ABSTRACT

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

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

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

Conclusion Children with moderate hearing loss have more difficulty in their ToM reasoning than hearing peers, despite the fact that their language abilities lie within the average range compared to test normative samples.
Engagement in social interactions is essential for the social-emotional development of children. In order to induce and maintain relationships, children need to learn that different people have different intentions, desires, and beliefs. The ability to apply such mental states to others is known as ‘Theory of Mind’ (ToM). Through ToM development, children will start to understand that our mental states explain our actions (e.g., dad chooses coffee for dessert because he prefers coffee over ice-cream). ToM development has been studied extensively over the last two decades (Wellman 1990; Dunn 1996). These studies revealed that both language and communicative abilities are very important for an adequate ToM development (see (Stanzione & Schick 2014) for a review). The importance of this relation has been illustrated previously by many studies in deaf children of hearing parents. Outcomes show severe delays in the ToM development of deaf children of hearing parents (Peterson & Siegal 1999; Courtin 2000; De Villiers & De Villiers 2000; Schick et al. 2007) that may continue to be problematic during adolescence (Pyers & Senghas 2009; Wellman et al. 2011). One explanation offered in the literature for these findings lies in the reduced abilities of parents (especially hearing parents who sign) to discuss abstract concepts such as thoughts and emotions compared to hearing-haring dyads (Moeller & Schick 2006). Children with moderate hearing loss (MHL) share the same mode of communication as their hearing parents. However, these children often still encounter language difficulties (Moeller et al. 2007; Moeller et al. 2015; Tomblin et al. 2015). Therefore, children with MHL are also potentially at risk for inadequate ToM development. Nevertheless, until now, no research has focused on the development of ToM in children with moderate hearing loss, which is the aim of this study.

Children with moderate hearing loss

A substantial number of children have hearing loss thresholds falling in the moderate range (40-70 dB HL). When wearing their hearing aids, children with MHL can function reasonably well in quiet areas and in one-on-one conversations. They can hear what is said when they are not disturbed by background noise that interferes with their hearing aids, their ability to recognize consonants, and directional hearing (Eisenberg 2007; McCreery et al. 2015a). However, the hearing capacities of these children are frequently overestimated. Children with MHL frequently encounter difficulties in fully understanding what is said in daily interactions, especially in noisy environments such as daycare centers and classrooms (Finitzo-Hieber & Tillman 1978). Children with hearing loss encounter difficulties in speech perception when listening to speech in noise (Yang et al. 2012). Furthermore, the children’s hearing aids (HAs) are often not fit optimally, which may negatively impact their hearing potential (McCreery et al. 2015; Tomblin et al. 2015). For the child’s surroundings, it is often difficult to understand what a child with MHL does hear and what input is missed. Diminished access to social conversations could potentially diminish their opportunities for social learning, which has ongoing consequences for their social-emotional development.
In studies on ToM development, the majority of research has focused on only one aspect of ToM development, that is, the understanding of (false) beliefs. Yet, Wellman and others emphasize on the importance of studying ToM in its broadest sense. Thereby, it is important to be aware of the fact that the acknowledgement of others’ intentions and desires precedes the understanding of others’ (false) beliefs (Wellman 2002). This was previously demonstrated in large studies examining the developmental sequence of ToM development in deaf children and children with an autism spectrum disorder. These studies show that deaf children generally show the same sequential pattern of ToM development as hearing peers, albeit slower (Peterson et al. 2005; Peterson & Wellman 2009). This delayed ToM development can have ongoing consequences for a child’s social development (Olson et al. 2011; Caputi et al. 2012).

Intention understanding
An essential precursor for the development of ToM is the ability to acknowledge others’ intentions (Sodian & Kristen-Antonow 2015). Growing consciousness of the fact that others’ actions are guided by their intentions teaches children to separate human beings from objects. Only by knowing someone else’s intentions, one can understand the person’s actions. To illustrate, the physical movement of an object from one person to the other can be interpreted as giving, sharing, loaning, returning, or trading something. Yet, without intention understanding, we do not know why actions happen. In typically developing children, intention understanding begins to emerge in the second year of life (Tomasello et al. 2005).

An important aspect of intention understanding is joint attention; the ability to share attention with someone else concerning an object or situation. Drawing someone’s attention to a certain situation increases language development and strengthens relationships. Studies in young children show equal levels of joint attention in deaf children with CI compared to age-related peers, whereas less engagement in joint attention was seen in deaf children without a CI (Tasker et al. 2010; Ketelaar et al. 2012; Cejas et al. 2014).

Desire understanding
The next important step in ToM development is the ability to acknowledge others’ desires and to be able to distinguish between one’s own and the other’s desires. Desire understanding gradually takes place after a child’s third birthday (Wellman et al. 2000). Abstract concepts such as taste allow children to understand the subjectivity of desires. For example, a child needs to learn to understand that dad does not like to eat cheese whereas the child herself really likes a cheese sandwich. Research on desire understanding in deaf children can be extracted from the work by Peterson (Peterson 2004; Peterson et al. 2005; Peterson & Wellman 2009; Wellman & Peterson 2013) and Remmel (Peters et al. 2009; Remmel & Peters 2009) who found no difference in desire understanding when comparing school-aged deaf children (with and without CI) to hearing preschoolers. Only one study compared preschoolers with CI to age-related hearing peers. When focusing on children with sufficient language comprehension, children with CI were able to appreciate the protagonist’s desire when it matched their own desire. Yet, they were outperformed by their hearing peers when the protagonist in the vignette had a dissimilar desire (Ketelaar et al. 2012).
Belief understanding

Classic false belief tasks include the change-of-location and the unexpected-content task. In both tasks, the child is questioned about the behavior of a story character. In the story, this character holds a belief that opposes the actual truth. Around the age of four, children start to appreciate other’s beliefs. Research shows equal levels of belief understanding in deaf children born to deaf parents compared to hearing peers born to hearing parents (see Stanzione and Schick 2014 for an overview) (Schick et al. 2007; Stanzione & Schick 2014). However, deaf children of hearing parents performed lower on false belief understanding than hearing children, with so-called late signers showing the least favorable results (Courtin 2000; Peterson et al. 2005). This difference can be explained by the quality and quantity of communication. Both deaf children who acquire oral communication and deaf children who acquire sign language relatively late (because it is their second language) may encounter limited participation in high-quality social interactions involving mental state talk, be it in school or with their family at home (Jeanes et al. 2000; Macaulay & Ford 2006; Ziv et al. 2013).

A limited number of studies on false belief understanding in deaf and hard of hearing children compared to hearing controls found no differences in ToM abilities. However, in these studies children were much older than the control group, making the groups difficult to compare (Peterson et al. 2005; Peters et al. 2009; Peterson & Wellman 2009; Remmel & Peters 2009; Levrez et al. 2012). Since the introduction of early identification of hearing loss and early cochlear implantation, results have changed. Because of early implantation, young children with CI had relatively better language skills. These improved language skills enabled them to join in conversations more often which could potentially stimulate their ToM skills. Consequently, studies started to compare children with CI to age-related peers. Yet, these studies in young children still found lower levels of belief understanding in preschoolers with CI as compared to hearing peers (Ketelaar et al. 2012; Sundqvist et al. 2014).

ToM and language

The relation between ToM and language abilities has been studied extensively. A meta-analysis examining this relation reported a strong relation between the two indices (Milligan et al. 2007). Since there has been an ongoing debate regarding the direction of causality between language and ToM development, this was one of the aims of this meta-analysis. Even though a bidirectional relationship was found in longitudinal studies (i.e., early language predicted later ToM development and early ToM skills predicted later language development), the relation reporting early language skills to be beneficial for later ToM development was significantly stronger than vice versa. However, this review only included studies that examined this relation in typically developing children.

In DHH children, the relation between language and ToM skills seems complex. False belief tasks for instance contain ‘mental state verbs’ and ‘if/then statements’. In order to understand such complex ToM tasks, a certain level of language and communication skills is needed to succeed. As a result it is often unclear what it is exactly that such tasks are measuring: the child’s ToM skills or their language capacities. Schick et al. therefore used
ToM tasks that required minimal language skills to measure ToM abilities in deaf children of hearing parents. Results showed that the deaf children in their study also performed lower on the low-verbal tasks compared to hearing children and deaf children of deaf parents, indicating the importance of access to communication with others. This statement was underlined by the fact that complement processing skills were found to predict performance on low-verbal ToM tasks, yet vocabulary comprehension skills did not (Schick et al. 2007).

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

**Present study**

The first aim of this study was to examine ToM abilities and its precursors in children with MHL compared to hearing children. Although children with MHL and their hearing caregivers share the same mode of communication (i.e. spoken language), it is also known that parents of children with hearing loss use less mental state talk in their conversations with their child (Ambrose et al. 2015). Additionally, due to various reasons children with MHL often still encounter (mild) language and communication problems (Tomblin et al. 2015). These difficulties could prevent them from fully benefiting from social interaction and incidental learning about others’ intentions, desires, and beliefs. We therefore hypothesized that children with MHL of hearing parents would have lower ToM skills than hearing children. The second aim of this study was to define the relation between language skills and the development of ToM in children with MHL and in hearing controls separately. We expected language skills to be positively related to both desire and belief understanding because a certain level of language is needed to develop these skills. We expected no difference in the strength of this relation between the two groups. The third aim of this study was to evaluate the developmental sequence of various ToM concepts both in children with and without MHL. Because of language difficulties, we expected a delayed but not qualitatively different development of ToM in children with MHL compared to peers with normal hearing.

**METHODS**

**Procedure**

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

For participation in the social-emotional assessments of the DECIBEL-study, children needed to fulfill additional inclusion criteria. Children needed to be at least 36 months old, their unaided hearing loss in the better ear should not exceed 70 dB HL, children had to use conventional hearing aids or bone conduction devices (BCD) and it was requested that their preferred mode of communication was either spoken, or sign-supported Dutch. This resulted in 74 children who were eligible for participation, and their parents were invited to participate. Finally, parents of 44 children gave informed consent (response rate 59.5%).

Children were visited at home. A researcher sat with the child in a quiet room and conducted several tasks which will be explained in more detail below. The session was video-recorded in order to score the child’s behavior afterwards. The camera was positioned so that both the experimenter and the child were recorded. This allowed for both observation of the child’s behavior as well as to check if all tasks were correctly performed by the experimenter. The sequence of the tasks and observations that were performed was as follows: 1) Intention understanding, 2) False belief task, 3) Similar desire task, 4) Imperative pointing, 5) Dissimilar desire task, 6) other tasks and observations not mentioned in this study, 7) Dissimilar desire task, 8) Declarative pointing, 9) other tasks and observations not mentioned in this study, 10) Similar desire task. Completion of the whole set of tasks and observations took approximately 35 to 45 minutes per child.

Parents were requested to complete several questionnaires to gain background information. Medical history and language scores were derived from the child’s medical files. A control group of hearing children was collected as part of another nationwide study. These children were previously described by Ketelaar et al. and were recruited from all over the Netherlands through mainstream primary schools and daycare centers (Ketelaar et al. 2012). From this large control sample, we were able to compose a subsample of 101 hearing children with a comparable age and sex distribution. Parents of children in the control group reported no history of hearing loss in their child.

Participants

All children were between 40 and 70 months old during home observations (mean age 57 months). Of the 44 children with MHL, 27 were boys (61.4%). Their hearing loss varied with a pure-tone-average between 35 and 70 dB HL in the better ear (mean loss 50 dB HL). Residual hearing was calculated by averaging unaided hearing thresholds at 500, 1,000 and 2,000 Hz. Six children had a hearing loss between 35 and 40 dB. One child used a BCD, all others used hearing aids. All but one were aided bilaterally. All children understood spoken language, yet five of them (11.4%) preferred to use sign-supported Dutch. Parents of seven children with MHL reported having hearing loss themselves. Three children with MHL had an additional handicap. One child was diagnosed with Turner syndrome, one child suffered...
from muscle-tone dysregulation and the third child had a mild hypotonic hemiparesis. These three children did not differ from the rest of the MHL group in age, language skills, or on any of the ToM tasks. The control group consisted of 101 children with normal hearing, 55 were boys (54.5%). Demographic characteristics of both groups are listed in Table 1.

Materials

Intention understanding

Comprehension of other people’s intentions was measured using three tasks. The ‘Intention Understanding task’ based on the design of Meltzoff (Meltzoff 1995) and adapted by Ketelaar et al. (Ketelaar et al. 2012) was used to define whether children understand others’ intentions when trying to achieve a certain goal, even if the person is unable to succeed. To illustrate this, in one of these tasks the researcher attempts to put a string of beads in a cup. After failing to get the string in the cup, she hands it over to the child. Children succeed if they put the string of beads in the cup. With each task (trying to stack two cups and fitting a tube in a slightly bigger one) the researcher makes three attempts before handing the task to the child. This results in a maximum score of three if all intentions are understood correctly.

The ‘Declarative Comprehension task’ measures joint attention (Colonnesi et al. 2008; Ketelaar et al. 2012). During this task, the researcher acts surprised and points to an object out of sight of the child. The researcher then looks back and forth between the object and the child. The subsequent behavior of the child was observed and children could receive up to three points when they looked at the object, looked at the researcher and, attempted to communicate about the object.

The third task to measure intention understanding was the ‘Imperative Comprehension task’ (Colonnesi et al. 2008; Ketelaar et al. 2012). This task starts with the researcher pointing towards an object that is within reach of the child but not of the researcher. After pointing towards the object, the researcher holds up her hand with the palm facing up to request the object. The child succeeds if he or she actively responds to this gesture either by handing over the object or refusing to do so (e.g., saying no, shaking his/her head). Three points were awarded if the child succeeded the first time. If not, up to two additional attempts were performed between the other tasks and the score decreased by one point each time until a score of zero was attained after three unsuccessful attempts.

Desire understanding

The acknowledgement of others’ desires was assessed using the ‘desire task’ (Ketelaar et al. 2012). This task uses vignettes to measures two types of desires: similar and dissimilar desires. In the similar desire condition, the child is presented with a picture showing two types of food (e.g., tomato and ice-cream). The child is asked what he or she prefers to eat. The researcher then tells a story about a boy who also likes the food that the child just chose. Then the child is asked: “Now the boy can choose a snack. What will the boy choose to eat?” This question is followed by two control questions: “Does the boy like [Snack 1]?” and “Does the boy like [Snack 2]?” The child is awarded one point if he or she answers all three
questions correctly. In the dissimilar desire task, the only difference is that the protagonist in
the story does not like the snack that the child preferred but instead likes the opposite snack.

Belief understanding
Belief understanding was measured using an adapted version of the traditional false-belief
Sally-Anne task by Baron-Cohen (Baron-Cohen et al. 1985; Ketelaar et al. 2012). In this task,
the child sees a drawing of a boy playing with his model airplane. The boy hides his plane
and leaves the scene. When the boy is away, a girl grabs the plane and hides it in a different
location. On the next drawing the boy returns and the child is asked: “Where will the boy
look for his plane?” This question is followed by two additional questions to check
comprehension: “Where did the boy hide his plane before he went away?” and “Where is the
plane now?”. One point was awarded only if the child was able to answer all three questions
correctly. All tasks mentioned above have previously been used in different clinical groups
with good reliability (Ketelaar et al. 2012; Broekhof et al. 2015).

Language
In order not to interfere with the regular evaluations of the child’s speech- and language
therapists, test scores were derived from the child’s medical files. Therefore, language scores
were not available from the hearing children. Receptive language abilities were assessed with
the verbal comprehension scale of the Dutch version of the Reynell Developmental Language
Scale (RLDS) (Van Eldik 1998). The word development and sentence development scales of
the Dutch version of the Schlichting Expressive Language Test (SELT) were used to assess
expressive language abilities. These language tests are used throughout The Netherlands to
assess language development, especially in high-risk groups. Raw scores are standardized
according to age using quotients in which the population mean in hearing children is 100 with
a standard deviation of 15. Language quotients within one standard deviation from the mean
are considered to be in the normal range (85-115).

Parent-reported language skills
Two scales of the Child Development Inventory (CDI) were used to assess language skills in
all participants (Ireton & Glascoe 1995). Parents completed 50 items that together represent
the Expressive Language scale and measures expressive communication ranging from simple
gestures and words to complex language (e.g., Asks questions beginning with “what” or
“where”). The Language Comprehension scale also consists of 50 items and relates to the
understanding of simple instructions to the understanding of complex concepts (e.g.,
Understands the meaning of at least six location words, such as “in, on, under, beside, top,
bottom, above, below”).

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

To define whether ToM development evolved in the same manner in both children with MHL and in hearing children, participants were grouped into four stages of increasing ability to successfully complete the desire and belief tasks (Wellman & Liu 2004; Peterson et al. 2005; Peterson & Wellman 2009). Because the Desire tasks each consisted of two vignettes, children needed to pass both tasks successfully in order to pass for this stage. ToM-Stage 1 was assigned when the child was unable to successfully complete any of the desire or belief tasks. Successful acknowledgement of similar desires resulted in assignment of the child to Stage 2. Stage 3 was assigned when a child also managed to acknowledge dissimilar desires. If a child mastered all ToM skills he or she was assigned to Stage 4. When other patterns were shown by the children, these were categorized as divergent. Categories were compared using the likelihood ratio test because some categories contained fewer than 5 participants.

Missing data
In the group of children with MHL, verbal comprehension scores were missing from 7 participants, word development scores were missing from 11 participants and sentence development scores were missing from 10 children. When conducting standard analyses such as ANCOVA’s and Pearson’s correlations, incomplete cases are automatically excluded from the analyses. Excluding these participants might give bias and would lower the power of our results. Therefore, missing language scores on the RLDS and the SELT were reconstructed using multiple imputations. This technique estimates a prediction model based on the complete cases and uses this model to predict outcomes of missing scores (Schafer & Graham 2002; Sterne et al. 2009; Van Buuren 2012; De Goeij et al. 2013; Netten et al. Accepted for publication). Language scores were predicted using the child’s age, language skills as reported by their parents (CDI), and observations during the ToM tasks. Ten imputations were performed because research has shown that this is a sufficient number to make a robust estimation of each unique data point (Sterne et al. 2009; Van Buuren 2012). Statistical analyses were carried out using the program SPSS version 23.0 (IBM 2013). One child with MHL refused to answer the dissimilar desire task. In analyses concerning desire understanding, this participant was excluded. Because of low language skills, one child was not able to perform the desire and false belief understanding task. This child was excluded in analysis that included these variables.
RESULTS

**Intention understanding**

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

**Desire understanding**

The ability to acknowledge others’ desires was assessed using a mixed ANCOVA with Desires (Similar and Dissimilar) as the within-subject variable, Group (MHL vs. hearing) as the between-subject variable and Age as the covariate. This analysis revealed a main effect for Group ($F(1.141) = 30.967, p < 0.001, \eta^2 = 0.18$) and Age ($F(1.141) = 12.714, p < 0.001, \eta^2 = 0.08$). On both Similar and Dissimilar desires, children with MHL scored lower than the hearing group (as indicated by the letter superscripts in Table 2). Older children were better in acknowledging others’ desires than younger children.

**Belief understanding**

The understanding of false beliefs was analyzed by logistic regression with Group (MHL vs. hearing) and Age as predictors. The outcomes in Table 3 show that children with MHL scored lower than hearing children on the false belief task. The Odds Ratio (OR) of 0.41 indicates that the chance of successfully completing the false belief task was lower in children with MHL. The understanding of false beliefs increased with age. The OR of 1.09 illustrates that the change of successfully acknowledging false beliefs increased when children were older.

**Language and ToM**

Children with MHL were found to have language quotients within the normal range compared to test normative samples ($M=92.5, M=94.9, \text{and } M=94.4$ for receptive language, word development, and sentence development, respectively. Parent-reported language skills were lower in the MHL group compared to the hearing control group ($t(46.422) = -4.276, p < 0.001, \text{and } t(50.419) = -3.326, p < 0.01$ for expressive language and language comprehension, respectively).
The relation between age and the ToM tasks was assessed first because age was thought to be a possible confounder of the relation between ToM and language abilities, as shown in Table 4. Pearson’s correlations revealed a positive relationship between age and all tasks in both groups. Partial correlations corrected for Age revealed a positive relation between both Expressive language and Language comprehension as reported by parents, and all ToM tasks. However, the relation between both parent-reported language indices and Similar desire was absent in the MHL group and significantly different from the hearing group (z = 2.12, p < 0.05, and z = 2.69, p < 0.01 for Expressive language and Language comprehension, respectively). This same pattern was seen in the relation between the Dissimilar desire task and Expressive language (z = 2.11, p < 0.05).

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

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

**Different stages of ToM development**

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

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

**DISCUSSION**

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

Both groups were equally able to understand others’ intentions. However, children with MHL had relatively more difficulties than hearing controls with interpreting others’ intentions when the other’s goal was not achieved compared to more directive intention understanding tasks. Perhaps the nature of the hand gestures in the joint attention tasks was much more explicit than in the intention understanding tasks. It has previously been found that parents of children with MHL show more directive communication towards their child than parents of hearing children (Pressman et al. 1999). Possibly, children with MHL are better used to this direct form of non-verbal communication using gestures to focus attention than to more indirect forms of communication where they need to interpret the situation before they understand what is going on. The hearing children on the other hand are relatively good in joint attention compared to the MHL group, this task only asks for a shared focus of attention, without having to participate actively.

Albeit most children with MHL showed sequences of ToM development similar to hearing children, one in four children showed a divergent pattern compared to one in ten in the hearing group. Children with MHL who showed such divergent sequences did not differ in their language abilities or in other hearing loss related factors such as age at detection of hearing loss or age at start of hearing amplification compared to those with normal sequences. However, we should interpret these results with care as these analyses were done in rather small groups. A lack of power could have prevented us from finding significant results. Because we were not able to identify factors that influenced such divergent development, we can only speculate about causes for divergent development. Possibly, the duration of testing was more exhausting for children with hearing loss. Since the belief understanding task was administered at the beginning of the test session, it may be that the children paid more attention than when administering the desire task at the end. In addition, beliefs were measured by a single task whereas to pass the (dis)similar desire tasks, children needed to succeed on the test twice resulting in a higher chance to fail one of them and obtaining a negative score. Yet, all tasks have previously been used successfully in different
clinical groups (i.e., preschoolers with a CI and preschoolers with an autism spectrum disorder) with reliable results (Ketelaar et al. 2012; Broekhof et al. 2015).

Despite their relatively good intention understanding skills, children with MHL fall behind compared to hearing peers on more language dependent skills such as desire and belief acknowledgement. In line with previous studies in children with more severe hearing loss wearing a CI, it is likely that a hearing loss may act as a barrier that prevents sufficient access to social communication in our sound-dominated world. This reduced ability to adequately receive social cues may cause a delay in ToM development (Ketelaar et al. 2012; Sundqvist et al. 2014). The relationship between ToM and hearing loss can be explained by several challenges that children with hearing loss and their families have to face. One aspect is the input children with hearing loss receive from their parents. In the first few years of life, parents provide the largest proportion of verbal input to the child. When parents talk about how others feel, what they want or wish for, they stimulate ToM understanding in their hearing children (Harris 2005; Taumoepeau & Ruffman 2006). However, research has shown that the quality of input that parents present to their child with MHL is frequently lower than in hearing children (Ambrose et al. 2015). As a result, children with MHL may encounter more difficulties increasing their language capacities. This in turn may prevent them from higher quality interactions that are essential in order to discuss abstract concepts such as other’s mental states and emotions.

However, what is said is not only important, but also how it is said. Both diversity in syntactic structures and the introduction of various speakers can positively influence ToM development (De Villiers & De Villiers 2000; Taumoepeau & Ruffman 2006; Bernard & Deleau 2007). Yet, parents of children with hearing loss often choose more simple and clear formulations when talking to their child. A relatively larger proportion of communication is also more directive in nature, aiming to instruct the child instead of discussing or explaining the child’s surroundings. Parents adjust the complexity of their language to the child’s language abilities (Ambrose et al. 2015). Although simple and clear communication can benefit language understanding in children with MHL, limited diversity of input may also hamper more complex language development in the long run. Again, diminished opportunities to learn about others’ perspectives may lead to less experience in ToM usage in children with MHL.

With the introduction of cochlear implantation, the focus of research on hearing loss has shifted. Improving and understanding the effects of this highly innovative technique became the goal of many funders and commercial companies for obvious reasons (Lederberg et al. 2013). But how about the children with moderate hearing loss? A recent special issue of Ear and Hearing discussing the Outcomes of Children with Hearing Loss (OCHL) Study by Moeller and colleagues addressed the challenges that children with MHL have to face. Among other things, this large longitudinal study revealed that children with MHL are still at risk for the development of language delays. The outcomes of the present study in which the language skills of children with MHL are in the low-normal range compared to test normative samples are in line with these findings. Despite their relatively normal language skills, the parent-reported language skills of children in the MHL group were below the average range.
These scores possibly better reflect children’s communication skills in daily life, because parents do not base their judgment on one particular moment but on the child’s average skills over a longer period in time. Because communicative abilities determine how well a child is able to join conversations with others, this may also better reflect their opportunities for incidental learning, which subsequently determines their social development. This is in line with the outcomes of the OCHL study in which qualitative aspects of conversations were important for a child’s language output (Ambrose et al. 2015; Tomblin et al. 2015). Our study is unique in providing insight into the relation between language skills and different aspects of ToM.

Parent-reported language skills were strongly related to ToM in the hearing controls. Yet, the relation between desire understanding and parent-reported language skills in children with MHL was almost absent. On the other hand, we found a relation between objective test-scores and desire understanding. It is possible that parents rate their child’s language skills in daily life, and take account for their lower communication skills in interactions with others and in noisy environments. They acknowledge the difficulties their child with MHL has in communication with others. This obviously differed from the quiet language-test settings in clinical surroundings. During the ToM observations in this study there was no time limit so children could take their time which might have benefitted their ToM outcomes compared to how they would have responded in hectic daily life. Still, this does not explain the absent relation between objective language tests and the dissimilar desire task. This absence could be the result of our study design. Children completed the false belief tasks relatively early and the dissimilar desire tasks relatively late during the test session. In addition, the dissimilar desire task was preceded by a rather difficult task that is not described in this study. Possibly, the children became tired and lost their concentration. Concentration difficulties are well known in children with various degrees of hearing loss (Bess & Hornsby 2014). Either way, this finding highlights the importance of this study. It aims to trigger both parents and professionals to be alert when it comes to ToM development in young children with MHL. It shows that although parents are well able to understand their child and professionals rate their language abilities to be within the average range, these children are at risk for delays in their social development. In addition, the outcomes of this study suggest it might be better to also focus on the child’s communicative abilities than to solely rely on language test scores (Tomblin et al. 2015).

Future research

We would like to point out that this study is a first attempt to address ToM-related difficulties in children with MHL. Some of the analyses were done in rather small groups and using a cross-sectional design. A second limitation of the current study concerns the absent language scores in the control group. Although norm-scores were available for typically developing (hearing) children, it would be more convenient to directly compare the two groups. Although a clear difference in ToM skills was found between the two groups, we feel that we are only able to hypothesize about a possible delay when focusing on the developmental patterns of ToM in young children with MHL. To confirm our findings, there is a strong need for longitudinal research that is able to link age, language and ToM abilities of increasing
difficulty to confirm causality and to focus on different developmental patterns in this specific group of young children. In addition, future research should also include participant and family-related factors that may influence social development like the cognitive abilities of the child (e.g., phonological working memory, executive functioning) and the socioeconomic status of the family as these factors are known to influence language skills and general development. This study was unable to show a direct link between hearing loss-related factors such as the age at detection or the age at first HA amplification and ToM. However, factors like audibility and early access to HA’s have been proven to influence language skills in MHL children and should therefore be integrated in future studies when studying social functioning in this group of children (Tomblin et al. 2015).

CONCLUSION

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

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AN, SK, and AK had full access to all of the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis. Study concept and design: SK, AK, AMOM, CR, FD, JF. Acquisition, analysis, or interpretation of data: All authors. Drafting of the manuscript: AN, CR. Critical revision of the manuscript for important intellectual content: All authors. Statistical analysis: AN. Obtained funding: AMOM, JF. Administrative, technical, or material support: All authors. Study supervision: CR, JF. Approval for this study was obtained through the Ethical Committee of the Leiden University Medical Center.
REFERENCES


Figure 1: Spread of ToM Stages by age across participants, separated for children with MHL and hearing controls.

Abbreviations: ToM Theory of Mind, MHL Moderate Hearing Loss

Note: Data points are overlapping in children of the same age assigned to the same ToM Stage.
Table 1: Demographic characteristics of participants

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

* p< 0.01, ** p< 0.001
† Categories: 0 = don't know, 1 =no education/primary education, 2 = lower general secondary education, 3 = higher general secondary education, 4 = college/university
‡ Raw scores

Abbreviations: MHL Moderate Hearing Loss, SD Standard Deviation, CDI Child Development Inventory, BCD Bone Conduction Device
Table 2: Mean scores on different aspects of ToM observations in both groups.

<table>
<thead>
<tr>
<th>Observation</th>
<th>MHL</th>
<th>Control</th>
<th>range</th>
</tr>
</thead>
<tbody>
<tr>
<td>Intention understanding</td>
<td>2.05 (1.03)\textsuperscript{a1}</td>
<td>2.32 (0.88)\textsuperscript{a1}</td>
<td>0-3</td>
</tr>
<tr>
<td>Joint attention</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Imperative Comprehension</td>
<td>2.62 (0.87)\textsuperscript{a2}</td>
<td>2.70 (0.72)\textsuperscript{a2}</td>
<td>0-3</td>
</tr>
<tr>
<td>Declarative Comprehension</td>
<td>2.57 (0.67)\textsuperscript{a2}</td>
<td>2.37 (0.58)\textsuperscript{a1}</td>
<td>0-3</td>
</tr>
<tr>
<td>Desires</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Similar</td>
<td>0.67 (0.38)\textsuperscript{a1}</td>
<td>0.93 (0.23)\textsuperscript{b1*}</td>
<td>0-1</td>
</tr>
<tr>
<td>Dissimilar</td>
<td>0.62 (0.42)\textsuperscript{a1}</td>
<td>0.89 (0.28)\textsuperscript{b1*}</td>
<td>0-1</td>
</tr>
<tr>
<td>False belief</td>
<td>0.44 (0.50)\textsuperscript{a}</td>
<td>0.63 (0.48)\textsuperscript{b†}</td>
<td>0-1</td>
</tr>
</tbody>
</table>

Abbreviations: MHL Moderate Hearing Loss, SD Standard Deviation

Letter-superscripts indicate differences at \( p < 0.05 \) on rows (between groups), number-superscripts indicate differences at \( p < 0.05 \) on columns (between tasks within groups).

* Groups differed on both desire tasks at \( p < 0.001 \)
† Groups differed on the false belief task at \( p < 0.01 \)
Table 3: Logistic regression predicting False belief understanding

<table>
<thead>
<tr>
<th></th>
<th>B (SE)</th>
<th>Wald</th>
<th>Odds Ratio</th>
<th>p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Constant</td>
<td>-4.273 (1.36)</td>
<td>9.91</td>
<td>0.14</td>
<td>0.24</td>
</tr>
<tr>
<td>Group</td>
<td>-0.887 (0.39)</td>
<td>5.11</td>
<td>0.41</td>
<td>0.002</td>
</tr>
<tr>
<td>Age</td>
<td>0.084 (0.02)</td>
<td>12.70</td>
<td>1.09</td>
<td>0.000</td>
</tr>
</tbody>
</table>

*Note. Model $\chi^2 (2) = 18.50, p < 0.001,*
*Group was dummy coded: 0=control group, 1= children with moderate hearing loss*
Table 4: (Partial) correlations between different aspects of ToM, parent-reported language skills, and age.

<table>
<thead>
<tr>
<th>Age</th>
<th>Language Comprehension (CDI)</th>
<th>Expressive Language (CDI)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>( r )</td>
<td>Partial ( r )</td>
</tr>
<tr>
<td></td>
<td>MHL</td>
<td>Control</td>
</tr>
<tr>
<td>Similar desire</td>
<td>.23*</td>
<td>.09</td>
</tr>
<tr>
<td>Dissimilar desire</td>
<td>.24*</td>
<td>.26*</td>
</tr>
<tr>
<td>False belief</td>
<td>.30**</td>
<td>.24*</td>
</tr>
</tbody>
</table>

Note: Partial correlations are corrected for age. Only when correlations between the two groups significantly differed (calculated using Fisher \( r \)-to-\( z \)), both coefficients are given separately.

Abbreviations: MHL Moderate Hearing Loss, CDI Child Development Inventory

*\( p < 0.01 \), **\( p < 0.001 \)
Table 5: Partial correlations in participants with MHL between different aspects of ToM observations, language test scores, and hearing loss related factors, corrected for age (N=43).

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

Abbreviations: HL Hearing Loss, RDLS Reynell Developmental Language Scales, SELT Schlichting Expressive Language Test

* p < 0.05, **p < 0.01, ***p < 0.001
Table 6: Different stages of ToM development.

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

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