

Salud mental

ISSN: 0185-3325

Instituto Nacional de Psiquiatría Ramón de la Fuente Muñiz

Charry-Sánchez, Jesús David; Ramírez-Guerrero, Sofía; Vargas-Cuellar, María Paula; Romero-Gordillo, María Alejandra; Talero-Gutiérrez, Claudia Executive functions in children and adolescents with hearing loss: A systematic review of case-control, case series, and cross-sectional studies Salud mental, vol. 45, no. 1, 2022, pp. 35-49
Instituto Nacional de Psiquiatría Ramón de la Fuente Muñiz

DOI: https://doi.org/10.17711/SM.0185-3325.2022.006

Available in: https://www.redalyc.org/articulo.oa?id=58269985006



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salud mental

Executive functions in children and adolescents with hearing loss: A systematic review of case-control, case series, and cross-sectional studies

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Received: 17 February 2021 **Accepted:** 12 August 2021

Citation:

Charry-Sánchez, J. D., Ramírez-Guerrero, S., Vargas-Cuellar, M. P., Romero-Gordillo, M. A., & Talero-Gutiérrez, C. (2022). Executive functions in children and adolescents with hearing loss: A systematic review of case-control, case series, and cross-sectional studies. Salud Mental, 45(1), 35-49.

DOI: 10.17711/SM.0185-3325.2022.006



ABSTRACT

Introduction. Children with hearing loss have been reported to perform lower in executive function and language tasks than their normal-hearing peers. Objective. To describe EF performance profile in children and adolescents with hearing loss. Method. Using different databases including PubMed, Scopus, and ScienceDirect, we conducted a systematic review of case-control, cross-sectional studies, and case series that evaluated executive function performance in children and adolescents with hearing loss with or without hearing aids, cochlear implants, and/or native sign language, since 2000 until April 2020. Fifteen studies were selected after quality assessment using Critical Appraisal Tools provided by Joanna Briggs Institute. Results. The studies differed in the assessment tools, and the results obtained by different authors were inconsistent. However, these studies revealed that children and adolescents with hearing impairment have lower performance in working memory, inhibition, cognitive flexibility, and attention than their normal-hearing peers. Discussion and conclusion. Executive function assessment tools are used indistinctively for both children with and without hearing loss. Consequently, as tools were designed for normal hearing population, results can significantly vary in the population with hearing impairment. Hence, it is important to establish a standardized protocol specifically adapted for this population.

Keywords: Children, cochlear implant, executive functions, hearing aids, hearing loss.

RESUMEN

Introducción. Se ha reportado que los niños con pérdida auditiva tienen un desempeño más bajo en pruebas de función ejecutiva y lenguaje en comparación con sus pares oyentes. Objetivo. Describir el perfil de desempeño en funciones ejecutivas en niños y adolescentes con pérdida auditiva. Método. Utilizando diferentes bases de datos incluidas PubMed, Scopus y ScienceDirect, se llevó a cabo una revisión sistemática de estudios de corte transversal, casos y controles y series de casos que evaluaron el desempeño en funciones ejecutivas de niños y adolescentes con pérdida auditiva con y sin audifonos, implante coclear y/o uso de lenguaje de señas desde 2000 hasta abril de 2020. Se seleccionaron 15 estudios usando el instrumento de evaluación de calidad del Instituto Joanna Briggs. Resultados. Los estudios utilizaron distintas herramientas de evaluación con resultados inconsistentes entre los diferentes autores. Sin embargo, los estudios reportaron que los niños y adolescentes con pérdida auditiva tienen un desempeño más bajo en memoria de trabajo, inhibición, flexibilidad cognitiva y atención que sus pares oyentes. Discusión y conclusión. Las herramientas de evaluación de función ejecutiva fueron empleadas indistintamente para niños con y sin pérdida auditiva. Teniendo en cuenta que las pruebas están diseñadas para la población oyente, los resultados pueden variar significativamente en la población con pérdida auditiva. De ahí la importancia de establecer un protocolo estandarizado adaptado para esta población.

Palabras clave: Niños, implante coclear, funciones ejecutivas, audífonos, pérdida auditiva.

INTRODUCTION

Executive functions (EFs) are a set of skills that facilitate planning, organizing, and structuring daily life activities and long-term life goals (Blair, 2017; Doebel, 2020). EF are principally mediated by the frontal lobes, specifically by the prefrontal cortex, with its right dorsolateral area involved with monitoring behavior and its left dorsolateral area related to verbal processing. Both dorsolateral areas, together with the superior medial frontal lobe, are required for shifting, whereas the inferior medial frontal area contributes to response inhibition (Blair, 2017; Jurado & Rosselli, 2007). EF performance is also associated with a series of neural circuits connecting the frontal lobes with subcortical structures (Goldstein, Naglieri, Princiotta, & Otero, 2014; Royall et al., 2002).

Although EF are not directly associated with the symbolic processing of information, they are related to its control and organization, as well as the coordination and programming of movement and behaviors directed towards a purposeful activity (Corbett, Constantine, Hendren, Rocke, & Ozonoff, 2009; Luria, 1977; Ustárroz, Molina, Lario, García, & Lago, 2012). EF also include selective attention, working memory, and cognitive flexibility, which are required for concept formation and perceptual activity (Anderson, 2002; Gilbert & Burgess, 2008; Ropovik, 2014). Furthermore, the structuring of logical syntax within coherent discourse and the modulation of behavior and affection in different scenarios are also considered to be EF-dependent abilities (Gilbert & Burgess, 2008).

The domains of EF have been described by different authors; some have proposed that there are three categories, while others have suggested that there are more than five (Ardila & Ostrosky-Solís, 2008; Diamond, 2013). However, most studies agree that EF include the following domains: inhibition, planning, working memory, cognitive flexibility, attention, problem solving, and reasoning, which are capacities that develop from early on in life (Flores-Lázaro, Castillo-Preciado, & Jiménez-Miramonte, 2014).

Multiple tools have been used to measure the different EF skills including the Behavior Rating Inventory of Executive Function (BRIEF) 2nd edition, BRIEF-Preschool, National Institutes of Health (NIH) Toolbox Cognitive Assessment Battery, Stroop test, Tower of London test, Go/No-Go task, Tower of Hanoi, Wisconsin Card Sorting Test, and subtests from the Wechsler Intelligence Scale for Children (WISC) (Chan, Shum, Toulopoulou, & Chen, 2008; Delgado-Mejía & Etchepareborda, 2013; Flores-Lázaro, Ostrosky-Solís, & Lozano-Gutiérrez, 2008; Soprano, 2003). The BRIEF is a standardized questionnaire designed for parents, teachers, and caregivers to evaluate a child's real-life performance in eight domains of executive functions including inhibition, shifting, emotional control, working memory, plan/organize, organization of materials, and

monitor. Contrastingly, other laboratory-based tools, such as the Stroop test and Tower of Hanoi, are carried out in a controlled environment to reduce biased results (Goldstein & Naglieri, 2014).

On the other hand, the WISC IV and V, provides information about executive functions by examining specific tasks that require working memory and processing speed. WISC V evaluates five primary index scores including Verbal Comprehension Index (VCI), Visual Spatial Index (VSI), Working Memory Index (WMI), Fluid Reasoning Index (FRI), and Processing Speed Index (PSI), of which WMI, FRI, and PSI provide important information regarding performance in executive function. Additionally, it measures complementary index scales which indirectly measure working memory such as Naming Speed Index, Symbol Translation Index, and Storage and Retrieval Index. Compared to WISC IV, the fifth edition includes a revision of instructions for children's better comprehension of evaluating tasks and simplifies scoring criteria. Furthermore, both tests require trained evaluators with experience in child assessment and in the application of the test (Lace et al., 2020; Pearson Assessment, 2018).

Certain physical, emotional, and social factors are required for the adequate acquisition of EF (Diamond & Lee, 2011). However, some conditions, such as hearing loss, may influence the optimal development of EF. Hearing loss is a treatable condition, which may interfere with normal neurodevelopment, especially in the acquisition of communication skills (Korver et al., 2017). Hearing loss can be classified according to the localization of damage (conductive, sensorineural, or mixed hearing loss), the degree of hearing loss (mild, moderate, severe, and profound), and its etiology (ASHA, 2016; Korver et al., 2017). Stevens et al. (2013) have reported that the prevalence of hearing loss in children between five to 14 years old can range from 1.0 to 2.2% and is more prevalent in boys than in girls. Furthermore, it has been found that as the population grows, the prevalence of hearing loss increases; in newborns, the prevalence is 1.33 per 1,000 live births, and 3.5 per 1,000 in adolescent population (Morton & Nance, 2006; Watkin & Baldwin, 2012).

Children with hearing loss may have language developmental delay of both comprehensive and expressive skills. Altered hearing afferences, either in the receptor or the transmission route, deprive the subject of adequate development of skills such as perception and auditory discrimination. The magnitude of the commitment will be related to the age of diagnosis and the beginning of rehabilitation, degree of hearing loss, and the use of hearing amplifiers or cochlear implant (Acosta Rodríguez, Ramírez Santana, & Hernández Expósito, 2017). Language allows nomination, categorization, and generalization of the surrounding environment, favoring the development of abstraction capacity. As experience is enriched, and new information is provided, these abilities mature and constitute the basic pillars for

cognition. Furthermore, language remains a key aspect in the capacity of self-monitor and self-regulation since the early developmental stages (Petersen, Bates, & Staples, 2014). As a result, the delay in language development affects performance in both verbal and non-verbal assignments, which compromises performance in tasks related to executive functions (Perszyk & Waxman, 2018).

Depending on the etiology and degree of hearing loss, the cognitive skills of these individuals vary. Nevertheless, the use of hearing aids or cochlear implants, combined with adequate language therapy will support the communication skills allowing them to perform tasks similar to those with normal hearing (Paluch et al., 2019; Yoshinaga-Itano, Sedey, Wiggin, & Mason, 2018). However, speech is not the only way to communicate, for example, native sign language subjects have been found to perform at the same level as their hearing peers in the Auslan working memory span task (Wang & Napier, 2013). Regarding the reading process, which involves functions such as attention, inhibition, and cognitive flexibility (Roldán, 2016), children with hearing loss, including cochlear implant users, have been found to perform lower in emergent literacy than their normal-hearing peers (Werfel, 2017). Furthermore, concept formation involves several abilities, including language, higher-order cognitive functions, and EF (Seel, 2012; Yoshida & Smith, 2003). Castellanos et al. (2015) reported that, despite the use of a cochlear implant, children with hearing loss perform significantly lower in concept formation and abstraction tasks than their normal-hearing peers.

Table 1

JBI quality tools results for final selected articles

			Quality	results
_	Author	Tool	(9	%)
1	Al-Salim, S. et al.	Cohort	9/11	(82%)
2	Beer, J. et al.	Cross sectional	6/8	(75%)
3	Beer, J. et al.	Case and controls	8/9	(89%)
4	Daza, M. T. et al.	Cohort	6/8	(75%)
5	Ead, B. et al.	Case and control	7/9	(78%)
6	Figueras, B. et al.	Case and control	7/10	(70%)
7	Hall, et al.	Case and control	10/10	(100%)
8	Holt, D. et al.	Cross sectional	6/8	(75%)
9	Kirby, B. et al.	Cross sectional	6/8	(75%)
10	Kronenberger, W. G. et al.	Cohort	8/10	(80%)
11	Nittrouer, S. et al.	Case and control	8/10	(80%)
12	Nunes, T. et al.	Case and control	7/9	(78%)
13	Pagliaro & Ansell	Cross sectional	6/8	(75%)
14	Surowiecki, V. et al.	Case and control	9/10	(90%)
15	Xuan, B. et al.	Case and control	7/9	(78%)

Note: Not all criteria were applicable to each article. The overall result (%) excluded the criteria not applicable for each study.

Despite expanding research in EF, there are few studies that have explored EF performance in children with hearing loss. The aim of this systematic review of the literature was to describe the qualitative characteristics of EF performance in children and adolescents with hearing loss, with or without hearing aids, cochlear implants, and/or native sign language, and to propose methods that provide information about EF in this population taking into account their specific language characteristics.

METHOD

Protocol registration was submitted to PROSPERO; however, it was rejected due to the high demand of submissions regarding the COVID-19 pandemic. We conducted literature searches of different databases, including PubMed, Scopus, and ScienceDirect. The terms included in the search were as follows: "sensorineural hearing loss, deafness, hearing loss impairment, inhibition, attention, and executive functions." The search syntax used for PubMed was as follows: ((((sensorineural hearing loss) OR deafness) OR hearing loss impairment) AND inhibition) AND executive functions.

The first search carried out by three authors JCS, SRG, MVC, without any date restriction, revealed few unrelated articles published before the year 2000; therefore, the cut-off point was taken from that year on. The last search date was April 2020. The inclusion criteria were as follows: articles with cross-sectional, case-control, and case series design publications after 2000, subjects were under 18 years old, and no language filter was used. By limiting the age group to children and adolescents under 18 years old, it is possible to better discriminate congenital hearing loss

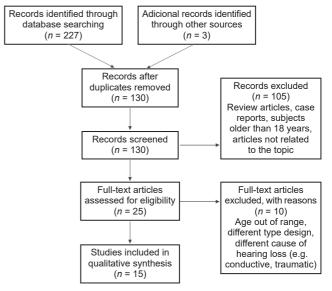


Figure 1. Prisma flow diagram.

from other multifactorial etiologies that induce hearing loss in adult population. A total of 228 articles were identified. After manually removing duplicates by the first three authors, 128 references remained. Of these, 105 articles were filtered and eliminated by title and abstract, particularly those unrelated to the topic or that did not comply with the established inclusion criteria. All authors participated at this stage. In all author's periodic meetings, reasons for eliminating articles were clearly stated and discussed in detail. The following articles were excluded: review articles, case reports, publications with subjects older than 18 years, and papers unrelated to the topic. Data from the remaining 23 studies were analyzed using full-text and quality assessment that was supported by the Critical Appraisal Tools provided by Joanna Briggs Institute (Joanna Briggs Institute, 2017).

Each quality assessment tool is specifically designed for each type of study (cohort, case control, etc.), which were applied to each article accordingly. The quality result, as shown in Table 1, reports the relationship between the number of items achieved by the article over the total number of items evaluated. The final percentage must be 70% or more for the article to be considered in the review (Joanna Briggs Institute, 2017).

Of those 23 studies, 10 articles were further excluded because they were unrelated to the objectives of the search, the age of participants was outside the range, and the variables measured were not related to EF. Finally, 15 articles were selected for qualitative synthesis. Appendix was performed independently for each article as shown in Supplementary Data. Due to the lack of a standardized assessment tool for

Table 2
Cognitive domains compromised correlated with the degree of hearing loss

Study	Year	Number of subjects with hearing loss	Degree of hearing loss	Cognitive domains compromised
Ead et al.	2013	7/14	Profound	- Complex verbal working memory - Verbal/Phonological processing
Daza et al.	2014	30/30	Severe-to-profound, Mild-to-mod- erate, cochlear implants and con- ventional hearing aids	- No significant difference
Al-Salim et al.	2020	65/100	Mild, cochlear implant	Phonological processingVocabularyWorking memoryExecutive functions
Kirby et al.	2019	24/24	Mild, hearing aid	- No significant differences
Beer et al.	2014	24/45	Profound, cochlear implant	Executive functionsAttention and inhibition
Surowiecki et al.	2002	48/48	Profound, Severe, Moderate-to-severe, cochlear implants	- No significant differences
Xuan et al.	2018	36/72	Profound	- Decision-making
Nittrouer et al.	2012	35/52	Severe-to-profound, Moderate, cochlear implants	- Emergent literacy - Oral language skills
Figueras et al.	2008	47/69	Profound, Moderate, Severe, co- chlear implants	- Intelligence
Holt et al.	2013	59/59	Cochlear implants	Inhibitory controlLanguage and vocabulary developmerShifting attentionWorking memory
Nunes et al.	2009	55/133	Moderate, Severe-to-profound, cochlear implants	- Multiplicative reasoning
Pagliaro & Ansell	2012	59/59	Mild, moderate, severe, profound, cochlear implants	- Problem-solving
Hall et al.	2018	71/116	Cochlear implant, sign language	- Inhibition - Working memory
Beer et al.	2011	45/45	Cochlear implant	- Inhibition - Working memory
Kronenberger et al.	2020	41/81	Cochlear implant	- Language - Inhibition - Working memory

children and adolescents with hearing loss, there was insufficient data for quantitative analysis, therefore, a qualitative analysis was preferred. This systematic review was conducted using the parameters established by the PRISMA Statement (Figure 1; Moher, Liberati, Tetzlaff, & Altman, 2009).

RESULTS

Among the 15 studies analyzed, there were seven case-control and six cross-sectional studies. The age range was from one to 18 years, with more frequent cases including those between the age ranges of six to 18 years. Some studies did not report the sex of the cases; those that reported sex found no significant difference between male and female participants. However, there was great variability in the number of subjects from seven to 71. Table 2 describes the main variables of each study.

Although all articles included objective measures for executive function, it was not possible to synthetize and compare them using the same criteria due to the variety of assessment tools used. Each study evaluated different skills: some used specific tests for children with hearing loss, while others applied tests designed for the general population. The assessment tools are outlined in Table 3.

As a standardized measure to classify hearing loss, most studies recorded the degrees of hearing loss based on pure-tone average (PTA). Only some studies used the classification established by the American Speech–Language–Hearing Association (ASHA, 2016; Clark, 1981; Table 4).

Some studies failed to report the methodology used to group subjects, so it remained unclear whether such grouping was based on hearing level or threshold ranges to establish the degree of hearing loss. Moreover, the studies that included children with cochlear implants reported variable durations of device use (from .5 to 16 years), as well as varying ages of implantation (from approximately one year to 3.5 years).

In terms of children with normal hearing, some studies recruited age- and sex-matched controls (Surowiecki et al., 2002), while others included subjects' siblings to control sociodemographic factors that may have an effect on children's overall performance (Ead, Hale, DeAlwis, & Lieu, 2013). The allocation of children with both sensorineural and conductive hearing loss to the same group in one study (Al-Salim, Moeller, & McGregor, 2020) contrasts with the rest of the studies, which only focused on sensorineural hearing loss.

There was no uniformity in the results obtained by different authors. While some reported success in different sets of skills, others reported a significantly lower performance on the same tasks (Figueras, Edwards, & Langdon, 2008).

Some studies found no significant difference in performance in EFs between children with normal hearing and those with hearing loss, independent of the degree of hearing loss and the type of hearing aid or cochlear implant (Beer et al., 2014; Daza, Phillips-Silver, Ruiz-Cuadra, & López-López, 2014; Figueras et al., 2008; Hall, Eigsti, Bortfeld, & Lillo-Martin, 2018; Kirby, Spratford, Klein, & McCreery, 2019; Nittrouer, Caldwell, Lowenstein, Tarr, & Holloman, 2012; Surowiecki et al., 2002). According to this finding, the following EFs of children with hearing loss was not different to those with normal hearing: inhibition, working memory, attention, visual attention, visual memory, cognitive flexibility, and planning/organizing (Beer, Kronenberger, & Pisoni, 2011; Beer et al., 2014; Figueras et al., 2008; Hall et al., 2018; Kirby et al., 2019; Kronenberger, Xu, & Pisoni, 2020; Surowiecki et al., 2002). Furthermore, children with hearing loss had similar comprehensive and expressive vocabulary and phonological skills as those with normal hearing (Daza et al., 2014; Nittrouer et al., 2012). Nonetheless, another study reported that children fluent in verbal or sign communication performed better than those who did not, independent of their hearing loss (Hall et al., 2018). Similarly, children with good family support, including maternal sensitivity, use of oral language, organization and control at home, supportiveness and cohesion among family members, family size, and education level, tended to have better emotional and inhibitory control (Holt, Beer, Kronenberger, & Pisoni, 2013). Conversely, several studies reported that children with hearing loss performed significantly lower than those with normal hearing in EFs, such as working memory, inhibition, cognitive flexibility, and attention (Beer et al., 2014; Figueras et al., 2008; Hall et al., 2018; Kirby et al., 2019; Kronenberger et al., 2020; Surowiecki et al., 2002).

DISCUSSION AND CONCLUSION

The role of language in overall performance of children and adolescents with hearing loss

The results of this systematic review indicate that children with deafness have a lower performance in hearing skills, from phonological discrimination to verbal reasoning, which are acquired later, regardless of whether the child had a cochlear implant or some other hearing aid. If stimuli are presented in a multiple-choice format or if recognition is limited to hearing, deaf children presented greater difficulties; however, if these were accompanied by visual clues, performance was improved (Al-Salim et al., 2020). Despite the use of cochlear implants or hearing aids, children with hearing loss do not have the same linguistic development as their hearing peers (Ambrose, Fey, & Eisenberg, 2012; Colin, Leybaert, Ecalle, & Magnan, 2013; Nittrouer et al., 2012). For example, James, Rajput, Brinton, and Goswami, (2008) found that children who had received cochlear

Table 3
Executive functions assessment tools used in the 15 studies

Assessment tools

Attention

- Child Neuropsychological Maturity Questionnaire computerized version
- The Intradimensional/Extradimensional Set Shift Task
- The Tower of London test
- Attention subtest of the NIH toolbox
- Attention Sustained subtest of the Leiter International Performance Scale

Intelligence

- Wechsler Abbreviated Scale of Intelligence
- Wechsler Intelligence Scale for Children IV
- The Picture Similarities subtest of the Differential Ability Scales
- Raven's Progressive Matrices
- The Hiskey-Nebraska Test of Learning Aptitude

Memory

Working memory

- Letter Span Tasks
- Counting Span Tasks
- Counting Recall subtest of the Automated Working Memory Assessment
- Nonword repetition task
- The Spatial Working Memory task

Visuospatial/spatial memory

- Visuospatial Memory Span Tasks
- Kaufman Assessment Battery for Children
- Memory for Designs subtest of the NEPSY-II
- The Pattern and Spatial Recognition test
- The Delayed Matching to Sample test
- The Paired Associates test
- Corsi block task

Short-term visual memory

- Memory of Faces task

Inhibition

- Go/No-Go task
- Flanker Inhibitory Control task

Executive functions

- Short-term memory task
- From NEPSY battery: Tower, Visual Attention, Design Fluency, and Knock and Tap
- Day-Night and One-Two tasks
- From de D-KEFS battery: Card Sorting test
- Behavior Rating Inventory of Executive Function
- Behavior Rating Inventory of Executive Function-Preschool
- Dimensional Change Card Sort
- NIH Toolbox Cognitive Assessment Battery
- Beery Developmental Test of Visual Motor Integration
- Decision-Making tasks: Iowa Gambling Task and Game of Dice Task

Language, vocabulary, speech, and phonological abilities

Language

- The Preschool Language Scale 4th edition
- Clinical Evaluation of Language Fundamentals preschool test
- The Auditory Comprehension subtest of the Preschool Language Scales 4th edition
- Nonword repetition task
- Spectral-temporally Modulated Ripple test
- Aided Speech Intelligibility Index
- The Expressive One-Word Picture Vocabulary test
- The British Picture Vocabulary Scale
- The Test for Reception of Grammar
- Bamford-Kowal-Bench Sentence List
- Consonant-Nucleus-Consonant word lists

Vocabulary

- Peabody Picture Vocabulary Test Revised
- Carolina Picture Vocabulary Test for Deaf and Hearing Impaired Children
- Vocabulary subtest of the Wechsler Abbreviated Scale of Intelligence II

Speech

- The Northwestern University Children's Perception of Speech closed consonant perception test

Phonological abilities

- Comprehensive Test of Phonological Processing
- Rhyme Judgment Requiring Picture Selection
- Psycholinguistic Assessments of Language Processing in Aphasia
- Initial Consonant Same-Different task
- The Final Consonant Choice task

Table 4
ASHA - Degrees of hearing loss

Degree of hearing loss	Hearing loss range (dB HL)
Normal	-10 to 15
Slight	16 to 25
Mild	26 to 40
Moderate	41 to 55
Moderately severe	56 to 70
Severe	71 to 90
Profound	91+

Note: (ASHA, 2016).

implantation at an early age had a lower performance in phonological awareness compared with the normal-hearing controls. However, Svirsky, Robbins, Kirk, Pisoni, and Miyamoto (2000) reported that cochlear implantation at an early age can improve language development when compared with children with conventional hearing aids. Furthermore, Figueras et al. (2008) reported that children with cochlear implants have a better response to auditory stimuli as well as improved speech and language skills compared with children with other hearing aids. Such differences in language development may contribute to the variability of the results of the studies included, not only between children with hearing loss and children with normal hearing, but also among children with different types of hearing aids.

The performance of deaf children in vocabulary tasks varied between the studies included. Grammar difficulties in children with hearing loss were also reported (Al-Salim et al., 2020; Daza et al., 2014; Figueras et al., 2008; Nittrouer et al., 2012). Variations in grammar skills in children with hearing loss can be attributed to greater delays in their syntax acquisition and difficulties in hearing essential morphemes compared with children with normal hearing, which represent an additional barrier in the learning of new words (Moeller & Tomblin, 2015). Such struggles with the acoustic-phonetic properties of spoken language mean that children with hearing loss have a limited access to linguistic input and, as a consequence, a reduction in language experience (Moeller & Tomblin, 2015).

Reading skills and mathematical problem solving are associated with language development, and children with hearing loss had less efficient reading and mathematical problem-solving skills; however, children with hearing loss were able to increase their reading ability via alternative routes, such as visual attention and visual memory (Daza et al., 2014; Nunes et al., 2009). Children with normal hearing, as well as those with hearing loss, have been shown to use similar strategies for mathematical problem solving (Pagliaro & Ansell, 2012).

Types of hearing loss, hearing aids and communication skills

Although some studies employed the same parameters to evaluate hearing loss, such as PTA, there was a great variability between the studies in the degree of hearing loss; indeed, some authors selected their own thresholds and did not consider the parameters established by the ASHA (2016) or the World Health Organization (2020). Hearing aids and cochlear implants also vary in their time of use, and there is no clear information regarding their functional aspects, which challenges the validity of this comparison. Nittrouer et al. (2012) identified moderately strong correlations between the age of implantation and both phonemic awareness and auditory comprehension. In addition, Niparko et al. (2010) found better language comprehension and expression in children with earlier cochlear implants. Therefore, the longer use of cochlear implants improves phonological awareness (Nittrouer et al., 2012). Additionally, Nicholas and Geers (2007) found higher performance levels in children who received a more advanced implant technology. This means that technological differences in cochlear implants used could contribute to variations in performance.

While some studies were limited to children with unilateral hearing loss only (Ead et al., 2013), others compared children with unilateral hearing loss, bilateral hearing loss, and those with cochlear implants within the same study (Al-Salim et al., 2020). Considering that children with unilateral hearing loss may have a normal hearing level in the unaffected ear, their performance is not comparable with that of children with bilateral hearing loss (Lieu, 2004). Differences between children with unilateral hearing loss and children with bilateral hearing loss or normal hearing are largely due to the change from binaural to monoaural sound inputs to the brain. This has been shown to affect the development of cognitive functions and the ability to localize sound (Lewis, Smith, Spalding, & Valente, 2018; Lieu, 2004; Schmithorst, Plante, & Holland, 2014).

It is noteworthy to mention that, in one study, children who used Spanish sign language were compared with spoken Spanish rehabilitated subjects, and their performance was similar to those reported by other authors (Daza et al., 2014). Other work included children that used native American sign language and showed no significant difference in EFs compared to children with cochlear implants, children with hearing loss, and normal hearing peers (Hall et al., 2018).

Children with hearing loss have developmental differences regarding communication skills and cognitive abilities depending on the etiology, degree of hearing loss, family support, early diagnosis, and type and time of rehabilitation, which make it difficult to compare them under the same criteria. However, defining a global developmental trend in executive function in this population outlines their strengths and weaknesses which can be used to better direct their rehabilitation (Korver et al., 2017; Niparko et al., 2010; The Joint Committee of Infant Hearing, 2019).

Executive function performance in hearing loss

The performance of children with hearing impairment on the tasks related to EF, such as working memory, inhibition, cognitive flexibility, and attention, tended to be inconsistent among the studies reviewed. Such inconsistencies could be attributed to the use of different methods of evaluation. (Hall, Eigsti, Bortfeld, & Lillo-Martin, 2017), which highlights the need for a standardized tool for this population.

Out of the fifteen articles analyzed, only three reported no significant differences between the population groups studied. One of them studied children with cochlear implants compared to children with hearing aids; therefore, there was no normal hearing control group to compare them with (Surowiecki et al., 2002). Another one focused on specific aspects such as comparing good and bad readers regarding phonological skills (Daza et al., 2014), while the other used tests like the Spectral-temporally modulated ripple test which is specific for the population with cochlear implants with no comparable results to normal hearing population (Kirby et al., 2019).

Kronenberger, Pisoni, Henning, and Colson (2013) mentioned that despite the prolonged use of cochlear implants, the performance in EFs, particularly working memory, verbal fluency, inhibition, and attention, was lower in deaf individuals compared to their normal hearing peers.

These findings are consistent with the period of deafness that occurs prior to the diagnosis and intervention which represent a critical moment in neurodevelopment. This means that children with hearing loss are deprived of important auditory information that influences language development (Kronenberger et al., 2013).

The studies that used the BRIEF and BRIEF-P showed that parents reported a lower performance in tasks related to attention, inhibitory control, and working memory; some authors also included shifting attention in this list (Beer et al., 2014; Hall et al., 2018; Kronenberger et al., 2020). These findings have been supported by other reports that children with cochlear implants presented with difficulties in working memory and inhibitory control scales, as well as in the behavioral regulation index, according to parent reports (Beer et al., 2011). Kronenberger et al. (2013) established that verbal skills are directly involved with EFs; therefore, children with hearing loss who present a delay in language acquisition are expected to have suboptimal development in processes used for directing and controlling thoughts and behavior, thereby explaining the parent-reported deficit in areas such as inhibitory control. It is worth noting that the articles included in this review only report the parents' perception of children's performance. Other studies have included both parent and teacher reports and have highlighted the differences between them regarding specific EFs. Sabat, Arango, Tassé, and Tenorio (2020) attribute this disagreement to the different skills that children are expected to acquire in the corresponding environment. As a result, children who are exposed to a constant learning of new concepts, as occurs in a classroom environment, require EF such as inhibition and cognitive flexibility to adapt effectively. In contrast, at home children are expected to develop more predictable adaptive skills involving other EF domains such as working memory (Sabat et al., 2020). Therefore, EF deficits can be perceived as more or less severe depending on the demand in the different settings.

Contributions and future implications

Given the range of causes and degree of hearing loss, time of diagnosis, and beginning of rehabilitation therapy, performance in EF is expected to vary. Furthermore, hearing loss in developed countries is mainly attributed to genetic causes, whereas in developing countries are more common hearing loss secondary to infectious and other preventable causes (Korver et al., 2017).

As seen in Table 3, a wide range of assessment tools were used. This indicates that there are no uniform criteria for the evaluation of EF in children with hearing loss. Despite the variability of assessment tools employed in the evaluation of this population, the selected studies indicated that children with hearing impairment had a lower performance in working memory, inhibition, cognitive flexibility, and attention measures (Botting et al., 2017). These findings highlight the importance of developing or adapting an objective, reliable, and standardized evaluating tool to assess EF according to this population's specific characteristics.

Despite the variability of tests and types of studies evaluated, there are evident weaknesses in EF performance in this population. This represents a therapeutic and rehabilitation target for them to access better long term educational and professional opportunities. Furthermore, a multidisciplinary team is required to improve the understanding of parents about their children's condition, such as the John Tracy Center. They provide structured support programs according to age-group, family structure, and specific individual characteristics (JTC, 2021).

Funding

None.

Conflict of interest

The authors declare they have no conflicts of interest.

Acknowledgments

We thank Nia Cason, PhD, from Edanz Group (https://en-author-services.edanzgroup.com/ac) for editing a draft of this manuscript.

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APPENDIX. Supplementary Data 1. Data extraction

Author / Year	Title	Design	No. subjects	Population description	Assessment test	Overall results
2013 2013	Pilot study of C cognition in children with unilateral hearing loss	Case-control	4	14 children between 9 to 14 years (6 boys, 8 girls); 7 cases had severe to profound unilateral sensorineural hearing loss, defined as PTA (500, 1000, 2000, 4000 Hz) ≥ 70 dB hearing level in the affected ear and PTA (500, 1000, 2000 Hz) < 20 dB hearing level in the better hearing ear with a threshold at 4000 Hz < 30 dB. 7 controls, siblings of the probands, with normal hearing, defined as PTA (500, 1000, 2000 Hz) < 20 dB hearing level with a threshold at 4000 Hz < 30 dB.	Wechsler Abbreviated Scale of Intelligence (IQ). Comprehensive Test of Phonological Processing (phonological processing). Executive function: Letter Span Tasks (simple verbal working memory); Counting Span Tasks (complex verbal working memory); Go/No-Go task (attention, inhibition). Letter classification tasks (processing speed).	Children with unilateral sensorineural hearing loss performed lower than controls in complex verbal working memory and phonological processing. Two verbal deficits were associated with unilateral hearing loss, as follows: 1. A decrease in the precision and efficiency of verbal processing when listening to unfamiliar verbal information. 2. Problematic executive function control when maintaining verbal information while simultaneously processing incoming and irrelevant verbal information. This demonstrates a low performance in double-task control networks in children with hearing loss.
Daza et al. 2014	Language skills C and nonverbal cognitive pro- cesses associat- ed with reading comprehension in deaf children	Cross-sectional	06	30 subjects (18 boys, 12 girls) aged between 8 and 16 years (mean age 10.7 ± 1.6 years) diagnosed with hearing loss before the age of 2 years. Out of the 30 children, 29 had normal-hearing parents, more than half had congenital hearing loss. 25 had sever-to-profound hearing loss and 5 had mild-to-moderate hearing loss. 23 of them communicated in spoken Spanish and 7 used Spanish sign language. Half of the subjects (15) used cochlear implants, and the other half used conventional hearing aids.	Language: Rhyme Judgment Requiring Picture Selection, Psycholinguistic Assessments of Language Processing in Aphasia (EPLA) (Phonological knowledge). Carolina Picture Vocabulary Test for Deaf and Hearlary). Executive functions: Child Neuropsychological Maturity Questionnaire (CUMANIN) computerized version (attention). Visuospatial memory, Kaufman Assessment Battery for Children (K-ABC) (spatial memory). Memory, of Faces task (short-term visual memory). Wechsler Intelligence Scale for Children-IV (non-verbal reasoning and concept formation). K-ABC (visuo-motor response).	There was no significant between-group difference in phonological skills. Good readers had a larger vocabulary and higher scores in attention, memory, and other executive functions. This suggests that although phonological skills were variable, children with hearing loss can achieve good levels of reading through an alternative route that is not based on spoken language, but rather on non-verbal cognitive processes such as visuospatial attention, visuospatial working memory, and other executive functions.
Al-Salim et al. 2020	Performance C of children with hearing loss on an audiovisual version of a nonword repetition task	Cross-sectional	100	100 subjects aged between 6 and 18 years, monolingual English speakers. 35 normal-hearing subjects (21 boys, 14 girls). 22 mild bilateral hearing loss (10 boys, 12 girls). 17 unilateral hearing loss (12 boys, 5 girls). 26 cochlear implant users (13 boys, 13 girls).	Language: Vocabulary subtest of the Wechsler Abbreviated Scale of Intelligence, second edition (WASI-II) (vocabulary). Nonword repetition task (NWR) (language). Executive functions: WASI-III (non-verbal problem solving). Counting Recall subtest of the Automated Working Memory. Flanker Inhibitory Control and Attention subtest of the NIH Toolbox (inhibition and attention).	Hearing loss altered phonological sensitivity, which resulted in decreased language skills when limited to an auditory stimulus. Performance improved when visual cues were included. All groups with hearing loss scored significantly lower on vocabulary and working memory domains than normal-hearing subjects, as well as in the executive functions test (inhibition and attention), with a moderate-to-large effect size.

Supplementary Data 1. Data extraction (continued)

Author / Year	Title	Design	No. subjects	Population description	Assessment test	Overall results
Kirby et al. 2019	Cognitive Abilities Contribute to Spectro-Temporal Discrimination in Children Who Are Hard of Hearing	nal	24	24 subjects (13 girls, 11 boys) between 6 and 13 years old with bilateral sensorineural hearing loss who had used a hearing aid for at least 1 year. The hearing threshold was established with a three-frequency PTA average of 39.4 dB for the right ear and 39.9 dB for the left ear.	Language: Spectral-temporally modulated ripple test (SMRT). Aided Speech Intelligibility Index (SII) (language). Non-verbal intelligence: WASI. Executive functions: Dimensional Change Card Sort (DCCS). NIH Toolbox Cognitive Assessment Battery (cognitive flexibility). Automated Working Memory Assessment (working memory). Nonword repetition tasks (verbal working memory).	Better general cognitive abilities were associated with a greater ability to discriminate spectro-temporal waves, regardless of the age of the children or SII. There was a partial correlation between non-verbal intelligence and visual working memory, whereby discrimination thresholds improved with higher non-verbal intelligence. There were no significant between-group differences in verbal working memory or language skills.
Beer et al. 2014	Executive functioning skills in preschool-age children with cochlear implants	Case-control	45	45 subjects between 3 and 6 years old. 24 (10 girls, 14, boys) with hearing loss diagnosed before 36 months of age, had profound bilateral hearing loss (defined as > 90 dB in the better-hearing ear), had received a cochlear implant before 3 years of age, lived in a monolingual English-speaking atmosphere. 21 (8 girls, 13 boys) with normal hearing.	Language: The Preschool Language Scale 4th edition (language). Non-verbal intelligence: The Picture Similarities subtest of the Differential Ability Scales. Executive functions: Memory for Designs subtest of the NEPSY-II (non-verbal visual memory). Attention Sustained subtest of the Leiter International Performance Scale (inhibition, concentration, attention). Berry Developmental Test of Visual-Motor Integration (organization, visuomotor integration). BRIEF-P (inhibition, working memory, planning/organizing).	The BRIEF-P results revealed that parents of children with profound hearing loss with a cochlear implant had a worse performance in executive processes, especially in attention and inhibitory control, as well as working memory. There were no differences between normal-hearing children and any of the hearing-impaired groups in terms of visual memory and organization, or in planning and organization domains, as reported by parents.
Surowiecki et al. 2002	Cognitive pro- cessing in chil- dren using co- chlear implants: the relationship between visual memory, atten- tion, and exec- utive functions and developing language skills	Case-control	84	48 subjects aged between 6 and 14.5 years with an average age of 9.39 years. All participants attended an oral-aural educational session, and all had prelingual hearing loss. No subjects were color blind. 24 nucleus cochlear implant users. 24 hearing aid users (11 children with a profound loss, 8 children with a severe loss, and 5 children with a moderate-to-severe loss). The two device groups were matched for age and sex.	Cognitive Abilities: Cambridge Neuropsychological Test Automated Battery (CAN-TAB). Language, Vocabulary, and Speech Perception Assessments: Bamford-Kowal-Bench (BKB) Sentences List, Consonant-Nucleus-Consonant (CNC) word lists, the Northwestern University Children's Perception of Speech (NUCHIPS) closed set consonant perception test, Peabody Picture Vocabulary Test Revised (PPVT-R), Clinical Evaluation of Language Fundamentals (CELF) preschool test, CELF-3. Visual Memory Subtest: The Pattern and Spatial Recognition test, the Delayed Matching to Sample (DMTS), the Paired Associates (PA) test. Attention and Executive Functions Subtest: The Intradimensional/Extradimensional Set-Shifting Task (ID/ED), The Spatial Span test, The Dower of London test.	There were no significant differences in various measures of attention, visual memory, and executive functioning between children who used either a cochlear implant or hearing aids, even when only those with profound hearing impairment were compared with matched implant users. This finding suggested that the cognitive skills in children with cochlear implants are not aided by the additional auditory information provided by the implant. The study demonstrated that the CELF-3 subtests require memory skills to various extents.

Supplementary Data 1. Data extraction (continued)

			No.			
Author / Year	Title	Design	subjects	Population description	Assessment test	Overall results
Xuan et al. 2018	Decision-Making in Adolescents with Profound Hearing Loss	Case-control	22	36 deaf adolescents. lowa Gambling Task. Mean age and years of education of the 36 deaf participants were 17.14 ± 2.05 years old and 8.53 ± 1.73 years (23 boys, 13 girls). Game of Dice Task. Mean age and years of education of the 36 deaf participants 16.99 ± 1.95 years and 8.25 ± 1.83 years (21 boys, 15 girls) 36 normal-hearing adolescents, participants were the same for the two tasks (age: 16.72 ± 1.14 years; years of education: 9.36 ± 1.25).	Intelligence test: Raven's Progressive Matrices, the Hiskey-Nebraska Test of Learning Aptitude. Decision-making tasks: Iowa Gambling Task, Game of Dice Task.	The deaf group performed significantly lower than the normal-hearing group on the Raven's Progressive Matrices and the Hiskey-Nebraska Test of Learning Aptitude. Furthermore, on the lowa Gambling Task and Game of Dice Task, the deaf group more often chose to make high-risk decisions, with higher gains but also higher losses.
Nittrouer et al. 2012	Emergent Literacy in Kindergart-ners with Cochle-ar Implants	Case-control	25	35 children with sensorineural hearing loss with three-frequency pure-tone averages > 50 dB HL in the better ear. 27 children with severe-to-profound hearing loss with a cochlear implant and 8 children with moderate hearing loss with hearing aids. 17 normal-hearing children.	Phonological awareness: Syllable counting, the Initial Consonant Same-Different task, the Final Consonant Choice task. Emergent literacy: The Qualitative Reading Inventory version 1. Oral language skills: The auditory comprehension subtest of the Preschool Language Scales-4, The Expressive One-Word Picture Vocabulary test, a 20-min language sample was recorded from each child, consisting of several personal narratives. Executive functions: Short-term memory task (six words: ball, coat, dog, ham, pack, and rake); for rapid serial naming, the color and object naming subtests of the Comprehensive Test of Phonological Processing were used.	Normal-hearing children performed significantly better than children with a cochlear implant in emergent literacy and in all three measures of the oral language skills. On the auditory comprehension and expressive vocabulary measures, children with hearing aids performed similarly to children with normal hearing, on the measure of narrative skills. There was no significant between-group difference in the executive functions tests. There were no significant differences for any measures between the hearing aid and cochlear implant groups.
Figueras et al. 2008	Executive Func- Cross-sectional tion and Language in Deaf Children	Cross-sectional	69	22 deaf children with a cochlear implant (mean age 9.8 ± 1.6 years). Mean length of implant use was 6.4 ± 2 years. 19 with profound hearing loss. 25 deaf children with hearing aids (mean age 10.8 ± 1.5 years) (4 with moderate hearing loss, 10 with severe hearing loss, 7 with profound hearing loss). 86% of children with cochlear implants and 56% of children with hearing aids were orally educated. The remaining children used the total communication approach. 22 normal-hearing children (mean age 10.2 ± 1.3 years).	Language: The British Picture Vocabulary Scale (BPVS) Long Form, the Test for Reception of Grammar, version 2 (TROG-2). Executive functions: Tower from the NEP-SY battery. Visual attention from the NEPSY battery. Design fluency from the NEPSY battery. Card and tap from the NEPSY battery. Day-Night and One-Two tasks. Card Sorting test from the D-KEFS battery.	Normal-hearing children performed significantly better in the BPVS and in the TROG-2 than the hearing aid and cochlear implant groups. TROG-2 results between the cochlear implant and hearing aid groups. Normal-hearing children performed significantly better on the Card Sorting attempted score, Card Sorting correct score, Day-Night/One-Two errors, Day-Night/One-Two time, Knock and Tap, and Tower rule violatine, Knock and Tap, and Tower rule violations than the hearing aid and deaf children. There were no significant between-group differences in the Tower, Visual Attention, Design Fluency, Design Fluency, repetitions, and Card Sorting repeated sorts scores. between the groups.

Supplementary Data 1. Data extraction (continued)

Author / Year	Title	Design	No. subjects	Population description	Assessment test	Overall results
Holf et al. 2013	Developmental Effects of Family Environment on Outcomes in Pediatric Cochlear Implant Recipients	Cross-sectional	29	Families of 59 prelingually deaf children with cochlear implants and without any additional disability were enrolled in the study. The parents had completed the FES as part of a longitudinal study on cochlear implant outomes. Children were between the ages of 1 to 18 years and length of device use ranged from 0.5 - 16 years. Children were separated into two groups: Preschool-Age group (younger than 5 years who had not started kindergarten; n = 20). School-Age group (older than 5 years who were in kindergarten or grade school; n = 39).	Family Environment Scale. Language: Peabody Picture Vocabulary Test-4. The Preschool Language Scales-4. The Clinical Evaluation of Language Fundamentals-4. Executive functions: Behavior Rating Inventory of Executive Function (Preschool version) and Behavior Rating Inventory of Executive Function.	Families of school-age children with cochlear implants reported higher levels of inhibitory control than families of preschool-age children. Both groups were similarly delayed in their language and vocabulary development. Regarding executive function, the sample exhibited more difficulties than expected in inhibiting behaviors, shifting attention, and working memory. Families with high levels of support and those with low conflict and high cohesiveness and organization tended to have preschoolers with fewer problems in emotional control. Families who had low conflict and high cohesiveness and organization tended to have school-age children with fewer problems with inhibitory control.
Nunes et al. 2009	Deaf Children's Informal Knowl- edge of Multipli- cative Reasoning	Case-control	Study 1: 106 Study 2: 27	Study 1: 28 deaf children. Mean age 6.5 years, in grades 1 and 2. Were recruited from seven schools for the deaf or conventional units for the deaf and hard of hearing. 12 deaf children had cochlear implants, 3 had moderate hearing loss, and 13 had severe-to-profound hearing loss, 78 normal-hearing children. Mean age of 6.2 years, and in grade 1. Were recruited from three schools with a varied intake in terms of their socio-economic background. Study 2: 27 deaf children with a mean age of 6.6 years. Recruited from seven special schools and mainstream schools with units for the deaf.	Matrices subtest of the BAS.	Deaf children performed worse than their nor- mal-hearing peers in multiplicative reasoning at the beginning of primary school. This was considered a performance discrepancy rath- er than a competence discrepancy. With a brief intervention, performance of the deaf children significantly improved to the same level as normal-hearing children when they were matched for cognitive ability.
Pagliaro & Ansell 2012	Deaf and hard of hearing students' problem-solving strategies with signed arithmetic story problems	Cross-sectional	20	59 children from 9 schools for the deaf and Individual interviews. hard of hearing across the United States. Of the 59 children, 3 had mild hearing loss, 5 had moderate hearing loss, 18 had severe hearing loss, and 33 had profound hearing loss, 34 children had at least one deaf parent, 21 had no deaf parents, and the remaining 4 were not known (2) or not reported (2). The children were aged from 5 to 9 years, with a mean age of 7.4 years. Children used American Sign Language.	Individual interviews.	For problem solving, both deaf children and normal hearing children used the same strategies, including modeling based on real events and counting. This was the most common strategy adopted for all kinds of problems.

Supplementary Data 1. Data extraction (continued)

			No.			
Author / Year	Title	Design	subjects	Population description	Assessment test	Overall results
Hall et al. 2018	Executive Function in Deaf Children: Auditory Access and Language Access	Case-control	116	116 children between 5 and 12.11 years. 45 normal-hearing children. 45 children with hearing loss with sign language. 26 children with a cochlear implant.	Brief interviews. Tower subtest of the NEPSY battery. Attention-Sustained subtest of the LIPS. Go/No-Go task. Corsi block task.	There was a significant difference in the BRIEF score for inhibition and working memory being lower in children with hearing loss. No significant differences were found on the Tower task, Attention-sustained, Go/No-Go, or Corsi block task scores. Go/No-Go and Corsi block task performance improved with age in children with hearing loss.
Beer et al. 2011	Executive func- Cross-sectional tion in everyday life: implications for young cochlear implant users	Cross-sectional	45	45 children with a cochlear implant children.	Executive functions: BRIEF. Speech and language: Clinical Evaluation of Language Fundamentals fourth edition, Peabody Picture Vocabulary Test, Lexical Neighborhood Test, Hearing in Noise Test for Children.	As revealed by the BRIEF, children with co- chlear implants performed above the mean t-score on the Inhibition and Working Mem- ory scales, and on the Behavioral Regulation Index.
Kronenberger et al. 2020	Longitudinal Cohort Development of Executive functioning and Spoken Lan- guage Skills in Preschool-Aged Children with Co- chlear Implants	Oohort	2	40 normal-hearing children. 41 children with a cochlear implant.	Nonverbal intelligence: Picture Similarities subtest of the Differential Ability Scales—Second Edition. Language: Peabody Picture Vocabulary Test–Fourth Edition, Preschool Language Scale—Fourth Edition. Executive functions: Forward Memory subtest of the Leiter International Performance Scale—Revised, Digit Span Forward subtest of the Wechsler Intelligence Scale for Children—Third Edition, Attention Sustained Total subtest scaled score of the Leiter-R, Behavior Rating Inventory of Executive Function—Preschool, Behavior Rating Inventory of Executive Function.	Children with cochlear implants had lower language scores than their normal-hearing peers, but scored similarly in inhibition, attention, and working memory. Regarding the BRIEF parent reports, children with cochlear implants scored lower than their normal-hearing peers in inhibition and working memory.