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Examining the Etiology of Reading Disability as Conceptualized by the Hybrid Model

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Abstract

A fairly recent definition of reading disability (RD) is that in the form of a hybrid model. The model views RD as a latent construct that is manifested through various observable unexpected impairments in reading related skills and through inadequate response to intervention. The current report evaluated this new conceptualization of RD from an etiological perspective. The sample consisted of 2737 twin pairs in first through fourth grade ($M_{\text{age}} = 8.52$) from the Florida Twin Project on Reading. Using twin analyses, results showed that a substantial proportion of genetic variance, a small proportion of shared environmental, and a small proportion of non-shared environmental variance was attributed to the RD factor. Findings suggest that the etiological architecture of RD closely mirrors the structure of the hybrid model of RD.

Keywords

reading disability; hybrid model; genetic; environmental; twins

One of the challenges the field of learning disabilities, including reading disability (RD) faces is that of definition. Despite extensive research on the RD definition for at least the past 30 years, agreement on it remains elusive (Fletcher, Stuebing, Morris, & Lyon, 2013). A promising way forward is to consider RD as a construct that is not associated with any specific, single deficit (indicator), but with a combination of indicators (e.g., unexpected impairment in sight word reading and unexpected impairment in associating written letters with their corresponding sounds). One recent example of such an approach is a hybrid model (Fletcher, Lyon, Fuchs, & Barnes, 2007; Fletcher et al., 2013; Spencer et al., 2014; Wagner, 2008). The hybrid model has been empirically supported in a study utilizing a phenotypic (a non-behavioral genetic) approach (Spencer et al., 2014). Phenotypic studies are one option

of addressing the issue of RD definition. A complementary avenue would be a behavioral genetic approach. In fact, an approach of describing RD etiology (genetic and environmental influences) has been advocated as a way of evaluating any new RD conceptualization (Fletcher, 1985; Kavale & Forness, 1987). As the hybrid model of RD is a fairly recent definition of RD, its etiological underpinnings have not been identified yet. Thus, the aim of the current study was to examine the extent to which RD as characterized in the hybrid model is influenced by genetic and environmental factors.

A hybrid model was proposed by Fletcher and colleagues (2007) following the recommendations introduced by the Learning Disabilities Summit convened by the U.S. Office of Special Education Programs (Bradley, Danielson, & Hallahan, 2002). The name of the model stems from the idea that characteristics of various RD models are combined into a single model. First, the feature of unexpected underachievement derives from low achievement and intra-individual differences models (Fletcher et al., 2013). Unexpected underachievement is conceptualized as individuals who struggle to learn to read despite the absence of known causes for poor achievement such as inadequate verbal aptitude, emotional disturbance, economic disadvantage, linguistic diversity, and inadequate instruction (Fletcher et al., 2007). Second, the concept of poor RTI stems from the response to intervention model (Fletcher et al., 2013). Poor RTI indicates that the reading problem is not simply the result of limited instruction, but that children who do not respond adequately to both instruction and intervention are considered RD (Fletcher et al., 2007; Spencer et al., 2014). Incorporating the elements of unexpected underachievement and poor RTI, the hybrid model of RD suggests that information on the RD construct be incorporated in the form of indicators that epitomize both characteristics as they are considered necessary inclusionary criteria in an RD definition (Fletcher et al., 2007).

Nascent empirical research has thus far yielded one published report on the hybrid model of RD. In a large-scale study (N = 31,339) Spencer and colleagues (2014) modeled RD as a continuous latent variable with four indicators (Figure 1, left panel). Indicators were based on both inclusionary characteristics for RD: (1) indicators that tapped unexpected underachievement – these were indicators of unexpected impairment in sight word reading and unexpected impairment in phonological decoding (i.e., decoding of nonsense words) and (2) an indicator of poor RTI. Aside from the two inclusionary components for RD, the model also included an indicator of impaired reading comprehension relative to listening comprehension. This indicator refers to the difficulty of comprehending written versus spoken passage due to difficulties in decoding in RD readers. Results from a confirmatory factor analysis showed that the RD hybrid model with the four indicators could be represented as a unidimensional structure. The model yielded an excellent fit ($\chi^2(2) = 263.4$; Comparative Fit Index = .99; Tucker Lewis Index = .97; Root Mean Square Error of Approximation = .065 with a 95% confidence interval between .058 and .071).

Spencer et al.'s study (2014) provided the first empirical support in the phenotypic literature for the definition of the hybrid model of RD. A genetically informed approach may complement the phenotypic approach by helping clarify the conceptualization of RD with respect to genetic and environmental origins underlying RD. Hence, a genetically sensitive study could assist in constructing a definition of RD based on the etiological structure of

RD. To address this point, our goal in the current report is to examine genetic and environmental influences on RD as it is operationalized by the Spencer et al.'s (2014) model. If our results demonstrate that RD factor's phenotypic variance reflects genetic and environmental variance, then such a finding would mirror the hybrid model's phenotypic structure and suggest genetic liability and environmental effects for RD as a latent factor. In so doing, the phenotypic conception of RD (Spencer et al., 2014) would be evaluated and complemented from an etiological perspective. In order to test this assumption directly, it is necessary to make use of a multivariate genetically informed approach such as a twin study.

The kind of multivariate genetically sensitive method in the twin study particularly well suited to test the underlying etiology of RD in the hybrid model is called a common pathway model (Figure 1, right panel). This method has the advantage of extending the Spencer et al.'s (2014) model in such a way that genetic and environmental factors are added to the RD factor and its indicators to account for etiological influences. On its own, the latent factor derived from the common pathway model is the same as the latent factor that is derived from standard phenotypic analyses. To our knowledge, the common pathway model has not been used in the reading literature thus far, however it is commonly applied in the clinical psychopathological literature. For example, the co-occurrence between indicators of ADHD hyperactivity-impulsivity, ADHD inattention, and emotional lability is primarily the result of a single latent factor, labeled "externalizing behaviors" (akin to our RD factor) (Merwood et al., 2014, p. 215).

In the common pathway model, the latent factor RD has its own (also called RD common or RD shared) genetic (A_1), shared (C_1), and non-shared environmental (E_1) factors, as indicated in the right panel of Figure 1. Genetic influences refer to the sum of genetic effects inherited from one's biological parents. Shared environmental influences are aspects of the environment that affect both twins and make them more similar, such as home and school environment. Non-shared environmental influences are those that are unique to each twin (as well as measurement error) and contribute to differences between twins, such as each twin's own experience. Genetic and environmental influences A_1 , C_1 , E_1 explain the proportion of variance associated with the latent factor RD (common variance). Apart from the RD common A_1 , C_1 , and E_1 factors, there are also genetic (A_{2-5}), shared environmental (C_{2-5}), and non-shared environmental factors (plus measurement error; E_{2-5}) that are unique to each individual indicator in the model. They are not shared with the RD factor. Genetic and environmental influences A_{2-5} , C_{2-5} , E_{2-5} explain the proportion of residual variance associated with each indicator (unique variance). Proportions of variance explained by both sets of factors (common plus unique variance) add up to 100%.

As previously noted, the common pathway model has not been used thus far to examine the etiology of RD. Nonetheless, genetic and environmental effects on RD have been studied extensively. The literature has shown highly consistent results, such that RD was influenced mainly by genetic factors and relatively less by shared environmental factors (Plomin & Kovas, 2005). This was the case regardless of whether RD was defined as a deficit in word recognition (e.g., Harlaar, Spinath, Dale, & Plomin, 2005; Hart, Petrill, & Kamp Dush, 2010; Kovas et al., 2007), reading fluency (e.g., Haworth et al., 2009), or reading comprehension (e.g., Haworth et al., 2009; Kovas et al., 2007). Genetic estimates for RD (in

terms of proportions of phenotypic variance accounted for) ranged from 47% to 74% (Friend, DeFries, & Olson, 2008; Harlaar, Spinath, Dale, & Plomin, 2005; Hart, Petrill, & Kamp Dush, 2010; Hawke, Wadsworth, DeFries, 2006; Haworth et al., 2009; Light & DeFries, 1995; Kovas et al., 2007). In contrast, estimates of shared environment were in the range from 0% to 32% (Friend, DeFries, & Olson, 2008; Hart, Petrill, & Kamp Dush, 2010; Haworth et al., 2009; Kovas et al., 2007).

In sum, it appears that RD owes largely to genetic influences and less to shared environmental influences. If the hybrid model is a viable way to define RD, then the biometric analysis will reveal these effects. Specifically, based on previous twin studies on RD (e.g., Harlaar, Spinath, Dale, & Plomin, 2005), paths to factors A_1 (genetic effects) and C_1 (shared environmental effects) were expected to be significant, with RD likely to be attributable primarily to genetic and less to shared environmental effects. Finding a larger piece of the variance explained at the RD common factor level rather than at the indicator unique level would suggest genetic liability and environmental effects on RD as a latent factor, which would be consistent with the basic idea behind the hybrid model. Even though we suspected that the indicators will account for the majority of variance through the RD factor, we also predicted that the indicators will retain some unique variance.

Method

Participants

The present sample was drawn from the Florida Twin Project on Reading (FTP-R; Taylor & Schatschneider, 2010). The ascertainment method for the FTP-R has been described in detail elsewhere (Taylor & Schatschneider, 2010; Taylor, Hart, Mikolajewski, & Schatschneider, 2013). Zygosity of twin pairs was determined by a parental five-item questionnaire obtained during intake into the FTP-R. The data presented in this study were used from the Florida's Progress Monitoring and Reporting Network (PMRN) database for four cohorts of twins from the FTP-R in order to maximize sample size for each indicator. For the indicators unexpected underachievement in sight word reading, unexpected underachievement in phonological decoding, and poor response to intervention, data were used for twins who were in first or second grade during the 2005–2006, 2006–2007, 2007–2008, 2008–2009 school years. These grades were chosen based on the prior report that also utilized the PMRN data (Spencer et al., 2014). For the indicator impaired reading comprehension relative to listening comprehension, data were used for the same twins who were in third or fourth grade during the 2007–2008, 2008–2009, 2009–2010 school years. These grades were also chosen based on the prior report (Spencer et al., 2014) and on the fact that reading comprehension starts developing in 3rd grade (Chall, 1983).

The final sample for this study comprised 2737 7- to 10-year old ($M = 8.52$, $SD = 2.04$, range = 6.54–10.77) twin pairs. Specifically, the sample included 918 monozygotic (MZ; 467 female-female, 451 male-male) and 1819 dizygotic (DZ; 459 female-female, 491 male-male, 869 female-male) twin pairs. The present report uses a general population sample, thus avoiding possible selection biases associated with placing arbitrary cutoff points to select RD sample. Racial composition of the present sample as reported in the PMRN was 1.62% Asian, 18.84% Black, 24.16% Hispanic, 0.15% Native American/Pacific Islander, 4.56%

Mixed, 48.08% White, and 2.59% with no reported information about race. The percentages here are similar to values reported by the U.S. Census Bureau for the state of Florida (<https://www.census.gov/quickfacts/table/RHI125215/12,00>). Regarding the socio-economic status, 33.54% of the twins did not apply or were not eligible for free or reduced price lunch, 31.91% were eligible for free lunch, 8.51% were eligible for reduced price lunch, and 26.04% had no information in regard to lunch status.

Procedures and Measures

Dynamic Indicators of Basic Early Literacy Skills (DIBELS) data were collected by schools at three time points throughout the school year (during the months of September, February, and April) and entered into the PMRN database. Data from Peabody Picture Vocabulary Test and Florida Comprehensive Assessment Test - Reading were also included in the PMRN. These two tests were administered once per school year; specifically, vocabulary test in February, and reading test during a 10 day testing window in the spring. All parents of twins provided informed consent for investigators to use their twins' PMRN data. The FTP-R project was approved by the Florida State University Institutional Review Board.

DIBELS Nonsense Word Fluency (NWF)—NWF is a measure of phonological decoding and measures children's ability to use letter-sound correspondence to decode words. Children are presented with printed vowel-consonant or consonant-vowel-consonant nonsense words (e.g., ov, sig, rav) and asked to verbally produce the individual letter sounds in each word, or verbally produce, or read, the whole nonsense word. The number of letter-sounds produced correctly in one minute is the final score. The alternate-form reliability is .83 in first grade. The median concurrent, criterion-related validity with DIBELS Oral Reading Fluency is .63 in second grade (Good et al., 2004).

DIBELS Oral Reading Fluency (ORF)—ORF is a measure of sight word reading and measures children's reading accuracy and speed with reading words in a connected text. Children read each of three grade-level passages aloud for one minute. Words omitted, substituted, and hesitations of more than three seconds are scored as errors. Words self-corrected within three seconds are scored as accurate. The number of words read correctly in one minute from each passage is recorded, and the median value from the three passages is taken as the final score. The alternate-form reliability is .95 in first and .98 in second grade (Good, Kaminski, Smith, & Bratten, 2001).

Peabody Picture Vocabulary Test (PPVT)—PPVT is a measure of receptive (hearing) vocabulary. The test is individually administered. The examiner presents a series of pictures to a student. There are four pictures to a page, and each is numbered. The examiner speaks a word describing one of the pictures and asks the student to point to or say the number of the picture that the word describes. The median alternate-forms reliability ranges from .88 to .96. PPVT is highly correlated with the measures of IQ. It has a correlation of .96 with the Verbal IQ score from the third edition of the Wechsler Intelligence Scales for Children (Dunn & Dunn, 1997).

Florida Comprehensive Assessment Test - Reading (FCAT)—FCAT is a measure of reading comprehension. Students are asked to read narrative and expository passages and answer multiple choice, short or long answer format items based on the content of the passage. Reliability for FCAT from Cronbach's alpha is .89 in third and .85 in fourth grade. The criterion-related validity with Stanford Achievement Test Series is .84 in third and .83 in fourth grade (Florida Department of Education, 2007).

Data Analyses

Operational definition of indicators—Operational definitions of indicators used in the present study were created using identical procedures as in Spencer and colleagues, 2014. Briefly, unexpected low achievement in sight word reading and phonological decoding were included to represent the notion of unexpected underachievement. ORF and NWF variables (the April scores) were regressed on PPVT, which served as a proxy for verbal aptitude. The two indicators were labeled ORFPPVT and NWFPPVT. To operationalize a discrepancy between reading and listening comprehension - reading comprehension being poorer than listening comprehension (Badian, 1999) - FCAT was regressed on PPVT, which served here and in previous reports as a proxy for listening comprehension (Spencer et al., 2014). The indicator was labeled FCATPPVT. To operationalize the poor response to intervention, student's low growth over the year (September, February, and April scores) and student's end-of-year low achievement (April score) in word reading (ORF) were taken into account. The slopes and end-of-year intercepts were converted to *z* scores and summed to create an RTI score that equally weighted slope and end-of-year intercept. The indicator was labeled RTI.

Descriptive statistics, correlational analyses, and overview of the twin method

—For the sake of clarity, descriptive statistics – means, standard deviations, ranges, skewness – were calculated on raw variables, that is prior to residualizing the variables ORF, NWF, and FCAT on PPVT and prior to standardizing all the variables. Because age and gender effects can bias twin analyses, and age and gender differences were not a focus of the current study, data were also corrected by residualizing on age, age-squared, and gender (McGue & Bouchard, 1984), and then standardized. It should be noted that data not residualized on age, age-squared, and gender produced almost identical results. Phenotypic (Pearson) correlations, all subsequent twin correlations, and structural equation modeling were calculated on residualized and standardized indicators.

Twin methodology allows for an examination of the variance and covariance of etiological (genetic and environmental) influences on a particular trait (indicator), or shared among traits (indicators). The assumptions under the twin method are that MZ twins share approximately 100% of their segregating genes whereas DZ twins share, on average, 50 %. Shared environmental factors are assumed to be 100% for both MZ and DZ twins if siblings are reared together. Non-shared environmental factors are not shared between the siblings for either MZ or DZ twins (Neale & Cardon, 1992). Model fitting is based on the comparison of the variance-covariance matrices in MZ and DZ twins and allows for the separation of the observed phenotypic variance into additive genetic (A), shared environmental (C), and non-shared environmental (E) components. In multivariate genetic

analyses, as well as partitioning the phenotypic variance of indicators, it is also the covariance between indicators that is decomposed into A, C, and E influences.

Intraclass (ICC) and cross-twin cross-trait (CTCT) correlations provide first indications to what extent genes and environments influence variation of a particular indicator and covariation of two indicators. Greater ICCs for MZ relative to DZ twins indicate the presence of additive genetic effects. By contrast, ICCs for MZ and DZ twins that are similar in magnitude and are significantly greater than 0 signify a lack of genetic effects but significant shared environmental influences. Finally, ICCs for MZ twins that are less than 1.0 indicate the presence of non-shared environmental factors. The CTCT correlation is calculated by correlating an indicator from one member of a pair with a different indicator from his/her co-twin. The interpretation of the CTCTs follows the same logic as that of the ICCs. Descriptive statistics and correlational analyses were conducted in SAS 9.4.

Structural equation modeling—Structural equation modeling was performed to obtain estimates of the A, C, and E on the indicators (ORFPPVT, NWFPPVT, FCATPPVT, and RTI). First, univariate analyses were conducted for the four indicators. Next, in accordance with the aim of our study, the common pathway model shown in the right panel of Figure 1 was tested. Genes (A_1), shared (C_1), and non-shared environment (E_1) together contribute to one latent factor (i.e., RD), which is responsible for the observed covariance between the indicators. Genetic (A_{2-5}), shared (C_{2-5}), and non-shared environmental (E_{2-5}) factors unique to each indicator are also incorporated in the model. Structural equation modeling analyses were conducted on all available data in the OpenMx package in R using a full information maximum likelihood estimator (Boker et al., 2011). Significance of parameter estimates was based on the 95% confidence intervals not including zero.

Results

Descriptive Statistics and Correlational Analyses

Means, standard deviations, ranges, and skewness on raw variables are presented in Table 1. No outliers were detected. Phenotypic correlations are summarized in Table 2. The pattern of phenotypic correlations indicated significant covariation across all combinations of indicators. The largest correlation was observed between impaired sight word reading (ORFPPVT) and RTI score, which was not surprising given that the RTI score equals a summed up *z*-score of sight word reading (ORF) slope and ORF end-of-year intercept. The lowest correlation was seen between impaired reading comprehension relative to listening comprehension (FCATPPVT) and impaired phonological decoding (NWFPPVT), however it was still moderate in magnitude and significant. As shown in Table 3, the pattern of ICCs, which consistently showed greater magnitude for MZ than DZ twins, suggested genetic influences on all indicators. Given that MZ ICCs were less than twice the magnitude of DZ ICCs and less than unity for all variables, there was also evidence for shared and non-shared environmental effects. The cross-twin cross-trait correlations (see Table 3) were consistently lower for DZ twins than for MZ twins, suggesting that genetic factors were at least partly responsible for the overlap between each combination of indicators. The moderate MZ

CTCTs also indicated that common non-shared environmental factors were likely to be important in explaining covariations among different indicators.

Structural Equation Modeling

Univariate results from the structural equation model fitting of the data for each indicator are displayed in Table 4. Univariate estimates were consistent with the ICCs and indicated the relative impact of genetic and environmental factors on each of the indicators.

A common pathway model was fit to examine the etiology of RD as conceptualized by the Spencer et al.'s hybrid model (2014) and results of the parameter estimates are shown in Figure 2. The RD common genetic factor (A_1) suggested significant (i.e. confidence interval did not include zero) genetic effects (pathway .74). This means that 55% of the common variance of RD factor was explained by genetic factors (squaring the path coefficient gives an estimated common variance of RD factor explained by genetic influences; $(.74)^2 = .55$). Also, common shared environmental (C_1) and non-shared environmental (E_1) factors indicated significant environmental effects (pathways .44 and .51, respectively) contributing to the RD factor. Thus, 19% of the RD common variance was explained by shared environmental effects, and 26% by non-shared environmental effects. These results support the hypothesis that the RD factor would mostly have genetic influences. As expected, shared and non-shared environment influence RD to a lesser degree than genetic effects. As for the genetic and environmental effects unique to each indicator, they are depicted in the lower part of Figure 2 (A_{2-5} , C_{2-5} , E_{2-5}). Indicators impaired reading comprehension relative to listening comprehension (FCATPPVT) and poor response to intervention (RTI) had significant unique genetic influences. In addition, indicators impaired phonological decoding (NWFPPVT), impaired reading comprehension relative to listening comprehension (FCATPPVT), and poor response to intervention (RTI) also had significant unique shared environmental influences and non-shared environmental influences plus possible measurement error.

The estimated total contributions of genetic, shared, and non-shared environmental effects in terms of variance explained on the four indicators of RD, and the proportion of each that is common to the RD factor versus indicator unique are presented in Table 5. Two results are particularly worthy of comment. First, as expected, results indicated that the majority of genetic influences were explained at the RD common factor level. For example, for the indicator impaired sight word reading (ORFPPVT), 52% of the variance was common to the RD factor. Only 1% of the variance in ORFPPVT was indicator unique. Common variance .52 represents 98% of the total variance for the indicator of ORFPPVT, whereas unique variance .01 represents 2% of ORFPPVT's total variance. A similar pattern in magnitude of the expressed variance at the common level was observed also for impaired phonological decoding (NWFPPVT) and poor response to intervention (RTI), but not for impaired reading comprehension relative to listening comprehension (FCATPPVT). FCATPPVT indicated a larger piece of the genetic variance being indicator unique (66%). The second interesting finding is that shared environmental variance was largely common to the RD factor only for indicators ORFPPVT and RTI (90% and 65%, respectively). In contrast, for indicators

NWFPPVT and FCATPPVT, nearly all of shared environmental variance was indicator unique (84% and 89%, respectively).

Discussion

The hybrid model of RD (Spencer et al., 2014) posits that RD is an unobserved latent variable which is manifested through a constellation of underlying deficits or indicators. Spencer and colleagues (2014) provided initial empirical evidence for the model. This study extended their work in an important way by evaluating their conceptualization of RD from an etiological perspective. Specifically, a common pathway model was conducted to test the assumption that the etiology of RD mirrors the phenotypic structure of RD in such a way that the latent factor of RD is itself influenced by genes and environment, providing a common pathway of etiology influences down to its indicators. Our results clearly showed that the underlying etiological architecture of RD closely reflects the phenotypic structure of the hybrid model of RD (Spencer et al., 2014), such that an appreciable proportion of genetic and environmental variance was attributed to the RD factor rather than its indicators. This suggests that Spencer et al.'s (2014) conception of RD in the form of the hybrid model may be accurate.

The RD factor was significantly and substantially influenced by genetic factors, as expected. Such a result is indicative of a broad shared genetic liability across all the RD indicators in the hybrid model. This is consistent with the findings of others who have found that RD measured either by word recognition, reading fluency, or reading comprehension assessments appears to be substantially heritable (e.g., Harlaar, Spinath, Dale, & Plomin, 2005; Haworth et al., 2009). Molecular genetic studies have shown that a set of genes is responsible for biological mechanisms involved in RD. For example, candidate genes such as DYX1-9, ROBO1, and DCDC2 have been proposed to be implicated in RD susceptibility (Carrion-Castillo et al., 2017). Overall, results of the current study provide additional evidence that a common set of genetic effects accounts for appreciable variation in RD.

Even though RD at the factor level is principally due to genetic influences, this should not deter our interests to consider environment and its prevention and intervention strategies as a way to, at least, partially compensate for genetic constraints. Consistent with the hypothesis and in line with prior work (e.g., Kovas et al., 2007), results demonstrated that RD factor was affected by small, yet significant shared environmental influences. Our findings suggest that there are shared environmental effects on the RD factor, common across all the indicators in the hybrid model. Shared environment could include aspects of home language and literacy environment and teacher, instruction, and/or school effects. For example, deficits in word decoding, a hallmark indication of RD, may manifest itself early on in oral language (Shaywitz & Shaywitz, 2005). Thus, home environmental sources such as parental influences toward creating environments of rich oral language (Burgess, Hecht, & Lonigan, 2002) may help remediate reading related skills in RD individuals. Turning to school environment, Byrne and colleagues (2010) found that teacher effects (more strictly, classroom effects) might be a minor source of variability in children's early reading related skills (decoding, reading comprehension, spelling). As for the quality of reading instruction effects, some reading training studies with RD children showed a growth rate improvement

in word recognition during the training periods (e.g., Wise & Olson, 1995). On the basis of prior research and current results, shared environmental factors appear to be important for RD, suggesting that home and/or school environments may be able to influence a child's RD to some extent.

RD factor also had small, but significant non-shared environmental effects. These are indicative of child-specific environmental influences only (i.e., less the error typically associated with this estimate due to the use of latent factor; Gayan & Olson, 2003). One example that might act as a non-shared environmental influence could be engagement in twins' individual reading experiences and print exposure. Eklund, Torppa, and Lyytinen (2013) examined to what extent second graders with RD and no RD differed from each other in task-focused behavior and print exposure. Results showed that children with RD had more task avoidance and were reading less as compared to typical readers. While both of these traits might be genetically influenced, print exposure could also indicate the importance of individual experiences with reading. There could be, however, many other examples of non-shared environments influencing RD, including idiosyncratic experiences, differential treatment by the parents and teachers in reading related activities, and, importantly, different perceptions of such experiences, even if the events appear to be the same for both twins. Each of these options, though, probably has only a small effect on a trait, such as RD (Kovas et al., 2007).

A useful feature of the common pathway model is to account not only for the factor level RD common variance, but also to evaluate the significance of the indicators' unique variance. Significant indicator unique effects in our study suggest that each of the indicators of RD partly represents something etiologically distinct. As such, these results are compatible with the intra-individual differences model (Fletcher et al., 2007) which posits that an individual with RD might have strengths in one cognitive process but weaknesses in the other. This means there is variability in the cognitive strengths and weaknesses displayed by individuals with RD, which could be due to differences in the etiology of the cognitive processes (akin to indicators in the hybrid model of RD). Our results suggest that unexpected underachievement in phonological decoding and impaired reading comprehension relative to listening comprehension seem to be more influenced by certain features of the home and/or school environments that are independent of those factors that also affect unexpected underachievement in sight word reading and poor response to intervention. Similarly, between 84-89% of the non-shared environmental effects on the two indicators were unique effects, some of which may be due to measurement error. Such findings are especially informative for prevention and intervention efforts.

For example, they suggest that prevention strategies at home and school level, and/or personal experience may be particularly able to influence a child's vulnerability to impaired phonological decoding and reading comprehension. Prior literature has indicated that interventions aimed at improving phonological abilities and comprehension in combination appear to be effective in children with RD (e.g., Gustafson et al., 2011; Snowling & Hulme, 2011; Wise, Ring, & Olson, 2000). More generally, our results imply that interventions should be developed to target multiple indicators of RD. Cognitive and linguistic processes, which underlie manifestations of RD, appear to be malleable. This indicates that children's

reading skills can generally be improved by effective intervention (Connor, Alberto, Compton, & O'Connor, 2014). If the findings of common genetic and environmental influences on RD are replicated, future research could explore whether there are intervention or prevention strategies that could ameliorate conditions leading to an RD and its indicators jointly.

Apart from implications for intervention, our results are directed to informing models aiming to determine the structure and definition of RD. While Spencer et al. (2014) provided evidence for the hybrid model of RD at the phenotypic level, evidence from the current study supports and complements the premise of the hybrid model from an etiological perspective. Substantial genetic and minor environmental effects are reflected at the RD common factor level, which mirror the theoretical expectations of the hybrid model and justify it as a feasible way to conceptualize RD. Results imply that children with RD have an underlying genetic risk to develop an RD. This genetic predisposition then reflects itself at the phenotypic level through individual indicators. An approach of clustering of related indicators into an RD factor, as posited in the hybrid model, seems to be a useful approach to capture the common etiology of particular deficits of RD into an entity.

Some limitations should be considered when interpreting the present results. First, even though the hybrid model captures major indicators of RD, it can be extended to account for other potentially important elements of RD, such as impaired phonological awareness, which is often viewed as a leading cause of RD (Snowling, 2000). Second, the loading on the RD common factor was weakest for the indicator impaired reading comprehension relative to listening comprehension (FCATPPVT; see Figure 2). This implies that this indicator did not best index the genetic liability and environmental effects common to all RD indicators, but rather it derived more than half of its genetic and environmental effects from factors specific to that deficit. This might raise the question whether FCATPPVT offers the level of measurement precision necessary to reveal that it etiologically belongs in the hybrid model. The current study attempted to evaluate the Spencer et al.'s (2014) RD conceptualization from an etiological perspective, thus the same variables as in the Spencer et al. (2014) study were created and included in the current study. Future work should attempt to improve the loading on the RD latent factor, for example, by conceptualizing the notion of impaired reading comprehension relative to listening comprehension using another listening comprehension measure. Previous study (Spencer et al., 2014) and the current study both used Peabody Picture Vocabulary Test (PPVT) as a proxy for listening comprehension. We acknowledge that PPVT may not have been the most accurate representation of it, however we used this measure because it represented the source of data available for our entire sample. The indicator impaired reading comprehension relative to listening comprehension (FCATPPVT) demonstrated high indicator unique non-shared environmental variance. This variance could be attributed to non-shared environmental effects, but also to measurement error. It is possible that the manipulation of the reading comprehension variable FCAT into the indicator form FCATPPVT created more measurement error.

All in all, the finding of the current study that genetic and environmental architecture of the hybrid model reflects its phenotypic structure provides support for the model. Despite the

initial success in showing that the model might be a way forward to RD definition, it is clear that the first phenotypic empirical study (Spencer et al., 2014) plus the current twin study are preliminary, and that the framework proposed by these two studies leads, hopefully, to further empirical research in this domain.

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Figure 1.

Left panel: Spencer et al.'s (2014) hybrid model of RD. Right panel: Common pathway model for the Spencer et al.'s (2014) hybrid model. RD = reading disability, RC = reading comprehension, LC = listening comprehension, RTI = response to intervention.

Figure 2.

Common pathway model for the Spencer et al.'s (2014) hybrid model with standardized path coefficients; significant paths are in bold [95% confidence intervals]. RD = reading disability, RC = reading comprehension, LC = listening comprehension, RTI = response to intervention.

Table 1

Descriptive Statistics for All Raw Variables (Pre Residualizing and Standardizing)

Variable (Measure)	M (SD)	N	Min	Max	Skew
1. Sight Word Reading (ORF)	85.84 (41.41)	3725	0.00	218.00	0.19
2. Phonological Decoding (NWF)	83.19 (39.13)	3727	0.00	232.00	0.50
3. Reading Comprehension (FCAT)	319.25 (58.53)	2686	100.00	500.00	-0.37
4. Vocabulary (PPVT)	93.26 (15.22)	1285	40.00	137.00	-0.06

Note. M = means, SD = standard deviation, N = number of twins, ORF = Oral Reading Fluency, NWF = Nonsense Word Fluency, FCAT = Florida Comprehensive Assessment Test – Reading, PPVT = Peabody Picture Vocabulary Test.

Table 2

Phenotypic Correlations

Indicator (Indicator Label)	1.	2.	3.
1. Impaired Sight Word Reading (ORFPPVT)	1		
2. Impaired Phonological Decoding (NWFPPVT)	.60 **	1	
3. Impaired Reading Comprehension Relative to Listening Comprehension (FCATPPVT)	.46 **	.34 **	1
4. Response to Intervention (RTI)	.86 **	.53 **	.42 **

Note.

**
p < .001.

Table 3

Intraclass and Cross-Twin, Cross-Trait Correlations

Indicator (Indicator Label)	MZ ICCs	DZ ICCs	1.	2.	3.	4.
1. Impaired Sight Word Reading (ORFPPVT)	.75**	.48**	–	.27**	.23**	.41**
2. Impaired Phonological Decoding (NWFPPVT)	.46**	.32**	.40**	–	.17**	.26**
3. Impaired Reading Comprehension Relative to Listening Comprehension (FCATPPVT)	.54**	.42**	.34**	.21**	–	.28**
4. Response to Intervention (RTI)	.70**	.46**	.64**	.37**	.34**	–

Note. – = N/A.

**

p < .001. Intraclass correlations (ICCs) are presented in columns 2 and 3. Cross-twin, cross-trait correlations (CTCTs) are in columns 4-7. The estimates for MZ twins are below the diagonal and for DZ twins above the diagonal.

Table 4

Univariate Estimates for Heritability (h^2), Shared Environmental (c^2), and Non-shared Environmental (e^2) Influences for All Indicators [with 95% Confidence Intervals].

Indicator (Indicator Label)	h^2	c^2	e^2
Impaired Sight Word Reading (ORFPPVT)	.52* [.40–.64]	.22* [.11–.33]	.26* [.22–.29]
Impaired Phonological Decoding (NWFPPVT)	.24* [.05–.41]	.21* [.07–.35]	.55* [.49–.63]
Impaired Reading Comprehension Relative to Listening Comprehension (FCATPPVT)	.29* [.09–.47]	.26* [.11–.41]	.45* [.37–.51]
Response to Intervention (RTI)	.48* [.42–.54]	.22* [.21–.27]	.30* [.28–.32]

Note.

* indicates significance based on confidence intervals not bounding zero.

Table 5
Estimates of the Total Contributions of Genetic, Shared, and Non-shared Environmental Effects for the Indicators

Indicator (Indicator Label)	Genetic			Shared Environment			Non-shared Environment		
	% Variance Total	% Variance Common to the RD Factor	% Variance Indicator Unique	% Variance Total	% Variance Common to the RD Factor	% Variance Indicator Unique	% Variance Total	% Variance Common to the RD Factor	% Variance Indicator Unique
1. Impaired Sight Word Reading (ORFPPVT)	53 ^c	52 ^a (98)	1 ^b (2)	21	19 (90)	2 (10)	26	26 (100)	0 (0)
2. Impaired Phonological Decoding (NWFPPVT)	22	17 (77)	5 (23)	23	6 (26)	17 (74)	55	9 (16)	46 (84)
3. Impaired Reading Comprehension Relative to Listening Comprehension (FCATPPVT)	29	10 (34)	19 (66)	24	3 (13)	21 (87)	47	5 (11)	42 (89)
4. Response to Intervention (RTI)	49	37 (76)	12 (24)	20	13 (65)	7 (35)	31	18 (58)	13 (42)

Note. Percentages in parentheses are proportions of common and unique variance for each type of effect (genetic, shared environmental, and non-shared environmental).

^a Multiplying the path coefficient from the factor A1 and the loadings from the RD factor – both indicated in Figure 2 – and squaring the product gives the proportion of the variance explained by common genetic factor. For example, $((.74) * (.98))^2 = .52$, which represents 52% of the variance explained by the common genetic factor for the ORFPPVT indicator. Same logic applies for common shared environmental and non-shared environmental factors.

^b Squaring the path coefficients from the factors A2-5 – again, indicated in Figure 2 – gives the proportion of variance explained by each unique genetic factor. For example, $(.11) * (.11) = .01$, which represents 1% of the variance explained by the unique genetic factor for the ORFPPVT indicator. Same logic applies for specific shared environmental and non-shared environmental factors.

^c Together, genetic factor for the ORFPPVT indicator explains $52\% + 1\% = 53\%$ of the total variance.