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Is orthographic knowledge a strength or a weakness in individuals with dyslexia? Evidence from a meta-analysis

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Abstract
The purpose of this meta-analysis was to examine if individuals with dyslexia (DYS) have a deficit in orthographic knowledge. We reviewed a total of 68 studies published between January 1990 and December 2019, representing a total of 7215 participants. There were 80 independent samples in the chronological-age (CA)-DYS comparison and 33 independent samples in the comparison between DYS and reading-level (RL) controls. A random-effects model analysis revealed a large effect size (Cohen's d = 1.17) for the CA-DYS comparison and a small effect size (Cohen's d = 0.18) for the RL-DYS comparison. In addition, we found significant heterogeneity in the effect sizes that was partly explained by the level of orthographic knowledge (effect sizes being higher for lexical than sub-lexical orthographic knowledge). These results suggest that individuals with dyslexia experience an orthographic knowledge deficit that is as large as that of phonological awareness and rapid automatized naming reported in previous meta-analyses.

Keywords Dyslexia · Meta-analysis · Orthographic knowledge · Surface dyslexia · Writing system

Developmental dyslexia, defined as a persistent and unexpected difficulty in developing age- and experience-appropriate word reading skills, is one of the most common learning disabilities affecting 5–10% of all school-age children (e.g., Snowling, Hulme, & Nation, 2020).

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Although the exact cause(s) of dyslexia remain unclear, researchers concur that most students with dyslexia have a phonological deficit that is manifested primarily in phonological awareness and rapid automatized naming tasks (e.g., Hulme & Snowling, 2013; Vellutino, Fletcher, Snowling, & Scanlon, 2004). Indeed, in their meta-analysis, Melby-Lervåg, Lyster, and Hulme (2012) found a large effect size (Cohen's $d = 1.37$) when comparing individuals with dyslexia to their chronological-age (CA) controls in phonological awareness tasks (children with dyslexia performed more poorly). Similarly, Araújo and Faísca (2019) reported a large effect size when comparing the two groups in rapid automatized naming tasks (Cohen's $d = 1.19$).

Even though difficulties in phonological processing skills have been documented in previous meta-analyses (e.g., Araújo & Faísca, 2019; Kudo, Lussier, & Swanson, 2015; Melby-Lervåg et al., 2012; Parrila, Dudley, Song, & Georgiou, 2020; Snowling & Melby-Lervåg, 2016; Swanson, Zheng, & Jerman, 2009), to our knowledge, only one meta-analysis has examined if individuals with dyslexia experience similar difficulties in orthographic knowledge,[1] and it included only studies with adults (Reis, Araújo, Morais, & Faísca, 2020). In their meta-analysis, the average effect size was found to be $d = 1.23$. Thus, the purpose of this meta-analysis was to replicate and extend Reis et al.'s meta-analysis by including also samples from studies with children and adolescents and by comparing the performance of individuals with dyslexia not only against their chronological-age (CA)–matched controls but also against their reading-level (RL)–matched controls. RL-matched designs are commonly used to test assumptions of causality following the logic that if the poor readers perform poorer than their RL-matched controls on task A assessing construct B, then construct B is a potential cause for dyslexia (e.g., Bradley & Bryant, 1978; Bryant & Goswami, 1986; see also Parrila et al., 2020, for a recent discussion on the use of RL-match designs).

Theoretical reasons for orthographic knowledge deficits in dyslexia

There are several theoretical reasons to expect deficits in orthographic knowledge by individuals with dyslexia. First, according to the dual-route theory of reading (e.g., Coltheart, 2005; Coltheart, Rastle, Perry, Langdon, & Ziegler, 2001), words are recognized by translating graphemes to phonemes (phonological route) or by mapping orthographic properties directly to the lexical entry (direct visual route). Consequently, we would expect difficulties in reading words to emanate from difficulties in processing skills underlying access and use of each route. Since orthographic knowledge is important for the development of sight word reading (e.g., Ehri, 2014), we would expect individuals who have a difficulty in accessing the direct visual route to also experience difficulties in orthographic knowledge. Likewise, in Chinese, if

[1] We acknowledge that terms like "orthographic awareness" and "orthographic skills" have also been used in the literature, particularly in studies conducted in Chinese (e.g., Lin, Mo, Liu, & Li, 2019; Yeung, Ho, Chan, & Chung, 2016). Even though we included all of these terms in our search process (see the "Method" section), for the purpose of this paper, we will consistently use the term orthographic knowledge. In addition, we acknowledge that various definitions of orthographic knowledge exist in the literature. For example, Stanovich and West (1989) defined orthographic knowledge as "the ability to form, store, and access orthographic representations of words" (p. 404), Newby, Recht, and Caldwell (1993) as "the rapid recognition of sight-words" (p. 73), Barker, Torgesen, and Wagner (1992) as "memory for specific visual/spelling patterns" (p. 47), and Manis, Custodio, and Szeszulski (1993) as "the ability to access visual-orthographic codes for specific words" (p. 65). In the context of Chinese, orthographic knowledge refers to children's knowledge of the positions, structuring, and functions of radicals; children's awareness of conventional rules in characters; and their ability to identify or distinguish real characters from a pool of pseudocharacters and visual symbols (see, e.g., Ho et al., 2007; Lin et al., 2019; Luo, Chen, Deacon, & Li, 2011).

children were to access their mental lexicon via the phonological route, then they would be slow readers due to the large number of homophones in the character corpora. If a visual input (i.e., Chinese character) stimulates multiple lexical entries via phonological mediation, then it should impede or delay the abstraction of semantics. For example, if a reader sees the graphic form “恕” [shu]4 (the number signals the tone) and systematically activates 26 characters pronounced [shu]4 in the mental lexicon, it should take some time to decide which one matches “恕” [shu]4. In contrast, if the reader is able to utilize direct access to map the orthographic unit to its semantics, this should speed up the process and ease the laborious procedure of transforming print to sound and meaning. Thus, we would expect orthographic knowledge to play an important role in reading Chinese and, at the same time, be a core deficit in dyslexia.

Second, deficits in orthographic knowledge would be expected on the basis of its theoretical connections with phonological awareness and rapid naming. According to the amalgamation hypothesis (Ehri, 1980), children first develop an awareness of the sounds of their written language (phonemic awareness) and knowledge of letter-sound correspondences. With the help of these skills, they begin to phonologically recode unknown words.[2] As they phonologically recode these words, the phonemes are bonded to the letters in the word; these letter-sound bonds help the child to construct orthographic representations of words. Thus, the ability to construct initial orthographic representations of words relies on children’s phonemic awareness. Given the findings of previous meta-analyses showing that individuals with dyslexia experience deficits in phonological awareness (see Kudo et al., 2015; Melby-Lervåg et al., 2012; Parrila et al., 2020), we should also expect deficits in orthographic knowledge. Similarly, Bowers and Wolf (1993) proposed that if children are slow in accessing and retrieving the names of letters (indexed by slow performance in rapid naming tasks), this would impact their ability to form high-quality orthographic representations of words. Previous studies have shown that children with naming speed deficits experience difficulties in orthographic knowledge (e.g., Bowers, Sunseth, & Golden, 1999; Powell, Stainthorp, & Stuart, 2014; Sunseth & Bowers, 2002).

Although we have good theoretical reasons to expect significant deficits in orthographic knowledge by individuals with dyslexia, evidence from empirical studies is mixed. In line with the theoretical expectations, some studies have shown that individuals with dyslexia experience significant difficulties in orthographic knowledge (e.g., Chung, Ho, Chan, Tsang, & Lee, 2010; Curtin, Manis, & Seidenberg, 2001; Diamanti et al., 2018a, b; Ho, Chan, Lee, Tsang, & Luan, 2004; Hultquist, 1997; Jiménez et al., 2008). In contrast, some studies have shown that children with dyslexia perform either better or equal to controls in orthographic knowledge (e.g., Chung et al., 2008; McArthur et al., 2013; Rothe, Cornell, Ise, & Schulte-Körne, 2015; Siegel, Share, & Geva, 1995). In fact, on the basis of these findings, Siegel et al. (1995) argued that orthographic knowledge might be a strength in individuals with dyslexia.

Moderators

In the presence of these diverse findings, it is reasonable to also expect significant heterogeneity in the effect sizes, which then requires an examination of the role of possible moderators.

[2] This is very similar to Share’s (1995) self-teaching hypothesis, according to which orthographic representations of words are built through phonological recoding, which functions as a self-teaching mechanism.

For the purpose of this meta-analysis, we examined the role of two moderators that are directly related to orthographic knowledge (i.e., level of orthographic knowledge and type of orthographic knowledge score) and the role of five moderators that are more generic (i.e., age, writing system, orthographic consistency, type of dyslexia, sample selection criteria) and are frequently encountered in meta-analyses of cognitive deficits in dyslexia (e.g., Araújo & Faísca, 2019; Kudo et al., 2015; Melby-Lervåg et al., 2012; Parrila et al., 2020).

Level of orthographic knowledge According to Apel (2011), orthographic knowledge consists of two levels: lexical and sub-lexical.[3] Lexical orthographic knowledge refers to the stored mental representation of known words. To measure lexical orthographic knowledge, researchers have mostly used the Orthographic Choice task (Olson, Wise, Conners, Rack, & Fulker, 1989). In this task, a real word is presented along with its pseudo-homophone (e.g., *rain – rane*) and individuals are asked to select the correctly spelled word. In turn, sub-lexical orthographic knowledge refers to knowledge of permissible language-specific orthographic patterns. To assess sub-lexical orthographic knowledge, researchers have mostly used the Word Likeness task in which individuals are asked to select which of the two juxtaposed pronounceable pseudowords (e.g., *filk – filv*) "looks like a real word" (i.e., it contains a permissible orthographic pattern). Because children practice reading and spelling whole words and not sub-lexical units and because learning of permissible orthographic patterns happens mostly implicitly (Treiman, 1993), we would expect children with less exposure to print and less reading practice (i.e., children with dyslexia) to experience more difficulties in lexical orthographic knowledge than in sub-lexical orthographic knowledge.

Type of orthographic knowledge task Even though most studies examining the role of orthographic knowledge in dyslexia have reported accuracy scores (e.g., Diamanti et al., 2018b; Sprenger-Charolles, Colé, Kipffer-Piquard, Pinton, & Billard, 2009; Szenkovits & Ramus, 2005), there are also studies that have reported response time scores (e.g., Ho, Chan, Chung, Lee, & Tsang, 2007; Jiménez-González & Valle, 2000; Martens & de Jong, 2006). No directional hypothesis could be articulated here as previous studies have not examined if the severity of the deficits varies according to the type of score used.

Writing system and orthographic consistency Effect sizes may also vary as a function of the writing system (alphabetic vs. non-alphabetic) or degree of orthographic consistency (high, medium, and low) among the alphabetic orthographies. For example, learning to read Chinese (a non-alphabetic orthography) imposes a high cognitive demand on orthographic knowledge because the smallest orthographic unit in Chinese, the stroke, does not convey any sound information, and therefore, Chinese characters cannot be sounded out through the application of letter-sound correspondences as in alphabetic writing systems. Thus, we would expect orthographic knowledge deficits in non-alphabetic orthographies to be more severe than in alphabetic orthographies. Likewise, because in consistent alphabetic orthographies (e.g., Finnish, Greek) every letter corresponds roughly to one sound, children can take advantage of these systematic relations to read words. Consequently, they would not need to rely heavily on their orthographic knowledge to read words (see Gagl, Hawelka, & Wimmer, 2015) and deficits in orthographic knowledge should not be as pronounced. In contrast, in inconsistent

[3] Some researchers have called the same levels "word-specific" orthographic knowledge and "general" orthographic knowledge (e.g., Bosse, Chaves, Largy, & Valdois, 2015; Zarić, Hasselhorn, & Nagler, 2020).

alphabetic orthographies (e.g., English, French) with equivocal letter-sound correspondences, children would have to develop flexible strategies in reading and rely on both phonological and orthographic processing skills. In these orthographies, deficits in orthographic knowledge should be more pronounced.

Type of dyslexia Because phonological dyslexics experience significant difficulties accessing and using the phonological recoding route in reading (Coltheart et al., 2001), they are forced to use the direct visual route. In this case, we would expect orthographic knowledge to be a relative strength. In turn, surface dyslexics would be more inclined to use the phonological recoding route, in which case orthographic knowledge would be a relative weakness. Indeed, in their study with phonological and surface dyslexics, Curtin et al. (2001) presented evidence in support of the aforementioned predictions.

Sample selection criteria A variety of approaches have been followed in selecting participants with dyslexia, and this may account for some of the variability in the effect sizes. For example, in some studies, children with dyslexia were selected on the basis of previous diagnosis (without any further information about how this diagnosis was made; e.g., Breznitz, 2002; Stoodley, Harrison, & Stein, 2006). In turn, in some other studies, children with dyslexia were selected based on teacher's nomination (e.g., Diamanti et al., 2018b; Wolff & Lundberg, 2003) or after teacher nomination and standardized assessment (e.g., Manis & Lindsey, 2008; Serrano & Defior, 2008).

The present study

The purpose of this meta-analysis was to examine if individuals with dyslexia have an orthographic knowledge deficit when compared to their chronological-age (CA)– and reading-level (RL)–matched controls. Based on the findings of previous meta-analyses examining phonological processing skills (e.g., Kudo et al., 2015; Melby-Lervåg et al., 2012; Parrila et al., 2020; Snowling & Melby-Lervåg, 2016) as well as the findings of Reis et al.'s (2020) meta-analysis with adult samples, we expected large differences between dyslexics and CA controls and small (non-significant) differences between dyslexics and RL controls.

Method

Data collection

To identify the studies for the meta-analysis, we first searched in the PsycINFO, ProQuest Educational, PubMed, Medline, ERIC, and Scopus computerized databases for publications between January 1990 and December 2019. A combination of terms related to orthographic knowledge (*orthographic knowledge* OR *orthographic awareness* OR *orthographic processing* OR *orthographic skills* OR *lexical knowledge* OR *sub-lexical knowledge* OR *orthographic choice* OR *word likeness* OR *homophone choice OR letter string choice OR lexical decision*) crossed with terms related to reading difficulties (*dyslexia* OR *dyslexic(s)* OR *reading disability(ies)* OR *learning* *disability(ies)* OR *reading disorder* OR *poor readers* OR at *risk readers* OR *special education*) was used to identify the initial pool of studies. Second, abstracts of peer-reviewed studies, dissertations, book chapters, reference lists of previous meta-analyses (Araújo & Faísca, 2019; Kudo et al., 2015; Melby-Lervåg et al., 2012; Parrila et al., 2020; Reis et al., 2020; Swanson et al., 2009), and journals publishing studies on dyslexia (*Journal of Learning Disabilities*, *Annals of Dyslexia*, *Dyslexia*, *Learning Disabilities Quarterly*, *Reading and Writing*, *Scientific Studies in Reading*, *Journal of Experimental Child Psychology*, and *Journal of Research in Reading*) were scrutinized. Finally, we contacted researchers who had published on the topic and asked them to share with us any of their unpublished data.

A total of 851 studies were initially identified (see Fig. 1). After removing any duplicated studies (e.g., studies identified through the database search and through our search in specialized journals), the second and third authors reviewed the abstract of the remaining 240 studies (all abstracts were written in English) and applied the following exclusion criteria:

(1) The study included participants who did not have dyslexia (i.e., participants with familial risk for reading disability, intellectual disability, non-specific learning disability, or language disorder).

(2) The study included "compensated" or "high-functioning" dyslexics. Given that "compensated" or "high-functioning" dyslexics have been defined in various ways in the literature (see Parrila, Georgiou, & Corkett, 2007, for a discussion), including these studies in this meta-analysis would end up introducing a significant amount of "noise" in our results.

(3) The study did not have CA- or RL-matched controls.

(4) The study used qualitative methodology, was a case study, or a literature review.

Fifty-four studies were excluded after applying these criteria. Subsequently, the first, second, and third authors fully reviewed the remaining 186 studies. All of these studies were written in English with the exception of one study written in Greek (Fella, 2017) and five studies written in Chinese (Dong et al., 2012; Liu, Liu, & Zhang, 2006; Yang, Ning, Liu, Pan, & Lu, 2009; Zhao, Bi, & Yang, 2012; Zheng, Huang, & Jing, 2007). As the first and last authors of this paper are native speakers of Greek and Chinese, respectively, they served as translators of these studies.

Before finalizing our list of studies for the meta-analysis, we further applied the following exclusion criteria:

(1) The study did not provide sufficient data to determine effect sizes.
(2) To avoid violation of the independence of effect sizes (including data from the same sample more than once), studies from the same author were examined for duplicate samples. Whenever a sample overlap occurred, we included the study that was published earlier and excluded the later studies. In longitudinal studies, we coded data only from the first time point.
(3) If a dissertation was subsequently published as an article, we only considered the article.
(4) The study included letter knowledge or spelling tasks as a single measure of orthographic knowledge.
(5) The study used an orthographic knowledge task (i.e., Orthographic Choice) to further subdivide their participants with dyslexia into high and low in orthographic knowledge prior to comparing them to CA or RL controls (see, e.g., Bekebrede, van der Leij, & Share, 2009; van der Leij & Morfidi, 2006).

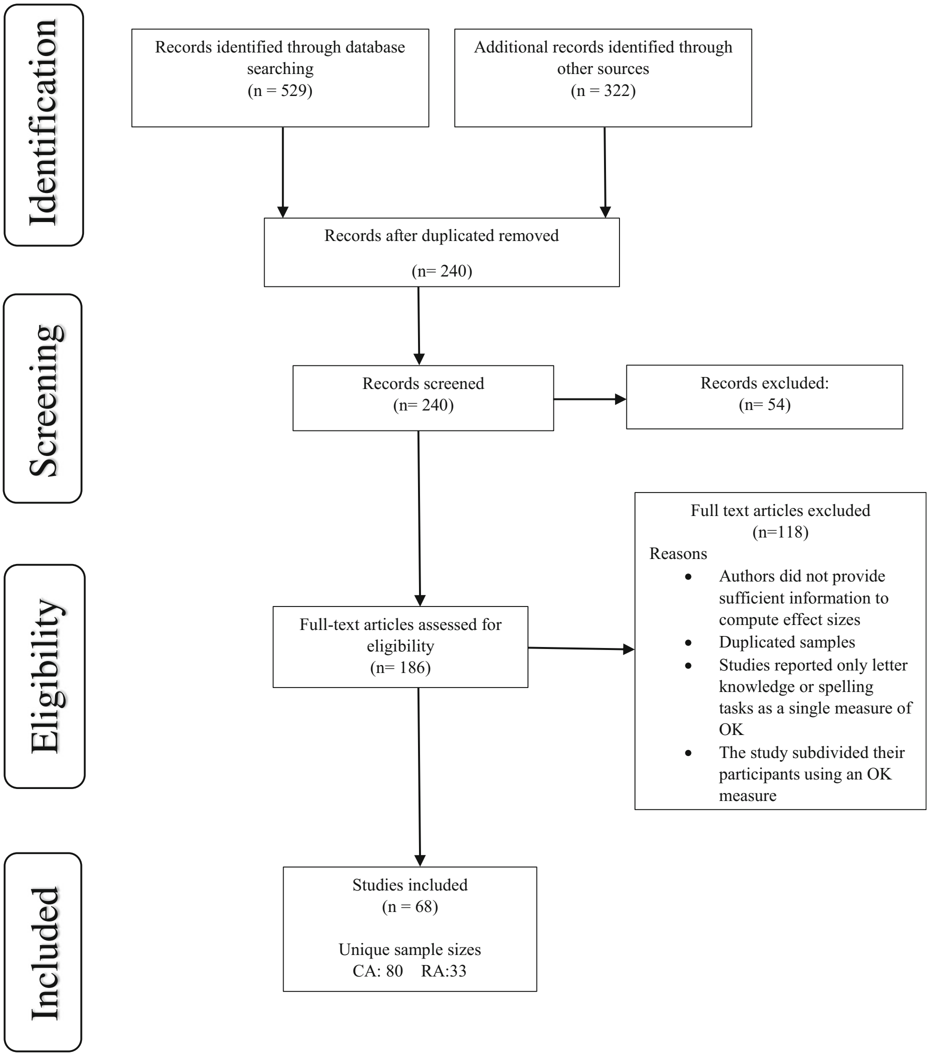

Fig. 1 Flow diagram for the search and inclusion on studies

(6) The study examined the double deficit hypothesis (DDH) in dyslexia using either an unselected sample of children or children who would not qualify as being dyslexics (e.g., children performing below the 35th percentile in a reading task). In the case of DDH studies with dyslexic children (e.g., Jiménez et al., 2008; Manis & Freedman, 2001), we used only the scores of the double deficit group and the control group in the analyses.

After these criteria were applied, our final sample comprised 68 studies, all published in refereed journals with the exception of two unpublished master's thesis (Barber, 2009; Tsantali, 2020) and one unpublished doctoral dissertation (Fella, 2017). A total of 80 unique samples were identified for the CA controls and 33 unique samples for the RL controls (see Appendices A and B in Supplementary Material, for information on the included studies).

Coding procedure

All of the studies that met the inclusion criteria were further coded independently by the second and third authors who are doctoral students with extensive experience in coding studies for meta-analyses. The data were recorded into two coding spreadsheets (one for the CA controls and one for the RL controls) and the intercoder agreement was calculated. The consensus rate was 96% for the CA controls coding and 98% for the RL controls coding. The few discrepancies in the coding were due to insufficient information provided in some studies about their participants and measures. The discrepancies between the coders were resolved after consulting the original study and after discussing the recorded data with the first author.

Recorded variables and coding procedures

The information extracted from each study was as follows: (a) means, standard deviations, and sample sizes from each group; (b) mean age of the participants at the time of the assessment; (c) type of dyslexia; (d) writing system/orthographic consistency; (e) type of orthographic knowledge task (lexical or sub-lexical); (f) type of score recorded (accuracy or response time); and (g) sample selection criteria.

Age The mean age of the samples in years and months was coded. The age ranged from 6 years and 6 months to 36 years.

Type of dyslexia We created three categories: (1) phonological dyslexia, (2) surface dyslexia, and (3) unspecified (this is when authors would say that they selected individuals with dyslexia without disclosing if their participants had phonological or surface dyslexia).

Writing system The writing system in which the study was conducted was first classified into two categories: alphabetic and non-alphabetic. The alphabetic category included the European languages and Hebrew (Verhoeven & Perfetti, 2017). The majority of the studies in the non-alphabetic category were conducted in Chinese (some of these studies were written in Chinese, refer to the "Data collection" section). There was also a study with Korean and a study with Hindi-speaking children (both of these studies were published in English). The alphabetic orthographies were further coded based on their orthographic consistency (see Seymour, Aro, & Erskine's, 2003, classification) in low, medium, and high consistency. English, French, Danish, and Hebrew comprised the low orthographic consistency group. Dutch, Portuguese, and Swedish comprised the medium orthographic consistency group, and Finnish, Greek, Italian, Spanish, German, and Norwegian the high orthographic consistency group.

Level of orthographic knowledge Operational criteria were first established in order to determine the separation of orthographic knowledge into two categories: lexical and sub-lexical. The lexical level of orthographic knowledge refers to specific knowledge of word-level orthographic representations (i.e., word-specific spelling). In Chinese, this would be similar to the task in which children are asked to select which of the two presented characters is a real character. The sub-lexical level includes permissible language-specific orthographic patterns that guide how words are generally represented in writing. In Chinese, this would be similar to the task in which children are asked to select which of the two presented pseudocharacters looks more like a real character (i.e., the radicals appear in a legal position). Subsequently, the orthographic knowledge tasks in each study were coded as lexical/sub-lexical orthographic knowledge tasks.

Type of orthographic knowledge score The score obtained from the orthographic knowledge tasks was coded into two categories: accuracy or response time.

Sample selection criteria The selection criteria used to identify the children in the dyslexia group were coded as (a) standardized test, (b) teacher nomination, (c) previous diagnosis, (d) school records, (e) teacher nomination plus tested on standardized and non-standardized tests, (f) non-standardized testing, and (g) previous diagnosis plus tested on standardized and non-standardized tests.

Moderators

In each study, we coded seven important moderators that could help us explain possible variability in the effect sizes: (a) age, (b) type of dyslexia, (c) writing system, (d) orthographic consistency, (e) level of orthographic knowledge, (f) type of orthographic knowledge score, and (g) sample selection criteria. Age was a continuous moderator. Type of dyslexia, writing system, orthographic consistency, level of orthographic knowledge, type of orthographic knowledge score, and sample selection criteria were categorical moderators. Detailed information on the recorded variables in every study is presented in Appendix A for the CA-matched controls and Appendix B for the RL-matched controls (see Supplementary Material).

Statistical analysis

The metafor package for the R statistical program (Viechtbauer, 2010) was used to perform the analyses. Effect sizes for studies involving group comparisons were computed with Cohen's d. When Cohen's d is positive, this means that the dyslexic group performed worse than the control group (CA or RL).

Overall effect sizes were estimated by calculating a weighted average of individual effect sizes (however, check also the "Results" section for analyses with all available effect sizes). Whether or not the overall effect size differed from zero was tested with a z test. 95% CIs were based on a random-effects model, which assumes that variation between studies can be systematic and not simply due to random error. For studies including both CA and RL control groups, a separate effect size was calculated for each of the two comparisons. Forest plots were used to present the distributions of effect sizes visually.

To examine the variation in effect sizes between studies, the Q test of homogeneity was used (Hedges & Olkin, 2014). A significant value on this test suggests that there is reliable variability between the effect sizes in the sample of studies. I^2 was used to determine the magnitude of the heterogeneity. I^2 is the proportion of total variation between effect sizes that is caused by real heterogeneity rather than chance. Moderator variables were also explored as potential sources of additional variance in the effect sizes. Linear models were used to predict the study's outcome from the moderator variables, both for the continuous (i.e., age) and categorical (i.e., writing system, level of orthographic knowledge, type of score, and sample selection criteria). The degree of difference between the subsets of studies was tested with a Q test and by comparing the effect sizes with CIs between the study subset.

Publication bias

To examine the relationship between the size of the effects from each study and their corresponding standard error, a Rosenthal's Fail-Safe N was computed. In a further step, we also conducted the Rank Correlation and Egger's Regression Tests to test for publication bias. In addition, we created funnel plots to assess the asymmetrical distribution of the studies around the mean effect size, which is also an indicator of publication bias (Borenstein, Hedges, Higgins, & Rothstein, 2009). In the funnel plot, the sample size is plotted on the y axis and the effect size on the x axis. In the presence of bias, the funnel should be asymmetric. Finally, in order to examine the impact of studies that might be missing from the analysis, we used the "trim and fill" method for random-effects models (Duval & Tweedie, 2000).

Results

The literature search and screening process resulted in 68 studies that were used in the meta-analysis: 64 of them included a CA-matched control group and 27 an RL-matched control group. These studies included 80 independent samples in the CA-DYS comparison and 33 independent samples in the RL-DYS comparison. There were 7215 participants represented, with sample sizes ranging from 8 to 279. The mean age reported in these samples ranged from 6.62 to 36.30 years.

Meta-analytic results

The random-effects model analysis demonstrated that the overall mean effect size of differences between the CA and DYS groups was significant (see Table 1 and Fig. 2 for the forest plot). The overall mean effect was 1.1742 (z = 14.8010, p < 0.0001, 95% CI = [1.0187, 1.3297]), favoring the CA group. For the RL-DYS comparison (see Table 1 and Fig. 3 for the forest plot), the overall mean effect size was 0.1811 (z = 4.7587, p = 0.0980, 95% CI = [−0.0111, 0.3733]).[4] The heterogeneity analysis further showed that the variation between studies was significant for both the CA-DYS (Q = 466.2654, I^2 = 84.81%, p < 0.0001) and the RL-DYS (Q = 100.6890, I^2 = 68.92%, p < 0.0001) group comparisons.

Moderator analyses

Only one significant moderator (level of orthographic knowledge) was found in the comparison between the CA and DYS groups (see Table 2). More specifically, the effect size was larger for lexical than sub-lexical orthographic knowledge (d = 1.2826 for lexical and d =

[4] Notice that similar results are obtained when using all possible effect sizes in the selected studies (186 in the CA-DYS comparison and 77 in the RL-DYS comparison) instead of using a weighted average of individual effect sizes. More specifically, when we reran the analyses using robumeta, we found that the overall mean effect in the CA-DYS comparison was 1.160 (p < 0.001, 95% CI = [1.000, 1.320]). In turn, the overall mean effect in the RL-DYS comparison was 0.183 (p = 0.072, 95% CI = [-0.0174, 0.383]).

Table 1 Meta-analytic results: overall standardized mean differences for the control and dyslexia group

Comparison	k	n	d	S.E.	Z value	p value	95% CI	Heterogeneity		
								I^2(%)	Q	p value
CA-DYS	80	CA: 3911 DYS: 2437	1.1742	0.0793	14.8010	<0.0001	[1.0187, 1.3297]	84.81	466.2654	<0.0001
RL-DYS	33	RL: 790 DYS: 694	0.1811	0.0980	1.8472	0.0647	[− 0.0111, 0.3733]	68.92	100.6890	<0.0001

Note. k = number of samples; n = total sample size; d = estimated Cohen's d in random-effects model; I^2 = the proportion of total variation caused by real heterogeneity; Q = Hedge's Q test of homogeneity

0.8376 for sub-lexical, $p = 0.0053$). As shown in Table 3, none of the moderators explained the variability in the effect sizes between the RL and DYS groups.

Publication bias

The results of the Fail-Safe N analysis suggested that the estimated effect sizes were reasonably stable. The results of Egger's Regression Test suggested the presence of publication bias in the model with the CA-DYS ($z = 3.9746$, $p < 0.0001$) comparisons (see Table 4). As suggested by the Rank Correlation Test, Kendall's taus for the CA-DYS group comparisons were significant (tau = 0.2380, $p = 0.0017$). No evidence of publication bias in the model for RL and DYS groups was recommended by Egger's Regression Test and Rank Correlation Test. Subsequently, the "trim and fill" analyses were performed for both CA-DYS and RL-DYS. The funnel plots indicated that studies were missing to the left of the mean (i.e., studies with effect sizes below the overall mean) (see Figs. 4 and 5). Therefore, the true effect size in these comparisons may be somewhat lower (corrected effect size for the CA-DYS = 1.1448, $p < 0.0001$ and corrected effect size for the RL-DYS = 0.1430, ns) than what has been reported in the initial analyses.

Discussion

The purpose of this meta-analysis was to examine if individuals with dyslexia have a deficit in orthographic knowledge when compared to their CA- and RL-matched controls. In line with our expectation, the effect sizes were large in the CA-DYS comparison (Cohen's $d = 1.17$) and small (but still significant) in the RL-DYS comparison (Cohen's $d = 0.18$). The effect size found in the CA-DYS comparison is very close to the one reported by Reis et al. (2020; $d = 1.23$) for studies with adult participants. Our effect sizes are also similar to the ones reported in previous meta-analyses examining other core deficits in dyslexia such as phonological awareness (e.g., Kudo et al., 2015; Melby-Lervåg et al., 2012; Parrila et al., 2020) and rapid automatized naming (e.g., Araújo & Faísca, 2019; Parrila et al., 2020). Taken together, these findings suggest that individuals with dyslexia experience difficulties in a variety of reading-related skills when compared to their CA controls. This, in turn, provides support to multiple-deficit models of dyslexia (e.g., Ho, Chan, Tsang, & Lee, 2002; McGrath et al., 2011; Pennington, 2006; see also Parrila et al., 2020). In addition, our findings suggest that the difficulties in orthographic knowledge persist into adulthood (the effect size in the adult group

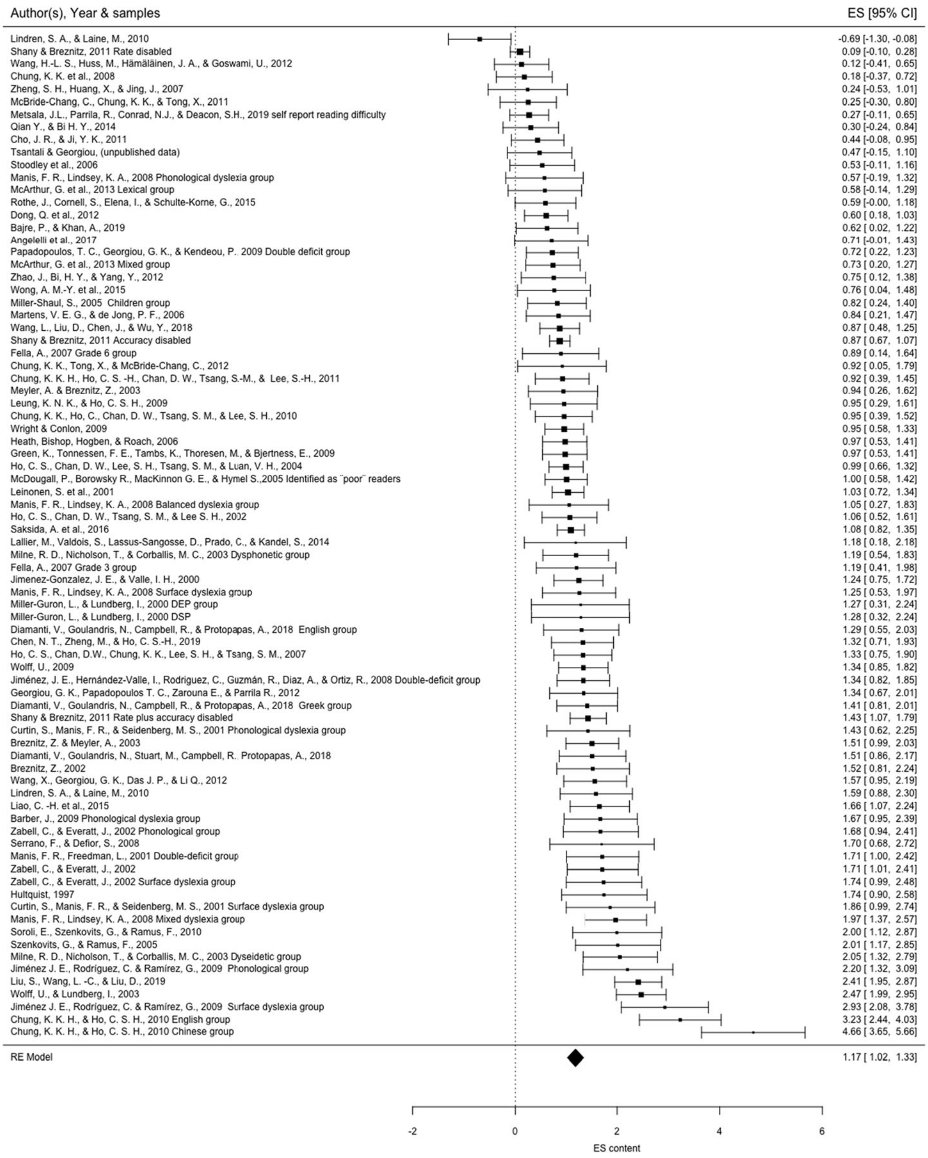

Fig. 2 Forest plot: strength of the standardized mean difference between CA and DYS groups

was a large as the effect size in younger ages) and can be found across writing systems, despite the fact that orthographic knowledge is somewhat differently defined in non-alphabetic orthographies such as Chinese.

In view of the dual-route theory of word reading (Coltheart et al., 2001), our findings suggest that individuals with dyslexia experience significant difficulties accessing and using both routes in word reading. Melby-Lervåg et al. (2012) have already reported a large effect size when comparing CA to DYS in phonological awareness (Cohen's d = 1.37), which is critical for phonological recoding. Similarly,

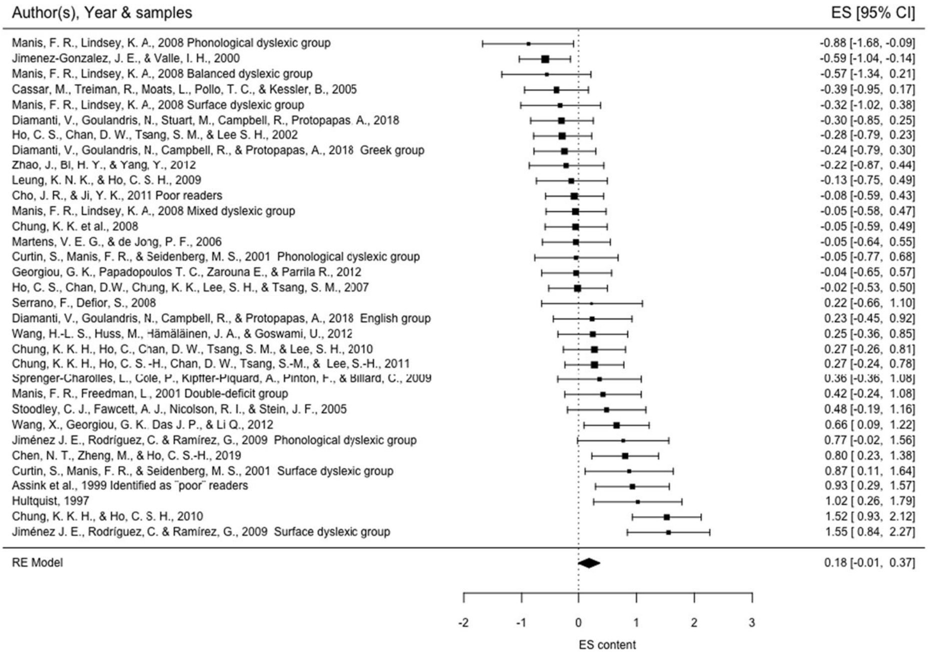

Fig. 3 Forest plot: strength of the standardized mean difference between RL and DYS groups

we have found a large effect size (Cohen's d = 1.17) in orthographic knowledge which is critical for the development of high-quality orthographic representations of words in the mental lexicon.

As expected, we also found significant heterogeneity in the effect sizes. Level of orthographic knowledge partly explained this heterogeneity in the CA-DYS comparison. There might be two explanations for this finding. First, as children practice reading and spelling whole words, it is more likely to acquire lexical orthographic knowledge than sub-lexical.[5] Consequently, earlier reading or spelling difficulties should impact more lexical than sub-lexical orthographic knowledge. Second, some researchers have argued that Orthographic Choice, the most popular lexical orthographic knowledge task, measures also reading ability (i.e., children who cannot read cannot answer the items; see Burt, 2006; Compton, Gilbert, Kearns, & Olson, 2020). To the extent this is true, then children with reading difficulties should perform more poorly on lexical orthographic knowledge.

None of the other moderators was significant. This means the effect sizes were relatively similar across different types of dyslexia, types of scores, writing systems, orthographic consistency, and sample selection criteria. Even though we did not have any specific expectations about the role of type of score and sample selection criteria, finding a non-significant effect of type of dyslexia and orthographic consistency caught us by surprise. In regard to type of dyslexia, it is possible that we did not have enough effect sizes to detect a significant difference. As can be seen in Table 2, the effect size in surface dyslexics was much larger (Cohen's d = 1.71) than in phonological dyslexics (Cohen's d = 1.21). However, these

[5] Notice though that some level of sub-lexical orthographic knowledge is present even at the onset of formal reading instruction (Cassar & Treiman, 1997; Treiman, 1993).

Table 2 Moderator analyses of the effect sizes in the CA-DYS group comparisons

Moderator variable	Number of effect sizes (k)	d	p value	95% CI	Difference in d (highest-lowest category)	Significance test (Q or β)	p value
1. Age	67					0.0190	0.1068
2. Type of dyslexia					0.5672	3.1958	0.2023
Phonological	8	1.2152	< 0.0001	[0.7056, 1.7247]			
Surface	6	1.7139	< 0.0001	[1.1210, 2.3069]			
Unspecified	54	1.1467	< 0.0001	[0.9580, 1.3354]			
3. Writing system					0.1366	0.6280	0.4281
Alphabetic	56	1.2162	< 0.0001	[1.0291, 1.4033]			
Non-alphabetic	24	1.0796	< 0.0001	[0.7984, 1.3609]			
4. Orthographic consistency					0.4470	1.9619	0.3750
Low	34	1.1307	< 0.0001	[0.9234, 1.3380]			
Medium	3	1.5777	< 0.0001	[0.9110, 2.2444]			
High	17	1.2834	< 0.0001	[0.9864, 1.5804]			
5. Selection criteria					1.0984	10.1082	0.1202
Standardized testing	20	0.9206	< 0.0001	[0.6217, 1.2196]			
Teacher nomination	2	2.0190	< 0.0001	[1.0903, 2.9476]			
Formal diagnosis	27	1.1113	< 0.0001	[0.8545, 1.3680]			
School records	4	1.0258	0.0037	[0.3332, 1.7185]			
Teacher nomination and tested	17	1.4987	< 0.0001	[1.1627, 1.8347]			
Non-standardized testing	2	1.0301	0.0260	[0.1232, 1.9370]			
Formal diagnosis and tested	8	1.2406	< 0.0001	[0.7815, 1.6997]			
6. Type of scores					0.0820	0.2706	0.6027
Accuracy	78	1.2066	< 0.0001	[1.0476, 1.3656]			
Response time	27	1.1246	< 0.0001	[0.8598, 1.3893]			
7. Level of Orth. Knowledge					0.4450	7.7820	0.0053
Lexical	60	1.2826	< 0.0001	[1.1110, 1.4542]			
Sub-lexical	25	0.8376	< 0.0001	[0.5763, 1.0990]			

Note. k = number of effect sizes; d = estimated Cohen's d for subsets of studies belonging to different categories of the moderator variable; Q = significant Q test value for categorical variables; β = regression coefficient in meta regressions for continuous variables

Table 3 Moderator analyses of the effect sizes in the RL-DYS group comparisons

Moderator variable	Number of effect sizes (k)	d	p value	95% CI	Difference in d (highest-lowest category)	Significance test (Q or β)	p value
1. Mean age RL/DYS	28/28					0.0707/0.0744	0.2681/0.2850
2. Type of dyslexia					0.7435	2.8026	0.2463
Phonological	3	− 0.0499	0.8820	[− 0.7086, 0.6088]			
Surface	3	0.6936	0.0339	[0.0527, 1.3345]			
Unspecified	23	0.1929	0.0804	[− 0.0233, 0.4092]			
3. Writing system					0.1066	0.2750	0.6000
Alphabetic	21	0.1399	0.2698	[− 0.1086, 0.3884]			
Non-alphabetic	12	0.2465	0.1207	[− 0.0648, 0.5578]			
4. Orthographic consistency					0.8495	1.8710	0.3924
Low	13	0.0803	0.6360	[− 0.2521, 0.4127]			
Medium	1	0.9298	0.1197	[− 0.2413, 2.1010]			
High	7	0.1387	0.5405	[− 0.3053, 0.5826]			
5. Selection criteria					0.6895	2.2398	0.8151
Standardized testing	7	0.2137	0.3418	[− 0.2269, 0.6542]			
Teacher nomination	1	− 0.2959	0.6045	[− 1.4157, 0.8239]			
Formal diagnosis	9	0.3233	0.0955	[− 0.0569, 0.7035]			
School records	3	0.3936	0.2573	[− 0.2874, 1.0747]			
Teacher nomination and tested	11	0.0669	0.7146	[− 0.2916, 0.4253]			
Non-standardized testing	0						
Formal diagnosis and tested	2	− 0.0052	0.9895	[− 0.7834, 0.7730]			
6. Type of scores					0.1716	0.7362	0.3909
Accuracy	31	0.2249	0.0225	[0.0317, 0.4182]			
Response time	10	0.0533	0.7592	[− 0.2877, 0.3943]			
7. Levels of Orth. Knowledge					0.1470	0.5902	0.4424
Lexical	24	0.2134	0.0739	[− 0.0206, 0.4474]			
Sub-lexical	14	0.0664	0.6570	[− 0.2266, 0.3594]			

Note. k = number of effect sizes; d = estimated Cohen's d for subsets of studies belonging to different categories of the moderator variable; Q = significant Q test value for categorical variables; β = regression coefficient in meta regressions for continuous variables

Table 4 Publication bias analyses

Comparison	Fail-Safe N	Egger's Method		Rank Correlation Test		Trim and Fill Procedure	
		z	*p*	Kendall's tau	*p*	Imputed	Corrected effect sizes
CA-DYS	35221	3.9746	< 0.0001	0.2380	0.0017	1	1.1448
RL-DYS	86	1.4074	0.1593	0.1856	0.1336	1	0.1430

numbers are based on 6 and 8 effect sizes, respectively, which is clearly not adequate to detect significant differences. Notably, the effect size in the "unspecified" category was even lower than in the other two categories (Cohen's $d = 1.14$). This likely indicates that the majority of participants in these dyslexia studies had phonological dyslexia.

The lack of significant effects of orthographic consistency was also unexpected in light of arguments that children in consistent orthographies (including children with dyslexia) rely mostly on small grain size units to read words correctly and fluently (e.g., Ziegler & Goswami, 2005; see also Gagl et al., 2015; Marinelli, Angelelli, Notarnicola, & Luzzatti, 2009). A possible explanation for this finding may relate to how children with dyslexia are selected in consistent orthographies (i.e., using speeded measures of reading). More specifically, some researchers have shown that children learning to read in consistent orthographies shift between strategies depending on task demands (e.g., Georgiou, Parrila, & Papadopoulos, 2008; Orsolini, Fanari, Tosi, De Nigris, & Carrieri, 2006). When asked to read accurately, they rely on letter-sound correspondences knowing that this strategy will help them decode correctly even long, unknown words. In contrast, when asked to read fluently, they rely on larger grain size units. Because in consistent orthographies researchers use reading fluency tasks to identify children with dyslexia, this may have resulted in picking up also orthographic knowledge difficulties.

Some limitations of the present meta-analysis are worth mentioning. First, even though some interactions (e.g., orthographic consistency X type of dyslexia) may be present, our relatively small sample size did not allow us to test for interaction effects. Second, some levels of the moderator variables had a small number of observations (e.g., type of dyslexia) and this may have prevented us from detecting significant effects for that moderator. Third, we did not examine how well the groups were matched on the reading tasks as this was beyond the scope of this meta-analysis. Recently, Parrila et al. (2020) showed that even though dyslexics were

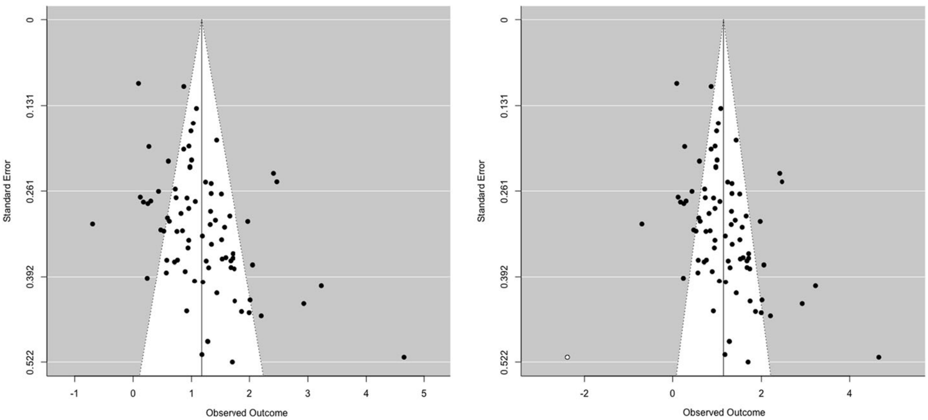

Fig. 4 Funnel plots for CA-DYS (left) and funnel plots with imputed samples for CA-DYS (right)

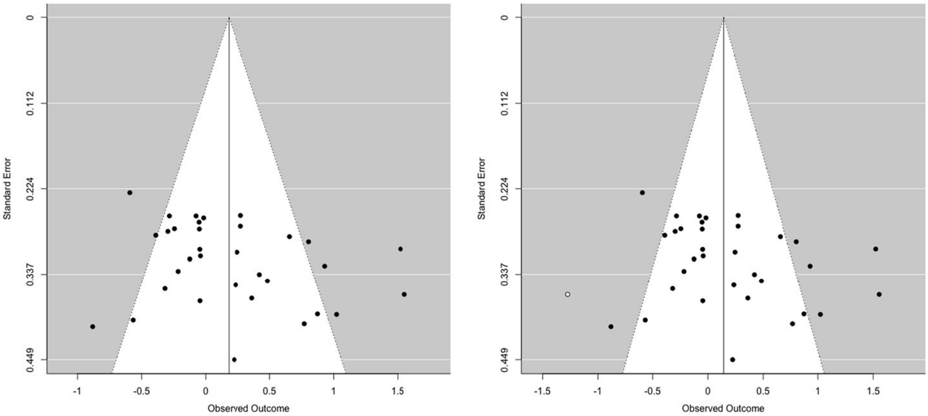

Fig. 5 Funnel plots for RL-DYS (left) and funnel plots with imputed samples for RL-DYS (right)

matched to their controls on one reading task (i.e., the task used to select them), they differed on other reading tasks. Obviously, imperfect matching may have significant implications when searching for core deficits in dyslexia. Finally, our "non-alphabetic" category included mostly studies conducted in Chinese. Thus, our findings may not generalize to other non-alphabetic languages.

Our findings have some important implications for assessment and intervention. Given that children and adults with dyslexia have a significant deficit in orthographic knowledge, researchers may consider including measures of orthographic knowledge when screening children for dyslexia. At the same time, researchers should explore ways to incorporate activities in orthographic knowledge in their intervention programs. Preliminary evidence from this kind of interventions has produced some promising results (e.g., Lovett et al., 2017; McMurray, in press; Morris et al., 2012).

To conclude, our findings add to those of previous meta-analyses (e.g., Araújo & Faísca, 2019; Melby-Lervåg et al., 2012; Parrila et al., 2020) by showing that individuals with dyslexia experience significant difficulties also in orthographic knowledge, but only when compared to CA controls and not to RL controls. However, in view of findings that earlier reading ability influences future orthographic knowledge (Conrad & Deacon, 2016), it is also possible that the observed difficulties of individuals with dyslexia in orthographic knowledge are partly a by-product of earlier reading difficulties. Future studies may want to explore the role of lexical and sub-lexical orthographic knowledge in groups of surface and phonological dyslexics.

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Declarations

Conflict of interest The authors declare no competing interests.

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*An asterisk precedes the studies that have been included in the meta-analysis.

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