Systematic Review

A meta-analysis of the executive function components inhibition, shifting, and attention in intellectual disabilities

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Abstract

Background Executive function is a concept for higher-order cognitive functions, which have the role of controller and modulator of cognitive abilities. The consensus in the literature is that people with an intellectual disability perform significantly lower on executive function tasks than groups matched on chronological age. The comparison with groups matched on mental age is less clear. Therefore, the objective of this meta-analysis was to investigate to what extent executive function is impaired in people with intellectual disability compared with a typically developing control group matched on mental age. It was also investigated if the executive function component and intellectual disability aetiology moderated the effect.

Methods Eligibility criteria were participants with intellectual disability ($IQ \leq 75$) without a dual diagnosis; a comparison group matched on mental age; executive function outcome reported in a group comparison study design with $n \geq 10$. Working

memory tasks and ratings of executive function were not included. The literature search yielded 6637 potentially interesting articles. Twenty-six studies (with 99 effect sizes) including 1395 participants were included in the quantitative synthesis.

Results A multilevel random-effects meta-analysis found that people with intellectual disability performed statistically significantly lower than the mental age-matched group on the executive function tasks, $g = -0.34$, 95% confidence interval = $[-0.53, -0.16]$. However, the heterogeneity between effect sizes was large. The intellectual disability aetiology moderator was significant, but it only reduced the heterogeneity marginally.

Conclusion The overall conclusion is that individuals with an intellectual disability have more problems with executive function tasks than mental age-matched controls. Limitations are the large unexplained variance and the remarkably high number (69) of different tests that were used, which make more detailed conclusions problematic. This meta-analysis implies that future studies need to be of better quality, to have higher power, and to a higher degree use the same executive function tests.

Keywords Attention, Executive function, Inhibition, Intellectual disability, Shifting

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Background

Rationale

People with an intellectual disability (ID) have low performance on executive function (EF) tasks (Danielsson *et al.* 2010; Costanzo *et al.* 2013), but the results are heterogeneous. One way to find overall patterns and investigate potential moderators that can explain differences in results is to conduct a meta-analysis (Lipsey and Wilson 2001). Therefore, the present study aimed to conduct a systematic review with an additional meta-analysis on EF in ID.

Executive function, which also can be called executive control or cognitive control, refers to a family of top-down mental processes needed when you must concentrate and pay attention, according to Diamond (2013). Different researchers have different views on whether EF is a unitary construct, a multifaceted construct, or both. Some argue that EF refers to several distinct abilities, which are only loosely related; others claim that there is one underlying ability, which is responsible for all EF abilities (Blair *et al.* 2005, pp. 192–193). However, the consensus in the literature is that EF consists of separated but related cognitive processes (Maricle and Avirett 2012). A similar view is presented in one of the most cited studies in the EF literature (Miyake *et al.* 2000). They treat inhibition, shifting and updating as the three target EF components, which are ‘not completely independent and seem to share some underlying commonality’ and therefore indicate both unity and diversity of EF (Miyake *et al.* 2000). Other studies (e.g. Menghini *et al.* 2010; Costanzo *et al.* 2013) use a broader definition which includes attention. In conclusion, EF can be defined in many ways and the different definitions are typically vague.

For the scope of this article, we needed a definition of EF with clear boundaries so that the search and coding of articles could be efficient. To avoid the problem of the vague EF definitions, we have defined EF as having the components attention, inhibition, shifting and updating. That is, EF as an umbrella term is not defined beyond the components, which will be defined in the coming paragraphs. Only laboratory measures of EF will be included because subjective ratings and measures of cognition correlate very low (refer to e.g. a meta-analysis on memory with $r = 0.06$ Crumley *et al.* 2014).

Attention refers to all aspects of human cognition that a person can control, and all aspects of cognition related to limited resources or capacity dealing with that, which is close to the Shiffrin (1988) definition. Attention can be further divided into for example selective attention, sustained attention, and self-regulatory attention.

The definitions of inhibition and shifting are in line with the Miyake *et al.* (2000) definition. Inhibition is defined as the ability to deliberately suppress an inappropriate, automatic response. Typical inhibition tasks include stroop and go/no-go tasks. Different Tower tasks belong to inhibition, following Miyake *et al.* (2000), even though some papers treat them as planning or problem-solving tasks. Shifting is the ability to switch or change between rules or tasks efficiently. Design fluency switching tasks and card sorting tasks with rule switching are included in shifting.

Updating involves active updating of information and decisions that are made on the updated information. This definition will exclude typical working memory tasks like dual-task span tasks. This is a deliberate choice because many test batteries for IQ include tests of working memory or short-term memory. Therefore, those tasks are part of the definition of the ID group. To avoid this methodological challenge and to make sure that the other EF tasks are not only a small part of the larger literature on working memory in ID (refer to for example a meta-analysis on EF in Down syndrome where short-term memory and working memory tasks accounted for 74% of their studies Tungate and Conners 2021), working memory tasks were not included in our definition of EF. Additionally, different aspects of working memory in people with ID are already summarised, for example, an integrative review (Lifshitz *et al.* 2016), book chapters (Henry 2011), and a meta-analysis of working memory training (Danielsson *et al.* 2015). One example of an updating task that is included in our definition is the N-back task, where the participant is to judge if one item (in a serially presented stream of items) is the same as the item presented N positions earlier (e.g. two positions earlier). In this task, the information needs to be updated after every item, and the decision is taken on the updated information.

No cognitive abilities can be measured without dependency on other cognitive abilities. This is also

true for EF, something that Miyake *et al.* (2000) called the task impurity problem. Even though the non-specific EF tasks lack specificity and sometimes even sensitivity, they are claimed to be well suited to assess severe EF deficits (Snyder *et al.* 2015). To handle this problem in the meta-analysis, tasks that to a large degree depend on several EF components were included in the EF component ‘combination’. Examples of tasks in this category are planning tasks, fluency tasks, and card sorting tasks.

In studies on people with ID, it is common to have a comparison group matched on mental age. This is based on the idea that ID is a delayed development and if the ID group is compared with a group matched on mental age (from here MA controls), the groups should perform on par, for more details, refer to, for example, Zigler and Balla (1982). In other words, the prediction based on the delayed development model would be to get an effect size of zero in this meta-analysis. The concept of mental age has been criticised (Thurstone 1926) and using comparison groups matched on mental age has its limitations (Burack *et al.* 2004; Jarrold and Brock 2004). However, most studies on EF in people with ID use a mental age comparison group as it is still the best available choice to investigate if a specific ability is delayed compared with general cognitive ability (Messer *et al.* 2021). Therefore, mental age will be the comparison group used in this meta-analysis.

Most EF studies on people with ID include one of the following experimental groups: ID with non-specific cause (NSID), Down syndrome (DS), Williams syndrome (WS) or Fragile X syndrome (FXS). Cross syndrome studies indicate differences between cognitive profiles of the four subtypes where NSID have a homogenous cognitive profile and the syndrome specific profiles seem to have clearer strengths and weaknesses, for example, people with DS and FXS perform well on visuospatial tasks (Kogan *et al.* 2004) and people with WS perform well on verbal tasks (Costanzo *et al.* 2013). The differences between the groups’ general cognitive profiles have also been found in studies on EF (Carney *et al.* 2013; Danielsson *et al.* 2016) even though studies with ID groups of different aetiologies are rare. For all these reasons, the low performance of the ID group on EF tasks can sometimes be explained by genetic factors, other times by an overall low intelligence level, as

suggested by Lanfranchi *et al.* (2010). To investigate this further, ID aetiology will be included as a moderator.

Objectives

To the best of our knowledge, no meta-analysis has been conducted on EF in people with ID without dual diagnosis so far. Therefore, this work unified and analysed the research, which has already been conducted. This was further specified in the following research questions:

- Do people with ID have impaired EF compared with MA controls?
- Do EF component and ID aetiology moderate the effect?

Overall, we expected to find a medium to large effect size. We expected a large heterogeneity between the effect sizes that could partly be explained by moderators.

Methods

This article follows an open science approach (e.g. Klein *et al.* 2018) by having material (full documentation of searches, search files, full-text screening exclusion for all articles), the dataset (coding of the included articles) and analysis code openly available online (<http://doi.org/10.17605/OSF.IO/NJSD3>). The analysis was not preregistered.

Information sources, search and study selection

For the identification of relevant studies, a literature search was conducted in the databases PubMed, Scopus, PsycINFO and Web of Science (core collection). The literature search (last updated 21 August 2019) was based on free text and predefined search words (different for each database) related to ‘intellectual disability’ AND ‘executive function’ with limitations to journal articles in English. A full search strategy for all databases can be found in <http://doi.org/10.17605/OSF.IO/NJSD3>.

The process in the meta-analysis followed the PRISMA model (Liberati *et al.* 2009) and included four main steps: identification, screening, eligibility and inclusion, for details, refer to the PRISMA flow diagram in Figure 1 that is adapted from

Figure 1. PRISMA flow diagram. [Colour figure can be viewed at wileyonlinelibrary.com]

(Moher *et al.* 2009) and the PRISMA checklist in Appendix A. The search for all four databases yielded 6582 potentially relevant results. Reference lists of finally included articles were scanned. Also, grey literature was requested at the European Conference on Psychological Theory and Research on Intellectual and Developmental Disabilities in Padua 2018, and authors who had published articles on EF in ID were contacted via email and asked for additional data,

matching the criteria. This led to 55 additional records. The total number after all duplicates that were removed was 3468. The number of full-text articles assessed for eligibility was 217. Twenty-six of all papers met the inclusion criteria. The other articles had to be excluded due to various reasons; refer to Figure 1 for details. All records in the title screening and abstract screening were assessed by at least one of three reviewers (H. D., M.S. and M.A.). If the

reviewer was uncertain, the record was included in the next round of screenings. The full-text screening was performed by two reviewers (H.D. and M.S.) and then checked by the other reviewer. Disagreements were solved by discussion. The coding of the articles was conducted by two coders (H.D. and M.S.) and checked for plausibility by the coders. Functions from the R packages Metagear (Lajeunesse 2016) and Revtools (Westgate 2019) were used to identify duplicates and to distribute the screening among the reviewers in the screening process.

Eligibility criteria

Studies had to meet (and report) the following criteria, in P(I)COS format (I for Intervention was not relevant), to be included in the present meta-analysis—participants: reported $IQ_{max} \leq 75$ of an ID group (including syndromes associated with an ID, e.g. DS, WS or FXS) without a dual diagnosis (such as ASD, ADHD, learning disability and dementia); Comparison: include a comparison group (without disabilities) matched on mental age (based on a standardised test); outcome: results on (our definition of) EF (attention, inhibition, shifting, updating and similar, but not working memory dual tasks, theory of mind tests) were reported with enough information to calculate an effect size; study design: group comparison with ≥ 10 participants per group.

Intellectual disability is generally defined as having $IQ < 70$, but given that there is an estimated margin of error of 5 points, which should be considered when making a diagnosis of ID (Schalock *et al.* 2010), we have used $IQ_{max} \leq 75$ as our criterion. Many studies have not reported IQ_{max} or IQ at all. To include more studies, IQ_{max} has been estimated where it is not reported. IQ was estimated as $100 * \text{mental age} / \text{chronological age}$ (for $CA < 20$ years). If IQ_{max} was not reported, it was estimated as $IQ_{mean} + 2 * IQ_{SD}$ if possible. If this also was not possible, studies with $IQ_{mean} < 50$ were estimated to have $IQ_{max} \leq 75$ and were also included.

Data collection process

To make analyses possible, different variables were extracted from the studies. They can be divided into demographic variables (publication year, sex ratio, chronological age, mental age, IQ and ID aetiology) and methodological variables (sample size, EF test

used, EF component and domain). Not all data were available for all studies. When a paper tested multiple ID groups, all ID groups that met the inclusion criteria were coded separately. When several EF measures were used in a paper, the most relevant measure (e.g. accuracy) was used if several measures were given for one test, but if the measures belonged to different tests, all effect sizes were included, even if they claimed to measure the same EF component. The coding of the EF components used the categories shifting, updating, inhibition, attention and combination. The categories follow our definition of EF and are an attempt to get some rough division of EF. The coding was hard to do because many tests claim to measure several EF components, and even when the same test was used in several studies, it was claimed to measure different aspects of EF.

Risk of bias in individual studies

It is recommended that meta-analyses include a risk of bias assessment to get an overview of the quality of the individual articles, which can be a base for analysis if the quality of the articles influences the results. Quality appraisal tools are commonly suitable for one specific design, for example, intervention studies, cohort studies or correlational studies. There is, however, to the best of our knowledge, no quality appraisal tool for the type of design (group comparisons) that this article is about. Therefore, we have created a short tool with four questions, influenced by several quality appraisal tools. The questions were (1) are the groups well matched on mental age? (2) Does the study have good statistical power? (3) Was the reliability of the test reported? and (4) Is the test used established? For each question, a score (range 0–2) was given and then a quality index was calculated as the sum of those scores (0–8). The matching on mental age was calculated as an effect size of the group difference, where an absolute group difference (in Cohen's d) of < 0.20 gave the score 2, < 0.40 gave the score 1, and score 0 was for studies with larger differences or studies where this could not be calculated because of lack of reporting. Statistical power was calculated for detecting a directional medium effect (Cohen's $d = 0.5$). Each effect got a score of 2 if the statistical power was $> 80\%$ power, 1 if it was $> 50\%$; and 0 if it was $< 50\%$. A reliability score of 2 was for studies that reported some type of reliability (e.g. Cronbach's

alpha or test–retest reliability) for the test of the study sample, a score of 1 if the tests reliability was reported on another sample, and 0 if it was not reported at all. For the last quality question, a score of 2 was given if the test used was referenced in the text, 1 if the test was an adaption of a referenced test and 0 if the test was new or not referenced at all.

Statistical analysis

The data was analysed using the Metafor package (Viechtbauer 2010) in R (R Core Team 2019) using R studio software (RStudio Team 2016) and the manuscript was formatted using papaja (Aust and Barth 2018) and citr (Aust 2018). We calculated each study's effect size Hedges' g , as recommended by Borenstein *et al.* (2009), based on means, SD s and n , with the MA controls as the reference group. For some effect sizes, the direction of the scale was reversed at the individual measurement level, so that negative effect size values reflected lower performance for the group with ID. The mean effect over all samples was estimated using the restricted maximum likelihood approach, as recommended by Van den Noortgate *et al.* (2013).

Many studies reported more than one effect size. To handle this dependency between effect sizes, a multilevel random-effects meta-analysis approach, as recommended by Assink and Wibbelink (2016), was used. This approach has one random effect for each study as an addition to the random effect for each effect size. The likelihood ratio tests yielded that the fit of the multilevel model was significantly better than the fit of the reduced models ($AIC_{full\ model} = 187.90$, $AIC_{reduced\ model\ without\ between-studies\ variance} = 238.99$, $AIC_{reduced\ model\ without\ within-studies\ variance} = 196.21$). Therefore, the multilevel random-effects analysis was used for all analyses.

The effect size heterogeneity was tested with the Metafor package's version of Cochran's Q test. How much of the total variation across studies was due to heterogeneity rather than sampling error (Higgins and Thompson 2002) is presented as a percentage (I^2) for both level 2 (variance within studies) and level 3 (variance between studies). Moderator analyses were conducted to test our second research question, using the coded variables ID aetiology and EF component as moderators. Subgroup analyses were carried out for significant moderators. Further, exploratory

moderator analyses were conducted on domain, chronological age, mental age and IQ.

It is recommended that publication bias is assessed with a variety of methods (Carter *et al.* 2017). However, only two methods were available for multilevel models in the Metafor package, so only a funnel plot and the rank correlation test examining Kendall's tau were used. For funnel plot analysis, asynchrony is an indicator of publication bias (Sterne *et al.* 2011). As a measure of sensitivity, Cook's distance was used to investigate potential outliers, with a cut-off of 1, as recommended by Cook (1979). Statistical power was calculated with the pwr package in r (Champely 2020) as a directional effect for a t test with two groups.

Results

The coded information for each effect size included in the study can be found in Tables 1 and 2 and the full references to the included articles are marked with * in the reference list. The 26 studies included in this meta-analysis were published across 16 different journals with the median publication year 2012 (1962–2020). The study means were weighted by the number of participants in each study. There were 1395 participants included, 677 with ID (55% male participants) and 718 MA-controls (55% male participants). The median number of ID participants was 21 (range 10–65), which equals having 80% power to detect group differences of Cohen's $d = -0.34$. The ID participants had mean chronological age of 18.83 years [range of study means 4.34–49.88], mean mental age of 6.31 years [1.88–11.17] and mean IQ of 46.50 [19.92–65.75]. The MA-controls had mean chronological age of 6.25 years [range of study means 1.93–11.17], mean mental age of 6.23 years [1.93–11.17], and mean IQ of 108.81 [94–124].

Out of the 99 included effect sizes, 16 belonged to attention, 37 to inhibition and 24 to shifting measures. There was only one study measuring updating, which is probably because our definition of EF excluded working memory tests, which other definitions of EF consider to be updating. In the moderator analyses, the updating study will be part of the category combination. Twenty-one effect sizes were categorised as combination which mostly included tests of planning, sorting and fluency. There was a large variety of tests used to assess EF.

Table 1 Information on the 99 effect sizes from the 26 studies

Study	EF component	Domain	ID aetiology	ID group				MA control group						Hedges' g	var
				n	IQ	MA	CA	M	SD	n	MA	CA	M	SD	
Borella <i>et al.</i> 2013	Inhibition	Verbal	DS	19	38.26	5.58	14.58	3.76	4.79	19	5.68	5.20	4.02	4.26	0.06
Borella <i>et al.</i> 2013	Inhibition	Verbal	DS	19	38.26	5.58	14.58	1.22	1.73	19	5.68	5.20	0.25	0.23	-0.77
Borella <i>et al.</i> 2013	Inhibition	Verbal	DS	19	38.26	5.58	14.58	0.80	0.79	19	5.68	5.20	1.47	0.96	0.75
Carney <i>et al.</i> 2013	Inhibition	Verbal	DS	25	44.17	6.03	13.64	7.08	4.44	26	6.53	6.14	5.58	3.21	-0.38
Carney <i>et al.</i> 2013	Inhibition	Visuospatial	DS	25	44.17	6.03	13.64	12.67	4.38	26	6.53	6.14	17.85	4.46	1.15
Carney <i>et al.</i> 2013	Combination	Verbal	DS	25	44.17	6.03	13.64	23.76	8.57	26	6.53	6.14	22.73	7.41	0.13
Carney <i>et al.</i> 2013	Combination	Visuospatial	DS	25	44.17	6.03	13.64	11.04	4.60	26	6.53	6.14	12.19	4.38	-0.25
Carney <i>et al.</i> 2013	Shifting	Verbal	DS	25	44.17	6.03	13.64	3.20	3.14	26	6.53	6.14	1.67	3.35	-0.46
Carney <i>et al.</i> 2013	Shifting	Visuospatial	DS	25	44.17	6.03	13.64	1.32	2.60	26	6.53	6.14	1.71	2.14	0.16
Carney <i>et al.</i> 2013	Inhibition	Verbal	WS	24	49.62	6.74	13.59	9.17	7.07	26	6.53	6.14	5.58	3.21	-0.65
Carney <i>et al.</i> 2013	Inhibition	Visuospatial	WS	24	49.62	6.74	13.59	22.09	7.79	26	6.53	6.14	17.85	4.46	-0.66
Carney <i>et al.</i> 2013	Combination	Verbal	WS	24	49.62	6.74	13.59	25.25	8.93	26	6.53	6.14	22.73	7.41	0.30
Carney <i>et al.</i> 2013	Combination	Visuospatial	WS	24	49.62	6.74	13.59	7.13	2.77	26	6.53	6.14	12.19	4.38	-1.35
Carney <i>et al.</i> 2013	Shifting	Verbal	WS	24	49.62	6.74	13.59	2.83	2.79	26	6.53	6.14	1.67	3.35	-0.37
Carney <i>et al.</i> 2013	Shifting	Visuospatial	WS	24	49.62	6.74	13.59	0.81	1.27	26	6.53	6.14	1.71	2.14	0.50
Carretti <i>et al.</i> 2010	Updating	Verbal	NSID	28		6.17	38.30	5.04	3.76	28	6.50	6.40	7.29	3.98	-0.57
Clunies-Ross 1972	Shifting	Visuospatial	NSID	40	49.60	5.79		10.80	14.30	40	5.98		7.30	12.40	-0.26
Clunies-Ross 1972	Shifting	Visuospatial	NSID	40	49.60	5.79		11.80	12.00	40	5.98		5.90	4.70	-0.64
Clunies-Ross 1972	Shifting	Visuospatial	NSID	40	49.60	5.79		5.40	7.90	40	5.98		1.10	2.30	-0.73
Cortier and McKinney (1968)	Shifting	Visuospatial	NSID	16	65.75	6.77	10.35	467.25	140.85	16	6.50		501.56	115.69	-0.26
Costanzo <i>et al.</i> 2013	Attention	Verbal	WS	15	53.00	6.70	17.60	33.80	10.80	16	6.90	7.40	35.40	4.40	-0.19
Costanzo <i>et al.</i> 2013	Attention	Visuospatial	WS	15	53.00	6.70	17.60	5.30	3.20	16	6.90	7.40	8.60	1.60	-1.28
Costanzo <i>et al.</i> 2013	Attention	Verbal	WS	15	53.00	6.70	17.60	14.20	6.60	16	6.90	7.40	8.00	3.70	-1.14
Costanzo <i>et al.</i> 2013	Attention	Visuospatial	WS	15	53.00	6.70	17.60	103.60	16.00	16	6.90	7.40	96.80	17.60	-0.39
Costanzo <i>et al.</i> 2013	Inhibition	Visuospatial	WS	15	53.00	6.70	17.60	16.40	6.20	16	6.90	7.40	25.70	3.20	-1.85
Costanzo <i>et al.</i> 2013	Combination	Verbal	WS	15	53.00	6.70	17.60	23.40	5.80	16	6.90	7.40	21.70	4.40	0.32
Costanzo <i>et al.</i> 2013	Combination	Visuospatial	WS	15	53.00	6.70	17.60	7.30	3.70	16	6.90	7.40	12.50	2.10	-1.70
Costanzo <i>et al.</i> 2013	Shifting	Verbal	WS	15	53.00	6.70	17.60	6.50	2.60	16	6.90	7.40	6.70	2.30	-0.08
Costanzo <i>et al.</i> 2013	Shifting	Visuospatial	WS	15	53.00	6.70	17.60	84.40	59.60	16	6.90	7.40	52.30	32.40	-0.66
Costanzo <i>et al.</i> 2013	Shifting	Visuospatial	WS	15	53.00	6.70	17.60	143.20	54.10	16	6.90	7.40	85.00	50.70	-1.08
Costanzo <i>et al.</i> 2013	Inhibition	Verbal	WS	15	53.00	6.70	17.60	1.40	0.80	16	6.90	7.40	0.70	0.70	-0.91
Costanzo <i>et al.</i> 2013	Inhibition	Visuospatial	WS	15	53.00	6.70	17.60	89.90	7.90	16	6.90	7.40	93.00	4.00	-0.49
Danielsson <i>et al.</i> 2012	Inhibition	Verbal	NSID	22	56.33	7.42	13.17	17.70	22.60	22	8.00	7.33	7.00	4.80	-0.64

Table 1. (Continued)

Study	EF component	Domain	ID aetiology	ID group				MA control group					Hedges' g	var
				n	IQ	MA	CA	M	SD	n	MA	CA	M	SD
Danielsson <i>et al.</i> 2012	Inhibition	Visuospatial	NSID	22	56.33	7.42	13.17	37.30	26.70	22	8.00	7.33	17.60	4.70
Danielsson <i>et al.</i> 2012	Combination	Verbal	NSID	22	56.33	7.42	13.17	0.95	0.83	22	8.00	7.33	2.10	1.10
Danielsson <i>et al.</i> 2012	Combination	Visuospatial	NSID	22	56.33	7.42	13.17	1.75	1.80	22	8.00	7.33	3.32	1.90
Danielsson <i>et al.</i> 2012	Shifting	Verbal	NSID	22	56.33	7.42	13.17	7.15	2.87	22	8.00	7.33	8.05	1.86
Danielsson <i>et al.</i> 2012	Shifting	Visuospatial	NSID	22	56.33	7.42	13.17	38.00	16.20	22	8.00	7.33	34.30	8.20
Danielsson <i>et al.</i> 2012	Combination	Verbal	NSID	22	56.33	7.42	13.17	13.60	8.60	22	8.00	7.33	24.45	5.10
Danielsson <i>et al.</i> 2012	Combination	Verbal	NSID	22	56.33	7.42	13.17	24.10	9.40	22	8.00	7.33	26.70	4.60
Danielsson <i>et al.</i> 2012	Combination	Visuospatial	NSID	22	56.33	7.42	13.17	8.55	5.57	22	8.00	7.33	9.82	3.26
Greer <i>et al.</i> 2017	Inhibition	Visuospatial	WS	11		11.17	42.58	500.65	64.56	13		11.17	490.67	59.54
Hocking <i>et al.</i> 2013	Inhibition	Visuospatial	WS	16		10.43	24.20	948.90	205.40	16	9.76	9.03	706.70	95.40
Hooper <i>et al.</i> 2008	Inhibition	Visuospatial	FXS	42	56.10	5.44	10.10	9.24	5.09	48	5.30		13.07	2.96
Hooper <i>et al.</i> 2008	Inhibition	Verbal	FXS	31	56.40	5.55	10.10	4.61	3.32	48	5.30		2.74	1.82
Hooper <i>et al.</i> 2008	Shifting	Verbal	FXS	14	54.10	5.59	10.10	12.50	5.50	48	5.30		3.68	5.07
Hooper <i>et al.</i> 2008	Combination	Visuospatial	FXS	38	56.30	5.54	10.10	16.18	15.98	48	5.30		57.86	47.75
Hooper <i>et al.</i> 2008	Inhibition	Visuospatial	FXS	35	56.60	5.55	10.10	3.89	2.47	48	5.30		8.61	3.43
Ikeda <i>et al.</i> 2014	Inhibition	Visuospatial	NSID	11	59.10	8.86	15.83	1.89	5.06	21	8.46	8.46	1.85	2.19
Ikeda <i>et al.</i> 2014	Inhibition	Visuospatial	NSID	11	59.10	8.86	15.83	2.27	5.06	21	8.46	8.46	1.41	2.76
Ikeda <i>et al.</i> 2014	Inhibition	Visuospatial	NSID	11	59.10	8.86	15.83	0.76	1.69	21	8.46	8.46	2.40	2.50
Ikeda <i>et al.</i> 2014	Inhibition	Visuospatial	NSID	11	59.10	8.86	15.83	1.53	2.13	21	8.46	8.46	2.99	3.27
Johns <i>et al.</i> 2012	Inhibition	Visuospatial	DS	13	55.93		14.80	0.83	20.13	15		5.70	0.97	6.63
Johns <i>et al.</i> 2012	Inhibition	Visuospatial	DS	13	55.93		14.80	0.54	25.36	15		5.70	0.75	16.92
Lanfranchi <i>et al.</i> 2010	Inhibition	Verbal	DS	15	37.90	5.75	15.17	13.87	2.90	15		5.75	15.67	0.82
Lanfranchi <i>et al.</i> 2010	Shifting	Verbal	DS	15	37.90	5.75	15.17	19.00	1.96	15		5.75	19.87	1.46
Lanfranchi <i>et al.</i> 2010	Shifting	Verbal	DS	15	37.90	5.75	15.17	12.80	2.43	15		5.75	15.40	3.50
Lanfranchi <i>et al.</i> 2010	Shifting	Visuospatial	DS	15	37.90	5.75	15.17	2.00	0.85	15		5.75	3.87	1.30
Lanfranchi <i>et al.</i> 2010	Inhibition	Visuospatial	DS	15	37.90	5.75	15.17	3.80	2.68	15		5.75	9.67	2.50
Lanfranchi <i>et al.</i> 2010	Combination	Verbal	DS	15	37.90	5.75	15.17	9.13	5.36	15		5.75	8.00	4.47
Lanfranchi <i>et al.</i> 2010	Combination	Verbal	DS	15	37.90	5.75	15.17	19.60	10.19	15		5.75	13.20	7.10
Lanfranchi <i>et al.</i> 2010	Attention	Visuospatial	DS	15	37.90	5.75	15.17	10.20	4.23	15		5.75	6.67	5.15
Menghini <i>et al.</i> 2010	Inhibition	Visuospatial	WS	15	53.30	6.10	19.11	18.00	6.00	15	6.90	7.60	26.10	3.50
Menghini <i>et al.</i> 2010	Shifting	Verbal	WS	15	53.30	6.10	19.11	6.60	2.80	15	6.90	7.60	7.30	1.90
Menghini <i>et al.</i> 2010	Shifting	Visuospatial	WS	15	53.30	6.10	19.11	72.50	47.00	15	6.90	7.60	44.10	21.70
Menghini <i>et al.</i> 2010	Shifting	Visuospatial	WS	15	53.30	6.10	19.11	133.80	56.60	15	6.90	7.60	89.60	56.40
Menghini <i>et al.</i> 2010	Inhibition	Visuospatial	WS	15	53.30	6.10	19.11	521.30	90.70	15	6.90	7.60	556.50	72.50

Table 1. (Continued)

Study	EF component	Domain	ID aetiology	ID group				MA control group						Hedges' g	var	
				n	IQ	MA	CA	M	SD	n	MA	CA	M			SD
Menghini <i>et al.</i> 2010	Inhibition	Verbal	WS	15	53.30	6.10	19.11	1.20	1.11	15	6.90	7.60	1.00	0.50	-0.23	0.13
Menghini <i>et al.</i> 2010	Inhibition	Verbal	WS	15	53.30	6.10	19.11	59.20	29.60	15	6.90	7.60	39.20	5.50	-0.91	0.15
Menghini <i>et al.</i> 2010	Attention	Visuospatial	WS	15	53.30	6.10	19.11	107.30	15.80	15	6.90	7.60	95.50	20.10	-0.64	0.14
Menghini <i>et al.</i> 2010	Attention	Visuospatial	WS	15	53.30	6.10	19.11	11.40	6.80	15	6.90	7.60	5.50	1.40	-1.17	0.16
Menghini <i>et al.</i> 2010	Attention	Verbal	WS	15	53.30	6.10	19.11	5.90	3.00	15	6.90	7.60	8.40	1.60	-1.01	0.15
Menghini <i>et al.</i> 2010	Attention	Verbal	WS	15	53.30	6.10	19.11	34.50	11.60	15	6.90	7.60	35.00	4.10	-0.06	0.13
Menghini <i>et al.</i> 2010	Combination	Verbal	WS	15	53.30	6.10	19.11	23.80	5.90	15	6.90	7.60	22.90	4.50	0.17	0.13
Menghini <i>et al.</i> 2010	Combination	Visuospatial	WS	15	53.30	6.10	19.11	7.80	3.80	15	6.90	7.60	12.50	2.20	-1.47	0.17
Mento <i>et al.</i> 2019	Attention	Visuospatial	DS	33	48.20	5.59	11.58	66.30	25.20	31	5.65	5.19	77.70	15.40	-0.54	0.06
Nader-Grosbois and Lefèvre 2011	Attention	Other	NSID	29		4.47	10.56	2.13	0.42	30	4.50	4.22	1.96	0.35	0.43	0.07
Nader-Grosbois and Lefèvre 2011	Attention	Other	NSID	29		4.47	10.56	2.50	0.44	30	4.50	4.22	2.59	0.48	-0.19	0.07
Nader-Grosbois and Vieillevoys 2012	Attention	Other	NSID	40		4.62	9.59	9.55	1.91	40	4.45	4.15	10.52	1.67	-0.54	0.05
Nader-Grosbois and Vieillevoys 2012	Attention	Other	NSID	40		4.62	9.59	9.93	1.89	40	4.45	4.15	10.26	1.40	-0.20	0.05
Numminen <i>et al.</i> 2001	Inhibition	Visuospatial	NSID	24		5.63	49.88	18.33	6.13	24	5.67	5.50	18.04	7.00	-0.04	0.08
Palmqvist <i>et al.</i> 2020	Combination	Visuospatial	NSID	65	46.57	8.43	18.10	0.04	1.11	62	8.55	7.84	0.03	0.78	0.00	0.03
Palmqvist <i>et al.</i> 2020	Combination	Verbal	NSID	68	46.35	8.39	18.10	38.30	12.10	62	8.55	7.84	37.00	8.73	0.12	0.03
Palmqvist <i>et al.</i> 2020	Shifting	Visuospatial	NSID	67	46.02	8.33	18.10	3.07	3.24	62	8.55	7.84	2.68	2.94	0.13	0.03
Pennington <i>et al.</i> 2003	Inhibition	Visuospatial	DS	28	30.65	4.50	14.68	4.95	1.72	28	4.50	4.92	5.19	1.44	-0.15	0.07
Pennington <i>et al.</i> 2004	Combination	Verbal	DS	28	30.65	4.50	14.68	20.19	8.94	28	4.50	4.92	16.78	6.47	0.43	0.07
Pennington <i>et al.</i> 2005	Combination	Visuospatial	DS	28	30.65	4.50	14.68	12.70	5.01	28	4.50	4.92	10.33	5.53	0.44	0.07
Pennington <i>et al.</i> 2006	Inhibition	Visuospatial	DS	28	30.65	4.50	14.68	0.66	0.19	28	4.50	4.92	0.56	0.13	0.61	0.07
Pennington <i>et al.</i> 2000	Shifting	Visuospatial	DS	20		5.27	20.02	1,096.20	251.50	20	5.43	5.49	1,258.00	277.90	0.60	0.10
Roberts <i>et al.</i> 2015	Inhibition	Visuospatial	DS	19	43.00	1.88	4.34	0.45	0.38	40	1.90	1.93	0.66	0.41	-0.52	0.08
Smith <i>et al.</i> 2019	Combination	Visuospatial	DS	18		7.60	30.30	84.90	15.60	18	7.60	7.60	87.00	16.70	-0.13	0.11
Traverso <i>et al.</i> 2018	Inhibition	Verbal	DS	32	44.30	6.20	14.48	14.38	8.65	65	6.40	5.91	10.80	4.97	-0.55	0.05
Traverso <i>et al.</i> 2018	Inhibition	Visuospatial	DS	32	44.30	6.20	14.48	4.13	2.09	65	6.40	5.91	5.22	1.31	-0.67	0.05
Traverso <i>et al.</i> 2018	Inhibition	Verbal	DS	32	44.30	6.20	14.48	10.30	4.08	65	6.40	5.91	10.58	4.30	-0.07	0.05
Traverso <i>et al.</i> 2018	Inhibition	Visuospatial	DS	32	44.30	6.20	14.48	10.63	1.54	65	6.40	5.91	13.82	3.78	-0.98	0.05
Van der Molen <i>et al.</i> 2012	Attention	Visuospatial	FXS	27	19.92	5.54	27.81	6.85	1.51	40		5.75	7.53	0.78	-0.59	0.06
Van der Molen <i>et al.</i> 2012	Attention	Visuospatial	DS	20	25.25	5.66	22.42	7.80	0.95	40		5.75	7.53	0.78	0.32	0.08

Table 1. (Continued)

Study	EF component	Domain	ID aetiology	ID group					MA control group					Hedges' g	var	
				n	IQ	MA	CA	M	SD	n	MA	CA	M			SD
Zigler and Unell 1962	Shifting	Visuospatial	NSID	10	53.00	6.10	11.50	3.90	2.33	10	5.90	5.10	4.20	2.39	-0.12	0.20
Zigler and Unell 1962	Shifting	Visuospatial	NSID	10	54.00	5.90	11.10	5.50	2.17	10	6.00	5.10	5.60	2.37	-0.04	0.20

CA, chronological age (years); DS, Down Syndrome; EF, executive function; FXS, fragile X syndrome; ID, intellectual disability; MA, mental age (years); NSID, non-specific intellectual disability; WS, Williams syndrome.

Altogether 69 different EF tests (18 tests for the 24 shifting measures, 9 tests for the 16 attention measures, and 28 tests for the 37 inhibition measures) were used; refer to Table 2 for details.

Overall, the ID group performed significantly lower than the MA controls, $g(k = 99) = -0.34$, $[-0.53, -0.16]$, $P = <0.001$. Figure 2 shows a forest plot of the meta-analysis. The range of the effect sizes was large $[-2.20, 1.15]$. The heterogeneity of the examined effect sizes was statistically significant $Q(98) = 390.57$, $P < 0.001$, and the estimation of the proportion of observed variation reflecting true variation between effect sizes was $I^2 = 76\%$ (49% within and 28 between studies), which is large ($>75\%$) according to Higgins *et al.* (2003). A large I^2 is an indication that moderator analyses are suitable.

The planned moderator analyses showed a significant moderating effect of ID aetiology (NSID, DS, WS and FXS), but not for EF component; refer to Table 3 for details on all moderator effects. The ID aetiology moderator reduced the between-study variance marginally (8%) and was followed up with subgroup analyses for the different ID subgroups. The smallest effect size was found for DS, $g(k = 32) = -0.15$, $[-0.44, 0.14]$, $P = 0.304$, and NSID, $g(k = 28) = -0.22$, $[-0.46, 0.02]$, $P = 0.068$, but was larger for WS $g(k = 33) = -0.63$, $[-0.86, -0.40]$, $P < 0.001$ and FXS $g(k = 6) = -0.97$, $[-1.65, -0.29]$, $P = 0.014$. Only the subgroups WS and FXS were statistically significantly <0 , but the NSID group was close. Because the planned moderators only explained a small part of the estimated true variance, exploratory moderator analyses were carried out for EF domain (verbal and visuospatial) and three variables in the ID group: chronological age, mental age and IQ, but none of the moderators were significant; refer to Table 3 for details.

Publication bias was indicated with a significant Ranktest (Kendall's $\tau = -0.19$, $P = 0.005$). A visual inspection of the funnel plot in Figure 3 does not indicate publication bias, as most studies lie symmetrically around the overall effect. However, the pattern of the plot is unusual. The outmost dashed line encloses the triangle, within which 99% of studies are expected to lie in the absence of bias and heterogeneity. In our case, 22 out of 99 data points lie outside this line. We conclude from this that publication bias is probably not a major problem, but there seems to be large heterogeneity.

Table 2 Information on the tests and quality of the studies included

Study	Test	Matching	Power	Reliability	Established test	Index
Borella <i>et al.</i> 2013	Animal Stroop adapted version	0.00	0.45	0	2	4
Borella <i>et al.</i> 2013	Proactive interference task	0.00	0.45	0	1	3
Borella <i>et al.</i> 2013	Directed forgetting	0.00	0.45	0	1	3
Carney <i>et al.</i> 2013	Verbal Inhibition Motor Inhibition task verbal	−0.53	0.55	2	2	5
Carney <i>et al.</i> 2013	Verbal Inhibition Motor Inhibition task motor	−0.53	0.55	2	2	5
Carney <i>et al.</i> 2013	Nepsy-II Word generation	−0.53	0.55	2	2	5
Carney <i>et al.</i> 2013	D-KEFS design fluency	−0.53	0.55	2	2	5
Carney <i>et al.</i> 2013	D-KEFS category switching	−0.53	0.55	2	2	5
Carney <i>et al.</i> 2013	D-KEFS switching version of design fluency	−0.53	0.55	2	2	5
Carney <i>et al.</i> 2013	VIMI verbal	0.19	0.54	2	2	7
Carney <i>et al.</i> 2013	VIMI motor	0.19	0.54	2	2	7
Carney <i>et al.</i> 2013	Category fluency	0.19	0.54	2	2	7
Carney <i>et al.</i> 2013	D-KEFS design fluency	0.19	0.54	2	2	7
Carney <i>et al.</i> 2013	D-KEFS category switching	0.19	0.54	2	2	7
Carney <i>et al.</i> 2013	D-KEFS switching version of design fluency	0.19	0.54	2	2	7
Carretti <i>et al.</i> 2010	Updating word span	−0.22	0.58	0	1	3
Clunies-Ross 1972	Extradimensional shift	−0.34	0.72	0	0	2
Clunies-Ross 1972	First reversal	−0.34	0.72	0	0	2
Clunies-Ross 1972	Stereotype shift	−0.34	0.72	0	0	2
Cortier and McKinney (1968)	Cognitive flexibility test battery of 5 tasks	0.32	0.40	2	0	3
Costanzo <i>et al.</i> 2013	SVN auditory selective attention	−0.22	0.39	0	2	3
Costanzo <i>et al.</i> 2013	Score task	−0.22	0.39	0	2	3
Costanzo <i>et al.</i> 2013	Sky search	−0.22	0.39	0	2	3
Costanzo <i>et al.</i> 2013	Bells test	−0.22	0.39	0	2	3
Costanzo <i>et al.</i> 2013	Tower of London	−0.22	0.39	0	2	3
Costanzo <i>et al.</i> 2013	Category Fluency Test	−0.22	0.39	0	2	3
Costanzo <i>et al.</i> 2013	Weigl Colour Form Sort Test	−0.22	0.39	0	2	3
Costanzo <i>et al.</i> 2013	Category Fluency Test alternative version	−0.22	0.39	0	2	3
Costanzo <i>et al.</i> 2013	Trail making test	−0.22	0.39	0	2	3
Costanzo <i>et al.</i> 2013	Forma forma task	−0.22	0.39	0	2	3
Costanzo <i>et al.</i> 2013	Stroop	−0.22	0.39	0	1	2
Costanzo <i>et al.</i> 2013	Go/no-go task	−0.22	0.39	0	2	3
Danielsson <i>et al.</i> 2012	Verbal Inhibition Motor Inhibition task verbal	−0.37	0.49	0	2	3
Danielsson <i>et al.</i> 2012	Verbal Inhibition Motor Inhibition task motor	−0.37	0.49	0	2	3
Danielsson <i>et al.</i> 2012	D-KEFS sorting	−0.37	0.49	0	2	3
Danielsson <i>et al.</i> 2012	D-KEFS sorting	−0.37	0.49	0	2	3
Danielsson <i>et al.</i> 2012	Delis–Kaplan Category fluency switching	−0.37	0.49	0	2	3
Danielsson <i>et al.</i> 2012	CANTAB ID ED	−0.37	0.49	0	2	3
Danielsson <i>et al.</i> 2012	D-KEFS letter fluency	−0.37	0.49	0	2	3
Danielsson <i>et al.</i> 2012	D-KEFS category fluency	−0.37	0.49	0	2	3
Danielsson <i>et al.</i> 2012	D-KEFS design fluency	−0.37	0.49	0	2	3
Greer <i>et al.</i> 2017	Oddball	0.10	0.32	0	1	3
Hocking <i>et al.</i> 2013	Go/no-go task	0.31	0.40	0	1	3
Hooper <i>et al.</i> 2008	Day-night	0.24	0.76	0	2	4
Hooper <i>et al.</i> 2008	Self-correction on The Contingency Naming Test	0.45	0.69	0	2	3
Hooper <i>et al.</i> 2008	The Contingency Naming Test subtest 3 errors	0.54	0.49	0	2	2
Hooper <i>et al.</i> 2008	WJ Planning	0.46	0.74	0	2	3
Hooper <i>et al.</i> 2008	Nepsy Tower	0.46	0.72	0	2	3
Ikeda <i>et al.</i> 2014	Real Animal Size Test control condition	0.23	0.37	0	2	3
Ikeda <i>et al.</i> 2014	Real Animal Size Test incongruent condition	0.23	0.37	0	2	3
Ikeda <i>et al.</i> 2014	Pictorial Animal Size Test control condition	0.23	0.37	0	2	3
Ikeda <i>et al.</i> 2014	Pictorial Animal Size Test incongruent condition	0.23	0.37	0	2	3
Johns <i>et al.</i> 2012	Animal reverse categorisation task		0.36	0	1	1

Table 2. (Continued)

Study	Test	Matching	Power	Reliability	Established test	Index
Johns <i>et al.</i> 2012	Hand reverse categorisation task		0.36	0	1	1
Lanfranchi <i>et al.</i> 2010	Day-night		0.38	0	2	2
Lanfranchi <i>et al.</i> 2010	Rule Shift Card Test 1		0.38	0	2	2
Lanfranchi <i>et al.</i> 2010	Rule Shift Card Test 2		0.38	0	2	2
Lanfranchi <i>et al.</i> 2010	Modified Card Sorting Test		0.38	0	2	2
Lanfranchi <i>et al.</i> 2010	Tower of London		0.38	0	2	2
Lanfranchi <i>et al.</i> 2010	FAS verbal fluency		0.38	0	2	2
Lanfranchi <i>et al.</i> 2010	Category fluency		0.38	0	2	2
Lanfranchi <i>et al.</i> 2010	Self-ordered pointing test		0.38	0	2	2
Menghini <i>et al.</i> 2010	Tower of London		0.38	0	2	2
Menghini <i>et al.</i> 2010	Category Fluency Test alternative version		0.38	0	2	2
Menghini <i>et al.</i> 2010	Trail making test		0.38	0	2	2
Menghini <i>et al.</i> 2010	Forma forma task		0.38	0	2	2
Menghini <i>et al.</i> 2010	Go/no-go task		0.38	0	2	2
Menghini <i>et al.</i> 2010	Stroop		0.38	0	1	1
Menghini <i>et al.</i> 2010	OppositeWorld		0.38	0	2	2
Menghini <i>et al.</i> 2010	Bells test		0.38	0	2	2
Menghini <i>et al.</i> 2010	Sky search		0.38	0	2	2
Menghini <i>et al.</i> 2010	Score task		0.38	0	2	2
Menghini <i>et al.</i> 2010	BVN auditory selective attention		0.38	0	2	2
Menghini <i>et al.</i> 2010	Category Fluency Test		0.38	0	2	2
Menghini <i>et al.</i> 2010	Weigl Colour Form Sort Test		0.38	0	2	2
Mento <i>et al.</i> 2019	Top-down and bottom-up temporal attention task	−0.04	0.63	0	2	5
Nader-Grosbois and Lefèvre 2011	Scoring grid for self-regulation joint attention	−0.08	0.60	2	2	7
Nader-Grosbois and Lefèvre 2011	Scoring grid for self-regulation attention	−0.08	0.60	2	2	7
Nader-Grosbois and Vieillevoys 2012	Scoring grid for self-regulation joint attention	0.17	0.72	2	2	7
Nader-Grosbois and Vieillevoys 2012	Scoring grid for self-regulation attention	0.17	0.72	2	2	7
Numminen <i>et al.</i> 2001	Tower of Hanoi	−0.02	0.52	0	1	4
Palmqvist <i>et al.</i> 2020	Planning tasks composite	−0.08	0.88	0	1	5
Palmqvist <i>et al.</i> 2020	Category fluency	−0.11	0.88	0	1	5
Palmqvist <i>et al.</i> 2020	BADS-C Playing cards	−0.16	0.88	0	2	6
Pennington <i>et al.</i> 2003	CANTAB Stockings of Cambridge	0.01	0.58	0	2	5
Pennington <i>et al.</i> 2004	NEPSY Verbal Fluency	0.01	0.58	1	2	6
Pennington <i>et al.</i> 2005	NEPSY Design Fluency	0.01	0.58	1	2	6
Pennington <i>et al.</i> 2006	Stopping task	0.01	0.58	1	2	6
Randolph <i>et al.</i> 2000	Modified forced choice RT task	−0.17	0.46	0	0	2
Roberts <i>et al.</i> 2015	A not B task	−0.04	0.55	0	1	4
Smith <i>et al.</i> 2019	Smith task	−0.03	0.43	0	0	2
Traverso <i>et al.</i> 2018	Preschool matching familiar figure task	−0.14	0.74	2	2	7
Traverso <i>et al.</i> 2018	Go/no-go task	−0.14	0.74	2	1	6
Traverso <i>et al.</i> 2018	Fish flanker task	−0.14	0.74	2	1	6
Traverso <i>et al.</i> 2018	Dots task	−0.14	0.74	2	2	7
Van der Molen <i>et al.</i> 2012	CANTAB ID ED		0.63	0	2	3
Van der Molen <i>et al.</i> 2012	CANTAB ID ED		0.56	0	2	3
Zigler and Unell 1962	Concept formation tasks		0.28	0	2	2
Zigler and Unell 1962	Concept formation tasks		0.28	0	2	2

Note. Matching is the mental age difference (in Cohen's d) between the intellectual disability (ID) group and the mental age (MA) group, Power is the power to detect a directed group difference (ID < MA) of Cohen's $d = 0.5$.

In the Test column, Test battery names are abbreviated: BVN, *Batteria per la Valutazione Neuropsicologica*; CANTAB, Cambridge Neuropsychological Test Automated Battery; D-KEFS, Delis-Kaplan executive function system; WJ, Woodcock-Johnson.

Figure 2. Forest plot of effect sizes (Hedges' g) and 95% confidence intervals for all 99 effects included in the meta-analysis, sorted by ID aetiology, and effect size.

A sensitivity analysis was conducted by investigating Cook's distance, but this did not indicate that one particular study had a large impact on the overall results. The largest Cook's distance

value was 0.05, which is well below the cut-off of 1. For the moderator analyses and the subgroup analyses, the Cook's distance was the highest (0.49) for the ID aetiology subgroup analysis for the FXS

Table 3 Information on the overall model, subgroup models for the four ID aetiologies and moderator analyses

Model	k	Effect size			Heterogeneity I^2		Cook's distance	Test of moderator	
		Hedges' g	95% CI	P	Within studies	Between studies		Q	P
Overall model	99	−0.34	[−0.53, −0.16]	<0.001	49	28	0.05		
Subgroups									
Subgroup NSID	28	−0.22	[−0.46, 0.02]	0.068	8	50	0.09		
Subgroup DS	32	−0.15	[−0.44, 0.14]	0.304	60	19	0.07		
Subgroup WS	33	−0.63	[−0.86, −0.4]	<0.001	68	0	0.12		
Subgroup FXS	6	−0.97	[−1.65, −0.29]	0.014	34	36	0.49		
Moderators									
EF component	99				49	29	0.27	0.31	0.82
ID aetiology	99				52	20	0.22	4.34	0.01
Chronological age	96				49	28	0.10	0.12	0.73
Domain	99				45	32	0.24	1.51	0.23
Mental age	97				48	29	0.46	0.27	0.60
IQ	89				52	25	0.38	2.12	0.15
Quality									
Quality index	99				51	25	0.15	0.76	0.39
Matching	99				52	25	0.26	0.53	0.47
Power	99				50	26	0.12	1.22	0.27
Reliability	99				49	27	0.08	1.08	0.30
Established test	99				49	28	0.23	0.69	0.41

CI, confidence interval; DS, Down Syndrome; EF, executive function; FXS, fragile X syndrome; ID, intellectual disability; NSID, non-specific intellectual disability; WS, Williams syndrome.

Figure 3. Funnel plot of effect size (Hedge's g) by the standard error (SE). Each dot represents one effect.

group in the Van der Molen *et al.* (2012) study. This is very high and has influenced the analysis, but it is not surprising that the Cook's distance was the highest for this study because this is the only FXS effect size

besides the five Hooper *et al.* (2008) effect sizes; refer to Table 3.

The risk of bias assessment results can be found in Table 2. As can be seen, there was a large variation in

quality. The mean quality index was 3.56. The means for the four quality indicators show that established tasks are used to a high degree (1.73), the MA matching between groups was suboptimal (0.94), few studies reported reliability (0.45) and the power was low (0.43). Analyses were made with the quality indicators as moderators, but neither the quality index nor the four quality indicators were significant moderators; refer to Table 3 for details.

Discussion

The present work investigated the current literature on EF in individuals with ID compared to MA-controls. The results can be summarised in four main points: (1) the heterogeneity between studies was large, (2) the ID group performed significantly lower than MA controls, ($g = -0.34$), (3) the moderator variables investigated could only explain a small part of the heterogeneity among studies and (4) publication bias is probably not a major issue in this field.

Do people with intellectual disability have impaired executive function compared with mental age controls?

The overall effect size shows that the ID group performs statistically significantly lower than the MA controls, which was also found for the subgroups WS and FXS (but not DS and NSID). Although we are not aware of another meta-analysis of EF in people with ID without a dual diagnosis, our results are similar to the findings of EF in similar groups. A meta-analysis of EF in people with fetal alcohol spectrum disorder and attention deficit hyperactivity disorder shows a significant effect (Kingdon *et al.* 2016) Hedges' g between -0.52 and -0.94 . A recent meta-analysis by Tungate and Connors (2021) also shows a significant effect, Cohen's $d = -0.87$ of EF in people with DS, but it should be noted here that their definition of EF differs from the one in our study, they included both short-term memory and working memory but excluded attention tasks.

Do executive function component and intellectual disability aetiology moderate the effect?

The second research question investigated whether the EF component and the ID aetiology significantly moderated the effect. The heterogeneity was large, as

indicated by an overall $I^2 = 76\%$, which is of similar magnitude as in the other meta-analyses mentioned (i.e. Tungate and Connors 2021, Kingdon *et al.* 2016 with $I^2 = 80\%$). This indicated that moderator analyses were suitable. The moderator ID aetiology was significant. It is specifically interesting that in the subgroups analysis, only WS and FXS (but not NSID and DS) were significantly different from zero.

This finding speaks for differences in the performance of EF tasks across syndromes. This is a finding that has been observed several times before (Carney *et al.* 2013; Costanzo *et al.* 2013). These syndrome-specific differences refer to EF performance in general, but are often expressed even more fine grained as that certain ID aetiologies have specific strengths or weaknesses in specific EF tasks, compared with MA controls. For example, people with WS seem to perform better on verbal inhibition tasks, compared with people with DS but lower on planning tasks (Costanzo *et al.* 2013). People with DS are considered to have specific strengths in visuospatial tasks but relative weaknesses in shifting and verbal tasks (Costanzo *et al.* 2013). Compared with other groups with ID, individuals with FXS seem to have strengths in visuospatial tasks (Kogan *et al.* 2004). Other studies, such as the meta-analysis by Tungate and Connors (2021), showed a significant effect also for the group of people with DS. Surprisingly, our DS subgroup was non-significant.

Those syndrome-specific findings are highly interesting, but due to the small number of studies in each ID aetiology group within our meta-analysis, inferences have large uncertainty. However, the syndrome-specific differences known from the literature have support from our analyses. The subgroups seem to have specific strengths to some of the EF components included in this meta-analysis, but these analyses are largely underpowered. Moreover, how far these task-specific strengths and weaknesses relate to difficulties in context-specific situations is a different question (Chan *et al.* 2008).

Surprisingly, the EF component was far from significant. From the literature on people without ID, we expected to find differences between EF components (e.g. Miyake *et al.* 2000). However, this is consistent with a meta-analysis on EF in DS (Tungate and Connors 2021), which only found one significant difference between EF components (which was short-term memory vs. inhibition). We hypothesize

that our result (EF component moderator nonsignificant) is due to the large number of different tests used to examine individual EF components.

Our findings (e.g. nonsignificant moderator EF component and subgroups NSID and DS) were potentially biased by our decision to exclude working memory measures. Our initial aim to exclude working memory measures was to avoid it being overrepresented. This could have created an opposite bias, as EF component updating was highly underrepresented in our results. Not having a representable number of updating tasks compared with the other included EF components might have biased the results concerning the moderator EF component and probably also the results on the different subgroups of ID aetiology, as syndrome specific differences for updating are possible. Our a priori decision on the definition of EF components makes it difficult to make a direct comparison with some other reviews such as the meta-analysis by Tungate and Conners (2021). Tungate and Conners (2021) included working memory and short-term memory tasks; however, they excluded attention measures. As mentioned in the rationale, EF can be defined in many ways, and the different definitions are typically vague. It is likely that different findings will be received by using different definitions of EF and therefore including different EF components. It is therefore noteworthy that our choice of the definition of EF, which includes inhibition, shifting, updating and attention, might have influenced the findings. Generalising conclusions regarding the overall EF construct should therefore only be made cautiously.

Mental age and chronological age moderators

None of the exploratory moderator variables chronological age, mental age, IQ and domain had a significant effect on the EF performance. From the literature (e.g. Miyake *et al.* 2001; Willner *et al.* 2010), it is known that there is a relationship between mental age and EF. One methodological challenge with the mental age moderator was that the reporting of mental age and IQ was not consistent. Some studies based their comparison on IQ, others on vocabulary skills and again others on fluid intelligence. Simultaneously, many only reported IQ or mental

age. Therefore, some mental age measures were only inferred by the given IQ score.

Even though the moderator IQ was not significant, it is worth mentioning that this might be due to a non-ideal coding of the variable. What was used in this meta-analysis was the mean IQ for every study. However, by reducing the IQ to the mean, a lot of information about the variable gets lost. It would be helpful to be able to divide the studies into mild, moderate and severe ID. However, as most studies have included participants with varying IQ levels, this distinction could not be applied. Still, we would expect differences between people with mild, moderate and severe ID.

As the experimental groups included for DS and FXS have a large range in mean chronological age (4.34 to 30.3 years), we expected that this could have a significant effect on the EF performance. The fastest development in EF abilities is between 5 and 8 years (Goldstein and Naglieri 2014, p. 9), but EF continues to develop into adulthood. The literature shows that the performance on EF tasks depends on the developmental stage in life (Luria 1980). In this meta-analysis, different steps in the development of EF were covered. Besides, it is known that the IQ of people with DS and FXS tends to decrease between early childhood and young adulthood. It is possible that the development in EF skills was at different levels.

Even though we did not find a significant effect for mental and chronological age, these different complex factors might have added to the heterogeneity of the results. Possibly, there is an underlying structure, which could not be uncovered by our analysis. A more detailed comparison should be made in future studies, investigating EF in ID longitudinally.

Limitations and bias

One concern when measuring EF is the close link to general cognitive abilities. As EF abilities are assumed to be significantly influenced by the level of intelligence, EF test results are difficult to interpret towards one specific EF component (Mahone *et al.* 2002). Many intelligence batteries measure EF, but often not in a detailed way. Therefore, a flexible battery approach is needed to test detailed hypotheses about EF (Goldstein and Naglieri 2014, p. 379). Most of the papers that were included in the present study

used a flexible battery approach. This led to further variability in the testing between studies. However, only a few studies included tests of all EF components. Some only examined one EF component, others had several measures per EF component. This can have a bias on the within-studies variance. Another limitation of our study is that we only included laboratory-based measures of EF, opposed to more ecological measures, such as the BRIEF (Gioia *et al.* 2000). This choice was a functional one, to keep the individual measures as comparable as possible. Still, it comes with a limitation concerning the generalisability to real-world behaviour (Chan *et al.* 2008), as ecological validity has been criticised for neuropsychological tests (Goldstein 1996).

In consideration of these methodological issues and their influence on the derived effect sizes, studies using standardised EF tasks and large sample sizes may produce more accurate data on the EF performance of individuals with ID. As there is no universally accepted definition of EF, there is also no clear agreement on which specific tasks measure which EF component. In conclusion, research on EF is diverse and often difficult to compare. There are many different EF tasks, which lead to low ecological validity (Ardila 2008). This was also the case here, as tests belonging to several different EF tasks were included. There were 69 different EF tests used to assess the 99 effect sizes included in the meta-analysis. Variations of Tower tests were the most frequently used test, but it was only used for six effect sizes. However, even though Tower tests are similar, they are not identical. Additionally, different measures were reported, for example, seconds to complete, the total number of errors made, levels achieved or the total number of correct responses. Using the standardised mean difference partly adjust for that, but the risk for a task-dependency effect (Welsh 2002) is still high.

The risk of bias assessment identified four studies (Nader-Grosbois and Lefèvre 2011; Nader-Grosbois and Vieillevoys 2012; Carney *et al.* 2013; Traverso *et al.* 2018) with high quality (score 7 of 8), but some studies only had a score of 1. The studies had on average very low power. The median number of participants in the ID group was 21, which gives 30% power to detect the average effect size ($g = -0.34$) found in this meta-analysis. This is very low power

and much lower than the rule of thumb of at least 80%.

Another quality issue is the MA matching between the groups. On average, the groups had a difference of Cohen's $d = 0.24$, which is of similar magnitude as the effect size found in the meta-analysis. When the groups differ that much in mental age and the studies have low power, it is not surprising that there is a large unexplained variance in this meta-analysis.

None of the publication bias tests indicated that publication bias was present. Studies on cognitive abilities in people with ID are generally publishable even if there are no group differences compared to MA controls for three different reasons. First, both difference and no difference results are interesting for this group because little is known about which cognitive functions are comparably intact and which are delayed. Second, often a control group matched on chronological age is included, and there is a difference between their performance and the ID groups' performance. Third, often many measures are taken in the same study. Typically, group differences vary in magnitude between tests. Efforts to get unpublished results were performed, but only one unpublished study was included, which has since been published (Palmqvist *et al.* 2020). Given this, it is not surprising that there were no strong signs of publication bias in this field.

Conclusions

Due to the extremely large heterogeneity, conclusions must be treated warily. This meta-analysis provides further evidence that people with ID perform below their mental age expectation on EF tasks. The heterogeneity in the data was large, which speaks for other moderating effects. When looking at the subgroups, there was only a significant effect for WS and FXS. The high variance levels indicate that research about EF is very heterogeneous. Several methodological issues could cause this. The typical study has a power of only 30% to detect the average effect size found in this meta-analysis. It is also remarkable that 69 different tests were used to assess the 99 effect sizes included in this meta-analysis. Our definition of EF, including inhibition, shifting, updating and attention might have influenced the findings. Generalising conclusions regarding the overall EF construct should therefore only be made

cautiously. To further advance the knowledge in this field, future studies should have higher power, be better matched on MA and to a higher degree use a smaller set of EF tests so that the same underlying construct is measured. The deepening of knowledge about EF in ID can help researchers, clinicians, caregivers and parents to improve interventions and help people with ID. The overall aim is to improve the quality of life of individuals with ID and their families.

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Conflict of interest

The authors have no conflicts of interest to report.

Data Availability Statement

The data that support the findings of this study are openly available in an Open Science Framework project at <http://doi.org/10.17605/OSF.IO/NJSD3>.

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A: Appendix

PRISMA 2009 Checklist

For the process of the meta-analysis, this checklist was used as a guideline. The checklist contains information about what information is included in this document and where it can be found (Liberati et al. 2009).^{1, 2}

Section/topic	#	Checklist item	Reported on page #
Title			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	1
Abstract			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	1–2
Introduction			
Rationale	3	Describe the rationale for the review in the context of what is already known.	3–5
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	5–6
Methods			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g. Web address), and, if available, provide registration information including registration number.	6
Eligibility criteria	6	Specify study characteristics (e.g. PICOS and length of follow-up) and report characteristics (e.g. years considered, language and publication status) used as criteria for eligibility, giving rationale.	7–8
Information sources	7	Describe all information sources (e.g. databases with dates of coverage and contact with study authors to identify additional studies) in the search and date last searched.	6–7
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	appendix A2
Study selection	9	State the process for selecting studies (i.e. screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	6–7
Data collection process	10	Describe method of data extraction from reports (e.g. piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	8
Data items	11		8

¹ Source: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi: 10.1371/journal.pmed1000097

² For more information, visit online (www.prisma-statement.org).

Section/topic	#	Checklist item	Reported on page #
		List and define all variables for which data were sought (e.g. PICOS and funding sources) and any assumptions and simplifications made.	
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was performed at the study or outcome level), and how this information is to be used in any data synthesis.	8–9
Summary measures	13	State the principal summary measures (e.g. risk ratio and difference in means).	10
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if performed, including measures of consistency (e.g. I^2) for each meta-analysis.	9–10
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g. publication bias and selective reporting within studies).	9–10
Additional analyses	16	Describe methods of additional analyses (e.g. sensitivity or subgroup analyses and meta-regression), if performed, indicating which were pre-specified.	10
Results			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	Fig. 1,6–7
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g. study size, PICOS and follow-up period) and provide the citations.	Tab. 1, References
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (refer to item 12).	12–13
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	Fig. 2, tab. 1
Synthesis of results	21	Present results of each meta-analysis performed, including confidence intervals and measures of consistency.	Fig. 2, tab. 1, 12–14
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (refer to item 15).	Tab. 2, 13
Additional analysis	23	Give results of additional analyses, if performed [e.g. sensitivity or subgroup analyses, meta-regression (refer to item 16)].	Tab 3,12–13
Discussion			
Summary of evidence	24	Summarise the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g. healthcare providers, users and policy makers).	13–15
Limitations	25	Discuss limitations at study and outcome level (e.g. risk of bias), and at review-level (e.g. incomplete retrieval of identified research and reporting bias).	16–17
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	18
Funding			
Funding	27	Describe sources of funding for the systematic review and other support (e.g. supply of data); role of funders for the systematic review.	Author details.docx

Full electronic search strategy

For the PRISMA checklist (8) 'Search', a full electronic search should be given. The additional files can be found online (<http://doi.org/10.17605/OSF.IO/NJSD3>)

Pubmed 2019-08-21

('intellectual disability'[TIAB] OR 'intellectual disabilities'[TIAB] OR 'mental retardation'[TIAB] OR 'down syndrome'[TIAB] OR 'williams syndrome'[TIAB] OR 'fragile x'[TIAB] OR 'intellectual disability'[MeSH Terms]) AND ('executive function'[TIAB] OR 'executive functions'[TIAB] OR 'executive functioning'[TIAB] OR 'executive dysfunction'[TIAB] OR 'shifting'[TIAB] OR 'switching'[TIAB] OR 'updating'[TIAB] OR 'inhibition'[TIAB] OR 'cognitive flexibility'[TIAB] OR 'cognitive control'[TIAB] OR 'executive control'[TIAB] OR 'executive controls'[TIAB] OR 'executive function'[MeSH Terms]) AND english[Language] AND Journal Article[ptyp]

1694 records

Saved in nine files called citation1-200.nbib to citation1601-1694.nbib

Scopus 2019-08-21

(TITLE-ABS-KEY ('intellectual disability') OR TITLE-ABS-KEY ('intellectual disabilities') OR TITLE-ABS-KEY ('mental retardation') OR TITLE-ABS-KEY ('down syndrome') OR TITLE-ABS-KEY ('williams syndrome') OR TITLE-ABS-KEY ('fragile x')) AND (TITLE-ABS-KEY ('executive function') OR TITLE-ABS-KEY ('executive functions') OR TITLE-ABS-KEY ('executive functioning') OR TITLE-ABS-KEY ('executive dysfunction') OR TITLE-ABS-KEY ('shifting') OR TITLE-ABS-KEY ('switching') OR TITLE-ABS-KEY ('updating') OR TITLE-ABS-KEY ('inhibition') OR TITLE-ABS-KEY ('cognitive flexibility') OR TITLE-ABS-KEY ('cognitive control') OR TITLE-ABS-KEY ('executive control') OR TITLE-ABS-KEY ('executive controls')) AND (LIMIT-TO (DOCTYPE, 'ar')) AND (LIMIT-TO (LANGUAGE, 'English'))

1870 records

Saved in one file called scopus1-1870.bib. This file only contains 1869 records, why a separate file was

created for the missing file (maehler *et al*) called scopus_maehler.bib

Psycinfo 2019-08-21

(TX 'intellectual disability' OR TX 'intellectual disabilities' OR TX 'mental retardation' OR TX 'down syndrome' OR TX 'williams syndrome' OR TX 'fragile x' OR SU 'Intellectual Development Disorder') AND (TX 'executive function' OR TX 'executive functions' OR TX 'executive functioning' OR TX 'executive dysfunction' OR TX 'shifting' OR TX 'switching' OR TX 'updating' OR TX 'inhibition' OR TX 'cognitive flexibility' OR TX 'cognitive control' OR TX 'executive control' OR TX 'executive controls' OR SU 'executive function') AND LA english AND PT Peer Reviewed Journal

1077 records

Saved in 1 file called psycinfo1-1077.bib

Web of Science core collection 2019-08-21

((TS = ('intellectual disability')

ORTS = ('intellectual disabilities') ORTS = ('mental retardation') ORTS = ('down syndrome')

ORTS = ('williams syndrome') ORTS = ('fragile x'))

AND (TS = ('executive function')

ORTS = ('executive functions') ORTS = ('executive functioning')

ORTS = ('executive dysfunction')

ORTS = ('shifting') ORTS = ('switching')

ORTS = ('updating') ORTS = ('inhibition')

ORTS = ('cognitive flexibility') ORTS = ('cognitive control')

ORTS = ('executive control')

ORTS = ('executive controls'))

Plus the limitors LANGUAGE: (English) AND DOCUMENT TYPES: (Article)

1941 records

Saved in four files named wos1-501.bib to wos1501-1941

Accepted 11 August 2021

Supporting Information

Additional Supporting Information may be found online in the supporting information tab for this article.

Data S1: Full text screening with reasons for exclusion.