Phonological Skills, Visual Attention Span, and Visual Stress in Developmental Dyslexia

Amanda Saksida Ecole Normale Supérieure, EHESS, CNRS, PSL Research University, and AREA Science Park, Scuola Internazionale Superiore di Studi Avanzati

Caroline Bogliotti
Ecole Normale Supérieure, EHESS, CNRS, PSL Research
University and Université Paris Ouest Nanterre

Jean-François Démonet Inserm, CHU Purpan and CHUV, University of Lausanne

> Catherine Billard Hôpital Bicêtre, APHP, Paris, France

Marie-France Le Heuzey Hôpital Robert Debré, APHP, Paris, France

Florence George Hôpital Salvator, Marseille, France Stéphanie Iannuzzi Ecole Normale Supérieure, EHESS, CNRS, PSL Research University and Inserm, CHU Purpan

Yves Chaix Inserm, CHU Purpan and Université de Toulouse III - Paul Sabatier

Laure Bricout
Ecole Normale Supérieure, EHESS, CNRS, PSL Research
University and Hôpital Bicêtre, APHP,
Paris, France

Marie-Ange Nguyen-Morel CNRS, Université Pierre Mendès France and Hôpital couple enfant, Grenoble, France

Isabelle Soares-Boucaud Hospices Civils de Lyon, Bron, France, and Centre Hospitalier Le Vinatier, Bron, France

> Johannes C. Ziegler CNRS, Aix-Marseille Université

Franck Ramus
Ecole Normale Supérieure, EHESS, CNRS, PSL Research University

Amanda Saksida, Laboratoire de Sciences Cognitives et Psycholinguistique (ENS, EHESS, CNRS), Ecole Normale Supérieure, PSL Research University, AREA di Ricerca Scientifica e Tecnologica di Trieste - AREA Science Park, and Language, Cognition and Development Lab, Scuola Internazionale Superiore di Studi Avanzati; Stéphanie Iannuzzi, Laboratoire de Sciences Cognitives et Psycholinguistique (ENS, EHESS, CNRS), Ecole Normale Supérieure, PSL Research University, Hôpital des enfants, and Imagerie Cérébrale et Handicaps Neurologiques, Inserm UMR 825, CHU Purpan; Caroline Bogliotti, Laboratoire de Sciences Cognitives et Psycholinguistique (ENS, EHESS, CNRS), Ecole Normale Supérieure, PSL Research University, and Laboratoire MODYCO CNRS UMR 7114, Université Paris Ouest Nanterre; Yves Chaix, Hôpital des enfants, CHU Purpan and Université de Toulouse III - Paul Sabatier; Jean-François Démonet, Imagerie Cérébrale et Handicaps Neurologiques, Inserm UMR 825, CHU Purpan, and Leenaards Memory Center, Department of Clinical Neurosciences, CHUV, University of Lausanne; Laure Bricout, Laboratoire de Sciences Cognitives et Psycholinguistique (ENS, EHESS, CNRS), Ecole Normale Supérieure, PSL Research University, and Centre de Référence sur les Troubles des Apprentissages, Hôpital Bicêtre, APHP, Paris, France; Catherine Billard, Centre de Référence sur les Troubles des Apprentissages, Hôpital Bicêtre, APHP; Marie-Ange Nguyen-Morel, Laboratoire de Psychologie et NeuroCognition UMR 5105 CNRS, Université Pierre Mendès France, and Centre de référence pour les troubles du langage et des apprentissages, Hôpital couple enfant, Grenoble, France; Marie-France Le Heuzey, Service de psychiatrie de l'enfant et de

l'adolescent, Hôpital Robert Debré, APHP, Paris, France; Isabelle Soares-Boucaud, Centre de référence pour les Troubles des Apprentissages, Hôpital Femme-Mère-Enfant, Hospices Civils de Lyon, Bron, France, and Service Hospitalo-Universitaire, Pôle de pédopsychiatrie, Centre Hospitalier Le Vinatier, Bron, France; Florence George, Centre de Références des troubles d'Apprentissages, Institut du développement de l'enfant et de sa communication, Hôpital Salvator, Marseille, France; Johannes C. Ziegler, Laboratoire de Psychologie Cognitive UMR 7290, CNRS, Aix-Marseille Université; Franck Ramus, Laboratoire de Sciences Cognitives et Psycholinguistique (ENS, EHESS, CNRS), Ecole Normale Supérieure, PSL Research University.

This research was supported by the European Commission (grant LSHM-CT-2005-018696), Ville de Paris, Agence Nationale de la Recherche (ANR-06-NEURO-019-01, ANR-11-BSV4-014-01, ANR-10-LABX-0087 IEC, ANR-10-IDEX-0001-02 PSL*), European Social Fund (TALENTS FVG Programme FP1362540002), and European Research Council (FP7/2007-2013 269502 PASCAL). We warmly thank Sylviane Valdois for providing the visual attention span test, Arnold Wilkins for providing the pattern glare test, and both of them for their support, advice, and criticism on a previous version of this paper.

Correspondence concerning this article should be addressed to Franck Ramus, Laboratoire de sciences cognitives et psycholinguistique, Département d'Etudes Cognitives, Ecole Normale Supérieure, PSL Research University, 29 rue d'Ulm, 75005 Paris, France. E-mail: franck.ramus@ens.fr

In this study, we concurrently investigated 3 possible causes of dyslexia—a phonological deficit, visual stress, and a reduced visual attention span—in a large population of 164 dyslexic and 118 control French children, aged between 8 and 13 years old. We found that most dyslexic children showed a phonological deficit, either in terms of response accuracy (92.1% of the sample), speed (84.8%), or both (79.3%). Deficits in visual attention span, as measured by partial report ability, affected 28.1% of dyslexic participants, all of which also showed a phonological deficit. Visual stress, as measured by subjective reports of visual discomfort, affected 5.5% of dyslexic participants, not more than controls (8.5%). Although phonological variables explained a large amount of variance in literacy skills, visual variables did not explain any additional variance. Finally, children with comorbid phonological and visual deficits did not show more severe reading disability than children with a pure phonological deficit. These results (a) confirm the importance of phonological deficits in dyslexia; (b) suggest that visual attention span may play a role, but a minor one, at least in this population; (c) do not support any involvement of visual stress in dyslexia. Among the factors that may explain some differences with previously published studies, the present sample is characterized by very stringent inclusion criteria, in terms of the severity of reading disability and in terms of exclusion of comorbidities. This may exacerbate the role of phonological deficits to the detriment of other factors playing a role in reading acquisition.

Keywords: developmental dyslexia, phonological deficits, visual stress, visual attention span

Supplemental materials: http://dx.doi.org/10.1037/dev0000184.supp

Developmental dyslexia is a common learning disorder affecting about 3% to 7% of the population (Lindgren, De Renzi, & Richman, 1985; Peterson & Pennington, 2015). It is defined as a specific deficit in reading acquisition that cannot be accounted for by low IQ, poor educational opportunities, or an obvious sensory or neurological damage (World Health Organization, 2011). It is quite remarkable that such a seemingly simple and circumscribed disorder has engendered a truly unique profusion of theories.

Because reading relies primarily on language and on vision, it is not surprising that most theories of dyslexia have postulated a deficit in one domain or in the other. The first descriptions of developmental dyslexia viewed it as a "congenital word blindness" (Hinshelwood, 1900; Morgan, 1896; Stephenson, 1907), and indeed, visual symptoms and hypotheses have dominated the best part of the 20th century (Dunlop, 1972; Hallgren, 1950; Orton, 1937). It is only in the 1970s, with the development of research on speech perception, that apparent visual confusions were reinterpreted as phonological ones and that the theory of a phonological deficit emerged and gradually became predominant (Brady & Shankweiler, 1991; Fischer, Liberman, & Shankweiler, 1978; Liberman, 1973; Shankweiler & Liberman, 1972).

However, debates have gone far beyond this simple distinction. In the language domain, although the hypothesis of a phonological deficit is widely accepted, there have been and still are discussions on whether or not more primary auditory deficits underlie the phonological deficit (Goswami, 2015; Hornickel & Kraus, 2013; Tallal, Miller, & Fitch, 1993). Furthermore, there are also debates on the specific nature of the phonological deficit (Boets et al., 2013; Ramus, 2014; Ramus & Ahissar, 2012; Ramus & Szenkovits, 2008). In the visual or visual-attentional domain, there is also more than one theory available (Vidyasagar & Pammer, 2010), including the magnocellular theory, according to which deficits in the magnocellular visual pathway may lead to poor binocular control and visual instability (Stein & Walsh, 1997), the sluggish attentional shifting theory, according to which reading deficits stem from spatial attention disorder (Gori & Facoetti, 2015; Hari & Renvall, 2001), visual stress (Irlen, 1991; Meares, 1980; Wilkins et al., 1984), and the visual attention span theory (Bosse,

Tainturier, & Valdois, 2007)—the latter two being discussed in further detail in the following paragraphs. Finally, a few theories appeal to deficits outside both the language and visual/attentional domains, such as an anchoring deficit, an automaticity deficit, or a deficit in perceptual noise exclusion (Ahissar, 2007; Nicolson & Fawcett, 2007; Sperling, Lu, Manis, & Seidenberg, 2005).

Overall, the accumulated data are broadly consistent with the existence of a majority subtype characterized by a phonological deficit, and one or several minority subtypes characterized by a visual or visual-attentional deficit. Additional subtypes of phonological and visual dyslexia might emerge from the consideration of underlying etiologies. Some earlier studies have attempted to adjudicate between different theories of dyslexia by testing them systematically against each other within the same individuals (Amitay, Ben-Yehudah, Banai, & Ahissar, 2002; Ramus, Pidgeon, & Frith, 2003; Ramus, Rosen, et al., 2003; White, Frith, et al., 2006; White, Milne, et al., 2006). Results were in favor of a widespread phonological deficit in dyslexia and against alternative theories. This conclusion was nevertheless limited by the small populations studied, and by the fact that it was obviously not possible to test all conceivable theories of dyslexia. In particular, the only visual theory of dyslexia that has been broadly tested by many independent teams is the magnocellular theory. Although, overall, many studies failed to support that hypothesis (Ramus, 2003; Skottun, 2000), it remains possible that other visual theories of dyslexia might be more successful in explaining the minority of dyslexic children who do not show a phonological deficit. It is the purpose of the present study to address two other visual theories of dyslexia: that of visual stress and that of a reduced visual attention span.

Visual stress (also known as Meares-Irlen syndrome; Irlen, 1991; Meares, 1980) is the inability to see without perceptual distortion and discomfort (Wilkins, Huang, & Cao, 2004). It has been linked to cortical excitability and migraine (Wilkins, 1995; Wilkins et al., 1984). Visual stress is particularly triggered by flickering stimuli and by certain geometric patterns, notably stripes at certain spatial frequencies (around 3 cycles/deg; Wilkins et al., 1984). It has been suggested that because text is a striped pattern,

it may provoke visual stress in certain individuals, thereby impairing reading performance (Wilkins & Nimmo-Smith, 1987). Indeed, there is some evidence that people with self-reported symptoms of visual stress show lower reading speed (Hollis & Allen, 2006; Wilkins, Lewis, Smith, Rowland, & Tweedie, 2001). There is also evidence that the use of colored overlays, which are supposed to reduce visual stress, may help increase reading speed (Bouldoukian, Wilkins, & Evans, 2002; Hollis & Allen, 2006; Wilkins et al., 2001), although some studies find no effect on reading even after long-term use of colored overlays (Ritchie, Della Sala, & McIntosh, 2011, 2012). Finally, there are frequent suggestions of an association between visual stress and developmental dyslexia. Three studies examined visual stress in groups of dyslexic children and reported a prevalence ranging from 35% to 47% (18% to 25% in the control group; Kriss & Evans, 2005; Singleton & Henderson, 2007; White, Frith, et al., 2006). However, these studies used diverse and debatable definitions of visual stress. Indeed, their main criterion was to what extent children improved their reading fluency by using their preferred colored overlay compared with reading the same text without an overlay. Yet benefit from using an overlay does not prove that the child had symptoms of visual stress to begin with, nor that visual stress was the cause of reading disability, any more than an improvement of attention skills following absorption of methylphenidate can provide a diagnosis of attention-deficit hyperactivity disorder (ADHD). In our view, only direct, deleterious symptoms should be taken as evidence for visual stress. One of these studies also collected symptoms of visual stress through a self-report questionnaire, and reported that dyslexic children reported slightly more symptoms of visual stress than controls, although they also responded positively more often to the control questions, suggesting a general response bias (Kriss & Evans, 2005). Across the three studies, the largest dyslexic group included 27 dyslexic children. Furthermore, visual stress has not been studied concurrently with other cognitive measures of dyslexia, such as the phonological deficit or other putative visual deficits. Thus, there remains a great need to assess symptoms of visual stress together with other cognitive deficits in a much larger population of dyslexic children.

The visual attention span is defined as the amount of distinct visual elements that can be processed in parallel in a multielement array (Bosse et al., 2007). Computational modeling suggests that a reduced visual attention span should impair reading performance and reading acquisition (Ans, Carbonnel, & Valdois, 1998). Indeed, some studies have reported that a substantial subset of dyslexic children show a reduced visual attention span. Furthermore, these studies suggest that most dyslexic children have either a visual attention span deficit or a phonological deficit, with few mixed cases (Bosse et al., 2007; Zoubrinetzky, Bielle, & Valdois, 2014; hereafter "Valdois and collaborators"). In these studies, visual attention span has typically been assessed by briefly flashing an array of five letters, and asking participants to verbally report either all five letters (global report) or a single, postcued letter (partial report). The usage of linguistic stimuli (letters) has led to criticism that poor performance in the report tasks might reflect verbal as much as visual attentional deficits (Hawelka & Wimmer, 2008; Ziegler, Pech-Georgel, Dufau, & Grainger, 2010). The debate is still ongoing, and some results suggest that dyslexic children show a visual attention span even when measured with entirely nonlinguistic stimuli (Goswami, 2015; Lobier & Valdois,

2015; Lobier, Zoubrinetzky, & Valdois, 2012). However, the letter report remains the standard way of testing the visual attention span (Peyrin et al., 2012; Zoubrinetzky et al., 2014). The theory of a visual attention span deficit in dyslexia is thus a potentially promising candidate to explain those cases of dyslexia that do not show a phonological deficit, and it is therefore important to further test it against both the phonological theory and alternative visual theories.

In summary, in the present study, we will assess concurrently, in a large population of French dyslexic and control children, three types of deficits that are potential explanations of dyslexia: the phonological deficit, a reduced visual attention span, and visual stress. We aim to estimate the relative prevalence of each type of deficit within the French dyslexic population, their potential overlap, and their explanatory value with respect to diagnostic category and to literacy skills.

Method

Participants

As part of a larger study on the genetic bases of dyslexia (Becker et al., 2014), we tested 282 French children, aged between 8 and 13 years and attending school Grades 3 to 7. One hundred sixty-four of them (109 boys and 55 girls) were children with dyslexia who were recruited in one of seven language and learning disability units in France (Toulouse, Kremlin-Bicêtre, Grenoble, Paris, Lyon, and Marseille). As a control group, 118 children (49 boys and 69 girls) with no reading problems were recruited either from schools in the same French cities as the dyslexic group or by announcements. Based on parental questionnaire, we excluded the children who had

- suffered a hearing loss,
- · uncorrected sight problems,
- · neurological injury,
- no parent speaking French from birth on,
- not been schooled in French,
- missed school for any period of 3 months or more,
- received a formal diagnosis of ADHD or specific language impairment (SLI), or
- received medication for epilepsy or behavioral problems.

General inclusion criteria for both dyslexic and control groups of participants were as follows:

- An attention scale score within the 95th percentile of the age-appropriate norm from the attention scale of the Child Behavior Check-List (Achenbach, 1991), filled by parents.
- A standard score higher than 7 on blocks or matrices subtests of the Wechsler Intelligence Scale for Children (WISC IV) battery (Wechsler, 2005).
- No score below -2 standard deviations on any of three oral language tests (word repetition, word retrieval, syntactic production). This was intended to exclude children with comorbid dyslexia and SLI, who were also part of the population of interest.

Children with dyslexia also met *both* of the following inclusion criteria:

 At least 18 months delay in text reading fluency as tested by the "Alouette," a standardized test for French reading (Lefavrais, 1967).

 At least 1.25 standard deviation below the gradeappropriate mean in isolated word reading fluency (including regular and irregular words; Jacquier-Roux, Valdois, & Zorman, 2002).

The control group met both of the following inclusion criteria:

- No more than 12 months of delay in text reading.
- No more than 0.85 standard deviation below the mean in isolated word reading.

Overall, this stringent set of inclusion and exclusion criteria was chosen to select dyslexic children with particularly severe and specific reading difficulties, while minimizing comorbidity with other disorders (in particular, SLI and ADHD).

Ethics approval for the study was granted by the Comité de Protection des Personnes of Hôpital Bicêtre (Le Kremlin-Bicêtre). Written informed consent was obtained from both parents of each child, and oral or written assent was obtained from each participant.

Protocol

Tests were administered individually in a quiet room at the hospital by psychologists or speech therapists. Children were tested in one or two sessions. When dyslexic participants had been administered the WISC IV battery and the reading tasks at the hospital less than one year before the study, those scores were registered without retesting.

Psychometric measures. Three of the 10 subtests of the WISC IV battery were administered to all participants: blocks, similarities, and digit span. For the majority of participants, matrix reasoning and comprehension were tested as well, and some participants were given the full WISC. The available scores were used to compute or estimate Verbal Comprehension Index and Perceptual Orientation Index (POI).

Attention screening. The 11 items of the attention scale of the Child Behavior Checklist (Achenbach, 1991) were filled by parents, who answered whether different patterns of behavior were present always (2 points), sometimes (1 point), or never (0 points) in their child. Based on current norms, children were considered as having inattention problems and were excluded if the total score was higher than 8 for girls and 9 for boys.

Reading tasks.

Text reading fluency. The "Alouette" reading test (Lefavrais, 1967) is a 265-word text without much meaning. All the words are real lexical entries, but some of them are extremely rare and not expected to be known by children. Participants had to read the text as quickly and accurately as possible in no more than 3 min. Scoring combined reading time and number of errors. This task provided an estimate of reading age.

Word/nonword reading accuracy/speed. ODEDYS (Jacquier-Roux et al., 2002) comprises three lists of 20 words each (regular, irregular, and nonwords). Children read each list as quickly and accurately as possible. For both word and nonword reading, combined accuracy/speed z scores were computed. The combined performance in regular and irregular word lists, but not in nonwords, was used for the inclusion criteria. For some analyses, however, separate accuracy and time scores are used.

Orthographic skills.

Word spelling (Martinet & Valdois, 1999). Sixty-six words were dictated to the children. For each trial, the target word was

pronounced first in isolation, then within a carrier sentence, and finally in isolation again (example: "Soucoupe. On a vu une soucoupe volante. Soucoupe" ["Saucer. Someone saw a flying saucer. Saucer"]). Twenty-two words had a regular and consistent spelling (example: soucoupe [saucer]), 22 had a spelling that was regular but inconsistent in the sense that at least one phoneme could plausibly be spelled in several ways (example: dentiste [dentist]), and 22 were irregular words (example: baptême [baptism]). Words were presented in a pseudorandom order in three lists of 22 words of equal difficulty, with categories interleaved. Total percentage of correct answers was calculated, irrespective of word category. When a child spelled fewer than three words correctly out of the first 22, the test was interrupted and the child's score multiplied by 3 (this stopping criterion was adopted because a 66-word dictation was judged too painful for very poor spellers, and going beyond the first 22 words provided little additional information in that case).

Orthographic choice (Sprenger-Charolles, Béchennec, Colé, & Kipffer-Piquard, 2005). Participants were shown triplets of words on a computer screen—only one correctly spelled and the other two differing by only one letter (example: tulipe [tulip], which is correct; tulippe, which has the same pronunciation but is visually different; tuliqe, which is visually similar but different in pronunciation). They were asked to decide as fast as possible which of the words was orthographically correct by pressing one of three keys. Response accuracy and speed were recorded.

Phonological assessment.

Phoneme deletion (Sprenger-Charolles et al., 2005). The two tests included 24 monosyllabic pseudowords, among which 12 had a consonant-vowel-consonant structure (e.g., "kip") and 12 had a consonant-consonant-vowel structure (e.g., "pra"). The time taken to repeat all items in the list without the initial phoneme was measured. The results of the two lists were summed to give a measure of accuracy and speed.

Spoonerisms. Participants were auditorily presented with pairs of words and were instructed to swap the first sound of each word and then pronounce the resulting nonwords. (e.g., "banana" and "candle" become "canana" and "bandle"). Ten items were presented and the total time was recorded, with a maximum of 30 s for each item.

Rapid automatized naming. Four series of 49 items (letters, digits, pictures, and colors) were presented on a computer screen. Participants were instructed to name them as rapidly as possible. Total naming time was the dependent measure.

Visual attention span.

Global letter report. In each trial, the participants were required to orally report the five letters of an unpronounceable string (e.g., R H S D M) presented for 200 ms in the center of a monitor screen. Font size and screen distance were adjusted such that the five letters spanned a visual angle of approximately 5.4°. The recorded score corresponded to the number of letters accurately reported across the 20 trials (maximum = 100).

Partial letter report. The participants were required to orally report a single cued letter among the five letters of each briefly presented string (e.g., T H F R D). At the offset of the letter string presentation, a vertical bar cue appears for 50 ms below one letter. Participants were asked to report the cued letter only.

The score was the number of letters accurately reported (maximum = 50).

In these tasks, participants were instructed to be as accurate as possible and no time pressure was applied (see Bosse et al., 2007).

Isolated letter identification. Because of the possibility that global and partial letter report scores are limited by single letter identification skills, our participants were also tested with the control task proposed by Bosse et al. (2007). Isolated letter identification consists of 10 consonants randomly presented (5 times each) in the center of the screen at different presentation durations (33, 50, 67, 84, and 101 ms). Each trial began with a central fixation point presented for 1,000 ms, followed by one letter. At the offset of the letter, a mask was displayed for 150 ms and participants were asked to name each letter immediately after its presentation. The score was the sum of letters accurately identified at each presentation time. To control for the impact of letter identification on global and partial letter report tasks, analyses were based on the residuals of these measures after regressing out letter identification scores.

Visual stress. Children were given a pattern glare test inspired by visual stress theory (Evans & Stevenson, 2008; Wilkins, 1995). Two patterns of striped lines were presented twice each in counterbalanced order. One of the gratings had a high spatial frequency (3 cycles/deg) and is meant to provoke visual stress in sensitive individuals. The other one has a lower spatial frequency (0.5 cycles/deg) and is used as a control for

response biases. Children had to fixate a cross at the center of each grating during 5 s and then answer five questions about whether lines are distorted, fuzzy, in color, appear to move, and whether other shapes can be seen. Each answer was scored 0 (not at all), 1 (yes, a little), or 2 (yes, a lot), and scores were summed across the five questions and the two repetitions of each condition to produce a score out of 20. Analyses were done on the residuals of the high spatial frequency scores after regressing out the lower spatial frequency scores.

Results

Descriptive Results

Group comparisons. Table 1 summarizes the results of the tasks performed by the two groups of participants. For group differences, effects sizes are indicated (Cohen's *d*). The two groups differed significantly in all the literacy, phonological, and visual tasks. In all these variables, dyslexic children performed more poorly than controls, with the exception of the pattern glare test, in which controls reported more symptoms of visual stress than the dyslexic participants, in both conditions. The two groups also differed slightly in age and in the POI. Therefore, when appropriate, the contribution of these variables was partialed out in the relevant analyses. For the subsequent analyses, the results from all literacy, phonological, and visual tasks were transformed into

Table 1
Results of Psychometric Tests, Reading, Phonological, and Visual Tasks

		Dyslexics			Controls		Group diff	erences	Effect size
Variables	n	Mean	SD	n	Mean	SD	F(1, 281)	p	Cohen's d
Age (months)	164	127.75	15.03	118	124.66	14.18	3.04	.08	.21
Grade	164	4.73	1.23	118	4.81	1.28	.34	.56	.06
Blocks (scaled score)	164	10.79	2.39	117	11.26	2.43	2.53	.11	.19
Similarities (scaled score)	160	11.05	2.90	117	13.53	2.67	52.80	<.001	.88
Digit span (scaled score)	161	7.14	2.55	116	10.17	2.77	88.44	<.001	1.15
Verbal Comprehension Index	160	104.26	14.62	117	118.14	16.78	53.71	<.001	.89
Perceptual Orientation Index	164	103.23	12.69	117	106.78	14.25	4.83	<.001	.27
Word reading accuracy (/20)	163	10.63	3.92	118	18.31	1.66	400.87	<.001	2.42
Word reading speed (s)	162	71.33	45.46	118	19.06	6.85	153.35	<.001	1.50
Reading lag (months)	164	-35.45	11.23	116	7.91	15.50	737.05	<.001	3.29
Text reading fluency (correct words/min)	164	38.21	20.55	116	116.11	28.67	701.55	<.001	3.21
Word spelling accuracy (%)	160	39.08	16.78	117	78.45	16.67	374.04	<.001	2.35
Nonword reading accuracy (/20)	160	10.91	4.02	118	17.19	2.05	241.51	<.001	1.89
Nonword reading time (s)	159	65.64	41.58	118	26.04	7.41	104.44	<.001	1.24
Orthographic choice accuracy (/10)	152	7.43	2.45	107	9.54	1.01	71.31	<.001	1.07
Orthographic choice speed (s)	152	53.30	35.40	107	22.53	8.55	77.59	<.001	1.11
RAN letters (s)	161	34.44	9.92	117	23.45	4.70	123.60	<.001	1.35
RAN digits (s)	162	34.27	8.77	117	24.02	5.37	125.80	<.001	1.36
RAN objects (s)	162	52.17	11.95	117	37.98	7.78	126.46	<.001	1.36
RAN colors (s)	161	51.75	15.48	116	36.56	8.78	90.65	<.001	1.16
Phoneme deletion accuracy (/24)	156	17.48	4.79	117	22.56	1.94	117.23	<.001	1.32
Phoneme deletion speed (s)	157	105.91	40.01	117	66.05	17.51	101.55	<.001	1.23
Spoonerisms accuracy (/10)	152	3.30	2.81	112	8.17	1.85	254.28	<.001	1.99
Spoonerisms speed (s)	151	248.28	56.48	111	126.23	45.42	351.27	<.001	2.34
Letter identification (/50)	159	41.13	7.31	115	45.76	4.22	37.17	<.001	.75
Global report (letters reported /100)	157	65.68	14.84	117	86.56	10.23	171.02	<.001	1.60
Partial report (letters correct /50)	158	37.83	6.65	114	42.91	5.31	45.55	<.001	.83
High frequency visual stress (/20)	149	2.05	2.49	96	3.13	2.70	10.22	.02	.42
Low frequency visual stress (/20)	149	1.19	1.79	96	2.30	2.15	19.09	<.001	.57

Note. Statistics and effect sizes are shown for group differences. RAN = rapid automatized naming.

normalized z scores based on the means and the standard deviations of the control group, separately for each grade.

Factor analysis of literacy variables. In order to assess the sources of variability across literacy tasks, we performed a principal component analysis of the following variables: word reading (ODEDYS, accuracy and speed), estimated reading lag (Alouette), word spelling (accuracy), nonword reading (accuracy and speed), and orthographic choice (accuracy and speed). Only one factor was extracted, accounting for 67.19% of the variance (see Table 2). This indicates that all literacy skills are highly intercorrelated in this population.

Nevertheless, for the purpose of examining whether some results might differ according to literacy subskills, we computed three composite variables: *Reading Accuracy*, as the average *z* score of word and nonword reading accuracy; *Reading Speed*, as the average *z* score of word and nonword reading speed as well as reading lag (based on text reading fluency), and *Orthography* as the average *z* score of word spelling, orthographic choice accuracy and speed.

Factor analysis of phonological and visual variables. Similarly, we performed an exploratory principal component analysis of all potentially explanatory variables, whether phonological or visual: rapid naming (speed); phoneme deletion (accuracy and speed); spoonerisms (accuracy and speed); digit span (accuracy); global, partial, and letter report (accuracy); and high- and low-frequency visual stress tests (subjective ratings). An oblique rotation (Oblimin) was chosen to allow factors to correlate with each other, which seems a more plausible assumption than having them orthogonal (Fabrigar, Wegener, MacCallum, & Strahan, 1999). Nevertheless, a principal component analysis with a Varimax rotation yielding orthogonal factors gave essentially the same picture (see Table 1 of the online supplemental materials).

Three components were extracted, accounting for a total of 62.64% of the variance (first component accounting for the 39.93% of variance, the second for the 12.86%, and the third for the 9.75 of variance; see Table 3). The first factor captured variance in all phonological tasks but rapid naming. It loaded more highly on accuracy than on speed scores. Furthermore, it captured variance in the three visual attention span tasks as well. Although it is surprising that such different sets of tasks might be lumped into a single factor, this is explained by the fact that, in this data set, performance in phonological and in visual-attentional tasks are substantially correlated (see online supplementary Table 2).

The second factor loaded mostly on the two visual stress tasks. Again, even though the high-rather than the low-frequency condi-

Table 2
Results of the Principal Component Analysis for the
Literacy Tasks

Variables	Loading
Word reading accuracy (ODEDYS)	.91
Word reading speed (ODEDYS)	.78
Reading lag (months) (Alouette)	.86
Word spelling accuracy	.85
Nonword reading accuracy (ODEDYS)	.83
Nonword reading speed (ODEDYS)	.78
Orthographic choice accuracy	.55
Orthographic choice speed	.68

tion is supposed to reflect visual stress, it turns out that the two variables were highly correlated and were therefore grouped into a single factor. Thus, most of the variance in these scores may actually reflect a general disposition to give lower or higher ratings regardless of condition.

The third factor loaded mostly on rapid naming tasks, as well as on the speed measures of spoonerisms and phoneme deletion. It therefore unambiguously reflects the speed component of phonological tasks.

Based on this exploratory factor analysis (EFA), one approach would be to simply continue the analyses using the factors as just defined. One drawback of that approach is that this may fail to distinguish cognitive dimensions that are theoretically important to distinguish. For instance, one of our goals is to compare the respective contributions of phonological skills and visual attention span, despite their being sufficiently correlated to appear in the same factor in the EFA. Another drawback is that the EFA may group together variables that we want to distinguish, because one is the control condition of another (e.g., letter detection and global report; low vs. high frequency visual stress). Finally, factors defined through an EFA carry many minor loadings that are not theoretically interpretable, that may not be stable, and that may therefore simply add noise. For all these reasons, we find it preferable to define theory-driven components that are simply informed by the EFA, like in our previous work (Ramus, Marshall, Rosen, & van der Lely, 2013; Ramus, Rosen, et al., 2003; White, Frith, et al., 2006). Because the EFA suggested that spoonerisms and phoneme deletion reaction time reflect partly accuracy and partly speed, we allowed these two variables to cross-load equally on phonological accuracy and speed. Thus, we computed a PHO-NOLOGICAL ACCURACY component, averaging the accuracy scores of spoonerisms, phoneme deletion and digit span, as well the response times of spoonerisms and phoneme deletion with a 0.5 loading (see Table 3). Similarly, we computed a PHONOLOG-ICAL SPEED component by averaging the three rapid naming scores, as well as the response time scores from phoneme deletion and spoonerisms with a 0.5 loading.

Despite the results of the EFA, we formed a separate component for visual-attentional tasks in order to assess them independently. We considered two versions of this factor. In their articles, Valdois and collaborators typically averaged global and partial report scores (Bosse et al., 2007; Zoubrinetzky et al., 2014). However, it may be argued that the global report measure, because it requires remembering five letters, loads on verbal short-term memory as well as on visual attention span. The partial report measure does not have this problem. Indeed, in the present data set, digit span's correlation with global report is 0.23 (p = .01), but is only 0.09 (p = .29) with partial report. Furthermore, performance in both measures is also potentially limited by children's ability to recognize briefly flashed letters and retrieve their names, which may conceivably be less automatized in dyslexic children. This was the point of including the single-letter identification task as a control, which indeed correlates with global report (R = 0.42; p < .01) and with partial report (R = 0.30; p < .01). Thus, in order to obtain a more conservative measure of VISUAL ATTENTION SPAN (VAS), we computed this component using partial report scores only, and by regressing out letter identification scores. Furthermore, for the purpose of comparing our results more closely with

Table 3
Principal Component Analysis of Phonological and Visual Tasks

	_	Factors of ploratory f analysis	actor		(Valdois version)			
Variables	1	2	3	Phonological accuracy	Phonological speed	Visual attention span	Visual stress	Visual attention span
Digit span	.70	06	35	1	0	0	0	0
Rapid naming – digits	.48	.10	91	0	1	0	0	0
Rapid naming – objects	.46	.21	89	0	1	0	0	0
Rapid naming – colors	.37	.05	88	0	1	0	0	0
Phoneme deletion accuracy	.54	.20	38	1	0	0	0	0
Phoneme deletion speed	.53	.06	61	.5	.5	0	0	0
Spoonerisms accuracy	.79	.29	46	1	0	0	0	0
Spoonerisms speed	.64	.21	69	.5	.5	0	0	0
Letter identification	.54	09	26	0	0	Regressed out	0	0
Global report	.84	.25	56	0	0	0	0	1
Partial report	.71	.08	29	0	0	1	0	1
High frequency visual stress	.05	.90	05	0	0	0	1	0
Low frequency visual stress	.09	.89	16	0	0	0	Regressed out	0

Note. Oblimin rotation and Kaiser normalization were used. The highest loading of each variable is shown in bold. Columns 4–7 give the loadings of the four theory-driven component scores. Column 8 gives the loadings of an alternative way to compute the visual attention span component score following Valdois and collaborators (Bosse et al., 2007; Zoubrinetzky et al., 2014).

those of Valdois et al., we also computed the average z score of global and partial report scores (VAS1).

Finally, in order to compute a VISUAL STRESS component, we used the high-frequency ratings, with the low-frequency ratings (control condition) regressed out. This ensured that this component reflected actual visual stress, unaffected by reporting bias. Descriptive statistics of these four components are given in Table 4.

Similar to the results with the raw scores, the differences between dyslexic and control groups are significant for the literacy tasks, phonological awareness tasks, and the visual attention span task, whereas the difference in the high frequency visual stress task has disappeared after controlling for the low-frequency condition.

In order to assess to what extent each of these four components contributes to predicting dyslexia and literacy skills, we then used them (a) in linear regressions to predict literacy skills, (b) in logistic regressions to predict group membership, and (c) in deviance analyses to compute the prevalence of deficits in each component and their overlap.

Predictors of Literacy Skills

As a first step, we observed correlations between literacy skills and phonological and visual skills, as represented by our composite variables. The results are presented in Table 5. Most notably, both VAS and VAS1 correlate highly with phonological and reading speed variables.

We also performed hierarchical linear regression analyses of reading accuracy, reading speed, and orthography (successively) as dependent variables, with phonological accuracy, phonological speed, visual attention span (VAS), visual stress, age, and POI indices as independent variables.

In order to first analyze the contribution of visual variables without phonological variables, age and POI were entered into the model in the first step, visual variables in the second step, and phonological variables in the third. Beyond age and POI, the visual attention span contributed an extra 4% of variance to reading accuracy, and 7% to reading speed. On the other hand, visual stress did not contribute any

Table 4
Group Differences on the Component Scores

		Dyslexics	S		Controls		Group 6	Effect size	
Component variables	n	Mean	SD	n	Mean	SD	F(1, 229)	p	Cohen's d
Literacy	164	-9.33	4.26	118	.00	1.00	542.82	<.001	2.81
Phonological speed	162	-3.71	2.36	118	.00	1.00	258.57	<.001	1.95
Phonological accuracy	162	-4.88	2.87	117	.00	1.00	310.67	<.001	2.14
VAS	158	82	1.38	114	.00	1.00	28.90	<.001	.66
VAS1	157	-1.55	1.55	114	.00	1.00	87.47	<.001	1.15
Visual stress	149	07	.95	96	.00	1.00	.31	.58	.07

Note. VAS = Visual attention span (partial report with letter report partialled out). VAS1 = Visual attention span calculated as in Valdois and collaborators (average of global and partial report).

Table 5
Partial Linear Correlations Between the Composite Variables After the Factor Analysis

Component variables	Reading speed	Orthography	Phonological speed	Phonological accuracy	VAS1	VAS	Visual stress
Reading accuracy	.523**	.655**	.654**	.650**	.305**	.136*	.022
Reading speed	1	.356**	.600**	.514**	.406**	.206**	.062
Orthography		1	.504**	.470**	.186**	.054	.071
Phonological speed			1	.590**	.309**	.176**	045
Phonological accuracy				1	.280**	.163*	.016
VAS1					1	.886**	.084
VAS						1	.034

Note. Correlations are reported after partialling out age, nonverbal IQ and digit span. VAS1 = visual attention span computed by averaging global and partial report following previous analyses; VAS = visual attention span computed by regressing letter report from partial report.

additional variance to any literacy component (Table 6, Model 2). Once phonological variables were added in the third step, VAS and visual stress did not play a significant role for any of the literacy components anymore, whereas phonological accuracy and speed played a significant role for all literacy components (Table 6A, Model

3). Repeating these analyses using Valdois's version of the visual attention span (VAS1) altered these results to the extent that under this analysis, VAS1 remained an important contributor of variance even when phonological skills were added to the model, but only for reading speed (see online supplementary Table 3A).

Table 6
Results of Hierarchical Linear Regression Analyses

		Reading accuracy					Reading s	speed		Orthography			
Model	Dependent variables	R^2	St. coeff. β	t	Sig.	R^2	St. coeff. β	t	Sig.	R^2	St. coeff. β	t	Sig.
				(A) Hi	erarchic	al line	ar regression an	alysis					
1.00	(Constant)	.09		-1.92	.06	.03		40	.69	.08		-3.06	.00
	Age		17	-2.76	.01		14	-2.14	.03		08	-1.28	.20
	POI		.24	3.91	.00		.09	1.44	.15		.27	4.32	.00
2.00	(Constant)	.13		-1.63	.11	.10		.05	.96	.09		-2.84	.01
	Age		16	-2.69	.01		13	-2.08	.04		08	-1.24	.22
	POI		.21	3.48	.00		.05	.84	.40		.25	3.99	.00
	VAS		.21	3.38	.00		.27	4.31	.00		.11	1.74	.08
	Visual stress		.01	.09	.93		.04	.65	.52		.06	.92	.36
3.00	(Constant)	.66		-1.12	.26	.51		.83	.41	.42		-2.73	.01
	Age		07	-1.79	.08		04	80	.43		.00	.01	.99
	POI		.11	2.81	.01		03	63	.53		.17	3.37	.00
	VAS		.00	.06	.96		.09	1.83	.07		05	-1.00	.32
	Visual stress		.03	.75	.45		.07	1.47	.14		.08	1.56	.12
	Phonological speed		.41	7.79	.00		.47	7.45	.00		.37	5.42	.00
	Phonological accuracy		.44	8.41	.00		.27	4.28	.00		.30	4.32	.00
			(B) Hierard	chical line	ar regre	ssion a	nalysis without	phonologi	cal spe	ed			
1.00	(Constant)	.16		-1.92	.06	.29		40	.69	.28		-3.06	.00
	Age		17	-2.76	.01		14	-2.14	.03		08	-1.28	.20
	POI		.24	3.91	.00		.09	1.44	.15		.27	4.32	.00
2.00	(Constant)	.32		-1.63	.11	.36		.05	.96	.31		-2.84	.01
	Age		16	-2.69	.01		13	-2.08	.04		08	-1.24	.22
	POI		.21	3.48	.00		.05	.84	.40		.25	3.99	.00
	VAS		.21	3.38	.00		.27	4.31	.00		.11	1.74	.08
	Visual stress		.01	.09	.93		.04	.65	.52		.06	.92	.36
3.00	(Constant)	.63		52	.61	.76		1.22	.22	.59		-2.23	.03
	Age		10	-2.40	.02		08	-1.49	.14		03	57	.57
	POI		.10	2.34	.02		04	73	.47		.17	3.07	.00
	VAS		.04	.86	.39		.13	2.45	.02		02	36	.72
	Visual stress		.01	.19	.85		.04	.86	.39		.06	1.12	.27
	Phonological accuracy		.71	15.76	.00		.57	10.72	.00		.54	9.69	.00

Note. (A) The variables were entered stepwise, with age and POI entered in the first step, VAS and visual stress in the second step, and phonological speed and accuracy in the third step. (B) The same analysis but with only phonological accuracy entered in the model. POI = perceptual orientation index. VAS = visual attention span.

^{*} Correlation is significant at p < .05 level. ** Correlation is significant at p < .01 level.

Because in Valdois et al.'s studies, phonological skills are represented solely by phonological accuracy, and because in the present data set, VAS seems particularly correlated with both reading and phonological speed (see Table 5), we ran a final analysis by entering only phonological accuracy in the model, leaving phonological speed out. In this final analysis, VAS becomes a significant explanatory factor for reading speed (Table 6B), whereas VAS1 explains variance in both reading sped and accuracy (see online supplementary Table 3B).

Predictors of Dyslexia

To examine to what extent the results from phonological accuracy, phonological speed, VAS, and visual stress tasks successfully predicted group membership, we performed a stepwise logistic regression analysis of these four variables. The analysis showed that the classification of participants into one of the two groups (controls and dyslexics) was highly accurate (95.9%) by including only phonological variables in Steps 1 and 2 (see Table 7). Visual attention span and visual stress were added in Steps 3 and 4 and contributed marginally, increasing the classification accuracy to 96.7% and 96.3%, respectively.

Repeating the same analysis using the VAS1 component, the classification of the groups was similarly successful, with the order of variable inclusion and model accuracy being almost identical to the previous analysis (see online supplementary Table 4).

Prevalence of Deficits

Finally, we identified each individual's deficit(s) by applying a -1.5 standard deviation cutoff criterion to each of the four explanatory components: PHONOLOGICAL ACCURACY, PHONOLOGICAL SPEED, VAS, and VISUAL STRESS. Figure 1 displays the number of dyslexic and control participants with each deficit and/or combination of deficits.

The most prevalent deficits were in phonological accuracy (92.1% of dyslexic vs. 8.5% of control participants) and in phonological speed (84.8% of dyslexic vs. 6.8% of control participants). Overall 97.6% of dyslexic participants had at least one or the other phonological deficit. Deficits in visual attention span affected 28.1% of dyslexic and 10.2% of control participants, whereas visual stress affected 5.5% of dyslexic and 8.5% of control participants. Furthermore, dyslexic participants showed a high degree of overlap between deficits, with 79.3% showing deficits in both phonological speed and accuracy, and 18.3% showing only one or the other deficit. All dyslexic participants with a visual attention span deficit also showed at least one form of phonological deficit, and none showed a pure visual attention span deficit. Similarly, no dyslexic child seemed to have visual stress without any concurrent phonological deficit.

For the purpose of comparison with the studies of Valdois and collaborators, we drew the scatterplot representing individual performance in phonological accuracy and VAS (see Figure 2). This figure can be compared with Figures 2 and 3 in Bosse et al. (2007) and with Figure 1 in Zoubrinetzky et al. (2014). In those studies, most dyslexic individuals showed either a phonological deficit, or a visual attention span deficit. In contrast, as shown in Figure 2, we find that most dyslexic children either had a pure phonological deficit, or a comorbid phonological and visual attention span deficit without a phonological accuracy deficit (and the analysis above showed that those three had a phonological speed deficit as well).

Because our conservative way of computing the visual attention span component (VAS, based on partial report only) might have led us to underestimate the prevalence of visual attention span deficits, we recomputed the prevalence of deficits using Valdois's version of the visual attention span (VAS1, mean of partial and global report). However, the results were almost identical to our previous analysis (see online supplementary Figure 1).

Table 7
Classification Into the Two Groups of Participants Using the Four Component Variables in a Logistic Regression Analysis

					Classifica	tion table	Model summary			
Variables in the equation	В	SE	p	Exp(B)	% Correct dyslexics	% Correct controls	-2 Log likelihood	Cox & Snell R ²	Nagelkerke R ²	
Step 1										
Phonological accuracy	2.15	.33	<.01	8.55	93.9	91.7	80.51	.64	.86	
Constant	3.15	.54	<.01	23.37						
Step 2										
Phonological speed	1.71	.44	<.01	5.50	95.9	95.8	43.73	.69	.93	
Phonological accuracy	2.15	.48	<.01	8.57						
Constant	5.40	1.09	<.01	221.34						
Step 3										
Phonological speed	1.99	.55	<.01	7.34	97.3	95.8	39.16	.69	.94	
Phonological accuracy	2.11	.47	<.01	8.21						
VAS	.83	.42	.05	2.29						
Constant	6.11	1.34	<.01	449.44						
Step 4										
Phonological speed	2.35	.65	<.01	10.46	96.6	95.8	32.92	.70	.95	
Phonological accuracy	2.66	.66	<.01	14.34						
VAS	1.07	.47	.02	2.92						
Visual stress	1.34	.62	.03	3.81						
Constant	7.47	1.79	<.01	1759.61						

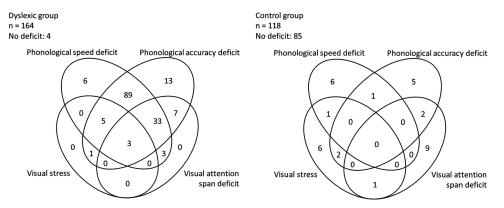


Figure 1. Prevalence of phonological and visual deficits and their combination in dyslexic and control groups of participants.

Finally, despite the results of the linear regression analyses showing that visual stress and visual attention span did not seem to contribute to explaining literacy skills beyond the contribution of phonological skills, we compared the literacy skills of children with a visual and a phonological deficit with those with a phonological deficit only. The results are given in Table 8.

This analysis suggests that dyslexic children with comorbid phonological and either visual attention span or visual stress deficits were not more reading impaired than children with a pure phonological deficit.

Discussion

The present study aimed to concurrently investigate three distinct types of cognitive deficits that are potential causes of devel-

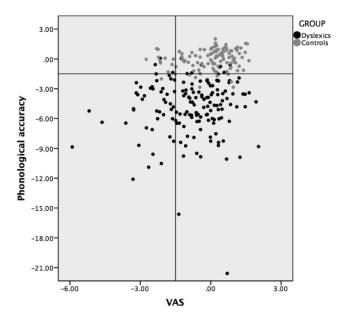


Figure 2. Scatterplot showing the distribution of dyslexic and control participants according to their visual attention span (VAS) and phonological accuracy deficit (z scores). The horizontal and vertical lines indicate the -1.5 standard deviation threshold.

opmental dyslexia: the phonological deficit, the visual attention span deficit, and visual stress. For this purpose, we administered the tests relevant to each hypothesis to a large French population of 164 dyslexic and 118 control children. Overall, our results are consistent with a major role of the phonological deficit, a minor role of the visual attention span, and no role of visual stress in developmental dyslexia.

Similar to many previous studies and consistent with metaanalyses (Kudo, Lussier, & Swanson, 2015; Melby-Lervåg, Lyster, & Hulme, 2012), we have found that dyslexic children are in general very impaired in phonological skills, as measured by tasks tapping phonological awareness, verbal short-term memory, and rapid naming. We observed that phonological deficits were highly prevalent in this group of dyslexic children (97%), and explained a large proportion of variance in literacy skills (from 40% for orthographic skills to 49% for reading speed and 59% for reading accuracy). A somewhat more original aspect of the present study is that, inspired by the results of an EFA suggesting two distinct sources of variance, we analyzed separately accuracy and speed in phonological performance. We thus distinguished, on the one hand, accuracy in phoneme deletion, spoonerisms, and digit span, and on the other hand, response times on the same measures as well as on rapid automatized naming. Nevertheless, we found that most dyslexic children (79%) were impaired on both phonological accuracy and speed. Both dimensions significantly contributed to the prediction of reading skills—phonological accuracy more so for reading accuracy and orthography, and phonological speed more so for reading speed—a result similar to that obtained in a large-scale cross-linguistic analysis (Landerl et al., 2013).

With respect to the visual attention span, we found that about 28% of dyslexic children were impaired in this measure, thus supporting its role in dyslexia. However, whereas Bosse et al. (2007) and Zoubrinetzky et al. (2014) reported a relatively strong dissociation between visual attention span and phonological deficits, we observed an important overlap. Indeed, all dyslexic children with an impaired visual attention span also showed a phonological deficit, either in accuracy or in speed, and none showed a pure visual attention span deficit. If one considers only phonological accuracy (as in the studies by Valdois and collaborators), three dyslexic children of 164 showed a pure visual attention span deficit. Furthermore, although visual attention span explained a

Table 8
Comparison of Subgroups Within the Dyslexic Group

	Group A Dyslexic children with phonological deficit only (n = 90)		Group B Dyslexic children with phonological deficit and visual stress (n = 9)		Grou Dyslexic with phor and v attention deficit (n	children ological isual n span	Groups A	& B	Groups A & C	
	Mean	SD	Mean	SD	Mean	SD	F(1, 148)	p	F(1, 111)	p
Reading accuracy	-5.85	2.85	-4.53	1.16	-5.57	2.38	2.79	.10	.49	.49
Reading speed	-11.00	7.57	-9.13	7.84	-12.40	11.14	.00	.98	1.26	.26
Orthography	-5.27	3.66	-3.83	3.39	-4.62	3.19	.05	.82	1.72	.19

Note. Mean z scores in dyslexic children with a pure phonological deficit compared with the ones with both phonological deficit and one of the visual deficits.

significant proportion of variance in literacy skills when entered first in the model (from 1% to 7%, depending on the literacy measure and on the visual attention span measure), it did not explain any additional variance on top of phonological skills, except when phonological speed was excluded from the model. Our results therefore contrast sharply with those of Valdois and collaborators in terms of the explanatory power of the visual attention span. Indeed, in the present study, only the existence of a few cases of dyslexia with a pure visual attention span deficit directly supports the hypothesis of the visual attention span deficit as an independent cause of developmental dyslexia. Most cases with a visual attention span deficit also show a phonological deficit, and for these cases, the presence of a visual attention span deficit does not seem to aggravate their reading disability. This is compatible with the view that the development of the visual attention span follows reading ability, and that a phonological deficit may therefore slow the development of the visual attention span. However, the argument cuts both ways: Because phonological skills are reciprocally influenced by reading ability, a reading disability initially induced by a visual attention span deficit is expected to delay the development of phonological skills, perhaps even to the point of meeting the criterion for a phonological deficit. Therefore, we cannot exclude that some cases in the present study actually had a visual attention span deficit as their primary cause of dyslexia, and a phonological deficit as a secondary outcome. Only longitudinal data would allow one to disentangle the causal pathways between phonological skills, visual attention span, and reading ability in the observed comorbid cases.

It remains unclear, to some extent, why our results are so different from those of Valdois and collaborators (Bosse et al., 2007; Zoubrinetzky et al., 2014). One thing that is particularly characteristic of the present sample is the high severity of the reading disability of the dyslexic children. Due to recruitment exclusively through hospital units, and to very stringent inclusion criteria, the group effect size is above 3 on the inclusion reading variables, above 2 on most literacy variables, and above 1 on all phonological variables (reaching 2 for spoonerisms). In z scores relative to the control group, some dyslexic children are as far as 10 standard deviations below controls on the phonological composite scores. This sample of dyslexic children is therefore much more severely impaired than those usually represented in the literature. The particularly high prevalence of the phonological deficit may be a consequence of this severity bias. From there, our

results follow relatively directly from the very high prevalence and severity of phonological deficits in the present sample of dyslexic children: Indeed, there was little room for another measure to explain many more cases or shares of variance.

If we compare our study more directly with those of Valdois and collaborators, the following observations can be made. First, our tests of the visual attention span were identical to those that they used. However, we adopted a relatively conservative approach to the measure of the visual attention span. In order to avoid potential confounds of the visual attention span with verbal memory and reporting ability and with letter retrieval, we used only the partial report measure, and we partialed out the control letter identification task. Reanalysis, presented as online supplemental materials using the more typical composite of partial and global report, confirms the hypothesis that methodological differences are reflected in the results: When the composite global and partial report variable is considered, it contributes to the variance in reading speed, although it is still not an important overall predictor of dyslexia. Furthermore, in the present study, we used a broad array of phonological tasks, and the EFA led us to compute two separate phonological components, one for accuracy and one for speed, whereas in the Valdois et al. study, only phonological accuracy is taken into account. Thus, they may have attributed to VAS variance that belonged to (unmeasured) phonological speed, and conversely, by adding one more phonological variable, we have reduced the chances of VAS to explain additional variance, which indeed becomes more important if we exclude phonological speed. Again, given the shared variance between VAS and phonological skills, only longitudinal data might allow us to tease their respective contributions apart. Finally, it is worth mentioning that, although we may have had different selection criteria from Valdois and collaborators in their studies, part of our sample (46 participants, of which 28 were dyslexic) was actually contributed by their clinic in Grenoble, and shows little sign of deviating from our general results, except in one respect: The sample from Grenoble exhibited a slightly less severe phonological accuracy deficit than other samples in this study (see online supplementary Table 5). This may reflect a recruitment bias, with that clinic being perhaps particularly attractive to children whose deficits have not been identified in other places. Thus, these children would show, on average, less severe phonological deficits (although here this manifests in phonological accuracy, not speed). Combined with the exclusive reliance of Valdois and collaborators on phonological

accuracy measures, this may be a partial explanation for their reporting relatively fewer cases of phonological deficit in their samples.

Turning to visual stress, we did not find any evidence that this phenomenon contributes to dyslexia, at least as measured using a subjective report of visual stress symptoms. Overall, children reported very few visual symptoms in absolute terms. Second, subjective reports on the stimulus meant to engender visual stress were highly correlated with reports on the control stimulus, suggesting that most of the variance in that measure reflected a general disposition to report symptoms, rather than a reaction to a specific spatial frequency. Thus, we partialed out reports on the control stimulus from those on the target stimulus in order to obtain a more specific measure of visual stress. On that measure, few children reported high symptoms, and, in total, 7% were beyond a 1.5-standard-deviation cutoff. If anything, more control than dyslexic children seemed to be affected. No dyslexic child (but five control children) seemed to be affected exclusively by visual stress. For those dyslexic children with a phonological deficit, reporting symptoms of visual stress was not associated with an increased severity of the reading disability. Our study differs from previous studies of visual stress in dyslexia by the criteria used for visual stress. Indeed, previous studies used improvement of reading fluency using a colored overlay as their main criterion. As argued in introduction, such improvement cannot by itself be taken as evidence for visual stress. It is interesting that some people can improve their reading fluency by using colored overlays, but this is not the same thing as saying that they had visual stress (or dyslexia) in the first place and that their disability was treated by the overlays. Overall, our results do not necessarily contradict the idea that visual stress exists and that it may affect some people's reading performance. However, the results do not support the hypothesis that visual stress can be the cause of such a severe reading disability as the one shown by the dyslexic children in the present study.

The conclusions that can be drawn from the present study are inevitably limited to the measures taken and to the population sampled. With respect to the measures, one potential limitation is that we have used visual attention span and visual stress measures provided by proponents of these deficits. In particular, given that letter-based measures of visual attention span have been criticized for not distinguishing visual attention and verbal components, our study is open to the same criticism. Because the visual attention span has been investigated by relatively few independent teams, we deemed it important to first attempt a direct replication with the same measures as in the original studies. Nevertheless, we have also attempted to take that criticism into account, by relying solely on partial report and by partialing out letter detection in our main analyses (while also using Valdois and collaborators preferred measures for a more direct comparison). At any rate, reliance on their measures would have been a problem if our results were blatantly in favor of their hypothesis, which is not the case.

Regarding the population sampled, ours is one of severely dyslexic French children. French has a relatively opaque orthography, so the results are expected to generalize at least to English and other similar languages (Landerl et al., 2013; Seymour, Aro, & Erskine, 2003; Ziegler et al., 2010), if not to all alphabetical languages. Given that visual stress has most often been tested in British populations, and visual attention span in French and British

populations, language should not be a source of divergence between the present and previous studies on these visual theories. As we have indicated, our results may also be skewed by our inclusion strategy. Unlike many studies, our dyslexic children are not just the 5%, 10%, or 15% poorest readers drawn from the general population. They have been recruited in specialized hospital units that tend to attract the most severe and complex cases of dyslexia. Furthermore, a stringent set of inclusion criteria ensured that all the children included had really severe reading disability, and excluded cases of comorbidity with low IQ, attention deficit, and oral language impairment. Thus, whereas there are, of course, a myriad of factors that influence reading acquisition and performance in the general population and in mild reading difficulties, including language, vision, attention, IQ, and many others, their relative contribution may have been much diminished by the selection of a dyslexic group that is both very severe and very specific. Our results suggest that when dyslexic children are selected in such a manner, the main (if not the only) way in which they differ from controls is that they show a very severe phonological deficit.

Nevertheless, we note that four dyslexic children out of 164 did not show any measurable phonological deficit, neither in accuracy nor in speed. These children did not show visual stress or a reduced visual attention span either, so their dyslexia remains unexplained by the current data set. This highlights another limitation of the present study, that is, that the results are limited to the theories investigated. We cannot speak about the relative prevalence and contribution of sluggish attentional shifting, anchoring deficit, perceptual noise exclusion, or other alternative theories to the reading ability of the children in the present sample. The only thing that we can say is that, given the high prevalence of phonological deficits in this population, there are very few cases left to demonstrate the independent contribution of another deficit to dyslexia. But it would be desirable to conduct other studies considering a broader range of potential causes of dyslexia, in order to obtain a clearer view of the different subtypes of dyslexia and of their relative prevalence.

References

Achenbach, T. M. (1991). Manual for the Child Behavior Checklist/4–18 and 1991 Profile. Burlington, VT: University of Vermont, Department of Psychiatry.

Ahissar, M. (2007). Dyslexia and the anchoring-deficit hypothesis. *Trends in Cognitive Sciences*, 11, 458–465. http://dx.doi.org/10.1016/j.tics.2007.08.015

Amitay, S., Ben-Yehudah, G., Banai, K., & Ahissar, M. (2002). Disabled readers suffer from visual and auditory impairments but not from a specific magnocellular deficit. *Brain: A Journal of Neurology, 125*, 2272–2285. http://dx.doi.org/10.1093/brain/awf231

Ans, B., Carbonnel, S., & Valdois, S. (1998). A connectionist multiple-trace memory model for polysyllabic word reading. *Psychological Review*, 105, 678–723. http://dx.doi.org/10.1037/0033-295X.105.4.678-723

Becker, J., Czamara, D., Scerri, T. S., Ramus, F., Csépe, V., Talcott, J. B.,
... Schumacher, J. (2014). Genetic analysis of dyslexia candidate genes in the European cross-linguistic NeuroDys cohort. *European Journal of Human Genetics*, 22, 675–680. http://dx.doi.org/10.1038/ejhg.2013.199
Boets, B., Op de Beeck, H. P., Vandermosten, M., Scott, S. K., Gillebert,

C. R., Mantini, D., . . . Ghesquière, P. (2013). Intact but less accessible phonetic representations in adults with dyslexia. *Science*, *342*, 1251–1254. http://dx.doi.org/10.1126/science.1244333

- Bosse, M. L., Tainturier, M. J., & Valdois, S. (2007). Developmental dyslexia: The visual attention span deficit hypothesis. *Cognition*, 104, 198–230. http://dx.doi.org/10.1016/j.cognition.2006.05.009
- Bouldoukian, J., Wilkins, A. J., & Evans, B. J. (2002). Randomised controlled trial of the effect of coloured overlays on the rate of reading of people with specific learning difficulties. *Ophthalmic and Physiological Optics*, 22, 55–60. http://dx.doi.org/10.1046/j.1475-1313.2002.00002.x
- Brady, S., & Shankweiler, D. (1991). *Phonological processes in literacy*. Hillsdale, NJ: Erlbaum.
- Dunlop, P. (1972). Dyslexia: The orthoptic approach. Australian Journal of Orthoptics, 12, 16–20.
- Evans, B. J., & Stevenson, S. J. (2008). The pattern glare test: A review and determination of normative values. *Ophthalmic and Physiological Optics*, 28, 295–309. http://dx.doi.org/10.1111/j.1475-1313.2008.00578.x
- Fabrigar, L. R., Wegener, D. T., MacCallum, R. C., & Strahan, E. J. (1999). Evaluating the use of exploratory factor analysis in psychological research. *Psychological Methods*, 4, 272–299. http://dx.doi.org/10.1037/1082-989X.4.3.272
- Fischer, F. W., Liberman, I. Y., & Shankweiler, D. (1978). Reading reversals and developmental dyslexia: A further study. Cortex: A Journal Devoted to the Study of the Nervous System and Behavior, 14, 496–510. http://dx.doi.org/10.1016/S0010-9452(78)80025-2
- Gori, S., & Facoetti, A. (2015). How the visual aspects can be crucial in reading acquisition? The intriguing case of crowding and developmental dyslexia. *Journal of Vision*, 15, 8. http://dx.doi.org/10.1167/15.1.8
- Goswami, U. (2015). Sensory theories of developmental dyslexia: Three challenges for research. *Nature Reviews Neuroscience*, 16, 43–54. http:// dx.doi.org/10.1038/nrn3836
- Hallgren, B. (1950). Specific dyslexia (congenital word-blindness); a clinical and genetic study. Acta Psychiatrica et Neurologica Supplementum, 65, 1–287.
- Hari, R., & Renvall, H. (2001). Impaired processing of rapid stimulus sequences in dyslexia. *Trends in Cognitive Sciences*, 5, 525–532. http:// dx.doi.org/10.1016/S1364-6613(00)01801-5
- Hawelka, S., & Wimmer, H. (2008). Visual target detection is not impaired in dyslexic readers. Vision Research, 48, 850–852. http://dx.doi.org/10 .1016/j.visres.2007.11.003
- Hinshelwood, J. (1900). Congenital word blindness. *Lancet*, 155, 1506–1508. http://dx.doi.org/10.1016/S0140-6736(01)99645-X
- Hollis, J., & Allen, P. M. (2006). Screening for Meares-Irlen sensitivity in adults: Can assessment methods predict changes in reading speed? *Ophthalmic and Physiological Optics*, 26, 566–571. http://dx.doi.org/10 .1111/j.1475-1313,2006,00401.x
- Hornickel, J., & Kraus, N. (2013). Unstable representation of sound: A biological marker of dyslexia. *The Journal of Neuroscience*, 33, 3500– 3504. http://dx.doi.org/10.1523/JNEUROSCI.4205-12.2013
- Irlen, H. (1991). Reading by the colors: Overcoming dyslexia and other reading disabilities through the Irlen method. New York, NY: Avery.
- Jacquier-Roux, M., Valdois, S., & Zorman, M. (2002). Outil de dépistage des dyslexies [Dyslexia screening tool]. Grenoble, France: Laboratoire de Cogni-Sciences IUFM.
- Kriss, I., & Evans, B. J. (2005). The relationship between dyslexia and Meares-Irlen Syndrome. *Journal of Research in Reading*, 28, 350–364. http://dx.doi.org/10.1111/j.1467-9817.2005.00274.x
- Kudo, M. F., Lussier, C. M., & Swanson, H. L. (2015). Reading disabilities in children: A selective meta-analysis of the cognitive literature. *Re-search in Developmental Disabilities*, 40, 51–62. http://dx.doi.org/10 .1016/j.ridd.2015.01.002
- Landerl, K., Ramus, F., Moll, K., Lyytinen, H., Leppänen, P. H. T., Lohvansuu, K., . . . Schulte-Körne, G. (2013). Predictors of developmental dyslexia in European orthographies with varying complexity. *Journal of Child Psychology and Psychiatry*, 54, 686–694. http://dx.doi.org/10.1111/jcpp.12029

- Lefavrais, P. (1967). *Test de l'Alouette* (2nd ed.) [The Lark test]. Paris, France: Editions du Centre de Psychologie Appliquée.
- Liberman, I. Y. (1973). Segmentation of the spoken word and reading acquisition. Bulletin of the Orton Society, 23, 64–77. http://dx.doi.org/ 10.1007/BF02653842
- Lindgren, S. D., De Renzi, E., & Richman, L. C. (1985). Cross-national comparisons of developmental dyslexia in Italy and the United States. *Child Development*, 56, 1404–1417. http://dx.doi.org/10.2307/1130460
- Lobier, M., & Valdois, S. (2015). Visual attention deficits in developmental dyslexia cannot be ascribed solely to poor reading experience. *Nature Reviews Neuroscience*, 16, 225. http://dx.doi.org/10.1038/nrn3836-c1
- Lobier, M., Zoubrinetzky, R., & Valdois, S. (2012). The visual attention span deficit in dyslexia is visual and not verbal. *Cortex: A Journal Devoted to the Study of the Nervous System and Behavior*, 48, 768–773. http://dx.doi.org/10.1016/j.cortex.2011.09.003
- Martinet, C., & Valdois, S. (1999). L'apprentissage de l'orthographe d'usage et ses troubles dans la dyslexie développementale de surface [Learning of conventional orthography and its disorders in surface developmental dyslexia]. L'Année Psychologique, 99, 577–622. http:// dx.doi.org/10.3406/psy.1999.28496
- Meares, O. (1980). Figure/background brightness/contrast and reading disability. Visible Language, 14, 13–29.
- Melby-Lervåg, M., Lyster, S. A. H., & Hulme, C. (2012). Phonological skills and their role in learning to read: A meta-analytic review. Psychological Bulletin, 138, 322–352. http://dx.doi.org/10.1037/a0026744
- Morgan, W. P. (1896). A case of congenital word blindness. *British Medical Journal*, 2, 1378. http://dx.doi.org/10.1136/bmj.2.1871.1378
- Nicolson, R. I., & Fawcett, A. J. (2007). Procedural learning difficulties: Reuniting the developmental disorders? *Trends in Neurosciences*, 30, 135–141. http://dx.doi.org/10.1016/j.tins.2007.02.003
- Orton, S. (1937). Reading, writing and speech problems in children. New York, NY: Norton.
- Peterson, R. L., & Pennington, B. F. (2015). Developmental dyslexia. Annual Review of Clinical Psychology, 11, 283–307. http://dx.doi.org/ 10.1146/annurev-clinpsy-032814-112842
- Peyrin, C., Lallier, M., Démonet, J. F., Pernet, C., Baciu, M., Le Bas, J. F., & Valdois, S. (2012). Neural dissociation of phonological and visual attention span disorders in developmental dyslexia: FMRI evidence from two case reports. *Brain and Language*, 120, 381–394. http://dx.doi.org/ 10.1016/j.bandl.2011.12.015
- Ramus, F. (2003). Developmental dyslexia: Specific phonological deficit or general sensorimotor dysfunction? *Current Opinion in Neurobiology*, 13, 212–218. http://dx.doi.org/10.1016/S0959-4388(03)00035-7
- Ramus, F. (2014). Neuroimaging sheds new light on the phonological deficit in dyslexia. *Trends in Cognitive Sciences*, 18, 274–275. http:// dx.doi.org/10.1016/j.tics.2014.01.009
- Ramus, F., & Ahissar, M. (2012). Developmental dyslexia: The difficulties of interpreting poor performance, and the importance of normal performance. *Cognitive Neuropsychology*, *29*, 104–122. http://dx.doi.org/10.1080/02643294.2012.677420
- Ramus, F., Marshall, C. R., Rosen, S., & van der Lely, H. K. J. (2013). Phonological deficits in specific language impairment and developmental dyslexia: Towards a multidimensional model. *Brain: A Journal of Neurology*, 136, 630–645. http://dx.doi.org/10.1093/brain/aws356
- Ramus, F., Pidgeon, E., & Frith, U. (2003). The relationship between motor control and phonology in dyslexic children. *Journal of Child Psychology* and Psychiatry, 44, 712–722. http://dx.doi.org/10.1111/1469-7610 .00157
- Ramus, F., Rosen, S., Dakin, S. C., Day, B. L., Castellote, J. M., White, S., & Frith, U. (2003). Theories of developmental dyslexia: Insights from a multiple case study of dyslexic adults. *Brain: A Journal of Neurology*, 126, 841–865. http://dx.doi.org/10.1093/brain/awg076

Ramus, F., & Szenkovits, G. (2008). What phonological deficit? The Quarterly Journal of Experimental Psychology, 61, 129–141. http://dx .doi.org/10.1080/17470210701508822

- Ritchie, S. J., Della Sala, S., & McIntosh, R. D. (2011). Irlen colored overlays do not alleviate reading difficulties. *Pediatrics*, 128, e932– e938. http://dx.doi.org/10.1542/peds.2011-0314
- Ritchie, S. J., Della Sala, S., & McIntosh, R. D. (2012). Irlen colored filters in the classroom: A 1-year follow-up. *Mind, Brain, and Education, 6*, 74–80. http://dx.doi.org/10.1111/j.1751-228X.2012.01139.x
- Seymour, P. H. K., Aro, M., & Erskine, J. M. (2003). Foundation literacy acquisition in European orthographies. *British Journal of Psychology*, 94, 143–174. http://dx.doi.org/10.1348/000712603321661859
- Shankweiler, D. P., & Liberman, I. Y. (1972). Misreading: A search for causes. In J. F. Kavanagh & I. G. Mattingly (Eds.), Language by ear and by eye: The relationships between speech and reading (pp. 293–317). Cambridge, MA: MIT Press.
- Singleton, C., & Henderson, L. M. (2007). Computerized screening for visual stress in children with dyslexia. *Dyslexia: An International Jour*nal of Research and Practice, 13, 130–151. http://dx.doi.org/10.1002/ dvs.329
- Skottun, B. C. (2000). The magnocellular deficit theory of dyslexia: The evidence from contrast sensitivity. *Vision Research*, 40, 111–127. http:// dx.doi.org/10.1016/S0042-6989(99)00170-4
- Sperling, A. J., Lu, Z. L., Manis, F. R., & Seidenberg, M. S. (2005). Deficits in perceptual noise exclusion in developmental dyslexia. *Nature Neuroscience*, 8, 862–863. http://dx.doi.org/10.1038/nn1474
- Sprenger-Charolles, L., Béchennec, D., Colé, P., & Kipffer-Piquard, A. (2005). French normative data on reading and related skills from EVA-LEC, a new computerized battery of tests (end Grade 1, Grade 2, Grade 3, and Grade 4). European Review of Applied Psychology, 55, 157–186. http://dx.doi.org/10.1016/j.erap.2004.11.002
- Stein, J., & Walsh, V. (1997). To see but not to read; the magnocellular theory of dyslexia. *Trends in Neurosciences*, 20, 147–152. http://dx.doi .org/10.1016/S0166-2236(96)01005-3
- Stephenson, S. (1907). Six cases of congenital word-blindness affecting three generations of one family. *Ophthalmoscope*, 5, 482–484.
- Tallal, P., Miller, S., & Fitch, R. H. (1993). Neurobiological basis of speech: A case for the preeminence of temporal processing. *Annals of the New York Academy of Sciences*, 682, 27–47. http://dx.doi.org/10 .1111/j.1749-6632.1993.tb22957.x
- Vidyasagar, T. R., & Pammer, K. (2010). Dyslexia: A deficit in visuo-spatial attention, not in phonological processing. *Trends in Cognitive Sciences*, 14, 57–63. http://dx.doi.org/10.1016/j.tics.2009.12.003
- Wechsler, D. (2005). WISC-IV: Echelle d'Intelligence de Wechsler pour Enfants (4th ed.) [Wechsler Intelligence Scale for Children]. Paris, France: Editions du Centre de Psychologie Appliquée.

- White, S., Frith, U., Milne, E., Rosen, S., Swettenham, J., & Ramus, F. (2006). A double dissociation between sensorimotor impairments and reading disability: A comparison of autistic and dyslexic children. Cognitive Neuropsychology, 23, 748–761. http://dx.doi.org/10.1080/02643290500438607
- White, S., Milne, E., Rosen, S., Hansen, P., Swettenham, J., Frith, U., & Ramus, F. (2006). The role of sensorimotor impairments in dyslexia: A multiple case study of dyslexic children. *Developmental Science*, 9, 237–255. http://dx.doi.org/10.1111/j.1467-7687.2006.00483.x
- Wilkins, A. J. (1995). Visual stress. Oxford, UK/New York: Oxford University Press. http://dx.doi.org/10.1093/acprof:oso/9780198521747 .001.0001
- Wilkins, A. J., Huang, J., & Cao, Y. (2004). Visual stress theory and its application to reading and reading tests. *Journal of Research in Reading*, 27, 152–162. http://dx.doi.org/10.1111/j.1467-9817.2004.00223.x
- Wilkins, A. J., Lewis, E., Smith, F., Rowland, E., & Tweedie, W. (2001).
 Coloured overlays and their benefit for reading. *Journal of Research in Reading*, 24, 41–64. http://dx.doi.org/10.1111/1467-9817.00132
- Wilkins, A., Nimmo-Smith, I., Tait, A., McManus, C., Della Sala, S., Tilley, A., . . . Scott, S. (1984). A neurological basis for visual discomfort. *Brain: A Journal of Neurology*, 107, 989–1017. http://dx.doi.org/ 10.1093/brain/107.4.989
- Wilkins, A. J., & Nimmo-Smith, M. I. (1987). The clarity and comfort of printed text. *Ergonomics*, 30, 1705–1720. http://dx.doi.org/10.1080/ 00140138708966059
- World Health Organization. (2011). *International Statistical Classification of Diseases and Related Health Problems 10th revision* (4th ed.). Geneva, Switzerland: World Health Organization.
- Ziegler, J. C., Bertrand, D., Tóth, D., Csépe, V., Reis, A., Faísca, L., . . . Blomert, L. (2010). Orthographic depth and its impact on universal predictors of reading: A cross-language investigation. *Psychological Science*, 21, 551–559. http://dx.doi.org/10.1177/0956797610363406
- Ziegler, J. C., Pech-Georgel, C., Dufau, S., & Grainger, J. (2010). Rapid processing of letters, digits and symbols: What purely visual-attentional deficit in developmental dyslexia? *Developmental Science*, *13*, F8–F14. http://dx.doi.org/10.1111/j.1467-7687.2010.00983.x
- Zoubrinetzky, R., Bielle, F., & Valdois, S. (2014). New insights on developmental dyslexia subtypes: Heterogeneity of mixed reading profiles. *PLoS ONE*, 9(6), e99337. http://dx.doi.org/10.1371/journal.pone 0099337

Received June 22, 2015
Revision received June 16, 2016
Accepted June 16, 2016