The link between hearing loss, language, and social functioning in childhood

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CHAPTER 4

TERRIBLE TWOS OR EARLY SIGNS OF **PSYCHOPATHOLOGY**? **DEVELOPMENTAL PATTERNS** IN EARLY IDENTIFIED PRESCHOOLERS WITH **COCHLEAR IMPLANTS** COMPARED TO HEARING CONTROLS

In revision

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ABSTRACT

Objective

Cochlear implantation (CI) has dramatically improved the lives of children who are deaf or hard of hearing. However, little is known about its implications for preventing the development of psychiatric symptoms in this at risk population. This is the first longitudinal study to examine the early manifestation of emotional and behavioral disorders and their risk and protective factors in early identified preschoolers with cochlear implants compared to hearing peers.

Design

Participants were 74 children with cochlear implants and 190 hearing controls between one and five years old (mean age 3;8 years). Hearing loss was detected using the Newborn Hearing Screening in The Netherlands and Flanders. Parents completed the Early Childhood Inventory-4, a well-validated measure to evaluate the symptoms of DSM-IV-defined psychiatric disorders, during three consecutive years. Language scores were derived from the child's medical notes.

Results

Children with cochlear implants and hearing controls evidenced comparable levels of disruptive behavior and anxiety/depression (which increased with age in both groups). Greater proficiency in language skills was associated with lower levels of psychopathology. Early cochlear implantation and longer duration of cochlear implant use resulted in better language development. In turn, higher early language skills served as a protective factor against the development of disruptive behavior symptoms.

Conclusion

This longitudinal study uniquely shows that the improvement of language skills lowers the development of early signs of psychopathology. Early identification of hearing loss and cochlear implantation help children to improve their language skills.

INTRODUCTION

Extensive research has shown that children who are deaf or hard of hearing (DHH) experience higher levels of psychopathology than hearing peers.[1] Symptoms of psychopathology (e.g., anxiety, depression, aggression, and disruptive behavior) have mainly been studied in DHH school-aged children and adolescents, but research has clearly shown that DSM-IV-defined psychopathology can already be detected in preschoolers.[2] Moreover, the presence of such symptoms of social and emotional dysregulation in early childhood is a risk factor for future behavioral problems, peer rejection, and poor academic achievements.[3,4] For DHH children to reach their full potential, it is necessary to identify any signs of psychopathology as early in life as possible. Universal hearing screening programs have been introduced worldwide to identify hearing loss and start intervention as soon as possible.[5,6] Because early identification of hearing loss and cochlear implantation (CI) have especially improved the speech and language development of young DHH children, this could also benefit their mental health. This study aimed to examine the development of early signs of psychopathology in early identified DHH toddlers with CI compared to hearing controls and to identify risk and protective factors.

Early signs of psychopathology

Detecting early signs of psychopathology in toddlerhood can be challenging because parents often regard behavioral tantrums as being 'part of the deal' (the terrible twos). Young children experience an increased urge for autonomy; they want to do things on their own. However, their motor skills are not yet fully developed to do so, their language skills prevent them from clearly communicating their needs, and their relatively immature emotion regulation and coping skills prevent them from adequately regulating their own emotions. These skills improve with age, and (among other factors) this results in decreasing levels of disruptive behavior and lower chances of developing symptoms of anxiety and depression as children grow into school age.[7] However, if for any reason one of these skills cannot develop properly, this can result in higher levels of psychopathology.[8] Previous studies indicated that children with language problems have emotional and behavioral problems, that are not always identified because of the lack of knowledge in this area.[9]

Childhood psychopathology has major impact on society. Not only because children need extra care and support, but also because of the child's future perspectives. Higher levels of disruptive interpersonal behavior, peer aggression and anxiety in childhood are strongly related to the development of depression and substance abuse in adulthood. The prevalence of disruptive disorders in early childhood (3-6 year) is around 11%.[10] The estimated prevalence of emotional disorders in preschool children is 3.9% for symptoms of anxiety and 1.3% for depressive symptoms.[10] Disruptive disorders in early childhood are linked to juvenile delinquency and criminal acts in adulthood, causing a serious burden for society.[8] Anxiety disorders are associated with all of the other major

classes of disorders and have been proven to be a precursors for the development of depression in adulthood.[11,12] Therefore, it is of the utmost importance to detect symptoms as early as possible, in order to prevent the development of symptoms later on in life, especially in high-risk groups.

Challenges associated with pediatric hearing impairment

Hearing impairment can cause language and communication problems, which often interferes with DHH children's ability to actively participate in communication with others.[13] Diminished participation in social situations affects DHH children's opportunities for incidental learning. Incidental learning is learning by occasion, without the intention to learn. This mainly takes place outside of school settings, in everyday situations. Through observation of others, children constantly pick up different behaviors and responses and learn to replicate this at later occasions.[14] Incidental learning is one of the cornerstones for socialization. Therefore, less opportunity for incidental learning has major consequences for DHH children's social learning.[15]

The introduction of newborn hearing screening programs has enabled earlier identification and consequently earlier intervention of hearing loss in Western societies. This has been proven beneficial for the child's speech, language, and socio-emotional development[5,6], especially for children with severe to profound losses who received a CI at a young age. In fact, with early intervention and implantation, the speech and language skills of children with CI are almost comparable to those of hearing peers.[16,17] Because an improvement in language skills can benefit communicative abilities, early intervention of hearing loss can have great potential for the child's social development.[4]

Psychopathology in DHH children

An increasing interest in the development of DHH children in its broadest sense has led to numerous studies examining emotional and behavioral difficulties in DHH children. The overall results show that DHH children experience higher levels of anxiety, depression, somatic complaints, aggression, and behavioral problems than hearing children, and that they more often encounter problems in relationships with peers.[1,18] When focusing solely on DHH children with CI, the picture becomes a little brighter. School-aged children with CI are reported to show equal levels of depression, anxiety and behavioral problems as their hearing peers.[19-22] Yet, there is conflicting evidence. A large Spanish study found higher levels of behavioral problems in children with CI compared to a matched hearing group.[23] A smaller study by De Giacomo et al. found more peer problems and emotional symptoms in children with CI compared to age-matched controls.[24] The differences in these findings are likely related to whether or not the study included children who received implants relatively late and thus had less time to benefit from their CI. Yet, this is in line with the conclusions the aforementioned studies all draw; mainly children with CI who had lower language and communication skills were at risk for developing social, emotional, or behavioral problems.

Why do DHH children experience higher levels of psychopathology? One important aspect of hearing loss is that it is known to cause language delays. These language delays may prevent children from adequately communicating with others and expressing what they feel, want, and need, which can have ongoing consequences. Through conversations with others, children incidentally learn about abstract concepts such as emotions. Learning about (others') emotions helps them to understand how they feel and how to deal with their feelings; it helps children to regulate their own emotions. Emotion regulation (which includes both coping with emotions as well as emotion expression) has been proven to be problematic for DHH children with CI.[25] The young children with CI in this study used less adequate coping strategies than the hearing control group. Together with an impaired capacity for adequately communicating their needs, these lower coping skills can lead to internalization of problems resulting in withdrawal, somatic complaints, anxiety and symptoms of depression. The children with CI also expressed more negative emotions than their hearing peers which was related to more behavioral problems. Language and communication difficulties may thus hamper incidental learning which can result in higher levels of various forms of psychopathology.

Little research has been conducted regarding the effect of (early) implantation on the behavior of young children. A recent systematic review and meta-analysis stated it remained unclear what the effect of implantation is on the development of emotional and behavioral difficulties in young children with CI.[18] In addition, researchers call for longitudinal research to define causal relationships and to study the effect of age on the development of behavioral problems in children with CI.[1,4,15,18,26] To the best of our knowledge, no study has examined the effect of early intervention on the development of early signs of psychopathology in preschool children with CI.

Present study

The aim of this study was threefold. The first aim was to compare the level of disruptive behavior and anxiety/depression symptoms between early identified DHH toddlers and preschoolers with CI and age-related hearing peers. The second aim was to compare the developmental patterns of these symptoms over time in the two groups. The third aim was to identify risk and protective factors for the development of psychopathology in both young children with CI and hearing peers.

As a result of early identification and implantation and in line with previous findings, we expected to find equal levels of psychopathology in children with CI compared to hearing children.[1,6,27] Second, because higher language skills enable children to better express themselves we expected that higher language skills would serve as a protective factor against the development of psychopathology. Third, it was expected that younger age at identification and implantation would serve as a protective factor to lower the chances of developing behavioral problems.

METHODS

Participants

This study was conducted as part of a larger longitudinal research project concerning the socio-emotional development of toddlers and preschoolers with CI and with normal hearing. The sample was comprised of 190 hearing children and 74 children with bilateral severe to profound hearing loss wearing CIs.[27-29] All children were between 1 and 5 years of age (mean age 44 months) at the start of the study. Children were excluded from this study if they had any other known disability besides their hearing loss.

Hearing loss was detected using early identification programs in the Netherlands and the Dutch-speaking part of Belgium. As such, children were implanted before their 3rd birthday. Thirty-four children (46%) were implanted bilaterally. Characteristics of all participants are shown in Table 1.

Procedure

Children with CI were recruited from nine different hospitals and counseling services in the Netherlands and the Dutch-speaking part of Belgium. Hearing children were recruited from schools and daycare centers all over The Netherlands. After identification of participants, information about the study was sent to their parents or caregivers. Information regarding hearing loss and speech- and language abilities of children with CI was collected from medical notes after obtainment of informed consent. For three consecutive years, parents were annually requested to complete several questionnaires concerning the socio-emotional development of their child and a list of background variables. Permission for this study was granted by the Medical Ethics Committee of the Leiden University Medical Center.

Materials

Early signs of psychopathology

The Early Childhood Inventory-4 (ECI-4) is a parent-report questionnaire containing 108 items that is widely used to assess the symptoms of DSM-IV-defined emotional and behavioral disorders.[30] Anxiety and depression symptom severity was calculated by summing the scores for the Major Depressive Disorder (11 items), Social Phobia (3 items), and Generalized Anxiety Disorder (14 items) scales from the ECI-4. To measure disruptive behavior we summed the scores of three ECI-4 scales: Peer Conflict Scale (10 items), Oppositional Defiant Disorder (8 items), and Conduct Disorder (10 items). We combined scales because prior research with young children shows that specific symptoms within the same domain (e.g., major depressive disorder and generalized anxiety disorder) often cannot be distinguished from each other and possibly represent the same underlying disorder in toddlers.[2]

Table 1. Demographic characteristics of participants

	Total study population N = 264	
	CI	**Controls**
No. of children	74	190
Age		
Mean - in months (SD) *	39.8 (12.7)	45.6 (13.9)
Range - in months	18 - 61	18 - 66
Gender		
Male (%)	47 (63.5)	97 (51.1)
Female (%)	27 (36.5)	93 (48.9)
Socioeconomic Status† (SD)	4.4 (1.3)	4.7 (1.0)
Language Skills		
CDI - Language Comprehension (SD) **	30.0 (13.3)	40.4 (11.8)
CDI - Expressive Language (SD) **	31.2 (13.0)	43.1 (11.2)
RDLS - Receptive Language (SD)	84.8 (17.5)	
SELT - Expressive Language (SD)	82.7 (14.6)	
Preferred mode of communication		
Oral language only (%)	27 (36.5)	
Sign-supported Dutch (%)	40 (54.1)	
Sign language only (%)	7 (9.5)	
Age at first hearing aid acquisition - in months (SD)	6.9 (6.7)	
Age at implantation - in months (SD)	16.8 (7.2)	
Duration of CI use - in months (SD)	22.6 (12.0)	

†The highest level of education of each parent and their net household income were categorized on a scale ranging from zero to five. Socio-economic status (SES) was calculated by averaging these three scores.
Abbreviations: CDI Child Development Inventory, RDLS Reynell Developmental language Scales, SELT Schlichting Expressive Language Test, CI Cochlear Implant, SD Standard Deviation
* $p < 0.01$, ** $p < 0.001$

Speech and language skills

The Child Development Inventory (CDI) is an extensive 300 item parent questionnaire that creates an accurate representation of the child's development in several domains.[31] From this parent-report, two scales were used in this study. The 'Expressive Language' scale includes 50 items that define the child's expressive communication by use of vocals, gestures, and verbal behavior. Language understanding was measured by parents using 50 items that account for the 'Language Comprehension' scale. Language skills were assessed in Wave 1 and Wave 3 (Table 2).

Receptive and expressive spoken language skills were tested using the Reynell Developmental language Scales (RDLS) and the Schlichting Expressive Language Test (SELT),

respectively.[32] These tests were administered as part of the post-implant rehabilitation program and therefore, scores are only available for the children with CI. Scores were derived from the child's medical notes and therefore only available at baseline. In 23 children, language scores were missing.

Table 2. Mean scores on psychopathology and language skills

	Baseline		Wave 2		Wave 3	
	CI	**Controls**	**CI**	**Controls**	**CI**	**Controls**
	n = 51	n = 159	n = 42	n = 109	n = 44	n = 77
ECI-4						
Anxiety / Depressive symptoms	0.15	0.16	0.18	0.18	0.20	0.19
Disruptive behavior	0.31	0.30	0.39	0.28	0.34	0.28
CDI						
Language Comprehension	30.0	40.4	na	na	44.0	48.7
Expressive Language	31.2	43.1	na	na	45.1	49.1

Abbreviations: CI Cochlear Implant, ECI Early Childhood Inventory, CDI Child Development Inventory, na not administered

Statistical analyses

The two groups (i.e., hearing and CI) were compared on demographic features using independent samples *t* tests for continuous variables and χ^2 test for dichotomous variables (Table 1). On average, children with CI were 5.8 months younger than the hearing children. We therefore decided to analyze our data using Linear Mixed Models (LMM) that allow us to correct for this difference in age. Assessment of model fit was evaluated using Akaike's information criterion. LMM were used to examine i.) differences in baseline levels of psychopathology, ii.) developmental changes of psychopathology over time, and between the two groups (hearing and CI), and iii.) risk and protective factors for the development of early signs of psychopathology over time. A p-value < 0.05 was considered statistically significant.

Multiple imputation of missing data

As almost inevitable in large longitudinal studies, we were confronted with missing data. Not all participants completed all three data waves and occasionally scores were missing from medical files for numerous reasons. Many statistical methods for analyzing datasets assume complete cases. Consequently, these analyses remove incomplete cases beforehand, introducing bias and a drop in statistical power.[33,34] To better deal with missing data, multiple imputations (MI) were used which involves filling in the missing data based on known characteristics of the participant and the relations observed in the data for other participants with complete data.[35-37] We were unable to find a pattern in the missing data and thus no relations between missing data and participant

characteristics such as age of degree of hearing loss were found. Using the MI technique, missing scores on the CDI and ECI from 23 children with CI and 31 controls were imputed together with missing scores on the variables to calculate the SES of the family (level of parental education and net income) in 16 children with CI and 33 controls. The following variables were entered into the imputation model to estimate missing values: age, hearing status, language test scores, outcomes on the CDI and ECI, SES of the family and gender. We performed ten imputations and analyzed the newly formed datasets using standard analyzing techniques. Pooled results are reported. Only missing data at baseline were imputed because the LMM technique is robust enough to correct for missing follow-up data in a longitudinal design.[38]

RESULTS

Language development

To confirm the extensively studied positive effect of early identification and implantation on the language development of young DHH children [5,6,16,17] we were interested in the development of expressive language and language comprehension over time in our sample. A multilevel LMM with Language Comprehension as the dependent variables, Time as the determinant for repeated measurement and Age, Age at implantation, and Time as the fixed effects revealed that younger age at implantation and longer duration of implant use increased Language Comprehension over time (t = -3.35, p < 0.001 and t = 3.80, p < 0.001, respectively). A comparable effect was found for Expressive Language development: (t = -3.58, p < 0.001 and t = 3.96, p < 0.001 for Age at implantation and Duration of implant use, respectively). Children with bilateral CIs had higher receptive language skills at baseline than unilaterally implanted children, t = -2.41, p < 0.05. This difference was not significant for expressive language skills (t = -1.99, p = 0.053 and , t = -1.91, p = 0.063 for word development and sentence development, respectively). On average, bilateral users were implanted five months earlier than children with only one CI, t = 3.15, p < 0.01.

Development of symptoms over time

In order to evaluate the development of anxiety/depression symptoms over time, a multilevel LMM with Time as the determinant for repeated measurements and Age, SES, Group, Language Comprehension, and Time as fixed effects was performed. A main effect was found for Time (t = -2.58, p < 0.01), Language Comprehension (t = -2.11, p < 0.05) and Age (t = 4.24, p < 0.001). No difference was found between the two groups. A better model fit was accomplished by adding an Age*Time interaction term to the model (t = -2.46, p < 0.05). Figure 1 shows the raw data for all three waves without the imputed values. The outcome of the LMM described above was plotted in this same figure. This line (which is based on the imputed dataset) shows that the relation between age and the development of anxiety/depression symptoms can best be described by means

of a parabola with a peak in symptoms at the age of 80 months. An increase in symptoms over time was seen in the younger children whereas a decrease of symptoms over time was found in children older than four at baseline.

A comparable model was run for the development of disruptive behavior symptoms and revealed an increase of symptoms with Age (t = 3.30, p < 0.001), which was qualified by an Age*Time interaction (t = -2.00, p < 0.05). No influence was found for Language Comprehension, Language Expression, Group or SES. The relation between age and the development of disruptive behavior symptoms can also be described by means of a parabola with a peak in symptoms at the age of 67 months. The raw data collected in all three waves is plotted in Figure 2. The plotted line is the function that can be derived from the LMM that is based on the imputed dataset.

Risk and protective factors for the development of symptoms

To identify risk and protective factors for the development of both anxiety/depression and disruptive behavior symptoms, the format of the database was changed into a long format with two time points instead of three (i.e., symptoms after one and two years). This way,

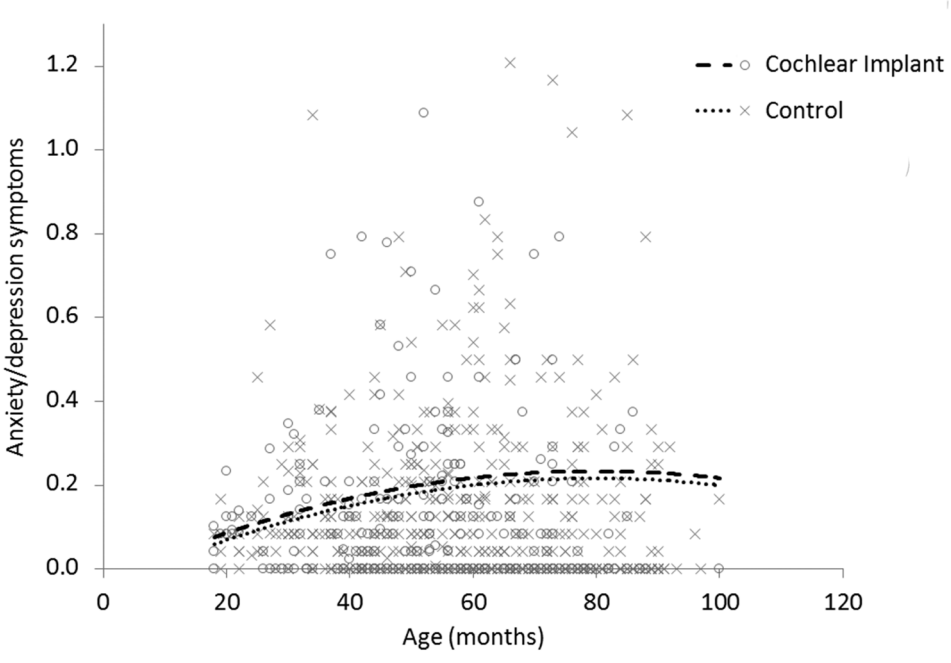

Figure 1. Development of anxiety/depression symptoms with age.
Outcomes on the anxiety/depression scale from the Early Childhood Inventory on all three waves (Y-axis) are plotted against the age of the child (X-axis) for children with CI (dots) and the control group (crosses). The lines describe the mean development of symptoms as age increases. No significant difference between the two groups was found.

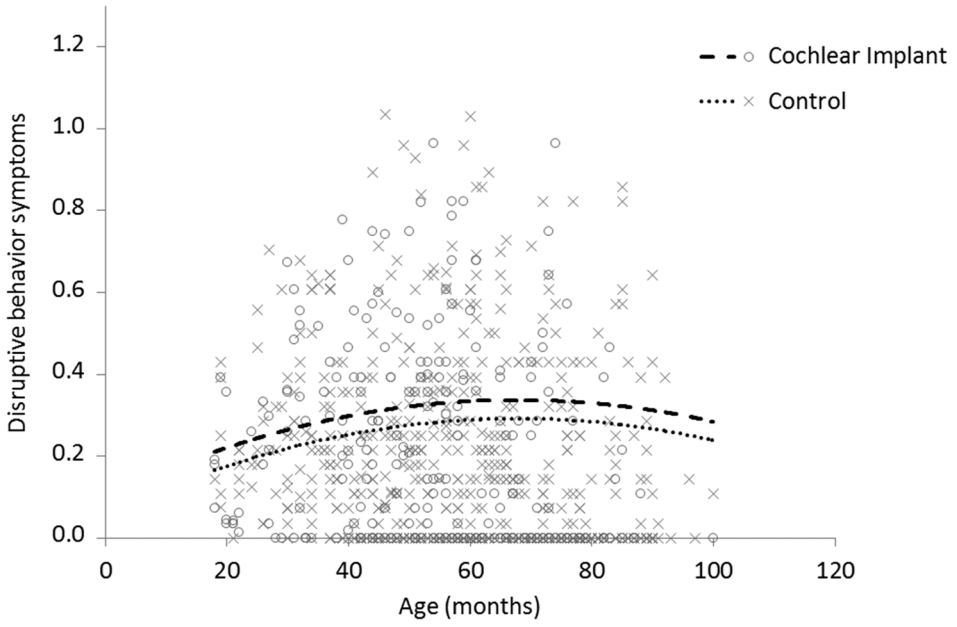

Figure 2. Development of disruptive behavior symptoms with age.
Outcomes on the disruptive behavior scale from the Early Childhood Inventory on all three waves (Y-axis) are plotted against the age of the child (X-axis) for children with CI (dots) and the control group (crosses). The lines describe the mean development of symptoms as age increases. No significant difference between the two groups was found.

the baseline levels of symptoms (measured at Wave 1) could be entered as covariates into the model to identify other unique predictors of psychopathology besides the level of symptoms at baseline. A LMM with anxiety/depression symptoms as the dependent variable and Group, Gender, Age, SES, Language Comprehension, Language Expression, and baseline level of anxiety/depression symptoms as fixed effects revealed a highly significant positive effect for baseline level of anxiety/depression symptoms only ($t = 9.14$, $p < 0.001$). Children that scored high at baseline remained to score high after two years.

A similar LMM with disruptive behavior revealed a positive effect for baseline level of disruptive behavior symptoms ($t = 7.53$, $p < 0.001$) and Age ($t = 1.97$, $p < 0.05$). For Language Comprehension and Expressive Language, a highly significant negative effect was found ($t = -2.87$, $p < 0.01$ and $t = -3.24$, $p < 0.001$, respectively). Higher levels of Language Comprehension and Expressive Language contributed to the prevention of the development of disruptive behavior, regardless of their hearing status.

The influence of CI on the development of symptoms

To identify the effect of several CI-related variables on the development of early signs of psychopathology, factors such as Age at implantation and Duration of implant use were

entered into a LMM with Age and Time as fixed effects. No effect was found for uni- or bilateral implantation or communication mode. No direct influence was found for the Age at first amplification, Age at implantation, or the Duration of CI usage. From the implanted children, spoken language test scores were available at baseline. Baseline spoken language scores had no influence on the development of symptoms.

DISCUSSION

This longitudinal study is the first to compare the developmental pattern of early signs of psychopathology between preschool children with CI and hearing controls. In line with our hypotheses and previous studies, preschoolers with CI showed levels of psychiatric symptoms equal to hearing peers.[1,6,27,39] Moreover, the developmental patterns of these symptoms were comparable for both groups. Symptoms of psychopathology increased with age, and higher language skills contributed to the prevention of disruptive behavior symptoms but not anxiety/depression symptoms in both groups. Within the group of children with CI, a younger age at implantation and longer duration of implant use led to an increased language development.

An increase in language skills over time led to a decrease of symptoms of depression and anxiety. This finding is in line with previous studies in typically developing children.[40] As children grow older, the levels of their language and communication skills are of increasing importance in determining the amount of social interaction they have access to. From these social interactions, children may learn new words and meanings and so the relationship between language and symptoms of anxiety and depression is reciprocal. Unfortunately, this reciprocity can also work in a negative way. When children encounter language delays, this may result in withdrawal from interactions. This in turn lowers their social interaction time which may result in loneliness, lower self-esteem and feelings of depression and social anxiety.[4]

Higher language skills at a young age help to protect against the development of early signs of disruptive behavior in our study. This can be explained by the fact that early language skills help children to communicate their needs and wishes. When children are less able to make themselves understandable to others, this causes frustration, resulting in higher levels of aggressive and disruptive behavior.[40]

This study shows that early identification and intervention of hearing loss through CI results in better language skills, as previously established in other studies.[6,16,17] The longitudinal design of this study uniquely adds insight into the protective effect of language skills on the development of early signs of psychopathology. A younger age at implantation increased the language skills of children with CI over time. Through improving language skills, an indirect effect of early intervention on the development of psychopathology was

shown. These findings support our hypothesis on the importance of language development for children's social-emotional development.

Possibly, the limited spread in both the age at implantation and the severity of symptoms prevents us from finding a direct relationship between age at implantation and levels of psychopathology. Furthermore, it is plausible that other factors that we did not measure in this study influenced the level of symptoms such as cognitive development, maternal sensitivity or parenting styles.[41] However, the absence of a direct causal relationship between age at intervention and the level of psychosocial development is consistent with previous literature.[39,42]

A factor that could have biased our results is the etiology of hearing loss in this group. Hearing loss due to meningitis for instance requires quick action from professionals as it is known to cause rapid ossification of the cochlea.[43] Children suffering from severe hearing loss after meningitis are therefore often implanted as soon as possible and also bilateral when possible, since waiting may decrease the chances for successful implantation and subsequent functioning of the implant. This is reflected in our data. The cause of hearing loss was more often found to be due to meningitis in the bilaterally implanted children than in the group of unilaterally implanted children. All nine bilaterally implanted children with a history of meningitis received their implants simultaneously. In addition, the bilaterally implanted children received their implants earlier than unilaterally implanted children. The question now is, how did this affect our data? Children who suffered from meningitis most likely had normal hearing prior to infection. This implies that the auditory cortex in the brain has been susceptible to auditory stimulation. Restoring auditory stimulation by CI may thus yield very good results in these children. On the other hand, it is also known that meningitis does not only affect the cochlea. It may also damage the auditory nerve and cause other neurological deficits which may result in lower hearing abilities and subsequent language development.[44] From this study, it therefore remains unclear what the role of bilateral implantation is on the development of language and psychosocial skills, which is a limitation of this study. Yet, the superior language skills that were found in the bilaterally implanted children when compared to unilaterally implanted children in this study are in line with previous research.[45,46] Future studies should address this important aspect. In addition, such studies should also include information regarding the type of amplification of the contralateral ear in unilaterally implanted children when comparing unilaterally and bilaterally implanted children.

Regardless of the child's hearing status, early signs of psychopathology increased with age with a peak in disruptive behavior symptoms around the age of five years and for anxiety/depression symptoms at approximately six years of age. In line with previous studies, both symptoms decreased in intensity afterwards.[47] This can be explained by the fact that by the age of six, children go to school, participate in sports and more frequently take part in social situations. Through incidental learning and by trial and error, they learn

how to respond in different social environments. Among other things, the development of language skills allows young children to increasingly express their selves and engage in communication with others.

CONCLUSION

To the best of our knowledge, this is the first longitudinal study that examines the development of early signs of psychopathology in DHH preschoolers with CI compared to hearing age-related peers. The results of this study shed new light on the development of early implanted children. The longitudinal design shows the clinical importance of early intervention on the development of language skills. It underlines the important effect of language development on the psychosocial functioning of DHH children.

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