



Executive functions in preschool and school-age cochlear implant users: do they differ from their hearing peers? A systematic review and meta-analysis

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Abstract

Purpose Executive functions (EF) play a fundamental role in planning and executing goal-driven behaviours. The purpose of this systematic review and meta-analysis was to investigate EF skills mastered by preschool/school-age cochlear implanted children (CIC) without morpho-functional abnormalities and to compare their outcomes with typically hearing children (THC).

Methods Bibliographic search for observational studies of any language/date up to 16 December 2022 was performed with the following electronic databases: PubMed, Scopus, and Web of Science. After removal of duplicates, 2442 records were subjected to a three-stage screening process and 83 potentially eligible articles were identified. A total of 15 studies was included in the final analysis: 9 articles directly meeting the eligibility criteria plus 6 more studies thanks to the authors sharing their data set, specifically for participants who met present inclusion criteria.

Results Meta-analysis showed a statistically significant difference only for verbal short-term memory, whereas group differences for visuospatial short-term memory and verbal/visuospatial working memory were not significant. For fluency skills, meta-analysis revealed statistical significance for the semantic fluency task but not for the rapid naming test. Qualitative analysis reflected group similarities in flexibility but CIC's difficulties in auditory attention/planning skills. Controversial findings for inhibitory control skills were observed.

Conclusions EF performance comparisons between CIC and THC show inter-skill and inter-test variances. Due to the paucity of existing studies, present findings should be interpreted with caution. Future research in this domain is strongly recommended.

Keywords Preschool · School-age · Cochlear implant · Executive functions · Systematic review · Meta-analysis

Introduction

Executive functions (EF) are a set of high order cognitive processes that are fundamental for planning and successfully executing goal-driven behaviours. They play a critical

role in individuals' social, emotional, and physical health and include multiple domains, such as short-term memory (STM), working memory (WM), sustained and selective attention, inhibition skills, cognitive or behavioural flexibility and planning/problem solving skills [1].

Auditory experiences are fundamental for the EF development, while auditory deprivation may alter them [2]. Cochlear implantation helps to enable functional hearing and spoken language development in children with severe-to-profound hearing loss [3]. Likewise, it has been reported how cochlear implanted children (CIC) are able to develop EF skills within the average range of typically hearing children (THC). However, some CIC (~ 1/3) seem to be at risk to show EF delays, with a rate that is 2–5 times greater than that observed in THC [4].

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A recent systematic review (SR) by Charry-Sanchez et al. [5] investigated EF skills in deaf or hard of hearing children (DHHC) with cochlear implants (CI) or hearing aids. However, heterogeneity in the study samples and task variations did not allow them to perform a meta-analysis. Nevertheless, the authors concluded that independently from the auditory prosthesis, published data on EF in DHHC were controversial: some studies reported no significance, whereas some others showed statistical significance in outcome differences between DHHC and TH. Another recent SR by Akçakaya et al. [6] studied STM and WM in long-term CI users. Performing a meta-analysis, the authors found evidence of significantly lower verbal STM and WM performances in adolescent and young adult CI users than those in TH peers. However, no conclusions were achievable for visuospatial STM and WM due to insufficient data.

To date, no SR is available on EF performance in CIC, specifically for the preschool or school-age period. Hence, there is a lack of clinical evidence for an important developmental period when timely and early intervention could be more effective in improving CIC's performance [7]. Such a question is particularly critical, since most of the existing studies on EF included CIC with morpho-functional abnormalities such as Mondini malformation, auditory neuropathy, and meningitis that may have negatively affected postoperative outcomes [4]. Indeed, it is not possible to know if significant outcome differences are resulted from neurophysiologic alterations that are typical to severe-to-profound hearing loss and/or from CI technological constraints or from contextual alterations.

The present SR with meta-analysis was designed to investigate the impact of CI on EF skills mastered by children with bilateral severe-to-profound congenital hearing loss without cochlear morpho-functional abnormalities. The following research questions were considered: do preschool and school-age congenitally deaf CIC without any additional disabilities perform similar to their TH peers in memory (STM and WM), attention, inhibition, flexibility, and problem-solving or are there significant performance differences between the two groups? Such findings are thought to provide useful knowledge to implement appropriate intervention programs during two developmental phases, where specific training may lead to significant performance improvements [7].

Methods

Protocol and registration

This SR and meta-analysis were performed following the instructions of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA—<http://www.prisma-statement.org>. Accessed 9 December 2021) and

was recorded in PROSPERO database (<http://www.crd.york.ac.uk/PROSPERO>) with the registration number CRD42022334543. The methods are reported according to the Preferred Reporting Items for SRs and Meta-Analyses (PRISMA) checklist. The present protocol is exempt from ethics approval, because this SR and meta-analysis retrieved and synthesized data from already published studies in which informed consent/ethical approval has already been obtained by the investigators.

Eligibility criteria

The PICO framework was followed to define the criteria for participants' inclusion, intervention, study design, and outcomes.

The participants were DHHC with bilateral severe-to-profound hearing loss (> 70 dB HL in the better ear) that was congenital or arose prior to 3 years of age. Their chronological age ranged from 3.0 to 12.9 years at the time of testing. Exclusion criteria were the presence of any associated neurocognitive comorbidity or developmental delays, a nonverbal IQ score ≤ 2 SD from the normative mean and the presence of any neuro-audiological/surgical complications (such as cochlear/cochlear nerve malformation, partial electrode insertion, auditory neuropathy) that may negatively impact postoperative outcomes. The intervention or exposure of interest was unilateral or bilateral cochlear implantation in DHHC within preschool age.

The control group was represented by TH with the following characteristics: same chronological age with CIC (3–12 years at the time of testing), pure-tone average thresholds (500–4000 Hz) < 20 dB HL and a monolingual environment. Exclusion criteria for TH were the presence of associated neurocognitive comorbidity or developmental delays and a nonverbal IQ score ≤ 2 SD from the normative mean.

Outcomes regarded the scores obtained at the following EF tests:

- Neuropsychological tests for attention skills: auditory or visual attention sustained/selective/shifting tasks.
- Neuropsychological tests for phonological, visuospatial and verbal STM and WM: i.e., nonverbal visual memory, forward and backward spatial span, block tapping task, location span, odd-one-out location span visual digit span, forward and backward digit span, reading span, counting span, sentence completion and recall, non-word repetition or recall tasks.
- Neuropsychological tests for verbal and visual fluency: i.e., rapid digit or naming tasks, retrieval fluency tasks, visual matching tasks, visual-perceptual fluency-speed and visual-motor fluency-speed.
- Neuropsychological tests for inhibitory control skills: i.e., stroop tasks, go-no-go tasks.

- Neuropsychological tests for flexibility skills: i.e., card sort tasks.
- Neuropsychological tests for planning/problem solving skills: i.e., tower test.

Cohort and case–control studies were considered as eligible study design. Expert opinions, practice guidelines, case reports, case series, conference abstracts, and book chapters were excluded.

Literature search

The search was performed through PubMed, Scopus, and Web of Science, using MeSH terms, keywords, or text searches on 16 December 2022, with different spellings and synonyms regarding CIC and the specific subdomains included in the executive functioning (child* AND cochlear implant* AND flexibil* OR attention* OR inhibit* OR working memory OR verbal fluenc* OR problem solving OR planning). The search string was adapted to fit the search criteria of each database (Supplementary Table 1). No language or date restrictions were applied. Grey literature was also searched through ClinicalTrials.gov and Google Scholar. The electronic search was completed by manual search of the references from articles identified for inclusion or from relevant review articles.

Study selection

With the aim to reduce random bias and errors, study identification was carried out by three independent review authors (VB, AS, GM) and selection/evaluation was performed by other two independent review authors (PM, IG). VB, AS, GM identified titles and abstracts of the studies using the search strategies, reference lists and grey literature search and removed duplicates. PM and IG reviewed them in terms of inclusion and exclusion criteria. A full-text analysis was performed for the articles that met the screening criteria. Disagreements on eligibility was solved by consensus or by a third reviewer (HDD).

Data collection process

Data were collected by LR and checked by MN. A third reviewer (HDD) was consulted in case of disagreement between them. LR and MN maintained email contacts with the Authors of included studies to ask relevant data as well as to clarify any study/data-related questions.

Data items

A standardized form was used to extract data from the included studies, collecting the following information for evidence synthesis:

- study identification (title, authors, year of publication, country, DOI, study design).
- study population (number of participants, age at testing, sociodemographic and audiological information).
- test details for each EF task.
- outcomes and results for every EF skill/measure.

Study quality and risk of bias assessment

Two review authors (CDV, AG) independently performed the evaluation of the quality and risk of bias assessment of the studies included in this SR. Discrepancies were resolved consulting a third reviewer (AS). The quality assessment was performed using the Newcastle Ottawa Scale (score range from 0 to 9) for cohort studies [8]: high quality (8–9 stars), medium quality (6–7 stars), and low quality (1–5 stars). The single item score and the total score for each study was recorded. The assessment was blindly performed, masking the names of authors and the journals, avoiding any potential bias and conflicts of interests during the process.

Synthesis of results

Articles were grouped according to the EF studied. A narrative synthesis of the main findings was done by comparing subjects with/without CIs. For authors who provided continuous data (i.e., mean, and standard deviation) of the same EF task, we combined results using the standardized mean difference (SMD) and 95% confidence interval (CFI) to perform a meta-analysis of group comparisons. It was assumed that the study samples were heterogeneous; hence, an inverse-variance random-effects model was used. The Cochran χ^2 test and the I^2 metric were used to test for heterogeneity [9]. Heterogeneity was considered statistically significant at p values < 0.05 , and substantial heterogeneity was defined as $I^2 > 50\%$. Since the number of studies for each analysis was always lower than 10, the Cochrane's suggestion [10] was followed, while the small study effect, potentially caused by publication bias, was not assessed. Given the limited availability of studies across all types of EF tasks, meta-regression analyses were not performed. All analyses were performed in Review Manager (RevMan), Version 5.4 (Cochrane Collaboration).

Results

A total of 3171 records were identified for screening. Following the removal of 729 duplicates, 2442 records were subjected to a three-stage screening process which resulted in 83 potentially eligible articles. Full texts were retrieved, and characteristics of studied samples were analysed according to the SR inclusion criteria. Nine studies respected all

the inclusion criteria and were directly included in the SR [11–19].

Despite study topics that were focus of interest of the present SR, most of the reviewed studies presented the following issues: overlapping data (five different groups of potentially overlapping studies were identified: 32 papers from Indiana-Atalanta-Ohio University, 7 from Linköping University, 6 from Florida University, 2 from St Louis-Texas University, 2 from Connecticut University), and/or having some participants not meeting the SR inclusion criteria (CIC outside the present chronological age range, CIC with morpho-functional alterations of the cochlea or children with mild-to-moderate hearing loss using traditional hearing aids). Therefore, the corresponding authors of these articles were contacted by email to ask confirmation of overlapping data or to request data sets specifically for the participants who met present inclusion criteria. A total of 43 emails were sent to the corresponding authors. Eleven authors (a total of 32 studies) confirmed overlapping data for their studies. These studies had also some subjects who did not fit the present inclusion criteria. Since the authors rejected our request to send their data excluding the participants who did not meet the present SR inclusion criteria, the corresponding studies were all excluded. Twenty-six authors (a total of 36 studies) did not respond, even though a second attempt was made to contact them. Therefore, also these studies were excluded as well.

Six authors shared their data set specifically for their participants who met present inclusion criteria and were included in the SR [20–25]. Finally, a total of 15 studies was definitively considered for the present SR (Fig. 1).

Characteristics of the included studies

Detailed information for the included studies is reported in Table 1. Quality of the included studies ranged from a score of 5 to 8 at New Castle–Ottawa scale (Table 2). Among a total of 15 included studies, 3 investigated attention skills [11, 15, 17], 4 did fluency skills [12, 14, 22, 24], and 11 did memory competencies [11–14, 16, 18–21, 23, 24], while 5 evaluated inhibitory control [20, 22–25], 2 did flexibility/set shifting [21, 23] and 2 did planning/problem solving skills [21, 24].

Due to the small number of studies and heterogeneity in tasks, meta-analysis was possible only for memory and fluency skills. Findings for attention, inhibitory control, flexibility, and planning skills could be only qualitatively described.

Attention skills

Only three studies (Table 3) referring to attention met the inclusion criteria [11, 15, 17].

Huber et al. [11] investigated selective visual attention through the Coding task of the German version of WISC, a task where the child is requested to transcribe rows of digit-symbol codes as quickly as possible. Data were available from 39 CIC and 17 TH peers: the CI group performed better than the TH one (CI mean score 11.1 with an SD = 2.6 vs TH mean score of 9.8 with an SD = 0.96). Performance differences were not statistically significant.

Sanei et al. [15] investigated auditory sustained attention on a group composed of 18 CIC, with a mean age of 9.43 years (SD = 0.84), all implanted below the age of 24 months, and compared their performance with that from 40 TH peers. CIC performed significantly worse than TH in all the measures (total score, inattention and impulsive errors, reduction span index).

Chen et al. [17] studied auditory selective attention in two different conditions (without distractors and with visual distractors) in a group of 22 CIC (age range from 5 to 8 years), comparing their skills with those of 16 TH peers. All CIC had bilateral congenital profound hearing loss and received their CI between 1.5 and 5 years of age, with only 4 subjects implanted within 2 years of age. Authors found that CI users had significantly longer reaction times than those from TH peers in both conditions and a poorer discrimination of auditory targets in the presence of visual distractors. They interpreted such CIC findings as indicative of an impaired auditory selective attention and a capacity-limited attentional mechanism across modalities, where visual stimuli interfered with auditory perception when visual and auditory stimuli were incongruent.

Memory skills

Memory represented one of the domains, where more studies were included in the present SR (Table 4). A meta-analysis was performed for both STM and WM skills based on verbal and visuospatial memory tasks.

Regarding STM, ten studies were included: eight studies investigated verbal STM, either using non-word repetition tasks [12, 14, 16, 20] or using forward digit span tasks–DSF [11, 14, 19, 24], while two studies evaluated visuospatial STM [11, 20]. Their forest plots are reported in Fig. 2.

For the non-word repetition abilities, the studies reported accuracy measures and altogether allowed us to analyse an overall sample of 96 TH and 86 CIC. The results of the meta-analysis (Fig. 2A) revealed a statistically significant difference between TH and CIC ($Z = 3.60$, $p = 0.0003$). CIC showed an average accuracy score lower than that of TH peers (SMD = -2.91 ; 95% CFI from -4.49 to -1.32). The effect size was large [26]. I^2 value (92%) reflected a large heterogeneity between the included studies.

Four studies were included in the meta-analysis of DSF for an overall sample of 60 TH and 82 CIC (Fig. 2B).

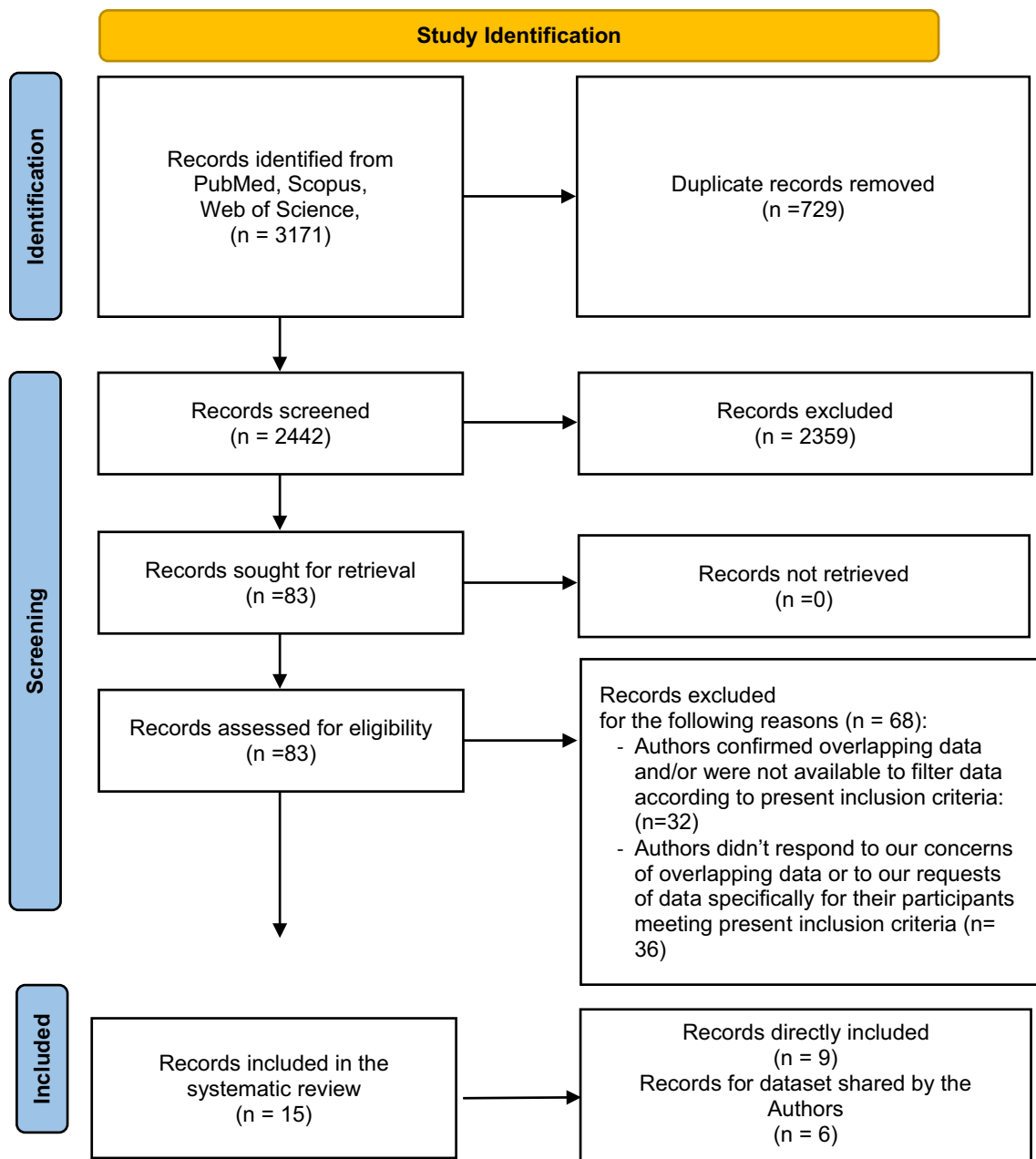


Fig. 1 Flow diagram for the study selection process

Statistically significant group differences in favour of THC were found for this aspect of memory as well ($Z=3.24$, $p=0.001$): CIC had on average a narrower span than TH (SMD = -0.93 ; 95% CFI from -1.49 to -0.37). The effect size was large [26]. Heterogeneity of the studies was moderate ($I^2=56\%$).

A different picture came from the visuo-spatial STM analysis based on the two studies (an overall sample of 46 THC and 55 CIC): CIC had a slightly lower mean span than TH peers (SMD = -0.12 ; 95% CFI from -1.03 to

0.78) but the difference was not statistically significant ($Z=0.27$, $p=0.79$) (Fig. 2C).

Eight studies were included for WM: 6 for verbal WM [11, 13, 16, 19, 23, 24], for an overall sample of 106 THC vs 120 CIC, and 2 for visuospatial WM [21, 23], for an overall sample of 135 THC vs 36 CIC (Fig. 3A, B). Botting et al. [21] included two different tasks to measure visuospatial WM (the Backward Spatial Span and the Odd one out span), but the meta-analysis was performed with the Backward Spatial Span—the same task of the second included study.

Table 1 General characteristics of the included studies for the qualitative and quantitative analyses

Authors (year)-Country	Sample size (number)	Chronological age	Gender F:M	Non verbal IQ	Hearing loss onset	Pre CI PTA	Age at CI	CI Use	Post CI PTA or speech perception	Listening mode	Communication mode	Studied EF considered for the SR
^a Grep et al. (2011)-Kentucky	CIC: 15 THC: 29	CIC: 89 (19.99) mths THC: 93 (11.38) months	CIC: 7:8 THC: 18:11	–	<3.5 years	–	32 (14.99) months	–	–	7: bimodal CIC 8: bilateral CIC	15: Oral 15: Verbal STM 15: Verbal Fluency 15: Inhibition control	Visuospatial STM Verbal STM Verbal Fluency Inhibition control
Huber et al. (2012)-Austria	Initially reported: CIC: 40 THC: 40 Real number varied in different tasks	CIC: 10.1 (1.3) years THC: 10.1 (1.3) years	CIC 21:19 THC 21:19	–	34: congenital 16: <24 months	> 70 dB	2.3 (1.2) years	7.8 (1.3) years	Sentences recognition: 75% (27.3%)	25: unilateral CIC 15: bilateral CIC	26: Oral 5: Total 9: Oral and total	Verbal STM and working memory Visuospatial STM Selective visual attention Verbal STM Verbal fluency
Lee et al. (2012)-Republic of Korea	CIC: 25 THC: 25	CIC: 64.76 (9.48) months THC: 64.36(8.53) months	CIC: 11:14 THC: 13:12	–	–	–	20.88 (3.88) months	–	–	–	–	–
López-Higes et al. (2015)-Spain	Early CIC: 19 Late CIC: 19 THC: 19	Early CIC: 116.32 (13.536) months Late CIC: 118.58 (14.072) months THC: 116.11 (14.749) months	Early CIC 7:12 Late CIC 8:11 THC 13:6	–	Prelingual (<24 months)	> 70 dB	Early CIC: 14.68–(5.69) months Late CIC: 41.89 (12.97) months	–	–	Bilateral: 47.0% early CIC and 21.1% late CIC Unilateral: 31.8% early CIC and 57.9% late CIC Bimodal: 21.1% both early and late CIC	–	Verbal working memory

Table 1 (continued)

Authors (year)-Country	Sample size (number)	Chronological age	Gender F:M	Non verbal IQ	Hearing loss onset	Pre CI PTA	Age at CI	CI Use	Post CI PTA or speech perception	Listening mode	Communication mode	Studied EF considered for the SR
^a Botting et al. (2017)-England	CIC: 33 THC: 121	CIC: 105.03 (21.57) months THC: 106.26 (16.52) months	THC 20:13 THC: 54:67	–	Congenital or <3 years	Left ear: 90.4 dB Right ear: 88.7 dB (20.1) dB	2.75 (1.15) years	–	–	12: bilateral-CIC 21: not reported	–	Visuospatial working memory Shifting Planning Verbal fluency Inhibition control
^a Marshall et al. (2017)-England	CIC: 22 THC: 124	CIC: 8.33 (1.69) years THC: 8.92 (1.5) years	CIC: 13:9 THC: 58:66	–	Congenital or <3 years	> 70 dB	2.9 (1.3) years	–	–	7: unilateral 8: not reported	14: Oral 8: oral and sign reported	Verbal fluency Inhibition control
Talli et al. (2017)-Greece	CIC: 15 THC: 15	CIC: 79.80 (14.80) months THC: 79.67 (13.85) months	CIC: 7:8 THC: 7:8	Raven CIC: 65.33 (22.55) percentile THC: 73 (20.42) percentile	Congenital or prelingual (<24 months)	> 70 dB	1.6–3.9 years	4.1 (1.7) years	–	15: unilateral-CIC 11: Oral signs 4: Oral and signs	–	Verbal working memory Verbal fluency
Sanei et al. (2018)-Iran	CIC: 18 THC: 40	CIC: 9.43 (0.84) years THC: 9.40 (0.75) years	CIC: 10:8 THC: 20:20	–	Congenital	–	<24 months	–	Speech recognition > 80%	18: unilateral 18: Oral	–	Sustained auditory attention
Akçakaya et al. (2019)-Turkey	CIC: 31 NH: 28	CIC: 101.6 (13) months NH: 101.8 (11.7) months	CIC: 17:14 NH: 14:14	Wechsler Intelligence Scale for Children-Revised (WISC-R) CIC: 112.7 (15.8) NH: 108.8 (9.9)	Congenital or prelingual (<24 months)	–	23.0 (7.3) months	78.0 (13.5) months	–	25: unilateral-CIC 6: bimodal-CIC	–	Verbal working memory Verbal fluency
Chen et al. (2019)-China	CIC: 22 THC: 16	CIC: 6.64 (0.99) years THC: 6.09 (1.5) years	CIC: 12:10 THC: 10:6	All subjects >25 percentile	Congenital	> 90 dB	Range 1.5–5 years	3.7 (1) years	Range 25–45 dB HL	22: unilateral-CIC	22: Oral	Auditory selective Attention with or without visual distraction

Table 1 (continued)

Authors (year)- Country	Sample size (number)	Chronological age	Gender F:M	Non verbal IQ	Hearing loss onset	Pre CI PTA Age at CI	CI Use	Post CI PTA or speech percep- tion	Listening mode	Commu- nication mode	Studied EF considered for the SR
Jing et al. (2019)-United States	CIC: 6 NH: 15	CIC: median 6.10 years (IQR 6 months) NH: median 5.2 years (IQR 10 months)	CIC: 4:2	Primary Test of Non-verbal Intelligence (PTONI) Median (IQR) CIC: 119.5 (20.5) NH: 121 (28)	Congenital	Range 9–21 months	–	Range 10–26 dB HL	4: bimodal CIC 5: Total 1: Total 2: bilateral CIC	5: Oral 1: Total	Non-verbal working memory
^a Figueroa et al. (2020)-Spain	CIC: 3 THC: 14	CIC: 12 years THC: 12 years	CIC: 2:1 THC: 12:2	CIC: 98.50 (1.68) THC: 101.96 ± 1.58	Congenital	1.78 (1.68) years	9 (0.28) years	–	2: unilateral CIC 1: bilateral CIC	3: Oral	Verbal working memory Visuos- patial working memory Inhibition control Shifting
Volpato (2020)- Italy	CIC: 13 THC: 13	CIC: 9.2 (range 7.9–10.8) THC: 9.0 (range 7.5–10.8) years	CIC 3:10	–	Congenital	Range 1.9–3.4 years	Range 4.5–8.6 years	≤ 30 dB HL	13: unilat- eral CIC	13: Oral	Verbal working memory
^a Jamsek et al. (2021)-Ohio	CIC: 35 THC: 62	CIC: 6.4 (1.5) years THC: 6.0 (1.6) years	CIC 19:16 THC 28:34	T score > 30 at Picture Similarities subtest of Dif- ferential Ability Scales-II	Congenital or prelingual (< 24 months)	< 3.5 years	4.9 (1.6) years	24.0 (6.0) dB HL	–	35: Oral	Inhibition control
^a De Giacomo et al. (2021)-Italy	CIC: 15 THC: 15	CIC: 8.1 (2.53) years THC: 7.99 (2.3) years	CIC: 6:9 THC: 5:10	Within 1 SD at not specified test	Congenital or prelingual (< 36 months)	1.6 (0.8) years	5.99 (2.3) years	–	12: unilat- eral CIC 3: bilateral CIC	15: Oral	Verbal STM and working memory Verbal flu- ency Inhibition control Planning

CIC cochlear implanted children, THC typically hearing children

^aReported data are not those published in the original study, but those of the CIC who respected the inclusion criteria for the present SR (database shared by the Authors)

Table 2 Quality and risk of bias assessment by New-Castle-Ottawa Scale for the included studies

Items	Selection		Comparability		Outcome		Total scores (n.)	Overall study quality
	Representativeness of exposed cohort	Selection of non-exposed cohort	Ascertainment of exposure	Demonstration of interest not present at start of study	Comparability of cohorts on the basis of the design or analysis	Assessment of outcome		
Gremp et al. (2011)-Kentucky	a	a	a	na	a	a	7	Medium
Huber et al (2012)-Austria	a	a	a	na		a	5	Low
Lee et al. (2012)-Republic of Korea	a	a	a	na		a	6	Medium
López-Higes et al. (2015)-Spain	a	a	a	na	a	a	7	Medium
Botting et al. (2017)-England	a	a	a	na	a	a	7	Medium
Marshall et al. (2018)-England	a	a	a	na	a	a	7	Medium
Talli et al. (2017)-Greece	a	a	a	na	aa	a	8	High
Sanei et al. (2018)-Iran	a	a	a	na		a	6	Medium
Akçakaya et al. (2019)-Turkey	a	a	a	na	a	a	7	Medium
Chen et al. (2019)-China	a	a	a	na	a	a	7	Medium
Jing et al. (2019)-United States	a	a	a	na		a	5	Low
Figuerola et al. (2020)-Spain	a	a	a	na	a	a	7	Medium
Volpato et al. (2020)-Italy	a	a	a	na	a	a	7	Medium
Jamsek et al. (2021)-Ohio	a	a	a	na	a	a	7	Medium
De Giacomo et al. (2021)-Italy	a	a	a	na	a	a	7	Medium

CIC cochlear implanted children, *THC* typically hearing children

^aEach asterisk represents whether individual criteria within the subsection was fulfilled

Table 3 Outcomes from the included studies for attention skills

Authors (year)-Country	Type of EF studied	Test	Measures	Scores	Statistics measures and <i>p</i> values	Study conclusions
Huber et al. (2012)-Austria	Visual selective attention	Coding subtest from Hamburger-Wechsler Intelligenz Test III	Correct answers	CIC: 11.1 (2.6) THC: 9.8 (2.5)	$t=0.96, p>0.05$	No differences
Sanei et al. (2018)-Iran	Sustained Auditory Attention	Sustained auditory attention capacity test (SAACT)	Inattention error Impulsive error Total score Reduction span index	Inattention error CIC: 2 (range 0–3) THC: 1 (range 0–3) Impulsive error: CIC: 2 (range 0–5) THC: 1 (range 0–3) Total score: CIC: 5 (range 1–7) THC: 2 (range 1–5) Reduction span index: CIC: 1 (range 0–2) THC: (range 0–1)	$p=0.002$ $p=0.002$ $p<0.001$ $p=0.001$	CIC performed lower than THC in all sustained auditory attention measures
Chen et al. (2019)-China	Auditory Selective Attention with or without visual distraction	Experimental Laboratory Auditory Selective Attention test without visual distraction	Reaction times (RT) Discriminability (d')	RT=CIC: 938.14 (112.81) THC: 790.30 (101.00) d' without visual distractor=CIC: 3.205 (0.954) THC: 3.759 (0.972) d' with visual distractor=CIC: 3.07 (1.03) THC: 3.99 (1.09)	RT= $F(1,36)=21.870, p<0.001$ d' without visual distractor= $F(1,36)=3.85, p=0.057$ $p=0.012$	RT=THC faster than CIC d' without visual distractor=trend for THC having higher discriminability than CIC d' with visual distractor=THC having higher discriminability than CIC

CIC cochlear implanted children, THC typically hearing children

Although CIC performed slightly worse than their TH peers in both modalities (verbal WM: SMD = -0.37 ; 95% CFI from -0.76 to 0.032 ; visuospatial WM: SMD = -0.37 ; 95% CFI from -1.18 to 0.44), the differences did not reach statistical significance (verbal WM: $Z=1.45, p=0.15$; visuospatial WM: $Z=0.89, p=0.37$). Heterogeneity was moderate (verbal WM: $I^2=46\%$; visuospatial WM: $I^2=47\%$).

Fluency skills

Four studies were included: 2 directly [12, 14] and other 2 with databases shared by the Authors [22, 24]. Details are reported in Table 5. Authors used different tasks to measure fluency: the rapid naming processing speed [12, 14] and the semantic fluency task [22, 24], so they were considered separately.

For the rapid naming processing speed (Fig. 4A), both studies recorded the time in seconds. A meta-analysis

including an overall population of 40 CIC and 40 THC did not show any statistically significant group differences (SMD = 0.25 ; 95% CFI from -0.30 to 0.79), with an overall effect Z value of 0.87 ($p=0.22$).

For the semantic categorical fluency, statistically significant differences were found by both De Giacomo et al. [24] and Marshall et al. [22]. The meta-analysis (Fig. 4B) based on an overall sample of 37 CIC and 139 THC showed significantly better performance in THC, with an SMD = -1.05 and a CFI ranging from -1.88 to -0.22 . The overall effect Z value was 2.48 ($p=0.01$). The effect size was large [26]. Heterogeneity between the studies was moderate ($I^2=68\%$).

Inhibitory control skills

For inhibitory control, 5 studies were included (details shown in Table 6), as the Authors sent us their database with participants fulfilling present inclusion criteria [20,

Table 4 Outcomes from the included studies for memory skills

Authors (year)-Country	Type of EF studied	Test	Measures	Scores	Statistics measures and <i>p</i> values	Study conclusions
Huber et al. (2012)-Austria	Verbal STM and working memory	Subtests from German version of WISC III	Digit span forward (DSF) Backward digit span (DSB) Corsi Block Tapping Test	DSF = 6.5 (1.6); THC: 7.2 BDS = 1.7 CIC: 5 (1.5); THC: 4.8 Block span 1 CIC: 6 (0.7); THC: 5.7 (0.9) Block span 2 CIC: 12.6 (2.1); THC: 11.9 (2.2)	DSF: $t = 1.43, p > 0.05$ DSB: $t = 0.4, p > 0.05$ Block span 1: $t = 1.38, p > 0.05$ Block span 2: $t = 1.12, p > 0.05$	DSF: No differences between groups DSB: No differences between groups Block span 1 and 2: No differences between groups
Lee et al. (2012)-Republic of Korea	Phonological memory	Non word repetition task	Correct answer percentage	CIC: 60 (20.87) THC: 78.6 (11.14)	$F(1,47) = 18.957$ $\eta^2_{\text{partial}} = 0.287, p < 0.001$	THC group performed significantly better than CIC group
López-Higes et al. (2015)-Spain	Verbal working memory	Subtest from WISC IV	Backward digit span	Early CIC: 3.74 (0.93) Late CIC: 3.26 (0.93) THC: 3.84 (0.69)	$U = 104.00; p < .03$ for Late CIC vs THC U not reported and p given as > 0.05 for Early CIC vs THC	Statistically significant differences for verbal working memory in Late CIC vs THC, but not for Early CIC vs THC
Talli et al. (2017)-Greece	Phonological and verbal STM	Athina test Subtest from battery EVALEC	Non-word (NW) repetition accuracy % span and processing speed Digit span forward and percentage score	NW accuracy CIC: 39.26 (16.06) THC: 80.37 (10.66) NW Processing speed (sec.) CIC: 236.80 (57.95) THC: 128.53 (31.26) NW span length CIC: 2.27 (0.80) THC: 4.47 (0.99) Digit percentage score CIC: 21.25 (14.67) THC: 48.96 (26.98) Digit span length CIC: 3.93 (0.80) THC: 4.87 (0.99)	NW accuracy $BF_{10} = 613.958, 783, \eta^2_p = 0.573, d = -3.02$ NW processing speed $BF_{10} = 17,290,000,000, \eta^2_p = 0.640, d = 2.33$ NW span length $BF_{10} = 6311.442, \eta^2_p = 0.458, d = -2.44$ Digit percentage score $BF_{10} = 56.476, \eta^2_p = 0.300, d = 1.28$ Digit span length $BF_{10} = 6.362, \eta^2_p = 0.206, d = -1.04$	Digit span: THC group more accurate than CIC and with a wider memory span length Non-word repetition: THC group faster, more accurate than CIC group and with a wider memory span length
Akçakaya et al. (2019)-Turkey	Phonological STM and verbal working memory	Subtest from WISC-R	Non-word repetition accuracy Backward digit span and correct score	Non-Word Repetition CIC: 6.7 (2.6) BDS/THC: 16.8 (1.8) BDS CIC: 5.6 (1.8) THC: 6.4 (1.5)	Non-word repetition $F(1, 57) = 1493.6, p < 0.001$ BDS $F(1, 57) = 9.8, p = 0.07$	Non-word repetition: significant difference between groups BDS: no significant difference between groups

Table 4 (continued)

Authors (year)-Country	Type of EF studied	Test	Measures	Scores	Statistics measures and <i>p</i> values	Study conclusions
Jing et al. (2019)-United States	Visuospatial STM	Working Memory Test Battery for Children (WMTB-C)	Block recall span forward	CIC: 4 (0.75) THC 4 (1)	z value=0.24, $p=0.59$	No significant difference between CIC and THC groups
^a Figuerola et al. (2020)-Spain	Verbal and visuospatial working memory	Psychology Experiment Building language battery	Backward digit span (BDS)BDS Corsi block span backward	CIC: 4.3 (0.58) THC 4.28 (0.82) Corsi block span CIC: 5 (0.87) THC 4.68 (1.07)	–	–
Volpato (2020)-Italy	Verbal STM and working memory	Subtests from TEMA, test of memory and learning	Forward digit span (FDS) Backward digit span (BDS)	FDS CIC: 8.23 (2.05) THC: 8.92 (2.50) BDS CIC: 9.69 (1.38) THC: 10.50 (1.78)	FDS $p > 0.05$ BDS $p > 0.05$	No significant difference between CIC and THC groups either for FDS and BDS
^a De Giacomo et al. (2021)-Italy	Verbal STM and working memory	Subtests from Batteria di Valutazione Neuropsicologica	Forward digit span (FDS) Backward digit span (BDS)	FDS CIC: 3.43 (1.18) THC: 5.29 (0.82) BDS CIC: 2.12 (0.91) THC: 3.64 (1.33)	FDS $Z = -3.876$, $p < 0.001$ BDS $Z = -2.513$, $p = 0.022$	Significant differences for both FDS and BDS in favour of THC group
^a Grep et al. (2011)-Kenya	Visuospatial sequential memory	Subtest from Wide Range assessment of memory and learning 2 (WRAML2)	Sequential memory	–	Memory difficult-to-name: $U = 35.0$, $p = 0.963$, $r = -0.024$ Memory easily nameable: $U = 31.5$, $p = 0.673$, $r = -0.109$	–
^a Botting et al. (2017)-England	Executive-loaded visuospatial working memory	Odd one out task Subtest from Wechsler non-verbal Scale of Ability	Odd one out span Backward spatial span	Odd one out span CIC: 7.64 (3.46) THC: 10.22 (4.39) Backward spatial span CIC: 4.73 (2.28) THC: 6 (1.94)	Odd one out span $t = 3.12$, $p = 0.002$ Backward spatial span $t = 3.21$, $p = 0.0016$	THC group performed significantly better than CIC group in both tasks of executive-loaded visuospatial working memory

CIC cochlear implanted children, THC typically hearing children

^aReported data are not those published in the original study, but those of the CIC who respected the inclusion criteria for the present SR (database shared by the Authors)

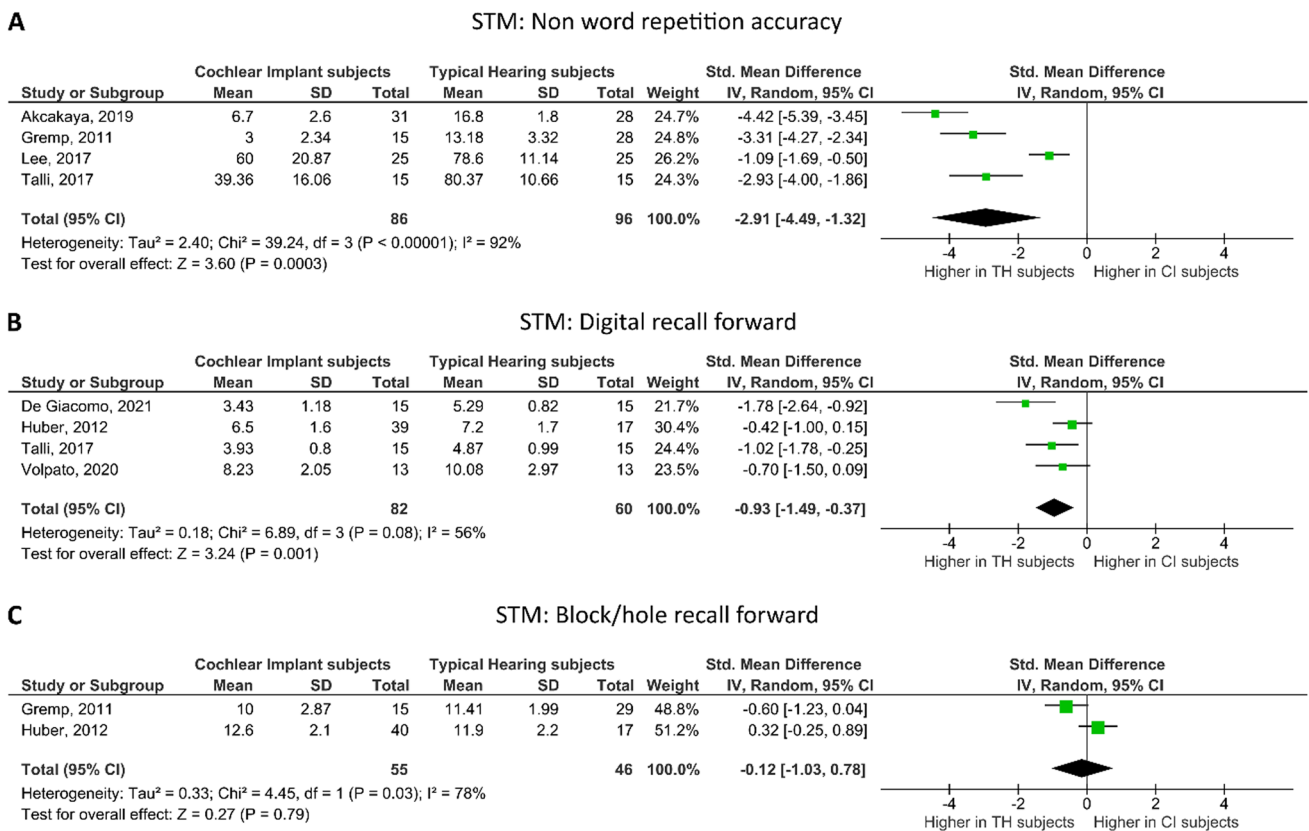


Fig. 2 Forest plot and meta-analysis for verbal and visuospatial short-term memory (STM)

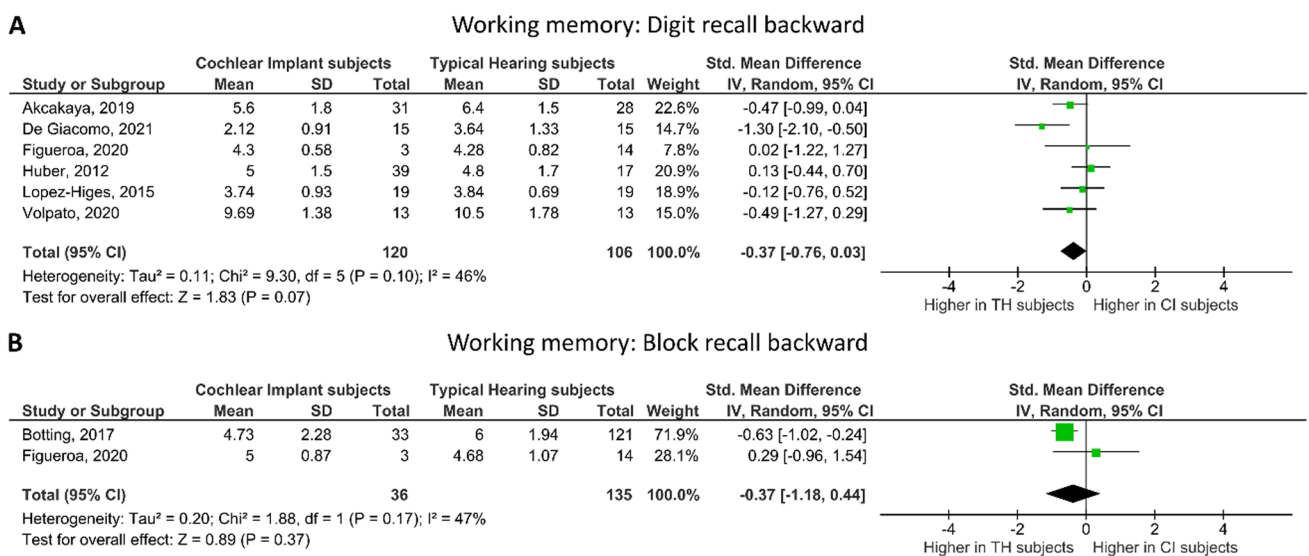


Fig. 3 Forest plot and meta-analysis for verbal and visuospatial working memory

22–25]. The studies used different tasks to measure inhibitory control: response to the task was verbal in three studies [20, 23, 24] vs non-verbal in the other two [22, 25]. Tasks were different in the process of inhibitory control involved:

for example, the conflict resolution in the Simon task used by Marshall et al. [22] requires excitatory biasing of task-relevant stimulus processing, while the Flanker or the Stroop tasks, used by Jamsek et al. [25] and Figuerola et al. [23],

Table 5 Outcomes from the included studies for fluency skills

Authors (year)-Country	Type of EF studied	Test	Measures	Scores	Statistics measures and <i>p</i> values	Study conclusions
Lee et al. (2012)-Republic of Korea	Rapid naming processing speed	RAN task-battery not specified	Processing speed (sec)	CIC: 213.76 (64.57) THC: 212.96 (63.24)	$F(1,47)=0.059$, $\eta_{\text{partial}}^2=0.001$, $p>0.05$	No differences in speed in rapid naming between groups
Talli et al. (2017)-Greece	Rapid naming processing speed	test battery EVA-LEC	Processing speed (sec)	CIC: 87.27 (26.89) THC: 75.07 (10.33)	$BF_{10}=16,275.555$, $\eta_p^2=0.485$, $p>0.05$	No differences in speed in rapid naming between groups
^a Marshall et al. (2017)-England	Categorical fluency	Semantic fluency task: animal category	Number of words in 60 s	CIC: 13.45 (5.9) THC: 17.55 (5.9)	$T=3.004$ $p=0.003$	THC group performed significantly better than CIC group
^a De Giacomo et al. (2021)-Italy	Categorical fluency	Semantic fluency task: home objects category	Number of words in 60 s	CIC: 32.8 (11.2) THC: 57.07 (18.42)	$Z=-3.351$ $p=0.001$	THC group performed significantly better than CIC group

CIC cochlear implanted children, THC typically hearing children

^aReported data are not those published in the original study, but those of the CIC who respected the inclusion criteria for the present SR (database shared by the Authors)

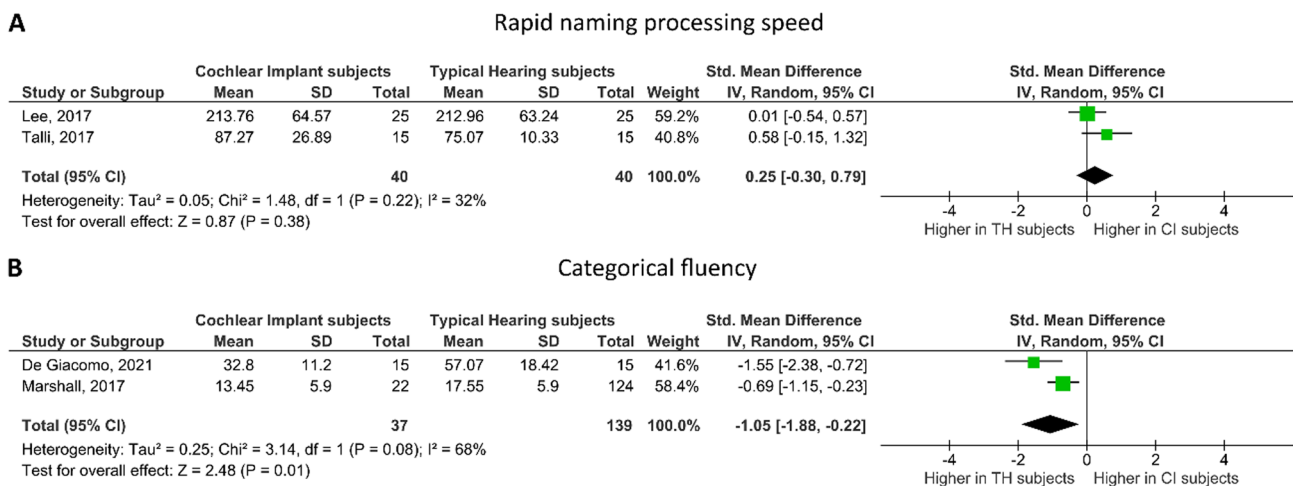


Fig. 4 Forest plot and meta-analysis for fluency

respectively, involve the inhibition of direct route response-priming processes [27]. Finally, score calculation differed significantly between the studies, passing from error score of De Giacomo et al. [24] to scaled score, deriving from a combination of number of errors and completion time in Grep et al. [20] to standard score derived by the combination of accuracy and reaction time in Jamsek et al. [25]. Due to the extreme heterogeneity of studies, a meta-analysis was not performed.

The qualitative comparison between the studies showed contrasting findings: similar performances between CI and TH groups emerged from Grep [20] and Marshal et al. [22] measures, whereas significant differences were reported by

Jamsek et al. [25] and De Giacomo et al. [24] (Table 6). In Figueroa et al. study [23], CI subjects had more accuracy with longer reaction times than TH, but no statistical analysis was performable as there were only 3 subjects respecting our inclusion criteria.

Flexibility skills

None of the studies met the inclusion criteria. Two authors [21, 23] sent us their database with the target participants only (Table 7).

Botting et al. [21] investigated the mediation role of EF in language skills for a group of 101 DHHC with

Table 6 Outcomes from the included studies for inhibition control skills

Authors (year)-Country	Type of EF studied	Test	Measures	Scores	Statistics measures and <i>p</i> values	Study conclusions
^a Gremp et al. (2011)-Kentucky	Verbal inhibition control	Inhibition task from Nepsy	Scaled score given by a combination of number of errors and completion time	CIC: 8 (3.24) THC: 9.76 (2.97)	$t = 1.8069$ $p = 0.08$	No significant differences between groups
^a Marshall et al. (2017)-England	Non-verbal inhibition control	Simon task	Interference score calculated by subtracting congruent from incongruent scores	CIC: -15.32 (17.02) THC: -11.58 (15.47)	$t = 0.97$ $p = 0.33$	No significant differences between groups
^a Figuerola et al. (2020)-Spain	Verbal inhibition control	Stroop task	Differences between congruent-incongruent accuracy, neutral-incongruent accuracy, congruent-incongruent reaction time, neutral-incongruent reaction time	Congruent-incongruent accuracy CIC: 0.006 (0.02) THC: 0.04 (0.05) Neutral-incongruent accuracy CIC: 0.0007 (0.04) THC: 0.04 (0.05) Congruent-incongruent reaction time CIC: 65.9 (14.15) THC: 29.2 (74.04) Neutral-incongruent reaction time CIC: 61.71 (11.44) THC: 56.36 (55.68)	–	–
^a Jamsek et al. (2021)-Ohio	Non-verbal inhibition control	Flanker Inhibitory Control and Attention Test	Standard score derived by the combination of accuracy and reaction time	CIC: 89.86 (15.39) THC: 100.77 (12.7)	$t = 3.760$ $p = 0.0003$	THC group performed significantly better than CIC group
^a De Giacomo et al. (2021)-Italy	Verbal inhibition control	Inhibition and control of Impulsive response from BVN battery	Error score	CIC: 15.69 (1.82) THC: 8.57 (+8.89)	$Z = -2.562$ $p = 0.006$	THC group made significantly less errors than CIC group

CIC cochlear implanted children, THC typically hearing children

^aReported data are not those published in the original study, but those of the CIC who respected the inclusion criteria for the present SR (database shared by the Authors)

CICs or hearing aids. The Authors sent us their database, so it was possible to select data from CIC who met our inclusion criteria. A total of 33 CIC (20 F, 13 M) with a mean age of 105.03 months ($SD = 21.57$) were compared to 121 THC (chronological age = 106.26 ($SD = 16.52$) months; 54 F, 67 M). Mean age at CI was 2.75 ($SD = 1.15$) years. Flexibility was tested with the Children's Colour Trails Test 1 and 2, computing the interference time between the two tests. In Test 1, children were only requested to draw a line as quickly as possible by connecting numbered circles from 1 to 15. In Test 2, they were asked to connect numbers always in ascending order but alternating pink and yellow circles that contain the number. THC had lower interference time than CIC: 29.94 ($SD = 17.05$) vs 34.63 ($SD = 19.9$), but the differences were not statistically significant ($t = 1.32, p = 0.190$).

The study from Figueroa et al. [23] reported results from the Plus-minus task, where participants had to perform a series of 90 mathematical operations divided into three blocks: in the first block, participants added 3 to each two-digit number; in the second one, they subtracted 3 from two-digit numbers; in the last block they had to add and subtract it alternately. Response accuracy and reaction time were recorded. The Authors sent us their complete database of 36 CIC and 54 THC, but only 3 CIC (2 F, 1 M) and 14 THC (12 F, 2 M) met present inclusion criteria, since all the other participants had a chronological age greater than 12.9 years. CIC, all implanted between 1.5 and 2 years of age, were more accurate but showed longer reaction time than TH peers (accuracy: 0.92 CI vs 0.88 TH; reaction time: 5519.51 ms CI vs 5285.82 ms TH). A statistical analysis was not possible.

Table 7 Outcomes from the included studies for flexibility skills

Authors (year)-Country	Type of EF studied	Test	Measures	Scores	Statistics measures and p values	Study conclusions
^a Botting et al. (2017)-England	Cognitive flexibility	Children's Color Trails Test 1 and 2	Interference score Additional time	CIC: 34.63 (19.9) THC: 29.94 (17.05)	$t = 1.318$ $p = 0.190$	No significant differences between the two groups
^a Figueroa et al. (2020)-Spain	Cognitive flexibility	Plus-minus task	Accuracy Reaction time	Accuracy CIC: 0.92 (0.05) THC: 0.88 (0.07) Reaction time CIC: 5519.51 (681.15) ms THC: 5285.82 (1011.5) ms	–	–

CIC cochlear implanted children, THC typically hearing children

^aReported data are not those published in the original study, but those of the CIC who respected the inclusion criteria for the present SR (database shared by the Authors)

Table 8 Outcomes from the included studies for planning skills

Authors (year)-Country	Type of EF studied	Test	Measures	Scores	Statistics measures and p values	Study conclusions
^a Botting et al. (2017)-England	Planning	Tower of London task	Additional moves score	CIC: 37.28 (23.65) THC: 30.32 (15.71)	$t = 2.0045$ $p = 0.047$	CIC group made more additional moves than THC group
^a De Giacomo et al. (2021)-Italy	Planning	Tower of London task from BVN	Correct score	CIC: 21.25 (4.45) THC: 30.43 (2.53)	$Z = -2.562$ $p < 0.001$	THC group performed significantly better than CIC group

CIC cochlear implanted children, THC typically hearing children

^aReported data are not those published in the original study, but those of the CIC who respected the inclusion criteria for the present SR (database shared by the Authors)

Planning/problem solving skills

Two studies were included [21, 24] as the authors of both studies sent us their database for children fulfilling our inclusion criteria (Table 8). Both the studies used the Tower of London test but used a different performance score: Botting et al. [21] measured the number of additional moves in comparison with the minimum number of possible moves required to complete the task, while De Giacomo et al. [24] evaluated the total number of correct answers. Nevertheless, both studies found statistically significant differences.

Botting et al. [21] reported a higher number of additional moves in the CI group: a mean number of 37.28 (SD=23.65) vs 30.32 (SD=15.71) in CIC and THC, respectively. The differences were statistically significant ($t=0.127, p=0.049$) but showed a small effect size ($d=0.32$).

In the study of De Giacomo et al. [24], CIC obtained a mean correct score of 21.25 (SD=4.45), while THC achieved a mean score of 30.43 (SD=2.53). The number of violation rule was 0 for THC and 3.22 (SD=2,76) for CIC. Both the measures showed significant differences at $p < 0.001$ and the effect size was high ($d=2.54$).

Discussion

In the last 20 years, there has been an increasing interest in cognitive processes such as EF that may play a critical role in individuals' social, emotional, and physical health. The aim of the present SR and meta-analysis was to compare preschool and school-age CIC's skills of attention, memory, flexibility, inhibition, fluency, and planning with those of TH peers. Present findings for group comparisons reflected inter-skill and inter-test performance variances.

Attention skills

Only 3 studies had characteristics to be included in the present SR: two focusing on auditory attention [15, 17] and one on visual attention [11].

Chen et al. [17] reported an impaired auditory selective attention and a capacity-limited attentional mechanism across modalities in CIC in comparison with TH peers: visual stimuli interfered with auditory perception when visual and auditory stimuli were incongruent. As identified also by Misurelli et al. [28] in adolescent CI users, auditory selective attention limitations seem to be present already in the first years of school-age (from 5 to 8 years). Such difficulties were reported by Sanei et al. [15] for sustained auditory attention as well.

Differently from the auditory modality, similar performances between CIC and THC were found by Huber et al. [11] for visual selective attention, probably due to

task characteristics, which requires space allocation of figures, an ability typically well-represented in DHHC [29]. However, findings from these studies are not generalizable due to their limited samples sizes. Furthermore, at present there are no other studies that investigated other types of attention, such as shifted attention.

Future research is needed to investigate the complexity of attentional processes because of their important role in cognitive functioning during typical development and aging [30]. Recent studies by Giallini et al. [31] and Nicastrri et al. [32] reflected the significant role of attention in elderly CI users' postoperative outcomes and in CIC's linguistic skills, respectively. Indeed, Nicastrri et al. study [32] indicated auditory selective attention as an independent predictor of lexical and morphosyntactic skills, accounting alone for 25% of observed variance.

Memory skills

Similar to the findings of Akçakaya et al. [6] in long term CI users, the present SR and meta-analysis in preschool/school-age CIC confirmed the presence of STM verbal memory limitations, for both non-word repetition and DSF tasks. Hence, such limitations seem to be present at early ages even in a sample without any morpho-functional abnormalities that may negatively influence postoperative outcomes. This seems an important finding that allows us to link more directly the observed performances to the negative effects of hearing loss and CI technical constraints [2].

Children of the included samples had severe/profound hearing loss and their age at implantation ranged from 1 to 4 years, reflecting duration variances of auditory deprivation. It is well-known that auditory experience is fundamental to provide temporal patterns that are at the bases of neurocognitive functioning. Early auditory deprivation prevents functional maturation, altering the normal processes of synaptogenesis and pruning. Indeed, only early intervention within the sensitive periods may help the central system to recover functional maturation, preventing further degenerative changes [2]. Best outcomes are achievable by strongly limiting auditory deprivation early in life, which for listening and language skills means to accomplish CI around the first year of age or even before [33]. However, no EF studies at present focused their attention on samples with so early age at implantation.

Observed limitations in verbal STM might also be linked to the continuative impoverished representation of auditory input delivered by the CI. As already discussed by Akçakaya et al. [6], perceptual limitations in CIC may alter automatic access of auditory information to the phonological storage area. Hence, the storage, recall, and processing of partially coded information may not be performed appropriately. Limited speech perception may also make CIC more sensitive to

be disturbed or distracted by irrelevant sound [34] as well as less able to use indexical/prosodic cue to help coding and storage of verbal material [35].

Finally, some authors such as Aubuchon et al. [36] considered the effects of other two important factors on verbal STM: the slower sub-vocal rehearsal and the longer memory scanning that characterize CIC. Some CIC could speak at a rate slower than TH peers, reducing the number of items that they can repeat, despite being stored within the time requested by the STM processing. Furthermore, longer memory scanner may impact auditory items' retrieval process from the long-term memory, limiting the quantity of items available for the STM.

For the visuospatial component of STM, Akçakaya et al. [6] could not perform a meta-analysis as they identified only one eligible study due to overlapping data. Conversely, present findings of meta-analysis showed that there were no statistically significant differences between CIC and THC, representing a novel finding and a starting point for future research in this cognitive domain. Indeed, such findings might be partially resulted from the block-recall task used by the authors. This is a spatial test, where there is no possibility to use language strategies such as verbal rehearsal to help memory storage and recall. This strategy is known to be benefited by TH subjects [37] and the impossibility to use it may have resulted in similar performances by the two groups. Future studies could investigate more complex visuospatial STM skills.

Finally, differently from Akçakaya et al. [6], the present meta-analysis on WM pointed out similar performances between CIC and THC for both verbal and visuospatial DSB. A possible explanation might be linked to the diverse nature of the STM vs WM tasks. The first task is more linked to the functioning of the phonological loop, that is at risk to be compromised in DHHC [38] and is strongly associated with language development and facility [39]. The second one, instead, seems to be more related to visual processing in both verbal and visuospatial form, since individuals tend to use a visual representation of numbers within a familiar visuospatial configuration, in preparation for providing the reversed sequence [40]. Moreover, as shown by the present meta-analysis, the visuospatial domain seems to result less compromised in CIC.

Fluency skills

Four studies were included for fluency skills analysis, but two independent meta-analyses were accomplished due to the use of two different study measures: rapid automatized naming (RAN) and verbal fluency. While RAN outcomes showed similar results of speed between CIC and THC, categorical fluency showed significantly better scores with a

large effect size in the TH group. These discrepancies might be linked to three different factors.

The first factor is the diverse grade of difficulties of the two tasks. In the first task, children have to name as quickly as possible a series of visually presented familiar symbols, such as digits, letters, colours or objects. In the second one, children are requested to recall within 1 min the highest number of items belonging to a given semantic category (i.e., colours, animals). Although both tasks rely on other more basic EF, such as verbal WM and inhibitory control, their weight is more pronounced in the categorical fluency, where no visual reference is present. For such a task, the phonological loop supports verbal information processing in real time and allows the activation of relevant content from long-term memory whenever necessary for ongoing actions [41]. Despite CI use, phonological loop is often compromised in DHHC [38]. In the RAN task, less efficient functioning of phonological loop might have been compensated by the high familiarity of the symbols in use. Furthermore, the semantic network in CIC is more condensed and less spread out than the THC's semantic network. This fact may significantly influence the rapidity and the readiness with which they could access and retrieve words from a specific semantic category, limiting the number of words that they are able to retrieve in 60s [42].

A second factor is that the speed of RAN was the only assessment, while no accuracy measure was reported. Despite similar results in speed between the two groups, children might have differed in the number of items correctly named.

Finally, due to limited number of studies included in this meta-analysis and small sample sizes, discrepancy might be merely due to the contextual characteristics of the samples themselves.

Inhibitory control skills

Five studies on inhibitory control were included in the present SR but their findings were contrasting. Two studies reported significant differences between CIC and THC, whereas three others showed similar performances. Task differences in inhibitory control measures might be one of the key reasons for the inconsistency between these studies. Furthermore, some subjective characteristics might have significantly influenced the findings.

First of all, inhibitory control skills are strongly linked to attentional processes. Sustained and focused selective attention are requested to suppress all non-relevant representation, to narrowly tied the contents of consciousness to the goals, and to remove once-relevant information that has become irrelevant due to a change in goals, context, task or situational demands [43]. It is a matter of fact that the

present SR highlights sustained and selective attention difficulties in CIC.

Second, inhibitory control is strictly linked to the Theory of Mind defined as the following abilities: to understand that others' desires and thoughts may differ from one's own, to attribute mental states (beliefs, intents, desires, emotions, and knowledge) to ourselves/others, and to consider another individual's mental state [44]. Theory of Mind might be compromised in DHHC in general and in CIC in particular [45].

Third, parents' psychological well-being and sensitivity are the other two crucial factors that may significantly influence inhibitory control development in CIC. Appropriate, cognitively stimulating and affectively engaging parental behaviors provide positive scaffolding for internalization of self-regulatory behaviors and inhibitory control skills in both THC and DHHC. Conversely, maternal depression or stress as well as familial excessive control and conflicts are significantly associated with poorer inhibitory control in DHHC [25]. Nevertheless, all these aspects may variably interact with each other, and such interactions may contribute to performance differences.

Flexibility skills

Very few research articles on flexibility skills in CIC were published so far. Two studies could be included in the present SR, again thanks to the Authors sharing their database specifically for CIC fulfilling present inclusion criteria. Similar to the original Figueroa et al. study [23] that did not find any significant differences between THC and CIC performance in flexibility skills, the three subjects included in the present SR showed the same tendency.

On the other hand, Botting et al. study [21] including 108 DHHC with different degrees of hearing loss, hearing technologies and aetiologies found a greater cognitive load for DHHC in completing the task, where the ability to switch between different sets of rules is needed. Conversely from Figueroa et al. study [23], once 33 CIC were sorted out of 108 DHHC, significant performance differences between CIC and THC disappeared. Namely, CIC showed readiness as good as their TH peers to selectively switch between mental processes and to generate appropriate behavioral responses. Indeed, by reducing selection bias, a positive effect of CI could be detected in flexibility skills development.

Therewithal, two considerations could be given here. First, the two included studies had small samples (a total of 36 subjects), limiting the possibility of generalizing present findings. Second, the two tests that they used, the Children's Colour Trials Test and the Plus-minus task, are both based on set shifting tasks. According to the hierarchy of cognitive flexibility outlined by Bunge and Zelazo [46], set shifting is

a lower-level form of cognitive flexibility. It is possible that CI allows DHHC to master the basic form of cognitive flexibility, while limitations may arise from more complex form of cognitive flexibility, as measured by task switching tests. There is the need to increment the research in this domain, varying the type and complexity of tasks, also considering the role that oral language skills, at risk to be compromised in CI users, have in holding up flexibility skills development, being the flexible rule-use driven by the linguistically mediated representation of the rules involved in the task, even when the tasks have apparently low verbal demands [47].

Planning/problem solving skills

Two studies with a total of 48 participants, all satisfying present inclusion criteria, were included in this SR [21, 24]. The analysis of outcomes concordantly confirmed the presence of planning difficulties in CIC, with an effect size from small in Botting et al. [21] to high in De Giacomo et al. [24].

Planning is a complex form of action that involves mental representation and/or behavioural execution of a consciously predetermined sequence of actions that could be adequate for achieving a task. It requires to analyse all the essential information to achieve an aim, to think of alternatives, to weigh and make choices, and to evolve a conceptual framework or structure, serving to direct activity [48]. For its structure, it can be considered as a complex EF skill, which from a point of view of an integrative framework could be considered as a set of lower-level skills, such as attention, WM and inhibition [49]. Limitations in these more basic EF may influence the readiness with which CIC achieve planning, where various rules need to be remembered and retrieved (memory), and automatic and more immediate actions need to be inhibited due to not being allowed or not being useful to the final aim (inhibition control).

Furthermore, planning seems to be mediated by verbal skills, in terms of inner speech or language-based reflection, that is the use of language to reflect and guide behaviour. This association was already demonstrated in THC studies comparing their peers with specific language impairments: in experimental conditions, where verbal mediation was disrupted through articulatory suppression, THC's planning performances get worse than a baseline situation, where language mediation is allowed. The dimension of this effect is greater for THC with relatively better language ability. The worsening phenomenon does not appear in children with specific language impairments that relies more on non-linguistic perceptual strategies for planning tasks [50].

Future research, specifically planned to experimentally control for all these aspects (low level EF and language tasks) may help to gain new insight in the CIC's planning skills.

Study limitations

Present findings should be interpreted with caution due to some considerations. First of all, the small number of studies in line with the present inclusion criteria limited the generalizability of our findings. Moreover, overlapping data by the majority of existing studies limited the overall sample size or did not allow us to perform a meta-analysis for some EF skills. Likewise, the effects of some demographic and audiological factors, such as age at test/implantation, preoperative auditory profile, listening mode (unilateral/bilateral/bimodal), and CI model on EF skills could not be addressed due to small size and heterogeneity of the study samples. Although we limited the present study population to CIC without any additional disabilities/morpho-functional abnormalities, it was not possible to control for other significant predictors, such as communication modality (i.e., exclusively oral language vs mixed mode), school setting (mainstream or special schools) or socioeconomic status. Finally, some Authors rejected our data request or did not respond to the emails, hence, their studies were not included. We do not know the effects that their findings could have had on the present SR and meta-analysis.

Conclusions and implications

The present SR and meta-analysis on preschool and school-age CIC confirmed the presence of significant limitations in their verbal STM and reflected for the first time their difficulties in auditory attention and planning skills. No conclusions could be driven for inhibitory control and fluency skills, while similar CIC and THC performances seem to be developed in the flexibility domain. It seems that the amount of clinical evidence is EF skill specific: memory received more attention than other skills (e.g., planning).

Future research is needed to investigate EF domains, where doubts are still present. Knowing the significant effects of early implantation and habilitation on postoperative performance, the knowledge we already have on deficits and possible mechanisms that underlie the most studied EF could guide the implementation of specific training programs, already in the first phases of postoperative (re)habilitative process, with particular attention to preschool age when delays are still not too big to be compensated.

Supplementary Information The online version contains supplementary material available at <https://doi.org/10.1007/s00405-023-08260-x>.

Data availability statement The data sets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Conflict of interest The authors declare that they have no conflicts of interest. This research was not financially supported.

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