

1 This is a post-print of: Netten, A. P., Rieffe, C., Soede, W., Dirks, E., Korver, A. M. H., Konings,
2 Briaire, J. J., Oudesluys-Murphy, A.M., Dekker, F.W., & Frijns, J.H.M., on behalf of the DECIBEL
3 Collaborative study group (2017). Can You Hear What I Think? Theory of Mind in Young Children
4 With Moderate Hearing Loss. *Ear and Hearing*, 38, 588-597, which was published at:
5 <http://dx.doi.org/10.1097/AUD.0000000000000427>

6

ABSTRACT

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

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

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

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

INTRODUCTION

33 Engagement in social interactions is essential for the social-emotional development of
34 children. In order to induce and maintain relationships, children need to learn that different
35 people have different intentions, desires, and beliefs. The ability to apply such mental states
36 to others is known as ‘Theory of Mind’ (ToM). Through ToM development, children will
37 start to understand that our mental states explain our actions (e.g., dad chooses coffee for
38 dessert because *he* prefers coffee over ice-cream). ToM development has been studied
39 extensively over the last two decades (Wellman 1990; Dunn 1996). These studies revealed
40 that both language and communicative abilities are very important for an adequate ToM
41 development (see (Stanzione & Schick 2014) for a review). The importance of this relation
42 has been illustrated previously by many studies in deaf children of hearing parents. Outcomes
43 show severe delays in the ToM development of deaf children of hearing parents (Peterson &
44 Siegal 1999; Courtin 2000; De Villiers & De Villiers 2000; Schick et al. 2007) that may
45 continue to be problematic during adolescence (Pyers & Senghas 2009; Wellman et al. 2011).
46 One explanation offered in the literature for these findings lies in the reduced abilities of
47 parents (especially hearing parents who sign) to discuss abstract concepts such as thoughts
48 and emotions compared to hearing-hearing dyads (Moeller & Schick 2006). Children with
49 moderate hearing loss (MHL) share the same mode of communication as their hearing
50 parents. However, these children often still encounter language difficulties (Moeller et al.
51 2007; Moeller et al. 2015; Tomblin et al. 2015). Therefore, children with MHL are also
52 potentially at risk for inadequate ToM development. Nevertheless, until now, no research has
53 focused on the development of ToM in children with moderate hearing loss, which is the aim
54 of this study.

55

56 **Children with moderate hearing loss**

57 A substantial number of children have hearing loss thresholds falling in the moderate range
58 (40-70 dB HL). When wearing their hearing aids, children with MHL can function
59 reasonably well in quiet areas and in one-on-one conversations. They can hear what is said
60 when they are not disturbed by background noise that interferes with their hearing aids, their
61 ability to recognize consonants, and directional hearing (Eisenberg 2007; McCreery et al.
62 2015a). However, the hearing capacities of these children are frequently overestimated.
63 Children with MHL frequently encounter difficulties in fully understanding what is said in
64 daily interactions, especially in noisy environments such as daycare centers and classrooms
65 (Finitzo-Hieber & Tillman 1978). Children with hearing loss encounter difficulties in speech
66 perception when listening to speech in noise (Yang et al. 2012). Furthermore, the children’s
67 hearing aids (HAs) are often not fit optimally, which may negatively impact their hearing
68 potential (McCreery et al. 2015; Tomblin et al. 2015). For the child’s surroundings, it is often
69 difficult to understand what a child with MHL does hear and what input is missed.
70 Diminished access to social conversations could potentially diminish their opportunities for
71 social learning, which has ongoing consequences for their social-emotional development.

72 **ToM development and hearing loss**

73 In studies on ToM development, the majority of research has focused on only one aspect of
74 ToM development, that is, the understanding of (false) beliefs. Yet, Wellman and others
75 emphasize on the importance of studying ToM in its broadest sense. Thereby, it is important
76 to be aware of the fact that the acknowledgement of others' intentions and desires precedes
77 the understanding of others' (false) beliefs (Wellman 2002). This was previously
78 demonstrated in large studies examining the developmental sequence of ToM development in
79 deaf children and children with an autism spectrum disorder. These studies show that deaf
80 children generally show the same sequential pattern of ToM development as hearing peers,
81 albeit slower (Peterson et al. 2005; Peterson & Wellman 2009). This delayed ToM
82 development can have ongoing consequences for a child's social development (Olson et al.
83 2011; Caputi et al. 2012).

84 Intention understanding

85 An essential precursor for the development of ToM is the ability to acknowledge others'
86 intentions (Sodian & Kristen-Antonow 2015). Growing consciousness of the fact that others'
87 actions are guided by their intentions teaches children to separate human beings from objects.
88 Only by knowing someone else's intentions, one can understand the person's actions. To
89 illustrate, the physical movement of an object from one person to the other can be interpreted
90 as giving, sharing, loaning, returning, or trading something. Yet, without intention
91 understanding, we do not know why actions happen. In typically developing children,
92 intention understanding begins to emerge in the second year of life (Tomasello et al. 2005).
93 An important aspect of intention understanding is joint attention; the ability to share attention
94 with someone else concerning an object or situation. Drawing someone's attention to a
95 certain situation increases language development and strengthens relationships. Studies in
96 young children show equal levels of joint attention in deaf children with CI compared to age-
97 related peers, whereas less engagement in joint attention was seen in deaf children without a
98 CI (Tasker et al. 2010; Ketelaar et al. 2012; Cejas et al. 2014).

99 Desire understanding

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

114 Belief understanding

115 Classic false belief tasks include the change-of-location and the unexpected-content task. In
116 both tasks, the child is questioned about the behavior of a story character. In the story, this
117 character holds a belief that opposes the actual truth. Around the age of four, children start to
118 appreciate other's beliefs. Research shows equal levels of belief understanding in deaf
119 children born to deaf parents compared to hearing peers born to hearing parents (see
120 Stanzione and Schick 2014 for an overview) (Schick et al. 2007; Stanzione & Schick 2014).
121 However, deaf children of hearing parents performed lower on false belief understanding
122 than hearing children, with so-called late signers showing the least favorable results (Courtin
123 2000; Peterson et al. 2005). This difference can be explained by the quality and quantity of
124 communication. Both deaf children who acquire oral communication and deaf children who
125 acquire sign language relatively late (because it is their second language) may encounter
126 limited participation in high-quality social interactions involving mental state talk, be it in
127 school or with their family at home (Jeanes et al. 2000; Macaulay & Ford 2006; Ziv et al.
128 2013).

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

140 **ToM and language**

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

150 In DHH children, the relation between language and ToM skills seems complex. False
151 belief tasks for instance contain 'mental state verbs' and 'if/then statements'. In order to
152 understand such complex ToM tasks, a certain level of language and communication skills is
153 needed to succeed. As a result it is often unclear what it is exactly that such tasks are
154 measuring: the child's ToM skills or their language capacities. Schick et al. therefore used

155 ToM tasks that required minimal language skills to measure ToM abilities in deaf children of
156 hearing parents. Results showed that the deaf children in their study also performed lower on
157 the low-verbal tasks compared to hearing children and deaf children of deaf parents,
158 indicating the importance of access to communication with others. This statement was
159 underlined by the fact that complement processing skills were found to predict performance
160 on low-verbal ToM tasks, yet vocabulary comprehension skills did not (Schick et al. 2007).

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

169 **Present study**

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

188 **METHODS**

189 **Procedure**

190 The children with MHL in this study were identified through the DECIBEL-study. DECIBEL
191 stands for Developmental Evaluation of Children: Impact and Benefits of Early hearing
192 screening strategies Leiden. The DECIBEL-study was conducted in The Netherlands between
193 2008 and 2010 to define the influence of early detection of hearing loss on the development
194 of young DHH children. This nationwide study identified all children who were born with

195 hearing loss between January 2003 and December 2005. Hearing loss was detected using
196 Otacoustic Emissions (OAEs) which enables identification of hearing loss of 35 dB HL or
197 more. The database consisted of 210 children with permanent bilateral hearing loss. Ethical
198 approval for the DECIBEL-study was obtained through the Medical Ethics Committee of the
199 Leiden University Medical Center (Korver 2010; Korver et al. 2010).

200 For participation in the social-emotional assessments of the DECIBEL-study, children
201 needed to fulfill additional inclusion criteria. Children needed to be at least 36 months old,
202 their unaided hearing loss in the better ear should not exceed 70 dB HL, children had to use
203 conventional hearing aids or bone conduction devices (BCD) and it was requested that their
204 preferred mode of communication was either spoken, or sign-supported Dutch. This resulted
205 in 74 children who were eligible for participation, and their parents were invited to
206 participate. Finally, parents of 44 children gave informed consent (response rate 59.5%).
207 Children were visited at home. A researcher sat with the child in a quiet room and conducted
208 several tasks which will be explained in more detail below. The session was video-recorded
209 in order to score the child's behavior afterwards. The camera was positioned so that both the
210 experimenter and the child were recorded. This allowed for both observation of the child's
211 behavior as well as to check if all tasks were correctly performed by the experimenter. The
212 sequence of the tasks and observations that were performed was as follows: 1) Intention
213 understanding, 2) False belief task, 3) Similar desire task, 4) Imperative pointing, 5)
214 Dissimilar desire task, 6) *other tasks and observations not mentioned in this study*, 7)
215 Dissimilar desire task, 8) Declarative pointing, 9) *other tasks and observations not mentioned*
216 *in this study*, 10) Similar desire task. Completion of the whole set of tasks and observations
217 took approximately 35 to 45 minutes per child.

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

226 **Participants**

227 All children were between 40 and 70 months old during home observations (mean age 57
228 months). Of the 44 children with MHL, 27 were boys (61.4%). Their hearing loss varied with
229 a pure-tone-average between 35 and 70 dB HL in the better ear (mean loss 50 dB HL).
230 Residual hearing was calculated by averaging unaided hearing thresholds at 500, 1,000 and
231 2,000 Hz. Six children had a hearing loss between 35 and 40 dB. One child used a BCD, all
232 others used hearing aids. All but one were aided bilaterally. All children understood spoken
233 language, yet five of them (11.4%) preferred to use sign-supported Dutch. Parents of seven
234 children with MHL reported having hearing loss themselves. Three children with MHL had
235 an additional handicap. One child was diagnosed with Turner syndrome, one child suffered

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

240 **Materials**

241 Intention understanding

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

251 The 'Declarative Comprehension task' measures joint attention (Colonnesi et al.
252 2008; Ketelaar et al. 2012). During this task, the researcher acts surprised and points to an
253 object out of sight of the child. The researcher then looks back and forth between the object
254 and the child. The subsequent behavior of the child was observed and children could receive
255 up to three points when they looked at the object, looked at the researcher and, attempted to
256 communicate about the object.

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

266 Desire understanding

267 The acknowledgement of others' desires was assessed using the 'desire task' (Ketelaar et al.
268 2012). This task uses vignettes to measure two types of desires: similar and dissimilar
269 desires. In the similar desire condition, the child is presented with a picture showing two
270 types of food (e.g., tomato and ice-cream). The child is asked what he or she prefers to eat.
271 The researcher then tells a story about a boy who also likes the food that the child just chose.
272 Then the child is asked: "Now the boy can choose a snack. What will the boy choose to eat?"
273 This question is followed by two control questions: "Does the boy like [Snack 1]?" and
274 "Does the boy like [Snack 2]?" The child is awarded one point if he or she answers all three

275 questions correctly. In the dissimilar desire task, the only difference is that the protagonist in
276 the story does not like the snack that the child preferred but instead likes the opposite snack.

277 Belief understanding

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

288 Language

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

300 Parent-reported language skills

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

309 **Statistical analyses**

310 To assess differences between the two groups on ToM abilities and precursors (mixed design)
311 analyses of covariance (ANCOVA’s) were used to test both between-group and repeated-
312 measures variables. Because the outcome on the False Belief task was dichotomous (i.e., pass
313 or not) logistic regression was used to predict the effect of group and age on belief
314 understanding. Pearsons’ correlations and partial correlations were used to identify the

315 relation between ToM skills and language abilities, taking the age of the child into account.
316 Fisher r -to- z transformations were used to compare if the correlation coefficients differed
317 between children with MHL and hearing controls.

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

330 Missing data

331 In the group of children with MHL, verbal comprehension scores were missing from 7
332 participants, word development scores were missing from 11 participants and sentence
333 development scores were missing from 10 children. When conducting standard analyses such
334 as ANCOVA's and Pearson's correlations, incomplete cases are automatically excluded from
335 the analyses. Excluding these participants might give bias and would lower the power of our
336 results. Therefore, missing language scores on the RLDS and the SELT were reconstructed
337 using multiple imputations. This technique estimates a prediction model based on the
338 complete cases and uses this model to predict outcomes of missing scores (Schafer & Graham
339 2002; Sterne et al. 2009; Van Buuren 2012; De Goeij et al. 2013; Netten et al. Accepted for
340 publication). Language scores were predicted using the child's age, language skills as
341 reported by their parents (CDI), and observations during the ToM tasks. Ten imputations
342 were performed because research has shown that this is a sufficient number to make a robust
343 estimation of each unique data point (Sterne et al. 2009; Van Buuren 2012). Statistical
344 analyses were carried out using the program *SPSS* version 23.0 (IBM 2013). One child with
345 MHL refused to answer the dissimilar desire task. In analyses concerning desire
346 understanding, this participant was excluded. Because of low language skills, one child was
347 not able to perform the desire and false belief understanding task. This child was excluded in
348 analysis that included these variables.

349

350

RESULTS

351 **Intention understanding**

352 The mean scores on outcomes of all observations are shown in Table 2. To assess if children
353 with MHL differed from hearing children in their ability to acknowledge others' intentions, a
354 mixed-design ANCOVA was performed with Intention understanding (Intention
355 understanding, Declarative pointing, and Imperative pointing) as the within-subject variable,
356 Group (MHL vs. hearing) as the between-subjects variable and Age as the covariate. No main
357 effects were found. An interaction effect was found for Intention understanding \times Group F_{HF}
358 (1.936, 267.225) = 3.063, $p < 0.05$, $\eta^2 = 0.02$. Age significantly influenced intention
359 understanding ($F(1, 138) = 3.971$, $p < 0.05$). Subsequent paired t-tests in both groups
360 separately revealed that children with MHL showed relatively lower Intention understanding
361 compared to Declarative and Imperative pointing (as indicated by the number superscripts in
362 Table 2). In the hearing group, children scored relatively higher on Imperative pointing as
363 compared to Declarative pointing and Intention understanding. Intention understanding
364 abilities increased with age.

365 **Desire understanding**

366 The ability to acknowledge others' desires was assessed using a mixed ANCOVA with
367 Desires (Similar and Dissimilar) as the within-subject variable, Group (MHL vs. hearing) as
368 the between-subject variable and Age as the covariate. This analysis revealed a main effect
369 for Group ($F(1,141) = 30.967$, $p < 0.001$, $\eta^2 = 0.18$) and Age ($F(1,141) = 12.714$, $p < 0.001$,
370 $\eta^2 = 0.08$). On both Similar and Dissimilar desires, children with MHL scored lower than the
371 hearing group (as indicated by the letter superscripts in Table 2). Older children were better
372 in acknowledging others' desires than younger children.

373 **Belief understanding**

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

381 **Language and ToM**

382 Children with MHL were found to have language quotients within the normal range compared
383 to test normative samples (M= 92.5, M=94.9, and M=94.4 for receptive language, word
384 development, and sentence development, respectively. Parent-reported language skills were
385 lower in the MHL group compared to the hearing control group ($t(46.422) = -4.276$, $p <$
386 0.001 , and $t(50.419) = -3.326$, $p < 0.01$ for expressive language and language comprehension,
387 respectively).

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

398 The six children with a PTA between 35 and 40 dB HL were compared with the 38
399 remaining children with a hearing loss between 40-70 dB. The parents of these six children
400 with mild hearing loss reported higher Expressive language scores ($t(39.10) = -3.715, p <$
401 0.01 than the parents of children with MHL. No difference was found in their Language
402 comprehension scores. We also observed better understanding of similar desires in the group
403 of children with mild hearing loss compared to the children with MHL ($t(11.87) = -2.691, p <$
404 0.05 . No differences were found in Intention understanding, Dissimilar desires or False belief
405 understanding between the two groups.

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

412

413 **Different stages of ToM development**

414 Children with MHL were more often in the lower ToM stages than their hearing peers ($\chi^2(4)$
415 $= 25.632, p < 0.001$). The various ToM stages can be found in Table 6. More than half of all
416 hearing children (54.4%) mastered all ToM skills compared to 25% of children with MHL. A
417 4 (ToM stages) x 2 (Group) mixed ANOVA with Age as the dependent variable revealed no
418 differences in age between the two groups in any of the ToM stages, although the overall
419 mean age per ToM stage was different ($F(3, 114) = 7.462, p < 0.001$. With increasing age,
420 children more often succeeded in the higher ToM stages. Figure 1 illustrates the relation
421 between the different ToM stages and age. Despite the fact that we did not find a difference
422 in age per ToM stage between the two groups, a tendency of hearing children reaching the
423 higher ToM stages earlier in life can be seen.

424 Approximately one-fourth (12; 27.3%) of children with MHL showed a divergent
425 sequence compared to 11 (10.9%) in the hearing group ($\chi^2(1) = 6.163, p < 0.05$). The
426 divergent sequences were so idiosyncratic that each appeared in only one or two children. For
427 reasons of clarity, these sequences were not visualized here. Compared to children with

428 normal developmental sequences, the children showing divergent sequences did not differ on
429 characteristics such as age and language capacities. When focusing only on the group of
430 children with MHL, no differences were found in age at detection, age at amplification of
431 first hearing device, degree of hearing loss, and language capacities when comparing children
432 with divergent sequences to those with the most common ToM development sequences.

433 **DISCUSSION**

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

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

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

469 clinical groups (i.e., preschoolers with a CI and preschoolers with an autism spectrum
470 disorder) with reliable results (Ketelaar et al. 2012; Broekhof et al. 2015).

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

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

500 With the introduction of cochlear implantation, the focus of research on hearing loss
501 has shifted. Improving and understanding the effects of this highly innovative technique
502 became the goal of many funders and commercial companies for obvious reasons (Lederberg
503 et al. 2013). But how about the children with moderate hearing loss? A recent special issue of
504 *Ear and Hearing* discussing the Outcomes of Children with Hearing Loss (OCHL) Study by
505 Moeller and colleagues addressed the challenges that children with MHL have to face.
506 Among other things, this large longitudinal study revealed that children with MHL are still at
507 risk for the development of language delays. The outcomes of the present study in which the
508 language skills of children with MHL are in the low-normal range compared to test normative
509 samples are in line with these findings. Despite their relatively normal language skills, the
510 parent-reported language skills of children in the MHL group were below the average range.

511 These scores possibly better reflect children's communication skills in daily life, because
512 parents do not base their judgment on one particular moment but on the child's average skills
513 over a longer period in time. Because communicative abilities determine how well a child is
514 able to join conversations with others, this may also better reflect their opportunities for
515 incidental learning, which subsequently determines their social development. This is in line
516 with the outcomes of the OCHL study in which qualitative aspects of conversations were
517 important for a child's language output (Ambrose et al. 2015; Tomblin et al. 2015). Our study
518 is unique in providing insight into the relation between language skills and different aspects
519 of ToM.

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

543 **Future research**

544 We would like to point out that this study is a first attempt to address ToM-related difficulties
545 in children with MHL. Some of the analyses were done in rather small groups and using a
546 cross-sectional design. A second limitation of the current study concerns the absent language
547 scores in the control group. Although norm-scores were available for typically developing
548 (hearing) children, it would be more convenient to directly compare the two groups.
549 Although a clear difference in ToM skills was found between the two groups, we feel that we
550 are only able to hypothesize about a possible delay when focusing on the developmental
551 patterns of ToM in young children with MHL. To confirm our findings, there is a strong need
552 for longitudinal research that is able to link age, language and ToM abilities of increasing

553 difficulty to confirm causality and to focus on different developmental patterns in this
554 specific group of young children. In addition, future research should also include participant
555 and family-related factors that may influence social development like the cognitive abilities
556 of the child (e.g., phonological working memory, executive functioning) and the
557 socioeconomic status of the family as these factors are known to influence language skills
558 and general development. This study was unable to show a direct link between hearing loss-
559 related factors such as the age at detection or the age at first HA amplification and ToM.
560 However, factors like audibility and early access to HA's have been proven to influence
561 language skills in MHL children and should therefore be integrated in future studies when
562 studying social functioning in this group of children (Tomblin et al. 2015).

563 **CONCLUSION**

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

570 **ACKNOWLEDGEMENT**

571 The authors would like to thank all children and their parents for their involvement and
572 participation in this study. We are also deeply grateful for the help of Lisanne Seekles and
573 Andrea Lievense for their extensive help with the data collection. The DECIBEL-study was
574 financially supported by the Heinsius-Houbolt Fund, the Willem-Alexander Children's Fund,
575 and the Wieger Wakinoerfund.

576 AN, SK, and AK had full access to all of the data in the study and take responsibility
577 for the integrity of the data and the accuracy of the data analysis. Study concept and design:
578 SK, AK, AMOM, CR, FD, JF. Acquisition, analysis, or interpretation of data: All authors.
579 Drafting of the manuscript: AN, CR. Critical revision of the manuscript for important
580 intellectual content: All authors. Statistical analysis: AN. Obtained funding: AMOM, JF.
581 Administrative, technical, or material support: All authors. Study supervision: CR, JF.
582 Approval for this study was obtained through the Ethical Committee of the Leiden University
583 Medical Center.
584

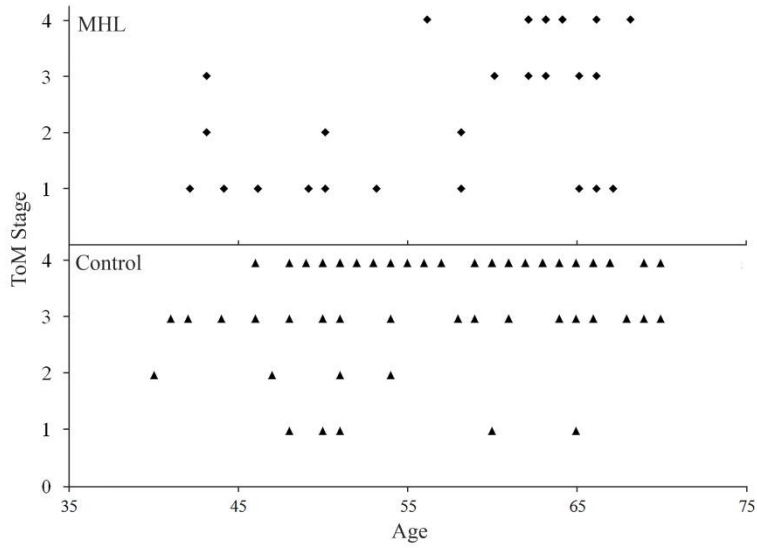
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725 Figure 1: Spread of ToM Stages by age across participants, separated for children with MHL
 726 and hearing controls.

727 *Abbreviations:* ToM Theory of Mind, MHL Moderate Hearing Loss

728 *Note:* Data points are overlapping in children of the same age assigned to the same ToM Stage.

729

Table 1: Demographic characteristics of participants

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

Abbreviations: MHL Moderate Hearing Loss, SD Standard Deviation, CDI Child Development Inventory, BCD Bone Conduction Device

* $p < 0.01$, ** $p < 0.001$

† Categories: 0 = don't know, 1 =no education/primary education, 2 = lower general secondary education, 3 = higher general secondary education, 4 = college/university

‡ Raw scores

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

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

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

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735 Table 3: Logistic regression predicting False belief understanding

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

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

736

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

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

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

Abbreviations: MHL Moderate Hearing Loss, CDI Child Development Inventory

p* < 0.01, *p* < 0.001

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

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

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

* $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$

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746 Table 6: Different stages of ToM development.

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

Abbreviations: ToM Theory of Mind, MHL Moderate Hearing Loss

- : Participant was not able to successfully complete this task

+ : Participant successfully completed this task

* $p < 0.05$

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