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Subtypes of developmental dyslexia: Testing the predictions of the dual-route and connectionist frameworks

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Abstract

We investigated the phonological and surface subtypes of developmental dyslexia in light of competing predictions made by two computational models of single word reading, the dual-route cascaded model (DRC; Coltheart, Rastle, Perry, Langdon, & Ziegler, 2001) and Harm and Seidenberg's connectionist model (HS model; Harm & Seidenberg, 1999). The regression-outlier procedure was applied to a large sample to identify children with disproportionately poor phonological coding skills (phonological dyslexia) or disproportionately poor orthographic coding skills (surface dyslexia). Consistent with the predictions of the HS model, children with "pure" phonological dyslexia, who did not have orthographic deficits, had milder phonological impairments than children with "relative" phonological dyslexia, who did have secondary orthographic deficits. In addition, pure cases of dyslexia were more common among older children. Consistent with the predictions of the DRC model, surface dyslexia was not well conceptualized as a reading delay; both phonological and surface dyslexia were associated with patterns of developmental deviance. In addition, some results were problematic for both models. We identified a small number of individuals with severe phonological dyslexia, relatively intact orthographic coding skills, and very poor real word reading. Further, a subset of controls could read normally despite impaired orthographic coding. The findings are discussed in terms of improvements to both models that might help better account for all cases of developmental dyslexia.

Keywords

reading disability; phonological dyslexia; surface dyslexia; phonological coding; orthographic coding; computational models

Introduction

The current investigation examines reading-related performance of children and adolescents in the context of two computational models of single word reading: Harm and Seidenberg's (1999) connectionist model and the Dual Route Cascaded model (DRC; Coltheart, Rastle, Perry, Langdon, & Ziegler, 2001). These two models offer contrasting accounts of the phonological and surface subtypes of developmental dyslexia. In a large sample of typically developing and dyslexic readers ages 8 to 13, we tested competing predictions arising from the models regarding the nature of these subtypes and their developmental course.

Phonological and surface dyslexia were first identified as subtypes of acquired dyslexia among previously skilled adult readers who had sustained brain damage. Phonological dyslexia is associated with a selective deficit in reading nonwords, while surface dyslexia is associated with a selective deficit in reading irregularly spelled words (Coltheart, 1985). The existence of this double dissociation provided some of the strongest evidence for a dual-route framework positing that skilled readers of English use two separable routes for reading aloud: a lexical orthographic route and a non-lexical phonological route.

Early evidence for developmental analogues of acquired phonological and surface dyslexia came from single case studies of children (e.g., Coltheart, Masterson, Byng, Prior, & Riddoch, 1983; Temple & Marshall, 1983) who showed differential impairment on either nonword or exception word reading. In a group study of impaired readers, Castles & Coltheart (1993) introduced the influential regression-outlier method. This method identifies children with “pure” phonological or surface dyslexia, who are below age level in only nonword reading or only exception word reading, respectively, as well as children with “relative” phonological or surface dyslexia. These relative cases are below age level in both processes, but one process is more impaired than expected based on the other. Using this method, Castles & Coltheart reported that most dyslexic children fit either the phonological or surface subtype profile. A number of other studies have since used the regression-outlier method and reported that at least half of dyslexic children in English, French, or Spanish (in which lexical route function is assessed with reading of high frequency words) can be classified as belonging to one of these two subtypes (Jimenez, Rodriguez, & Ramirez, 2009; Manis, Seidenberg, Doi, & McBride-Chang, 1996; Sprenger-Charolles, Cole, Lacert, & Serniclaes, 2000; Stanovich, Siegel, & Gottardo, 1997; Ziegler et al., 2008). A substantial minority of dyslexic children are similarly impaired at both nonword and exception word reading and do not fit either subtype. Of course, the proportion of children who meet subtype criteria depends on the particular cutoff used to determine “poorer than expected” performance, with most studies using a 90% or 95% confidence interval. On balance, the evidence indicates that the phonological and surface forms of developmental dyslexia represent two ends of a continuum, rather than qualitatively distinct subtypes (Castles, Datta, Gayan, & Olson, 1999; Griffiths & Snowling, 2002). Nonetheless, comparison of children falling toward one end of the continuum or the other can provide important information about the development of the cognitive architecture for single word reading (Castles, Bates, & Coltheart, 2006).

Overview of the Models

The DRC, which offers a fully-specified computational instantiation of the dual-route framework, reads words aloud using two separate procedures. In the direct (lexical) route, orthographic input selects an entry in the orthographic lexicon via interactive activation, which in turn activates the appropriate phonological output. The indirect, or nonlexical, route, takes orthographic input, parses it into graphemes, converts the graphemes into their corresponding phonemes via a set of explicit rules, and then assembles these phonemes into a word for output. Only the lexical route can read exception words, since these words break rules of grapheme-phoneme correspondences. Only the nonlexical route can read nonwords, since these have not been encountered before and are not in the orthographic lexicon. Both routes contribute to successful reading of regular words. The DRC was developed to account for skilled adult reading; the model does not learn or change over time. However, its proponents argue that it offers a coherent account of developmental reading problems (Jackson & Coltheart, 2001; Castles et al., 2006), and the model has been used to simulate individual differences in developmental dyslexia (Ziegler et al., 2008). According to the traditional dual-route view, phonological dyslexia (developmental or acquired) arises from differential damage to the non-lexical route, while surface dyslexia arises from differential damage to the lexical route. Systematic lesioning of the DRC has been used to more precisely specify the nature of the impairments in acquired phonological and surface dyslexia. Damage to the DRC’s orthographic input lexicon produces acquired surface dyslexia (Coltheart et al., 2001). No single lesion can reproduce all of the specific patterns displayed by different patients with acquired phonological dyslexia. Some patients’ data are well simulated by damage to the grapheme-phoneme conversion procedures, while for others, damage to the phoneme system is required (Nickels, Biedermann, Coltheart, Saunders, & Tree, 2008).

Harm and Seidenberg's (1999) connectionist model (henceforth, the HS model) differs from the DRC in several important ways and offers an alternate account of developmental dyslexia subtypes. The HS model is based conceptually on the triangle model (Seidenberg & McClelland, 1989) and was designed to account for the development of normal reading as well as developmental dyslexia. The model does not include explicit rules about grapheme-phoneme correspondences; instead, rule-like behavior emerges over the course of network training based on probabilistic constraints. Since children typically approach the task of learning to read with a well-developed phonological system (Fowler, 1991), the HS model was first trained to develop phonological attractors, such that over time, interactive activation in a phonological network was pulled toward familiar pronunciations for English words. Next, the network was trained to "read;" a set of orthographic inputs was connected to the phonological network via a set of hidden units, and the back propogation through time algorithm was used to teach the model to produce the correct phonological output in response to a given orthographic input. Thus, in contrast to the DRC and several other computational models of single word reading, the HS model reads all words (and pseudowords) via a single procedure.

The HS model simulated the phonological and surface subtypes of developmental dyslexia by causing different types of damage to the network before training. The prevailing view of developmental dyslexia is that most cases are caused by underlying impairments in phonological representations (Peterson & Pennington, 2012). To produce phonological dyslexia, Harm and Seidenberg (1999) impaired the model's phonological representations with damage to the phonological network. Mild damage led to a pattern of pure phonological dyslexia (normal exception word reading and impaired nonword reading), while more substantial damage led to a pattern of relative phonological dyslexia (impairments in both nonword word and exception word reading, with the former deficit being more pronounced). Based on these findings, Harm and Seidenberg (1999) advanced a severity hypothesis. They argued that all pure cases of phonological dyslexia should be mild cases, while cases of mixed or relative phonological dyslexia should result from a more severe phonological impairment.

Based on empirical findings that children with surface dyslexia perform similarly to younger, typically developing readers on a variety of reading-related tasks (Manis et al., 1996; Sprenger-Charolles et al., 2000; Stanovich et al., 1997; see also Sprenger-Charolles, Siegel, Jimenez, & Ziegler, 2011), Harm and Seidenberg (1999) conceptualized surface dyslexia as "reading delay dyslexia." To model a reading delay, Harm and Seidenberg slowed the overall reading acquisition of their model in four different ways—providing less training (meant to simulate a lack of reading experience), reducing the learning rate, degrading the orthographic input, and removing some of the hidden units. In every case, the model showed a pattern of relative surface dyslexia (impairments in both exception word and nonword reading, with the former deficit being more pronounced). The HS model did not simulate pure surface dyslexia.

In contrast to the HS model, implementations of the triangle model that include a semantic network (Plaut et al., 1996; Harm & Seidenberg, 2004) provide two routes from print to naming: one from orthography to phonology, and one from orthography through semantics to phonology. However, these models differ from the DRC in that the division of labor between the two routes is not "clean"; instead, both routes contribute to successful reading of regular words, exception words, and pseudowords. Harm and Seidenberg's reading delay account of developmental surface dyslexia contrasts with the standard triangle-model account of acquired surface dyslexia, in which selective impairments in exception word reading arise from damage within the semantic pathway (Plaut, McClelland, Seidenberg, & Patterson, 1996; Woollams, Lambon Ralph, Plaut, & Patterson, 2007). Thus, an important question is whether the developmental form of surface dyslexia can be adequately understood without considering contributions from a semantic network. This issue is addressed at several points in the current investigation.

Some aspects of the DRC and connectionist learning models have been integrated in the connectionist dual-process (CDP) approach, which has successfully simulated a wide range of phenomena in normal word reading and acquired dyslexia (Perry, Ziegler, & Zorzi 2007, 2010; Zorzi, 2010; Zorzi, Houghton, & Butterworth, 1998b). The CDP+ model (Perry et al., 2007) retains a discrete dual-route architecture and encompasses the localist, lexical route of the DRC. However, the nonlexical route is implemented via a two-layer assembly (TLA) network in which regularities between graphemes and phonemes are learned through training; no explicit rules are specified. Notably, the German version of CDP+ provided a better fit to the human data for nonword reading than the DRC (Perry, Ziegler, Braun, & Zorzi, 2010). Various types of damage to the TLA could be used to model developmental phonological dyslexia. Of interest, Zorzi, Houghton & Butterworth (1998a) demonstrated that in the context of a small training set, adding a hidden layer to the TLA biased the network towards a more item-specific, lexicalist strategy and reduced its ability to generalize to novel items, thus showing some similarity to developmental phonological dyslexia. Note that this account of phonological dyslexia remains more consistent with the classic dual-route view than the HS model. Specifically, the underlying impairment is attributed to deficient grapheme-phoneme conversion, in contrast to the HS account in which the causal deficit lies in phonological representations. Recently, the CDP+ model has been “scaled up” to allow for accurate naming of bisyllabic words (Perry et al., 2010).

Overview of the Current Investigation

The DRC and HS models offer an example of the indeterminacy problem: both can account for patterns of observed data, namely, the existence of phonological and surface subtypes of developmental dyslexia. To better discriminate between the models, we used their accounts of the dyslexia subtypes to generate competing predictions and then we evaluated those predictions empirically. The current investigation includes six studies: Study 1 identifies children meeting a phonological or surface dyslexia pattern in a large sample and examines their reading-related skills, while Studies 2–6 test questions related to dyslexia subtypes that arise from the two models. Each of these five studies is described very briefly below, and in more detail in the introduction to the relevant study.

Study 2 tests the severity hypothesis of pure versus relative phonological dyslexia. According to the HS model, pure cases of phonological dyslexia result from a mild phonological deficit, while relative cases of phonological dyslexia result from a more severe phonological deficit. According to the DRC, pure phonological dyslexia results when only the nonlexical route is damaged (regardless of severity), while relative phonological dyslexia results when the lexical route is also damaged, albeit to a lesser degree. Although the DRC offers a coherent account of pure subtypes, it is more challenged to explain the preponderance of mixed cases of developmental dyslexia. Dual-route proponents (Castles et al.,2006; Jackson and Coltheart, 2001) have argued that while the lexical and nonlexical routes may function independently in healthy adults, their development is likely to be intertwined over the course of reading acquisition. For example, the ability to sound out unfamiliar words via the nonlexical route should promote the development of the orthographic lexicon (Share, 1995). Further, children may infer information about grapheme-phoneme correspondences by analyzing spelling-sound relationships in words they have learned to recognize (Fletcher-Flinn & Thompson, 2004). Thus, an initial deficit localized to one route would eventually impair the other route as well. Study 3 tests whether pure cases are more prevalent among younger dyslexic readers. The HS model makes the opposite developmental prediction for phonological dyslexia. Damage to the phonological network resulted in an *increasing* discrepancy between nonword and exception word reading over time as the impaired model settled into a "strategy" of reading based on item-specific information. Study 4 tests a final differential prediction of the two models by asking whether surface dyslexia is best characterized as a reading delay. According to the dual-route view, surface dyslexia results from pathological performance of the lexical route, and children with surface dyslexia should exhibit poorer lexical-orthographic skill than even younger, typically developing readers matched on overall reading ability. According to the HS model, surface dyslexia results from a general delay in the process of reading acquisition, and so children with surface dyslexia should perform similarly to reading-level controls.

In addition to these studies testing differential predictions of the models, the current investigation includes two further studies addressing questions relevant to both models. The DRC and HS model both assume that phonological and surface dyslexia result from qualitatively different underlying impairments. Study 5 uses a multiple-groups path analysis to test this assumption, thus evaluating the validity of these developmental dyslexia subtypes. Finally, Study 6 explored individual differences in reading skills among typically developing children. We tested whether controls with unbalanced reading profiles would show subthreshold forms of phonological or surface dyslexia, or alternatively, whether they appeared to support normal word reading by compensating for a deficit in lexical or nonlexical processing.

Study 1

The primary purpose of Study 1 was to identify individuals meeting criteria for phonological or surface dyslexia subtypes using a sample from the Colorado Learning Disabilities Research Center (CLDRC) that is much larger than those used in other regression-outlier studies. The dyslexic sample in the current study is more than 6 times larger than the largest dyslexic sample employed in a regression-outlier subtyping study to date. Identifying greater numbers of individuals meeting one subtype or the other will provide for more powerful tests of the predictions generated by the models of single word reading.

A second advantage of conducting a subtyping study within the CLDRC is that the measures employed have been selected to have high reliability (typically 0.80 or higher) and validity, and participants complete multiple measures of the relevant constructs. Previous subtyping studies have typically estimated nonlexical-phonological and lexical-orthographic abilities using a single measure, sometimes with as few as 30 stimuli. By creating composites based on multiple measures, the current study offers increased reliability in identification of dyslexic subtypes.

Method

Participants—Participants in this and all subsequent studies represented a subsample of the CLDRC. This population-based sample of twins and their families has been accumulated since 1982 from 27 school districts across the state of Colorado. First, school records are used to identify all twin pairs between the ages of 8 and 18 years. Then, those twin pairs in which at least one of the twins has a history of reading and/or attentional problems are invited to the laboratory to undergo an extensive battery of psychometric tests. A comparison group of twin pairs in which neither member has a history of reading or attentional problems is selected as controls and tested on the same battery of tests. Current analyses included participants who completed testing between October of 1990 and

February of 2006. Exclusionary criteria for both the control and the dyslexic groups included having low verbal and performance IQ on the Wechsler Intelligence Scale (i.e., both below 85), obvious neurological or sensory deficits, and a first language other than English. Because ceiling effects were evident among older adolescents for some of the measures assessing reading component skills, the current investigation only included individuals ages 8 to 13.

The criteria for dyslexia was determined with a discriminant function analysis developed by DeFries (1985) using separate samples of nontwin individuals with and without a history of significant reading problems. The discriminant function produced an age-corrected reading composite score based on the Reading Recognition, Spelling, and Reading Comprehension subtests of the Peabody Individual Achievement Test (PIAT; Dunn & Markwardt, 1970). Any participants in the dyslexic group who did not meet criteria based on the discriminant function were removed from the current study, as were any participants in the control group who did meet dyslexia criteria based on the discriminant function. When both members of a twin pair met criteria for either the control or dyslexic group, one was chosen at random for the current study. The final groups included 319 controls and 437 children with dyslexia. Demographic statistics for the two groups are reported in Table 1. The groups were of very similar mean ages ($d = 0.06$). The proportion of males was slightly higher in the dyslexic group ($\phi = .10$), which is expected in a population-based sample (Rutter et al., 2004). On average, mothers of control participants had completed approximately one additional year of education than mothers of participants with dyslexia ($d = 0.57$). This discrepancy likely reflects the facts that dyslexia is familial (Pennington & Olson, 2005) and associated with lower levels of educational achievement (Boetsch, Green, & Pennington, 1996; Vogel & Holt, 2003). There was not a significant group difference in race.

Procedure—Participants were tested individually, in a quiet room, at the University of Colorado at Boulder. They completed a large battery of cognitive, language, and literacy tests. A subset of measures from the battery was used in the current analyses.

Measures

Subtyping measures—Phonological coding (PC, equivalent to nonlexical route function in a dual-route architecture) was assessed with a nonword reading test (Olson, Forsberg, Wise, & Rack, 1994; Olson, Wise, Connors, Rack, & Fulker, 1989) and a phonological choice test (Olson et al., 1994). The nonword reading test was presented in two blocks and included 85 items of varying difficulty levels (e.g., strale, lobsel). The phonological choice task consisted of 60 items requiring participants to select which of three printed nonwords would sound like a real word (e.g., *beal*/*bair*/*rabe*).

Orthographic coding (OC, equivalent to lexical route function in a dual-route architecture) was assessed with an orthographic choice test (Olson et al., 1994) and a homophone choice test (Olson, Forsberg, & Wise, 1994). The orthographic choice test included 80 real word/pseudohomophone pairs (e.g., *easy-eazy*, *fue-few*, *salmon-sammon*) presented in two blocks. The Homophone Choice task required participants to select which of two homophones presented on the computer screen answered a question asked orally by the computer ("Which is a flower?" *rose*/*rows*). There were 65 items.

A subset of participants in the current study (n = 213 with dyslexia and 178 controls) also completed an exception word reading test that included 30 irregularly spelled words of varying difficulty (e.g., *island*, *choir*) (Castles & Coltheart, 1993).

Single word recognition measures—Oral single word reading skill was measured with the PIAT Reading Recognition subtest and the Time Limited Word Recognition Test

(TLWRT; Olson et al., 1994). The TLWRT includes 182 words of increasing difficulty. Basal levels are set using screening items and the test is discontinued once subjects have failed to initiate a correct response within 2 seconds on 10 of the last 20 items (or when the end of the list is reached). Performance on each test was converted to an age-corrected, standardized z-score by regressing on age and age squared for the control participants. The two z-scores were then averaged to create a single word reading composite score.

Cognitive correlates of reading—Participants completed two measures of phonological awareness (PA): phoneme deletion (Olson et al., 1994) and pig Latin (Olson et al., 1989). The phoneme deletion task consisted of 6 practice and 40 test trials, and required subjects to repeat a nonword, then remove a specific phoneme (when done correctly, a real word resulted—e.g., "Say 'prot.' Now say 'prot' without the '/r/'.") The pig Latin task required participants to take the first sound from the front of a word, put it at the end, and add the sound /ay/. For example, "boat" would become "oat-bay". The child received 5 demonstrated examples, 9 practice trials with feedback, and 45 experimental trials with no feedback. All words were within the listening vocabulary of elementary-school aged children. Performance on each PA test was converted to an age-corrected, standardized z-score by regressing on age and age squared for the control participants. The two z-scores were then averaged to create a PA composite score.

Phonological memory was measured with a nonword repetition task including items of varying number of syllables and phonemic complexity. The 40 items described by Gathercole, Willis, Baddeley, & Emslie (1994) were included, as well as 40 more complex items designed to avoid ceiling effects with older participants. The 80 trials are presented without feedback. The task was presented using a CD player. A response period of 2 seconds was allowed between trials. Responses are scored for phonemic accuracy off line, and raw scores were converted to an age-corrected, standardized z-score by regressing on age and age squared for the control participants.

Rapid naming (RN) was assessed with the Colors and Pictures subtest of the rapid naming paradigm (Denckla & Rudel, 1976). Participants were presented with a series of stimuli (either colored circles or pictures of objects) and then asked to identify the items orally as quickly as possible. Each subtest has a maximum time allowance of 15 seconds. Performance on each RN test was converted to an age-corrected, standardized z-score by regressing on age and age squared for the control participants. The two z-scores were then averaged to create an RN composite score.

Vocabulary knowledge was measured with the Vocabulary subtest from the Wechsler Intelligence Scale for Children--Revised (Wechsler, 1974). For primary analyses, we created age-corrected standardized scores by regressing raw scores on age and age squared for the control participants.

Print exposure was measured by presenting children with the names of books, some of which were real and some of which were not, and asking them to identify those that they knew. Raw scores (percent hits-percent misses) were converted into standardized z-scores by regressing on age and age squared for the control participants.

Results and Discussion

Percent correct scores on each of the six primary subtyping measures (each block of pseudoword reading, phonological choice, each block of orthographic choice, and homophone choice) were converted to z-scores based on the means and standard deviations for all participants in the current study. Z-scores on the relevant measures were averaged to create PC and OC composites. Standardized, age-corrected scores were created by regressing these composites on age and age-squared for the control group. The mean age-corrected PC score for the dyslexic sample was −2.46 and the mean age-corrected OC score was −1.89, making it clear that the dyslexic group as a whole showed substantial impairment in both component processes. A deficit in either PC or OC was defined as a standardized residual of −1.5 or less, a cutoff that identifies approximately 7% of the control group. By this criterion, 80.1% of children with dyslexia had a PC deficit and 58.6% had an OC deficit.

Following Castles and Coltheart (1993), individuals meeting criteria for "pure" phonological or surface dyslexia were identified. In that study, all individuals who met criteria for a deficit in one process but not in the other were included in the pure subgroups. However, because this procedure can identify individuals who show only minimal differences between the two processes (e.g., standardized residuals of −1.51 versus −1.49) a slightly more stringent criterion was adopted in the current study. Pure phonological dyslexia was defined as a deficit in PC (≤ −1.5) and an OC score within one standard deviation of the control group mean (i.e., ≥ −1). Similarly, pure surface dyslexia was defined as a deficit in OC and an age-corrected PC score within one standard deviation of the control group mean. By these criteria, 15.6% of the dyslexic sample (n=68) exhibited a pure phonological pattern, and just 2.3% (n=10) exhibited a pure surface pattern. In addition, 12.4% of the dyslexic sample did not meet criteria for a deficit in either PC or OC. This group, which has not usually been clearly delineated in previous studies, was called the "mild" subgroup based on the assumption that relatively spared PC and OC skills would relate to less pronounced real-world reading difficulty.

Individuals meeting a relative phonological, relative surface, or mixed dyslexia pattern were then identified among the remaining dyslexic participants. First, the (not age-corrected) PC composite was regressed on the (not age-corrected) OC composite for control subjects, and the resulting regression equation was used to compute a standardized residual for all participants in the study. Low scores on this residual indicated poorer PC than expected given an individual's own OC abilities. Children with dyslexia who had scores of −1.5 or less were identified as having relative phonological dyslexia. Similarly, the OC composite was regressed on the PC composite for control subjects, and the equation used to calculate a standardized residual that indexed OC ability relative to PC ability. Participants with dyslexia who had scores of −1.5 or less were identified as having relative surface dyslexia. In the current study, 41.0% of children with dyslexia (n=179) exhibited a relative phonological pattern and 7.8% (n=34) a relative surface pattern. A mixed dyslexic subgroup was also identified in two ways. First there were children with dyslexia who did not meet criteria for any of the other dyslexic subtypes. Their age-corrected performance was at least 1 SD below the control group mean for both OC and PC, as well as at least 1.5 SD below the control group mean on at least one of the variables. Further, they did not meet regression-outlier criteria for either relative phonological or surface dyslexia, indicating they were not differentially impaired at either process. In addition, some individuals met criteria for *both* relative phonological and surface dyslexia and were included in the mixed subgroup. Though this seems paradoxical, it can occur when performance on both word types is extremely poor, since the predicted scores regress to the mean. Altogether, 21.1% of children with dyslexia (n=92) were included in the mixed group.

Overall, 56.6% of children with dyslexia met criteria for either pure or relative phonological dyslexia and 10.1% for pure or relative surface dyslexia, with 33.3% not fitting either subtype pattern (i.e., they were either mild or mixed). The prevalence of dyslexia subtypes reported by earlier studies using the regression-outlier approach has varied substantially, perhaps due to methodological differences (e.g., different age ranges, different defining measures, slightly different regression cut-offs). Compared to previous studies of English speakers, current prevalence estimates are at the high end of the range for phonological dyslexia and at the low end of the range for surface dyslexia. It is possible that the exclusion of children with low verbal IQ related to lower rates of surface dyslexia, although some previous studies have excluded children on a similar basis (Stanovich et al., 1997; Manis & Bailey, 2008). Alternatively, lower rates of surface dyslexia may have arisen from more reliable measurement of OC. Supporting this idea is that fact that surface dyslexia has been found to be less stable longitudinally than phonological dyslexia (Manis & Bailey, 2008).

Table 2 summarizes the breakdown of the dyslexic sample into the six possible subtypes including demographic information as well as performance on the subtyping measures, single word reading composite, and cognitive correlates of reading. We conducted a series of one-way ANOVAs with follow-up Tukey post-hoc tests to compare subgroups. Although the sample sizes for the subgroups were quite different, variances were generally similar. Gender and ethnicity ratios were compared with chi-square analyses. Ages were not equivalent across groups (F (5, 431) = 10.91, $p < .001$; η_p^2=.11); in general, children meeting a relative dyslexia pattern were younger, on average, than children meeting criteria for the other subtype patterns. This finding is relevant to the differential predictions generated by the models of single word reading and will be explored more fully in Study 3. Age differences among subtypes have not been reported in previous studies. Castles & Coltheart (1993) did not report ages by subtypes, and Stanovich et al. (1997) studied a much narrower age range than used in the current study. Manis et al. (1996) found no age differences by subtype in a sample that spanned a wide age range, but used a much smaller sample size.

Gender ratios also differed across subtypes ($\chi^2(5)$ = 19.23, p = .002, ϕ = .21). The pure phonological group included more girls than boys, while all other subgroups included more boys than girls. Gender differences have not typically been reported in this literature, and thus, this finding should be replicated by future studies. There were no significant differences for maternal years of education or race. We investigated whether cohort effects influenced subtype membership, which might arise due to changes in reading instruction between 1990 and 2006. Our impression is that over this period, the emphasis on phonics instruction increased in Colorado public schools, which could conceivably lead to lower rates of phonological dyslexia. However, there was no evidence for subgroup differences in date of testing ($p > .3$; η_p^2=.11).

As expected, there were subtype differences for both the PC (F (5, 431) = 169.11, $p<.001$; η_p^2=.66) and OC age-corrected composites (F (5, 431) = 127.05, $p<.001$; η_p^2=.60). By definition, phonological dyslexics had weaker PC scores while surface dyslexics had weaker OC scores. Further, pure and mild subtypes had milder deficits than relative subtypes (see Study 2 for a further exploration of pure versus relative phonological dyslexia). Differences in component process scores related to subgroup differences on the single word recognition composite (F (5,431) = 27.13, $p < .001$; η_p^2=.24) and on the exception word reading task (F (5, 207) = 12.40, $p < .001$; η_p^2=.23). Again, mild and pure subtypes tended to have a less pronounced deficit than relative subtypes. There was not evidence for a difference in severity of reading problem between phonological and surface dyslexics, even on the exception word reading task. In fact, across the full set of participants in the current investigation, exception word reading correlated similarly with the PC and OC composites (r-values of .77 and .75, respectively; both p-values < .001). The magnitude of the correlation between PC and OC was also similar (r = 0.70, $p < .001$). This unpredicted finding is more consistent with the HS model than the DRC, since it suggests that the various reading tasks may tap similar underlying mechanisms.

There were also subgroup differences on most cognitive correlates of reading, including PA (F (5, 424) = 38.17, $p < .001$; η_p^2=.31), PM (F (5, 429) = 4.75, $p < .001$; η_p^2=.05), and RN

($F(5, 431) = 2.69$, $p = .021$; $\eta_p^2=.03$). Following considerable previous research (Bowey & Rutherford, 2007; Castles et al. 1999; Sprenger-Charolles et al., 2011), individuals with phonological dyslexia demonstrated poorer PA than individuals with surface dyslexia. Further, consistent with the predictions of the HS model, relative phonological dyslexia was associated with a larger PA deficit than was pure phonological dyslexia. Subgroup differences in PM were smaller, and Tukey post-hoc tests revealed few meaningful differences. These findings are discussed further in Study 2. For RN tasks, the pattern of means suggested that individuals with surface dyslexia performed more poorly than did individuals with phonological dyslexia, but post-hoc tests did not confirm this result. However, a follow-up independent-samples t-test comparing all phonological dyslexics (pure or relative) to all surface dyslexics on the RN composite was statistically significant, with a moderate effect size ($t(289) = 2.48$, $p = .014$; $d = 0.41$). Again, although the sample sizes were unequal, the variances were very similar for the two groups. The suggestive evidence for an association between surface dyslexia and RN deficits echoes earlier findings of a relationship between RN and OC skills in typical reading development (Manis, Seidenberg, & Doi, 1999) and raises the possibility of overlap between different subtyping schemes for developmental dyslexia (i.e., phonological/surface/mixed versus phonological awareness/rapid naming/double deficit of Wolf and colleagues (Wolf & Bowers, 1999)). There was no evidence for subgroup differences in vocabulary ($p > .3$; $\eta_p^2=.01$). Although evidence highlights a link between semantic skill and OC in acquired dyslexia (Woollams et al., 2007) and across the full range of individual differences (Nation & Cocksey, 2009), this issue has been less explored with regard to phonological/surface dyslexia subtypes. No subtype differences in print exposure were detected ($p > .05$; $\eta_p^2=.02$). Some researchers have proposed that reduced print exposure would lead to a surface dyslexia profile (e.g., Stanovich et al., 1997), but direct empirical evidence on this issue has been limited (though see Castles et al., 1999).

Study 2

The primary purpose of Study 2 is to test the severity hypothesis of pure versus relative phonological dyslexia generated by the HS model. This hypothesis holds that pure phonological dyslexia results from a mild phonological deficit and relative phonological dyslexia from a moderate to severe phonological deficit. So, on average, pure cases should have milder PC impairments as well as milder difficulties on other measures of phonological processing, such as phonological awareness (PA) and phonological memory (PM). The strong version of this severity hypothesis is that all pure phonological dyslexics should exhibit relatively mild phonological impairments; in other words, there should be no children with severe pure phonological dyslexia. Because dual-route proponents acknowledge that PC and OC skill are correlated and argue that the nonlexical and lexical routes interact in development, this approach does not generate a competing prediction for group mean differences in severity of the phonological deficit for pure versus relative phonological dyslexics. However, the dual-route approach does hold that there should be some individuals with a severely impaired nonlexical route and a fully functional lexical route.

One previous study reported a small number of individuals with severe pure phonological dyslexia (Castles et al., 2006). If this result is replicated in the current data set, the presence of this pattern must be explained by something other than a strict severity hypothesis. The dual-route framework would offer one possible explanation. A second alternative would be that some protective factor allows these individuals to partly compensate for the severity of the phonological deficit. We included measures of both vocabulary knowledge and rapid naming, since previous research has identified these as important risk/protective factors that interact with phonological deficits in the development of word reading problems (Bishop et al., 2009; Snowling et al., 2007). We also measured print exposure and maternal education, which may capture environmental effects that have been hypothesized to relate differentially to OC (Castles et al., 1999; Jimenez et al., 2009).

Method

Participants—All participants meeting criteria for pure or relative phonological dyslexia were included in the current study (total n = 247).

Measures—Measures of PC, OC, word recognition, PA, PM, RN, vocabulary and print exposure described in Study 1 were used.

Results

Means, standard deviations, and statistical comparisons (Tukey post-hoc tests) for the pure and relative phonological dyslexia groups for OC, PC, word recognition PA, PM, RN, vocabulary, and print exposure are shown in Table 2. Effect sizes (Cohen's d) were as follows: PC: 1.12; OC: 2.43; word recognition: 0.96; PA: 0.63; PM: −0.14; RN: 0.17; vocabulary: 0.23; print exposure: 0.33. By definition, the relative group performed more poorly on the OC composite than the pure group. Evidence for a larger phonological deficit in the relative than pure group was found on the PC and PA composites, though this did not hold for PM. Not surprisingly, greater difficulty on both OC and PC related to poorer single word reading for the relative as compared to the pure phonological dyslexics. There were no significant differences for the pure and relative subgroups on vocabulary, RN, print exposure, or maternal education.

To test the strong version of the severity hypothesis, we next examined individual scores on the age-corrected PC composite. 82.4% of individuals with pure phonological dyslexia (n = 56) had a mild PC deficit in that their score was better than the mean of the relative phonological dyslexic group. However, 17.6% (n = 12) of pure cases had a moderate to severe PC deficit, defined as a score below the relative phonological subgroup mean. These 12 individuals can be considered to have "severe pure phonological dyslexia." Table 3 summarizes demographic information and the PC, OC, word recognition, PA, PM, RN, vocabulary, and print exposure scores for these 12 individuals in comparison to the relative phonological dyslexia group means. There were more girls than boys in this group, consistent with findings in the pure phonological group as a whole, and a wide age range was represented.

Further evidence for a large phonological deficit came from the finding that all 12 individuals performed below the mean of the group with relative phonological dyslexia for either PA or PM, with 8 individuals being below the relative phonological mean on both these variables. Five of the 12 cases performed even more poorly than the mean of the group with relative phonological dyslexia on the single word reading composite. So, for nearly half of the individuals with severe pure phonological dyslexia, a severe PC deficit related to very poor single word reading despite relatively spared OC. There was evidence that vocabulary and RN serve as important additional risk/protective factors in determining reading outcome for this group. Of the 5 individuals with severe pure phonological dyslexia whose single word reading was worse than the relative phonological dyslexia mean, all 5 also had a worse vocabulary score and 4 had a worse RN score. Conversely, of the 7 individuals with severe pure phonological dyslexia whose single word reading was better than the relative phonological dyslexia mean, 6 also had a better vocabulary score and 6 had a better RN score. Nine of 12 individuals with severe pure phonological dyslexia had lower levels of maternal education or print exposure than the group with relative phonological dyslexia.

Given the small number of individuals in the severe pure phonological group, one possibility is that the pattern can be accounted for by measurement error. Perhaps the true OC abilities of these individuals are actually poorer than their scores suggest. Fortunately, the twin design of the CLDRC allows a means of addressing this issue. Since monozygotic (MZ) twins share all of their genes and much of their environment, their scores on cognitive and literacy tests should be quite similar, provided the measures are reliable. Six of the 12 children with severe pure phonological dyslexia had an MZ twin in the CLDRC sample. These twins had been removed from current analyses, but their scores on the relevant measures are displayed in Table 3. Of the six severe pure phonological dyslexia cases with an MZ twin, two of their co-twins (IDs 9a and 10a) could also be categorized as having severe pure phonological dyslexia. An additional two co-twins (IDs 7a and 11a) showed a very similar pattern, but just missed the cut-off to be classified as having severe pure phonological dyslexia on either the OC or PC composite. A fifth co-twin (12a) met criteria for pure phonological dyslexia, but the PC deficit was not severe. There was only one twin pair for whom the OC composite was quite different across the two members (IDs 8 and 8a). Overall, the existence of a small number of children with severe pure phonological dyslexia did not appear to be due entirely to measurement error.

Discussion

Study 2 compared the size of the phonological deficit in children with pure versus relative phonological dyslexia. The HS model predicts that relative phonological dyslexia should be associated with a larger PC deficit than pure phonological dyslexia. The weak version of this prediction relates to group mean differences, while the strong version extends to all individual cases.

As predicted by the HS model, children with relative phonological dyslexia had more severe PC and PA deficits, on average, than children with pure phonological dyslexia. However, the more severe phonological deficit did not extend to nonword repetition. The relationship between PA and reading is bidirectional (Morais et al., 1979; Castles et al., 2011), and over time, poor reading can cause poor PA. This reciprocal relationship has been less clearly established for PM (though see Nation & Hulme 2011). Thus, it may be that relative phonological dyslexia is associated with greater PC deficits, but not necessarily greater deficits in underlying phonological representations. This explanation is more consistent with the DRC account of phonological dyslexia (deficit grapheme-phoneme conversion) than with the HS account (damage to the phonological network). Alternatively, given that PM was measured with a single variable, the current pattern of results may simply reflect lower reliability for nonword repetition than for the PA or PC composite.

The strong version of the HS prediction was not confirmed. Consistent with previous research (Castles et al., 2006), we identified a small number of individuals with relatively spared OC skill despite a severe PC deficit. The existence of these individuals is problematic for the HS model, particularly because an examination of MZ twin scores indicated that the pattern could not be explained away by measurement error. At first, this pattern might appear more consistent with a damaged nonlexical route and spared lexical route within a dual-route architecture. However, it is puzzling that the single word reading skill of many individuals in this group should be so poor, since all real words could be effectively handled by a functioning lexical route. Along with poor single word reading, many individuals in this group had low levels of maternal education and exposure to print. This result is somewhat surprising, since the relatively intact OC skills of this group might have been expected to be linked to higher levels of maternal education and print exposure.

Thus, we identified a small number of individuals whose pattern of performance on single word reading and component process measures was problematic for both the HS model and the dual-route approach. There was some evidence that lexical/semantic skills (expressive vocabulary or rapid naming) represented additional risk/protective factors in determining reading outcome for individuals with a severe PC deficit and relatively spared OC. This interpretation is consistent with research documenting associations of both lexical phonology and semantic knowledge to single word reading (Nation & Cocksey, 2009). Although the dual-route approach conceptually includes semantics, semantic knowledge plays only an ancillary role in oral reading in the dual-route framework. In contrast, semantics plays a fundamental role in oral reading in the triangle model, but developmental dyslexia subtypes have not been explicitly modeled in a version of the triangle model with an implemented semantic network (Harm & Seidenberg, 2004). The current results suggest that this model might account for all cases of developmental dyslexia better than either the DRC or the HS model does.

Study 3

The purpose of Study 3 was to investigate the prevalence of pure phonological and surface dyslexia at different ages. One problem for a dual-route view has been to account for the preponderance of mixed cases of developmental dyslexia (including relative phonological and surface dyslexia). If reading component processes are achieved by separable cognitive mechanisms, why would so many individuals with dyslexia exhibit deficits in both? According to proponents of a dual-route architecture, isolated deficits in one route (i.e., pure cases) should be more common early in literacy acquisition (Castles et al., 2006). These authors have argued that the acquisition of each route impacts the other, and so a single deficit may lead to secondary deficits in the other route. Therefore, the prevalence of pure cases should diminish with age.

The HS model offers a different explanation of the preponderance of mixed cases, at least for relative phonological dyslexia. According to that viewpoint, a mild deficit in phonological representations leads to an impairment in PC only, and a moderate to severe deficit in phonological representations leads to impairments in both PC and OC. Study 2 tested this severity hypothesis of pure versus relative phonological dyslexia. A prediction that follows is that pure phonological dyslexia should be found at older as well as younger ages, provided the underlying impairment is mild. In fact, results from the HS simulations suggest that pure phonological dyslexia should become more common with development. Specifically, models with phonological deficits exhibited an increasing discrepancy between nonword and exception word reading over training, as they settled into a “strategy” of reading based on item-specific information. The HS model did not simulate pure surface dyslexia, and so makes no prediction about its developmental course.

Method

Participants—The current study included all participants with pure or relative phonological or surface dyslexia (total n = 291).

Results

First, the sample was divided into age bands of similar n. Because younger ages are more heavily represented in the CLDRC, age bands were wider at older ages. Though the widths of the age bands were established based on practical reasons, it also makes theoretical sense to have a narrower range at younger ages, when there is more rapid developmental change in single word reading ability. The age bands included were: 8.0–8.4 years (n = 54), 8.5–8.9 years (n = 36), 9.0–9.4 years (n = 47), 9.5–9.9 years (n = 42), 10.0–10.9 years (n = 40), 11.0–11.9 years (n = 38), and 12.0–13.9 years (n = 34). For each age band, we calculated the prevalence of pure phonological dyslexia (as a function of all cases of phonological dyslexia) and pure surface dyslexia (as a function of all cases of surface dyslexia) (see Table 4).

Discussion

This study tested the prediction of dual-route proponents that pure cases of phonological and surface dyslexia should be more common early in literacy development, before a deficit in one route has had time to impair acquisition of the other route. There was no evidence to support this prediction. Instead, the prevalence of pure cases increased with age, for both phonological and surface dyslexia. The growing prevalence of pure phonological dyslexia with age was consistent with HS model simulations showing that in the presence of a phonological deficit, the network settles into a strategy of relying more and more heavily on item-specific information.

A limitation of the current study is that it used a cross-sectional, rather than longitudinal, design. Longitudinal research on dyslexia subtypes is quite limited, but has demonstrated that factors such as intervention and type of instruction can lead to changes in subtype identification over time (Olson, 2011). A more recent longitudinal study found that the stability of phonological dyslexia over two years was higher than that of surface dyslexia (Manis & Bailey, 2008). Despite this limitation, the current research makes an important contribution by confirming and extending the pattern of findings across previous studies. In particular, Stanovich et al., (1997) used a younger sample and reported higher rates of mixed cases in comparison to other regression-outlier studies. We provide convergent evidence for this pattern within a single study using identical measures and subtype definitions across age groups.

A second limitation of the current study is that the youngest participants were eight years old and thus had already received approximately two years of formal literacy instruction. Therefore, our results do not rule out the possibility that pure phonological or surface dyslexia is substantially more common in six-and seven-year-old children.

Study 4

The purpose of Study 4 was to investigate whether or not surface dyslexia is best characterized as “reading delay dyslexia.” According to proponents of a dual-route architecture, surface dyslexia results from a differentially impaired lexical route. Therefore, surface dyslexics should demonstrate poorer OC skills than even younger, typically developing children matched for overall reading skill (as long as floor effects are avoided in the comparison group). According to the HS model, surface dyslexia results from a general delay in the process of reading acquisition, such that surface dyslexics should demonstrate similar OC and PC in comparison to reading level (RL) controls.

Previous studies have used two different approaches to evaluate whether or not surface dyslexia is best characterized as a reading delay. First, the performance of individuals with surface dyslexia (identified using the regression-outlier method with chronological age, or CA, controls) has been compared directly to RL controls on measures of PC and OC. Second, the regression-outlier method has been used to identify individuals with surface dyslexia with regard to RL, rather than CA, controls. In other words, the criteria for surface dyslexia depended on the regression equation for the relationship between PC and OC for RL controls. To date, results from both methods have been more consistent with the predictions of the HS model. In direct comparisons, group mean differences have not been statistically significant in studies of English speakers (though see Jimenez et al., 2009, for a positive finding in Spanish-speaking children). With the second subgroup identification method, the prevalence of surface dyslexia has been extremely low (Manis et al., 1996;

Sprenger-Charolles et al., 2000; Stanovich et al., 1997). We report results from both approaches below. The first method represents the more direct and sensitive measure of whether the performance of surface dyslexics mirrors that of younger, typically developing readers. Though previous studies have not found significant group differences using this approach, sample sizes have been relatively small and group means have been in the direction predicted by the dual route approach. The larger sample size of the current study provides increased power to detect significant group differences if they do exist.

Method

Participants—For the first approach, participants included a subset of children with surface dyslexia (pure or relative) and a subset of control participants, selected to be similar in terms of raw score on the PIAT Reading Recognition test. Groups were selected on the basis of chronological age to avoid the statistical pitfalls associated with individually matching on reading skill (Miller & Chapman, 2001). Selecting controls younger than 8.4 years (n = 34; mean age = 8.18 years; mean reading raw score (SD) = 39.53 (5.90)) and children with surface dyslexia older than 9.0 years (n = 23; mean age = 10.76 years; mean reading raw score (SD) = 37.48 (7.52)) produced groups that did not differ in terms of reading level. A group of children with phonological dyslexia (age > 10.5 years; n = 82; mean age = 11.85 years; mean reading raw score (SD) = 37.94 (6.98)) was also chosen to be similar in reading level to both groups. Univariate ANOVAs confirmed that reading level did not differ by group (F<1; η_p^2=.01) but chronological age did (F (2, 136) = 161.97, p <. 001; η_p^2=.73).

For the second approach, subsets of all dyslexic and control participants were selected to be similar in terms of reading level. All participants with dyslexia older than 11.5 years and all control participants younger than 9.0 years provided a good match (dyslexic mean reading raw score (SD) = 42.90 (6.87); control mean reading raw score (SD) = 43.23 (6.47); p >.7). This procedure resulted in 104 participants with dyslexia and 74 controls.

Measures—Measures of PC, OC, word recognition and exception word reading described in Study 1 were used.

Results

First, we compared the performance of participants with surface or phonological dyslexia directly to RL controls. A repeated-measures ANOVA was conducted with within-subjects factor of reading component process composite raw score (PC, OC) and between-subjects factor of group (surface dyslexia, phonological dyslexia, RL control). There was a main effect of component process (F (2, 136) = 10.97, p < .001; η_p^2=.08) reflecting the fact that collapsed across group, scores were slightly lower for the PC than the OC composite. There was also a main effect of group (F (2, 136) = 20.46, p = .008; η_p^2=.07). Although the three groups were quite similar in terms of real word reading level, there were small group differences on the scores collapsed across component process (children with surface dyslexia: −0.41; children with phonological dyslexia: −0.29; RL controls: 0.00). Most importantly, there was a large component process by group interaction (F (2, 136) = 142.56, p < .001; η_p^2=.68) reflecting the fact that each of the three groups showed a different pattern of performance on the two tasks. The interaction remained significant when just the participants with surface dyslexia were compared to controls (F (1, 55) = 21.00, p < .001; η_p^2=.28), as well as when just the participants with phonological dyslexia were compared to controls (F (1, 114) = 99.02, p < .001; η_p^2=.47). Follow-up t-tests indicated that the participants with surface dyslexia performed significantly more poorly than RL controls on the OC composite (t (55) = 4.83, p < .001; *d* = 1.30) but not the PC composite (p > .8; *d* = 0.06). In contrast, participants with phonological dyslexia performed more poorly than RL controls on the PC composite (t (43.4) = 5.81, p < .001; *d* = 1.48), and *better* than RL controls on the OC composite (t (114) = 2.07, p = .04; *d* = −0.42). Results are shown in Figure 1.

The critical question for discriminating between the two models concerns the performance of participants with surface dyslexia relative to RL controls on measures of lexical route function. Thus, we also used a t-test to compare raw scores of these two groups on the exception word reading task. Of the children selected for this study, this task had been administered to 12 of those with surface dyslexia and 25 RL controls. Consistent with results from the OC composite, children with surface dyslexia performed significantly more poorly than RL controls (t (35) = 2.21, p = .03; *d* = 0.81).

We followed these analyses with a second approach that identified participants with dyslexia who fit the phonological or surface pattern relative to RL, rather than CA, controls. By definition, this approach can only identify individuals fitting a relative (not pure) phonological or surface pattern. Regression equations predicting PC from OC and vice versa were computed in the control participants selected as RL controls for a subset of all participants with dyslexia. These regression equations were then used to identify children with phonological dyslexia (worse PC than predicted by OC) and surface dyslexia (worse OC than predicted by PC) as described in Study 1. 26.0% (n=27) of the subgroup of participants with dyslexia (age > 11.5 years) exhibited a pattern of phonological dyslexia, just 1.9% (n = 2) of the subgroup exhibited a pattern of surface dyslexia, and the remaining 72.1% (n = 75) of the sample did not meet criteria for either subtype.

Discussion

This study used two approaches to evaluate whether either surface or phonological dyslexia is well described as a reading delay. Proponents of the HS model have argued that only phonological dyslexia represents developmental deviance, with surface dyslexia representing a developmental delay. Proponents of the dual-route approach argue that both subtypes reflect qualitatively abnormal reading development. Most previous studies supported a reading delay account of surface dyslexia and reported that surface dyslexics performed comparably to RL controls on a variety of word reading tasks. Further, when subtypes were identified relative to RL controls, the surface dyslexia pattern all but disappeared.

The current study produced somewhat different results. When compared directly to RL controls, individuals with both surface and phonological dyslexia showed an atypical pattern of performance, with selective impairment in their defining component process. Children with surface dyslexia also underperformed RL controls on an exception word reading task. Though the difference in pattern of performance compared to RL controls was more striking for children with phonological dyslexia, it was statistically significant for both groups. The difference between the current result and those of previous studies appears to be at least partly due to the increased power offered by the increased sample size. To evaluate a reading delay account of surface dyslexia, the critical comparison is between individuals with surface dyslexia and RL controls on the subtype-defining measure (i.e., exception word reading, orthographic coding). The current study found that participants with surface dyslexia performed significantly more poorly, with a large effect size. As described more fully below, previous studies of English speakers have also found medium to large effect sizes for this comparison, but with smaller sample sizes, the difference has not been statistically significant.

Manis et al. (1996) found that individuals with surface dyslexia did not differ significantly from RL controls on the subtype defining measures of exception word reading or nonword reading. However, numerically, individuals with surface dyslexia performed worse than RL controls at exception word reading (*d* = 0.74). These authors also administered an orthographic choice task similar to the one included in the current OC composite, not to define subtypes but as a validation task. Again, although the difference in performance was not statistically significant, RL controls performed better than participants with surface dyslexia (*d* = 0.52). There was essentially no mean difference between the two groups on a validation task designed to tap phonological processing (*d* = 0.04). Stanovich et al. (1997) reported that children with surface dyslexia did not differ significantly from RL controls on the defining measures of exception word reading and nonword reading. However, children with surface dyslexia obtained numerically lower exception word reading scores (*d* = 0.57) yet numerically higher nonword reading scores than RL controls (*d* = 0.67).

Cross-linguistic results provide further evidence against the reading delay account. Spanish-speaking children with surface dyslexia performed significantly more poorly than RL controls on a homophone comprehension task (Jimenez et al, 2009; *d*=1.61). Finally, Sprenger-Charolles et al. (2000) reported that French-speaking children with surface dyslexia did not differ from RL controls on irregular word reading time or accuracy, though again the means were in the predicted direction (*d*-values of 0.32 and 0.26, respectively). Unexpectedly, children with surface dyslexia also performed significantly more poorly than RL controls on nonword reading accuracy. This result may have related to a speed-accuracy trade-off, since children with surface dyslexia obtained lower nonword accuracy scores than even children with phonological dyslexia, but read the nonwords significantly faster.

Given the pattern of results across studies, we speculate that meta-analysis would confirm that when children with dyslexia are selected for differentially poor lexical/orthographic processing (e.g., exception word reading) relative to same-age peers, they also display poorer performance on the defining measure than do RL controls. Such a finding would be consistent with current results.

Using a second approach to test the reading delay account of surface dyslexia, previous studies reported that the surface dyslexic pattern essentially disappeared when the regression equation was computed for RL controls (Manis et al., 1996: 1 case; Sprenger-Charolles et al., 2000: 3 cases; Stanovich et al., 1997: 1 case; Stanovich et al. reanalysis of Castles and Coltheart, 1993 data: 2 cases). This finding was mirrored in the current study, with just two out of 104 children meeting criteria for surface dyslexia. The somewhat contradictory results from the two approaches probably relate to differences in the effect size required to find a positive result. In order to meet criteria for surface dyslexia under the regression-outlier method, participants needed to have exception word reading or orthographic coding scores at least 1.5 SD below that predicted by their nonword reading. As the above discussion makes clear, obtained effect sizes for mean differences between children with surface dyslexia and RL controls have generally been smaller than that.

In summary, results from the current study are more consistent with the dual-route approach than the HS model in providing evidence that surface dyslexia cannot be accounted for solely by a reading delay. The failure of previous investigations to find differences between participants with surface dyslexia and RL controls was likely due at least in part to insufficient power. Current results agree with previous studies in demonstrating that the performance of phonological dyslexics represents a pattern of developmental deviance (Rack, Snowling, & Olson, 1992), probably to an even greater degree than that of surface dyslexics, but it was clear that neither subtype performed identically to younger, typically developing readers.

Study 5

Studies 2 through 4 tested differential predictions about dyslexia subtypes generated by the DRC and HS models. This study tests an assumption that is shared by both models.

According to the DRC, phonological dyslexia is caused by a differentially impaired nonlexical route and surface dyslexia by a differentially impaired lexical route. According to the HS model, phonological dyslexia is caused by impaired phonological representations and surface dyslexia by a general reading delay. Though the two approaches disagree about the nature of the underlying impairments, they agree that qualitatively different problems cause the word reading deficit in the two subtypes. According to both views, the severity of the underlying impairment should determine the severity of the real-word reading problem. For example, the HS model with mild damage to phonological representations could read more words than could the model with moderate damage to phonological representations. Thus, both models posit that members of each subgroup are impaired at real word reading primarily because of difficulties with one component skill or the other (PC or OC). Because substantial individual differences in reading skill exist within each subgroup, a prediction that follows is that the PC composite should be the stronger predictor of reading for children with phonological dyslexia, while the OC composite should be the stronger predictor for children with surface dyslexia. This prediction was tested using multiple-group path analysis, with the two component skills as IVs and the single word reading composite as the DV. The invariance of path weights was tested across children with phonological dyslexia, surface dyslexia and controls.

In addition to testing this assumption that is shared across models, we conducted more exploratory analyses to investigate factors that may underlie PC and OC deficits and thus cause poor reading for each subgroup. Specifically, in multiple-group path analyses, we evaluated whether phonological skills (PA or PM) differentially relate to word reading in phonological dyslexia, while print exposure differentially relates to word reading in surface dyslexia. Such findings would be consistent with the HS model, since PA or PM provide a means of estimating integrity of underlying phonological representations, and one form of reading delay could result from reduced exposure to print. However, these analyses will not necessarily help discriminate between the models, because the DRC is silent as to distal causes of OC or PC dysfunction.

Method

Participants—All participants meeting criteria for phonological or surface dyslexia, as well as all controls, were included in the current study (total n = 716).

Measures—Measures of PC, OC, word recognition, PA, PM, RN, and print exposure described in Study 1 were used.

Results

First, we examined variances of the relevant variables by group. Since some of the variables were used to define the subtypes, it was possible that groups could show restricted variance that would influence the results. There were some differences in variance for participants with phonological versus surface dyslexia on the PC composite (phonological dyslexia SD: 0.69; surface dyslexia SD: 0.97; Levene's $F < .05$) and the OC composite (phonological dyslexia SD: 1.08; surface dyslexia SD: 0.84; Levene's $F < .05$). This is a conservative bias, because the reduced variance in each subgroup on the defining measure makes it less likely to find the result predicted by both models—that PC is the stronger predictor of word reading in phonological dyslexia and that OC is the stronger predictor of word reading in surface dyslexia. Variances for all other variables used in this study were comparable across subgroups according to Levene's test.

A simple path model was constructed with the two component process composites predicting the real word reading composite. Path weights by group are shown in Figure 2. The equivalence of path weights across groups was formally tested with multiple-group analyses. Forcing equal path weights across the groups with phonological and surface dyslexia groups significantly degraded model fit ($\chi^2(2) = 12.50$, $p = .002$). Children with phonological dyslexia and controls were well described by similar path weights ($\chi^2(2) = 2.14$, $p > .3$), while children with surface dyslexia differed significantly from controls ($\chi^2(2) = 9.52$, $p = .009$). For both controls and children with phonological dyslexia, PC more strongly predicted word reading than did OC, while children with surface dyslexia showed the opposite pattern.

A second analysis used a similar approach to predict word reading from PA and print exposure. Constraining path weights to be equal across the groups with phonological and surface dyslexia did not significantly degrade model fit ($\chi^2(2) = 1.62$, $p > .4$). However, each dyslexia subgroup showed a significantly different pattern of weights from controls (phonological: $\chi^2(2) = 31.34$, $p < .001$; surface: $\chi^2(2) = 8.90$, $p = .012$). Results by group are shown in Figure 3. Both subgroups with dyslexia showed fairly balanced contributions of PA and print exposure to word reading, while in the control group, the unique contribution of PA was twice as large as that of print exposure.

We conducted two follow-up analyses to better understand these results. First, we substituted PM (nonword repetition) for PA based on the argument that it may provide a more "clean" measure of underlying phonological representations. However, path weights remained comparable across the subgroups with dyslexia. Second, based on results from Study 1 and 2 suggesting an association between OC skills and RN, we substituted the RN composite for the print exposure composite. Again, however, there was not evidence for different path weights across the subgroups with phonological and surface dyslexia.

Discussion

This study tested the shared assumption of the DRC and HS models that phonological and surface dyslexia result from qualitatively different underlying impairments, and that children with phonological dyslexia are poor readers primarily because of PC difficulties, while children with surface dyslexia are poor readers primarily because of OC difficulties. Results of multiple-group path analyses supported this assumption. For children with phonological dyslexia and controls, PC made a large unique contribution to single word reading, while the unique contribution of OC was much smaller. In contrast, for children with surface dyslexia, OC made a large unique contribution to word reading, while PC did not contribute significant unique variance. These results agree with a large body of previous research demonstrating that for the majority of typically developing and dyslexic readers, individual differences in word reading skill relate most strongly to individual differences in PC ability (see Vellutino et al., 2004). However, children with surface dyslexia demonstrated a qualitatively different pattern of performance, supporting the validity of surface dyslexia as a meaningful subtype. The fact that children with surface dyslexia (and not children with phonological dyslexia) showed a different pattern from controls is problematic for claims that only phonological dyslexia represents developmental deviance and that surface dyslexia represents developmental delay.

In a second set of analyses, we investigated the relative contributions of deficits that have been hypothesized to be distal to problems with reading component skills, including PA and print exposure. Despite evidence that the proximal causes of phonological and surface dyslexia (i.e., PC and OC deficits) are at least partly different, this study generated no evidence for differences in the possible underlying causes that we tested. Instead, phonological skills and print exposure both made similar, moderate, unique contributions to real word ability for children with either phonological or surface dyslexia.

Study 6

The final study investigated individual differences in reading-related skills among typically developing children. The regression-outlier method inevitably identifies some controls who meet criteria for a phonological or surface pattern. In fact, there was no indication of heteroskedasticity in the regressions used to identify subtypes, which means that unbalanced word reading profiles are equally likely to occur in both good and poor readers (see also Bowey & Rutherford, 2007; Bryant & Impey, 1986). Previous studies using the regression-outlier method have not described the reading-related performance of typically achieving children with unbalanced word reading profiles. The current study compares the word-reading and reading-related abilities of three groups of controls: those with poorer PC than OC (phonological pattern), those with poorer OC than PC (surface pattern), and those with balanced PC and OC abilities.

Research has demonstrated that that dyslexia exists on a continuum with normal reading (e.g., Shaywitz et al., 1992). Thus, control participants with differentially poor PC or OC may display subthreshold phonological or surface dyslexia. Alternatively, these participants might be able to support fully normal word reading through a protective factor or factors that allows for compensation of the PC or OC deficit. Both computational models acknowledge that regular word reading can be supported in multiple ways, such that different divisions of labor can theoretically give rise to similar overt performance. Different divisions of labor in normal reading have been explicitly modeled by versions of the triangle model with an implemented semantic network (Harm & Seidenberg, 2004; see also Woollams et al., 2007). Thus, a weakness in PC might be compensated for by a strength in semantics. We included measures of several cognitive correlates of reading that might serve as protective factors to allow normal word reading despite PC or OC deficits.

Method

Participants—Participants included the 23 controls who met criteria for a phonological pattern (18 pure and 5 relative) and the 13 controls who met criteria for a surface pattern (11 pure and 2 relative). The term “pattern” is used in this study in place of “dyslexia,” since the controls by definition do not have dyslexia. However, criteria used to identify participants were identical to those described in Study 1. A group of controls with balanced PC and OC abilities was identified for comparison. These controls (n = 54) showed a less than 0.5 SD discrepancy between their observed score on each component process and that predicted by the other component process.

Measures—Measures of PC, OC, word recognition, exception word reading, PA, RN, and vocabulary described in Study 1 were used.

Results

First, the ages of the three groups were compared with a univariate ANOVA, which revealed that the three groups were not equivalent ($F(2, 87) = 3.14$, $p = .049$; $\eta_p^2=.07$). The ages of the phonological and surface pattern controls were similar (9.50 and 9.75 years, respectively), with the balanced controls being slightly older (10.38 years). However, none of the differences were significant in follow-up Tukey post-hoc tests. Thus, while the effect is small, results suggest that among typical readers, an unbalanced word reading profile may be more common earlier in literacy development.

Next, the scores of the three groups on the age-corrected PC, OC, word recognition, exception word reading, spelling, PA, RN, and vocabulary measures were compared with univariate ANOVAs and follow-up Tukey post-hoc tests. Results are summarized in Table 5. By definition, there were group differences on the PC and OC composites. In addition, there were significant group differences for word recognition ($F(2, 87) = 25.42$, $p <.001$; $\eta_p^2=.37$), exception word reading ($F(2, 44) = 5.33$, $p =.008$; $\eta_p^2=.20$), and PA ($F(2, 87) = 26.77$, $p <.001$; $\eta_p^2=.38$), but not RN or vocabulary. In general, phonological pattern controls performed more poorly than balanced controls on all literacy measures and PA. In contrast, surface pattern controls performed comparably to balanced controls on measures of word reading, PC, and PA, and only demonstrated a deficit in the defining measure of OC. Surface pattern controls also tended to underperform balanced controls on exception word reading ($d = 0.93$), though the effect did not reach significance in a Tukey post-hoc test corrected for multiple comparisons.

To further understand the performance of controls exhibiting a phonological or surface pattern, it is useful to consider their performance in comparison to the pure phonological and surface dyslexia subgroups. For ease of reference, scores for these subgroups with dyslexia are reproduced in Table 5. Controls exhibiting a phonological pattern appeared to have a sub-threshold form of phonological dyslexia. Relative to children with pure phonological dyslexia, phonological pattern controls showed smaller deficits on phonological tasks (PC: $d = 0.79$; PA: $d = 0.52$) as well as on tasks emphasizing lexical processes (RN: $d = 0.51$; Vocabulary: 0.75). Perhaps as a result, the average word reading deficit was smaller in phonological pattern controls than in children with pure phonological dyslexia ($d = 1.51$). In contrast, controls exhibiting a surface pattern showed distinctly different performance from children with pure surface dyslexics. The OC skills of surface pattern controls were numerically poorer than those of the dyslexic subgroup ($d = -0.19$), but their word reading skills were markedly better ($d = 1.24$). The surface pattern controls performed better than children with pure surface dyslexia on all cognitive correlates of literacy, including PA ($d = 1.02$), RN ($d = 2.46$), and vocabulary ($d = 1.13$).

Discussion

This study investigated the reading-related abilities of a sample of participants who have been largely ignored by previous research: control participants who display unbalanced profiles in terms of reading component processes. We explored whether these individuals would display sub-threshold phonological or surface dyslexia, or, alternatively, whether they would appear to support fully normal word reading by compensating for a PC or OC deficit.

Results differed for controls with PC versus OC deficits. There was a significant cost to having differentially impaired PC skills: controls exhibiting a phonological pattern essentially demonstrated sub-threshold phonological dyslexia, and were poorer at single word reading than their peers with intact PC abilities. This finding agrees with previous studies documenting that dyslexia exists on a continuum with normal reading and highlights some of the difficulties inherent in searching for discrete subtypes of dyslexia. However, a small subset of controls had differentially impaired OC skills with little apparent cost. Their word reading skills were not significantly different from those of controls exhibiting a balanced pattern. Further, the comparison between this group and the pure surface dyslexic group was striking. On average, the OC skills of the control subgroup were numerically worse, but their word reading abilities were better, as was their performance on measures emphasizing lexical/semantic skills (RN and vocabulary) and phonological skills. One possibility is that a phonological deficit alone is sufficient to impair word reading skill, but an OC deficit is impairing only when it occurs in conjunction with a second deficit in lexical semantics. This explanation is broadly consistent with the notion of different divisions of labor in normal reading, as demonstrated by some implementations of the triangle model (Harm & Seidenberg, 2004; Plaut et al., 1996). In the context of the DRC, the surface pattern controls appear to support normal word reading performance with an intact nonlexical route.

These results can thus be understood in the context of either model, but they also raise some challenges for both models. Both tests contributing to the word recognition composite include some exception words. Thus, in a dual-route framework, the surface pattern group ought to incur at least some cost on these tasks, although it is possible that we did not have sufficient power to detect a small group difference. These results also challenge proponents of the HS model to address why some individuals whose component process profiles reflect what the model conceptualizes as a reading delay are not delayed in terms of real-word reading abilities.

General Discussion

The current investigation identified children fitting a phonological or surface dyslexia profile in a large sample spanning ages 8 to 13 years. We then tested predictions concerning these subtypes of dyslexia arising from two computational models of single word reading, the DRC and HS models. Our results did not clearly support one model over another. Instead, the primary contribution of the current paper may be to identify areas of improvement for both models. Below, we summarize the key findings of this investigation in terms of how they bear on the two models and suggest some directions for future work.

Consistent with the predictions of both models and with substantial previous research, we found subgroups of children meeting criteria for either phonological or surface dyslexia. Given that PC, OC, and word reading are all strongly but less than perfectly correlated, it is essentially inevitable that individuals can be identified as fitting one subtype profile or another. Since the relationships among these variables are generally mutivariate normal, however, the subtypes do not represent discrete categories (Castles et al., 1999; Pennington, Santerre-Lemmon, Rosenberg, MacDonald, Boada, et al., 2012). Nonetheless, we found support for the validity of the distinction between groups of children from two ends of the distribution. Cognitive correlates of reading showed somewhat different patterns for the two subgroups. Further, in a multiple-groups path analysis, children with phonological dyslexia appeared to be poor readers primarily because of PC impairments, while children with surface dyslexia appeared to be poor readers primarily because of OC impairments. Notably, the rate of surface dyslexia (and of pure surface dyslexia in particular) was lower in the current research than in previous regression outlier studies of English-speaking children. This finding may partly reflect our exclusionary criteria, but we believe it may also relate to more reliable measurement of OC skills. This result appears to be more consistent with the HS model, which emphasizes the central role of phonology in word reading, than the DRC, though it is not necessarily problematic for the DRC. Also consistent with the predictions of the HS model, we found that on average, pure phonological dyslexia is associated with a milder phonological impairment than relative phonological dyslexia. The prevalence of pure phonological dyslexia (in comparison to relative phonological dyslexia) increased with age, suggesting that children with phonological impairments may increasingly rely on item-specific information to read as they grow older. This finding is consistent with the HS model and problematic for the DRC.

Consistent with the predictions of the DRC and problematic for the HS model, we found that surface dyslexia is not well characterized as a reading delay and that children with surface dyslexia display poorer lexical/orthographic skills than even younger, typically developing children matched on reading level. Further, in a multiple-groups path analysis examining the unique contributions of PC and OC skills to word reading, only children with surface dyslexia showed a qualitatively different pattern from controls, which is also problematic for a reading delay account.

We also reported results that are problematic for both models. First, we discovered a small number of pure phonological dyslexics who had relatively spared OC abilities despite a severe phonological deficit. Many of these children had very poor real-word reading abilities. This finding contradicts the strong version of the severity hypothesis generated by the HS model, and is also inconsistent with the dual-route approach, since children ought to be able to read real words reasonably well with an intact lexical route. Second, we found that having disproportionately poor lexical/orthographic skills need not incur a cost. A group of control participants had OC skills on par with the pure surface dyslexic group and yet read as well as control subjects with balanced PC/OC skills.

The failure of the models to fully account for the data suggests that both could be improved with inclusion of additional components. Our results indicate that addition of a semantic network would be an important next step for any computational model of word reading that attempts to account for all cases of developmental dyslexia. Although the dual-route framework conceptually includes semantics, semantic knowledge is not represented in the DRC. The triangle model has historically highlighted the importance of semantics in reading, and some implementations of the triangle model have included a semantic network (Harm & Seidenberg, 2004; Plaut et al., 1996; Woollams et al., 2007). However, these implementations have not yet been used to model developmental dyslexia.

In the current investigation, predictions of the HS model bearing on phonological dyslexia were largely supported, but this was less true for predictions bearing on surface dyslexia. Using a related computational model, Plaut and colleagues (1996) modeled acquired surface dyslexia with damage to a semantic network. We propose that such an approach might also be relevant to developmental surface dyslexia. Consistent with this proposal, children with poor reading comprehension and associated weaknesses in semantics had adequate PC skills but selective deficits in reading low frequency and irregularly spelled words (Nation & Snowling, 1998). Furthermore, in typical 7-year-olds, measures of lexical/semantic knowledge showed stronger associations with irregular than regular word reading (Nation & Cocksey, 2009).

An important challenge to the dual-route account of developmental dyslexia subtypes has been the prevalence of cases with apparent damage to *both* the nonlexical and lexical routes (including children meeting criteria for relative phonological or surface dyslexia). One suggestion has been that early deficits localized to one route generalize over the course of development. In Study 3, we found no support for this suggestion. In fact, our results were consistent with previous research suggesting that the dissociation between lexical and nonlexical processing increases with age, at least for some children (Sprenger-Charolles et al., 2000).

Ziegler and colleagues (2008) provided a different explanation for the preponderance of mixed cases through a participant-based modeling approach with the DRC. The study included a group of French-speaking children with dyslexia who were classified into phonological and surface subtypes according to the regression-outlier method. The children completed several tasks designed to measure the functioning of component reading processes as specified within the DRC. For example, a confrontation naming task was used to measure access to the phonological lexicon, the word superiority effect was used to measure access to the orthographic lexicon, and a phoneme awareness task was used to measure efficiency of the nonlexical route. On average, phonological and surface dyslexic children showed deficits in multiple component processes (including tasks designed to index the functioning of both the lexical and nonlexical routes), leading to the conclusion that these subtypes do not arise from single dissociated deficits. The researchers then ran a series of simulations with the DRC based on individual data. For each dyslexic child, noise was added to those component processes that were impaired, and the model’s performance reading nonwords and exception words was reported. These simulations were effective in reproducing the phonological and surface dyslexia patterns. Thus, the impaired models reproduced the dissociation between nonword and exception word reading without a clear dissociation in the (presumably) underlying impairments. The study therefore accounts for the frequency of mixed cases of dyslexia by arguing that both phonological and surface dyslexia typically result from impairments in both routes.

Ziegler et al. (2008) took a novel approach to modeling individual differences in dyslexia and reported some striking results. Nonetheless, some aspects of their conclusions are puzzling. The original motivation for a dual-route model with separable routes for reading aloud was a behavioral double dissociation among brain-damaged patients, and dual-route proponents have long argued that this dissociation extends to developmental reading problems. Ziegler et al. (2008) propose that the behavioral dissociation exists but does not arise from route-specific impairments. Such an account is logically possible, but seems counter-intuitive in light of the historical roots of a dual-route model. Thus, it would be important to know the results of a similar participant-based modeling approach in models with a single procedure for reading words (such as the HS model) or two procedures that do not have a “clean” division of labor between the routes (such as implementations of the triangle model that include a semantic network).

Our findings also highlight some important future directions for behavioral studies. First, we proposed that a phonological deficit alone may be sufficient to impair reading acquisition, but that a lexical/orthographic deficit is impairing only when it occurs in conjunction with a second deficit. Future studies should test this proposal with a range of second deficits. There has recently been increased attention to the role of semantics in reading, and current results do highlight lexical/semantic skill as a potentially important deficit. Other interesting possibilities include processing speed (McGrath et al., 2011), nonverbal IQ (Catts, Fey, Tomblin, & Zhang, 2002), verbal short-term memory (Griffiths & Snowling, 2002), and morphosyntax (Peterson, Pennington, Shriberg, & Boada, 2009). Second, the current study suggested some intriguing findings about the developmental course of PC and OC skills in both typically developing and impaired readers. We suggested that for children with phonological dyslexia, these skills become increasingly discrepant over time, perhaps reflecting the emergence of compensatory strategies. In contrast, results from Study 6 suggested that for typically developing children, and unbalanced profile might be more likely earlier in reading acquisition. However, we lacked a longitudinal design and were thus unable to address questions such as the stability of subtype classification over time and effects of intervention, type of instruction, and reading experience. Definitive evidence regarding the clinical implications of these subtypes is lacking, and important questions concern whether subtype membership informs issues such as prognosis or treatment response. A future longitudinal study should measure instructional and treatment factors in modeling the developmental trajectories of PC and OC skills for children with phonological or surface dyslexia as well as normally achieving readers.

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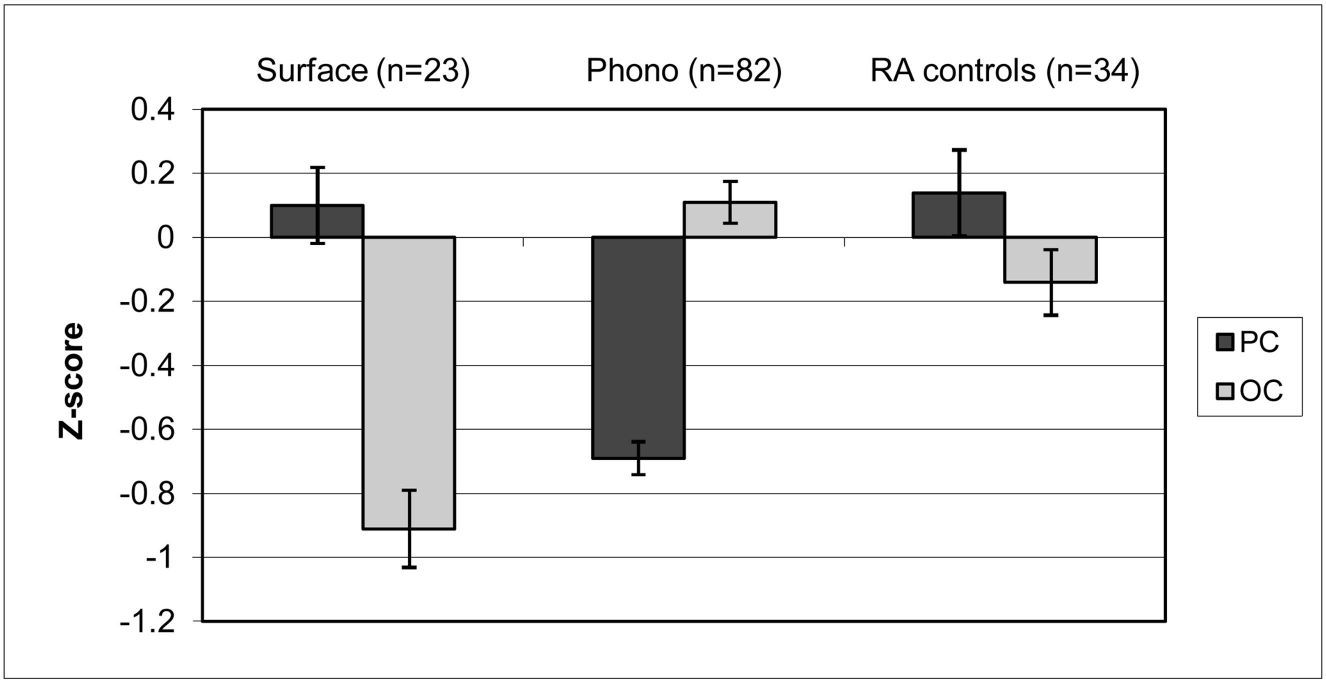

Figure 1. Group by component process interaction
Surface = Group with surface dyslexia
Phono = Group with phonological dyslexia

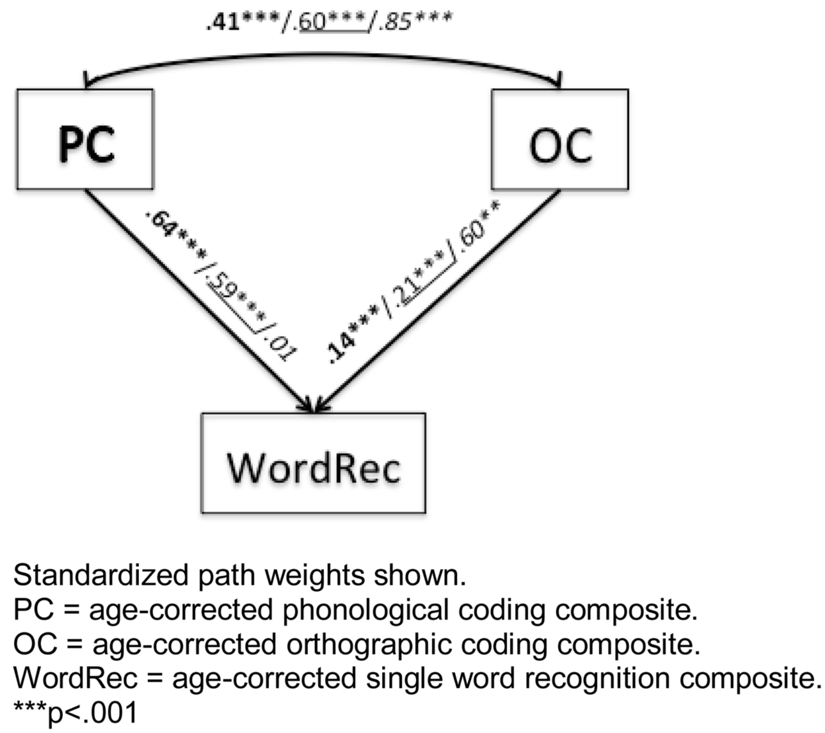

Figure 2. Predicting single word reading from component skills

Bold = Controls (n = 377). Underline = Participants with phonological dyslexia (n = 246). *Italics = Participants with surface dyslexia (n = 93)*.

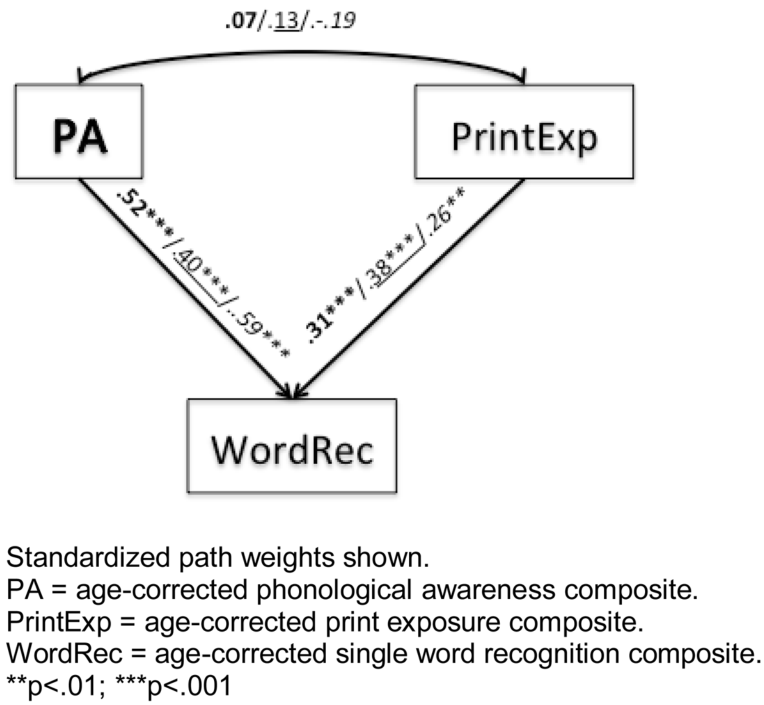

Figure 3. Predicting single word reading from phonological awareness and print exposure

Bold = Controls (n = 377). Underline = Participants with phonological dyslexia (n = 246). *Italics = Participants with surface dyslexia (n = 93).*

Table 1

Demographic Information for Control and Dyslexic Groups

	Control	Dyslexic
N	319	437
Age in years	10.34 (1.56)	10.24 (1.59)
% female	56.4%	43.6%**
Mother years of education	15.28 (2.27)	13.99*** (2.28)
% Caucasian	83.8%	78.9%

** Control vs. dyslexic group difference: $p < .01$.

*** Control vs. dyslexic group difference: $p < .001$.

Table 2

Means (and Standard Deviations) for Demographic Variables, Reading Component Processes, Single Word Reading, and Cognitive Correlates for Dyslexic Subtypes

	Mild	Pure phonological	Pure surface	Relative phonological	Relative surface	Mixed
n	54	68	10	179	34	92
Age (years)	10.99 (1.74)[a]	10.45 (1.68)[a, b]	11.27 (1.99)[a, b]	9.88 (1.41)[b, c]	9.14 (1.09)[c]	10.65 (1.49)[a]
% female	44.4%	69.1%	40.0%	44.7%	32.4%	39.1%
% Caucasian	89.1%	80.4%	100.0%	72.9%	82.1%	80.0%
Mother ed (years)	13.62 (2.41)	13.82 (2.31)	13.80 (2.57)	13.84 (2.29)	14.06 (2.10)	14.04 (2.21)
PC[1]	−0.90 (0.41)[a]	−2.56 (0.67)[b]	−0.58 (0.31)[a]	−3.26 (0.61)[c]	−2.14 (0.79)[d]	−2.09 (0.52)[d]
OC[2]	−0.88 (0.51)[a]	−0.51 (0.35)[b]	−2.13 (0.41)[c]	−2.24 (0.85)[c]	−3.45 (0.67)[d]	−2.22 (0.64)[c]
Word rec[3]	−1.60 (0.49)[a]	−2.03 (0.69)[b]	−1.76 (0.79)[a, b, c]	−2.75 (0.77)[d]	−2.43 (0.79)[b, c, d]	−2.37 (0.68)[c]
Exception words	−1.60 (0.85)[a]	−2.13 (1.05)[a]	−2.01 (1.37)[a, b, c]	−3.68 (1.82)[b, c]	−3.55 (1.59)[b, c, d]	−2.47 (1.41)[a, d]
PA[4]	−0.44 (0.78)[a]	−2.11 (1.42)[b]	−0.64 (1.22)[a, c]	−3.07 (1.51)[d]	−1.76 (1.76)[b, c]	−1.54 (1.23)[b, c]
PM[5]	−0.82 (1.15)[a, b]	−1.47 (1.41)[a]	−0.57 (0.90)[a, b]	−1.28 (1.30)[a]	−1.46 (1.23)[a, b]	−0.75 (1.08)[b]
Vocabulary	−1.34 (1.00)[a]	−1.19 (0.93)[a]	−1.56 (1.13)[a]	−1.43 (1.08)[a]	−1.09 (1.27)[a]	−1.31 (1.05)[a]
Vocabulary ss[5]	9.19 (2.07)	9.78 (2.20)	8.70 (2.58)	8.88 (2.67)	9.50 (3.26)	9.25 (2.50)
Rapid Naming	−0.53 (1.03)	−0.65 (0.84)	−1.32 (0.62)	−0.81 (0.99)	−1.10 (1.01)	−0.94 (1.02)
Print Exposure	−1.01 (0.90)	−1.11 (1.00)	−1.21 (0.99)	−1.41 (0.86)	−1.21 (1.05)	−1.25 (1.05)

Note. Cells with the same superscript within a row do not differ at the $p<.05$ level (Tukey). All scores represent age-corrected z-scores unless otherwise noted

[1] Phonological coding composite.

[2] Orthographic coding composite.

[3] Single word recognition composite.

[4] Phonological awareness composite.

[5] Phonological memory.

[5] Vocabulary scaled score

Table 3

Individual Scores for the 12 Children with Pure Phonological Dyslexia whose PC Deficit was Not Mild [and Their MZ Co-Twins, When Available]

ID	Age	Gender	Mother education (years)	PC[1]	OC[2]	Word rec[3]	PA[4]	PM[5]	Vocab[6]	RN[7]	PrintExp[8]
1	11.13	F	13*	−3.45*	−0.77	−3.59*	−4.16*	−4.77*	−1.80*	−1.33*	−1.97*
2	10.13	M	14	−3.29*	−0.28	−1.63	−3.86*	0.26	0.99	−0.14	1.68
3	9.93	F	12*	−3.90*	−0.15	−3.00*	−3.29*	−3.30*	−2.51*	−1.25*	−1.43*
4	8.21	F	15	−3.40*	−0.21	−1.91	−5.05*	−2.12*	−1.10	−0.82	−0.24
5	11.37	M	13*	−3.74*	−0.83	−2.79*	−2.80	−3.83*	−2.51*	−2.53*	−1.62*
6	9.04	F	16	−3.61*	−0.47	−2.39	−3.13*	−2.76*	−0.97	−1.42	−0.46
7	12.66	F	13*	−3.86*	−0.78	−3.38*	−3.90*	−0.41	−2.30*	−2.28*	−1.30
[7a]				[−3.59*]	[−1.15]	[−2.95*]	[−4.40*]	[−1.62*]	[−1.72*]	[−1.71*]	[−1.30]
8	8.83	M	12*	−3.42*	−0.90	−2.58	−2.62	−3.92*	−0.58	0.13	−0.32
[8a]				[−3.45*]	[−2.33*]	[−2.60]	[−5.14*]	[−2.69*]	[−1.16]	[−0.56]	[−2.25*]
9	9.47	F	13*	−3.72*	−0.50	−1.86	−4.55*	−1.66*	−0.78	−1.26	−1.16
[9a]				[−3.70*]	[−0.97]	[−2.44]	[−3.29*]	[−0.76]	[−0.40]	[−1.33*]	[−0.42]
10	11.94	F	10*	−3.39*	0.06	−2.20	−3.69*	−0.96	−0.68	−2.10*	−2.93*
[10a]				[−3.47*]	[−0.06]	[−2.64]	[−4.55*]	[−1.87*]	[−2.02*]	[−3.56*]	[−2.44*]
11	12.04	F	12*	−3.83*	−0.38	−2.93*	−3.87*	−3.69*	−2.45*	−0.22	−1.71*
[11a]				[−2.95]	[−0.16]	[−2.21]	[−4.93*]	[−1.88*]	[−3.03*]	[−0.42]	[−0.48]
12	8.78	F	13*	−3.30*	−0.08	−1.88	−4.42*	−1.44*	−1.49*	0.40	−1.43*
[12a]				[−1.71]	[−0.36]	[−0.07]	[−2.55]	[−1.74*]	[−2.06*]	[0.67]	[0.80]
Relative phonological mean (standard deviation)	9.88 (1.41)		13.84 (2.29)	−3.26 (0.61)	−2.24 (0.75)	−2.75 (0.77)	−3.07 (1.51)	−1.28 (1.30)	−1.43 (1.08)	−0.81 (0.99)	−1.41 (0.86)

[1]Phonological coding composite.

[2]Orthographic coding composite.

[3]Word recognition composite.

[4] Phonological awareness composite.

[5] Phonological memory.

[6] Vocabulary.

[7] Rapid naming composite.

[8] Print exposure.

[*] Poorer score on a given measure than the mean of the group with relative phonological dyslexia.

Table 4

Prevalence of pure subtypes by age

Age band (years)	Mean age (years)	% Pure Phonological Dyslexia (n pure/n all phonological cases)	% Pure Surface Dyslexia (n pure/n all surface cases)
8.0–8.4	8.23	20.51 (8/31)	6.67 (1/15)
8.5–8.9	8.74	20.00 (6/30)	0.00 (0/6)
9.0–9.4	9.28	20.93 (9/43)	25.00 (1/4)
9.5–9.9	9.76	34.29 (12/35)	28.57 (2/7)
10.0–10.9	10.49	21.62 (8/27)	33.33 (1/3)
11.0–11.9	11.48	33.33 (11/33)	20.00 (1/5)
12.0–13.9	13.00	46.67 (14/30)	100.00 (4/4)

A visual inspection of the data revealed that the prevalence of pure cases did not decrease with age. A bivariate correlation between the mean age for each age band and the percentage of pure cases was large, positive, and statistically significant in both cases (phonological: $r = 0.86$, $p = .02$; surface: $r = .85$, $p = .02$).

Table 5

Ages, Component Process, and Word Recognition Scores for Controls Exhibiting Balanced PC/OC Skills, a Phonological Pattern, and a Surface Pattern. Scores for Subgroups with Pure Phonological and Surface Dyslexia are Included for Comparison.

	n	Age	PC[1]	OC[2]	WordRec[3]	Exception	PA	RN	Vocab
Control: Balanced pattern	54	10.38 (1.64)	0.13[a] (0.60)	0.00[a] (0.77)	0.23[a] (0.71)	0.06 (1.01)	0.16[a] (0.72)	0.05 (0.98)	−0.02 (0.89)
Control: Phonological pattern	23	9.50 (1.19)	−2.09[b] (0.40)	−0.51[b] (0.64)	−1.05[b] (0.58)	−0.89 (0.68)	−1.35[b] (1.23)	−0.22 (0.67)	−0.44 (0.94)
Control: Surface pattern	13	9.75 (1.36)	−0.10[a] (.75)	−2.35[c] (0.63)	0.13[a] (1.02)	−0.89 (1.08)	0.29[a] (0.67)	0.19 (0.61)	−0.24 (0.89)
Pure phonological dyslexia	68	10.45 (1.68)	−2.62 (0.72)	−0.51 (0.34)	−2.04 (0.68)	−2.13 (1.05)	−2.11 (1.42)	−0.65 (0.84)	−1.19 (0.93)
Pure surface dyslexia	10	11.27 (1.99)	−0.65 (0.31)	−2.15 (0.57)	−1.92 (0.71)	−2.01 (1.37)	−0.64 (1.22)	−1.32 (0.62)	−1.56 (1.13)

Notes. Cells with the same superscript within a column do not differ at the $p<.05$ level. Scores of pure phonological and surface dyslexic subgroups are included in this table for ease of reference, but these groups were not included in statistical comparisons.

[1]Standardized, age-corrected phonological coding composite;

[2]Standardized, age-corrected orthographic coding composite;

[3]Standardized, age-corrected word recognition composite