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Citation for published version (APA):

Document status and date:
Published: 01/01/2016

DOI:
10.1016/j.ridd.2015.12.018

Document Version:
Publisher's PDF, also known as Version of record

Document license:
Taverne

Please check the document version of this publication:
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Download date: 16 Sep. 2023
Cognitive and familial risk evidence converged: A data-driven identification of distinct and homogeneous subtypes within the heterogeneous sample of reading disabled children

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ARTICLE INFO

Article history:
Received 10 April 2015
Received in revised form 1 November 2015
Accepted 5 December 2015
Available online 27 February 2016

Keywords:
Subtypes of reading disability
Data-driven clustering
Cognitive profiling
Familial risk and environmental risk factors
Dyslexia deficits

ABSTRACT

The evident degree of heterogeneity observed in reading disabled children has puzzled reading researchers for decades. Recent advances in the genetic underpinnings of reading disability have indicated that the heritable, familial risk for dyslexia is a major risk factor. The present data-driven, classification attempt aims to revisit the possibility of identifying distinct cognitive deficit profiles in a large sample of second to fourth grade reading disabled children. In this sample, we investigated whether genetic and environmental risk factors are able to distinguish between poor reader subtypes. In this profile, we included reading-related measures of phonemic awareness, letter-speech sound processing and rapid naming, known as candidate vulnerability markers associated with dyslexia and familial risk for dyslexia, as well as general cognitive abilities (non-verbal IQ and vocabulary). Clustering was based on a 200 multi-start K-means approach. Results revealed four emerging subtypes of which the first subtype showed no cognitive deficits underlying their poor reading skills (Reading-only impaired poor readers). The other three subtypes shared a core phonological deficit (PA) with a variable and discriminative expression across the other underlying vulnerability markers. More specific, type 2 showed low to poor performance across all reading-related and general cognitive abilities (general poor readers), type 3 showed a specific letter-speech sound mapping deficit next to a PA deficit (PA-LS specific poor readers) and type 4 showed a specific rapid naming deficit complementing their phonological weakness (PA-RAN specific poor readers). The first three poor reader profiles were more characterized by variable environmental risk factor, while the fourth, PA-RAN poor reader subtype showed a significantly strong familial risk for dyslexia. Overall, when we zoom in on the heterogeneous phenomenon of reading disability, unique and distinct cognitive subtypes can be identified, distinguishing between those poor readers more influenced by the role of genes and those more influenced by environmental risk factors. Taking into account this diversity of distinct cognitive subtypes, instead of looking at the reading disabled sample as a whole, will help tailor future diagnostic and intervention efforts more specifically to the needs of children with such a specific deficit and risk pattern, as well as providing a more promising way forward for genetic studies of dyslexia.

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Abbreviations: PA, phonological/phonemic awareness; LS, letter-speech sound processing; RAN, rapid automatized naming.

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http://dx.doi.org/10.1016/j.ridd.2015.12.018
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1. Introduction

While most children become fluent readers without much effort within a few years of reading instruction, a considerable number of children experience great difficulties in acquiring adequate literacy skills. Developmental dyslexia, a specific learning disability resulting in reading and spelling impairments despite normal intelligence and proper educational instruction (Lyon, Shaywitz, & Shaywitz, 2003; Peterson & Pennington, 2012), is only one possible cause of the reading failure experienced by the heterogeneous group of poor readers (Heim & Grande, 2012; Menghini et al., 2010; Pennington et al., 2012; van Bergen, van der Leij, & de Jong, 2014). Consequently, there have been numerous attempts to classify distinct and coherent cognitive profiles of reading failure. Traditionally, classification studies have divided poor readers into pre-defined categories, based on existing theoretical insights regarding the etiology of the different reading profiles, such as the ‘IQ-reading performance discrepancy’ hypothesis (Rutter & Yule, 1975), the ‘phonological-core variable-difference’ model (Stanovich, 1988) or according to a specific word decoding deficit or a broader decoding-comprehension deficit, i.e., the ‘simple view of reading’ (Gough & Tunnner, 1986). These earlier studies have significantly influenced the ongoing debate on what constitutes dyslexia and have at least supported the possibility to distinguish between a specific reading disabled, or dyslexic profile, and a more general poor, or garden-variety profile (Gough & Tunnner, 1986; Rutter & Yule, 1975; Stanovich, 1988). In more recent years however, new developments in the genetic underpinnings of reading disability added greatly to the current definition of dyslexia as a specific learning disability with a neurobiological origin and strong genetic disposition to develop reading difficulties (Byrne et al., 2006; Castles, Datta, Gayan, & Olson, 1999; Grigorenko, 2001; Pennington & Olson, 2008; Williams & O’Donovan, 2006). Reading problems experienced by dyslexic children are highly heritable and run in families (Pennington & Olson, 2008). Approximately 40–65% of children at familial risk are expected to develop dyslexia (Blomert & Willems, 2010; Pennington & Leffly, 2001; Pennington & Olson, 2008; Scarborough, 1990), indicating that the chances to develop dyslexia, given a dyslexic parent or sibling, amount to at least 10 times the population prevalence (i.e., 4–5%, Blomert, 2005). Although having a familial history of dyslexia is one of the strongest risk factors of the disorder (Thompson et al., 2015), it should not be considered as a pure genetic component (Plomin, Reiss, Hetherington, & Howe, 1994: Rutter & Silberg, 2002). Since at-risk children share both genes and home literacy environment with their close relatives, gene–environment interaction has been studied to understand reading deficits better. It has been shown that children with general impairments across various cognitive abilities are often more specially influenced by environmental risk factors and less by familial risk (Castles et al., 1999; Gayan & Olson, 2001; Grigorenko, 2001; Rack & Olson, 1993; Wadsworth, Olson, Pennington, & Defries, 2000). A neuroimaging study supported two subtypes demonstrating the existence of two distinct brain activation profiles characterizing a primarily genetic poor reader type and an environmentally influenced more generally impaired type (Shaywitz & Shaywitz, 2005). One relevant question now is whether this environmental poor reader subtype can be differentiated from the poor reader subtype with a familial risk of dyslexia based on distinct, underlying cognitive deficit patterns. The present clustering study, for the first time, aims to investigate emerging cognitive subtypes of reading disability related to at-risk status based on history of dyslexia as well as environmental factors. We hypothesize that if the development of reading difficulties of a certain cognitive subtype of reading disability is influenced more strongly by a familial predisposition or alternatively more strongly by environmental risk factors, that this influence will also be reflected in their underlying cognitive deficit pattern. The possibility of identifying unique poor reading profiles will help tailor future diagnostic and intervention efforts more effectively than is currently possible.

Several important cognitive risk factors, or ‘vulnerability markers’, have been identified as important behavioral markers useful for the identification of reading disability. Traditional single deficit accounts of dyslexia point to weakness in phonological awareness (PA) as the core deficit in dyslexia (Adams, 1990; Goswami & Bryant, 1990; Ramus, 2003; Snowling, 2000; Vellutino, Fletcher, Snowling, & Scanlon, 2004). However, more recently a growing tendency emerged to view neurodevelopmental disorders like literacy impairment as the product of various interacting vulnerability markers with distinct, behavioral profiles as a result (Bishop, 2006, 2008; Pennington & Olson, 2008; Snowling, 2008, 2012). Family studies of dyslexia confirm that there is indeed a spectrum of reading disorders (Snowling & Hulme, 2012). Although a single PA deficit explanation of dyslexia is under discussion, it is undisputed that PA is a main characteristic of reading disability (e.g., Ramus et al., 2003; Sunseh & Bowers, 2002). Reading disabled children at increased familial risk for dyslexia show significantly weaker PA skills than non-risk controls (e.g., Carroll, Mundy, & Cunningham, 2014). In turn, both impaired and unimpaired at-risk children show PA problems (Puolakanaho et al., 2008; Snowling, Gallagher, & Frith, 2003), resulting in a parametric increase of reading problems (i.e., healthy < unimpaired at-risk < impaired at-risk: Boets, Wouters, Van Wieringen, & Ghesquiere, 2007; Elbro, Borstrom, & Petersen, 1998; Moll, Loff, & Snowling, 2013; Pennington & Leffly, 2001; Snowling et al., 2003). Although it was always assumed that PA plays a causal role in reading acquisition (Snowling, 2000; Vellutino et al., 2004; Wagner & Torgetsen, 1987), this has been opposed by others who indicated that a PA deficit develops in close relation with the developing reading deficit (Blomert & Willems, 2010; Castles & Colheart, 2004; Castles, Wilson, & Coltheart, 2011; Morais, Cary, Alegria, & Bertelson, 1979; Perfetti, Bell, & Hughes, 1987). Genetic studies also indicated that a PA deficit is associated with a genetic predisposition of dyslexia which, although often differing in severity, can be present in both affected and unaffected family members (Berninger et al., 2006; Snowling, 2008).

Another candidate vulnerability marker of dyslexia is the ability to rapidly name familiar visual symbols such as objects, letters or colors, known as Rapid automated naming (RAN) (Berninger, Abbott, Billingsley, & Nagy, 2001; Berninger et al., 2006; Bowers & Wolf, 1993). RAN is thought to reflect efficient and fast matching of visual and phonological codes and is
profiles without making a priori assumptions on the nature and number of the expected subtypes. An advantage of such data-driven, analysis in large representative samples of children which provides the opportunity to explore distinct etiologies of dyslexia (Byrne et al., 2006; Davis et al., 2001; Gayan & Olson, 2001; van Bergen, de Jong, Maassen, & van der Leij, 2014) and RAN performance shows a similar parametric variation from healthy to at-risk non-dyslexic readers to at-risk dyslexic readers, as does PA (Pennington & Leffly, 2001; Puolakahaho et al., 2008).

A third candidate vulnerability marker of reading disability is letter-speech sound processing (LS). This process can be further divided into two sub-processes: letter-to-sound mapping and automatic letter-sound integration. Letter-to-sound mapping is relatively fast. Most children learn this within months of reading education (Seymour, Aro, & Erskine, 2003). In contrast, automatic integration of letter-speech sound pairs takes much longer, even in normal readers (Froyen, Bonte, Van Atteveldt, & Blomert, 2009; Froyen, Van Atteveldt, Bonte, & Blomert, 2008). Poor readers are significantly slower in processing LS pairs than normal readers over six primary school grades, indicating persistent problems to efficiently and automatically integrate letters to speech sounds (summarized in Blau, Van Atteveldt, Ekkebus, Goebel, & Blomert, 2009; Blau et al., 2010; Blomert & Vaessen, 2009; Blomert, 2011; Froyen, Willems, & Blomert, 2011). This LS learning deficit is already present in a core group of children at-risk for dyslexia before they start to read (Blomert & Willems, 2010). Recent neuroimaging results showed that the reduced neural LS integration activity can be moderately improved by an intensive letter-speech sound coupling training next to regular reading instruction (Zaric et al., 2015). Similar to PA and RAN, at-risk non-dyslexic as well as at-risk dyslexic children were both found to demonstrate slow acquisition of letter-sound speech knowledge (Puolakahaho et al., 2008; Snowling et al., 2003).

A fourth potential reading-related risk factor might be verbal working memory (VWM). Although reduced performance on tasks of verbal working memory involving phonologically represented material are often reported in struggling readers (Beneventi, Tønnessen, Erland, & Hugdahl, 2010; Berninger et al., 2006; Georgiou, Das, & Hayward, 2008), the exact role of a VWM deficit in dyslexia has been disputed. For example, it has been shown that memory span deficits in dyslexic readers are mainly restricted to tasks with a phonological component (Blomert & Vaessen, 2009; Carroll & Snowling, 2004; Felton & Brown, 1990; Tijms, 2004) whereas their performance on tasks without this verbal component (e.g., spatial working memory) is comparable to that of non-dyslexic readers (Jeffries & Everatt, 2004; Kibby, Marks, Morgan, & Long, 2004). This has led to the debate whether VWM impairments are uniquely related to poor reading outcome, independent of phonological problems (Georgiou et al., 2008; McCallum et al., 2006) or not (Tijms, 2004). Nevertheless, a VWM deficit seems to be a candidate vulnerability marker since it predicts reading development (Georgiou et al., 2008; McCallum et al., 2006) and is subject to genetic effects already prior to schooling (e.g., Byrne et al., 2006). VWM deficits are shared by at-risk reading impaired and at-risk non-impaired children (Pennington & Leffly, 2001).

Finally, we return to the validity of an IQ-reading performance discrepancy criterion traditionally used to identify reading disability assuming that the cause of reading failure might vary between low and high IQ readers. Current working definitions have not fully eliminated its use as an exclusionary criterion, defining dyslexia to be unexpected and a ‘specific’ learning deficit (e.g., Lyon et al., 2003). The use of discrepancy criterion in the diagnosis of dyslexia has been challenged from its inception though. Behavioral findings reveal that discrepant and non-discrepant poor readers did not differ in terms of long-term prognosis, severity of their PA deficits or response to intervention (for a review; Jiménez, Siegel, O’Shanahan, & Ford, 2009; Stanovich & Siegel, 1994; Stuebing et al., 2002; Stuebing, Barth, Molfeese, Weiss, & Fletcher, 2009). Practical and psychometric problems such as caused identification of dyslexia solely based on an IQ-reading discrepancy or strict cut-off scores to become (Lyon, 1989, 1995). Moreover, recent neuroimaging studies confirm the lack of validity of the IQ-reading discrepancy definition of dyslexia by showing that regardless of IQ, dyslexic readers show similar functional organization of the brain network involved in phonological processing of print (Simos, Rezaie, Papanicolaou, & Fletcher, 2013; Tanaka et al., 2011). Although the behavioral and neuroimaging profiles of poor readers do not seem to differ as a function of IQ, recent genetic twin studies found that the degree of genetic influences does vary as a linear function of IQ. More specific, reading failure in high-IQ dyslexics are due to substantially genetic causes whereas the etiology of reading failure in low-IQ dyslexics seem to be due to more environmental causes (Knopik et al., 2002; Wadsworth et al., 2000; Wadsworth, Olson, & DeFries, 2010). The existence of two potential etiologies for childhood reading disability was confirmed by a neuroimaging study by Shaywitz and Shaywitz (2005) indicating a predominantly genetic, specifically impaired reading impaired type with IQ scores over 100 and a more environmentally influenced type with IQ scores below 100. Thus, although the same genetic factors could be operating across varying levels of IQ, genetic influence is stronger in high-IQ dyslexics which may require different or more thorough remediation.

Previous poor reader classification attempts focused on the above described cognitive vulnerability markers are rather limited and can be divided into two major categories. The first category employs a top-down, theory-driven approach, classifying subjects into one of the hypothesized subtypes by means of a predefined division (Compton, DeFries, & Olson, 2001; Wolf & Bowers, 1999; Zoubrinetzky, Bielle, & Valdois, 2014). The second type of classification studies employs bottom-up, data-driven, analysis in large representative samples of children which provides the opportunity to explore distinct profiles without making a priori assumptions on the nature and number of the expected subtypes. An advantage of such
data-driven classification over theory-driven studies is that they circumvent possible limitations of the boundaries of the theoretical model and look beyond this adopted framework to find different, emerging subtypes (Heim et al., 2008; King, Giess, & Lombardino, 2007; Morris et al., 1998; Pacheco et al., 2014). Several data-driven studies focused on reading abilities in unselected samples of children, but not on the underlying cognitive deficits (e.g., Buly & Valencia, 2002; Pierce, Katzir, Wolf, & Noam, 2007). Others have investigated reading problems as one manifestation of learning disabilities amongst a widespread range of deficits in general language, reading, perceptual, motoric and other cognitive processes (e.g., Morris et al., 1998; Rourke, 1985; Satz & Morris, 1981). The data-driven classification study by King et al. (2007) did focus on reading impaired children, using PA and RAN as main clustering variables. Results revealed evidence for three impaired subtypes of distinct underlying cognitive deficits, namely one phonological deficit profile, a rapid naming deficit profile and a profile reflecting a double deficit in both domains. Another more recent clustering study by Pacheco et al. (2014) focused on a wider range of cognitive abilities when profiling dyslexic children with normal-range IQ scores, including measures of PA, RAN, VWM and vocabulary. Results suggested a cluster with phoneme deletion and RAN deficiencies and a cluster with phonological processing difficulties (phoneme deletion and digit span) without a RAN deficit. A limitation was the small sample size of 37 children. The present clustering attempt will further explore this broader cognitive spectrum of poor reading across these reading-related vulnerability markers in a much larger and representative sample of grade 2 to 4 poor readers. We will moreover also take into account recent major advances in the genetic underpinnings of reading disability by adding information on the familial occurrence of reading problems in addition to possible environmental influences. Since genetic influence varies with the level of IQ, we moreover included non-verbal IQ as a general cognitive clustering variable not as an inclusion criteria like in the study of Pacheco and colleagues (2014).

1.1. The present study

The present, data-driven clustering study was executed within a large sample of second to fourth grade poor readers between 7 and 10 years old. We aimed to explore the possibility of identifying distinct types of reading disability and investigated whether these cognitive profiles can differentiate between poor readers with a family history of dyslexia and those influenced mainly by environmental factors. We hypothesize that a strong familial risk should be reflected in disabled reading skills and also in underlying cognitive deficits. Cognitive clustering was based on a 200 multi-start K-means approach with the following vulnerability markers of reading failure: phonological awareness (PA), rapid naming (RAN), letter-speech sound processing accuracy and response time (LS) and verbal working memory (VWM) as clustering variables, next to general cognitive abilities of non-verbal IQ and vocabulary. Subsequently, the obtained subtypes were compared on characteristics of familial risk (i.e., first-degree family member with severe childhood reading and spelling difficulties), environmental risk factors (i.e., socio-economic status and reading performance level at schools) and co-morbidity characteristics (i.e., arithmetic deficits and ADHD characteristics).

2. Methods

2.1. Participants

The current sample of poor readers investigated in the present study consisted of 334 children (i.e., 155 girls; n = 134 in grade 2; n = 111 in grade 3, n = 89 in grade 4) obtained from two larger samples:

- The first sample is an unselected school sample of 1717 children (i.e., all children in one classroom were evaluated) enrolled in 13 primary schools spread across 5 different regions in the Netherlands. This sample was originally used for the standardization of the 3DM test battery (i.e., psycho-metric Differential Diagnosis Dyslexia Maastricht (3DM), Blomert & Vaessen, 2009);
- The second sample consisted of 108 children attending regular primary school that were also referred to a specialized dyslexia institute in their region to further investigate their reading or reading-related development. These children were also part of the norming and validation study by Blomert and Vaessen (2009) and tested at the dyslexia institute.

Children were labeled as poor readers if they performed one standard deviation below the mean norm score (i.e., percentile score <16) on a Dutch standardized word reading fluency test of the 3DM test battery (Blomert & Vaessen, 2009). Based on this selection criterion the following poor reading samples emerged:

- In the first sample, 257 children of 1717 met the present’s study poor reader criterion;
- In the second sample, 77 of 108 children were labeled as poor readers.

All children were native Dutch speakers. Note that the Dutch school system approves the use of a handful of reading instruction protocols of which the basic principles of instructions focus on developing awareness of the basic phonological structure of written language in addition to learning the important letter-speech sound relations in isolation and in the context of structured words. As a consequence, the reading instruction and interventions provided at Dutch schools are highly comparable. This is further guaranteed by the national protocol for reading intervention, or the pupil evaluation
system, dictating schools how to monitor and remediate children struggling with their literacy development. An informed consent from all parents/caregivers and permission from the Ethical Faculty Committee was obtained. Age and mean performance (raw and standardized \( z \)-scores) of the poor reader sample on literacy tasks and on the general cognitive and the reading-related tasks subsequently used for clustering are presented in Table 1. We used SPSS (version 21). The overall poor reader sample was on average 8.9 (\( \pm 0.99 \)) years old and showed general cognitive abilities that fell within the average range. They moreover showed a disabled performance on all reading and spelling tasks (confirming the employed poor reader selection criterion). Finally, the poor readers demonstrated poor to low-average PA, LS, RAN and VWM skills.

2.2. Procedure

Children were tested at their school or dyslexia institute in a quiet room. Tasks were administered individually, in a fixed order by trained project workers (except for the classically administered non-verbal IQ). Independent Dutch national norms for the 3DM test battery range from grade 1 to the end of grade 6 of primary school and all subtest were computerized, using a specially designed response-box to precisely measure accuracy and/or response time in milliseconds. Independent Dutch national norms were also available for all other included tasks (norm range included in the task description below) and were all paper-to-pencil tasks. Please note that the performance from all children from the original norming and validation study of the 3DM study by Blomert and Vaessen (2009) were used for the present study. Thus, the 3DM tests (or other included measures) were not administered previously, excluding a possible influence of multiple testing with the same test battery.

2.3. Tasks

2.3.1. Poor reader criterion – Reading fluency (3DM)

Children read aloud as many singly presented words as quickly as possible on 3 levels (high-frequent, low-frequent, pseudo-words, 75 items in 30 s per level on 5 screens, 15 items). Fluency was an overall composite score over 3 levels expressed in number of correct words (items/second) and used to define the poor reader criterion (i.e., the lowest 16th percentile). Test-retest reliability coefficient was 0.95 (reported in the test manual of 3DM).

2.3.2. Initial clustering variables

2.3.2.a. Phonemic awareness – Phoneme deletion (PA 3DM). The task presented 23 pseudo-words via headphones. The child deleted a speech sound at different positions and pronounced the resulting pseudo-word (e.g., “/dauk/ – /d/, what is left?”).
Accuracy (% correct) and response time (seconds/item) was measured but RT was excluded from further analyses due to floor accuracy performance in 1/3 of children (i.e., ≤5 items correct). The task had an internal consistency of 0.85 for accuracy and 0.93 for speed (reported in the test manual of 3DM).

2.3.2.b. Rapid naming (RAN 3DM). The child named items on a sheet (letter and digit task) as quickly as possible (i.e., 2 sheets per task, 15 items per sheet in different order). Response time was mean RT over two sheets. The naming task had a split-half reliability of 0.80 (reported in the test manual of 3DM).

2.3.2.c. Letter-speech sound processing (LS 3DM). Two tasks were used to measure accuracy and response time of letter-speech sound knowledge, a letter-speech sound identification task and a letter-speech sound discrimination task. In the letter-speech sound identification (LSSI) task, a phoneme was presented over headphones (in total 45 unique trials) simultaneously with four letters (or letter combinations) appearing on a screen (e.g., /b/ and ‘b’ ‘d’ ‘t’ ‘p’). The child identified the corresponding letter-speech sound pair by pressing the button matching to the correct letter. Accuracy (% correct) was measured as well as response time (s/item) was measured for both tasks. The accuracy scores of the LSSI and LSSD tasks had an internal consistency of respectively 0.72 and 0.82 and the speed scores of the LSSI and LSSD tasks had an internal consistency of respectively 0.90 and 0.96 (reported in the test manual of 3DM).

2.3.2.d. Verbal working memory (VWM). Digit span (WISC-R) required children to repeat a digit sequence in forward or backward order (de Bruyn, van der Steenen, & van Haasen, 1986). The sequence length increased from 2 to 9 digits and 2 trials per sequence length were offered. The task was stopped if both trials of a given length were incorrect. Accuracy score was the sum of the largest correctly repeated forward and backward sequence. The digit span test had a split-half reliability of 0.78 (reported in the test manual WISC-R). The Dutch version of WISC-R has norms for children between 6 and 16 years of age.

2.3.2.e. Non-verbal IQ. RAVEN-CPM required children to identify missing segments required to complete a larger pattern to measure non-verbal IQ at schools (Van Bon, 1986) For the Dutch version of this task, reliabilities of 0.80 and higher were reported. WISC-R-NL Block design measured IQ at the dyslexia institutes (WISC-R: de Bruyn et al., 1986). Children copied geometric designs with four or nine plastic cubes. The reliability coefficient for the Block Design subtest in a Dutch population was between 0.85 and 0.89. To compare RAVEN and WISC performance, both scores were transformed into an IQ estimate (median = 100, SD = 15). The transformed IQ scores did not differ significantly for RAVEN and WISC, respective values were 98.26 (14.64) and 101.42 (14.35), t(1) = 2.77, p > 0.05. Normative data for RAVEN-CPM ranged from 5 to 11 years of age (for norm range of WISC-R see VWM section above).

2.3.2.f. Receptive vocabulary. Children matched one of four pictures to an orally presented word (RAKIT; Bleichrodt, Drenth, Zaal, & Resing, 1988). Accuracy was expressed in % correct (maximum items 60). The test has an internal consistency of 0.81 (reported in the test manual Rakit). Dutch norms were available for children from 4 to 11 years of age.

2.3.2.g. Baseline response time (3DM). An animated figure (20 in total) appeared in one of four squares on the screen. The child identified the location of the figure by pressing the corresponding button as fast as possible. The task has an internal consistency of 0.93 (reported in the test manual of 3DM).

2.3.3. Comparing clusters: Reading and spelling

2.3.3.a. Reading fluency. Children read aloud single words as quickly as possible in the presented time: One-Minute test (OMT: high and low frequent words, 1 min, Brus & Voeten, 1973), Klepel (pseudo-words, 2 min, van den Bos, Lutje Spelberg, Scheepstra, & de Vries, 1994) and 3DM reading fluency used for the initial poor reader selection (for a description see above). Fluency was expressed in correctly read words (items/second). Test-retest reliabilities are reported to be over 0.80 for the OMT, 0.89 for the Klepel and 0.95 for 3DM reading fluency. Dutch norms ranged from grade 1 to the first class of secondary school for both the OMT and the Klepel (for norm range of 3DM see Procedure section above).

2.3.3.b. Spelling. Two spelling tasks were administered. Spelling-to-dictation (PI-dictee, Geelhoed & Reitsma, 1999): 135 words were dictated to the child in a sentence (i.e., from simple words to multi-syllable words with complex spelling rules) until 8 or more incorrect spellings in one block of 15 sentences were made. The internal consistency of this standardized test is between 0.90 and 0.96 (reported in the PI-dictee test manual). Dutch norms are available for children from grade 1 to 6. Computerized spelling task (3DM): an auditory word (e.g., /boom/) was presented via headphone simultaneously with a visual, incomplete word (e.g., /b___m/). Below the incomplete word, four options of the missing part were presented (e.g., ‘oo’, ‘a’, ’o’, ‘aa’). The child selected the option to complete the word correctly by pressing a corresponding button. Accuracy was expressed in % correct. The task accuracy measure has an internal consistency of 0.80 (reported in the manual of 3DM).
2.3.4. Comparing clusters: Reports on familial risk, co-morbidity and environmental factors

The emerging subtypes were compared on familial risk for dyslexia, environmental risk factors (school performance level, socio-economic status (SES) based on parental education and on neighborhood status) and co-morbidity characteristics were obtained through parent-ratings on a reading-status questionnaire (FADD, Blomert, unpublished).

2.3.4.a. Familial risk for dyslexia. A child was defined as at-risk for dyslexia if at least one first-degree family member (i.e., a parent or sibling) indicated to have suffered from severe childhood reading and spelling difficulties.

2.3.4.b. Environmental factors. Parental Education Level SES. Parental education was assessed on a 5-point scale ranging from ‘primary school’ to ‘vocational bachelor or university level’ (i.e., 5 levels ranging from low, low average, average, high average or high parental education). For the current analyses we defined two socio-economic status categories: poor parental education for low to low average and normal parental education for the remaining level. Neighborhood SES. This neighborhood social status score was adopted from extensive research by the Netherlands Institute for Social Research (Knol, 2012). A factor analyses defined the status score of each Dutch postal code area based on four characteristics of neighborhood residents within each area: mean income, percentage low incomes, percentage of low educated residents and percentage of unemployed residents (i.e., scores ranged from −7.25 to 3.19, a low score indicates low neighborhood status, average score in 2010 was of 0.17). For the current analysis, we defined two neighborhood social status categories: poor neighborhood status with a status score of 1 standard deviation below the mean norm and a normal neighborhood status.

2.3.4.c. School reading performance level. Within the original, unselected grade 1–6 school sample (n = 1717) from which the present grade 2–4 poor reader sample (n = 334) was selected, we evaluated the overall proportion of poor reading children based on the 3DM reading fluency performance of 1 SD below the norm (i.e., the 16th percentile). Overall proportions of poor readers were set for all 13 participating schools. Next, we ranked the 13 schools based on a median cut-off (i.e., ≤13.33%) we divided the schools in two categories: lower reading level schools and higher reading level schools. More specific, 7 schools were labeled as low proportion of poor reader schools and 6 schools as high proportion of poor reader schools.

2.3.4.d. Co-morbidity characteristics. Parent-ratings on the FADD questionnaire indicated presence/absence of severe arithmetic difficulties for a minimum period of 6 months. Parent-ratings on the AVL questionnaire (Scholte & van der Ploeg, 2005) consisting of 18 questions on attention, concentration and (hyper)activity indicated whether the child exhibited behavioral symptoms of ADHD for the last 6 months or more. The overall score resulted in three categories; ADHD symptoms were low, moderate or considerably present, of which the latter category was considered ADHD behavior.

3. Results

3.1. Data preparation

The present data-driven, clustering approach investigated the possibility to identify distinct types of underlying cognitive deficit patterns in a large sample of poor readers and subsequently explored the relation between these subtypes and familial risk and environmental factors contributing to reading failure. This statistical classification approach was previously employed to successfully identify distinct cognitive subtypes within an independent heterogeneous sample of children with arithmetic difficulties (Bartelet, Ansari, Vaessen, & Blomert, 2014). We now apply it in the context of reading disability. Note that all variables scores were nationally normed measures (i.e., normally distributed and corrected for age ensuring that a child’s performance is compared to children that received a similar amount of reading instruction). For all analyses, the scores on each measure were transformed into standardized z-scores to ensure that differences in measurement scale did not influence the results.

In order to distinguish theoretically meaningful latent variables we first conducted an exploratory factor analysis (principal components with oblimin rotation) on the eight reading-related vulnerability markers: phonological awareness accuracy (PA), rapid naming speed (RAN) letters and digits, letter-speech sound (LS) identification and discrimination accuracy and response time, and verbal working memory accuracy (VWM). LS response time tasks were corrected for possible individual motor response differences (i.e., tendency to work fast/slow resulting in a possible speed/accuracy trade-off) prior to the factor analysis. Specifically, we regressed baseline response time (with a similar design to the LS tasks) on the LS RT to subsequently compute corrected RT residuals. This baseline RT tasks was thus not separately included in the factor or subsequent analyses. Note that post hoc comparisons revealed that baseline response time was comparable across emerging clusters (type 1: $M = -0.07$, $SD = 1.08$, type 2: $M = -0.16$, $SD = 1.03$, type 3: $M = -0.15$, $SD = 1.04$ and type 4: $M = -0.21$, $SD = 1.19$, $F(3, 330) = 0.22$, p = 0.88). The factor solution for the current reading disabled sample resulted in the following five theoretically relevant factors with eigenvalues of at least 0.70 (Jolliffe, 1986) and a communality estimate of at least 0.70 (MacCallum, Widaman, Zhang, & Hong, 1999): phonological awareness (PA; accuracy), rapid naming (RAN; reaction time on letters and digits), letter-speech sound accuracy (LS accuracy; on identification and discrimination), letter-speech sound fluency (LS rt; reaction time on identification and discrimination) and verbal working memory (VWM;
3.2. Identifying clusters of reading disability

Nonverbal IQ, vocabulary performance and the five identified factors were entered as variables in the subsequent clustering analyses conducted with SPSS (version 21). We firstly conducted an Agglomerative hierarchical clustering approach (Ward’s method) as a pre-processing step in our data analyses. Specifically, this bottom-up clustering approach provides us with the mechanism for monitoring data merging, on the one hand and, on the other, a strategy to determine the optimal number of clusters representing the data. The analysis initially starts with every single case within a separate cluster, considering each individual case separate from all the others. Subsequently, cases are merged (i.e., agglomerated) together into new clusters based on highest similarity in each following iteration. Similarity is based on the lowest increase in error of sum-of-squares or in other words the smallest distance to the cluster center (i.e., SUMD). This merging process is continued until all of the data is merged into one cluster (Field, 2005; Sarstedt & Mooi, 2014). Thus, clusters generated in early stages are nested in those generated in later stage which is reflected in the hierarchy of the clustering tree plot. The results can be described in a dendrogram (Fig. 1 – bottom) and a plot of the mean sum-of-squares (mean SUMD) as a function of $K$ number of clusters (Fig. 1 – top). The dendrogram visualizes the hierarchy within the final cluster in which each iteration merge is represented by a binary tree. Interpreting the height between the different nodes and its leaf indicated that the largest threshold distances can be seen at four clusters. The optimal four cluster solution best representing the data was further confirmed by the observed ‘elbow’ at $K = 4$ (i.e., while the average sum-of-squares monotonically decreased for increasing $K$ (1 to 4), this decrease reduces markedly for $K > 4$) in the mean SUMD plot. Hierarchical agglomerative clustering thus provided us with the optimal indication of the number of clusters represented in our data. However, using this approach for clustering alone, although leading to a cluster grouping of our data, only leads to one possible (and possibly sub-optimal) solution. Hence, we applied a second step, employing $K$-means clustering.

Iterative partitioning $K$-means multi-start (200 runs) clustering approach (Euclidean distance) provided us with a mechanism for assessing different configurations (cluster solutions) based on multiple initializations to subsequently identify the cognitive profiles of these four clusters estimated to be the best fit of the current data. $K$-means clustering performs an iterative fitting process to form the specified optimal number of clusters ($K = 4$). This is done by selecting a set of cluster seeds as a first guess of the starting point as the initial cluster centers. Subsequently, each case is assigned to one of the four clusters it most closely resembles, after which the cluster mean is re-calculated. This process continues until the clusters stabilize. The choice of this initial starting point for clustering can greatly affect the resulting classification which may be a sub-optimal representation of the data (King et al., 2007). However, by re-sampling the data 200 times using MATLAB (2001), using randomly chosen, cluster centers to start clustering from, we circumvent the well-known problem of local optima. We moreover can guarantee that the final four cluster solution obtained (i.e., the most optimal fit out of 200 runs) represented the clustering result with the minimal sum of average squared distances within clusters. In other words, we would ensure maximal cluster stability and obtain the result that reduces the Euclidean distance most effectively, represents the global optima or best fit of the data (Steinley, 2006). The best fitting four-cluster solutions resulting from this multi-start $K$-means revealed the following cognitive profiles of reading disability (standardized $z$-scores of the mean performance on the classification variables per cluster are presented in Table 3, for a visual representation of this data see Fig. 2):

**Type I** ($n = 86$) holds poor readers with average general cognitive IQ, vocabulary and average performance on PA, RAN, LS and VWM ($z$-scores ranging between 0.46 and $–0.50$). This type thus showed difficulties with reading without showing underlying general of reading-related cognitive deficits and was therefore labeled *reading-only impaired readers*. 

### Table 2

Results of exploratory factor analysis.

<table>
<thead>
<tr>
<th>Factors</th>
<th>PA acc</th>
<th>RAN digits</th>
<th>LSacc</th>
<th>LSrt</th>
<th>VWM acc</th>
<th>Eigen values</th>
</tr>
</thead>
<tbody>
<tr>
<td>PA acc</td>
<td>0.89</td>
<td>0.01</td>
<td>0.10</td>
<td>–0.05</td>
<td>–0.20</td>
<td>2.39</td>
</tr>
<tr>
<td>RAN digits</td>
<td>–0.19</td>
<td>0.81</td>
<td>0.08</td>
<td>–0.01</td>
<td>0.27</td>
<td>0.79</td>
</tr>
<tr>
<td>RAN letters</td>
<td>0.18</td>
<td>0.86</td>
<td>–0.7</td>
<td>0.04</td>
<td>0.22</td>
<td>0.81</td>
</tr>
<tr>
<td>LSSI acc</td>
<td>0.11</td>
<td>0.02</td>
<td>0.90</td>
<td>0.17</td>
<td>0.13</td>
<td>0.82</td>
</tr>
<tr>
<td>LSSD acc</td>
<td>–0.04</td>
<td>–0.03</td>
<td>0.80</td>
<td>–0.22</td>
<td>–0.12</td>
<td>0.76</td>
</tr>
<tr>
<td>LSSI rt</td>
<td>–0.10</td>
<td>0.03</td>
<td>0.09</td>
<td>0.76</td>
<td>–0.15</td>
<td>0.76</td>
</tr>
<tr>
<td>LSSD rt</td>
<td>0.03</td>
<td>0.00</td>
<td>–0.06</td>
<td>0.90</td>
<td>0.03</td>
<td>0.81</td>
</tr>
<tr>
<td>VWM acc</td>
<td>0.18</td>
<td>0.01</td>
<td>–0.02</td>
<td>0.08</td>
<td>–0.91</td>
<td>0.90</td>
</tr>
<tr>
<td>Eigen values</td>
<td>2.39</td>
<td>1.88</td>
<td>1.13</td>
<td>0.93</td>
<td>0.80</td>
<td></td>
</tr>
</tbody>
</table>

*Note.* Principal component analysis with oblimin rotation. PA, phonological awareness – phoneme deletion; RAN, rapid naming; LSSI, letter–speech sound identification; LSSD, letter–speech sound discrimination; VWM, verbal working memory; acc, accuracy; rt, response time.
Type 2 \((n = 76)\) showed low IQ and vocabulary (z-scores of \(-0.84\) and \(-0.81\), respectively) which were in correspondence to their low-to-poor performance across all reading-related skills, indicated by impaired PA (z-score of \(-1.35\)), low-average RAN (z-score between \(-0.75\) and \(-0.84\)), low-average-to-poor LS accuracy and RT (z-score between \(-0.52\) and \(-1.07\)) and poor VWM (z-score of \(-0.99\)). Thus, this type consisted of general poor readers.

Type 3 \((n = 79)\) poor readers exhibited low-average IQ scores (z-score of \(-0.46\)) and normal vocabulary (z-score of 0.06) next to a specific impairment in LS processing (Accuracy z-score between \(-1.09\) and \(-1.54\)) and PA (z-score of \(-1.16\)). Their performance on RAN and VWM performance where in the average norm range (z-score between \(-0.55\) and \(-0.10\)). Note that these children performed average on LS processing speed. This type was labeled PA-LS specific poor readers.

Type 4 \((n = 93)\) poor readers showed average IQ and vocabulary (i.e., a z-score of 0.55 on both measures) discrepant to their impaired RAN and PA performance (respective z-scores were \(-1.38\) to \(-1.47\) and \(-0.94\)). The performance on LS accuracy, LS RT and VWM was in the low average range (z-scores ranging between \(-0.49\) and \(-0.72\)). This type was labeled as PA-RAN specific poor readers. Note. A cross-validation confirmatory analysis (i.e., randomly split the present poor reader sample in half to repeat the clustering approach for each sub-sample using the 200 multi-start procedure) yielded the same four cognitive types of poor reading as described above. To further confirm that a four-cluster solution was the best fit for the present data, we also explored a five and six-cluster solution. Both analyses revealed the same important clusters as four-cluster solution, and although additional cluster(s) emerged, none corroborated any poor reader types predicted by theoretically relevant models of poor reading (e.g., no additional speed/RAN-only disabled type of poor reading, predicted by the double-deficit hypothesis), confirming the validity of the emerging optimal four cluster solution.

Fig. 1. Identifying optimal number of clusters. (Top) – Total sum-of-squared within cluster distances (SUMD) as a function of k-number of clusters. Note that the total SUMD shows an ‘elbow’ at \(K = 4\) and that after that the decrease of total SUMD is markedly reduced, indicating four as the optimal number of clusters representing the data. (Bottom) – Dendrogram plot of the agglomerative hierarchical binary cluster tree (Wards linkage, Euclidean distance). The Dendrogram is collapsed over lower branches to increase visibility of the plot (some nodes in the plot thus correspond to more than one data point). The largest threshold distances (i.e., height between the node and the leaf) can be seen at four clusters.
3.3. Comparison of types of poor readers on clustering variables

Validation and interpretation are the two last steps of a clustering process. We firstly compared the obtained four types of poor reading on the initial cluster variables to be able to investigate on which specific clustering variables significant differences occurred between the poor reading subtypes. The subtypes of poor reading were compared on the clustering variables by means of analyses of variance (ANOVA). Type 1 (reading-only impaired reader) performed within the average range on all general cognitive and markers. Due to the already extensive nature of the results section, we list the most interesting significant differences between the other three subtypes of poor readers (Table 3 summarizes the comparisons and post hoc analyses, Bonferroni-corrected).

The three poor reader types in which we identified variable underlying patterns of cognitive deficits all showed a PA deficit, with type 2 (general poor readers) showing the most impaired PA skills. Significantly poorer LS accuracy was characteristic of type 3 (PA-LS specific poor readers), while type 4 (RAN-PA specific poor readers) and type 2 (general poor readers) did not differ. A significant RAN deficit was specific for type 4 (PA-RAN poor readers) revealing an inferior performance as compared to type 3 (PA-LS specific poor readers) and type 2 (general poor readers). Less impaired LS RT was found for type 3 (PA-LS specific poor readers) compared to type 2 (general poor readers) and type 4 (PA-RAN specific poor readers). Low VWM was characteristic of type 2 (general poor readers), while type 3 (PA-LS specific poor readers) and type 4 (PA-RAN specific poor readers) showed no difference. As for the general cognitive performance, type 3 (PA-LS specific poor readers) and type 4 (PA-RAN specific poor readers) IQ performance was within average range with the later showing superior IQ skills. The general poor readers showed below average IQ. Finally, Vocabulary was significantly better in type 4 (PA-RAN specific poor readers) than the general poor readers but not as compared to type 3 (PA-LS specific poor readers).

3.4. External validity of the obtained poor reading types

Next, the obtained poor reading subtypes were compared on external validation variables, independent of the initial classification variables. First, age and reading and spelling fluency of the four subtypes were compared by means of an ANOVA (Table 4 lists mean z-scores and standard deviations per subtype and the comparisons and post hoc analyses, Bonferroni-corrected). The reading-only impaired type was significantly younger (i.e., 5 months) than the other three types of poor reading that did not differ. However, there was no significant difference between the proportion of children attending grade 4, grade 5 or grade 6 between the subtypes. This subtype also showed significantly less impaired reading fluency skills on all three reading fluency measures than the other three types that did not differ in their severity of reading deficit. This poor reader type actually performed low average both spelling tasks, while the other three types that did not differ significantly, performed below average.

Second, familial risk for dyslexia and environmental risk factors (i.e., SES based on parental education, SES based on social neighborhood status and poor school performance level) expressed in proportions were compared (Table 5 lists the proportions per subtype in addition to the chi-square comparison). The PA-RAN specific deficit poor readers (type 4) showed a significantly larger familial risk (63%) as compared to the other three types that did not differ from each other (19% in type 1, 24% in type 2 and 20% in type 3). Please note that information for 7 children in the PA-RAN type was missing. Familial risk

Table 3
Mean performance and standard deviations on general cognitive variables and reading-related vulnerability markers per poor reader type.

<table>
<thead>
<tr>
<th>Type</th>
<th>Type 1 Read-only</th>
<th>Type 2 Gen poor</th>
<th>Type 3 PA-LS</th>
<th>Type 4 PA-RAN</th>
<th>F-value</th>
<th>Post hoc</th>
</tr>
</thead>
<tbody>
<tr>
<td>n</td>
<td>86</td>
<td>76</td>
<td>79</td>
<td>93</td>
<td></td>
<td></td>
</tr>
<tr>
<td>IQ</td>
<td>0.01</td>
<td>0.01</td>
<td>0.01</td>
<td>0.01</td>
<td>46.07*</td>
<td>1 &gt; 3 &gt; 4</td>
</tr>
<tr>
<td>Vocabulary</td>
<td>0.46</td>
<td>0.46</td>
<td>0.46</td>
<td>0.46</td>
<td>46.49*</td>
<td>1 &gt; 3 &gt; 4</td>
</tr>
<tr>
<td>LSSI acc</td>
<td>0.14</td>
<td>0.14</td>
<td>0.14</td>
<td>0.14</td>
<td>97.11*</td>
<td>1 &gt; 3 &gt; 4</td>
</tr>
<tr>
<td>LSSI rt</td>
<td>–0.50</td>
<td>–0.50</td>
<td>–0.50</td>
<td>–0.50</td>
<td>189.21*</td>
<td>1 &gt; 3 &gt; 4</td>
</tr>
<tr>
<td>VWM</td>
<td>0.17</td>
<td>0.17</td>
<td>0.17</td>
<td>0.17</td>
<td>146.21*</td>
<td>1 &gt; 3 &gt; 4</td>
</tr>
</tbody>
</table>

Read-only, reading-only impaired readers; Gen Poor, general-poor readers; PA, phonological awareness – phoneme deletion; RAN, rapid naming; LSSI, letter–speech sound identification; LSSD, letter–speech sound discrimination; VWM, verbal working memory; acc, accuracy; rt, response time; performance of all tasks expressed in standardized z-scores.

* p < 0.01. For a visual representation of the data see Fig. 2.
Fig. 2. Visual representation of the cognitive profiles of the poor reader subtypes. Average performance expressed in standardized z-scores (x-axis) with standard error bars on the general cognitive variables and reading-related vulnerability markers (y-axis) per poor reader type. Voca, vocabulary; VWM, verbal working memory; PA, phonological awareness – phoneme deletion; RAN, rapid naming; dig, digits; let, letters; LSSI, letter-speech sound identification; LSSD, letter-speech sound discrimination; acc, accuracy; RT, response time. For a summary of z-scores, standard deviations and post hoc analyses (Bonferroni-corrected) see Table 3.
was based on at least one first-degree family member (sibling or parent) suffering from severe childhood literacy difficulties since official dyslexia diagnoses in parental generations are rare in the Netherlands. We did however have information on proportions of impaired first-degree family members who also had an official dyslexia diagnosis and results showed that official diagnosis of either a sibling or parent was more common in PA-RAN poor readers (50%) compared to the other poor reader types (19% in type 1, 22% in type 2 and 31% in type 3). The general poor and PA-LS specific poor readers showed a significantly higher proportion of children with a poor SES based on parental education than the PA-RAN specific and reading-only impaired poor readers, respective proportions were 69% (type 2), 68% (type 3), 24% (type 4) and 33% (type 1).

Similarly, the general poor and PA-LS specific poor readers showed a significantly higher proportion of children with poor social SES based on neighborhood social status than the PA-RAN specific and reading-only impaired poor readers, respective proportions were 50% (type 2), 51% (type 3), 31% (type 4) and 33% (type 1). The information of SES was missing for respectively proportions were 50% (type 2), 51% (type 3), 31% (type 4) and 33% (type 1). The information of SES was missing for one poor reader type (19% in type 1, 22% in type 2 and 31% in type 3). The general poor and PA-LS specific poor readers showed a significantly higher proportion of children attending school with a lower school reading performance level than the PA-RAN specific and reading-only impaired poor readers, respective proportions were 69% (type 2), 68% (type 3), 24% (type 4) and 33% (type 1).

Finally, gender (proportions of girls) and co-morbidity characteristics (i.e., arithmetic difficulties and ADHD) were compared. There was no difference in gender between the poor reader types, showing similar proportions of girls: reading-only impaired reader type (50%, 21 of 42), general poor (51%, 29 of 57), PA-LS specific (43%, 34 of 79) and PA-RAN specific (47%, 44 of 93), \( \chi^2(3) = 1.30, p = 0.73 \). Proportions of parental concern on arithmetic difficulties was very small and similar across poor reader types: reading-only impaired reader type (6%, 3 of 42), general poor (7%, 4 of 57), PA-LS (8%, 6 of 79) and PA-RAN (7%, 6 of 86), \( \chi^2(3) = 0.23, p = 0.97 \). The number of children with a formal ADHD diagnosis was very small and did not differ between types: reading-only impaired reader type (0%), general poor (1%, 1 of 76), PA-LS specific (0%) and PA-RAN specific (4%, 3 of 86), \( \chi^2(3) = 5.70, p = 0.13 \).

---

**Table 4**

Mean reading and spelling performance per poor reader type.

<table>
<thead>
<tr>
<th>Poor Reader Type</th>
<th>Type 1</th>
<th>Type 2</th>
<th>Type 3</th>
<th>Type 4</th>
<th>F-value</th>
<th>Post hoc</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age</td>
<td>M</td>
<td>SD</td>
<td>M</td>
<td>SD</td>
<td>M</td>
<td>SD</td>
</tr>
<tr>
<td>Reading fluency in correct items/second</td>
<td>OMT</td>
<td>1.40</td>
<td>0.60</td>
<td>1.62</td>
<td>0.56</td>
<td>1.65</td>
</tr>
<tr>
<td>Spelling fluency in correct items</td>
<td>OMT</td>
<td>1.40</td>
<td>0.50</td>
<td>1.42</td>
<td>0.55</td>
<td>1.42</td>
</tr>
</tbody>
</table>

---

**Table 5**

Proportions of familial and environmental risk factors per poor reader type.

<table>
<thead>
<tr>
<th>Poor Reader Type</th>
<th>Type 1</th>
<th>Type 2</th>
<th>Type 3</th>
<th>Type 4</th>
<th>( \chi^2 )</th>
<th>Comparison</th>
</tr>
</thead>
<tbody>
<tr>
<td>Fam Risk</td>
<td>%</td>
<td>n</td>
<td>%</td>
<td>n</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Par SES</td>
<td>%</td>
<td>n</td>
<td>%</td>
<td>n</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Soc SES</td>
<td>%</td>
<td>n</td>
<td>%</td>
<td>n</td>
<td></td>
<td></td>
</tr>
<tr>
<td>School read</td>
<td>%</td>
<td>n</td>
<td>%</td>
<td>n</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

---

Read-only, reading-only impaired readers; Gen Poor, general poor readers; PA-LS, PA-LS specific poor readers; PA-RAN, PA-RAN specific poor readers; Fam Risk, familial risk for dyslexia; Par SES, parental education SES; Soc SES, social neighborhood status; School read, lower school reading performance level; \( \chi^2 \), chi-square test.

* \( p < 0.05 \).
** \( p < 0.01 \).
4. Discussion

The present data-driven classification approach identified profiles of cognitive deficits in a large sample of grade 2–4 poor readers and subsequently explored the relation between these cognitive profiles and familial history risk and environmental risk factors contributing to reading failure. Four stable subtypes of reading disability emerged from cluster analyses: the reading-only impaired readers (type 1), the general poor readers (type 2), the PA-LS specific poor readers (type 3) and the PA-RAN specific poor readers (type 4). They are based on the following four cognitive, candidate risk factors known to mark vulnerability for reading failure: phonological awareness (PA), rapid naming (RAN), letter–speech sound processing (LS), and verbal working memory (VWM), next to general cognitive abilities (i.e., IQ and vocabulary). The reading-only impaired readers (type 1) show no impairments on any clustering variables despite their reading failure. The other three identified types of poor reading (general poor readers, PA-LS specific, PA-RAN specific) share a PA deficit and reveal a specific and unique deficit pattern across the other cognitive clustering variables. The general poor readers (type 2) demonstrate a general impaired performance on all vulnerability markers in accordance with their inferior general cognitive skills. The PA-LS specific poor readers (type 3) exhibit a specific deficit in LS in addition to the shared PA deficit, with average RAN, low average IQ and average vocabulary. The PA-RAN poor readers (type 4) reveal impaired PA and RAN, low average LS in combination with average IQ and vocabulary. The reading-only impaired type 1 poor readers are significantly younger (5 months) than the other three poor reader types and attend schools classified as lower reading performance schools (i.e., 69% of children attend one of the 13 participating schools ranked as lower reading performance schools compared to approximately 45% in the other three subtypes of poor reading). The general poor readers (type 2) and the PA-LS specific poor readers (type 3) are characterized by overall poorer SES based on poorer parental education (i.e., two-third of poor readers are characterized by a socio-economic status level within the low to low-average categories, which was around one-third in the PA-RAN specific and reading-only impaired poor readers) and poorer neighborhood social SES (i.e., half of the general poor and PA-LS specific poor readers show a poor social status score of 1 SD below the norm as compared to one-third in the PA-RAN specific and reading-only impaired poor readers). On the other hand, the poor readers in the PA-RAN specific profile (type 4) are at a significant higher familial risk for dyslexia (i.e., two-third of the PA-RAN specific poor readers have one or more first-degree family member(s) with severe reading difficulties, which was only around one-fifth of poor readers in the other three profiles). Finally, the four subtypes show no clinically significant co-morbidity (i.e., occurrence of ADHD and arithmetic problems was very low and non-discriminative).

The present results confirmed the existence of heterogeneity at the level of cognitive deficits in reading disability (Menghini et al., 2010; Pennington et al., 2012; van Bergen, van der Leij et al., 2014) and support the view that it is possible to identify distinct poor reading subtypes within the heterogeneous group that constitutes reading disabled children (e.g., Heim & Grande, 2012; Heim et al., 2008; King et al., 2007; Morris et al., 1998; Pacheco et al., 2014). The reading-only impaired type 1 poor readers without underlying general cognitive deficits seems puzzling. These poor readers score within the normal standardized range on all reading-related and general cognitive abilities despite their reading difficulties. A closer look reveals that this subtype has significantly less impaired reading fluency skills than the other three subtypes with cognitive deficits underlying their reading difficulties, and also is the only poor reader type scoring within average range on spelling. Results show that these poor readers are significantly younger than in the other three subtypes, making it possible that specific underlying cognitive deficits are not yet revealed. Although younger, type 1 poor readers however did not attend a significantly lower school grade, indicating that their non-specific reading deficit is not due to not yet being exposed to higher grade reading instruction. These poor readers did however attend schools with a lower reading performance level based on high proportions of poor readers at that school (i.e., two-thirds of this subtype compared to 44% in type 2, 43% in type 3 and 46% in type 4, although the proportion of the latter group should be interpreted with care since PA-RAN poor readers were tested at their dyslexia institute and school information was missing in 40% of this subtype). As for the comparison between the other three subtypes, although we acknowledge that factors beyond the scope of the current study may explain why type 1 children showed poor reading skill in the absence of other deficits and adequate intellectual ability, the school reading performance level may indicate that a possible explanation might lie in inadequate reading instruction at school, which seems more easily counterattacked by more adequate or intensive instruction.

The other three identified poor reader profiles did show a unique and distinct cognitive deficit pattern underlying their reading problems. Firstly, all three subtypes of poor reading share a PA deficit which is in line with findings from other classification attempts (e.g., Fletcher et al., 1994; Morris et al., 1998) and corroborates the phonological-core variable difference model (Stanovich & Siegel, 1994). This model suggests that poor readers with lower intellectual capacities and educational opportunities (i.e., IQ-reading congruent type) are more likely to show a more generally impaired profile across general and reading-related abilities (i.e., type 2 in the current study) compared to those disabled readers with relatively higher IQs (i.e., IQ-reading discrepant type) with a more specific reading-related deficit (i.e., type 3 and 4). Although this model predicts that the phonological deficit of the IQ-reading discrepant poor readers would be more severe, the current findings indicated that the PA deficit in the general poor readers was worse consistent with their overall cognitive deficit profile. Nevertheless, results are in compliance with various reports that show that all poor reading types are well predicted by PA deficits, irrespective of IQ level (e.g., Fletcher et al., 1994; Stanovich, 1991; Stanovich & Siegel, 1994). In line with this are the results of recent neuroimaging studies confirming that brain activation patterns during phonological processing of print are similar for low-IQ and high-IQ poor readers (Simos et al., 2013; Tanaka et al., 2011). The present result moreover supports the generally accepted reciprocal correlation between phonological awareness and reading (Blomert & Willems,
Next to a shared PA deficit, a differential expression of underlying cognitive deficits across other domains was found in these three poor reader subtypes. The generally impaired poor readers (type 2) reveal a rather poor and non-differentiated cognitive profile across all reading-related risk markers and general cognitive skills (being the only profile with impaired verbal working memory, for similar VVM results in general poor readers see for instance Daal & Leij, 1999; De Jong, 1998). In contrast, the two specifically impaired poor reader profiles each show a unique reading-related deficit, in addition to the common PA deficit. More specifically, the PA-LS specific poor readers show poor letter-speech sound accuracy processing skills, next to a PA deficit. The PA-RAN specific type demonstrates additional impaired rapid naming skills, suggesting a relation between PA and RAN. Overall, we can conclude that these results do not directly fit into a double-deficit framework proposed by Wolf and Bowers (1999). One would expect a PA-only deficit type, a RAN-only deficit type and a type showing deficits in both domains (for classification results pointing in this direction see King et al., 2007; Morris et al., 1998). The double-deficit hypothesis expects the two deficits to be independent and additive, resulting in a more severe form of dyslexia in the double-deficit subtype as compared to the single deficit subtypes. However, the present results reveal no ‘pure’ RAN-only core deficit type, independent of PA problems (corroborating the findings of other behavioral studies directly investigating the main assumptions of the double-deficit hypothesis; Badian, 1997; Bowers & Ishaïk, 2003; Pennington, Cardoso-Martins, Green, & Leffly, 2001; Vaessen et al., 2009; Vukovic & Siegel, 2006; Wagner & Torgesen, 1987). Pennington et al. (2001) for example only found 1 child within their sample of 71 dyslexic readers with a RAN specific impairment. Similarly, Vaessen et al. (2009) indicated that only 5% of their sample showed RAN difficulties without affected PA skills, leaving them to conclude that RAN is merely a different (speeded) manifestation of the same underlying deficit in the phonological domain, with the unique addition that RAN problems might reflect the inefficient and slower cross-modal matching of visual-orthographic codes (also see Araújo et al., 2011). The present data cannot contribute directly to the discussion on whether RAN reflects cross-modal ability uniquely and independent of phonological processing. Nevertheless our results demonstrate that RAN is a useful cognitive clustering variable, able to differentiate between distinct cognitive profiles within the heterogeneous sample of poor readers, where traditional PA tasks alone cannot.

The finding of an LS specific deficit, next to a shared PA deficit, in our PA-LS specific poor readers (type 3) is in line with the few previous studies focusing on this vulnerability marker. Learning letter-sound pairs is found to be an immediate problem for a substantial part of kindergarten children at-risk for reading failure, already before reading instruction (Blomert & Willems, 2010). Moreover, older poor readers are slower in LS mapping skills than normally developing peers up to the end of primary school (Blomert & Vaessen, 2009). In fact, while normal readers improve this skill up to grade 6, poor readers show a premature halting after grade 3. This extended weakness or even inability to efficiently map letters-sound pairs in dyslexic children is further confirmed by recent brain imaging studies (Blau et al., 2010; Froyen et al., 2011) and even indicated far beyond primary school in dyslexic adults (Blau et al., 2009; for a summery see Blomert, 2011). Although LS processing is a seemingly important vulnerability marker of poor reading it has, to our knowledge, never been included in previous poor reading classification studies (Fletcher et al., 1994; King et al., 2007; Morris et al., 1998; Pacheco et al., 2014). At least, the present result of a distinct poor reader type with LS problems indicates the importance of including such a differentiating measure in future classification research, especially given the information it can provide for subsequent diagnostic and intervention efforts.

Finding distinct cognitive patterns of deficits in three of the four emerging poor reader profiles suggests a possible difference in underlying etiology through which these three subtypes arrive at their similar poor reading and spelling levels. Our results are in line with previous findings of a primarily genetic poor reader type and an environmentally influenced impaired type (Shaywitz & Shaywitz, 2005). The general poor readers (type 2) and the PA-LS specific poor readers (type 3) are characterized by environmental risk factors. Both subtypes of poor reading show significantly poorer socio-economic status (SES) on two measures generally agreed to cover the three most influential components of SES (i.e., the cultural component of parental education, the social component of labor market position and the economical component of family income; Israel, Beaulieu, & Hartless, 2001). More specific, around two-thirds of the children in type 2 and 3 come from families with a low to low-average parental education (compared to one-third in the PA-RAN and reading-only impaired poor readers). In accordance, they also reveal a poorer neighborhood social status composite score based on average family education, income and occupation status of the child’s area of residence (i.e., half of the children scored 1 SD below the norm as compared to one-third of the children in the reading-only impaired type and PA-RAN specific profiles). The PA-RAN specific poor readers (type 4) on the other hand exhibit a stronger familial predisposition to the reading failure they experienced: more than two-thirds of children have one or more first-degree family members with dyslexia, whereas only one-fifth show a familial risk in the other three types. Although only genetic testing (e.g., twin study designs) can detangle how much variation in a trait is related to genes or environmental factors and, family history of dyslexia is not a pure genetic trait (Pennington & Olson, 2008; Plomin et al., 1994; Rutter & Silberg, 2002), being at familial risk is one of the strongest predictive factors of the disorder (Thompson et al., 2015). It is possible that parents with higher SES (based on parental education or higher neighborhood scores) are more aware of possible reading difficulties in their children, over-reporting family history of dyslexia on our parental reports. Results however showed that parents with average to above average education level
showed similar proportions of at-risk indications (30%) to parents lower educational level (36%), \( \chi^2 = 1.17, p = 0.28 \). Families with an average to high social neighborhood status score also showed comparable proportions of at-risk indications (33%) compared to families with a lower score (30%), \( \chi^2 = 0.51, p = 0.47 \). Similarly, parent reports on family history for dyslexia may also be higher in parents from children already referred to a specialized dyslexia institute (sample 2) compared to parents from school children with poor reading skills (sample 1). Results nonetheless showed no difference in at-risk indications between the samples with 30% at-risk in sample 1 and 39% at-risk in sample 2, \( \chi^2 = 1.88, p = 0.17 \). Taken together, these results seem to support the validity of parental report of family risk of reading failure.

The PA-RAN subtype with increased familial risk for dyslexia also shows significantly higher IQ than the other subtypes influenced more by environmental risk factors. Specifically, the IQ scores of type 4 are approximately 1 SD higher than for the PA-LS type 3 poor readers and up to 1.4 SD higher than for the general type 2 poor readers. Finding two types of etiologies, one predominantly influenced by familial risk and characterized by high IQs discrepant to their poor reading profile (i.e., type 4) and one primarily environmentally influenced with lower to poor intellectual capabilities (type 2 and 3), is well in line with previous recent genetic behavioral results (Castles et al., 1999; Friend, DeFries, & Olson, 2008; Knopik et al., 2002; Rack & Olson, 1993; Wadsworth et al., 2000, 2010) and neuroimaging results (Shaywitz & Shaywitz, 2005). Finding support for the existence of these two possible etiologies for childhood reading could still mean that the same familial risk factors are operative in both etiologies, but that the PA-RAN specific type with high IQs is a more homogeneous environmental subtype and this in turn results in this profile showing a higher impact of familial risk for dyslexia (also see Wadsworth et al., 2000, 2010 for similar results). Thus, these findings do not suggest that different familial risk factors are influencing different types of reading disability as a function of IQ, but the proportion of variance accounted for by a familial predisposition may vary since a differential environmental impact might be at play. It is not only known that there is a significantly higher heritability of reading deficits in children with higher IQs (Wadsworth et al., 2000), it is also shown that heritability of dyslexia increases linearly with high levels of parental education (Friend et al., 2008). Consistently, our PA-RAN children at increased familial risk for dyslexia, characterized by higher IQs, also showed a more favorable socio-economic background (based on parental education and income). Although it is still unknown which proximal environmental factors mediate such a gene–environment interaction (Peterson & Pennington, 2012), possible candidates include language and (pre)literacy environments provided to children by their parents at home. The PA-RAN children at increased risk for dyslexia who come from families with higher parental education might thus enjoy a more supportive home literacy environment. A recent genetic study even suggested that children with increased genetic predispositions evoke more cognitively stimulating experiences from their environments (Tucker-Drob & Harden, 2012). Interestingly, previous studies also reveal that, in addition to PA, difficulties with RAN (the deficit specific to type 4) are found to be more related to genetic risk (e.g., Gayan & Olson, 2001; Petrill, Deater-Deckard, Thompson, DeThorne, & Schatschneider, 2006; Samuelsson et al., 2005), while letter name and sound knowledge problems (the deficit specific to type 3) is mostly influenced by shared home/preschool environmental factors and only to a small degree by genetic influences (e.g., Byrne et al., 2006, 2013; Samuelsson et al., 2005). It is thus conceivable that a more enriching home environment support might have ameliorated presence of LS weakness in our PA-RAN subtype, but not in our PA-LS poor readers from less favorable SES backgrounds. Reading and reading-related deficits due to a substantially larger genetic influence may in turn require more intensive remediation efforts though (Wadsworth et al., 2010).

Taken together, the present clustering approach was able to confirm the existence of distinct and homogeneous subtypes of poor reading substantially characterized by either familial risk factors or environmental risk factors, purely based on cognitive vulnerability markers commonly used for the diagnostics of dyslexia. And although IQ has fallen out of favor for sole diagnosis of dyslexia based on discrepancy or cut-off criteria (Stanovich & Siegel, 1994; Stuebing et al., 2002), the way IQ forms distinct patterns with these cognitive vulnerability markers as well as family history of dyslexia, seems informative and supports the idea that IQ can provide more information on the cause of the reading deficit experienced by the child (Friend et al., 2008; Wadsworth et al., 2000, 2010) as well as have a valuable attribution for exclusion purposes in diagnostic processes (Coltheart & Jackson, 1998; Rack & Olson, 1993). It may be noted that these clustering results are supported by the use of a data-driven, bottom-up approach, making no a priori assumptions on either the number or theoretical nature of subtypes, beyond the included clustering variables. A possible limitation of the study might be that these included cognitive vulnerability markers are based on one cognitive task only. A recommended improvement would be to include more than on measure per marker to increase stability of each cognitive, clustering construct. Moreover, we do not exclude the existence of additional, emerging subtypes if more than the currently employed classification measures were included. Finally, given that the relative severity and impact of cognitive deficits are commonly found to vary as a function of reading expertise and age (Landerl & Wimmer, 2008; Vaessen & Blomert, 2010), we recommend utilizing periodic reevaluation to determine whether the obtained latent clustering of deficits continues to be appropriate.

The present classification study provides evidence for distinct and coherent cognitive deficit patterns of poor reading. Despite a shared deficit in phonological awareness, a variable expression of other underlying cognitive deficits was found across several reading-related vulnerability markers and general cognitive domains, which differentiated between a poor reader type with a predominantly familial predisposition and two other poor readers that where characterized most by poor environmental factors. While letter-speech sound difficulties where characteristic in one of the environmentally influenced profiles (the other being the general poor reader type), a rapid automated naming deficit was specific for the poor reader type at elevated familial risk for dyslexia. The present results indicate that it is useful to include information on familial and environmental risk when identifying subtypes of poor reading and subsequently define tailored intervention efforts.
Moreover, it might also be a promising way forward for genetic testing studies to not just look at the reading impaired group as a whole as is usually done, but look at the heterogeneous group of poor readers with finer clustering resolution, focusing on underlying cognitive subtypes. From a methodological point of view, we could demonstrate that using large sample size and the external validation of the different subtypes in combination with a multi-start re-sampling method ensured the optimal number and stability of clusters representing the present data.

Acknowledgements

We thank the schools, children, parents and assistants for their enthusiastic participation and contribution. We are grateful to the Regional Institute of Dyslexia (RID) for their assistance in participant recruitment. The authors also wish to thank Dr. Arie van der Lugt and the four reviewers for their useful comments on the manuscript. This research was partly supported by a grant (608/001/2005) of the Dutch National Board of Health Care Insurance to our co-author Prof. Dr. Leo Blomert, who passed away on November 25, 2012.

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