Meta-Analysis



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Associated With Reading in Intellectual

Disability: A Systematic Review and

Cognitive and Language Abilities

Abstract

Cognitive and language abilities influence literacy outcomes in individuals with intellectual disability (ID). The aim of this study was to investigate abilities associated with decoding and reading comprehension in individuals with ID with a systematic review and a correlational meta-analysis. A total of 26 studies with 27 samples and 1,137 participants were included in the meta-analysis. The results showed that reading comprehension was significantly related to decoding (r = .63), vocabulary (r = .51), listening comprehension (r = .43), and IQ (r = .32). Decoding was significantly related to phonological awareness (r = .52), phonological short-term memory (r = .46), rapid automatized naming (r = -.44), vocabulary (r = .34), chronological age (r = .26), IQ (r = .30), visual short-term memory (r = .24), and executive-loaded working memory (r = .32). Limitations connected to unexplained heterogeneity and study quality were found and discussed. This meta-analysis showed that variables identified in studies of typically developing children are relevant for individuals with ID indicating that theories about literacy could reasonably be applied to this population.

Keywords

reading ability, decoding, reading comprehension, intellectual disability

Being able to read is important for a person's independence and participation in society. Many individuals with intellectual disability (ID) struggle with both reading comprehension and decoding (Afacan & Wilkerson, 2022; Lemons et al., 2013; Ratz & Lenhard, 2013; Wei et al., 2011). ID is a developmental condition that most recently has been defined by limitations in intellectual functioning (often measured by an IQ test), and adaptive behavior in the form of social, conceptual, and practical skills (American Psychiatric Association [APA], 2022; Schalock et al., 2021). Historically, research on individuals with ID has been sparse (Bishop, 2010), but there appears to be a growing interest in studying the academic skills of students with ID (Cannella-Malone et al., 2021). Despite this interest, the underlying factors related to reading difficulties in individuals with ID remain unclear. This contrasts with the considerable progress that has been made in the understanding of reading disabilities such as dyslexia (Snowling et al., 2020), and reading comprehension difficulties (e.g., Elwér et al., 2013; Groen et al., 2019; Kelso et al., 2022). To systematically review the available evidence about reading in individuals with ID is an important first step in increasing our understanding of this process.

Our systematic review and meta-analysis investigates which underlying factors are associated with reading comprehension and decoding ability in individuals with ID. These two abilities were chosen because of their widespread recognition as primary objectives of reading instruction and are the most common ways of assessing reading ability. Numerous variables are hypothesized to influence the progression of reading comprehension and decoding, and these vary depending on which theoretical framework is adopted. A brief overview of reading comprehension and decoding is provided to identify relevant variables for the meta-analysis and to show how discrepancies between findings provide a rationale for a systematic review.

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Reading Comprehension

Reading comprehension research in typically developing children has generated two important theoretical positions about underlying mechanisms and predictor variables. One is the Simple View of Reading (SVR, Gough & Tunmer, 1986), where reading comprehension is explained as the product of decoding and listening comprehension. There are several definitions of decoding, Gough and Tunmer (1986) defined it as the ability to read isolated words quickly, accurately, and silently. They chose not to use the term word recognition, to emphasize that decoding denotes the use of letter-sound correspondence. We adopt this definition, with the modification that words being decoded should be read aloud (to ensure an accurate assessment). Many studies have found support for this framework, both in typically developing individuals (e.g., Lervåg et al., 2018) and in individuals with reading comprehension difficulties (Catts et al., 2006).

Other research work takes a broader perspective and suggests that additional abilities might be of importance when explaining and predicting reading comprehension. Vocabulary has been shown to play a significant role in explaining the variance in reading comprehension, over and above decoding and listening comprehension (Braze et al., 2007; Ouellette & Beers, 2010). These findings support another theoretical framework, namely the Lexical Quality Hypothesis (LQH, Perfetti, 2007), where the quality of lexical representations and vocabulary size are assumed to affect reading comprehension directly. A large, Dutch longitudinal study found support for both the SVR and the LQH (Verhoeven & van Leeuwe, 2008); decoding, vocabulary, and listening comprehension all had substantial relationships with children's reading comprehension.

In addition to decoding, listening comprehension, and vocabulary, there are other predictors that often emerge as important for reading comprehension in groups without ID, such as IQ (Hulslander et al., 2010), grammatical skills (Muter et al., 2004), verbal working memory (e.g., Cain et al., 2004; Seigneuric & Ehrlich, 2005; Swanson & Howell, 2001), and visuospatial working memory (VSWM) (Bayliss et al., 2005; Pham & Hasson, 2014).

Investigations of the SVR and LQH in individuals with ID are sparse, but some studies have found that the components of the SVR play a crucial role in explaining reading comprehension in populations with ID (Roch & Levorato, 2009; van Wingerden et al., 2014, 2017; Verhoeven & Vermeer, 2006). For example, Mervis et al. (2022) found that decoding and listening comprehension accounted for 79% of the variance in reading comprehension in a relatively large sample of English-speaking 9-year-old children with ID, who also had Williams syndrome (WS). In addition, other variables have been shown to explain a significant amount of variance in reading comprehension, such as

nonverbal IQ in Dutch-speaking children and second-language learners with ID (van Wingerden et al., 2017, 2018; Verhoeven & Vermeer, 2006), vocabulary and sentence comprehension in Italian-speaking children with DS (Levorato et al., 2009), and phonological awareness and letter-sound knowledge in French-, German-, and Dutch-speaking children with ID (Sermier Dessemontet & de Chambrier, 2015; van Wingerden et al., 2017, 2018).

Decoding

It appears that for individuals with typical reading development and with dyslexia, the most important underlying factors in explaining the variance in decoding ability are phonological awareness, rapid automatized naming (RAN), and letter-sound knowledge (Melby-Lervåg et al., 2012; Moll et al., 2014; Schatschneider et al., 2004; Torgesen et al., 1997). However, studies also have suggested that other abilities could be of relevance for decoding ability, such as phonological short-term memory (STM, Moll et al., 2014), vocabulary (Ouellette, 2006), visual STM (Kibby et al., 2015; Kulp et al., 2002), and working memory (Christopher et al., 2012).

Many studies of individuals with ID report similar results as studies on typically developing children, namely that phonological awareness, RAN, and letter-sound knowledge correlate strongly with decoding skills (Barker et al., 2014; Kennedy & Flynn, 2003; Mervis et al., 2022; Pezzino et al., 2021; Saunders & DeFulio, 2007; Sermier Dessemontet & de Chambrier, 2015; Soltani & Roslan, 2013). Some studies have also emphasized the importance of phonological memory in explaining decoding ability in individuals with mixed etiology ID (Channell et al., 2013; Conners et al., 2001), Down syndrome (DS, Byrne et al., 2002), and nonspecific ID (Henry & Winfield, 2010). In addition, studies report that vocabulary and grammatical comprehension are significantly related to decoding ability, especially in participants with DS (Boudreau, 2002; Byrne et al., 2002; Cardoso-Martins et al., 2009; Naess et al., 2011), whereas for individuals with WS, there seems to be a correlation between decoding and visuospatial abilities (Mervis et al., 2022). Regarding the association between decoding ability and IQ, the literature contains different findings. Some studies point toward a strong relationship between decoding and IQ (Levy, 2011; van Tilborg et al., 2014), whereas others fail to find this association (Boudreau, 2002; Conners et al., 2001).

Thus, for individuals with ID, several variables have been identified as predictors of decoding abilities. Some correspond to those often identified in typically developing groups, that is, phonological awareness, RAN, and letter-sound knowledge; in addition, other variables have sometimes been identified, that is, IQ, vocabulary, grammatical comprehension, and phonological memory. This range of variables makes it important to systematically review the literature.

The Current Study

As has been outlined, investigations of individuals with ID have identified a number of variables associated with both decoding and reading comprehension. However, because studies often produce contradictory results, and it is common for studies to involve different sets of variables, there is a need for a systematic review of reading research focusing on these individuals. Furthermore, a review and analysis can help to identify whether or not decoding and reading comprehension show a similar or different pattern of findings to students with typical development. A similar pattern suggests that future research might evaluate the forms of reading instruction used with typical students in relation to those with ID, different patterns would suggest a need to develop new forms of reading instruction adapted to the specific needs of individuals with ID. A systematic review and a meta-analysis were conducted targeting the variables that have been shown to have significant relationships with decoding or reading comprehension in earlier studies of individuals with ID. One aim was to identify cognitive and language abilities significantly associated with decoding and reading comprehension in individuals with ID. A second aim was to address two key questions: (a) whether or not these associations were similar to those in typically developing groups and (b) whether or not the associations aligned with the principles of the SVR and the LQH.

Method

Search Strategy

We searched four databases, PubMed, PsycInfo, Web of Science, and ERIC (Educational Resources Information Center). The search strategy included a combination of keywords related to reading (reading, literacy, decoding, word recognition), ID (ID, mental retardation, mental deficiency, intellectual developmental disorder, developmental disability), and relation (relation, relationship, prediction, correlation, regression, association). A full description of the keywords for each database is in the online supplements. No filters were used during the search. We performed several searches to ensure that articles published or made available since the first search were found and included in the final meta-analysis. The first search was conducted in August 2017, the second search in March 2021, and the third search in August 2022. The first search yielded 1,677 articles (1,333 after removing duplicates). For the subsequent searches, the same search strategy was adopted. One database had changed the interface of its advanced search; consequently, a minor change was made to the search strategy (see online supplements). The second and third searches yielded 3,040 and 3,457 articles, respectively (1,202 and 387 articles,

respectively, after removing duplicates within the searches and duplicates of those found in the previous searches).

In addition to the systematic search, other methods were used to find additional records. The first author sent inquiries for file drawer data to all 23 authors of already included articles who had valid email contact information in the publications, and 10 replied. Furthermore, the first author performed the following additional search methods: reference lists in all the included articles were scanned for suitable articles, articles citing the included articles were identified via Google Scholar and screened (completed in May 2021), and a request for file drawer data was made in a poster presentation at the 2019 Society for Scientific Studies of Reading conference.

Inclusion and Exclusion Criteria

For the abstract and full-text screening, the inclusion criteria were the presence of: (a) measures of decoding and/or reading comprehension; (b) correlational data; (c) a sample with a mean IQ at or below 70, and a maximum (i.e., range) IQ of 85 (any standardized assessment of cognitive ability was approved. Whenever multiple measurements were reported, the order of preference was non-verbal, full scale, and verbal); (d) a minimum sample size of 10 participants; and (e) participants with nonspecific ID (unknown etiology), Down syndrome, WS, or mixed etiology (i.e., participants with various aetiologies within the same sample). The most recent and current definitions of ID give much more emphasis to adaptive abilities (APA, 2022); however, the most usual way to operationalize ID in research has been through a measure of IQ or related cognitive ability, as a result, our inclusion criteria reflected this approach. The meta-analysis included articles in dissertations, and all articles had to be written in English. Articles were excluded if the focus was another syndrome, another disability, if the participants also had autism, or if the article was a review. Data collection and reporting were guided by the directions of Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA, Page et al., 2021). For a more detailed description of identified records, of included and excluded records, and the reasons for exclusion, see Figure 1. The PRISMA checklist and an abstract that adheres to the PRISMA guidelines are in the online supplements.

Screening

Two of the authors screened the abstracts and full-text articles independently. After initial abstract screening, the following number of articles remained for full-text screening after each search: first search (k=166), second search (k=199), and third search (k=34). Inter-rater reliability figures were calculated for the abstract screening in the second and third search using Cohen's κ , and the agreement ranged between 0.56 and

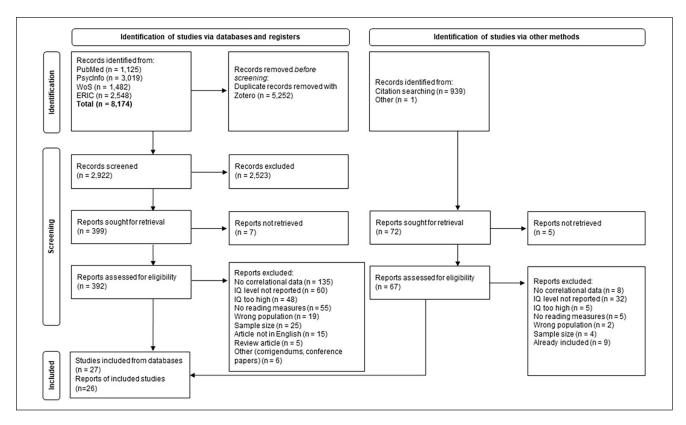


Figure 1. PRISMA Flowchart Showing Number of Identified Records, Number of Included and Excluded Records, and the Reasons for Exclusion.

0.70 which is considered as fair to good agreement according to Fleiss et al. (2003). The percentage agreement ranged between 91% and 96%. The two authors marked each study as "include," "exclude" or "unsure," and only the studies marked with "exclude" from both authors were excluded before the next step. Studies marked as "unsure" and any studies where there was a difference between the two authors were discussed and resolved. The screening process was conducted in Zotero (first search) and Excel (second and third search). Through the systematic searches and the screening processes, we identified 24 articles that met our inclusion criteria. In addition, the inquiries for file drawer data and forward and backward citation searches made by the first author yielded one additional article plus one additional data set, which was later published. Hence, a total of 26 articles were identified and included in the meta-analysis. In addition, the study by Levy (2011) involves two independent samples, one with DS and one with nonspecific ID. These samples are reported separately in the primary study and thus treated as two separate studies in this metaanalysis, meaning that the final set consists of 27 studies.

Coding

Two of the authors coded the descriptive data and the correlations. Agreement between coders was 93% for the

descriptive data and 96% for the correlations. All disagreements were discussed and resolved. The articles in this meta-analysis were mainly cross-sectional studies. There also were some relevant longitudinal studies (k = 3) and for these, the pre-test data was used. Our meta-analysis targeted variables that have been found to correlate with reading comprehension and decoding in research on individuals with ID and with typical development. Hence, we coded all pairwise correlations between the following variables: reading comprehension, decoding, phonological awareness, listening comprehension, vocabulary, phonological STM, visual STM, executive-loaded working memory (ELWM), RAN, IO, chronological age, letter-sound knowledge, and grammatical comprehension. The mean age reported in the primary studies was coded in months, and when age was reported in years in the primary study it was converted to months by the coding author.

Quality Assessment

The quality of the included articles was assessed with the AXIS tool (Downes et al., 2016), which was created by an expert panel for the purpose of critically appraising cross-sectional studies. In addition, the tool also provides a user-friendly guidance document. Note that the AXIS tool was

used to critically appraise the pre-test data from three longitudinal studies that we coded and analyzed; it was not used to assess the methodological quality of the longitudinal designs as only the pre-test data was of interest for our meta-analysis. The AXIS tool consists of 20 items divided into sections that correspond with the outline of a scientific article (one item in the method section is "Was the sample size justified?"). Each item was scored with yes, no, or unsure. The unsure option was used when there was not enough information provided in the primary article to make a decision. The quality assessment did not aim at excluding studies from the meta-analysis, but rather at providing information that could aid in rating the certainty of evidence and the interpretation of the results. Two of the authors conducted the quality assessment independently. Assessments were compared, and any differences were discussed and resolved.

Data Analysis

The number of relevant variables varied across the primary studies. Hence, some of the coded correlations were reported in many of the primary studies, whereas some correlations only occurred in one or two primary studies. To minimize the risk of bias where the result is driven by a few primary studies, we decided to only analyze correlations between variables that were reported in five or more studies. As a result, our meta-analysis included correlations between decoding and these variables: phonological awareness, listening comprehension, vocabulary, phonological STM, visual STM, ELWM, RAN, IQ, and chronological age and between reading comprehension and these variables: decoding, listening comprehension, vocabulary, and IQ.

All analyses were made using R (R Core Team, 2017), and the following R packages: metafor (Viechtbauer, 2010), tidyverse (Wickham, 2017), readxl (Wickham & Bryan, 2019), knitr (Xie, 2015), dplyr (Wickham et al., 2019), and robvis (McGuinness, 2019). The manuscript was formatted using papaja (Aust & Barth, 2017), and citr (Aust, 2016). The effect size of interest in this meta-analysis was the correlation coefficient r. Effect sizes were calculated using the escalc() function in the metafor package based on the studies' reported correlation coefficients and sample sizes. As recommended in the metafor package, the correlations were transformed to z-scores using Fisher's r-to-z transformation to reduce bias, and these scores were then used in the analyses. In a meta-analysis, a fixed-effects model or randomeffects model could be used. A fixed-effects model assumes that there is one underlying effect that is the same for all studies. A random-effects model assumes that the effect could be different in each study, depending on, for example, participant characteristics (different mean IQ in different studies) or design of the studies. For the current meta-analysis, it was reasonable to assume that the underlying effect could be different across studies and, therefore, we used a random-effects model for the analyses. In some cases, primary studies used multiple measures for the same variable (e.g., subtests of phonological awareness such as blending and elision). These effect size estimates could not be assumed to be independent and to deal with the dependencies we used a robust variance estimation (Pustejovsky & Tipton, 2022) for the affected correlations. The random-effects model was fitted on the *z*-transformed data, and the effect sizes were then back-transformed to correlations for easier interpretation of results.

To test the degree of heterogeneity between studies we used three different heterogeneity measures. Cochran's O-test was conducted to examine whether heterogeneity was different from zero. As this test is sensitive to the number of studies included, the I^2 -statistic was used to determine the proportion of observed variance reflecting true variation between effect sizes (Higgins et al., 2003). We used the categorization proposed by Higgins et al. (2003), namely that I^2 values of 25%, 50%, and 75% are regarded as low, moderate, and high, respectively. Finally, the amount of betweenstudy heterogeneity (τ^2) was estimated using the restricted maximum-likelihood estimator (Viechtbauer, 2005). We used the same rule as implemented by Hjetland et al. (2020), namely that a τ^2 larger than 0.1 indicates a large variation between studies. To assess whether study quality could explain heterogeneity in the results, we calculated an overall study quality indicator for each study. We did this by assigning different scores to the three risk of bias levels, where low risk of bias gave 2 points, some concerns (or not reported) gave 1 point, and high risk of bias gave 0 points. Study quality was used as a moderator in the analyses that included at least 10 primary studies, because performing moderator analyses on smaller samples can introduce other problems (Higgins et al., 2019). Publication bias was tested using two methods, namely funnel plots and the Rank Correlation test examining Kendall's τ. Publication bias is indicated through asymmetrical funnel plots and a significant Rank Correlation test. We used Cook's distances to identify studies that were likely to be influential in the context of the model (Viechtbauer & Cheung, 2010). Studies with Cook's distance larger than the median plus six times the interquartile range of Cook's distances were considered to be influential.

The Grading of Recommendations Assessment, Development, and Evaluation (GRADE) tool (Balshem et al., 2011) is a valuable assessment of the certainty of intervention effects; because our meta-analysis concerns correlations, we used a modified version of GRADE to obtain measures of certainty (see Yousefifard & Shafiee, 2023 for a discussion on this topic). This involved four different indicators: (a) an overall assessment of risk of bias, (b) inconsistency of results (a confidence interval [CI] larger than 0.3), (c) imprecision of results (an *I*² larger than 50%), and (d) a significant publication bias test.

Table 1. Average Correlations, Heterogeneity Measures, and Publication Bias Measures for the Outcome Variables Decoding and
Reading Comprehension, and Their Associated Variables.

	n	Effect sizes					Heterogeneity				Publication bias	
Variables		k	r	p(r)	CI lower	CI upper	Q	p (Q)	I ²	τ2	Kendall's τ	þ (Kendall's τ)
Reading compreher	sion											
Decoding	11	10	0.63	0.00	0.37	0.80	157.11	0.00	91.88	0.23	0.00	1.00
Listening comprehension	8	8	0.43	0.00	0.33	0.52	5.42	0.61	4.09	0.00	0.50	0.11
IQ	9	9	0.32	0.00	0.13	0.48	24.37	0.00	69.84	0.05	-0.06	0.92
Vocabulary	8	8	0.51	0.00	0.31	0.66	21.84	0.00	77.52	0.08	0.55	0.06
Decoding												
Phonological awareness	29	10	0.52	0.00	0.43	0.60	52.69	0.00	45.96	0.02	-0.08	0.54
Phonological STM	19	9	0.46	0.00	0.40	0.52	15.95	0.60	0.02	0.00	-0.29	0.09
RAN	13	6	-0.44	0.00	-0.56	-0.30	30.04	0.00	57.23	0.02	0.13	0.57
IQ	21	18	0.30	0.00	0.17	0.42	38.51	0.01	50.12	0.03	0.15	0.35
Chronological age	8	7	0.26	0.03	0.03	0.46	12.90	0.07	52.38	0.03	-0.04	0.90
Vocabulary	15	12	0.34	0.00	0.19	0.48	28.24	0.01	56.29	0.03	0.23	0.23
Listening comprehension	9	8	0.12	0.12	-0.04	0.27	13.46	0.10	42.44	0.02	0.08	0.75
Visual STM	10	5	0.24	0.01	0.11	0.37	10.33	0.32	25.50	0.01	-0.02	0.93
ELWM	10	5	0.32	0.01	0.11	0.50	22.92	0.01	60.23	0.03	0.07	0.78

Note. n = number of effect sizes, k = number of studies.

Results

In this section, we outline the findings from our analyses about the variables associated with reading comprehension and decoding, and the quality assessment of the data. In the discussion, we summarize these findings to address our research questions. Descriptive information about the primary studies can be found in the Supplements, Table S1. The included studies were published between 1960 and 2021. There were 1,137 participants from 27 different samples included in the meta-analysis. The primary studies focused on ID with different etiology, namely nonspecific/ unknown etiology (k = 5, n = 176), Down syndrome (k = 176) 10, n = 169), and WS (k = 2, n = 420). Some studies included participants with mixed etiology (k = 3, n = 32), and several studies did not specify the etiology of the ID (k = 7, n = 340). The mean chronological age of the participants was 196 (range = 92-594) months (M in years = 16, range in years = 8-50). The mean IQ of the participants in 25 out of 27 samples was 56.52 (range of means in primary studies 42-68.80). Two studies were excluded from the calculation of the mean. First, the mean and maximum IQ scores from the study by Nash and Heath (2011) are t-scores from BAS-II (M = 22.77, max = 37), where a score of 30 is equivalent to a standard IO score of 70 and a score of 40 is equivalent to a standard IQ score of 85. Second, the study by Gupta (1985) did not report an exact mean IQ score but reported that the mean IQ was below 70. Average effect sizes and heterogeneity measures for both outcome variables and their associated variables are provided in Table 1. For information about excluded studies, see Supplements.

Reading Comprehension

Reading comprehension correlated significantly with all four variables, with strong correlations for decoding (r = .63, p = <.001) and vocabulary (r = .51, p = <.001), and moderate correlations for listening comprehension (r = .43, p = <.001) and IQ (r = .32, p = .001).

The heterogeneity measures found in Table 1 indicate a high proportion of true heterogeneity between studies for decoding and vocabulary. The proportion of true heterogeneity between studies for listening comprehension was low, and for IQ it was moderate. A moderator analysis was performed on the association between reading comprehension and decoding (k=10) using study quality as a moderator, and it was not significant (p=.437). The average effect sizes for each variable associated with reading comprehension are visualized in Figure 2. Forest plots visualizing all effect sizes from the primary studies can be found in Figures S1–S4 in the Supplements.

The rank correlation test was nonsignificant for all variables associated with reading comprehension (see Table 1),

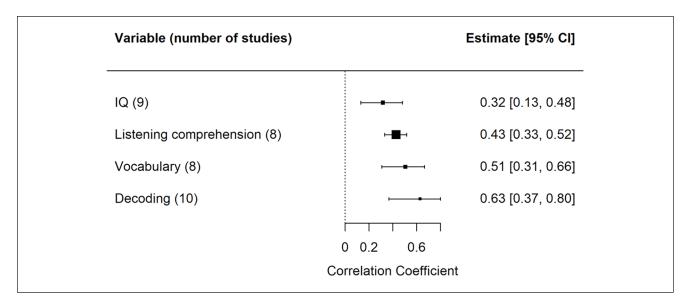


Figure 2. The Average Effect Sizes for Reading Comprehension and Its Associated Variables.

indicating that no publication bias was present. However, a visual inspection of the funnel plots revealed a slightly asymmetrical distribution for both listening comprehension and vocabulary. The funnel plots can be found in Figure S14 in the Supplements. For results from sensitivity analyses, please see Supplements.

Decoding

Decoding correlated significantly with all the chosen variables, except for listening comprehension (r=.12, p=.119). Decoding strongly correlated with phonological awareness (r=.52, p=<.001). Furthermore, moderate correlations were found with phonological STM (r=.46, p=<.001), vocabulary (r=.34, p=<.001), RAN (r=-.44, p=<.001), IQ (r=.30, p=<.001), and ELWM (r=.32, p=.014). Finally, correlations in the lower range were found with chronological age (r=.26, p=.035) and visual STM (r=.24, p=.007).

The heterogeneity measures in Table 1 indicate low true heterogeneity between studies for phonological STM, listening comprehension, and visual STM. A moderate proportion of true heterogeneity between studies was found for phonological awareness, RAN, IQ, chronological age, vocabulary, and ELWM. A moderator analysis was performed on the association between decoding and the following variables: phonological awareness (k = 10), IQ (k = 18), and vocabulary (k = 12) using study quality as a moderator. Study quality was a significant moderator for the association with phonological awareness accounting for 47.35% of the heterogeneity (p = .010), but not for the association with IQ (p = .560), or vocabulary (p = .522).

The average effect sizes for each variable associated with decoding are visualized in Figure 3. Forest plots visualizing all effect sizes from the primary studies can be found in Figures S5–S13 in the Supplements. Note that the most common way of measuring RAN is for the participant to name items as fast as possible. Consequently, the lower the score the better the performance, and a negative correlation is expected with literacy measures. RAN was reported in six primary studies, and three of them originally reported negative correlations. Two of the studies used a "per minute" score, and hence the correlations were converted to negative scores. One study reported a positive correlation, despite using the common test procedure, and this correlation was not converted.

The Rank Correlation test was non-significant for all variables associated with decoding (see Table 1), indicating that no publication bias was present. A visual inspection of funnel plots revealed quite symmetrical distributions. The funnel plots can be found in Figures S15–S16 in the Supplements. The funnel plot of RAN in Supplemental Figure S16 shows a wide distribution along the x-axis, which is more indicative of heterogeneity between studies than publication bias. For results from sensitivity analyses, please see Supplements.

Quality Assessment

The primary studies all showed similar patterns in the quality assessment. Studies generally described clear aims (k = 20), and used a design that was appropriate for their aims (k = 20). All studies except one failed to justify their sample size (k = 25) and the vast majority did not report about the use of a sampling frame (k = 25), meaning that they also

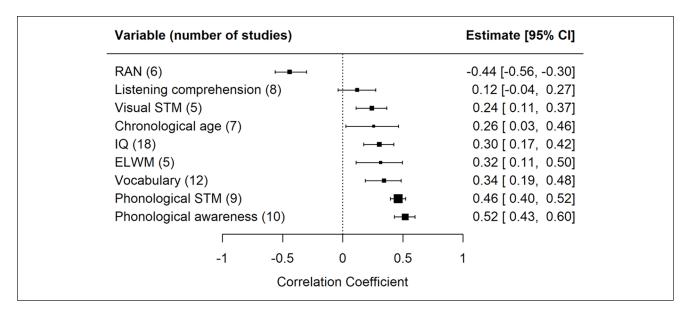


Figure 3. The Average Effect Sizes for Decoding and Its Associated Variables.

failed to provide a sample that can be regarded as representative for the population (k=26). None of the studies provided information about non-responders (k=26), therefore a decision about non-response bias was not possible to make. The majority of the studies used appropriate tests for measuring the outcome variables (k=22), but not all studies used standardized tests (k=11). In general, studies described their methods in a way that could enable replication (k=24) and the basic data was adequately described (k=24). The discussions and conclusions were most often justified by the results (k=22), but many studies failed to discuss limitations of the study (k=10). Results regarding certainty of evidence, and a risk of bias plot (see Figure S17) are in the Supplements.

Discussion

This meta-analysis concerning individuals with ID showed that reading comprehension is significantly associated with decoding, listening comprehension, IQ, and vocabulary and that decoding is significantly associated with phonological awareness, phonological STM, RAN, IQ, chronological age, vocabulary, visual STM, and ELWM. The discussion will focus on the strong and moderate associations, associations in the low range will not be discussed further.

Variables Associated With Reading Comprehension

This meta-analysis showed that reading comprehension was strongly associated with decoding and moderately associated with listening comprehension. This is similar to research findings about typically developing children (Lervåg et al., 2018), and children with reading comprehension difficulties (Catts et al., 2006), and these associations support the SVR (Gough & Tunmer, 1986). However, other variables also were associated with reading comprehension, something that is not predicted by the SVR. For example, vocabulary was also shown to correlate strongly with reading comprehension, a relationship that has been identified in research on typically developing children as well. Ouellette (2006) found that measures of receptive vocabulary breadth and depth of vocabulary knowledge accounted for 28.5% of the variance in reading comprehension in a sample of typically developing students in Grade 4, even when decoding was taken into account. In another study, Ouellette and Beers (2010) argued for a not-so-SVR when their results showed that vocabulary accounted for 15.3% of the variance in reading comprehension in typically developing students in Grade 6, even when decoding and listening comprehension were also entered into the analysis.

These findings about the relevance of vocabulary to reading comprehension support the LQH (Perfetti, 2007), where both vocabulary size and quality of the reader's lexical representations are assumed to affect reading comprehension directly. The strong associations between reading comprehension and decoding, listening comprehension, and vocabulary in the current analysis indicate that combining the theoretical frameworks might be a successful way of explaining reading comprehension in individuals with ID. Other support for a combined model comes from Verhoeven and van Leeuwe (2008) who found in a longitudinal study on typically developing children, that a combined model of decoding, listening comprehension, and vocabulary predicted reading comprehension. In another study, focusing on children with ID enrolled in special education classes,

reading comprehension was predicted by decoding and a composite variable of vocabulary and syntactic skills (Verhoeven & Vermeer, 2006).

Furthermore, the current study found a moderate correlation between reading comprehension and IQ. This is consistent with findings from Verhoeven and Vermeer (2006), where reading comprehension was predicted by non-verbal IQ, decoding and language skills, in a sample of 10- and 12-year-old children with ID. Studies on typically developing children have also found significant correlations between IQ and reading comprehension both in early and later grades (Hulslander et al., 2010; Scarborough, 1998). In a longitudinal study by Hulslander et al. (2010), full scale IQ was found to be the only significant longitudinal predictor of later reading comprehension over and above decoding abilities and initial measures of reading comprehension. In contrast, two other longitudinal studies did not find any predictive contribution of IQ when earlier reading skills were controlled for (Cunningham & Stanovich, 1997; Scarborough, 1998).

Our meta-analysis revealed many similarities between the pattern of correlations between reading comprehension and other cognitive variables in individuals with ID and individuals with typical development. In addition, although the pattern of correlations in individuals with ID was found to be consistent with the SVR, the presence of other significant correlations supported the LQH. This suggests that a combined theoretical framework is needed to explain reading comprehension in both typical development and the development of individuals with ID.

Variables Associated With Decoding

Our meta-analysis suggests that the ability to process speech sounds (phonological awareness, RAN, and phonological STM) is related to decoding measures. There was a strong and significant correlation between decoding and phonological awareness and a moderate and significant correlation with RAN. Similar findings have been reported in a large body of research on typically developing children (Melby-Lervåg et al., 2012; Moll et al., 2014; Schatschneider et al., 2004), and also correspond with previous research on individuals with ID (Barker et al., 2014; Kennedy & Flynn, 2003; Saunders & DeFulio, 2007). A moderate and significant association was found between phonological STM and decoding, which has been reported in studies on individuals with ID (Channell et al., 2013; Conners et al., 2001; Henry & Winfield, 2010).

Decoding also showed a significant and moderate correlation with vocabulary. The association between decoding and vocabulary have been shown in several studies focusing on individuals with DS (Boudreau, 2002; Cardoso-Martins et al., 2009; Naess et al., 2011). Notably, the meta-analytic review by Naess et al. (2011) found that

vocabulary, not phonological awareness, predicted non-word decoding. It could be the case that the moderate association between vocabulary and decoding found in the present study is driven by the primary studies focusing on individuals with DS. There are no clear answers as to why vocabulary seems to be important for decoding in the DS group. Boudreau (2002) suggested that language skills could be the foundation for building literacy skills, or that it might be the case that both language and literacy draw upon a third independent cognitive process.

Furthermore, this meta-analysis showed that decoding correlated moderately with IQ. The relationship between decoding and IQ has been debated in the literature. For children with typical development, it appears that IQ is not related to decoding ability (Gustafson & Samuelsson, 1999; Stanovich, 2005). However, for the population with ID this is still an open question. van Tilborg et al. (2014) found that non-verbal intelligence was the only significant predictor of word decoding in a group with non-specific ID, explaining a total of 33% of the variance, even though letter knowledge and phonological awareness were used as predictors in the regression. In contrast, a study by Conners et al. (2001) found that two groups with mixed etiology ID divided by level of decoding ability did not differ on an IQ measure. Instead, the better decoders in the study scored significantly higher on measures of language ability and phonemic awareness, which is more in line with research on typical readers and individuals with dyslexia. Boudreau (2002) also found only a weak relationship between nonverbal mental age and decoding (both decoding of words and nonsense words) in a group with DS. The moderate association found in the current study indicates that IQ could play an important part in the development of decoding ability for individuals with ID, although this possibility needs to be evaluated in investigations of the shared variance between IQ and other predictor variables.

Decoding also correlated moderately with ELWM. The role of working memory for reading has mainly been emphasized in relation to reading comprehension, but there are studies on typically developing children suggesting that working memory plays an important role in decoding as well (Christopher et al., 2012). It would be reasonable to assume that working memory has a larger impact on decoding during the early stages of development when decoding is expected to be an effortful task. Many individuals with ID, both children and adults, decode words with an alphabetic strategy (Ratz & Lenhard, 2013), which could explain the association between decoding and ELWM found in this meta-analysis. However, it also is the case that in typical readers there is a relationship between working memory and decoding even at higher grades where an alphabetic strategy would not be expected (Peng et al., 2017). Further research is needed to understand the reasons for this relationship in individuals with ID.

As with reading comprehension, in individuals with ID the pattern of correlations between decoding and other variables was similar to the pattern reported in relation to those with typical development. An interesting exception to this was the correlation between decoding and IQ. In individuals with ID, unlike those with typical development, this correlation was found to be significant; and this could be a useful topic for future research.

Limitations and Future Directions

The tests of heterogeneity indicated significant betweenstudy variability for many variables. A standard procedure is to explore if between-study heterogeneity can be explained by moderators. However, due to the small number of primary studies in most analyses, moderator analyses were only feasible for four associations. These analyses identified study quality as a significant moderator for only one association, between decoding and phonological awareness.

The quality assessment showed that all primary studies had a potential risk of bias, often related to participant recruitment and sample size justification. These are common challenges in disability research due to smaller population sizes. About half of the studies also had a high risk of bias because they used potentially unreliable, study-specific measures.

A more general limitation of the meta-analysis is the possibility of measurement error. Because individuals with ID have intellectual difficulties, a test of visual STM could instead be an assessment of the degree to which the participant with ID understood the instruction. This phenomenon is discussed by van Wingerden et al. (2018), who suggested that the cognitive demands of phonological awareness tasks could lead them to capture higher-order skills like working memory. This potential limitation is a concern with participants who have cognitive difficulties, although validated standardized tests can help to address the issue.

Except for two associations, certainty of evidence was rated as low or very low, mostly due to the risk of bias, inconsistency, and imprecision of results. As previously discussed, the frequent use of unreliable measures likely contributed to these limitations. In addition, researcher decisions regarding study inclusion may have influenced heterogeneity. Our decision to include studies across different aetiologies enabled a meta-analysis by increasing the number of primary studies but may have introduced a larger amount of heterogeneity.

A further consideration is the influence of orthographic transparency, as primary studies were conducted in languages with varying levels of transparency. Readers of transparent orthographies tend to read faster and more accurately than those of opaque orthographies (Patel et al., 2004). Moreover, the predictors of decoding appear to

differ depending on orthographic transparency, with RAN consistently predicting reading fluency, whereas the relationship between decoding and phonological awareness is complex and interactive (Landerl et al., 2019). Although no clear patterns related to orthographic transparency were observed in the forest plots, this factor warrants further investigation in future research. One reviewed study compared German- and French-speaking children with ID, finding that spoken language did not predict progress in word and non-word reading (Sermier Dessemontet & de Chambrier, 2015). Because the orthographic transparency of German and French are not fundamentally different, a comparison between languages with greater orthographic differences would provide a more stringent evaluation of these effects. For example, a comparison between Frenchand English-speaking individuals might have produced other results as onset entropy (an index of orthographic transparency) is almost twice as high for English compared with French (Ziegler et al., 2010).

The limitations of our systematic review and meta-analysis reflect many of the more general limitations of research concerned with reading, and especially research concerned with ID. Future studies should address these issues while ensuring a high level of transparency when reporting their findings. Adopting open science practices to enhance reproducibility and transparency would be highly beneficial.

Conclusion

This meta-analysis focussed on the predictor variables of reading comprehension and decoding in individuals with ID. When considering our findings, it is important to bear in mind the limitations we have identified. The analysis has confirmed that many variables identified in studies on typically developing children and other groups are related to decoding and reading comprehension in individuals with ID. An exception to this general pattern was that IQ correlated moderately with decoding in individuals with ID, whereas studies on individuals with typical development indicate that there is no significant relationship between these two variables (Gustafson & Samuelsson, 1999; Stanovich, 2005). Furthermore, this meta-analysis indicates support for a combination of the SVR and the LQH as reading comprehension is significantly related to decoding, listening comprehension, and vocabulary. All this taken together confirms that theoretical frameworks describing the relationships between components of reading in typically developing children and other groups could reasonably be applied to individuals with ID.

Consequently, our results imply that abilities associated with reading comprehension and decoding in individuals with ID are similar to those of typically developing children and struggling readers. This indicates that support and interventions developed for typically developing children and

struggling readers might be cautiously applied to students with ID, with the addition of feasible adaptations. This is in line with findings from intervention research that reading interventions could be very effective for students with ID if the delivery and context, not the content, are adapted to student needs (see, for example, Allor et al., 2014).

Supplemental Material

Please find the following supplementary material below: additional figures, tables, and results. Please find the following supplementary material on OSF https://doi.org/10.17605/OSF.IO/U7P8G: data files, analysis script, PRISMA checklist, PRISMA abstract, and a detailed search strategy.

Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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