ISSN: 2083-389X eISSN: 2084-3127

F Literature analysis/search G Funds collection

# **CHILDREN WITH COCHLEAR IMPLANTS:** LITERATURE REVIEW Contributions: A Study design/planning B Data collection/entry C Data analysis/statistics D Data interpretation E Preparation of manuscript

Magdalena Krysztofiak<sup>1,A-B,D-F</sup>, Agnieszka Pluta<sup>2,1,A,D-G</sup>

- <sup>1</sup> Faculty of Psychology, University of Warsaw, Warsaw, Poland, Poland
- <sup>2</sup> Bioimaging Research Center, World Hearing Center, Institute of Physiology and Pathology of Hearing, Warsaw, Poland

THEORY OF MIND DEVELOPMENT IN DEAF

Corresponding author: Agnieszka Pluta, Bioimaging Research Center, World Hearing Center, Institute of Physiology and Pathology of Hearing, Mokra 17, 05-830, Kajetany, Poland; email: a.pluta@ifps.org.pl

#### **Abstract**

Theory of mind (ToM) is the mental capacity that allows us to represent the mental states (beliefs, desires, emotions) of other people, infer them from situational cues, and predict their behavior. According to the standard view, the most important milestone in ToM development - the ability to pass the false belief test (FBT) - emerges around four years of age. FBT requires one to understand that the beliefs of others are independent from reality and from one's own beliefs, and that their behavior can be predicted by their mental states. Previous research has indicated that deaf and hard-of-hearing children born into hearing families (DoH) are at risk of delayed ToM development due to restricted social interactions. However, these findings are unclear for DoH children who receive cochlear implants (CIs) and whose hearing is partially restored. In this review, we summarize research on the development of ToM in DoH children with cochlear implants (CIs). We describe how language (vocabulary and syntax) influences ToM. Finally, we discuss the nature of social interactions that facilitate ToM development in children with typical hearing as well as in DoH children with CIs.

Key words: theory of mind • deafness • cochlear implants • social developmen

# ROZWÓJ TEORII UMYSŁU U DZIECI GŁUCHYCH I NIEDOSŁYSZĄCYCH BĘDĄCYCH UŻYTKOWNIKAMI IMPLANTÓW ŚLIMAKOWYCH. PRZEGLĄD LITERATURY

#### Streszczenie

Teoria umysłu (theory of mind, ToM) to zdolność poznawcza, która pozwala reprezentować wewnętrzne, nieobserwowalne stany psychiczne (przekonania, pragnienia, emocje) innych ludzi, wywnioskować je ze wskazówek sytuacyjnych i na ich podstawie przewidywać zachowania innych osób. Zgodnie ze standardowym poglądem kamień milowy w rozwoju ToM – zdolność do przejścia testu fałszywych przekonań (false belief test, FBT) – dzieci osiągają w wieku około czterech lat. FBT wymaga zrozumienia przekonań innych osób jako niezależnych zarówno od rzeczywistości, jak i od prawidłowych przekonań obserwatora oraz zrozumienia, że zachowanie ludzi można przewidzieć na podstawie ich stanów psychicznych. Wcześniejsze badania wykazały, że dzieci niesłyszące i niedosłyszące urodzone w rodzinach słyszących (deaf of hearing parents, DoH) są narażone na opóźnienie w rozwoju ToM z powodu ograniczonych interakcji społeczno-komunikacyjnych. Jednak wyniki te są niejasne w przypadku dzieci DoH, które otrzymują implanty ślimakowe (CI), zatem ich słuch może być częściowo przywrócony. W tej recenzji prezentujemy badania dotyczące rozwoju ToM u dzieci DoH, które są użytkownikami implantów ślimakowych (CI). Opisujemy, jak język (słownictwo i składnia) wpływa na rozwój ToM. Na koniec omawiamy naturę interakcji społecznych, które ułatwiają rozwój ToM u dzieci z typowym słuchem, a także u dzieci DoH będących użytkownikami CI.

Słowa kluczowe: teoria umysłu • głuchota • implanty ślimakowe • rozwój społeczny

#### Introduction

Typically, at around age four, children undergo a striking developmental change in the way they perceive their social world. They start to understand that thoughts and feelings shape human behavior, enabling them to "enter the community of minds" [1]. This ability is called Theory of Mind (ToM) and is considered by developmental psychologists to be the "most important development in early childhood social cognition" [2]. It allows the child to understand that the knowledge and perspective of others can be different from their own [3]. Therefore, ToM is important for successful social functioning and affects many aspects of social life, including peer popularity (e.g. [4]). Moreover, it

is worth noting that poor social perception poses adverse risks to mental health.

ToM ability progresses in typically-developing children according to a particular sequence based on age [5]. Around the age of four, children undergo a conceptual change and start to explicitly reason about the mental states of others, which manifests in the ability to pass the traditional false belief test (FBT) [3,6]. The FBT assesses whether the child can predict the behavior of another person, even if the other person is acting on a false belief that differs from objective reality and the child's own knowledge. The FBT is therefore considered to be the "litmus test of Theory of Mind" [7]; however, there are also other methods that assess

ToM ability in infants, children, and adults [8,9]. Furthermore, ToM development is a gradual process, reflected in several milestones that children reach in infancy and later childhood [10].

ToM is a multifaceted concept, requiring the integration of a number of components. It can be broken down into at least two subcomponents: affective ToM (inferring the feelings of another person) and cognitive ToM (understanding the beliefs of another person), which comprise both distinct as well as overlapping neural processes [11]. It has also been suggested that there are additional abilities connected with ToM development – in particular, language skills [12]. Furthermore, the quality of conversations about mental states with caregivers has also been suggested to be an important factor in ToM development [13]. Therefore, deaf children of hearing parents (DoH) - who might have less access to spontaneous mentalistic-type conversations and who frequently show delays in language acquisition – are at risk of slower ToM development [14,15]. At the same time, medical and technological advances provide more options for the treatment of children born with profound hearing loss. These children can benefit from cochlear implants (CIs), which, by restoring a degree of hearing, can allow them to acquire spoken language [16,17]. However, in terms of conversations with their hearing parents, deaf children with CIs might have qualitatively different experiences compared to their peers with typical hearing, since their parents are likely to simplify conversations to adjust to the assumed cognitive ability of their child [18]. Hence, deaf children with CIs can still experience limited conversational input early in life, they can manifest difficulties in some language tasks, and possibly experience delayed ToM development [14].

This line of argument highlights that specific features of the early social interactions with caregivers, along with language development, have important effects on the subsequent development of social cognition, but this remains underexplored in children with hearing loss who use CIs.

The main aim of this article is to review the existing literature on ToM development and assess the environmental factors that might affect mentalizing in profoundly deaf children of hearing parents (DoH) who use cochlear implants. We decided to focus exclusively on DoH children with CIs who use oral language as their preferred mode of communication. The reason is that previous research has shown that deaf children of deaf parents, who communicate in sign language, develop ToM in line with the timetable of children with typical hearing [19,20] due to a typical linguistic experience and access to conversational interaction with their caregivers. However, studies on prelingually DoH children who use CIs are still scarce and provide inconsistent results regarding ToM competencies.

The first section outlines the concept of ToM, including theoretical and methodological frameworks behind its development, ending with the relationship between ToM and language. The following section focuses on profoundly deaf children who use CIs, further expanding on the relationship between ToM, language, and conversation. Finally, we set out research questions that need to be explored by future studies on social cognition development in DoH.

# Theoretical and methodological framework of ToM

There are various theoretical accounts that explain the development and mechanisms of ToM and they focus on different aspects of this ability.

According to "theory-theory" accounts, ToM development is similar to building a scientific theory. Children are compared to scientists who, through learning and experimentation, reorganize their theories in order to explain the causal relationships between mental states and behavior [21]. Simulation accounts of ToM posit that it is rooted in a mechanism of adopting the perspective of others and imagining their mental states, relying on a system of mirror neurons [22]. Modular accounts of ToM, such as that of Leslie et al. [23], argue that there is an innate mechanism corresponding to the ability to reason about mental states.

However, ToM could also be viewed in social constructivist terms, as proposed by Carpendale and Lewis [24]. Their account emphasizes the role of social interactions and communication in ToM development. They propose that "children gradually construct social understanding through the regularities they experience in interacting with others" [24]. They also outline the role of language competency and the quality of conversations in the development of a child's social understanding. Their account corresponds with studies of deaf children of hearing parents, as the delay in development of ToM in this group can be explained by limited conversational input [14].

There are several methods available to study ToM. Task variations might depend on the age of the participant or the ToM system being studied (explicit or verbal; implicit or automatic, nonverbal), since ToM can be divided into two systems which differ in terms of cognitive control and awareness [25]. Some measures also allow to assess how the parents perceive their child's social understanding [26,27]. The most common method of assessing explicit ToM is the false belief test. One version of this test is the change-of-location task [6]. This paradigm was devised by Wimmer and Perner [28] in the form of a puppet play, and was later modified by Baron-Cohen et al. [29] during their research with children with autism spectrum disorder. In this task, participants observe a scene with two protagonists named Sally and Anne. Sally places a marble in a basket and leaves the room. During her absence, Anne transfers Sally's marble to a box. When Sally returns, the participants are asked a question concerning the beliefs of the protagonist: "Where will Sally look for her marble?" [29]. In order to pass the Sally–Anne task, participants have to acknowledge the protagonist's false belief and point to the previous location of the marble (the basket). Studies have shown that typically developing children are able to successfully pass this test around the age of four [6]. Children aged three and younger fail this test and point to the actual location of the object, not accounting for the lack of knowledge of the protagonist [6].

In addition to the change-of-location task, there are also other methods of assessing false belief understanding, such as the unexpected contents tasks (the "Smarties" box task) or unexpected identity tasks [30]. In the unexpected

contents task, the child is shown an object with something unexpected inside (e.g., a box of sweets filled with pencils). Next, the child is asked what a child who had not been shown the unexpected contents would say is in the box. To succeed, children have to understand that the other child could not know the truth and would therefore have a false belief. Similarly, in the unexpected identity task, the child is also presented with a surprising object, but the properties of the object are unexpected (e.g., a sponge that looks like a stone) [31]. These tasks require a direct description of another person's mental state and consequently engage an explicit and intentional system of ToM.

Although most ToM studies focus on children, this ability can also be studied in adults. There are special paradigms adjusted for adult studies that also enable more ecological assessments of ToM and focus on the understanding of more advanced concepts, such as metaphors or faux pas [9].

Finally, some measures also assess how caregivers perceive their child's understanding of mental states [26,27]. For example, the Theory of Mind Inventory has been used in studies of typically developing children as well as of children with autism spectrum disorder or children with hearing loss to identify challenges specific to these populations [26,27]. The advantage of this type of measure is that they include everyday manifestations of ToM and take into account parental observations and expertise regarding their child.

Thus, reasoning about mental states can be studied in various ways and in a wide range of participants of different ages, giving measures of the developmental stage of ToM ability.

#### **Development of ToM**

Development of ToM can be viewed as a gradual process [5,24]. Passing the standard false belief test around the age of four is a pivotal point for ToM; however, there are several social development milestones that children reach before this point. These skills include joint attention (which is the shared focus of two individuals on an object), the ability to recognize others' emotional states, the knowledge that people act according to their intentions, understanding the causes and consequences of emotions, and pretend play. Moreover, social development also continues as children progress into middle and later childhood when they develop more advanced ToM concepts and can reason about mental states in a more refined manner [5,10].

Infants already demonstrate a considerable interest in social interaction. Even 6-month-olds distinguish between the motion of inanimate and animate objects and "interact dyadically" with objects and people [32]. Around their first birthday, infants undergo what Tomasello (e.g. [32]) has described as a "nine-month revolution", as at this stage they start to engage in joint attention. Joint attention refers to the ability of an infant to share a focus on an object or event with an adult in a triadic interaction. For example, a child might point to a toy on the table to draw the parent's attention or follow the parent's gaze to the toy. Therefore, joint attention is based on the understanding that both the infant and adult share a focus on the object, but from different perspectives [3]. Children develop pretend play around

18-24 months (e.g. [33]). Leslie [33] argued that the ability to engage in a "shared pretense" (e.g., understanding that someone imagines that a banana is a telephone) relies on the same representational mechanism as later understanding of explicit false beliefs. Subsequently, toddlers and young preschoolers are able to understand desires and intentions, which are precursors to false belief understanding [5]. Finally, around the age of four, children start to understand that someone can hold false beliefs about the world; they can then pass the standard false belief test (FBT), which is considered critical in ToM development [6]. There is also evidence that children pass the FBT around the same age across different cultures [34]. The critical age for false belief understanding is a matter of some debate, as the test itself may be too difficult for younger children due to linguistic and executive demands. Thus, simplified versions of the FBT can improve a child's performance [8]. However, a meta-analysis by Wellman et al. [6] supported the claim that a conceptual change in the understanding of beliefs occurs around preschool age, questioning the view that it only appears around this age due to test difficulty. Currently, there is still discussion about whether success on simplified tasks represents early false belief understanding or a distinct competence [3]. Later, children are able to understand that someone can have a belief about another person's belief (known as a second-order belief) [35]. Finally, during later childhood children are able to understand irony, metaphors, and faux pas, which can be described as "advanced ToM" [10,36].

The emergence of ToM concepts during subsequent stages of life supports the idea that this ability is multifaceted and not limited to false belief understanding. Furthermore, this developmental process can be viewed in a much broader context. Linguistic and family factors, such as the child's level of language competency or the frequency and quality of talk about mental states in parent-child interactions, may contribute to variance in ToM [12,13]. Furthermore, progress in ToM might be altered or delayed in populations with atypical language development [5]. Several studies have highlighted the case of deaf children of hearing parents, as these children can display different trajectories of acquiring ToM concepts - mainly false belief understanding - in comparison to their peers with typical hearing (e.g. [5,14,37,38]). These results can be explained in terms of the relationship between language and ToM, which will now be discussed.

## Language and ToM

Bretherton et al. [39] have previously emphasized the connection between the emergence of explicit ToM and a child's ability to verbally refer to mental states, suggesting the importance of language development for ToM. Since then, studies have confirmed the existence of a relationship between language and ToM ability – in particular false belief understanding [12,40]. The role of language in ToM development can also be conceptualized in terms of social constructivist accounts, since the quality of everyday conversations has been shown to contribute to a child's later understanding of mental states [13].

There is ongoing discussion on the nature of the relationship between language and false belief understanding. As summarized by Farrar et al. [40], some studies emphasize the role of general language competency, including syntactic and semantic ability (e.g. [41,42]), while others indicate that there are specific aspects of language that might be more important for false belief understanding, such as complement syntax (e.g. [43]). According to the second account, complementation is necessary for the representation of false beliefs [43]. Understanding special structures such as "Sally thinks that the marble is in the basket" might be especially important for false belief understanding, as they reflect a subjective perspective of the situation [40,44]. However, other studies argue that for typically developing children, complementation is not always uniquely related to false belief understanding and point towards the role of general language measures such as semantic or syntactic abilities [40]. General syntax, which includes various syntactic forms, enables the child to represent and track the relations and changes in the classic change-of-location task, thereby contributing to false belief understanding [42,45]. For example, de Rosnay et al. [41] measured children's syntactic understanding with the Test for the Reception of Grammar and found that it was significantly related to their performance on the FBT. Also, according to a meta-analysis by Milligan et al. [12], syntax, among other language measures, was significantly related to false belief understanding.

Moreover, in line with social constructivist accounts, conversations with parents might also be important for the child's ToM development [13,24]. Devine and Hughes [13] found that talk about mental states predicted the child's false belief understanding after one year. Taumoepeau and Ruffman [46] suggested that parents could "scaffold" their child's mental state understanding through the use of mentalistic language. Parental input can be assessed by self-report (e.g. [47]) or, more qualitatively, through the analysis of real-life conversations (e.g. [38,44]). Peterson and Slaughter [47] devised the Maternal Mental State Input Inventory (MMSII) in order to measure a mother's preference for explanations of everyday interactions (e.g., preparing a birthday surprise). They found that mothers who preferred more elaborate explanations with references to mental states had children who exhibited higher false belief understanding than their peers (e.g., by explaining that dad will be surprised with his birthday present because he does not know what is inside the box). Tompkins [44] studied conversations between parents and children during shared storybook reading and also found that references to mental states were positively related to the child's performance on the FBT. Furthermore, a recent meta-analysis of family correlates of ToM by Devine and Hughes [48] also confirmed the impact of parental mental state talk on the development of children's false belief understanding.

Finally, the relationship between language and false belief understanding can be elucidated by studying specific populations in which language development differs from the typical model, such as deaf children raised by hearing parents [49]. The experiential view of cognitive development assumes that the child's language skills affect how much they can access and understand conversations that refer to the mind. Consequently, delayed acquisition of language – which is frequent in deaf children of hearing parents – might hamper the later development of ToM [14,20]. On the contrary, deaf children who use sign language and whose parents are

native signers do not show delays in explicit ToM, as their experience in conversations has not been constrained [20].

# Language and conversation in deaf children with CIs

In deaf children of hearing parents, the hearing loss can have a significant impact on speech, and indirectly affect academic achievement or other aspects of life, such as social functioning [20,50-53]. However, the spread of hearing screening programs has contributed to earlier diagnoses and interventions for hearing loss [54,55]. Furthermore, the invention of cochlear implants (CIs), an electrical hearing prosthesis that provides access to environmental sounds and spoken language, has provided the opportunity for habilitation of children with profound hearing loss [56]. CIs provide access to sensory input, improving the perception of sound and acquisition of spoken language, leading to a larger proportion of children approaching the spoken language levels of their peers with typical hearing [51,52,56]. Improvements in communication abilities after implantation are also reported to positively affect the child's relationship with family members and peers [57].

Nevertheless, it should be borne in mind that it takes substantial time for young children or infants to adjust to a CI as an aid to hearing - and so CIs may not be sufficient to overcome slower development in several aspects of spoken language (e.g. [38,58]). Indeed, it has been reported that deaf children with CIs experience delays in various domains of spoken language development, including grammar and pragmatic skills [59] and lexical comprehension [60]. The variability in spoken language outcomes in children following cochlear implantation remains quite high. Previous studies have shown that spoken language performance in children following cochlear implantation is influenced by the age at implantation and access to conversations [58]. The main idea behind early cochlear implantation is that children who are implanted earlier have better spoken language outcomes because they will have experienced a shorter period of auditory deprivation and had more opportunities to engage in vocal interaction with their caregivers than children who are implanted later [61].

Geers and Sedey [52] assessed children's spoken language skills first during elementary school and then at high school, finding that deaf children who had earlier implantation – and who had thus experienced a shorter period of auditory deprivation – had better spoken language skills later in life. However, a number of those children still encountered difficulties in connected discourse and abstract reasoning tasks.

The importance of early implantation for spoken language acquisition can be explained by the hypothesis of sensitive or critical periods in language development, an idea supposing that there is a biologically determined period of life when language can be acquired more easily and after which language becomes increasingly difficult to acquire. Although it is difficult to precisely identify the optimal time for language acquisition, there is a general consensus that earlier implantation is better than later implantation, as it minimizes the gap between the child's chronological age and their linguistic age [58,61]. However, despite conflicting results of studies

comparing language development in deaf children who received implants before the age of 1 to those who received CIs between 1 and 2 years of age (which suggest a small or nonexistent advantage in the case of younger children), there are consistent results showing that deaf children implanted later than 2 years of age lag behind their peers with typical hearing in language development. We can therefore conclude that the period before 2 years of age is critical for language development [62].

Over the years, the consensus regarding the critical period for cochlear implantation has changed – from up to 5 years previously to 12 months of age now [58]. However, in a recent review and meta-analysis, Duchesne and Marschark [58] emphasized the variability in studies examining the relationships between age at cochlear implantation and vocabulary and grammar outcomes. While many of the reviewed studies found significant relationships between early implantation and better language outcomes, the authors conclude that other factors, including the family environment, should also be considered when trying to explain the observed variability.

The relationship between CIs and language might also depend on the language domain of interest, as some language skills might be more difficult than others. For example, Geers et al. [63] compared the performance of 5- and 6-year-old deaf children with CIs to their peers with typical hearing on various language measures. They found that syntactic tasks were more difficult than vocabulary tasks. Conversely, Boons et al. [64] did not find any strong or weak aspects of language development in school-aged deaf children with CIs compared to their peers with typical hearing. However, after a qualitative analysis of systematic errors in language tasks, they found that deaf children with CIs made more severe errors in syntactic tasks than the control group.

There could also be differences in terms of the quality of conversations displayed by parents of deaf children with CIs and parents of children with typical hearing, especially when it comes to the discussion of topics that can't be seen or pointed to [18]. Conversations are important for language acquisition, but they also affect the development of social perceptions, since parents tend to guide their children's perception of the environment and improve their reasoning about the cognitive and emotional states of others. For instance, a study by Morgan et al. [18] found that hearing mothers of deaf infants (using CIs or hearing aids) engaged in less effective exchanges with their children and made fewer references to mental states than the mothers of hearing infants. The authors hypothesized that hearing parents of deaf children might try to adjust the quality of their conversations to the perceived level of understanding of their child.

Current research on deaf children who use CIs has extended beyond functional communication, as improved language acquisition can also affect social development and ToM abilities [65]. In order to fully understand the relationship between cochlear implantation and ToM, different types of predictors should be included in the studies: age at implantation, level of specific language abilities, and family factors, including the quality of parental conversational input and the parents' perceptions of their child's social functioning.

# ToM development in deaf children with CIs

The development of ToM in deaf children of hearing parents and, more specifically, in DoH who use CIs, has gained increased attention. The majority of studies conducted in the field aim to answer the question of whether deaf children with CIs who are raised by hearing parents struggle with ToM compared to their peers with typical hearing. The results are inconsistent and the underlying mechanisms will now be highlighted.

The first study on ToM development in deaf children of hearing parents was performed by Peterson and Siegal in 1995 [37]. They found that the majority of school-aged signing deaf children (who were raised by hearing parents) failed the standard false belief test. Peterson [14] examined deaf children with cochlear implants separately, making a novel contribution to the study of deafness and ToM. She found that deaf children with CIs performed worse on the FBT in comparison to preschoolers with typical hearing and suggested that deaf children with CIs could have delayed ToM reasoning. Even though CIs improve development in terms of communication and socialization [66], other researchers have also found that deaf children with CIs exhibit delayed ToM development, in particular false belief understanding, in comparison to their peers with typical hearing [67-71]. For example, Yu et al. [71] reported that only 3% of deaf preschoolers with CIs (aged 3-6) succeeded in the false belief task, a dramatically low success rate since the majority of hearing preschoolers pass the false belief task around the age of 5 [6].

Similar results were shown by Ketelaar et al. [67] who demonstrated that 2–4 year-old deaf children with CIs lagged in false belief understanding behind their peers with typical hearing. Nevertheless, the ability to ascribe early ToM concepts (understanding desires and intentions) to others was intact in this group of deaf children with CIs. A different pattern was demonstrated by Meristo et al. [72], who showed that deaf children with CIs did not differ from their peers with typical hearing on the verbal false belief task. However, they failed the implicit false belief task, meaning they were unable to spontaneously anticipate another person's belief.

In contrast, Remmel and Peters [73] did not find differences in ToM ability between deaf children with CIs and their peers with typical hearing. Ziv et al. [74] also reported no difference between a group of 20 deaf children with CIs who used spoken language as their main mode of communication (mean age = 6.6 years; mean age at implantation = 2.5 years) and 23 peers with typical hearing in either affective perspective-taking or in change-of-location false belief understanding. Although the study revealed a relatively high average success rate of deaf children with CIs on tasks measuring different domains of social development, the authors highlighted greater heterogeneity in ToM performance among deaf children with CIs than in children with typical hearing. This means that high rates of variability in ToM skills are widespread in deaf children with CIs even after many years of CI experience.

Delay in false belief understanding might also affect the development of advanced ToM concepts, as suggested by the recent study of Figueroa et al. [75]. They showed that

deaf adolescents aged 12–16 years with CIs had a lower understanding of second- or higher-order beliefs and of understanding multiple perspectives requiring mentalizing. Marschark et al. [69] have even reported differences in advanced ToM in deaf adults with CIs.

Results of studies of ToM in deaf children with CIs are mixed, but the majority of studies report delayed performance on ToM tasks in comparison to peers with typical hearing. Some show that there are partial deficits depending on the task. Few indicate no differences between these groups. However, there are various factors that might contribute to the performance on ToM tasks in deaf children with CIs, such as language skills and access to conversations about mental states (see [49] for a review of ToM and language development in DoH), age at implantation, family correlates, and executive functions. These factors are now described.

Spoken language abilities have been found to be associated with false belief understanding in deaf children with CIs (e.g. [14,43,73]). There are few studies aiming to determine which domains of language account for much of the success in ToM tasks in deaf children with CIs (for a review of the relationship between false belief understanding and language skills, including children who are deaf, see [40]). For example, Schick et al. [20] demonstrated that both vocabulary and comprehension of syntactic complements were significant independent predictors of success on ToM tasks. Remmel and Peters [73] showed a higher correlation between ToM performance and general syntactic proficiency than between ToM score and measures of complement syntax in deaf children with CIs.

In general, research with deaf children with CIs supports the idea that language experience affects ToM development and that delayed language acquisition is the key predictor of hampered ToM development of deaf children with CIs. There are a number of studies demonstrating this relationship (e.g. [14,43,71,73]). For example, Yu et al. [71] showed in a longitudinal study that deaf children with CIs who had more advanced language ability had better ToM growth. However, in the previously mentioned study by Ketelaar et al. [67], the authors demonstrated that despite having the same level of spoken language skills, deaf children with CIs still lagged behind their peers with typical hearing in false belief understanding. Furthermore, there might be a possible confounding effect of linguistic demands on the standard explicit false belief test, as it requires that the child follows the course of a story and answers test questions. The issue of linguistic demands on the standard FBT has also been raised by researchers working on nonverbal (implicit) ToM in infants [8], who proposed that failure on these tasks might be due to processing difficulties. However, de Villiers and de Villiers [43] and Pluta et al. (in press) [76] found that deaf children with CIs had difficulties reasoning about false beliefs even when the linguistic demands of the task were minimized.

These results might also be attributed to limited conversational input early in life, as hearing parents could find it difficult to interact with their deaf child until he or she acquires a sufficient level of spoken language development after cochlear implantation and effective hearing habilitation. Consistently, ToM impairment has not been

reported in deaf children whose parents are native signers and could freely communicate with their child, supporting the hypothesis that limited access to language could impact ToM development in profoundly deaf children who are raised in a hearing culture [49]. Some have also emphasized that better language skills might provide enhanced access to conversations about mental states (e.g. [73]). Peterson [38] proposed certain potential mechanisms underlying delayed ToM in DoH children with CIs. She explained that deaf children are deprived of exposure to mental state conversations until they manage to master enough spoken language to be able to follow their parents' spoken conversations. Subsequently, they can gradually start to acquire receptive and expressive language (words referring to mental states such as "know" or "believe") as well as the syntax (sentential complements and relative clauses) necessary for conversations about mental states [38]. Moreover, a study by Moeller and Schick [77] found that the frequency of maternal references to mental states was related to false belief understanding in deaf children. This resonates with studies of children with typical hearing, which also emphasized the importance of a cognitively and socially stimulating environment for ToM development [13,46].

Sundqvist et al. [68] found that children who were implanted earlier (before 27 months) performed better on emotional ToM tasks than children who were implanted later (after 27 months). However, this finding has not been confirmed in other studies and some have advised that age at implantation should not be treated as the only factor explaining the delay in ToM experienced by deaf children with CIs [65]. Moreover, as described earlier, age at implantation is related to language skills.

A recent review by Marschark et al. [65] suggested that in order to fully understand the relationship between ToM and CIs, other factors, such as family correlates should also be taken into account. Previous studies focusing on children with typical hearing found that the family's socioeconomic status and number of siblings contributed to the child's ToM and false belief understanding [48]. Other factors could also be attachment security or the parent's propensity to be attuned to their child's mental states – thoughts, desires, emotions, and intentions (also referred to as mindmindedness) [48,78].

Another factor that could be important in terms of deaf children with CIs is the quality and quantity of joint attention between the child and caregiver [79]. For example, MacGowan et al. [80] found that joint attention scores between hearing mothers and deaf children aged from 17 to 41 months (including deaf children with CIs) were positively related to the child's social competence as reported by the mother. Interestingly, this relationship was not observed in the hearing dyads. Furthermore, including parental assessment of the child's ToM ability in everyday settings could also help provide further perspectives on the characteristics of deaf children with CIs [26].

Finally, apart from the linguistic, conversational, and family factors, executive functions have also been indicated as a possible factor contributing to the performance on ToM tasks (in particular, the FBT) [81]. A positive relationship between executive functions and ToM development

has been confirmed in children with typical hearing as well as in deaf children with CIs [70].

It is also worth noting that the equivocal results of studies on ToM development in deaf children with CIs might partly result from differences in the recruitment procedures used, since groups are heterogeneous in terms of age at implantation and preferred mode of communication (spoken or signed language) (e.g. [67,73]). Moreover, Ziv et al. [74] highlighted the greater heterogeneity in ToM performance among deaf children with CIs than in controls with typical hearing. This is important, as it suggests that assessment of ToM should be routinely done in deaf children with CIs in order to identify individuals who need tailored interventions with additional ToM training.

#### Interventions to promote ToM in deaf children

Several studies have provided evidence that mentalizing abilities might be enhanced via specific intervention programs designed to focus a child's attention on the mental states of others. This could be achieved by training caregivers to talk more elaborately about past events with their children, or by teaching children the language used to talk about the mental states of others [82]. For example, the training designed by Wellman and Peterson [83], using cartoons with thought bubbles, helped school-aged deaf children understand that different people might have different representational mental states. After the training, children scored higher on false belief scales than did the control groups without training. It was also demonstrated that ToM can be scaffolded with explicit instructions [49] or through using fiction books in order to engage the child in exploring the topic of thoughts and feelings [84]. Additionally, interactions between deaf parents and deaf children could serve as a model for interactions in the population of DoH children. By mimicking deaf parents of deaf children, hearing parents might learn how they can adjust their behavior to effectively adapt to their child's needs in terms of visual input (in particular, prior to implantation when their children have no auditory input). This might facilitate the development of joint attention (prior to CI implantation), which is an important prerequisite of ToM ability [79]. Additionally, closer collaboration between practitioners working with deaf children and researchers is still needed. Beazley and Chilton [85] conducted qualitative interviews with five educators of deaf children (including deaf children with CIs) in terms of ToM development. In some parts of the interviews, practitioners described techniques they were using that could support ToM ability in children, such as book sharing, role play, or "speech" and "thought" bubbles. However, although most participants were familiar with the concept of ToM, they were unsure about its definition or implications for deaf children in their everyday practice. Nevertheless, practitioners expressed their expectations towards future ToM research in terms of supporting their work and strengthening the collaboration.

### **Summary**

Understanding the development of ToM during childhood has practical relevance because success on tests of ToM correlates with many important aspects of social life, including mental health in general. To date, there have only been a few studies that focus on ToM development in deaf children with CIs, and the results of these studies are mixed due to confounding factors (e.g., participants varied substantially in terms of age at implantation and their preferred mode of communication, parental hearing status, or linguistic complexity of the ToM tasks [67,73]).

The majority of these studies report that false belief understanding is delayed in deaf children with CIs. Considering the fact that the protocol for when a child should receive a CI has changed recently (cochlear implantation is now performed in children under 12 months of age [86]), there is a pressing need to conduct studies on children who were implanted early in order to determine how severe ToM delays are (if any) in this unique population.

Studies of deaf children of hearing parents further confirm a positive relationship between language, conversations about mental states, and ToM ability, which has been previously indicated in studies of children with typical levels of hearing [12,13]. Thus, it is not deafness per se, but rather delayed spoken language development and restricted early access to abstract mind-related discourse, that are the key factors explaining ToM delays in DoH [38]. There are also different variables that might explain and/or mediate this relationship, such as the age at which the child started to receive auditory input, level of language ability and access to conversations about mental states, socioeconomic status, and parental education [18,26,65,82].

Furthermore, to the best of the authors' knowledge, no study of deaf children with CIs has yet adopted a social constructivist perspective and investigated the relationship between the quality of social interactions (including the propensity to use mental state talk) and ToM development in DoH children who received their implant in infancy.

Future studies could also further inform rehabilitation programs and provide practical guidelines for therapists and parents of deaf children with CIs. Moreover, based on existing studies on ToM in DoH, clinicians should consider including assessments of mentalizing ability in interventions offered to this population.

# Acknowledgements

This work was supported by the Polish National Science Center (Narodowe Centrum Nauki; Grant No. 2017/25/B/ HS6/01624) awarded to AP. This article is part of the first author's Master's thesis.

### References

- Nelson K, Plesa Skwerer D, Goldman S, Henseler S, Presler N, Walkenfeld F. Entering a community of minds: an experiential approach to 'Theory of Mind.' Hum Dev, 2003; 46: 24–46.
- Astington JW, Edward MJ. The development of theory of mind in early childhood. In: Tremblay RE, Boivin M, Peters RD, editors. Encyclopedia on Early Childhood Development [online at https://www.child-encyclopedia.com]. 2010.

- Tomasello M. How children come to understand false beliefs: a shared intentionality account. Proc Natl Acad Sci, 2018; 115: 8491–8.
- Slaughter V, Imuta K, Peterson CC, Henry JD. Meta-analysis of theory of mind and peer popularity in the preschool and early school years. Child Dev, 2015; 86: 1159–74.
- Peterson CC, Wellman HM. Longitudinal theory of mind (ToM) development from preschool to adolescence with and without ToM delay. Child Dev, 2019; 90: 1917–34.
- Wellman HM, Cross D, Watson J. Meta-analysis of theory-ofmind development: the truth about false belief. Child Dev, 2001; 72: 655–84.
- Bauminger-Zviely N. False-belief task. In: Volkmar FR, editor. Encyclopedia of Autism Spectrum Disorders. New York, NY: Springer; 2013.
- 8. Scott RM, Baillargeon R. Early false-belief understanding. Trends Cogn Sci, 2017; 21: 237–49.
- Turner R, Felisberti FM. Measuring mindreading: a review of behavioral approaches to testing cognitive and affective mental state attribution in neurologically typical adults. Front Psychol, 2017; 8: 47.
- Brüne M, Brüne-Cohrs U. Theory of mind: evolution, ontogeny, brain mechanisms and psychopathology. Neurosci Biobehav Rev, 2006; 30: 437–55.
- Pluta A, Łojek E. Architektura funkcjonalna teorii umysłu.
   Podejście neuropsychologiczne. Warszawa: Wydawnictwa Uniwersytetu Warszawskiego; 2014.
- Milligan K, Astington JW, Dack LA. Language and theory of mind: meta-analysis of the relation between language ability and false-belief understanding. Child Dev, 2007; 78: 622–46.
- Devine RT, Hughes C. Let's talk: parents' mental talk (not mindmindedness or mindreading capacity) predicts children's false belief understanding. Child Dev, 2019; 90: 1236–53.
- Peterson CC. Theory-of-mind development in oral deaf children with cochlear implants or conventional hearing aids. J Child Psychol Psychiatry, 2004; 45: 1096–106.
- Kossewska J. Językowe i kulturowe wyznaczniki rozwoju teorii umysłu u dzieci głuchych. Ann Univ Paedagog Cracoviensis Stud Psychol, 2012; 5: 67–84.
- Kral A, O'Donoghue GM. Profound deafness in childhood. New Engl J Med, 2010; 363: 1438–50.
- Niparko JK, Tobey EA, Thal DJ, et al. Spoken language development in children following cochlear implantation. JAMA, 2010; 303: 1498–506.
- Morgan G, Meristo M, Mann W, Hjelmquist E, Surian L, Siegal M. Mental state language and quality of conversational experience in deaf and hearing children. Cogn Dev, 2014; 29: 41–9.
- Courtin C, Melot A-M. Metacognitive development of deaf children: lessons from the appearance–reality and false belief tasks. Dev Sci, 2005; 8: 16–25.
- Schick B, Villiers PD, Villiers JD, Hoffmeister R. Language and theory of mind: a study of deaf children. Child Dev, 2007; 78: 376–96
- Gopnik A, Wellman HM. Why the child's theory of mind really is a theory. Mind Lang, 1992; 7: 145–71.
- Gallese V, Goldman A. Mirror neurons and the simulation theory of mind-reading. Trends Cogn Sci, 1998; 2: 493–501.
- Leslie AM, Friedman O, German TP. Core mechanisms in 'theory of mind.' Trends Cogn Sci, 2004; 8: 528–33.
- Carpendale JI, Lewis C. Constructing an understanding of mind: the development of children's social understanding within social interaction. Behav Brain Sci, 2004; 27: 79–96.
- Mahy CE, Moses LJ, Pfeifer JH. How and where: theory-of-mind in the brain. Dev Cogn Neurosci, 2014; 9: 68–81.

- Hutchins TL, Allen L, Schefer M. Using the theory of mind inventory to detect a broad range of theory of mind challenges in children with hearing loss: a pilot study. Deaf Educ. Int, 2017; 19: 2–12.
- Hutchins TL, Prelock PA, Bonazinga L. Psychometric evaluation of the Theory of Mind Inventory (ToMI): a study of typically developing children and children with autism spectrum disorder. J Autism Dev Disord, 2012; 42: 327–41.
- Wimmer H, Perner J. Beliefs about beliefs: representation and constraining function of wrong beliefs in young children's understanding of deception. Cognition, 1983; 13: 103–28.
- Baron-Cohen S, Leslie AM, Frith U. Does the autistic child have a "theory of mind." Cognition, 1985; 21: 37–46.
- Perner J, Frith U, Leslie AM, Leekam SR. Exploration of the autistic child's theory of mind: knowledge, belief, and communication. Child Dev, 1989; 19: 689–700.
- Flavell JH, Flavell ER, Green FL. Development of the appearance–reality distinction. Cognit Psychol, 1983; 15: 95–120.
- Tomasello M, Rakoczy H. What makes human cognition unique? From individual to shared to collective intentionality. Mind Lang, 2003; 18: 121–47.
- 33. Leslie AM. Pretense, autism, and the theory-of-mind module. Curr Dir Psychol Sci, 1992; 1: 18–21.
- Callaghan T, Rochat P, Lillard A, et al. Synchrony in the onset of mental-state reasoning: evidence from five cultures. Psychol Sci, 2005; 16: 378–84.
- Perner J, Wimmer H. "John thinks that Mary thinks that..." attribution of second-order beliefs by 5- to 10-year-old children. J Exp Child Psychol, 1985; 39: 437–71.
- Baron-Cohen S, O'Riordan M, Stone V, Jones R, Plaisted K. Recognition of faux pas by normally developing children and children with Asperger syndrome or high-functioning autism. J Autism Dev Disord, 1999; 29: 407–18.
- Peterson CC, Siegal M. Deafness, conversation and theory of mind. J Child Psychol Psychiatry, 1995; 36: 459–74.
- 38. Peterson CC. Theory of mind and conversation in deaf and hearing children. In: Marschark M, Knoors H, editors. The Oxford Handbook of Deaf Studies in Learning and Cognition. Oxford (UK): Oxford University Press; 2020, 213–31.
- 39. Bretherton I, McNew S, Beegly-Smith M. Early person knowledge as expressed in gestural and verbal communication: when do infants acquire a "Theory of Mind"? In: Lamb ME, Sherrod LR, editors. Infant Social Cognition. Hillsdale, NJ: Erlbaum; 1981, 333–73.
- Farrar MJ, Benigno JP, Tompkins V, Gage NA. Are there different pathways to explicit false belief understanding? General language and complementation in typical and atypical children. Cogn Dev, 2017; 43: 49–66.
- 41. de Rosnay M, Pons F, Harris PL, Morrell JMB. A lag between understanding false belief and emotion attribution in young children: relationships with linguistic ability and mothers' mental-state language. Br J Dev Psychol, 2004; 22: 197–218.
- Slade L, Ruffman T. How language does (and does not) relate to theory of mind: a longitudinal study of syntax, semantics, working memory and false belief. Br J Dev Psychol, 2005; 23: 117–41.
- de Villiers PA, de Villiers JG. Deception dissociates from false belief reasoning in deaf children: implications for the implicit versus explicit theory of mind distinction. Br J Dev Psychol, 2012; 30: 188–209.
- Tompkins V. Mothers' cognitive state talk during shared book reading and children's later false belief understanding. Cogn Dev, 2015; 36: 40–51.

- Astington JW, Jenkins JM. A longitudinal study of the relation between language and theory-of-mind development. Dev Psychol, 1999; 35: 1311–20.
- 46. Taumoepeau M, Ruffman T. Stepping stones to others' minds: maternal talk relates to child mental state language and emotion understanding at 15, 24, and 33 months. Child Dev, 2008 Apr; 79: 284–302
- Peterson C, Slaughter V. Opening windows into the mind: mothers' preferences for mental state explanations and children's theory of mind. Cogn Dev, 2003; 18: 399–429.
- Devine RT, Hughes C. Family correlates of false belief understanding in early childhood: a meta-analysis. Child Dev, 2018; 89: 971–87.
- Stanzione C, Schick B. Environmental language factors in theory of mind development: evidence from children who are deaf/ hard-of-hearing or who have specific language impairment. Top Lang Disord, 2014; 34: 296–312.
- 50. Svirsky MA, Teoh S-W, Neuburger H. Development of language and speech perception in congenitally, profoundly deaf children as a function of age at cochlear implantation. Audiol Neurootol, 2004; 9: 224–33.
- 51. Zgoda M, Lorens A, Skarzynski H. Partial deafness treatment in children: educational settings after 5 to 7 years of cochlear implant use. J Hear Sci, 2020; 2: 70–4.
- Geers AE, Sedey AL. Language and verbal reasoning skills in adolescents with 10 or more years of cochlear implant experience. Ear Hear, 2011; 32: 398–48S.
- 53. Geers AE, Hayes H. Reading, writing, and phonological processing skills of adolescents with 10 or more years of cochlear implant experience. Ear Hear, 2011; 32(Suppl 1): 49S-59S.
- 54. Skarżyński PH, Ludwikowski M. Hearing screening around the world. In: Hatzopoulos S, Ciorba A, editors. An Excursus into Hearing Loss. IntechOpen; 2018.
- 55. Skarżyński H, Piotrowska A. Screening for pre-school and schoolage hearing problems: European Consensus Statement. Int J Pediatr Otorhinolaryngol, 2012; 76: 120–1.
- 56. Skarżyński H, Mielnik-Niedzielska G, Kochanek K, Niedzielski A, Skarżyński PH, Lorens A. Standardy jakości stosowania implantów ślimakowych u niemowląt, dzieci i młodzieży. Nowa Audiofonologia, 2018; 7(1): 7–15.
- Obrycka A, Padilla JL, Putkiewicz-Aleksandrowicz J, Lorens A, Skarzynski H. Partial deafness treatment in children: a preliminary report of the parents' perspective. J Hear Sci, 2020; 2: 61–9.
- Duchesne L, Marschark M. Effects of age at cochlear implantation on vocabulary and grammar: a review of the evidence. Am J Speech Lang Pathol, 2019; 28: 1673–91.
- Rinaldi P, Baruffaldi F, Burdo S, Caselli MC. Linguistic and pragmatic skills in toddlers with cochlear implant. Int J Lang Commun Disord, 2013; 48: 715–25.
- Caselli MC, Rinaldi P, Varuzza C, Giuliani A, Burdo S. Cochlear implant in the second year of life: lexical and grammatical outcomes. J Speech Lang Hear Res, 2012; 55: 382–94.
- McKinney S. Cochlear implantation in children under 12 months of age. Curr Opin Otolaryngol Head Neck Surg, 2017; 25: 400–4.
- 62. Bruijnzeel H, Ziylan F, Stegeman I, Topsakal V, Grolman W. A systematic review to define the speech and language benefit of early (<12 months) pediatric cochlear implantation. Audiol Neurootol, 2016; 21: 113–26.</p>
- 63. Geers AE, Moog JS, Biedenstein J, Brenner C, Hayes H. Spoken language scores of children using cochlear implants compared to hearing age-mates at school entry. J Deaf Stud Deaf Educ, 2009; 14: 371–85.

- 64. Boons T, De Raeve L, Langereis M, Peeraer L, Wouters J, Van Wieringen A. Expressive vocabulary, morphology, syntax and narrative skills in profoundly deaf children after early cochlear implantation. Res Dev Disabil, 2013; 34: 2008–22.
- 65. Marschark M, Duchesne L, Pisoni D. Effects of age at cochlear implantation on learning and cognition: a critical assessment. Am J Speech Lang Pathol, 2019; 28: 1318–34.
- 66. Bat-Chava Y, Martin D, Kosciw JG. Longitudinal improvements in communication and socialization of deaf children with cochlear implants and hearing aids: evidence from parental reports. J Child Psychol Psychiatry, 2005; 46: 1287–96.
- Ketelaar L, Rieffe C, Wiefferink CH, Frijns JH. Does hearing lead to understanding? Theory of mind in toddlers and preschoolers with cochlear implants. J Pediatr Psychol, 2012; 37: 1041–50.
- 68. Sundqvist A, Lyxell B, Jönsson R, Heimann M. Understanding minds: early cochlear implantation and the development of theory of mind in children with profound hearing impairment. Int J Pediatr Otorhinolaryngol, 2014; 78: 538–44.
- Marschark M, Edwards L, Peterson C, Crowe K, Walton D. Understanding theory of mind in deaf and hearing college students.
   J Deaf Stud Deaf Educ, 2019; 24: 104–18.
- Liu M, Wu L, Wu W, Li G, Cai T, Liu J. The relationships among verbal ability, executive function, and theory of mind in young children with cochlear implants. Int J Audiol, 2018; 57: 881–8.
- Yu C-L, Stanzione CM, Wellman HM, Lederberg AR. Theoryof-mind development in young deaf children with early hearing provisions. Psychol Sci, 2021; 32: 109–19.
- Meristo M, Strid K, Hjelmquist E. Early conversational environment enables spontaneous belief attribution in deaf children. Cognition, 2016; 157: 139–45.
- 73. Remmel E, Peters K. Theory of mind and language in children with cochlear implants. J Deaf Stud Deaf Educ, 2009; 14: 218–36.
- 74. Ziv M, Most T, Cohen S. Understanding of emotions and false beliefs among hearing children versus deaf children. J. Deaf Stud Deaf Educ, 2013; 18: 161–74.
- 75. Figueroa M, Darbra S, Silvestre N. Reading and theory of mind in adolescents with cochlear implant. J Deaf Stud Deaf Educ, 2020; 25: 212–23.
- Pluta A, Krysztofiak M, Zgoda M, Wysocka J, Golec. K, Wójcik J, Włodarczyk E, Haman M. False belief understanding in deaf children with cochlear implants. J Deaf Stud Deaf Educ, 2021 (in press).
- Moeller MP, Schick B. Relations between maternal input and theory of mind understanding in deaf children. Child Dev, 2006; 77: 751–66.
- Szpak M, Białecka-Pikul M. Links between attachment and theory of mind in childhood: meta-analytic review. Soc Dev, 2020; 29: 653–73.
- 79. Kotowicz J. W jaki sposób można wspomagać rozwój umiejętności podzielania uwagi głuchego dziecka słyszących rodziców? [How to support the development of the ability to divide attention in a deaf child of hearing parents?]. Studia Paedagogica, 2014; 3: 97–110.
- MacGowan TL, Tasker SL, Schmidt LA. Differences in established joint attention in hearing–hearing and hearing–deaf mother–child dyads: associations with social competence, settings, and tasks. Child Dev, 2020.
- Devine RT, Hughes C. Relations between false belief understanding and executive function in early childhood: a meta-analysis. Child Dev, 2014; 85: 1777–94.
- 82. Hale CM, Tager-Flusberg H. The influence of language on theory of mind: a training study. Dev Sci. 2003; 6: 346–59.
- Wellman HM, Peterson CC. Deafness, thought bubbles, and theory-of-mind development. Dev Psychol, 2013; 49: 2357.

- 84. Chilton H, Beazley SM. Reading the mind or only the story? Sharing fiction to develop ToM with deaf children. Commun Disord Q, 2018; 39: 466–76.
- 85. Beazley S, Chilton H. The voice of the practitioner: sharing fiction books to support the understanding of theory of mind in deaf children. Deaf Educ Int, 2015; 17: 231–40.
- 86. Holcomb M, Smeal M. Pediatric cochlear implantation: who is a candidate in 2020? Hear J, 2020; 73(7): 8–9.