Psicothema

Psicothema 2019, Vol. 31, No. 4, 384-392
doi: 10.7334/psicothema2019.107

ISSN 0214 - 9915 CODEN PSOTEG

www.psicothema.com

Impairment of cognitive memory inhibition in individuals with intellectual disability: A meta-analysis

Elena Palomino, José María López-Frutos, Juan Botella, and María Sotillo
Universidad Autónoma de Madrid

Abstract

Background: Cognitive inhibition impairment is intimately related to the forgetfulness of relevant information. This meta-analysis aims to synthesise the evidence of impaired function of cognitive inhibition processes over memory in individuals with intellectual disability (ID). **Method:** Eleven studies were selected and analysed and included a total of 683 participants. The studies were categorised according to variables such as the task used, the processes involved, the sensory modalities and the method. **Results:** Despite the small sample of studies, the results revealed significant difficulties with cognitive memory inhibition (CMI) tasks in individuals with ID compared with typical development (TD) individuals (d = 0.62). CMI problems were found in all life stages except the 19-45-year-old stage. In this stage, there was a smaller amount of evidence even though it included the 31-40-year-old range, during which premature aging has been observed in ID. **Conclusions:** An impairment of CMI in people with ID was observed. More studies are needed to more reliably assess the potential moderating role of age and other factors.

Keywords: Cognitive inhibition, intellectual disability, executive function, memory.

Resumen

Afectación de la inhibición cognitiva sobre la memoria en personas con discapacidad intelectual: un meta-análisis. **Antecedentes:** la afectación de la inhibición cognitiva se encuentra íntimamente relacionada con el olvido de información relevante. Este meta-análisis tiene como objetivo conocer si los procesos de inhibición cognitiva sobre la memoria están afectados en personas con discapacidad intelectual (DI). **Método:** se seleccionaron y analizaron 11 estudios que incluyeron un total de 683 participantes. Los artículos fueron categorizados en función de la tarea utilizada, los procesos implicados, las modalidades sensoriales y el método. **Resultados:** a pesar del número de estudios, se observaron dificultades significativas en inhibición cognitiva sobre la memoria (ICM) en personas con DI, en comparación con personas con desarrollo típico (d = 0.62). Estas dificultades se observaron en todas las etapas cronológicas, excepto de 19 a 45 años. En esta etapa, la evidencia fue escasa, a pesar de incluir el rango de los 31-40 años, donde se ha observado presencia de envejecimiento prematuro en personas con DI. **Conclusiones:** se observaron dificultades en ICM en personas con DI. Se necesitan más estudios para evaluar de forma más exhaustiva el papel potencialmente moderador de la edad y de otros factores.

Palabras clave: inhibición cognitiva, discapacidad intelectual, función ejecutiva, memoria.

Since the pioneering research by Luria (1966) on executive alterations and by Lezak (1982), who first used the term "executive functions (EFs)", EFs have come to be considered fundamental in biopsychosocial development (Diamond, 2013). They are believed to play a role in processes such as conflict resolution, goal formulation, behaviour planning and information retrieval.

The conceptual definition of EFs is elusive. Most models define them as a set of high-level cognitive processes whose purpose is to regulate cognition and motor action and adjust behaviour (Friedman & Miyake, 2017; Hasher & Zacks, 1988). EF components include cognitive flexibility, working memory and inhibition (Anderson & Bjork, 1994; Miyake et al., 2000).

Received: April 14, 2019 • Accepted: July 30, 2019
Corresponding author: Elena Palomino
Facultad de Psicología
Universidad Autónoma de Madrid
28049 Madrid (Spain)
e-mail: jml.frutos@.uam.es

Inhibition (inhibitory control) is defined as a process that controls other processes—attention, memory, thinking, language, emotions and motor behaviours—(Amieva, Phillips, Della Sala, & Henry, 2004). Inhibition is involved in situations in which preferential internal or external interfering predispositions must be addressed. In memory processes, inhibition intervenes in the active maintenance of information in the short term and in the recovery of memory traces, preventing other information from interfering (Anderson & Weaver, 2009). Hasher and Zacks (1988) distinguish three processes in cognitive memory inhibition (CMI): (a) access processes to control the information that enters the operative memory; (b) deletion processes that monitor the information that is removed from the operative memory; (c) restraint processes to prevent information that is not appropriate for the task in progress from entering the operative memory. Anderson (2007) focused on the role of cognitive inhibition over long-term memory, indicating that it is thought to prevent interference between the distracting information and the information that we wish to recover or maintain active. To prevent the interference, inhibition acts on

the undesirable memory trace to produce a potentially reversible and gradual change that makes it less accessible and thereby difficult to recover (Anderson & Bjork, 1994). Research findings on memory inhibitory control suggest that when inhibition reduces the accessibility of a memory trace, it is more difficult to recover in the future; therefore, this process has been related to forgetfulness. To synthesise, the basic function of the inhibition is to facilitate the recovery and maintenance of the information to be remembered. To achieve this, it controls the competing responses and the non-entry into the awareness of distracting memories (Hasher & Zacks, 1988).

CMI has been studied in individuals of different ages. In young individuals with typical development (TD), an increase in interference is observed when increasing the retention interval, the similarity of the material or the amount of material to remember (Corman & Wickens, 1968). Likewise, in young population other authors have observed a decrease in memory when there is an interference (Clapp, Rubens, & Gazzaley, 2010). In elderly populations with TD, research reveals that as the individual ages, CMI decreases (Gazzaley, Cooney, Rissman, & D´Esposito, 2005).

Given that natural aging influences CMI functioning, we consider the involvement of these processes when there is premature aging, as occurs in certain individuals with intellectual disability (ID) (Devenny et al., 2004; Roth, Sun, Greensite, Lott, & Dietrich, 1996; Zigman, 2013).

Difficulties in executive control in individuals with ID at different developmental stages were found (Greer, Riby, Hamilton, & Riby, 2013) in inhibition, working memory, planning and problem solving (Lanfranchi, Jerman, & Vianello, 2009), parallel to involution processes in adults with ID between 35 and 40 years old (Hawkins, Eklund, James, & Foose, 2003).

The research about CMI processes in people with ID has a fundamental relevance; thus, they are closely linked, on the one hand, to the selection of the correct memory trace for the task and, on the other hand, to the prevention of the presence of distractors. Therefore, these processes have an ecological implication on the effective recovery of information.

The objective of this meta-analysis was to synthesise the empirical evidence available about whether CMI is impaired in people with ID compared to people with typical development. The potential moderating role of several variables were also analysed. This meta-analysis would be the first—to the authors' knowledge—to study the state of cognitive inhibition over memory in a population with ID. This knowledge is especially important because of the close relationship between an impairment of CMI and the presence of forgetfulness.

Method

This meta-analysis was performed following the PRISMA statement. A statement -based on evidence- widely recognized in the scientific community to elaborate meta-analysis with quality standards (Strech & Sofaer, 2016).

Literature Sampling

Information sources and search strategy. We conducted a search of the studies of CMI in individuals with ID in the following databases: PsycINFO, ERIC, PsycARTICLES, ScienceDirect, MEDLINE and PubMed. The following descriptors were used: *developmental disorders*, *developmental disabilities*, *intellectual development disabilities*, *executive function*, *memory*, *aging, inhibition*, *retroactive inhibition*, *proactive inhibition*, *memory control*, *inhibitory control*, *interference control*, *cognitive inhibition*, *cognitive control*, *intellectual disability*, *mental retardation*, *intellectual deficiency*, *cognitive impairment*, *cognitive processes* and *cognitive development*. The publication type was limited to "all journals" published from 1973 to July 2018. To reduce publication bias, formal sources (in article databases) and informal sources (conference proceedings and doctoral theses) were reviewed.

Eligibility criteria. We selected studies that met the following inclusion criteria: (a) studies of individuals with ID, regardless of whether a particular syndrome was specified (e.g., Down syndrome, Williams syndrome), with at least one control group (with TD); (b) studies published in English, French and Spanish; (c) studies that used experimental memory tasks in situations of inhibitory mnesic control. The exclusion criteria consisted of (a) studies in which it was uncertain that the processes assessed involved CMI; (b) narrative reviews and empirical articles that subjectively evaluated CMI; (c) individuals with acquired brain damage, a diagnosis of comorbidity, epilepsy, mixed diagnoses or autism spectrum disorder.

Instruments

The statistical analyses and the publication bias were carried out with the SPSS macros of Lipsey and Wilson (2001) and the Metafor R package (Viechtbauer, 2010), whereas the forest plot was obtained through Review Manager (2008).

Procedure

Result of the study selection process. After the database searches and the inclusion and the exclusion criteria a total of 11 studies remained (Figure 1). The articles included in this meta-analysis are marked with an asterisk (*) in the references section.

Data extraction. To avoid biases, prior to the search and selection of articles, the quantitative and categorical variables potentially moderating the results were analysed. Following Lipsey (2009), these variables were classified as substantive, methodological and extrinsic. The substantive moderating variables were developmental stage, average ages (and their standard deviations), distribution by gender of each sample, institutional origin, social class, diagnosis and level of severity as well as context (i.e., country and context of evaluation). The categorical methodological variables were between-group comparison mode, diagnostic method, dependent variable used, type of task, input and output modality, pre-experimental practice, systems of memory assessed and stimulus type. The extrinsic variables were the year of publication, the source and the professional background of the first author. The quantitative methodological moderating variables were initial sample size, statistical sample size, differences between average ages, differences between the percentages of the distribution by gender, duration of the tasks, quality index (Sanduvete, 2008) and the statistical data. Finally, it should be noticed that age was the only variable that was analysed in two ways, as a categorical variable depending on the stage of development and as a quantitative variable when using the chronological mean age of the groups.

Regarding developmental stage, papers were distributed according to the chronological age of participants: two of childhood (0-11 years), six of adolescence (12-18), one of adult (19-45), one of older adults (46-64) and one of elderly individuals (over 65). Two researchers independently coded all the variables. The kappa coefficients were calculated for the categorical variables and the intraclass correlations for continuous variables. The inter-rater reliability was on: the quality index (average = .99, minimum = .91, maximum = 1), the risk of bias items (average = .99, minimum = .91, maximum = 1), the standardized mean difference (average = 1, minimum = 1, maximum = 1), the categorical moderator variables (average = .99, minimum = .86, maximum = 1) and the continuous moderator variables (average = 1, minimum = 1, maximum = 1). The discrepancies between the two evaluators were solved with a third independent coder.

Analysis of the risk of bias. To assess the risk of bias in each study (i.e., reliability and validity), an analysis of the selection, execution, detection and mortality processes was performed based on Wright, Brand, Dunn and Spindler (2007). For selection bias, the selection process of the participants was analysed. For execution bias, whether all groups received the same instructions and whether all participants followed the same procedure was assessed. For detection bias, the degree of agreement was assessed when there were several evaluators. Sample mortality bias was also considered. Similarly, we analysed the fulfilment of pre-specified results, completeness in the data processing, other sources of bias and author-indicated conflicts of interest. In addition, the risk of bias was analysed with the scale of Sanduvete (2008) to assess the individual quality of each study. The benefits of this scale are: a quality index, a qualitative description of the characteristics of each work, a content validity study carried out by experts and it is applicable to any type of design (Carro, 2016).

Data analysis

The effect size index was the standardised mean difference between the ID and TD groups, $d = [c(m)] \cdot [(\bar{X}_{ID} - \bar{X}_{TD}/\hat{S}]$: $c(m)$ was a correction factor for small sample sizes (Borenstein, Hedges, Higgins, & Rothstein, 2009), and $\hat{S}$ was the squared root of the pooled estimate of the common population variance (Botella & Sánchez-Meca, 2015; Lipsey & Wilson, 2001). For number of words remembered and negative priming tasks, the order of the groups in the formula was changed so that all the positive values for d indicated better performance in the TD group. To interpret

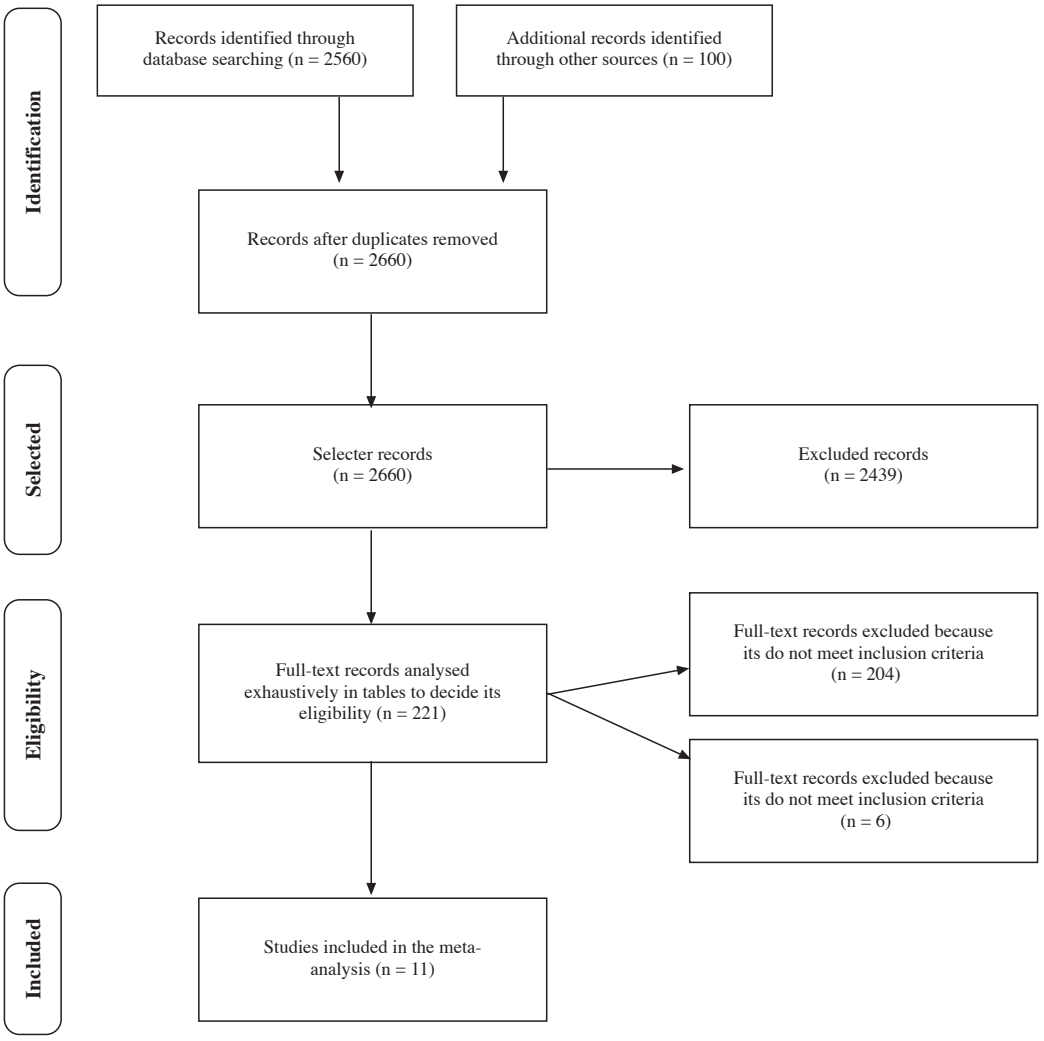

Figure 1. PRISMA flow diagram

the magnitude of the effect size, the conventions of Cohen (1988) were used (d = 0.2 small, d = 0.5 medium, d = 0.8 large).

The d values were analysed assuming a random-effects model for obtaining a pooled effect size estimate and 95% confidence intervals and for analysing any moderating variable (Borenstein et al., 2009). Heterogeneity of the effect size estimates was assessed with the Q statistic and the I^2 index (Huedo-Medina, Sánchez-Meca, Marín-Martínez, & Botella, 2006). Significance of the moderator variables was determined by assessing the model misspecification for the ANOVAs (categorical moderators) and meta-regressions (quantitative moderators).

The first part of the results is based on the combined effect size estimation. In addition, direction was considered, as was the strength of the relationship quantified by the effect sizes. The second part of the results focused on the presence or absence of heterogeneity, the description of the combined estimates of the categories of the categorical variables that did not have at least three studies within the categories, the statistical analysis of the categorical variables that did meet this criteria and the meta-regression analysis for the main quantitative moderators. In addition, we assessed publication bias through the Egger's regression test (Egger, Smith, Schneider, & Minder, 1997), the visual inspection of the funnel plot (Borenstein et al., 2009) and the trim-and-fill method (Duval & Tweedie, 2000). We calculated the fail-safe number (Rosenthal, 1979).

Results

Descriptive characteristics of the studies

Table 1 includes the detailed information for each primary study.

Evaluation of the risk of bias

The risk of selection process bias was lower in studies with larger effect sizes. Among the studies with medium effect sizes, one presented a medium risk by using relatives of individuals with ID for the control group. Regarding execution bias, the group of studies that offered more information on this process were those studies

Table 1
Detailed information for each primary study

Author (year)	Developmental stage	Quality index	Initial/statistical sample size	% of women and difference of %	MA/CA Mean ages (SD)	Mean (SD)	Type of task (stimulus type)	Memory system (Input/Output modality)
Baker (2011)	1	7.5	ID: 40/39 TD: 40/40	–	MA. ID: 5.26 (0.68) TD: 4.8 (0.96)	ID: 23.58 (7.38) TD: 26.4 (7.62)	N-Back (Pictures)	WM (Vi/Ve)
Belacchi (2014)	2	7	ID: 42/42 TD: 42/42	ID: 57.14 TD: 47.62 Difference: 9.52	MA. ID: 5.6 (-) TD: 5.6 (-)	ID: 4.52 (2.04) TD: 5.95 (2.63)	WM verbal 2 (Words)	WM (A-Vi/Ve)
Borella (2013)	2	7.5	ID: 19/19 TD: 19/19	ID: 63.15 TD: 57.89 Difference: 5.26	MA. ID: 5.6 (2) TD: 5.6 (2)	ID: 0.95 (0.91) TD: 0.47 (0.61)	Distracter inhibition (Words)	EM (A/Ve)
Brega (2008)	5	8	ID: 47/47 TD: 41/41	ID: 0 TD: 0 Difference: 0	CA. ID: 68.2 (-) TD: 64.5 (-)	ID: 6.1 (3.3) TD: 8.8 (4)	RAVLT (Words)	WM (A/Ve)
Carretti (2010)	3	8	ID: 28/28 TD: 28/28	ID: 46.43 TD: 46.43 Difference: 0	MA. ID: 6.2 (1.6) TD: 6.6 (1.4)	Cohen's d = 0.42	Selective span (Words)	WM (A/Ve)
Danielsson (2010)	4	10.5	ID: 46/46 TD: 92/92	ID: 45.7 TD: 45.7 Difference: 0	CA. ID: 63.2 (8.1) TD: 63.2 (8)	ID: 2.41 (1) TD: 2.87 (0.85)	Executive load at encoding (Words)	WM (A/Ve)
Lanfranchi (2004)	1	7.5	ID: 18/18 TD: 18/18	–	MA. ID: 5.42 (10) TD: 5.17 (7)	ID: 1.33 (0.48) TD: 3.61 (2.28)	Study 1 task 3 (Words)	WM (A/Ve)
Lanfranchi (2009)	2	8.5	ID: 20/20 TD: 20/20	–	MA. ID: 4.10 (0.9) TD: -	Cohen's d = 0.6	Selective span (Words)	WM (A/Ve)
Merril (1996)	2	7.5	ID: 18/18 TD: 18/18	–	CA. ID: 17.8 (1.2) TD: 19.1 (1.8)	F (1, 34) = 5.15*	1 (Letter)	WM (Vi/M)
Odekirk (2006)	2	8	ID: 30/30 TD: 30/30	–	CA. ID: 16.16 (1.22) TD: 16.36 (0.99)	F (1, 58) = 5.11*	Identity (Letter)	WM (Vi/M)
Sampaio (2008)	2	7.5	ID: 14/14 TD: 14/14	ID: 50 TD: 64.29 Difference: 14.29	CA. ID: 16.79 (5.68) TD: 17.93 (6.1)	ID: 4.79 (1.42) TD: 6.79 (1.63)	CVLT (Words)	EM (A/Ve)

Note: Developmental stage: 1 = children; 2 = adolescents; 3 = adults; 4 = older adults; 5 = elders. ID = intellectual disability; TD = typical development; - = information not provided in the original articles; *SD*= standard deviation; WM= working memory; EM= episodic memory; A= auditory; Vi= visual; Ve= verbal; M= motor; MA = mental age; CVLT = California Verbal Learning Test; RAVLT = Rey Auditory Verbal Learning Test; CA = chronological age. In this figure, MA or CA is presented depending on whether the comparison made by the authors is, respectively, by mental or chronological age. Standard deviations of mental comparisons are in months. Standard deviations of chronological comparisons are in years
* $p < .05$

with larger effect sizes. Regarding detection bias, most of the articles used tests or tasks adequate for evaluating the research objective. However, the studies offered little information on the number of evaluators and statistical coherence among them. Generally, a low risk of mortality was found. The results of the quality scale were slightly higher in the studies with medium effect sizes.

Combined and study-level effect size estimates

The effect size estimate was d = 0.62, 95% CI [0.47, 0.78]. Therefore, the estimation of the effect size showed, as expected, a significant impairment in CMI among individuals with ID compared to TD individuals.

All directions of calculated effect sizes reflected a better execution in the group of people with TD. However, there were some discrepancies regarding the presence or absence of statistically significant differences between groups. The individual effect sizes of the studies are shown in Table 2 and are graphically represented by a forest plot elaborated with Review Manager (Figure 2).

Table 2
State of CMI processes in people with ID

Author	Year	d	95% CI	DV	Population
Baker	2011	0.37	[-0.07, 0.82]	Words	FXS
Belacchi	2014	**0.60**	[0.17, 1.04]	Words	DS
Borella	2013	0.61	[-0.04, 1.26]	Intrusion	DS
Brega	2008	**0.74**	[0.30, 1.17]	Words	FXS-TA
Carretti	2010	0.41	[-0.12, 0.94]	Words	ID-WSA
Danielsson	2010	**0.51**	[0.15, 0.87]	Words	ID-WSA
Lanfranchi	2004	**1.35**	[0.63, 2.07]	Words	DS
Lanfranchi	2009	0.59	[-0.05, 1.22]	Words	DS
Merrill	1996	**0.76**	[0.09, 1.44]	Priming	ID
Odekirk	2006	**0.59**	[0.07, 1.10]	Priming	ID
Sampaio	2008	**1.27**	[0.46, 2.08]	Words	WS

Note: DV = dependent variable; Words= number of words; FXS = Fragile X syndrome; DS = Down syndrome; ID = intellectual disability; ID-WSA= intellectual disability without specific aetiology; WS = Williams syndrome; FXS-TA= Fragile X syndrome with tremor/ataxia. **d** = statistically significant effect; d = statistically non-significant effect

Heterogeneity

The heterogeneity statistic was $Q(10) = 9.02$, $p = .530$, reflecting an absence of statistical heterogeneity beyond what would be expected by mere random sampling of individuals. Consistent with these data, the I^2 = 0% index was interpreted as reflecting statistical homogeneity. Despite the absence of heterogeneity, we assessed categorical and quantitative moderators for theoretical reasons.

Statistical analysis of categorical moderating variables

The most relevant categorical moderating variables at the theoretical level were age, the diagnostic aetiology of the participants, the dependent variable used, the type of task, the memory systems studied, the level of severity, the level of intelligence and the type of comparison (Table 3). The first five did not meet the criterion of having at least three studies within each category, so only the combined estimate of the categories of each of them was calculated. The next two, the severity of the diagnosis and the level of intelligence, could not be analysed because this information did not appear in most of the articles. Finally, the type of comparison variable did meet the criterion of having three studies in each category; therefore, an exhaustive statistical analysis of this moderating variable was carried out.

In developmental stages, the effect was significantly different from zero for all age groups except for the adult age group. In all the stages—except for the adults—the people with ID had more difficulties in CMI than the population without ID.

In terms of the aetiology variable, all effect sizes were statistically significant. In all the aetiologies evaluated, the group with ID had less execution in CMI processes. If the categories with a greater number of studies are compared, we observe a greater magnitude of the effect size in the groups with Down syndrome (DS) than in those with unspecified intellectual disability. The differences between the group with DS and the group without ID were greater than between the group with unspecified ID and the group without ID.

For the dependent variables, the statistically significant effect sizes appeared in the variable number of words remembered and in

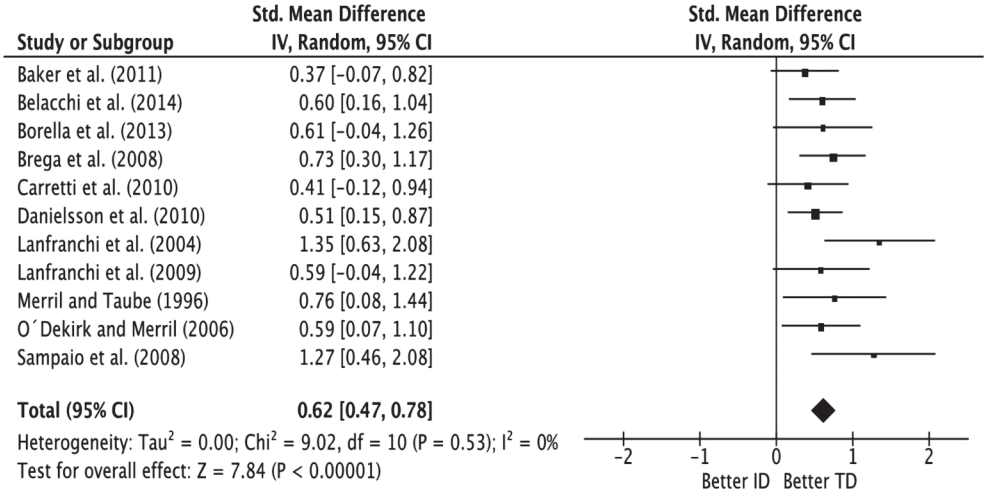

Figure 2. Forest plot

the negative priming tasks. In the intrusions variable, which only one study included, the effect was not statistically different from zero. In both—the number of words remembered and in negative priming—the group without ID shows better performance than the group with ID.

All effect sizes were statistically significant in the moderator type of task. Regardless of the type of task used, the group with ID showed more difficulties. Effect size in experimental tasks was interpreted as a medium effect and standardized test size as a large effect. The differences between the groups were greater in standardized tasks.

Inside the memory system categories, the effect was significantly different from zero. Regardless of the memory system evaluated, the group without ID had better performance. In tasks involving the operative memory system, the magnitude of the effect size was medium, whereas if the tasks involved episodic memory, the magnitude of the effect size was large. The differences between the groups were greater if an episodic memory task was used.

Last but not least, a statistically significant effect different from zero was observed in all categories of type of comparison. In all the types of comparison used, the group with ID had more difficulties than the group without ID. However, no differences were observed among the categories according to the type of comparison. The difficulties were similar in all types of comparison. That is, we observed an effect in all categories, but those effects were not different from each other. In addition, the individual effect sizes within each category were homogeneous with respect to their mean effect. Therefore, we can say that there was no variability to explain between effect sizes.

Finally, all statistical calculations were fit with fixed and random effects models, the same conclusions were reached with both.

Statistical analysis of quantitative moderating variables

The quality index and the mean chronological age were considered for theoretical reasons as the two main quantitative variables. Therefore, both were analysed through meta-regression. The results of the meta-regression did not show a significant association between the effect size on one side and both the quality of the study and the chronological age (Table 4). Therefore, the heterogeneity of *d* estimation is not explained by any of these two variables.

Publication bias

Egger's regression test found cues of asymmetry in the funnel plot (p = .049), as can be observed in Figure 3. However, the trim and fill procedure did not yield any missing study on either side of the figure. In any case, because the asymmetry found in the visual inspection was due to the two studies with larger effect size estimates at the left side of the figure, we performed a sensitivity analysis to assess the consequences of its presence. The combined estimate of the effect size with the other nine studies was still robust and significant (d = 0.56, 95% CI [0.40, 0.72]). Rosenthal's fail-safe number is 145. This number is much larger than the reference for this analysis (k·5 + 10 = 65). Thus, although there might be some publication bias in this set of studies, the effect of such bias could be an overestimate in the combined effect size but is not yielding an artificial non-existent effect.

Table 3
Summary of categorical moderating variables analysis

	k	d	95% CI	
Age				
Childhood	2	0.64	[0.26, 1.02]	
Adolescence	6	0.67	[0.44, 0.91]	
Adult stage	1	0.41	[-0.12, 0.94]	
Older adults	1	0.51	[0.15, 0.87]	
Elderly	1	0.73	[0.30, 1.17]	
Aetiology				
Down syndrome	4	0.72	[0.43, 1.01]	
Intellectual disability	4	0.54	[0.30, 0.78]	
Fragile X syndrome	2	0.56	[0.25, 0.87]	
Williams syndrome	1	1.27	[0.46, 2.08]	
Dependent variable				
Number of words	8	0.62	[0.44, 0.79]	
Negative priming	2	0.65	[0.24, 1.06]	
Intrusions	1	0.61	[-0.04, 1.26]	
Type of task				
Experimental task	9	0.58	[0.41, 0.75]	
Measured tests	2	0.85	[0.47, 1.24]	
Memory systems				
Operative memory	9	0.60	[0.43, 0.76]	
Episodic memory	2	0.87	[0.36, 1.37]	
Type of comparison				$Q_B(1) = 0.27$ (p = .603)
Mental	6	0.58	[0.36, 0.80]	$Q_{W1}(5) = 5.61$ (p = .346)
Chronological	5	0.66	[0.44, 0.88]	$Q_{W2}(4) = 3.14$ (p = .535)

Note: k = number of studies

Table 4
Summary of quantitative moderating variable analysis

	k	b	95% CI	
Quality	11	-0.05	[-0.19, 0.08]	$Q_{model}(1) = 0.56$ (p = .455) $Q_{model}(1) = 8.46$ (p = .489)
Mean chronological age	11	-0.00	[-0.01, 0.01]	$Q_{model}(1) = 0.12$ (p = .735) $Q_{model}(1) = 8.90$ (p = .446)

Note: k = number of studies

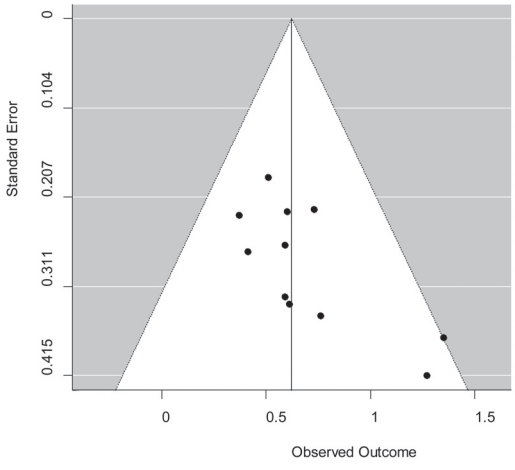

Figure 3. Funnel plot

Discussion

The main result of this meta-analysis is that the individuals with ID experience more difficulties than the controls in CMI tasks. The estimated effect size ($d = 0.62$) shows that the difference between the control group and the ID group is statistically significant and has a medium magnitude. As far as we know, this is the first time that a combined effect size was obtained, indicating the degree of impairment of the CMI processes in people with ID.

The effect sizes of all included individual studies had the expected direction, greater impairment of the CMI in people with ID, although in some of the primary studies did not show statistically significant differences (Figure 2), probably due to lack of statistical power. This result is important because the CMI processes intervene in the effective recovery of memory traces, and an impairment of these processes can lead to forgetfulness of valuable information (Anderson & Weaver, 2009; Hasher & Zacks, 1988).

Despite the presence of homogeneity, for theoretical reasons we analysed a set of potentially moderating variables. Some authors, aware that the results could be related to the level of intelligence or severity (Patterson, Rapsey, & Glue, 2013). However, we did not have enough information from the studies to carry them out. We fit categorical and meta-regression models with the moderators that had at least three studies within each category. With variables that did not meet this condition, we calculated a combined estimate of the categories.

For the developmental stage, we observed a significant effect at all stages except the adult. In studies with people with ID, the children and adolescence stages had the greatest number of studies (Table 1). In this meta-analysis, little research studied the adult, older adult and elderly populations with ID. However, more studies are needed in adults with ID to know about possible premature aging in CMI.

Regardless of the type of aetiology, we observed that people with DS were the ones who moved the furthest away from the control group. This pattern is similar to previous studies (Bower & Hayes, 1994; Varnhagen, Das, & Varnhagen, 1987).

Within the analysis of dependent variables, priming tasks and number of words contained more studies and, in them, inter-group discrepancies were observed. In terms of the type of task—experimental tasks or standardised tests—the differences between the groups were significant in both. When standardised tests were used, the differences between the groups were greater—due to good psychometric properties and less error variance—.

In the analysis of memory systems, regardless of the system used, people with ID experienced more difficulties than TD people. These results are similar to those observed in memory systems and executive function by other authors (Lanfranchi, Baddeley, Gathercole, & Vianello, 2012). Within the categorical variables, we fit that moderator to a categorical model. It was observed that people with ID had greater difficulties in implementing CMI processes and that this occurred regardless of the type of comparison used.

In the variables where meta-regression was applied, both the quality index and the mean chronological age, these variables did not account for a significant part of the variance in the effect size values. Finally, taking into account the number of studies included in this meta-analysis, it was relevant to carry out an analysis of the publication bias. Even after removing the two papers that caused the observed asymmetry, the combined effect size remained robust.

The main limitation of this meta-analysis was the small number of published studies. Despite this limitation, we were able to carry out analyses of a large number of variables thanks to the fact that the 11 articles provided very specific information and had good methodological quality.

The authors wish to emphasize that despite the fact that the initial temporal criterion of the search was 1973 and that both ancient (mental retardation or intellectual deficiency) and current descriptive (intellectual disability or cognitive impairment) were used, of the 11 articles only one was before 2000.

The assessment of the data set reveals the need for additional studies of CMI in people with ID, older than 18 years old, particularly from 31 to 45 (the range related to premature aging in ID). Research in this range will help us to understand the development (and possible involution) of the CMI processes in the adults with ID.

The limited number of studies also makes it difficult to analyze the publication bias and its role as a potential threat to the conclusions. Given that with so few studies the asymmetry tests (like the Egger's test) are unstable, the conclusions that are derived from them are not very reliable. A more robust diagnosis of the presence of publication bias around this question must wait until there is a greater number of studies. However, the small sample of studies has not prevented the fail-safe number from successfully exceeding Rosenthal's criteria. This allows us to conclude with confidence that although the estimate of the effect size could be inflated by a potential publication bias, the very existence of the relationship between the variables is not challenged by that threat.

Likewise, a greater number of studies would offer the possibility of studying moderating variables such as intelligence or severity level. Moreover, with respect to future research, a greater number of studies is necessary to enable the comparison of CMI processes separately depending on the aetiology of ID. Also, it is necessary empirical studies which compares more specific age ranges with similar experimental tasks. A better understanding of CMI processes would enable us to assess the effectiveness of CMI intervention programs and design better ones, for the fulfilment of the right to have the best possible quality of life.

Acknowledgements

Part of this work has been supported by "Autour des Williams". Project "Inhibitory processes and memory mechanisms in adults with Williams syndrome: A neuropsychological and functional connectivity approach using magnetoencephalography".

References

Amieva, H., Phillips, L. H., Della Sala, S., & Henry, J. D. (2004). Inhibitory functioning in Alzheimer's disease. *Brain: A Journal of Neurology, 127*, 949-964. doi:10.1093/brain/awh045

Anderson, M. C. (2007). Inhibition: Manifestations in long-term memory. In Y. Dudai, R. Roediger, E. Tulving, & S. Fitzpatrick (Eds.), *The science of memory: Concepts* (pp. 295-299). New York: Oxford University Press.

Anderson, M. C., & Bjork, R. A. (1994). Mechanisms of inhibition in long-term memory: A new taxonomy. In D. Dagenbach & T. Carr (Eds.), *Inhibitory processes in attention, memory and language* (pp. 265-326). San Diego: Academic Press.

Anderson, M. C., & Weaver, C. (2009). Inhibitory control over action and memory. In L. Squire (Ed.), *The new encyclopedia of neuroscience* (pp. 153-163). Oxford: Academic Press.

*Baker, S., Hooper, S., Skinner, M., Hatton, D., Schaaf, J., Ornstein, P., & Bailey, D. (2011). Working memory subsystems and task complexity in young boys with fragile X syndrome. *Journal of Intellectual Disability Research, 55*, 19-29. doi:10.1111/j.1365-2788.2010.01343.x

*Belacchi, C., Passolunghi, M. C., Brentan, E., Dante, A., Persi, L., & Cornoldi, C. (2014). Approximate additions and working memory in individuals with Down syndrome. *Research in Developmental Disabilities, 35*, 1027-1035. doi:10.1016/j.ridd.2014.01.036

*Borella, E., Carretti, B., & Lanfranchi, S. (2013). Inhibitory mechanisms in Down syndrome: Is there a specific or general deficit? *Research in Developmental Disabilities, 34*, 65-71. doi:10.1016/j.ridd.2012.07.017

Borenstein, M., Hedges, L. V., Higgins, J. P. T., & Rothstein, H. R. (2009). *Introduction to meta-analysis*. Chichester: Wiley.

Botella, J., & Sánchez-Meca, J. (2015). *Meta-análisis en ciencias sociales y de la salud* [Meta-analysis in social and heatlh sciences] (2015 ed.). Madrid: Síntesis.

Bower, A., & Hayes, A. (1994). Short-term memory deficits and Down syndrome: A comparative study. *Down Syndrome Research and Practice, 2*, 47-50. doi:10.3104/reports.29

*Brega, A. G., Goodrich, G., Bennett, R. E., Hessl, D., Engle, K., Leehey, M. A., … Grigsby, J. (2008). The primary cognitive deficit among males with fragile X-associated tremor/ataxia syndrome is a dysexecutive syndrome. *Journal of Clinical and Experimental Neuropsychology, 30*, 853-869. doi:10.1080/13803390701819044

*Carretti, B., Belacchi, C., & Cornoldi, C. (2010). Difficulties in working memory updating in individuals with intellectual disability. *Journal of Intellectual Disability Research, 54*, 337-345. doi:10.1111/j.1365-2788.2010.01267.x

Carro, E. H. (2016). *Methodological advances in the evaluation of the quality of scientific production* (Doctoral dissertation, University of Sevilla). Retrieved from https://idus.us.es

Clapp, W. C., Rubens, M. T., & Gazzaley, A. (2010). Mechanisms of working memory disruption by external interference. *Cerebral Cortex, 20*, 859-872. doi:10.1093/cercor/bhp150

Cohen J. (1988). *Statistical power analysis for the behavioral sciences* (2nd ed.). New Jersey: Lawrence Earlbaum Associates.

Corman, C. D., & Wickens, D. D. (1968). Retroactive inhibition in short-term memory. *Journal of Verbal Learning and Verbal Behavior, 7*, 16-19. doi:10.1016/S0022-5371(68)80157-4

*Danielsson, H., Henry, L., Rönnberg, J., & Nilsson, L. G. (2010). Executive functions in individuals with intellectual disability. *Research in Developmental Disabilities, 31*, 1299-1304. doi:10.1016/j.ridd.2010.07.012

Devenny, D. A., Krinsky, S. J., Kittler, P. M., Flory, M., Jenkins, E., & Brown, W. T. (2004). Age-associated memory changes in adults with Williams syndrome. *Developmental Neuropsychology, 26*, 691-706. doi:10.1207/s15326942dn2603_3

Diamond, A. (2013). Executive functions. *Annual Review of Psychology, 64*, 135-168. doi:10.1146/annurev-psych-113011-143750

Duval, S., & Tweedie, R. (2000). A nonparametric "trim and fill" method of accounting for publication bias in meta-analysis. *Journal of the American Statistical Association, 95*, 89-98. doi:10.1080/01621459.2000.10473905

Egger, M., Smith, G. D., Schneider, M., & Minder, C. (1997). Bias in meta-analysis detected by a simple, graphical test. *British Medical Journal, 315*, 629–634. doi:10.1136/bmj.315.7109.629

Friedman, N. P., & Miyake A. (2017). Unity and diversity of executive functions: Individual differences as a window on cognitive structure. *Cortex, 86*, 186-204. doi:10.1016/j.cortex.2016.04.023

Gazzaley, A., Cooney, J. W., Rissman, J., & D'Esposito, M. (2005). Top-down suppression deficit underlies working memory impairment in normal aging. *Nature Neuroscience, 8*, 1298-1300. doi:10.1038/nn1543

Greer, J., Riby, D. M., Hamiliton, C., & Riby, L. M. (2013). Attentional lapse and inhibition control in adults with Williams Syndrome. *Research in Developmental Disabilities, 34*, 4170-4177. doi:10.1016/j.ridd.2013.08.041

Hasher, L., & Zacks, R. T. (1988). Working memory, comprehension, and aging: A review and a new view. *Psychology of Learning and Motivation, 22*, 193-225. doi:10.1016/S0079-7421(08)60041-9

Hawkins, B. A., Eklund, S. J., James, D. R., & Foose, A. K. (2003). Adaptive behavior and cognitive function of adults with Down syndrome: Modeling change with age. *Mental Retardation, 41*, 7-28. doi:10.1352/00476765(2003)041%3C0007:ABACFO%3E2.0.CO;2

Huedo-Medina, T. B., Sánchez-Meca, J., Marín-Martínez, F., & Botella, J. (2006). Assessing heterogeneity in meta-analysis: Q statistic or I^2 index? *Psychological Methods, 11*, 193-206. doi:10.1037/1082-989X.11.2.193

Lanfranchi, S., Baddeley, A., Gathercole, S., & Vianello, R. (2012). Working memory in Down syndrome: Is there a dual deficit? *Journal of Intellectual Disability Research*, *56*, 157-166. doi:10.1111/j.1365-2788.2011.01444.x

*Lanfranchi, S., Cornoldi, C., & Vianello, R. (2004). Verbal and visuospatial working memory deficits in children with Down syndrome. *American Journal on Mental Retardation, 109*, 456-466. doi:10.1352/0895-8017(2004)109<456:VAVWMD>2.0.CO;2

*Lanfranchi, S., Jerman, O., & Vianello, R. (2009). Working memory and cognitive skills in individuals with Down syndrome. *Child Neuropsychology, 15*, 397-416. doi:10.1080/09297040902740652

Lezak, M. D. (1982). The problem of assessing executive functions. *International Journal of Psychology, 17*, 281-297. doi:10.1080/00207598208247445

Lipsey, M. W. (2009). Identifying interesting variables and analysis opportunities. In H. Cooper, L. V. Hedges, & J. C. Valentine (Eds.), *The handbook of research synthesis and meta-analysis* (pp. 147-158). New York: Russell Sage Foundation.

Lipsey, M. W., & Wilson, D. B. (2001). *Practical meta-analysis*. Thousand Oaks: Sage Publications.

Luria, A. (1966). *Higher cortical functions in man* (2012 ed.). New York: Springer-Verlag.

*Merrill, E. C., & Taube M. (1996). Negative priming and mental retardation: The processing of distractor information. *American Journal on Mental Retardation, 101*, 65-71. Retrieved from http://emerrill.people.ua.edu/

Miyake, A., Friedman, N. P., Emerson, M. J., Witzki, A. H., Howerter, A., & Wager, T. D. (2000). The unity and diversity of executive functions and their contributions to complex "frontal lobe" tasks: A latent variable analysis. *Cognitive Psychology, 41*, 49-100. doi:10.1006/cogp.1999.0734

*O'Dekirk, M., & Merrill, E. C. (2006). Inhibition of return and negative priming by persons with and without mental retardation. *American Journal on Mental Retardation, 111*, 389-399. doi:10.1352/0895-8017(2006)111[389:IORANP]2.0.CO;2

Patterson, T., Rapsey, C. M., & Glue, P. (2013). Systematic review of cognitive development across childhood in Down syndrome: Implications for treatment interventions. *Journal of Intellectual Disability Research, 57*, 306-318. doi:10.1111/jir.12037.

Review Manager. (2008). RevMan (Version 5.0) [Computer program]. Copenhagen: The Nordic Cochrane Centre.

Rey, A. (1958). *L'examen clinique en psychologie*. [The clinical examination in psychology]. Oxford: Presses Universitaires de France.

Rosenthal, R. (1979). The file drawer problem and tolerance for null results. *Psychological Bulletin*, *86*, 638-641. doi:10.1037/0033-2909.86.3.638

Roth, G. M., Sun, B., Greensite, F. S., Lott, I. T., & Dietrich, R. B. (1996). Premature aging in persons with Down syndrome: MR findings. *American Journal of Neuroradiology, 17*, 1283-1289. Retrieved from http://www.ajnr.org/

*Sampaio, A., Sousa, N., Férnandez, M., Henriques, M., & Gonçalves, O. F. (2008). Memory abilities in Williams syndrome: Dissociation or developmental delay hypothesis? *Brain and Cognition, 66*, 290-297. doi:10.1016/j.bandc.2007.09.005

Sanduvete, S. (2008). *Methodological innovations in continuous assessment* (Doctoral dissertation, University of Sevilla, Spain). Retrieved from http://fondosdigitales.us.es/

Strech, D., & Sofaer, N. (2012). How to write a systematic review of reasons. *Journal of Medical Ethics*, *38*, 121-126. doi:10.1136/medethics-2011-100096

Tipper, S. P. (2001). Does negative priming reflect inhibitory mechanisms? A review and integration of conflicting views. *Quarterly Journal of Experimental Psychology: Human Experimental Psychology, 54*, 321–343. doi:10.1080=713755969

Varnhagen, C. K., Das, J. P., & Varnhagen, S. (1987). Auditory and visual memory span: Cognitive processing by TMR individuals with Down syndrome or other etiologies. *American Journal of Mental Deficiency*, *91*(4), 398-405.

Viechtbauer, W. (2010). Conducting meta-analyses in R with the metafor package. *Journal of Statistical Software, 36*(3), 1-48. doi:10.18637/jss.v036.i03

Wright, R. W., Brand, R. A., Dunn, W., & Spindler, K. P. (2007). How to write a systematic review. *Clinical Orthopaedics and Related Research, 455*, 23-29. doi:10.1097/BLO.0b013e31802c9098

Zigman, W. B. (2013). Atypical aging in Down syndrome. *Developmental Disabilities Research Reviews, 18*, 51-67. doi:10.1002/ddrr.1128