A New Look at Theory of Mind in Children With Ocular and Ocular-Plus Congenital Blindness

Sander Begeer, Marjolein Dik, Marieke J. voor de Wind, Doreen Asbrock, Michael Brambring, and Sabina Kef

Structured abstract: Introduction: Delays in theory of mind (ToM) of children who are congenitally blind have often been attributed to the absence of visual and social experiences. However, these delays could also be partly due to neural factors. In some children, the blindness itself has neural causes (ocular-plus blindness). Children whose blindness has an ocular-plus cause may be more delayed in ToM than children with blindness due to ocular causes. Methods: In the current study, performances of children with congenital ocular-plus blindness \((n = 22)\) and congenital ocular blindness \((n = 9)\) were compared with sighted children \((n = 103)\) on ToM tasks designed for children with blindness. Results: Compared with sighted children, ToM performance was delayed in children with ocular-plus blindness, but not in children with ocular blindness. Discussion: ToM development in children with congenital blindness could be related to factors other than the loss of a sensory function and the lack of visual social and communicative experiences. Implications for practitioners: The specific ToM deficits in children with ocular-plus blindness may help in developing new research paradigms that consider delays in ToM in children with congenital blindness.

Theory of mind (ToM) refers to the understanding that mental states such as beliefs and desires govern human behavior.

We wish to thank all the children who participated in the study and all the students who helped with the data collection.

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Previous studies have indicated a delay in the development of ToM in children with congenital blindness, despite their normal IQs (see, for example, Green, Pring, & Swettenham, 2004). Although the lack of vision and social experience is an obvious explanation for these delays—possibly in conjunction with additional impediments—there may also be alternative explanations more closely linked to the neuronal development of children with blindness. In fact, for many children, the cause for the blindness itself is linked to the optic...
neural pathways (hereafter referred to as “ocular-plus blindness”). These underlying neurological mechanisms may also influence the development of ToM abilities. Therefore, ToM delays might be more pronounced in children with ocular-plus blindness compared with children whose blindness does not involve the optic neural pathways (ocular blindness). The current study focused on ToM abilities of children with ocular-plus blindness compared with children with ocular blindness.

Blindness is defined by the World Health Organization as having visual acuity of less than 3/60 to no perception of light (World Health Organization, 2010). Blindness affects about .01% to .04% of all living births (Gilbert & Awan, 2003). Ocular and ocular-plus congenital blindness can be distinguished based on the cause of blindness located either before the optic tract (ocular blindness) or within the optic tract and further in the brain tissue (ocular-plus blindness). The optic tract is composed of a chain of four nerve cells. The first two neurons form the retina, and the third is the optic nerve. In the lateral geniculate nucleus, a part of the thalamus, the optic tract relays to the fourth neuron, projecting to the occipital lobe or visual cortex. The ocular-plus versus ocular partition in the visual function has been a subject of debate (Colenbrander, 2009; Dutton & Bax, 2010; Frebel, 2006). Although it was previously believed that the partition was made at the third neuron level, more recent studies show that information processing starts at the level of the retina (Roska, Molnar, & Werblin, 2006), and the retina activates brain plasticity (Morishita & Hensch, 2008; Sugiyama et al., 2008). Therefore, we define ocular-plus blindness as those diagnoses involving the retina and other parts of the optic tract and brain tissue, and ocular blindness as those diagnoses only involving all nonretinal parts of the eye globe.

ToM understanding is generally tested with false belief tasks. In these tasks, children are tested for their ability to reflect on a story character that has a false belief about a certain situation (for example, because the character is unaware that an object has been relocated). Typically developing children generally pass these tasks at around 6 years of age (Wellman, Cross, & Watson, 2001). Various studies have indicated delayed ToM development in children with congenital blindness, primarily based on standard false-belief tasks that were adapted for children with blindness (Green et al., 2004; McAlpine & Moore, 1995; Minter, Hobson, & Bishop, 1998; Peterson, Peterson, & Webb, 2000). These delays are usually ascribed to the absence of visual experiences in social and communicative interactions (see Sonksen & Dale, 2002, for a more multifaceted explanation). It can indeed be argued that the lack of vision prevents the development of important ToM precursors, such as sharing experiences based on joint visual attention and visual observations of subjective states (Bedny, Pascual-Leone, & Saxe, 2009; Minter et al., 1998). These explanations are very plausible. However, their plausibility might have caused researchers to overlook additional neuronal explanations for impaired ToM skills in children with congenital blindness.

The possibility of shared underlying neurological mechanisms for ToM delays and congenital blindness has received little scientific attention. This is remarkable because severe visual impairment or blindness...
often (77%) coincide with additional non-ophthalmic disorders or impairments (Rahi & Cable, 2003). For instance, 10% of children die within a year of being diagnosed with congenital blindness (Rahi & Cable, 2003). In addition, a common cause (5% to 20%) of congenital blindness is retinopathy of prematurity (ROP), an eye disease affecting prematurely born children. ROP coincides with additional disabilities in 68% to 74% of all cases. The percentage of comorbid neurological disabilities varies between 30% and 49% in children with ROP (van Sorge et al., 2011). In addition to the high incidence of comorbid disabilities in individuals with congenital blindness, there are several other reasons for focusing on a possible neuronal basis of their delayed ToM development.

Individual differences in the social abilities of children with blindness are great. Some show strikingly poor social responsiveness, whereas others seem to respond without limitations (Hobson, Lee, & Brown, 1999). This heterogeneity of social behavior has been linked to different factors that underlie the blindness (Hobson & Bishop, 2003). For instance, although many researchers agree that autism spectrum disorders are generally more prevalent in children with congenital blindness than in sighted children (11.6% versus 0.6%; Centers for Disease Control and Prevention, 2007; Mukaddes, Kilincaslan, Kucukyazici, Sevketoglu, & Tuncer, 2007), high rates of autism have been shown in particular when the blindness was caused by premature birth affecting the retina (ROP; Ek, Fernell, Jacobson, & Gillberg, 1998; Green et al., 2004; Msall et al., 2000; Mukaddes et al., 2007; Peterson et al., 2000). Furthermore, disorders such as ROP and bilateral optic nerve hypoplasia have been linked to general social impairments (Ek, Fernell, & Jacobson, 2005; Msall et al., 2000). This may suggest common underlying neurological mechanisms giving rise to both visual and social limitations. However, any information about the prevalence of autism in children with and without blindness should be interpreted with care, because diagnosing autism in children with blindness can be problematic.

From an anatomical perspective, neural mechanisms related to ocular-plus blindness may be involved in ToM development as well. The optic tract passes through the subcortical area of the limbic system, known for its relation to emotional behavior (Kahle & Frotscher, 2003). Studies of young infants with perinatal problems report serious damage to thalamic and subcortical brain areas that are important to both visual and emotional processing (Ricci et al., 2006; Volpe, 2009). Parts of the limbic system, in particular the amygdala and thalamus, are strongly interconnected and have massive projections to the medial prefrontal cortex. This aspect of the limbic system is associated with consciousness and ToM (Frith & Frith, 2003; Kobayashi, Glover, & Temple, 2007) and the superior temporal sulcus, which can be related to recognition and interpretative abilities (Perner & Aichhorn, 2008). Though adults with congenital blindness have been found to utilize the same ToM network as sighted individuals (Bedny et al., 2009), it remains unclear whether ocular and ocular-plus blindness affect ToM competence to the same extent.

In sum, the lack of visual and social experiences may not be the only or main cause for delayed ToM development in
children with congenital blindness. Delays in the development of ToM may be more specific to children with ocular-plus blindness than those with ocular blindness. In the current study, using data from Asbrock’s (2008) research, we investigated whether, compared with sighted children, ToM delays are more pronounced in children with ocular-plus or ocular blindness. The measures used rely exclusively on tactile and auditory experiences (Asbrock, 2008; Brambring & Asbrock, 2010). If ToM delays in children with blindness are mainly due to the absence of visual and social experiences, then we would expect to find no significant differences between the performances of children with ocular-plus and ocular blindness. However, if ToM abilities are delayed in children with ocular-plus blindness but not with ocular blindness, this would substantiate the hypothesis that common neural mechanisms involved in visual, as well as mental, processing influence ToM development more than the visual and social experiences.

Method

Participants

Due to the low prevalence of congenital blindness in general, and ocular congenital blindness in particular, children with blindness were recruited through regular and special schools in two countries: Germany and the Netherlands. Congenital blindness was confirmed by medical reports and professional partners of the cooperating institutions for visually impaired children. Further selection criteria for all participants were: no autism spectrum diagnosis (or no suspicion of autism), no cognitive delays, and typical memory skills. Out of an original sample of 49 children with congenital blindness, 18 children were not included in the final analyses due to later onset of blindness (n = 3), below-average language abilities (n = 3) or memory skills (n = 3), or unknown causes of blindness (n = 9). A final sample of 31 children with congenital blindness, 19 girls and 12 boys from Germany and the Netherlands, aged 4 to 9 years old (M = 7.1, SD = 1.4), were divided into two groups. The ocular-plus blind group (n = 22) included children with ROP (n = 5), Leber’s congenital amaurosis (n = 13), Norrie’s disease (n = 2), optic nerve hypoplasia (n = 1), and optic nerve atrophy (n = 1). An ocular blind group (n = 9) included children with microphthalmus (n = 6), congenital cataracts (n = 1), and infantile glaucoma (n = 2). All participants with blindness had no additional impairments, had no problems with language ability and memory span, and were integrated in mainstream (pre)school systems. A typically developing control group included 103 children without visual impairments, 53 girls and 50 boys, aged 4 to 7 years old (M = 5.1, SD = 0.5), who showed no sensory or cognitive impairments, no autism spectrum disorders, and adequate cognitive skills (see Table 1 for participant details). The study was approved by the medical ethical committee of the University of Bielefeld and was carried out according to the standards of the Declaration of Helsinki (2000). Informed consent was obtained from all families.

Material

ToM test

The ToM test used in this study included nine tactile and auditory first-order false-belief tasks, which were
specifically constructed for children with blindness, including material and simple actions that children were familiar with through their own tactile experiences (Brambring & Asbrock, 2010). All tasks involved simple actions and could be categorized as spatial, auditory, or tactile. The first spatial task (Task 1) was modeled on the original visual Smarties tasks created by Perner, Frith, Leslie, and Leekam (1989), and included a hamburger box that contained a sock. After discovering its content, the child was asked to predict what his or her friend would say about the content of the closed box (McAlpine & Moore, 1995). The second spatial task (Task 2) included a small closet with six drawers, which alternatively contained a toy shark or a toy dinosaur, except for the last drawer, which contained a box. Before opening the last drawer, it was checked whether children understood the repetitive content. Finally, after discovering the content of the last drawer, the child was asked what his or her friend would think was in the last drawer before opening it.

The first auditory task (Task 3) was an audio recording of a person counting from 1 to 5. The CD was then paused and the child was asked how he or she thought the CD would continue. When the CD was played again, a telephone ring was heard rather than a “6.” Children then were tested on their perspective-taking ability by asking what their friend would think would follow the first five counted numbers they heard. The second auditory task (Task 4) included a set of five small wooden stairs with buttons on each step that produced sounds alternating between a loud and a soft tone. The sixth step included an unexpected siren, and children were asked to predict what a friend would think would be the last tone. The third auditory task (Task 5) included a read-aloud story about Person A leaving the room and Person B changing locations. The child was then asked to predict where Person A would look for Person B.

The tactile texture task (Task 6) included a horizontal shelf with six compartments that could be touched. The first five compartments alternated between rugged and smooth materials. The last compartment was filled with soft stuffing. The child was asked memory questions.

<table>
<thead>
<tr>
<th>Table 1</th>
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<tbody>
<tr>
<td>Characteristics of the participants.</td>
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<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Ocular-plus blind</th>
<th>Ocular blind</th>
<th>Sighted</th>
</tr>
</thead>
<tbody>
<tr>
<td>n</td>
<td>22</td>
<td>9</td>
<td>103</td>
</tr>
<tr>
<td>Gender (female or male)a</td>
<td>11/11</td>
<td>8/1</td>
<td>53/50</td>
</tr>
<tr>
<td>Chronological age (mo)b</td>
<td>81.7 (17.9)</td>
<td>92.7 (13.0)</td>
<td>61.0 (6.1)</td>
</tr>
<tr>
<td>Short-term memory (K-ABC)c</td>
<td>10.7 (2.0)</td>
<td>10.6 (1.8)</td>
<td>9.2 (2.3)</td>
</tr>
<tr>
<td>Verbal skillsd</td>
<td>57.9 (5.8)d</td>
<td>54.8 (6.5)d</td>
<td>–</td>
</tr>
<tr>
<td>Nationality (German or Dutch)</td>
<td>14/8</td>
<td>8/1</td>
<td>103/0</td>
</tr>
<tr>
<td>Premature birth</td>
<td>12</td>
<td>7</td>
<td>–</td>
</tr>
</tbody>
</table>

a No difference between gender rates in groups, \( \chi^2 = 5.54, df = 2, p = .09. \)
b No difference between ocular-plus and ocular blind children, \( F(1,29) = 2.7, p = .11; \) difference between ocular-plus blind, ocular blind, and sighted children, \( F(2,131) = 80.16, p < 0.001. \)
c No difference between ocular-plus and ocular blind children, \( F(1,29) = .01, p = .92; \) difference between ocular-plus blind, ocular blind, and sighted children, \( F(1,131) = 4.93, p < 0.01. \)
d No difference between ocular-plus and ocular blind children, \( F(1,20) = 1.35, p = .26. \)
and was asked to predict how a friend
would think the last compartment would
feel.

The first tactile form task (Task 7) was
a toothbrush that had a spoon on its other
side. The experimenter held the spoon
part in her hand, so the child could only
feel the toothbrush. The child had to guess
what was in the hand of the experimenter,
followed by memory and perspective-
taking questions. The second tactile form
task (Task 8) included five bags with ob-
jects, alternating between a die and a bris-
tle. A sixth bag included an unexpected
object—a pacifier. Memory and perspective-
taking questions were asked about a
friend’s expectation of the content of the
sixth bag. The third task (Task 9) in-
cluded three objects that could contain
another object: a basket, a box, and a
wallet. Together with one of their family
members, children put a coin in one of the
containers. In the absence of the family
member, the child and the experimenter
moved the coin to another container. The
child had to predict where the parent
would look for the coin. Brambring and
Asbrock (2010) provide a full description
of this ToM test. The reliability of this
test has been found to be acceptable both
in the present study (a = .78) and in a
previous study (a = .85; Asbrock, 2008).
Furthermore, the test is unrelated to mem-
ory and language skills (Brambring et al.,
2010).

Short-term memory test
The number-recall test, a subtest of the
Kaufman Assessment Battery for Chil-
dren (K-ABC; Kaufman & Kaufman,
1983), was administered to measure
short-term memory. This auditory task is
commonly used with children with blind-
ness without adaptations, and has shown
excellent validity and reliability (Kiese-
Himmel, 2007).

Language test
Verbal abilities of 3- to 5-year-olds were
assessed with memory tasks for sentences
and phonological working memory tests
from the German Language Develop-
ment Battery (SETK 3-5; Grimm, 2001).
The SETK 3-5 has shown discriminant
validity with the standardized nonverbal
K-ABC (Melchers & Preuss, 1992) and
with the Viennese Development Test for
children (Kastner-Koller & Deimann,
1998). From 6 years old, children were
administered a productive test from the
German Basic Competencies for Reading
and Writing (Basiskompetenzen für Lese-
Rechtschreibleistungen; Stock, Marx, &
Schneider, 2003), in which subjects had to
identify pseudo words presented auditorily,
and the Imitating Grammatical Structures, a
subtest of the German language develop-
ment task (Heidelberger Sprächentwick-
lungstest; Grimm & Schöler, 1991). Both
measures have shown good reliability and
validity in typically developing children
(Ptok & Buller, 2006). The Dutch children
received the Wordform Production task, a
comparable subtest of the Dutch language
battery (De taaltest voor kinderen), which
has shown excellent test–retest reliability in
typically developing children (van Bon &
Hoekstra, 1982). All scores on the language
tasks were converted into standardized t
scores.

Procedure
Prior to testing, a questionnaire was sent
to families, teachers, and professional coun-
selors to ensure that, according to them,
their children had no autism spectrum
diagnosis, had no cognitive delays, and had adequate memory skills. All children were tested in familiar environments at home or school. Each session began by ensuring that all children could name the objects, the auditory or tactile properties of the stimuli, and the features of the different locations. Each child was given a short warm-up task to familiarize him or her with the material. All sessions were videotaped and transcribed. Experimenters received extensive training before they tested the children.

**Scoring**

Following the scoring procedure of Brambring et al. (2010), all tasks with verbal information included control questions to check the child’s recall of the information, and a check of correct naming of the identity, location, and surface of objects. Each test question was preceded by a memory question and a temporal cue, which focused the child on what he or she thought was the correct answer. Perspective-taking questions were correct if the children predicted that their friend would give a similar answer as they gave themselves. Responses were incorrect when the child referred to an unexpected outcome. Each correct response was awarded 1 point, and a total score ranging from 0 to 9 was calculated based on the nine perspective-taking questions.

**Results**

Differences between the summed ToM scores in children with ocular-plus blindness, children with ocular blindness, and sighted children were analyzed with an ANCOVA, controlling for children’s age, gender, and short-term memory. A main effect of diagnosis was observed, $F(2,125) = 8.24, p < 0.001, d = .58$.

Planned contrasts showed an expected worse performance of children with ocular-plus blindness ($M = 5.72, SD = 3.57$) than both sighted children ($M = 7.63, SD = 1.28; t(125) = -3.86, p < .001, d = .79$) and children with ocular blindness ($M = 8.33, SD = .71; t(125) = 1.99, p < .05, d = .41$). However, no difference in ToM scores was found between children with ocular blindness and sighted children, $t(125) = .48, p = .63$.

To control for possible differences in ToM scores between the German- and Dutch-speaking participants, an additional analysis was conducted that included only German children, sighted or blind. The same pattern of results emerged, that is, a main effect of diagnosis, $F(2,116) = 7.94, p < 0.001, d = .58$, and differences on TOM scores between children with ocular-plus blindness and sighted children, $t(116) = -3.79, p < .001, d = .78$, and between children with ocular-plus and ocular blindness, $t(116) = 2.22, p < .05, d = .45$, but no difference on ToM scores between children with ocular blindness and sighted children, $t(116) = .56, p = .58$. In addition, using a Welch $F$ ratio to control for the unequal group sizes confirmed the previously described contrasts.

**Discussion**

The study presented here showed that ToM abilities of children with ocular-plus blindness were delayed compared with those of sighted children. The ToM abilities of children with ocular blindness did not diverge from those of sighted children. Moreover, the ToM performances of children with ocular blindness were significantly better than those of children with ocular-plus blindness. Although replication
is needed before strong conclusions can be made, these tentative findings suggest that ToM development in children with congenital blindness could be related to factors other than the loss of a sensory function and the lack of visual, social, and communicative experiences (Bedny et al., 2009). Although additional studies should rule out whether the two groups of blind children were distinct on other external factors, ToM development may also be related to neuronal mechanisms involved in ocular-plus blindness; for example, the plasticity mechanism in which the retina has an important role, as described by Sugiyama et al. (2008), or, much broader, the mechanisms involved in ciliopathy in general (D’Angelo & Franco, 2009), which also involves retinal development. The current results cannot be explained by differences in age, gender, or verbal abilities of our participants. Rather, optic neural pathway involvement may be an important clue in the development of ToM deficits in children with congenital blindness. This suggestion is consistent with earlier indications of a vulnerability in ocular-plus blind groups for developmental setback regressions (Cass, 1996), with the association between the Lebers congenital amaurosis and autism (Rogers & Newhart-Larson, 1989), or with ROP and autism, the last most probably mediated by brain damage and largely independent of the blindness per se (Ek et al., 1998).

The current findings seem to suggest that an important clue for ToM development may be found in the overlapping subcortical brain areas for visual and mental (social-emotional) functioning. Before such a suggestion can be offered, however, two alternative explanations for the ToM deficit in children with ocular-plus blindness should be considered. First, it is feasible that children with ocular-plus blindness more often show a comorbid autism spectrum diagnosis (Mukaddes et al., 2007). In our study, however, this argument may tentatively be dismissed, because children with blindness and comorbid autism spectrum diagnoses were excluded from the current study; nevertheless, it should be acknowledged that diagnosing autism in children with blindness is very complicated. It should be noted that very few earlier studies on children with blindness have explicitly controlled for autism, and thus may, in fact, report findings that are due to autism rather than blindness, as these children may also have been autistic (and delayed on ToM abilities) (Hobson, 2002). It is an important challenge for future studies to disentangle the confusing contribution of blindness and autism in the explanation of the social problems of children with (ocular-plus) blindness. Genetic research in autism, especially that concerning ciliary genes influencing many aspects of organ development, including the retina, provide important clues for these future studies (D’Angelo & Franco, 2009).

It should also be emphasized that ocular-plus congenital blindness may be due to a wide variety of factors. In our study, we differentiated five types of ocular-plus blindness, but it has proven very difficult to gain large enough samples of children with congenital blindness for analyzing these subgroups, even when collecting data across several countries, because the ocular group is small in number and difficult to find. Future studies will have to confirm, and possibly elaborate on, the ToM delays of children with specific types of ocular-plus congenital blindness.
Therefore, it may be premature to search for common neurological mechanisms, and more research and hypotheses are needed on different types of ocular-plus blindness—if possible, combined with ciliary genetic information, which is more and more available—using adequate assessment tools for both autism and ToM development (and offered social experiences).

Additional limitations of the study include a lack of assessment data on the presence of autism in the children with blindness. In addition, we relied on a relatively small group of children with blindness and a large comparison group. Analyzing more children with blindness may provide a better picture of the variance within this group. However, additional analyses did confirm the current results when controlling for unequal group sizes.

It is not yet possible to offer an explanation on the nature of the suggested common neurological mechanisms underlying ToM delays and congenital blindness. The current findings do suggest a connection between optic neural pathway involvement and ToM development of children with ocular-plus blindness, whereas children who have ocular blindness may be relatively unaffected by ToM delays. Considering the low number of cases of ocular blindness found in this large group of children with congenital blindness, future studies are required to confirm our findings.

References
Declaration of Helsinki. 52nd WMA General Assembly, Edinburgh, Scotland, October 2000.


