.

A recent study by Quittner and colleagues (2013) demonstrated that parental behaviors, including language stimulation and maternal sensitivity, had a major impact on language outcomes in children with CI. Nonetheless, parents



might continue to experience difficulties conversing about abstract concepts such as emotions with their implanted children, despite these children having adequate concrete language skills. In a study by Zaidman-Zait (2008), 39% of parents reported that they experienced difficulties communicating with their children with CI. A previous study by the current authors demonstrated that children with CI who had well-developed general language understanding still had an impaired theory of mind in comparison to NH age mates (Ketelaar, Rieffe, Wiefferink, & Frijns, 2012b). Additionally, emotion language was unrelated to implantation timing in the current study, whereas general language was better developed in children who had been implanted at a younger age, or who had been using their implant for a longer time. Together, these findings validate that professionals and parents should actively try to promote the emotion language skills of children with CI in order to enhance their ability to understand their own and other people's (emotional) behavior, and to further raise their level of social skills.

The outcomes of this study should be interpreted with caution and represent a preliminary step towards understanding the interplay between moral and social behavior. We included a specific subset of children with CI, who were implanted before age three, had hearing parents, and no apparent mental health disorders. Generalization to the whole population of children with CI, of which approximately one third is reported to have an additional disability (Fortnum et al., 2002; Holden-Pitt & Diaz, 1998), is problematic. Furthermore, the correlational and cross-sectional nature of the study precludes drawing any conclusions as to causation. Another point which needs to be addressed concerns the type of data gathered for the purpose of this study. Although parents are a well-informed source regarding their young children's behavior, their reports on their children's social and externalizing behavior may have been biased. Parents of children with CI in particular may not have been aware of young children's normative behavior, and could have been overly positive regarding their children's capacities. Likewise, we cannot be certain that children's responses to the experimental tasks designed to induce moral emotions were a genuine reflection of how they would behave in a real-life situation. Therefore, adopting a multi-informant approach in future studies is advisable. Clearly, more (longitudinal) research is necessary to confirm the current findings.

Moreover, the outcomes of this study give rise to new research questions. For example, regarding which factors contribute to children's moral development. As moral development in children with CI turns out to be impaired, it is even more important to gain an understanding of the underlying reasons for this impairment. This might provide us with valuable information on how moral development in these children could be promoted. Attention could be directed at children's ToM understanding as a likely contributing factor. In addition, it might also be worthwhile to assess parent-child communication more directly instead of by means of children's language skills, i.e., through observational measures. Nonetheless, despite its limitations, this study constitutes an important first step in shedding more light on the moral development of children with CI, and its relation to social functioning. The outcomes of this study demonstrate that young children with CI have a less well-developed moral sense than their NH peers. Older children with CI did however express moral emotions more often than younger children with CI, which suggests a delayed rather than a qualitatively different moral development. At this young age, impaired expression of moral emotions did not have negative consequences for social functioning in children with CI. In contrast, moral behavior did turn out to be important for NH children's social functioning, but only in terms of positive social behavior.





6

Children with cochlear implants and their parents: The influence of parenting on children's social-emotional functioning

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Abstract

Parenting a child with a hearing impairment is challenging and at times stressful, and might cause parents of children with hearing impairments to employ more adverse parenting styles as compared to parents of children with normal hearing. Parenting styles, in turn, are known to impact children's social-emotional development. Children with hearing impairments may rely even more on their parents regarding their social-emotional development than their peers with normal hearing who can interact with and learn more easily from others outside the family. Identifying the role parenting styles play in the social-emotional development of children with hearing impairments who have cochlear implants could help advance these children's well-being. Therefore, this study aimed to compare parenting styles of parents with children with normal hearing and of parents with children with a cochlear implant, and to examine the relations between parenting styles and two key aspects of children's social-emotional functioning: emotion regulation and empathy. Ninety-two parents with normal hearing and their children (aged 1 to 5 years old) with either normal hearing or with a cochlear implant participated in the study. Parents filled in questionnaires concerning their parenting styles (i.e., positive, negative and uninvolved), and regarding the extent to which their children expressed negative emotions (i.e., anger and sadness) and empathy. Furthermore, an emotion regulation task measuring negative emotionality was administered to the children. No differences in reported parenting styles appeared between parents of children with normal hearing and parents of children with cochlear implants. Additionally, negative and uninvolved parenting were related to higher levels of negative emotionality in both groups of children. The initially found relation between positive parenting and children's empathy disappeared when language skills were added to the regression model. In conclusion, children's hearing status did not impact parenting styles, which might be a result of the support parents of children with cochlear implants receive during their enrollment in the rehabilitation program following implantation. Uninvolved parenting in particular turns out to be related to emotion dysregulation in children. Future research should address the directionality of the relations between parenting styles and children's social-emotional functioning.

Acknowledgments. The authors thank the parents and children who were willing to participate in this study. Additionally, we thank Bernadette Vermeij and Anke Otten-Koens for collecting the data for the CI group, and several Psychology students from Leiden University for assisting with the data collection for the NH group.

Introduction

Currently, most children with severe to profound hearing impairments in the Western world receive a cochlear implant (CI) (Bradham & Jones, 2008; De Raeve & Lichtert, 2011; Fortnum et al., 2002; Hyde & Power, 2006; Johnston, 2004; Preisler et al., 2002), a device that electrically stimulates the auditory nerve, bypassing the damaged part of the ear. The positive effects of this technology are undisputable. Numerous studies have shown that auditory and language skills improve in the majority of children with hearing impairments (HI) following cochlear implantation (e.g., Boons et al., 2012a; De Raeve, 2010; Ganek et al., 2012; Nikolopoulos et al., 1999; Niparko et al., 2010; Svirsky et al., 2000). A by comparison much smaller number of studies has been dedicated to children's social-emotional development following cochlear implantation. In general, these studies show that Quality of Life increases and that symptoms of psychopathology and social problems diminish (Bat-Chava, Martin, & Kosciw, 2005; Huttunen et al., 2009; Huttunen & Valimaa, 2010; Nicholas & Geers, 2003; Schorr, Roth, & Fox, 2009; Theunissen et al., in revision). Notwithstanding, the social and emotional development of young children with a CI is often not yet on a par with their peers with normal hearing (NH). For example, young children with CI exhibit impairments on verbal as well as nonverbal tasks of emotion understanding (Ketelaar et al., 2012a, 2013; Most & Michaelis, 2012; Peterson, 2004; Wang et al., 2011; Wiefferink et al., 2013; Wiefferink et al., 2012a). Identification of factors that influence these children's social-emotional development is vital if we want to promote their well-being.

Bronfenbrenner's (1979) social-ecological model proposes that a child's development is not only affected by intrapersonal factors, such as temperament or physiology, but also by interpersonal factors, such as family, neighborhood, and school. Parents in particular play an important role in the life of young children. Therefore, this study aims to examine the extent to which different



parenting styles are related to aspects of social-emotional functioning in young children with CI, compared to their NH peers.

Two key aspects of social-emotional functioning are emotion regulation and empathy. Emotion regulation is defined as the ability to manage one's emotions in ways that are adaptive to one's social functioning (Denham et al., 2003; Morris et al., 2007). Although regulation can entail decreasing as well as increasing the intensity, frequency and duration of either negative or positive emotions, particularly the ability to decrease negative emotions is found to be important with respect to social functioning (Eisenberg et al., 2001; Eisenberg et al., 2003; Zeman, Cassano, Perry-Parrish, & Stegall, 2006). Whereas emotion regulation involves one's own emotions, empathy is directed at other people's emotions. Empathy denotes the ability to acknowledge and be affected by other people's emotions. It motivates to respond to others in distress, for example by offering consolation or support (McDonald & Messinger, 2011; Rieffe et al., 2010). Empathy has been referred to as the 'social glue' in relationships, and is considered to be an important determinant of children's social functioning (Baron-Cohen & Wheelwright, 2004; Eisenberg & Miller, 1987; Jolliffe & Farrington, 2006).

Parenting a Child with a Hearing Impairment

Parents play a crucial role in many areas of child development, including, but not limited to, language and social-emotional functioning (Amato & Fowler, 2002; Morris et al., 2007; Newland & Crnic, 2011; Stack et al., 2010; Vaccari & Marschark, 1997; Van Aken et al., 2007). This might apply even more to parents of children with HI. For these children, who are often faced with communication difficulties and corresponding social problems (Fellinger, Holzinger, & Pollard, 2012; Kouwenberg et al., 2012; Van Eldik et al., 2004; Wolters et al., 2011), opportunities for incidental learning by overhearing conversations between other people are assumed to be limited (Vaccari & Marschark, 1997). At the same time, parents of children with HI (with or without CI) encounter more challenges and experience more stress concerning their role as parents than parents of children with NH (Hintermair, 2006; Quittner et al., 2010), which might have a detrimental effect on their child-rearing practices and consequently, on the development of children with HI.

Most parents who are told their child has a severe to profound hearing loss are facing an unfamiliar situation, taking into consideration that

approximately 90% of deaf children are born to hearing parents (Mitchell & Karchmer, 2004). Moreover, shortly after they have learned about their child's hearing loss, parents have to make decisions with life-long consequences for their child, including if and when their child should receive a CI. The decision to opt for a CI can be stressful for parents. A recent survey conducted among a large sample of parents of children with CI revealed that, although the majority of parents reached a decision within three months time, approximately half of the parents considered it stressful to decide on cochlear implantation (Hyde, Punch, & Komesaroff, 2010). Yet, another study with a same-size sample reported that most parents did not find the decision particularly difficult to make because they felt that cochlear implantation without a doubt was the best option for their child (Sach & Whynes, 2005).

Once parents have decided to proceed with cochlear implantation, they will likely encounter another emotional and stressful period following the implantation. Zaidman-Zait (2008) reported that general stress levels of 16% of parents with a child with CI were in the clinical range. Parents have to deal with uncertainties about the needs and future of the child. For example, should their child be addressed in sign language or spoken language, and should their child attend mainstream or special education? Furthermore, parents have to find a balance between the amount of time they ought to invest in for example supporting their child at home, taking sign language courses, or visiting the hospital on the one hand, and the amount of time they have to spend at work or doing household chores on the other hand. Meeting all of these demands could take a toll on family life, and many parents indeed feel that they are unable to spend enough time with their partner or with the other children in the household (Sach & Whynes, 2005). When parents were asked to describe the everyday problems they encountered within the context of parenting a child with CI, 26% of parents commented on the demands of, and parents' own responsibilities concerning the rehabilitation process (Zaidman-Zait, 2008).

Another stressor experienced by hearing parents of children with HI is the mismatch between communication modes, when their child has a low ability to communicate in spoken language (Zaidman-Zait, 2008). Most parents lack fluency in sign language, which makes it difficult for them to have conversations with their children (Vaccari & Marschark, 1997). Several studies have shown that parent-child interactions occur less frequently and/or extensively with children with HI than with children with NH (Barker et al., 2009; Gale & Schick, 2009;



Prezbindowski et al., 1998). Discussing abstract topics, such as emotions, is particularly difficult for parents of children with HI (Vaccari & Marschark, 1997), and could cause social-emotional difficulties in these children (Moeller & Schick, 2006; Peterson & Siegal, 2000).

Cochlear implantation could potentially reduce these communication problems, because it makes spoken language much more attainable to children with hearing losses in the severe to profound range (Ganek et al., 2012; Niparko et al., 2010). One of the main outcomes of cochlear implantation parents hope for and expect is that their children will be able to use spoken language as a mode of communication (Nikolopoulos, Lloyd, Archbold, & O'Donoghue, 2001; Zaidman-Zait & Most, 2005). The few studies that have examined whether this goal was reached indicated a shift from sign language toward spoken language in children following cochlear implantation (Huttunen & Valimaa, 2010; Watson, Archbold, & Nikolopoulos, 2006; Watson, Hardie, Archbold, & Wheeler, 2008). It should be noted, however, that a CI does not ensure optimal spoken language development right away, if at all. Nor does it make up for the fact that these children were sound-deprived before the implantation took place. A recent review by Ganek and colleagues (2012) showed that spoken language was often still delayed in these children, despite early implantation. These continued language issues are also acknowledged by parents. In a study by Zaidman-Zait (2008), 39% of parents reported that they encountered difficulties when communicating with their child with CI in spoken language.

In sum, parents of children with CI experience stressors which are unique to this group (Hyde et al., 2010; Sach & Whynes, 2005), and which negatively impact both the quality and the quantity of communication (Barker et al., 2009; Gale & Schick, 2009; Prezbindowski et al., 1998), and consequently, these children's social-emotional development (Moeller & Schick, 2006; Peterson & Siegal, 2000). Moreover, parental stress among parents of children with CI could be associated with more adverse parenting styles.

Parenting Styles

Parenting styles refer to a set of parenting practices and strategies that parents employ during child-rearing. Three main categories of parenting styles are authoritative, authoritarian and neglectful parenting (Baumrind, 1966; Maccoby & Martin, 1983). Authoritative parenting, which we will call positive parenting, is characterized by high responsiveness and high demandingness. Parents

who employ this parenting style are sensitive to their child's needs, interact in positive and meaningful ways with their children, praise their children when they have done something good, and will set clear standards while granting their children autonomy. A positive parenting style has been found to be related to adaptive social-emotional functioning in children with NH (Kawabata et al., 2011; Stack et al., 2010). Past studies suggest less sensitivity and responsiveness among mothers of children with HI than among mothers of children with NH (Meadow-Orlans, 1997; Meadow-Orlans & Spencer, 1996). Yet, a study on the related subject of maternal emotional availability (which subsumes sensitivity, among other things) revealed no differences between parents of 1-year-old children with HI or with NH (Pressman, Pipp-Siegel, Yoshinaga-Itano, Kubicek, & Emde, 1998).

Authoritarian parenting, which we will call negative parenting, is characterized by low responsiveness and high demandingness. Parents employing this style are quite strict with their children, they have a tendency to use a lot of disciplining, without allowing much room for discussion. A negative parenting style is found to hamper a healthy social-emotional development in children with NH (Kawabata et al., 2011; Newland & Crnic, 2011; Stack et al., 2010). Parents of children with HI are reported to show more of an inclination towards using physical discipline (Knutson et al., 2004), which suggests that they are more likely to employ a negative parenting style than parents of children with NH.

Neglectful parenting, which we will call uninvolved parenting, is characterized by low responsiveness as well as low demandingness. This parenting style can involve ignoring or disregarding the child, and showing inconsistent parental behaviors, which causes unpredictability for the child. Uninvolved parenting has been related to more aggressive behavior in children with NH (Kawabata et al., 2011; Van Aken et al., 2007). To our knowledge, this particular parenting style has not been examined among parents of children with HI.

It is important to determine whether this pattern of parenting styles found in children with HI can also be observed in implanted children. Moreover, parenting styles might show a different relation to social-emotional development in children with CI than in children with NH. Although no studies on this topic are available, studies concerning the influence of parenting on language development do point in this direction. For example, a recent study by



Quittner and colleagues (2013) showed that maternal sensitivity had as much of an effect on language development of children with CI as the age at implantation, suggesting that parenting styles might even have a stronger influence on the development of children with CI compared to their peers with NH. Similarly, parental involvement could be even more crucial for children with HI than for children with NH, because parents need to learn how to communicate with their child with HI, for example by taking sign language courses and adhering to prerequisites such as facing the child when speaking or signing. Moeller (2000) found that parental involvement in an intervention program (in which parents were enrolled in after their child was diagnosed with a HI) was the most important predictor of vocabulary skills in children with HI among a range of factors studied (e.g., non-verbal IQ and child's age at enrollment in the program).

Even after the CI is implanted and children with HI are able to perceive sounds, continued parental involvement is required. Children have to learn to assign meaning to sounds in order to understand and use spoken language. To support this, children in the Netherlands are enrolled in a rehabilitation program after implantation. As described elsewhere in this manuscript, participation in such programs require parents to frequently visit the hospital, doing speech exercises with their children at home, taking their children to specialized play groups, and so on. Sarant and colleagues (2009) found that a higher socioeconomic status (SES) was related to more parental involvement in these rehabilitation programs which, in turn, positively influenced children's language skills.

Current Study

The first aim of the present study is to examine whether parents of children with a CI employ different parenting styles from parents of children with NH. With the exception of the study by Quittner and colleagues (2013) on the influence of maternal sensitivity on children's language development, we found no studies concerning parenting styles of parents who have a child with a CI. Although the literature reveals that parents of children with HI (without CI) use more negative and less positive parenting styles (Knutson et al., 2004; Meadow-Orlans, 1997; Meadow-Orlans & Spencer, 1996), we do not know whether this also applies to parents of children with CI. Because parenting a child with CI is demanding, requiring strong and positive involvement, we expect that parents of children with CI will report involved parenting equal to, or more than parents of children with NH.

The second aim of this study is to explore the relations between parenting styles and indices of social-emotional functioning. Links have been established between parenting and emotion regulation skills in children with NH (e.g., Eisenberg et al., 2003; Morris et al., 2007; Zeman et al., 2006), as well as between parenting and empathy in children with NH (e.g., Cornell & Frick, 2007; Valiente et al., 2004). To our knowledge, no studies have been conducted thus far examining these links in children with HI (with or without CI). Parents may exert a stronger influence on the lives of children with CI as compared to children with NH. Consequently, we expect stronger relations between parenting styles and children's social-emotional functioning in children with CI than with NH. Additionally, we expect SES and children's language skills to affect these relationships.

Method

Participants

The total sample participating in this study consisted of 92 children aged 1 to 5 years old: 46 children with CI and 46 children with NH. The samples were matched on age, sex, and SES (calculated from maternal education and net household income). Additionally, gross and fine motor skills were assessed using the Dutch version of the Child Development Inventory (CDI; Ireton & Glascoe, 1995). As IQ cannot be reliably measured in very young children, motor skills were taken as an indication of cognitive development (cf. Piek, Dawson, Smith & Gasson, 2008) on which we could compare the samples. After removing five items referring to balance, because HI children are known to frequently have problems with their organ of balance (Gheysen et al., 2008), from the gross motor scale, no differences appeared between the two samples. All children were born to hearing parents and had no (other) disabling conditions. Additionally, all children with CI had severe to profound prelingual hearing loss, and had been implanted before their third birthday. Seventy-two percent of the sample had 12 months or more experience with their (first) CI. Half of the sample was unilaterally implanted, the other half was bilaterally implanted. Parents reported on their children's daily CI use on a 4-point scale ((almost) never, sometimes, often, (almost) always). All but two children were wearing their CI (almost) always, the remaining two were wearing their CI often. The majority (65%) of children with CI preferred to use



some form of signed language instead of relying solely on spoken language. Detailed characteristics of both samples are reported in Table 1.

Table 1 Characteristics of the Samples

	CI (<i>n</i> = 46)	NH (<i>n</i> = 46)
Age, mean (SD), mo	37.7 (14.2)	37.6 (14.1)
Age, range, mo	14 - 61	15 - 61
Sex – <i>n</i> (%)		
Male	28 (61%)	28 (61%)
Female	18 (39%)	18 (39%)
Socioeconomic status		
Maternal education, mean (SD) ^a	3.53 (0.76)	3.73 (0.50)
Net household income, mean (SD) ^b	3.69 (1.17)	3.94 (1.00)
Motor development		
Gross motor skills, mean (SD)	0.68 (0.23)	0.74 (0.23)
Fine motor skills, mean (SD)	0.65 (0.22)	0.67 (0.24)
CI characteristics		
Age at implantation, mean (SD), mo	16.0 (6.7)	
Age at implantation, range, mo	7 - 35	
Time with (first) CI, mean (SD), mo	21.6 (12.7)	
Time with (first) CI, range, mo	2 - 45	
Preferred mode of communication, No. (%)		
Oral language only	16 (35%)	
Sign-supported Dutch	21 (46%)	
Sign Language of the Netherlands	3 (7%)	
Combination of communication modes	6 (13%)	
Spoken language skills		
RDLS, Understanding, mean (SD)	87.09 (20.64)	
SELT-W, Production, mean (SD)	92.03 (17.03)	
SELT-S, Production, mean (SD)	85.94 (13.48)	

Note. RDLS = Reynell Developmental Language Scales; SELT-W = Schlichting Expressive Language Test, Word; SELT-S = Schlichting Expressive Language Test, Sentence

^a(1 = no/primary education, 2 = lower general secondary education, 3 = higher general secondary education, 4 = college/university).

^b(1 = less than €15,000, 2 = €15,000 - €30,000, 3 = €30,000 - €45,000, 4 = €45,000 - €60,000, 5 = more than €60,000).

Materials

Negative Emotionality

The *Negative Emotion Expression* scale (8 items) from the Emotion Expression Questionnaire (EEQ; Rieffe et al., 2010) indicates the frequency and intensity of children's negative emotion expressions, and the extent to which they are easily calmed when angry or sad. Parents rated the degree to which each item described their child on a 5-point response scale, ranging from 0 = (*almost*) *never* to 4 = (*almost*) *always*.

The *Emotion Regulation Task* (Wiefferink et al., 2012a) examines children's responses to a frustrating event. Children were handed a bottle to open after the test leader demonstrated it could be opened easily. Yet, children did not know that the bottle had a child-proof safety cap. The test leader waited 30 to 60 seconds and coded the child's responses on a checklist (3 items) before helping the child to open the bottle. An example item is 'The child shows a negative facial expression'. The test leader scored each item on a 3-point scale, ranging from 0 = *not at all* to 2 = *a lot*.

The 5-point *Negative Emotion Expression* scale was transformed to a 3-point scale and subsequently combined with the *Emotion Regulation Task*, resulting in the scale *Negative Emotionality* (11 items). Mean scores across items were calculated for each child. The internal consistency of this composite scale per group is acceptable (Cronbach's $\alpha > .70$, see Table 2).

Empathy

The *Empathy Questionnaire* (EmQue; Rieffe et al., 2010) is a 19-item parent-report instrument that measures children's empathic behavior in daily social situations. Parents rated the degree to which each behavior was displayed by their child in the past two months on a 3-point scale, ranging from 0 = *never* to 2 = *often*. Subsequently, mean scores across all items of the questionnaire were calculated. The internal consistency of the questionnaire per group is good (Cronbach's $\alpha \geq .80$, see Table 2).

Parenting styles

Following Van Aken and colleagues (2007), we used scales from various parenting questionnaires. These scales were subsequently combined to reflect the three parenting styles under examination: positive parenting, negative parenting, and uninvolved parenting.



The scale *Positive Parenting* was a combination of the Responsivity scale from the Dutch Parenting Questionnaire (Gerris et al., 1993), the scale Reinforcement of good behavior from the Alabama Parenting Questionnaire (Shelton, Frick, & Wootton, 1996), and the scale Positive interactions from the Parent Practices Scale (Strayhorn & Weidman, 1988). This resulted in a 16-item scale.

The scale *Negative Parenting* was a combination of the Discipline scale from the Parent Behavior Checklist (Fox, 1994) and the scale Overreaction from the Parenting Scale (Irvine, Biglan, Smolkowski, & Ary, 1999). This resulted in a 10-item scale.

The scale *Uninvolved Parenting* was a combination of the scale Love withdrawal from the Dutch Parenting Questionnaire (Gerris et al., 1993) and the Inconsistency scale from the Alabama Parenting Questionnaire (Shelton et al., 1996). This resulted in a 10-item scale.

Parents rated how often they showed each parenting behavior on a 5-point scale, ranging from 0 = (almost) never to 4 = (almost) always. Subsequently, mean scores across the items of the scales were calculated. The internal consistencies of the scales are acceptable (Cronbach's $\alpha > .70$, see Table 2).

Language skills

Two measures were used to assess children's language skills. A questionnaire was administered to parents of both groups of children in order to assess everyday language skills, regardless of modality (i.e., signed or spoken language). Additionally, data on the spoken language skills of children with CI were obtained via hospital or counseling service.

The *Comprehension* and *Expression* scales from the Dutch version of the Child Development Inventory questionnaire (Ireton & Glascoe, 1995), each containing 50 items, were administered to parents. Items on both scales addressed syntactic, pragmatic, semantic, and intelligibility aspects. Parents indicated for each item whether it described their child's abilities (0 = no, 1 = yes). Because we were interested in the general language skills of children, not just their spoken language abilities, parents were asked to answer "yes" when their child mastered the topic in either spoken or sign language. For each scale, scores on all 50 items were summed and subsequently divided by the total number of items, resulting in a mean score ranging between 0 and 1. The internal consistencies of these scales are good (Cronbach's $\alpha > .90$, see Table 2).

Spoken language understanding and production skills of children with CI are frequently assessed as part of the rehabilitation process. The most commonly used language tests in this respect in the Netherlands are the Dutch version of the Reynell Developmental Language Scales (RDLS) to measure language understanding, and the Schlichting Expressive Language Test (SELT) to measure word and sentence production (Van Eldik, 1998). The RDLS and SELT provide age norms based on typically developing children with NH ($M = 100$, $SD = 15$). The most recent RDLS and SELT scores of children with CI in our sample were provided by hospitals and counseling services. For 11 children with CI (24%), recent language scores were either not available, or children were assessed with other, incomparable tests. To examine whether the subsample of children with CI for whom Reynell and Schlichting scores were available was comparable to the subsample for whom these scores were not available, we performed independent sample t tests on age, parenting styles and social-emotional functioning. No significant differences between the subsamples were detected on any of the study variables ($p > .05$).

Procedure

Children with CI were recruited from various counseling services and hospitals all over the Netherlands. Children with NH were recruited from day-care centers and schools throughout the Netherlands. Recruitment letters to eligible participants were dispersed through these organizations. Once informed consent from the parents was obtained, children were tested individually in a quiet room. Children with CI who communicated wholly or partially in some form of signed language were tested by a researcher who was familiar with spoken and sign language. Parents filled in questionnaires concerning parenting behaviors, their children's behaviors, and variables such as SES and age at implantation. The study was approved by the university's medical ethics committee.



Table 2 Psychometric Properties and Mean Scores for all Variables

	No. of items	Min-Max	Cronbach's alpha		Mean scores (SD)		
			CI	NH	CI	NH	Total
<i>Social-emotional functioning</i>							
Negative Emotionality	11	0-2	.72	.73	0.59 (0.21)	0.61 (0.22)	
Empathy	19	0-2	.80	.86	0.86 (0.30)	0.90 (0.32)	
<i>Parenting styles</i>							
Positive Parenting	16	0-4	.74	.86	3.25 (0.28)	3.09 (0.38)	3.17 ¹ (0.34)
Negative Parenting	10	0-4	.84	.86	1.02 (0.48)	1.05 (0.56)	1.03 ² (0.52)
Uninvolved Parenting	10	0-4	.73	.79	0.88 (0.47)	0.89 (0.52)	0.89 ³ (0.49)
<i>Language skills (signed or spoken)</i>							
CDI, Comprehension	50	0-1	.97	.99	0.58 (0.28)	0.68 (0.32)	
CDI, Expression	50	0-1	.97	.97	0.62 (0.27)	0.71 (0.35)	

Note. CDI = Child Development Inventory

Note. Different number-superscripts indicate differences at $p < .05$ on columns.

Statistical Analyses

To examine group differences in parenting styles and children's social-emotional functioning, a multivariate analysis of variance and independent samples t tests were carried out. Relationships between parenting style and indices of social-emotional functioning were examined by means of Pearson's correlations. Fisher r to Z transformations were used to examine whether correlations between variables were stronger in one group than in the other. Subsequently, hierarchical regression analyses (method enter) were performed to examine the unique contributions of parenting style, SES and language skills on children's social-emotional functioning. In order to examine whether the contributions of the various parenting styles differed between the groups, interactions between the centered independent variables and group (dummy-coded; 0 = NH, 1 = CI) were added to the regression analyses.

Results

Table 2 shows the mean scores for the two indices for children's social-emotional functioning (Empathy and Negative Emotionality), the three parenting styles (Positive, Negative and Uninvolved), and language skills (Language Comprehension and Language Expression). Independent samples *t* tests for Negative Emotionality and Empathy revealed no differences between the groups ($t(90) = 0.43, p = .670, \eta^2 = .00$ and $t(90) = 0.49, p = .629, \eta^2 = .00$ respectively). A 2 (Group: CI, NH) \times 3 (Parenting style: Positive, Negative, and Uninvolved) multivariate analysis of variance produced a main effect for Parenting style ($F(2, 150.30) = 722.95, p < .001, \eta^2_p = .89$) but not for Group ($F(1, 90) = 0.62, p = .434, \eta^2_p = .01$). Post-hoc paired *t* tests revealed that parents in both groups of children most often showed positive parenting behaviors, followed by negative parenting behaviors. Uninvolved parenting behaviors were shown least often.

Independent samples *t* tests demonstrated equal language skills in both groups as reported by parents on the CDI (Comprehension: $t(90) = 1.59, p = .116, \eta^2 = .03$; Expression: $t(85.67) = 1.40, p = .165, \eta^2 = .02$). This measure was insensitive to language modality (i.e., spoken or signed language). Yet, the spoken language skills of children with CI, as assessed with the RDLS and SELT were not up to par. Scores in Table 1 show that, on average, children with CI scored one *SD* below the norm of children with NH. Yet, there was considerable variability within the CI group.

Partial correlations corrected for age between the parenting styles showed that Positive Parenting correlated negatively with Negative Parenting and Uninvolved Parenting. In turn, Negative Parenting was positively related to Uninvolved Parenting. The strength of the correlations of Age and parenting styles with children's social-emotional functioning did not differ between the groups ($p > .05$), except for Age \times Negative Emotionality. Age was more strongly correlated to Negative Emotionality in the CI group than in the NH group ($Z = -2.71, p = .007$). All other correlations were collapsed over the two groups. As can be seen in Table 3, Negative and Uninvolved Parenting were positively related to Negative Emotionality, but unrelated to Empathy. Positive Parenting was unrelated to both indices for children's social-emotional functioning.



Table 3 Pearson Correlations between Parenting Styles Indices for Social-Emotional Functioning, and Indices for Language (Corrected for Age)

	1	2	3	4	5
Age	-.20	.46***	.32**	.27 / .70***	.33**
1. Positive Parenting	-	-.32**	-.42***	.02	.16
2. Negative Parenting		-	.51***	.21*	.00
3. Uninvolved Parenting			-	.24*	.10
4. Negative Emotionality				-	.03
5. Empathy					-
6. CDI, Language Comprehension	.15	.01	-.04	.08	.35**
7. CDI, Language Production	.17	.02	-.12	.00	.34**
8. RDLS	.23	.09	-.33	-.05	.19
9. SELT-W	.22	.23	.12	.27	.41*
10. SELT-S	.22	.27	.01	.22	.35*

Note. CDI = Child Development Inventory; RDLS = Reynell Developmental Language Scales; SELT-W = Schlichting Expressive Language Test, Word Development; SELT-S = Schlichting Expressive Language Test, Sentence Development

Note. Correlations with the RDLS, SELT-W and SELT-S are available for the CI group only

Note. Only when correlations significantly differed in strength, these are provided separately for the NH and CI group respectively.

After correcting for age, Language Comprehension nor Language Expression, as measured with the CDI, were related to any of the parenting styles. Furthermore, Language Comprehension and Language Expression were also unrelated to Negative Emotionality, but were positively related to Empathy. A similar pattern was revealed for children with CI regarding their spoken language skills (i.e., RDLS and SELT scores). After correcting for age, spoken language skills of children with CI were unrelated to any of the parenting styles or to Negative Emotionality. However, spoken language production (on word and sentence level), but not spoken language understanding, turned out to be positively related to Empathy (Table 3).

To examine unique relations of parenting style, age, and language skills with children's social-emotional functioning, regression analyses were carried out separately for each index of social-emotional functioning. Group, Age and the Parenting Styles were included in Step 1. In Step 2 we added Language Comprehension as a possible mediator. Language Expression was not included because of the high correlation with Language Comprehension ($r = .96, p < .001$). The interaction terms of Parenting Styles with Group were included in Step 3. The regression models (Table 4) show significant explained

variance. Age and Uninvolved Parenting were uniquely related to Negative Emotionality. Adding Language Comprehension did not improve the model, nor did adding the interaction terms of Parenting Styles with Group. Age and Positive Parenting were related to Empathy (Step 1). However, after including Language Comprehension in Step 2, relations of Age and Positive Parenting with Empathy disappeared, and only Language Comprehension was uniquely related to Empathy. Adding the interaction terms of Parenting Styles with Group did not produce a better model. The regression analyses were also carried out including SES but this did not result in an increase in model fit. Additionally, regression analyses were run including Language Expression instead of Language Comprehension, but this produced similar outcomes. Therefore, these results were omitted.

Table 4 Hierarchical Regression Analysis for Negative Emotionality and Empathy (N = 92)

	Negative Emotionality			Empathy		
	R^2_{adj}	β	p	R^2_{adj}	β	p
<i>Step 1</i>	.26***			.12**		
Group		-.09	.352		-.11	.294
Age		.37	<.001		.32	.005
Positive Parenting		.17	.106		.27	.023
Negative Parenting		.13	.261		-.03	.814
Uninvolved Parenting		.24	.046		.23	.076
<i>Step 2</i>	$\Delta R^2 = ns$.21***		
Group					.03	.765
Age					-.30	.174
Positive Parenting					.17	.138
Negative Parenting					-.06	.649
Uninvolved Parenting					.22	.079
Language Comprehension					.70	.002
<i>Step 3</i>	$\Delta R^2 = ns$			$\Delta R^2 = ns$		

Note. Empathy: $\Delta R^2 = .09$ for Step 2 ($p = .002$).

* $p < .05$; ** $p < .01$; *** $p < .001$



Discussion

Parenting styles have repeatedly been found to have a major effect on the development of children with NH in all areas, including emotion regulation, mental health, and academic achievement (Amato & Fowler, 2002; Lieb et al., 2000; Morris et al., 2007). The finding that negative and uninvolved parenting are associated with negative emotionality confirms prior research which reported that these parenting styles hamper the social-emotional development of children (Kawabata et al., 2011; Stack et al., 2010). Yet, it also extends our knowledge by demonstrating that this association not only applies to children with NH, but also to children with CI. The relatively high level of covariance between negative and uninvolved parenting might explain why only uninvolved parenting was uniquely related to negative emotionality in children.

This study failed to confirm the direct influence of positive parenting on children's social-emotional lives, but instead suggests an indirect effect, mediated by children's language skills. This might be a consequence of the age range chosen in this study. At these ages, language is still very much in development. Prior research with children with NH in this age range showed high levels of externalizing behavior, which was, among other things, attributable to their limited capacity for expressing themselves verbally, which led to frustration and anger (Alink et al., 2006). Our study showed that those children who had better language skills displayed more positive behavior, which probably allowed parents to approach them in a more positive way. Nonetheless, despite the children in this study being in a difficult age period, most parents still favored a positive parenting style over the two other styles, possibly keeping a healthy balance in their approach towards their children.

In light of this study, an outcome of utmost importance was the absence of any differences between parents of children with NH or with CI. Parents of children with HI usually report more parental stress than parents of children with NH (Quittner et al., 2010), but at least for parents of children with CI this does not seem to negatively affect their parenting practices. Potentially, parents of children with CI have increased access to (social) support in comparison to parents of children with HI who do not have a CI. Following cochlear implantation, children and their parents in the Netherlands take part in a rehabilitation program which includes regular visits to the audiologist, speech therapist and psychologist, as well as house calls by a family counselor. This

provides them with frequent opportunities to gain advice from professionals on how to raise their child to the best of their abilities.

No differences between the groups were found regarding two aspects of children's social-emotional functioning: emotion regulation and empathy. This is a very positive outcome given that studies among children with HI without CI have repeatedly shown that these children experience difficulties concerning their social-emotional functioning (e.g., Barker et al., 2009; Theunissen et al., 2014; Wolters et al., 2011). The findings from the current study indicate that a CI benefits children's development in other areas than the auditory and language domains. However, whereas some aspects of social-emotional functioning are intact, others are impaired in children with CI. Recently, some studies conducted with children with CI have revealed impairments in emotion recognition skills (Wang et al., 2011; Wiefferink et al., 2013) and theory of mind (Ketelaar et al., 2012a; Peterson, 2004). When we combine all findings, it appears that children with CI are able to deal with their own emotions (empathy and emotion regulation), but fall behind when other people's emotions are concerned (emotion recognition and theory of mind). More research is necessary to confirm this hypothesis.

Another positive, yet surprising finding was that language skills, as reported by parents, of children with CI were not delayed. Although a CI is known to benefit children's language development, a large number of studies have shown that many children with CI still fall behind their peers with NH (Ganek et al., 2012). And indeed, our study confirmed prior research by demonstrating below-average scores on two commonly used measures which specifically assess the spoken-language skills of children with CI (i.e., Reynell and Schlichting; Van Eldik, 1998). In other words, we found a discrepancy between language as evaluated by parents and as assessed by a formal test. An explanation for this discrepancy is that parents were instructed to evaluate language skills regardless of language modality (i.e., signed or spoken language), as opposed to solely evaluating children's skills in spoken language. Additionally, the parent questionnaire assesses everyday language (i.e., 'follows simple instructions', 'uses at least ten words/signs', 'can reproduce short stories'), whereas the Reynell and Schlichting assess more narrow knowledge by asking children to point at objects which are named (i.e., 'where is the chair?'), or to name objects themselves.



In conclusion, for both groups alike, negative and especially uninvolved parenting appear to be a risk factor for emotion dysregulation. The relationship between positive parenting and children's social-emotional functioning is less straight-forward. Note that the cross-sectional nature of this study does not allow us to make firm statements about the directionality of the relationship between parenting style and social-emotional functioning of children with CI. This relationship is most likely a bidirectional one. Longitudinal studies conducted with children with NH show that parenting style is predictive of children's social-emotional functioning, but can also be predicted by children's behavior (Stack et al., 2010; Zhou et al., 2002). The outcomes of this study do not give any reason to assume that these relations might be different for parents and their children with a CI.





7



General discussion

The overall aim of this thesis was to expand our current knowledge of the social-emotional well-being of young (1- to 5-year-old) hearing-impaired (HI) children who have received a cochlear implant (CI) before the age of 3 years old. The participants were children with CI and a comparison group of children with NH from all over the Netherlands and the Dutch-speaking part of Belgium. All children were born to hearing parents and had no apparent (additional) disabilities. Various key aspects of social-emotional functioning (i.e., theory of mind, empathy, and moral emotions) were studied by means of a combination of tasks, behavior observations, and parent questionnaires. Children with CI were compared with NH children regarding their level of functioning on each individual aspect of social-emotional development. In addition, relations between various social-emotional skills were examined in each group separately. Moreover, the role of factors that potentially influenced children's social-emotional functioning was studied. These included CI-related variables such as experience with the device and age at implantation, but also language skills and parenting styles.

First, an overview of the main findings from this thesis will be provided in an effort to tie these outcomes together and paint a comprehensive picture of the strengths and weaknesses in the social-emotional functioning of children with CI. Subsequently, these findings will be compared to findings from other studies with children with CI, in order to shed some light on the influence of current trends in the field of cochlear implantation on children's development. Implications for clinical practice are discussed next. The chapter is concluded with some directions for future research.

Main Findings

HI children (without CI) are known to experience social-emotional difficulties. For example, in comparison to NH children, their Theory-of-Mind (ToM) development is delayed, they exhibit elevated levels of behavior problems, and have problems interacting with peers (e.g., Barker et al., 2009; Kouwenberg et al., 2012; Moeller & Schick, 2006; Peterson & Siegal, 2000; Theunissen et al., in press; Van Eldik et al., 2004; Woolfe et al., 2002). The picture that emerged from the various studies included in this thesis is that HI children who have been fitted with a CI were delayed on some, but not all, aspects of social-emotional functioning

compared to NH peers. Children with CI showed no delays regarding aspects of social-emotional functioning that require no or very little language, and which typically develop during infancy in NH children. Additionally, social behavior of children with CI turned out to be on a par with their NH peers. However, those aspects of social-emotional functioning that require more complex reasoning, and which are acquired through communication and socialization, were found to be impaired in children with CI. In sum, an important conclusion that can be drawn from these outcomes is that, at least to some extent, CIs appear to benefit HI children's functioning in other areas than merely the auditory and language domain.

Social-Emotional Functioning of Children with CI

Some social-emotional skills of children with CI were on a par with their NH peers, whereas other skills were less well-developed. Here, these will be discussed in more detail, starting with those aspects of social-emotional functioning that were intact, before turning to those that appear to be impaired in children with CI.

No differences were observed between children with CI and NH children regarding empathy (chapter 3). Empathy denotes the capacity to experience the other person's emotion, and is a key factor in promoting appropriate (pro) social behavior (Baron-Cohen & Wheelwright, 2004; Eisenberg & Miller, 1987; Jolliffe & Farrington, 2006). Parents filled in questionnaires about their children's empathic behavior in everyday situations, towards peers as well as adults. Children's empathic reactions were also observed in an experimental setting, where the experimenter simulated an emotion (happiness, pain/sadness, and anger) and scored the children's responses. Parents as well as experimenters reported similar levels of empathic behavior in both groups of children. Moreover, higher levels of empathy were associated with higher levels of social competence in both groups of children alike.

It should be noted that the empathic behaviors measured in this study were mostly indicative of children's underlying capacity for affective empathy. This component of empathy, reflecting the ability to vicariously experience the other person's emotion (McDonald & Messinger, 2011), is assumed to be inborn and can typically already be observed in infancy (Decety & Meyer, 2008). As the affective part of empathy appears to be hard-wired, we did not expect any delays or impairments to arise in children with CI. Whether children actually understood the emotion and its antecedents, which makes up the cognitive



component of empathy (Baron-Cohen & Wheelwright, 2004; Jolliffe & Farrington, 2006), could not be inferred from the findings of this study. However, we have reason to assume that cognitive empathy will be impaired in children with CI. A study by Wiefferink and colleagues (Wiefferink et al., 2013) on largely the same sample of children demonstrates that the capacity for understanding emotions is impaired in children with CI. Children with CI were less likely than NH children to attribute emotions correctly to a protagonist in a prototypical emotion-evoking situation. Moreover, certain components of ToM understanding are also impaired in children with CI, as will be discussed in more detail later. ToM is assumed to be crucial to the development of cognitive empathy. In fact, Blair (2005, p. 699) claims that “cognitive empathy is effectively Theory of Mind”. Together, this could be indicative of corresponding impairments in cognitive empathy in these children, but future studies should confirm this.

Like empathy, the ability to acknowledge that other people’s actions are intentional develops at an early age. This capacity for intention understanding is regarded as a component of ToM. Children with a well-developed ToM understand that people’s mental states are subjective, and that these mental states govern people’s behavior (Wellman, 1990), which is very helpful in making social situations more predictable. Toward the end of their first year of life, typically-developing children understand that other people’s pointing gestures toward an object (or event) during social interactions are an attempt to direct their attention to that object. Children will respond to this bid by alternating their gaze between the object and the person who pointed to it, thereby establishing joint attention (Tomasello et al., 2007). Around the same age, typically-developing children start to infer a person’s desired goal from their failed attempts to reach this goal (Meltzoff, 1995). Children with CI performed equally well as NH children on tasks measuring these abilities (chapter 4). Because intention understanding typically develops in infancy, observing similar levels of intention understanding in these 1-to-5-year-old children may not necessarily imply that children with CI develop intention understanding along the same timeline as NH children. It may merely mean that they have caught up by this age. Nonetheless, we also observed similar levels of intention understanding in a subsample of children under 30 months of age.

Regarding children’s social functioning, parents of children with CI in our studies reported equal levels of social competence and behavior problems compared to parents of NH children (chapters 3 and 5). Also, no differences were

found regarding the extent to which children in each group displayed negative emotions, which was taken as an index of emotion (dys)regulation (chapter 6). It should however be noted that, using largely the same sample of children with CI, Wiefferink and colleagues (2012a) did find a difference between children with CI and NH children on negative emotionality, but only as reported by parents. The study included in the current thesis combined different measures (i.e., an observation task and a parent report) to calculate an overall index for children's negative emotionality, whereas Wiefferink and colleagues examined outcomes on these measures separately.

In sum, some very positive outcomes were noted for children with CI, particularly if we take into account that prior studies consistently indicated higher levels of behavior problems and problematic social functioning for (older) HI children without CI (Barker et al., 2009; Kouwenberg et al., 2012; Theunissen et al., in press; Van Eldik et al., 2004). Notwithstanding these positive outcomes for children with CI, impairments were detected in some areas of social-emotional functioning. Revisiting ToM, children with CI had impaired understanding of two other, more complex components of ToM: desires and false beliefs. Children were asked to predict the behavior of protagonists in short stories, taking into account the protagonists' desires and false beliefs. These tasks were administered in signed or spoken language, and were accompanied by pictures. Understanding of desires and beliefs starts to develop from the age of 2 or 3 years old in typically developing children (Wellman, 1990). The outcomes signify that children with CI find it difficult to predict other people's behavior based on their mental states, particularly when these differ from their own (chapter 4). This is consistent with studies among HI children without CI (cf. Peterson & Siegal, 2000), and also corroborates previous findings among children with CI (Macaulay & Ford, 2006; Peterson, 2004). The current study extends our knowledge by demonstrating that a delay in ToM understanding is already present in children with CI under 5 years old, who have received their implant early (before age 3 years) in comparison to children included in previous studies (who were implanted up till the age of 6 years). On a positive note, our results indicate that children with CI develop an understanding of desires before they start to understand false beliefs. Paired with the finding that intention understanding does not seem to be impaired in these children, this implies a sequence of development which resembles that of the control group of NH children in our study, and of prior studies with typically developing



children (Colonnesi et al., 2008; Wellman & Liu, 2004). This is indicative of a delayed, rather than a qualitatively different development of ToM. Over time, these children will presumably catch up with their NH peers, as is the case for HI children without CI. A longitudinal study by Peterson (2009) demonstrated that the majority of HI children did not pass ToM tasks until the age of 10 years old. Whether children with CI will catch up to their NH peers at an earlier age than has been found for HI children without CI needs to be addressed in future studies.

A well-developed ToM helps children to behave appropriately during social interactions, but a strong moral sense might be just as important. It is people's moral sense which motivates them to do the right thing, and to treat others nicely. Conversely, a lack of morality is associated with antisocial and criminal behavior (Holmqvist, 2008; Mealey, 1995; Menesini & Camodeca, 2008). As with ToM, perspective-taking skills are required in order to judge your behavior through the eyes of others. If the behavior is not up to par or causes harm to someone else, shame or guilt are experienced. If the behavior exceeds expectations, pride is experienced (Barrett, 1995; Lewis et al., 1992; Tracy & Robins, 2004a). Whereas shame and guilt communicate that you feel bad about your transgression and you want to make amends in order to maintain the relationship, pride is a sign of dominance and enhances one's social status within the group (Stearns & Parrott, 2012; Tracy & Robins, 2004a). Moral emotions emerge at a later age than the basic emotions, and are assumed to develop in interaction with the social environment. In other words, at first children are explicitly and implicitly taught by their parents and others in their direct surroundings how to behave, and when they should feel and express moral emotions. Over time, these external evaluations are internalized and children will develop their own moral compass.

Given the communication and ToM difficulties that HI children generally experience, we assumed that it would be difficult for children with CI to learn about moral emotions. As expected, children with CI expressed less shame/guilt than their NH peers following staged emotion-evoking events such as failure on a mastery task or damaging another person's property. In addition, children with CI also showed less signs of pride than NH children when they succeeded on a mastery task (chapter 5). Again, this may suggest a lack of insight into other people's perspectives. Impaired expression of moral emotions, even in the presence of overt feedback on the child's performance by the experimenter,

may reflect lack of a sense of what is generally regarded as reprehensible or admirable by others. Contrary to our expectations, we did not find a negative relation between moral emotions and behavioral problems in either group. This could be explained by the fact that parents in both groups reported quite low levels of behavioral problems in their children. In addition, NH children who displayed more shame/guilt and more pride were reported to be more socially competent, whereas this relation was not found for children with CI. We could hypothesize that at this young age, impaired expression of moral emotions does not damage the social relations of children with CI yet. Parents of children with CI, who were the source of information on their children's social competence, might also be more forgiving regarding their children's social behavior than parents of NH children. Follow-up studies should examine whether moral emotions become more important for the social functioning of children with CI over time.

Underlying Factors

Deafness in itself does not cause social-emotional difficulties. Instead, we hypothesized that difficulties may arise as a result of underlying factors such as language delays or parenting issues. Moreover, a large body of studies has established that CI-related variables such as age at implantation play a substantial role in language development (e.g., Boons et al., 2012a; Ganek et al., 2012; Geers & Nicholas, 2013; Niparko et al., 2010), and we therefore aimed to assess whether this also accounted for inter-individual differences in social-emotional functioning.

A differentiated picture emerged with respect to the language skills of children with CI. On measures of spoken language understanding and production (i.e., Reynell and Schlichting), children with CI scored approximately one standard deviation below the norm of NH children (chapters 3, 5, and 6). However, we also asked parents to report on their children's language skills, and found no differences between children with CI and NH according to this source of information (chapter 6). Two explanations for these seemingly contradictory findings come to mind. The first is that the Reynell and Schlichting tests only measure spoken language skills, whereas parents reported on children's language skills regardless of language mode (i.e., spoken or signed). It could be that language understanding and production skills of children with CI are up to par when they can switch between and combine language modes, but



not solely by means of spoken language. The second explanation might be that parents are a biased source of information regarding their children's language skills. Parents are quite skilled at understanding their toddler's emerging speech, even when no one else can make sense of it. Likewise, parents of children with CI might be able to understand what their child is trying to say even though the words aren't pronounced correctly. Nonetheless, the child might fail when these spoken language skills are tested formally.

Note that while the study in chapter 6 indicates similar language skills in children with CI and NH children according to their parents, Wiefferink and colleagues (Wiefferink et al., 2012a), using the same parent questionnaire, did report delays in language understanding and production for children with CI as compared to their NH peers. A closer inspection of the data from the Wiefferink et al. study revealed that some parents did not fill in the questionnaire on children's language skills. After recalculating the mean ages per group based on complete cases only, children in the NH group turned out to be six months older on average than children in the CI group, which could explain the difference in language skills between the groups.

Language is assumed to play a major role in children's social-emotional development. The mismatch between communication modes of deaf children and their hearing parents and deaf children's language delays are thought to hamper these children's social-emotional development (Moeller, 2007; Moeller & Schick, 2006; Peterson & Siegal, 2000; Vaccari & Marschark, 1997). Yet, in our studies, language turned out not to play a major role in the social-emotional functioning of children with CI. Neither formally measured spoken language skills (Reynell and Schlichting), nor their understanding and production of everyday language (spoken or signed) as reported by parents were related to social-emotional functioning when age was controlled for (chapters 3 through 6). An explanation for the absence of relations between language and social-emotion functioning could be that the language measures employed did not sufficiently tap into specific language domains which are important for social-emotional development. It has been suggested that mental-state talk in particular is associated with emotional functioning (Adrian, Clemente, Villanueva, & Rieffe, 2005; Moeller & Schick, 2006; Ruffman et al., 2002). Important to note in this respect is that although everyday language skills (as reported by parents) were intact, emotion language skills were impaired (chapter 5). Children with CI knew and used less words (regardless of language mode)

related to emotions and mental states than NH children. This could imply that children with CI are competent language users who can handle themselves when it comes to concrete language (i.e., naming objects, telling left from right) in their preferred language mode, but that they are less competent when dealing with abstract language (i.e., mental states, emotions). Peterson and Siegal (2000) stated that parents of HI children without CI find it hard to talk to their children about abstract matters, which may be due to a lack of fluency in sign language (Vaccari & Marschark, 1997). The parents participating in our research arguably experienced similar difficulties when discussing abstract matters with their children with CI, given that the spoken language skills of these children were below average. Although this could offer an explanation for the impaired emotion vocabulary of children with CI, ill-developed emotion language skills did not account for their delays in emotional functioning. In contrast with expectations, emotion language was unrelated to (moral) emotional functioning in both groups of children. Emotion language was, however, positively related to social functioning in the CI group only (chapter 5).

Regarding parenting styles, no differences were observed between parents of children with CI and parents of NH children (chapter 6). Parents in both groups reported to predominantly practice a positive parenting style, which is characterized by responsiveness and sensitivity towards children's needs, and is associated with adaptive social-emotional functioning in NH children (Kawabata et al., 2011; Stack et al., 2010). Moreover, no differences were found between the groups on any of the parenting styles (i.e., positive, negative, and uninvolved). This stands in contrast to prior research, which indicated less positive and more negative (i.e., harsh, punitive) parenting practices among parents of HI children without CI compared to parents of NH children (Knutson et al., 2004; Meadow-Orlans, 1997; Meadow-Orlans & Spencer, 1996).

The effect of parenting styles on children's social-emotional functioning turned out to be equally strong in both groups. Parents who more frequently adopted a negative or uninvolved parenting style had children who experienced more difficulties regulating their negative emotions (i.e., anger and sadness). Positive parenting, on the other hand, was related to more empathic behaviors in children. Yet, this relation was modulated by children's capacity for language understanding. Children with better language abilities may have been more able to express their needs verbally, instead of having to resort to outbursts of sadness or anger. In turn, this could have made it easier for parents to



approach these children in a positive manner. Although we set out to measure the influence of parenting styles on children's social-emotional functioning, the data was collected cross-sectionally, which means relations could very well be the other way around, or - which is most likely - of a reciprocal nature.

In contrast to findings from language studies, age at implantation and duration of implant use (corrected for chronological age) were unrelated to most indices of social-emotional functioning, except for pride (chapter 5). Yet, by no means we want to suggest that early implantation is not important with respect to children's social-emotional development. It should be noted that the range with respect to the age at which children in our sample were implanted was small (i.e., on average at 16 months of age, and before age 3 years). Lack of sufficient variability regarding age at implantation could explain the absence of a relation with social-emotional functioning. Alternatively, we could hypothesize that there is a sensitive period during which children need to be implanted in order to gain optimal outcomes regarding social-emotional functioning. Research suggests that such a window of opportunity exists with regard to language outcomes. Children implanted under the age of 2 years are most likely to achieve age-appropriate language skills (Boons et al., 2012a; Ganek et al., 2012).

Trends in Cochlear Implantation: Past, Present and Future

The field of cochlear implantation is a dynamic one. Not only with respect to the technology involved, but also regarding aspects such as candidacy criteria, preservation of residual hearing, optimal age at implantation, and unilateral versus bilateral implantation. We should therefore also adopt a dynamic stance with respect to studying the social-emotional development of children with CI. It does not suffice to assess social-emotional skills in one cohort of children at one particular point in time. In order to get a broader perspective of the benefits of cochlear implantation for children's social-emotional development, we should also look at the past and the future.

Recently, two theses on the social-emotional development of 9-to-16-year-old HI children (with and without CI) in the Netherlands were completed (Kouwenberg, 2013; Theunissen, 2013). Zooming in on only the children with CI, these studies provide an overview of - with all due respect - the past. Hearing

losses in these children were identified late (i.e., not with the neonatal hearing screening), they were implanted late (i.e., on average at approximately 4 years of age) according to current standards, and with somewhat older technology. The overall picture that emerged from these theses is that children with CI showed less problems regarding their social-emotional functioning than their HI peers without CI. Children with CI showed equal levels of internalizing (i.e., depression, anxiety, somatic complaints) and externalizing problems (i.e., aggression, delinquency, symptoms of behavior disorders) compared to NH peers. HI children without CI showed higher levels of internalizing and externalizing behavior compared to NH children. Children with CI did experience more social difficulties than NH children. They more often felt victimized and experienced lower friendship qualities.

Past and present seem to be somewhat at odds with each other. Although the theses by Kouwenberg (2013) and Theunissen (2013) provide no information on the emotional skills of older children with CI, their studies do indicate some problems in the social domain. Studies included in the current thesis, on the other hand, showed a largely equal level of social development in children with CI or with NH. There are two plausible explanations for this finding. Improvements in technology and earlier implantation could have prevented social problems from arising in the younger sample of children with CI. Alternatively, problems may not be visible in young children with CI, but could appear later on, because social demands increase when children grow older. Only longitudinal studies could resolve this matter.

Recently, the rate of bilateral implantation has been increasing. Currently in the Netherlands, children under 5 years of age by default are implanted bilaterally, preferably receiving both implants simultaneously. To make predictions about future developments in the social-emotional functioning of children with CI, we draw from studies which have examined the benefits of bilateral versus unilateral implantation in other areas of development. Children who underwent bilateral implantation are reported to have better auditory (i.e., lateralization and localization of sounds) and speech recognition skills over children with a unilateral implant. This in turn may improve their ability for incidental learning because it improves their ability to overhear other people's conversations in noisy situations such as classrooms (cf. Boons et al., 2012b). An increase in incidental learning in social situations would provide children with CI with more opportunities to learn social-emotional skills, and could



in turn improve their social-emotional functioning. Of course, future studies comparing social-emotional skills of children who are implanted bilaterally to those of children who are implanted unilaterally should be carried out to confirm this hypothesis.

Implications for Clinical Practice

The finding that better language skills were not associated with better social-emotional functioning has important implications from a clinical perspective. Language development of children with CI in the Netherlands is carefully monitored, and these children receive extensive support in the form of speech therapy following implantation. On the basis of our results, however, we cannot assume that children with CI whose language development is progressing well, will also show a healthy social-emotional development. Therefore, professionals working with children with CI should be made aware that these children, despite having (re)gained a sense of hearing following implantation, and even in the presence of adequately developing language skills, could still present with considerable difficulties in the social-emotional domain.

In order to distribute monetary and time resources most efficiently, interventions should ideally be targeted at those children with CI who are most at risk for developing problems in their social-emotional functioning. Studies included in the theses by Kouwenberg (2013) and Theunissen (2013) have shown that children who are in special versus mainstream education, and who prefer some form of sign language over spoken language are more at risk for developing social-emotional problems. However, we should keep in mind that these children are in special education and rely on sign language for a reason. They are likely to experience difficulties which caused them to be placed in special education, and which prevent them from developing spoken language. Nonetheless, this constitutes important information and professionals should be extra vigilant when working with these children. Belonging to an ethnic minority group could also pose a risk for children's social-emotional development. Parents from minority group might experience difficulties communicating with professionals about the care for and needs of their child with CI, which could stand in the way of the rehabilitation process of these children (cf. Wiefferink, Vermeij, & Uilenburg, 2012b). This thesis has also

revealed that the way parents raise their children (i.e., their parenting style) may either promote or hamper children's social-emotional development. A negative parenting style puts children at risk for emotion dysregulation. Finally, low SES has been associated with problematic social-emotional development in typically developing children (Wadsworth & Achenbach, 2005). Growing up in a low-SES household may therefore also be a risk factor for children with CI.

Given that some impairments in the social-emotional domain were already apparent in children with CI below the age of 5 years in our studies, we could assume that these problems may only grow worse when children grow up and more sophisticated social-emotional behavior is expected of them. Providing professionals who are involved in the rehabilitation of children with CI with an easy and systematic way to monitor progress over time could make an important difference in the lives of these children. This will allow for early detection of delays and, in turn, early intervention if needed. For this reason, the data collected for this research project as well as for the research project with 9-to-16-year-old HI children described in the theses by Kouwenberg (2013) and Theunissen (2013) have been made available to professionals by means of the website www.emotieweb.nl. Professionals who sign up through this website are trained to administer the tasks and receive all materials required to assess the social-emotional functioning of an individual child in their care. Scores on the tasks and questionnaires are entered into the scoring program on the website and compared to norm scores of NH children as well as to norm scores of HI children (with CI), which were calculated based on the scores of the children who took part in these research projects.

Results from the studies in this thesis pinpoint the areas which are most likely to be problematic for individual children with CI, and at which interventions could be targeted. Impairments were predominantly found in the emotional functioning of children with CI, not in their social functioning. However, this does not preclude social difficulties from arising later on. Problems in the emotional domain may not only become more apparent over time, but these may also cross over into the social domain. In children this young, social skills are still very much in development and may currently be judged as sufficient by adults. Yet, when children grow older, interactions with peers become more important and peers are bound to be less forgiving than parents towards social behavior that deviates from the norm. As discussed previously, Kouwenberg (2013) found that older children with CI more often than NH



children experienced social difficulties. Improving social skills of children with CI by means of an intervention may prove to be quite difficult. How do you teach children to improve their social relationships? Instead, interventions could better be targeted at enhancing children's underlying emotional competence in order to keep their social skills up to par over time.

Making children aware of other people's perspectives, their drives and inner thoughts, helps them to make sense of other people's behavior, and to adjust their own behavior accordingly. Parents are to play a critical role in this respect. They may be advised to more overtly teach their children with CI social-emotional skills. Picture-book reading with extra attention for the story characters' emotions and motives behind behavior could be helpful (Adrian et al., 2005; Ruffman et al., 2002). Another approach could be for parents to explain their own behavior and emotions during daily activities with their children, or those of other people with whom the child interacts. Teaching children schemas, prototypical ways to behave in certain situations, could also improve their understanding of appropriate social behavior. For example, in the case of their child having a birthday party, parents could tell their children beforehand that they are expected to say thank you and smile when they are given a present, even when they do not particularly like the present, because it is the polite thing to do and that way they will avoid hurting the feelings of the person giving them the present.

While parents should be encouraged to be actively involved in their children's social-emotional development, they should at the same time avoid being too intrusive or directive during interactions with their children. This has been reported to occur among parents of HI children (cf. Spencer & Meadow-Orlans, 1996) and could actually hamper children's development. Instead, parents should be encouraged to allow their child to initiate interactions and to be responsive to the child's interests, thereby creating reciprocity in interactions. Several programs have been developed to help parents become responsive communication partners. One of these is the comprehensive Hanen Program. This program uses video feedback, role play and group discussion to teach parents to allow their children to take the lead in communication attempts, and to respond appropriately to these attempts. An adaptation of the Hanen Program tailored to the specific needs of HI children helped parents to be less controlling and more responsive during communication with their young children with CI (Harrigan & Nikolopoulos, 2002).

Programs like the Hanen Program are ultimately aimed at improving children's communication skills through the empowerment of parents. Nonetheless, these programs could indirectly also benefit children's social-emotional development, because parent-child communication provides a platform for learning social-emotional skills. There are, however, other programs which are specifically aimed at improving children's social-emotional skills. A well-known example is the PATHS (Promoting Alternative Thinking Strategies) Curriculum. This school-based prevention program teaches children a better understanding of emotions and how to regulate emotions, as well as problem-solving skills (Greenberg, Kusche, Cook, & Quamma, 1995). The downside to this program is that it runs at schools, which first of all means that the school has to be willing to employ this program, and second, delays in social-emotional development may have already started to develop before children first go to school.

Directions for Future Research

Several directions for future research stemmed from the current research project, and were alluded to briefly throughout the discussion. The most important ones are outlined here. First, there is a need for longitudinal studies concerning the social-emotional development of children with CI in order to, for example, make causal inferences regarding the relations between emotional and social functioning, or between parenting practices and child development. The children who participated in this research project have in fact been retested multiple times with a one-year interval, so the first steps toward this longitudinal approach have already been taken. Following the same group of children for a number of years will also provide us with the opportunity to see whether children with CI who have been implanted relatively early will eventually catch up with their NH peers regarding those aspects of emotional functioning they were currently found to fall behind on.

Second, the finding that children's language abilities were largely unrelated to their social-emotional functioning means the search is on to identify which variables are causing the delays in emotional functioning in children with CI. The actual content and process of communication between parents and their children with CI seems a likely candidate in this respect.



Future studies should employ ecologically valid ways of assessing parent-child communication in order to observe exchanges of mental-state language, but also parental sensitivity and responsiveness during interactions.

Third, because children with CI are known to often still experience language delays (cf. Ganek et al., 2012), it is difficult to disentangle social-emotional difficulties from language difficulties. Children with CI might have been prevented from showing their true potential because verbal requirements of the tasks designed to measure their social-emotional skills were too high, even though efforts were made to keep these as low as possible. Alternatively, language might be a prerequisite of social-emotional skills, meaning that the delays in emotional functioning that were found are an accurate reflection of their capacities. Some preliminary evidence that the latter is the case stems from a study with preverbal HI infants. Meristo and colleagues (2012) used eye tracking as a means to assess NH and HI infants' belief understanding. Results suggest that even in infancy, HI children have less of an understanding of the relation between beliefs and behavior than NH children. This study included a small sample of only 10 HI children, half of whom had a CI. Replication of this study with a larger sample is called for.

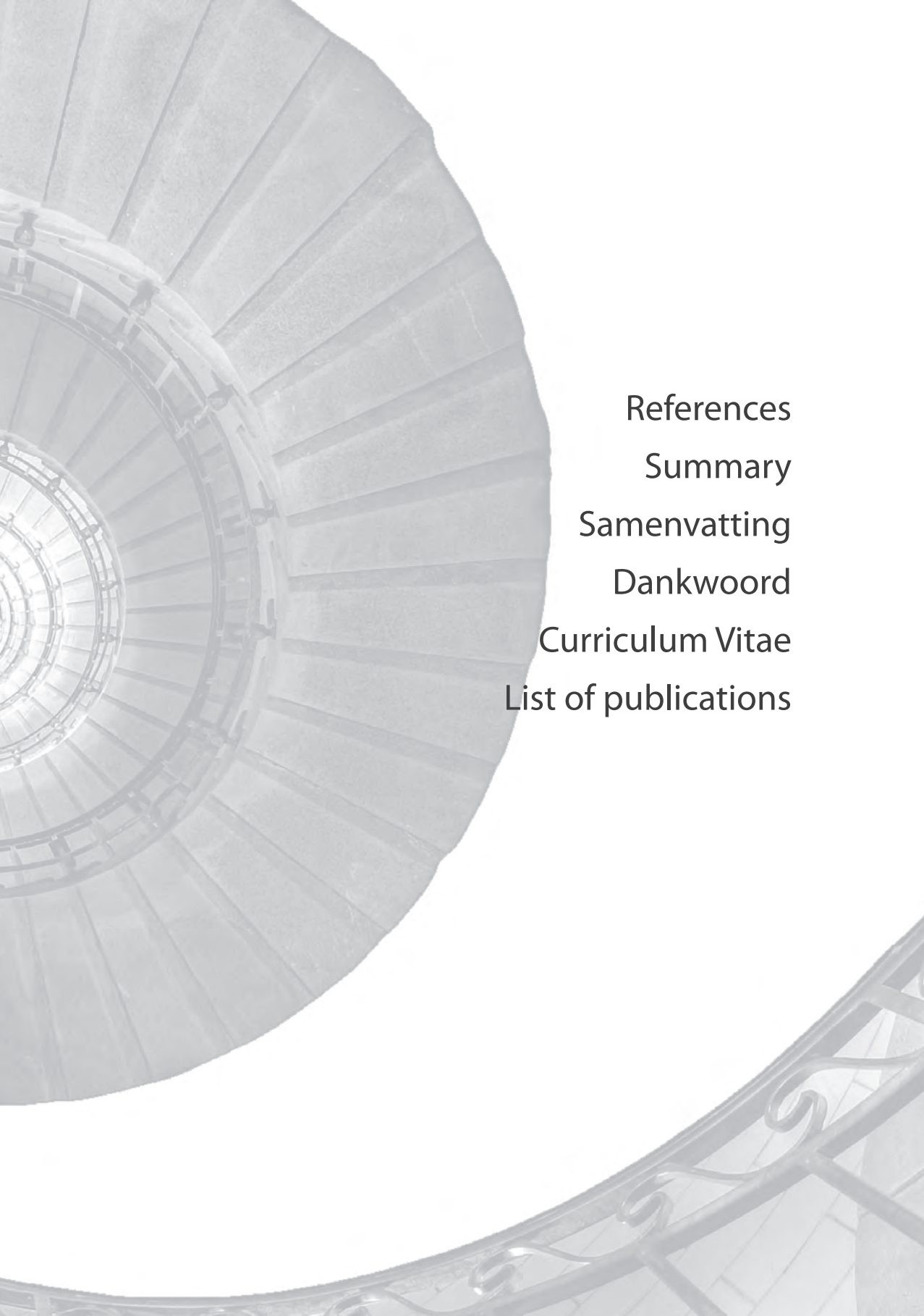
Fourth, future studies should try to identify child or family factors that increase the risk of a delayed social-emotional development in children with CI. For example, the role of SES or ethnicity needs to be examined, as well as the influence of being implanted bilaterally as opposed to unilaterally. A related and final point that needs to be addressed concerns the heterogeneity of the population of children with CI, and its dynamic nature. Although we aimed to draw conclusions about the current status quo of young children with CI in the Netherlands, there is of course no such thing as 'the average child'. In this population in particular, many children present with other difficulties besides their hearing impairment, for example an autism spectrum disorder. The extent to which children benefit from their CI also differs considerably from one child to the next. Children with additional disabilities, with deaf parents, or who were implanted beyond 3 years of age were excluded from the study. This enabled us to draw conclusions about the unique effect of a CI on children's social-development. Yet, it makes it harder to generalize these findings to those children with CI who did not fit our inclusion criteria.

The research presented throughout this thesis is a first attempt to shed light on distinct aspects of the social-emotional development of young children

with CI. Although clearly a lot remains to be examined yet, this research provides important new insights into these children's social-emotional development following cochlear implantation.







References

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Summary

Each year between 150 and 200 children in the Netherlands are born with a hearing loss (Korver, 2010). In Western countries nowadays, a large proportion of children with a severe to profound degree of hearing loss receives a cochlear implant (CI) (De Raeve & Lichtert, 2011; Hyde & Power, 2006). This device converts sounds to electrical signals which are transmitted to the auditory nerve. These signals are perceived by the brain as sounds. Completely deaf people are able to hear with a CI, although not to the extent of people without hearing loss.

Since the implementation of cochlear implantation as a remedy for hearing loss, research has been conducted to assess its outcomes. To date, most of this research was primarily focused on the consequences of cochlear implantation for sound perception and speech and language acquisition. A large number of studies has demonstrated that cochlear implantation leads to improvements in these areas, particularly for children who have been implanted before the age of 2 years old (Boons et al., 2012a; Connor et al., 2006; De Raeve, 2010; Ganek et al., 2012; Niparko et al., 2010). Up till now, not much attention has been paid to the consequences of cochlear implantation on other areas of development, such as the social-emotional development, even though it is well known that children with hearing impairments who do not have a CI experience social-emotional difficulties to a greater extent than children without hearing impairments (Barker et al., 2009; Kouwenberg et al., 2012; Theunissen et al., 2014; Van Eldik et al., 2004; Wolters et al., 2011). The main question that underlies this thesis is whether the social-emotional development of young children with a CI (aged 1 to 5 years old) is comparable to that of hearing children, and which factors influence this development.

Chapter 1 provides some background information concerning the development of children with hearing impairments and the influence of a CI on this development. Subsequently, social-emotional development is explained from a functionalist perspective. This perspective entails that emotions function to make ourselves as well as other people in our surroundings aware of our goals, desires, needs, and so on (Keltner & Haidt, 1999; Parkinson, 1996; Scherer, 2000). From this perspective it is explained that, as they grow older, children become increasingly able to recognize and regulate their own emotions, and to recognize other people's emotions and respond to these appropriately

(Denham et al., 2003; Pons et al., 2004). Next, an overview is provided of prior research, which indicates that social-emotional development of children with hearing loss is often impaired. Two factors which could affect this development are referred to: parenting style and communication. Lastly, the heterogeneity of the population of children with hearing loss is being considered, and the goals and research questions of this thesis are described.

Chapter 2 concerns the development of a parent-report questionnaire to measure empathy in young children: the EmQue. Empathy plays an important role in social interactions with other people. It helps to understand what someone else is feeling and to respond appropriately, for example by comforting or helping the other person (Decety & Jackson, 2004; Hoffman, 1987). The parent-report questionnaire was developed with the aim to assess three different levels of empathy, as described by Hoffman (1987): emotional contagion, attention to others' feelings, and prosocial behavior. The results show that this three-factor structure is present in the questionnaire. Moreover, the questionnaire turns out to be valid. In conclusion, the EmQue is a useful instrument to measure empathy in young children.

The EmQue is employed in the study described in *Chapter 3* to assess empathy in hearing children and children with CI, and to examine whether empathy is important for social competence in both groups of children. In addition, empathic reactions of children were observed in response to emotions that were simulated by the experimenter. As expected based on the literature (e.g., Eisenberg et al., 2006), empathy was associated with social competence in hearing children. This relation was also found for children with CI. In addition, the ability to acknowledge other people's emotions was also important for social competence in children with CI, whereas this was not the case for hearing children.

A very positive outcome was that children with CI did not show a delay compared to their hearing peers concerning social competence or empathy. It should be noted however, that the indices used to measure empathy in this study predominantly assessed whether children were affected by another person's emotion (affective empathy), and to a lesser extent whether they could understand the other person's emotion (cognitive empathy). This latter aspect of empathy is related to Theory of Mind (ToM) (Blair, 2005), which develops less well in children with hearing loss (Peterson & Siegal, 2000).

Comparing ToM skills in children with CI and hearing children is the focus of *Chapter 4*. A large number of studies among children with hearing loss without CI has unequivocally demonstrated that ToM is delayed in this population (Peterson, 2009; Peterson & Siegal, 2000; Russell et al., 1998; Terwogt & Rieffe, 2004; Woolfe, Want, & Siegal, 2002). Children with hearing loss are less able than hearing children to taken another person's perspective. They experience difficulties understanding that another person's behavior is motivated by that person's intentions, desires and beliefs, and that these are not necessarily in accordance with their own intentions, desires or beliefs. Findings among children with CI were less consistent and the focus of prior studies was mainly on one aspect of ToM: whether children with CI were able to understand that someone else may act on the basis of a belief that they know to be incorrect (false belief). In the study described in this chapter, the other important aspects of ToM are also being examined.

The results show that children with CI were as able as hearing children to understand other people's intentions. They were however less able to predict other people's behavior based on their desires (which were incongruent with the participants' desires), or based on their false beliefs. Even when children with insufficient language skills were excluded, a difference between children with CI and their hearing peers could still be observed. Language skills were not related to ToM skills in children with CI, whereas they were in hearing children. Language skills assessed during this study were of a general nature, while research among hearing children and children with hearing loss without CI demonstrated that particularly mental-state language is important for ToM development (Moeller & Schick, 2006; Ruffman, Slade, & Crowe, 2002). It is feasible that a imbalance exists between general language skills and mental-state language skills in children with CI.

The study in *Chapter 5* examines the extent to which moral emotions have developed in children with CI as compared to hearing children, and whether there is a relation with social functioning. Moral emotions such as guilt, shame and pride are evoked when people evaluate their own behavior against the prevailing norms and values (Eisenberg, 2000; Tangney, Stuewig, & Mashek, 2007). This ability for self evaluation implies a certain level of ToM skills, considering that one has to be able to see oneself through other people's eyes.

As expected based on the ToM delays that were observed in children with CI, these children also expressed moral emotions to a lesser degree than hearing children in response to failing or succeeding a task. Previous research among hearing children and adults established a relation between moral emotions and social functioning. Moral emotions motivate people to adhere to prevailing norms and values, whereas a lack of moral emotions is associated with antisocial behavior and even psychopathy (Barrett, 1995; Holmqvist, 2008; Mealey, 1995; Menesini & Camodeca, 2008; Olthof, 2012; Stearns & Parrott, 2012). Findings from the study described in this chapter showed that in hearing children moral emotions were related to more positive social behavior, but not to less negative behavior. In children with CI no relation between moral emotions and social functioning was found at all. At this young age, children with CI showed equal levels of social functioning as their hearing peers, even though their moral compass seemed to be less well developed.

Parents are the focus of *Chapter 6*, in which the influence of parenting style on children's social-emotional functioning is being examined. The mostly hearing parents of children with CI are confronted with hard decisions and stressful situations, such as the decision to opt for a CI, communication difficulties, and extra care and counseling for their child (Hyde, Punch, & Komesaroff, 2010; Sach & Whynes, 2005; Zaidman-Zait, 2008). This could negatively affect the way parents raise their children with CI. Research among hearing children and their parents demonstrated that a negative parenting style, in which parents are strict and not very responsive to their children's needs, can have negative consequences for children's social-emotional development. This also applies to an uninvolved parenting style, in which children are ignored and receive inconsistent responses. A positive parenting style on the other hand, in which parents support and encourage their children, is related to a better social-emotional development (Kawabata, Alink, Tseng, Van Ijzendoorn, & Crick, 2011; Newland & Crnic, 2011; Stack, Serbin, Enns, Ruttle, & Barrieau, 2010; Van Aken et al., 2007).

Prior studies among parents of children with hearing loss without CI showed that they more often employ a negative parenting style and less often a positive parenting style in comparison to parents of hearing children (Knutson, Johnson, & Sullivan, 2004; Meadow-Orlans, 1997; Meadow-Orlans & Spencer, 1996). From the study described in this chapter it became clear that this is not

the case for parents of children with CI. No differences were found concerning parenting style between parents of hearing children and parents of children with CI. In both groups of children a negative or uninvolved parenting style was related to more display of negative emotions. A positive parenting style was however not directly related to better social-emotional functioning in children. This relation seems to be mediated by children's language skills; children with better language skills display more positive behavior (in this case empathy) and might therefore be approached in a more positive way by their parents.

In the concluding *Chapter 7* the main outcomes of the studies included in this thesis are outlined to paint a picture of the strengths and weaknesses in the social-emotional development of young children with CI. Based on these studies it can be concluded that a CI does not only lead to improvements in the auditory and language domains but also, to a certain extent, in the social-emotional domain. It turns out that children with CI do not fall behind hearing children concerning their social functioning, whereas research among (older) children with hearing loss without CI does point to social difficulties (Barker et al., 2009; Kouwenberg et al., 2012; Theunissen et al., in press; Van Eldik et al., 2004). Compared to hearing children, children with CI did exhibit delays on some aspects of social-emotional development that are learned by means of communication and socialization, such as ToM and moral emotions. However, language levels did not play a major role in these delays. Implications of these outcomes for clinical practice are discussed and recommendations for future research are offered.

Samenvatting

Per jaarkomen in Nederland 150 tot 200 kinderen ter wereld met een gehoorverlies (Korver, 2010). Tegenwoordig krijgt in Westerse landen een groot deel van de kinderen met een ernstige of zeer ernstige mate van gehoorverlies een cochleair implantaat (CI) aangemeten (De Raeve & Lichtert, 2011; Hyde & Power, 2006). Dit apparaat zet geluiden om in elektrische signalen die worden doorgegeven aan de gehoorzenuw. Door de hersenen worden deze signalen waargenomen als geluid. Met behulp van een CI kunnen ook volledig dove mensen horen, hoewel niet op hetzelfde niveau als mensen zonder gehoorverlies.

Sinds cochleaire implantatie wordt toegepast als remedie voor gehoorverlies, wordt er ook onderzoek gedaan naar de uitkomsten ervan. Dit onderzoek heeft zich tot nu toe voornamelijk toegespitst op de gevolgen van cochleaire implantatie voor het kunnen waarnemen van geluid en voor de spraak- en taalontwikkeling. Een groot aantal studies heeft aangetoond dat cochleaire implantatie tot verbetering leidt op deze gebieden, voornamelijk voor kinderen die onder de leeftijd van 2 jaar zijn geïmplantieerd (Boons et al., 2012a; Connor et al., 2006; De Raeve, 2010; Ganek et al., 2012; Niparko et al., 2010). Er is tot op heden echter nog maar weinig aandacht besteed aan de gevolgen van cochleaire implantatie op andere gebieden van de ontwikkeling, zoals de sociaal-emotionele ontwikkeling. Dit terwijl het bekend is dat kinderen met een gehoorverlies zonder CI in grotere mate problemen ervaren op sociaal-emotioneel gebied dan normaal horende kinderen (Barker et al., 2009; Kouwenberg et al., 2012; Theunissen et al., 2014; Van Eldik et al., 2004; Wolters et al., 2011). De vraag die in dit proefschrift centraal staat, is of de sociaal-emotionele ontwikkeling van jonge kinderen met een CI (1 tot 5 jaar oud) vergelijkbaar is met de ontwikkeling van normaal horende kinderen en welke factoren van invloed zijn op deze ontwikkeling.

In *Hoofdstuk 1* wordt een achtergrond geschetst van de ontwikkeling van kinderen met een gehoorverlies en wordt de invloed van het CI op deze ontwikkeling beschreven. Vervolgens wordt de sociaal-emotionele ontwikkeling uitgelegd vanuit het functionalistisch perspectief. Dit perspectief stelt dat emoties een duidelijke functie hebben om zowel onszelf als anderen in onze omgeving bewust te maken van onze doelen, wensen, behoeftes, enzovoorts (Keltner & Haidt, 1999; Parkinson, 1996; Scherer, 2000). Vanuit dit perspectief

wordt uitgelegd dat kinderen als ze ouder worden steeds beter in staat zijn om hun eigen emoties te herkennen en te reguleren, maar ook om emoties van anderen te herkennen en hier gepast op te reageren (Denham et al., 2003; Pons et al., 2004). Vervolgens wordt aan de hand van eerder onderzoek beschreven dat de sociaal-emotionele ontwikkeling van kinderen met een gehoorverlies vaak minder goed verloopt en worden twee factoren benoemd die hier mogelijk aan ten grondslag liggen: opvoedstijl en communicatie. Tot slot wordt de heterogeniteit van de populatie kinderen met een gehoorverlies belicht en worden de doelen en onderzoeksvragen die centraal staan in dit proefschrift besproken.

In *Hoofdstuk 2* wordt de ontwikkeling van een oudervragenlijst voor het meten van empathie bij jonge kinderen besproken: de EmQue. Empathie speelt een belangrijke rol in de sociale interactie met andere mensen. Het zorgt ervoor dat je begrijpt wat een ander voelt en hier gepast op kunt reageren, bijvoorbeeld om de ander te troosten of te helpen (Decety & Jackson, 2004; Hoffman, 1987). De oudervragenlijst is ontwikkeld met het doel om drie verschillende niveaus van empathie te meten, zoals beschreven door Hoffman (1987): emotionele besmetting, aandacht voor andermans gevoelens en prosociaal gedrag. Uit de resultaten blijkt dat deze driefactor structuur inderdaad aanwezig is in de vragenlijst. Bovendien is aangetoond dat de vragenlijst valide is. Concluderend kan gesteld worden dat de EmQue een bruikbaar instrument is om empathie bij jonge kinderen in kaart te brengen.

De EmQue wordt in de studie die beschreven staat in *Hoofdstuk 3* gebruikt om empathie bij horende kinderen en kinderen met een CI te meten en te onderzoeken of empathie bij beide groepen belangrijk is voor sociale competentie. Daarnaast werden ook de empathische reacties van kinderen op een door de testleider gesimuleerde emotie geobserveerd. Zoals verwacht op basis van de literatuur (o.a. Eisenberg et al., 2006) bleek empathie bij horende kinderen geassocieerd te zijn met sociale competentie. Deze relatie werd ook gevonden bij kinderen met een CI. Bij kinderen met een CI bleek daarnaast het vermogen om andermans emoties te onderkennen van belang te zijn voor de sociale competentie, terwijl dit bij horende kinderen niet het geval was.

Een zeer positieve uitkomst was dat kinderen met een CI geen achterstand in sociale competentie of in empathie lieten zien ten opzichte van hun horende leeftijdsgenoten. Hierbij moet wel opgemerkt worden dat de in deze studie gebruikte indices voor empathie voornamelijk in kaart brachten of

kinderen konden meevoelen met een ander (affectieve empathie) en in mindere mate of zij de emotie van de ander ook begrepen (cognitieve empathie). Dit laatste aspect van empathie is gerelateerd aan Theory of Mind (ToM) (Blair, 2005) , waarvan bekend is dat dit minder goed ontwikkeld is bij kinderen met een gehoorverlies (Peterson & Siegal, 2000).

Het vergelijken van de ToM vaardigheden van kinderen met een CI en horende kinderen is de focus van **Hoofdstuk 4**. Een grote hoeveelheid studies bij kinderen met een gehoorverlies zonder CI heeft onomstotelijk aangetoond dat ToM vertraagd ontwikkelt bij deze populatie (Peterson, 2009; Peterson & Siegal, 2000; Russell et al., 1998; Terwogt & Rieffe, 2004; Woolfe et al., 2002). Kinderen met een gehoorverlies zijn minder goed in staat dan horende kinderen om zich te verplaatsen in de gedachtewereld van een ander. Zij hebben er moeite mee om te begrijpen dat het gedrag van een ander gestuurd wordt door diens intenties, verlangens en overtuigingen, en dat die niet per se overeen hoeven te komen met die van henzelf. Wat betreft de populatie kinderen met een CI waren de uitkomsten minder consistent en lag de focus van eerdere studies voornamelijk op één aspect van ToM, namelijk of kinderen met een CI konden begrijpen dat iemand anders kan handelen op basis van een overtuiging waarvan zij weten dat die niet klopt (false belief). In de studie die in dit hoofdstuk besproken wordt, worden ook de andere belangrijke aspecten van ToM onderzocht.

Uit de resultaten blijkt dat kinderen met een CI in dezelfde mate als horende kinderen in staat waren om de intenties van een ander te begrijpen. Zij waren echter minder goed in staat om het gedrag van een ander te voorspellen op basis van diens (niet met de proefpersoon overeenkomende) wensen, of op basis van diens false belief. Ook wanneer kinderen met onvoldoende taalvaardigheden werden uitgesloten, bleef er een verschil bestaan tussen kinderen met een CI en hun horende leeftijdsgenoten. Taalvaardigheden waren ook niet gerelateerd aan ToM vaardigheden bij kinderen met een CI, terwijl dit bij horende kinderen wel het geval was. De taalvaardigheden die gemeten werden in deze studie waren echter van algemene aard, terwijl onderzoek bij horende kinderen en bij kinderen met gehoorverlies zonder CI heeft aangetoond dat vooral zogenaamde mental state taal, taal die betrekking heeft op mentale toestanden, van belang is voor de ToM ontwikkeling (Moeller & Schick, 2006; Ruffman et al., 2002). Het is mogelijk dat er bij kinderen met CI een disbalans is tussen algemene taalvaardigheden en mental state taalvaardigheden.

In *Hoofdstuk 5* wordt onderzocht in hoeverre morele emoties ontwikkeld zijn bij kinderen met een CI in vergelijking met horende kinderen en of er een relatie is met sociaal functioneren. Morele emoties zoals schuld, schaamte en trots ontstaan wanneer individuen hun eigen gedrag evalueren ten opzichte van de geldende normen en waarden (Eisenberg, 2000; Tangney et al., 2007). Dit vermogen tot zelfevaluatie impliceert een zekere mate van ToM vaardigheden, immers men moet zichzelf kunnen zien door de ogen van een ander.

Zoals verwacht op basis van de gevonden ToM achterstanden bij kinderen met een CI, bleken deze kinderen ook in mindere mate dan horende kinderen morele emoties te tonen in reactie op het falen of slagen op een taak. Eerder onderzoek bij horende kinderen en volwassenen heeft aangetoond dat er een verband is tussen morele emoties en sociaal functioneren (Barrett, 1995; Holmqvist, 2008; Mealey, 1995; Menesini & Camodeca, 2008; Olthof, 2012; Stearns & Parrott, 2012). Morele emoties zorgen ervoor dat men zich houdt aan de geldende normen en waarden, terwijl een gebrek aan morele emoties juist gerelateerd is aan antisociaal gedrag en zelfs psychopathie. Uit de in dit hoofdstuk beschreven studie bleek echter dat morele emoties bij horende kinderen wel gerelateerd waren aan meer positief sociaal gedrag maar niet aan minder negatief gedrag. Bij kinderen met een CI werd zelfs helemaal geen verband tussen morele emoties en sociaal functioneren gevonden. Kinderen met een CI toonden op deze jonge leeftijd eenzelfde niveau van sociaal functioneren als hun horende leeftijdsgenoten, ondanks een ogenschijnlijk minder goed ontwikkeld moreel kompas.

In *Hoofdstuk 6* staan de ouders centraal en wordt onderzocht wat de invloed van opvoedstijl is op het sociaal-emotionele functioneren van kinderen. De veelal horende ouders van kinderen met een CI worden geconfronteerd met moeilijke beslissingen en stressvolle situaties, zoals de keuze voor een CI, communicatiemoeilijkheden en extra zorg en begeleiding voor hun kind (Hyde et al., 2010; Sach & Whynes, 2005; Zaidman-Zait, 2008). Dit zou negatieve gevolgen kunnen hebben voor de manier waarop ouders hun kinderen met een CI opvoeden. Uit onderzoek bij horende kinderen en hun ouders is gebleken dat een negatieve opvoedstijl, waarbij ouders streng en weinig responsief zijn wat betreft de behoeftes van hun kind, negatieve consequenties kan hebben voor de sociaal-emotionele ontwikkeling van kinderen. Dit geldt ook voor een onverschillige opvoedstijl, waarbij het kind genegeerd wordt en inconsistente

reacties krijgt. Een positieve opvoedstijl daarentegen, waarbij ouders hun kinderen steunen en stimuleren, is juist gerelateerd aan een betere sociaal-emotionele ontwikkeling (Kawabata et al., 2011; Newland & Crnic, 2011; Stack et al., 2010; Van Aken et al., 2007).

Onderzoek bij ouders van kinderen met een gehoorverlies zonder CI heeft aangetoond dat zij vaker een negatieve en minder vaak een positieve opvoedstijl hanteren dan ouders van horende kinderen (Knutson et al., 2004; Meadow-Orlans, 1997; Meadow-Orlans & Spencer, 1996). Uit de resultaten van de in dit hoofdstuk beschreven studie bleek echter dat dit niet het geval is bij ouders van kinderen met een CI. Er waren geen verschillen in opvoedstijl tussen ouders van horende kinderen en van kinderen met een CI. Voor beide groepen kinderen bleek dat een negatieve of onverschillige opvoedstijl gerelateerd was aan meer vertoon van negatieve emoties. Een positieve opvoedstijl was echter niet direct gerelateerd aan het sociaal-emotioneel functioneren van kinderen. Deze relatie lijkt te worden gemedieerd door het taalvermogen van kinderen; kinderen met betere taalvaardigheden vertonen meer positief gedrag (in dit geval empathie) en worden daardoor wellicht ook positiever benaderd door hun ouders.

In het afsluitende *Hoofdstuk 7* worden de belangrijkste uitkomsten van de in dit proefschrift opgenomen studies op een rijtje gezet om een beeld te schetsen van de sterke en zwakke kanten in de sociaal-emotionele ontwikkeling van jonge kinderen met een CI. Op basis van deze studies kan de conclusie getrokken worden dat een CI niet alleen op het gebied van auditieve en taalvermogens een verbetering teweeg brengt maar ook, tot op zekere hoogte, op sociaal-emotioneel gebied. Kinderen met een CI blijken op sociaal vlak niet achter te lopen op horende kinderen, terwijl onderzoek bij (weliswaar oudere) kinderen met een gehoorverlies zonder CI wel wijst op sociale moeilijkheden (Barker et al., 2009; Kouwenberg et al., 2012; Theunissen et al., in press; Van Eldik et al., 2004). Kinderen met een CI vertoonden wel een achterstand ten opzichte van horende kinderen op een aantal aspecten in de sociaal-emotionele ontwikkeling die aangeleerd worden door communicatie en socialisatie, zoals ToM en morele emoties. Het taalniveau van kinderen met een CI bleek hier echter geen grote rol in te spelen. Implicaties van deze uitkomsten voor de praktijk worden besproken en er worden aanbevelingen gedaan voor toekomstig onderzoek.

Dankwoord

Terugkijkend op de afgelopen jaren ben ik erg blij dat ik de sprong gewaagd heb toen Professor Carolien Rieffe me benaderde of ik mee wilde werken aan een nieuw onderzoek. Dit onderzoek bleek te gaan over de sociaal-emotionele ontwikkeling van jonge dove kinderen met een cochleair implantaat en heeft uiteindelijk geleid tot dit proefschrift.

Allereerst gaat dan ook mijn dank uit naar Carolien voor de geboden kans en voor alles wat ze sinds dat eerste aanbod in me geïnvesteerd heeft. Jouw enthousiasme en inzicht zijn een inspiratie voor me. Ik waardeer onze samenwerking erg en kijk ernaar uit om ook de komende jaren veel mooie nieuwe onderzoeken met je op te zetten.

Ook mijn andere promotor, Professor Johan Frijns, wil ik graag bedanken voor zijn altijd scherpe blik en waardevolle adviezen tijdens dit project en bij de vele versies van manuscripten die hij onder ogen heeft gehad. De samenwerking met de afdeling KNO van het LUMC is altijd bijzonder prettig en waardevol gebleken. Met name dank ik Anke Otten-Koens voor haar betrokkenheid bij het testen van de kinderen met CI en het ontwikkelen van het testmateriaal.

Ik wil hier ook graag mijn co-promotor Dr. Karin Wiefferink bedanken voor de fijne samenwerking door de jaren heen. Je stond altijd klaar om mee te denken of iets voor me door te lezen. Ook de NSDSK is een hele fijne partner geweest om mee samen te werken aan dit onderzoek. Ik dank met name Bernadette Vermeij voor het testen van de kinderen met CI en voor haar hulp bij het ontwikkelen van nieuw testmateriaal.

Alhoewel dit proefschrift draait om kinderen met een cochleair implantaat, is ook Centrum Autisme betrokken geweest bij dit onderzoek. Ik dank vooral Lex Stockmann en Annette van Zijp. Ook dank ik Sigrid Kok voor haar hulp bij de dataverzameling en haar waardevolle input bij het ontwikkelen van het testmateriaal.

Verder gaat mijn grote dank uit naar de ouders en kinderen die hebben meegewerkt aan het onderzoek en de organisaties die het mogelijk hebben gemaakt om hen te benaderen en die veelal de faciliteiten hebben geboden om de kinderen te testen.

In de afgelopen jaren heb ik met vele fijne collega's mogen samenwerken bij de afdeling Ontwikkelingspsychologie van de Universiteit Leiden. Maartje,

Lucinda, Stephanie, Sigrid, Cédric, Neeltje, Evelien, Anouk, ik ben blij dat ik jullie heb leren kennen.

De achterban heeft het tijdens mijn promotieonderzoek soms zonder mij moeten stellen. Ik beloof dat ik de komende tijd wat aan mijn eigen sociaal functioneren ga doen. Jary en Ingrid, dank dat jullie er altijd voor me zijn. Ook ben ik dankbaar voor mijn lieve broer en schoonzus, en fantastische neefje. Tot slot: papa en mama, voor jullie nooit aflatende steun en liefde.

Curriculum Vitae

Lizet Ketelaar (1980) was born in Alphen aan den Rijn. She graduated from the Groene Hart Lyceum in Alphen aan den Rijn in 1998. Lizet obtained her Master's degree in Developmental Psychology from Leiden University in 2004. After obtaining her Master's degree, she was offered a teaching job at the Developmental Psychology unit of Leiden University, facilitating workgroups for several courses and supervising Bachelor and Master theses. In 2007, Lizet started working with Prof.dr. Carolien Rieffe on a new research project concerning the social-emotional development of children with cochlear implants. In 2008, she started as a PhD student on this project, under supervision of Prof.dr. Carolien Rieffe and Prof.dr.ir. Johan Frijns. Lizet continues to work as a teacher and researcher at the Developmental Psychology unit of Leiden University.

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