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Dyslexia heterogeneity: cognitive profiling of Portuguese children with dyslexia

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Abstract Recent studies have emphasized that developmental dyslexia is a multiple-deficit disorder, in contrast to the traditional single-deficit view. In this context, cognitive profiling of children with dyslexia may be a relevant contribution to this unresolved discussion. The aim of this study was to profile 36 Portuguese children with dyslexia from the 2nd to 5th grade. Hierarchical cluster analysis was used to group participants according to their phonological awareness, rapid automatized naming, verbal short-term memory, vocabulary, and nonverbal intelligence abilities. The results suggested a two-cluster solution: a group with poorer performance on phoneme deletion and rapid automatized naming compared with the remaining variables (Cluster 1) and a group characterized by underperforming on the variables most related to phonological processing (phoneme deletion and digit span), but not on rapid automatized naming (Cluster 2). Overall, the results seem more consistent with a hybrid perspective, such as that proposed by Pennington and colleagues (2012), for understanding the heterogeneity of dyslexia. The importance of characterizing the profiles of individuals with dyslexia becomes clear within the context

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of constructing remediation programs that are specifically targeted and are more effective in terms of intervention outcome.

Keywords Reading disorders · Dyslexia · Profile analysis · Portuguese orthography

Introduction

Developmental dyslexia has been described as a neurobiologically based learning-specific deficit related to the acquisition of reading and writing skills, including difficulties with accurate or fluent word recognition and poor spelling and encoding abilities (Lyon, Shaywitz, & Shaywitz, 2003). According to the World Health Organization (2008), reading deficits observed in children with dyslexia cannot be explained by low intelligence, poor educational opportunities or evident sensory or neurological damage. Although there has been a consensus in the literature regarding the presence of phonological impairments in individuals with dyslexia (for a review, see Ramus & Szenkovits, 2008; Ramus et al., 2003), some recent studies have suggested that this deficit may not be the core impairment of all dyslexic cases (see, for example, Pennington et al., 2012; Vidyasagar & Pammer, 2010). The idea that this disorder could have heterogeneous cognitive characteristics is increasingly discussed in the literature (Menghini et al., 2010), and considerable effort is currently being invested in trying to identify the existence of distinct cognitive profiles for reading disorders (Chung, Ho, Chan, Tsang, & Lee, 2010; Hedman, 2012; Heim et al., 2008; Jimenez et al., 2011).

There are several theoretical frameworks according to which the diversity of children with dyslexia can be characterized and grouped (Coltheart, Curtis, Atkins, & Haller, 1993; Pennington, 2006; Ramus et al., 2003; Snowling, 1981; Ziegler et al., 2008). For example, the single-phonological-deficit account assumes that a deficit in phonological processing is necessary and sufficient to cause dyslexia (Ramus et al., 2003; Snowling, 1981) and does not predict the existence of qualitatively different subtypes in dyslexia. From a different perspective, Coltheart et al. (1993) proposed a dual-route model for reading, which allows for descriptions of dyslexia deficits localized at the lexical (orthographic), nonlexical (phonological) or both routes for reading, which would generate surface, phonological and deep dyslexia, respectively (Coltheart, Rastle, Perry, Langdon, & Ziegler, 2001). Pennington (2006) emphasized the multiple-deficit nature of dyslexia, arguing for a probabilistic and multifactorial etiological model based on the argument that dyslexia cannot be accounted for by a single deficit. According to this model, the current understanding of the neuropsychology of reading disorders suggests that at least three cognitive risk factors are involved. Thus, in addition to the defining difficulties with written language, individuals with dyslexia are expected to have deficits in at least three areas of cognition: processing speed, phonological awareness, and language skills (Pennington & Bishop, 2009).

More recently, Menghini et al. (2010) added support to the perspective of dyslexia as a multifactorial deficit, documenting impairments in individuals with dyslexia on both phonological and non-phonological tasks. Thus, the multiple-deficit perspective on developmental dyslexia suggests that cognitive profiling (cognitive phenotype characterization) is essential for the understanding of dyslexia subtypes.

Wolf and Bowers' double-deficit hypothesis (1999) can be situated somewhere between the single-deficit approach and the multifactorial model for dyslexia. According to this hypothesis, both phonological and rapid-naming deficits are associated with reading impairments, establishing three subtypes of individuals with dyslexia: an exclusively phonologically impaired group, an exclusively rapid-automatized-naming deficit group, and a third group with impairments in both skills. O'Brien, Wolf and Lovett (2012) aimed to verify the double-deficit hypothesis using taxometric classification techniques in a large sample of children with reading difficulties. The authors found a group of children with dyslexia characterized by phonological deficits as well as a group with intact phonological skills but with naming speed and reading fluency difficulties. The hypothetic subtypes for the double-deficit hypothesis have been supported by other empirical evidence. Using clustering techniques, King, Giess and Lombardino (2007) classified 93 English-speaking children with developmental dyslexia and 93 children without reading disability matched for gender and age. They found no clusters in the unimpaired children and three distinct clusters in children with dyslexia, corresponding to the prediction of the double-deficit hypothesis (a phonological deficit cluster, a rapid-naming-deficit cluster, and a third cluster with both deficits). They also identified a fourth cluster that aggregated older children with typical phonological and rapid naming skills, many of them having benefited from previous reading therapy and thus possibly being 'compensated' readers. Similar findings were obtained in a recent study of Portuguese children with dyslexia (Araújo, Pacheco, Faísca, Petersson, & Reis, 2010). Overall, these results support the double-deficit hypothesis and suggest the existence of an association between rapid naming skills and reading performance, independent from phonological contributions, highlighting how the double-deficit model emphasizes the role of variables other than phonological processing in reading. This has subsequently been corroborated by several studies showing that rapid automatized naming, together with phonological skills, is an important predictor for reading success (see Kirby, Georgiou, Martinussen, & Parrila, 2010, for a recent review). However, Pennington et al. (2012), in a study involving 165 children with dyslexia, found that more than half the cases did not fall into one of the three subtypes defined by the double-deficit model. Based on their findings, Pennington et al. (2012) argued that there are multiple possible pathways to reading disorders, which translates to multiple subtypes of dyslexia. Some subtypes are expected to have difficulties resulting from a single deficit, and others are expected to have more generalized difficulties that emerge across multiple deficits. Thus, according to these authors, all competing models of dyslexia are possible.

Taken as a whole, there is still an unresolved discussion about the single- versus multiple-deficit models in the dyslexia literature, with phonological core deficit and double-deficit accounts still prevalent in addition to the multiple-deficit models proposed more recently. Within the context of this discussion, Pennington et al.

(2012) propose a hybrid model that encompasses all competing models of dyslexia and thus allows for the occurrence of either single or multiple deficit individual profiles.

The orthography by which children learn to read has been identified as one factor that influences both reading skill acquisition and the expression of dyslexia. Orthographies can be classified along a depth (inconsistency) continuum regarding their syllabic complexity and symbol-sound consistency (see Seymour et al., 2003, for a classification of European orthographies). For example, Portuguese orthography is characterized as presenting a simple syllabic structure with medium depth and as being more irregular for spelling than for reading (i.e., grapheme-phoneme conversion has less exceptions in reading), which explains its designation as an asymmetric orthography (Seymour et al., 2003; Sucena, Castro, & Seymour, 2009). Seymour et al. (2003) compared children at the end of the first grade from 13 different alphabetic orthographies, observing that reading accuracy was close to the ceiling in shallow orthographies but that children acquiring more complex, inconsistent orthographies were still struggling to master reading skills. To examine the influence of orthographic systems in reading, Ziegler and Goswami (2005, 2006) proposed the grain size theory, which suggests that shallow orthographies with high symbol-sound consistency are acquired more easily than are complex and deep orthographies with a high proportion of irregular spelling. According to this theory, in shallow orthographies reading is based on smaller units, which contrasts with more deep orthographies in which highly inconsistent phoneme-grapheme mappings require readers to use larger phonological units, adopting more flexible reading strategies. If children learning to read in different orthographies need to address phonological units of various sizes, then the cognitive mechanisms underlying reading acquisition and dyslexia are expected to be different among orthographies. Recently, some cross-linguistics studies have addressed this question with typical (Vaessen et al., 2010; Ziegler et al., 2010) and impaired (Landerl et al., 2013) readers. In general, phonological processing and rapid automatized naming are associated with reading accuracy and speed across orthographies; phonological processing seems to be a stronger predictor than is rapid automatized naming for word reading accuracy, whereas rapid automatized naming is a stronger predictor for time-locked reading measures (Norton & Wolf, 2012). However, the relative contributions of these predictors seem to be modulated by orthographic depth and reading expertise. That is, phonological processing may be more relevant in deep orthographies in which its influence remains for a longer period of time, whereas children reading more shallow orthographies will shift away earlier from a predominant reliance on phonology. In contrast, orthographic consistency seems to not influence the contribution of rapid naming to reading fluency, indicating that the strength of the relationship between rapid naming and reading is equally strong in deep and shallow orthographies (Vaessen et al., 2010). The interplay between the different predictors according to the orthographies seems to be explained by the fact that grapheme-phoneme mappings in shallow orthographies are simpler to learn compared with deep orthographies in which it is more difficult to grasp the basic principle of the alphabetic structure. The existence of ambiguous grapheme-phoneme correspondences in a deep orthography requires children to deal with differently sized units and to develop multiple phonological decoding strategies (as suggested by Ziegler & Goswami, 2005), explaining the stronger contribution of phonological processing in these orthographies compared with children learning to read in more shallow orthographies.

The differential effects of orthography on reading predictors are equally relevant to reading deficits. For instance, in shallow orthographies such as Spanish or German (e.g., Escribano, 2007; Wimmer, 1993, 1996), word reading accuracy is not a major problem for poor readers, and deficits mainly show up in reading speed scores. For deep orthographies such as English, however, poor readers are characterized by low scores on both accuracy and speed. These results suggest that dyslexia in a shallow orthography does not prevent the development of satisfactory decoding skills, as happens in deep orthographies. Altogether, these findings seem to imply that in addition to the cognitive heterogeneity that may characterize dyslexia, we have to consider that the characteristics of a reading disorder also might depend on the structure (deep vs. shallow) of its orthography. Therefore, the need to develop more flexible inclusion criteria for each of the possible specific cognitive profiles emerges, and the imperative to better characterize the profiles of individuals with dyslexia arises, to guarantee that the needed intervention can be better directed.

Considering that dyslexia symptoms might be heterogeneous, our goal in this study was to characterize the cognitive profiles of Portuguese children with dyslexia using a hierarchical cluster approach. We decided to aggregate participants based on the most commonly used cognitive measures associated with reading performance: phonological awareness, rapid automatized naming, verbal short-term memory, vocabulary, and intelligence. Both phonological awareness and rapid automatized naming have consistently been associated with reading performance and reading impairment (Wolf & Bowers, 1999). We also used tests of vocabulary knowledge and verbal short-term memory, measures that are consistently related to reading skills: oral vocabulary knowledge promotes written word recognition (Ouellettte & Beers, 2010), and verbal short-term memory is thought to facilitate reading (Jackson & Myers, 1982; Martinez-Perez, Majerus, & Poncelet, 2012). Although all children need to have normal-range cognitive ability for a dyslexia diagnosis, we added a nonverbal intelligence measure to our analysis. Recent research (Ferrer, Shaywitz, Holahan, Marchione, & Shaywitz, 2010) suggests that reading and intelligence might be uncoupled in individuals with dyslexia, in contrast to typical readers. If this is the case, intelligence can be considered a variable that might differentiate reading cognitive profiles within dyslexia disorders.

Method

Participants

Thirty-six children with dyslexia (22 males and 14 females; mean age ± standard deviation = 9.5 years ± 1.14; minimum–maximum = 7.5–11.7 years) attending regular school from the 2nd to 5th grade (children per grade: 2nd grade = 8;

3rd grade = 11; 4th grade = 16; 5th grade = 1) participated in this study. Eight children had repeated a school year, and three had repeated two school years. The participants were recruited from schools to clinics specializing in psychological assessment, and they all had a clinical diagnosis of dyslexia and no history of neurological, emotional, or attentional problems. For the inclusion criteria, participants had to have a normal-range nonverbal IQ performance (above the 25th percentile), and their reading speed scores had to be significantly below the expected grade mean level—at least 1.5 SDs below the age/grade mean defined by the Portuguese normative sample (Reis et al., 2011) for the Portuguese version of the Differential Diagnosis Dyslexia Maastricht Battery (Blomert & Vaessen, 2009). The results on the cognitive (phonological awareness, rapid automatized naming, vocabulary, verbal short-term memory, and nonverbal intelligence) and alphabetic (reading words, pseudowords, and spelling) measures are tabulated in Table 1.

Materials and Procedure

Participants were assessed on reading, spelling, phonological awareness, and rapid automatized naming skills using the Portuguese version (Reis et al., 2011) of the Differential Diagnosis Dyslexia Maastricht Battery (3DM Battery; Blomert & Vaessen, 2009). In addition, vocabulary knowledge and verbal short-term memory span were assessed using vocabulary and digit-span tasks taken from the Wechsler Intelligence Scale for Children (WISC-III; Wechsler, 2006). The nonverbal intelligence measure was obtained through Raven Coloured Progressive Matrices—Parallel form (Raven, Raven, & Court, 2009).

Reading words and pseudowords (RWR and PWR)

The 3DM reading test is a time-limited reading-aloud task composed of three lists of high- and low-frequency words (RWR) and pseudowords (PWR). For each list, the children had 30 s to read aloud as many words as possible. Each list was composed of 75 stimuli distributed on five sheets (15 stimuli per sheet) of increasing difficulty with respect to the number of syllables (2–4), syllabic structure (with and without consonant clusters), and phoneme–grapheme correspondence rules (regular and irregular). The stimulus from the high-frequency (e.g., ⟨*lata*⟩; frequency range 55–397) and low-frequency lists (e.g., ⟨*lota*⟩; frequency range 10–54) were selected from among the 1,000 most frequent words of a 600-thousand-token lexical database developed from 42 reader books used in Portuguese primary schools between the first and fourth grades (Faísca, Bramão, Araújo, Pacheco, & Reis, 2006). The pseudowords were derived from the high-frequency words, which were separated into syllables and then rearranged to form pseudowords (e.g., ⟨*lano*⟩). Reading speed was computed as the number of correctly read stimuli per second. The percentage of correctly read stimuli was taken as the reading accuracy measure. The test–retest reliability for the 3DM reading measures was high ($r > 0.9$).

Table 1 Results on cognitive and alphabetic measures (raw scores and z scores: mean ± SD), and collinearity statistics for cognitive measures (VIF)

Tasks	Raw scores Mean ± SD	Z scores Mean ± SD	Variance inflation factor (VIF)
PD	35.5 ± 23.26	−1.35 ± 0.578	1.52
RAN	0.85 ± 0.208	−0.95 ± 0.911	1.12
DS	8.9 ± 2.04	−1.06 ± 0.841	1.28
VOC	16.4 ± 4.66	−0.37 ± 0.853	1.38
NVIQ	80.0 ± 11.67	0.43 ± 0.723	1.03
RWR			
RWR (speed)	0.31 ± 0.185	−1.92 ± 0.351	–
RWR (accuracy)	70.4 ± 20.71	−1.93 ± 0.982	–
PWR			
PWR (speed)	0.29 ± 0.153	−1.79 ± 0.488	–
PWR (accuracy)	69.5 ± 21.86	−1.41 ± 0.901	–
SP	61.7 ± 16.53	−1.50 ± 0.685	–

Real word reading (RWR-speed: correct items/second; RWR-accuracy: percentage of correct items), pseudoword reading (PWR-speed: correct items/second; PWR-accuracy: percentage of correct items), spelling (SP: percentage of accuracy), phoneme deletion (PD: percentage of accuracy), rapid automatized naming (RAN: correct items/second), digit span (DS: number of repeated series), vocabulary (VOC), and non verbal intelligence (NVIQ: percentage of accuracy)

Spelling (SP)

In the 3DM spelling task, a complete word was presented auditorily (e.g., */'sapu/*), while part of the word was simultaneously presented visually (e.g., ⟨*sa_*⟩). The child had to complete the visual stimulus by choosing the correct letter(s) from among four alternatives. Syllabic structure (simple or complex), number of syllables (two, three, and four), position of the omitted syllable (initial, medial, and final), grapheme-phoneme correspondence (from 1:1 mapping to exceptions), and frequency (high and low) were accounted for. This task comprised 96 words, and accuracy was considered the measure for spelling skills (reliability: *Cronbach's* $\alpha = 0.920$).

Phoneme deletion (PD)

Phonological awareness was evaluated using a phoneme deletion task composed of 44 pseudowords. Each auditorily presented pseudoword had to be repeated by the child after he or she excluded a specific phoneme (e.g., */dul/minus/d/=/ul/*). Accuracy was taken as the phonological awareness measure (*Cronbach's* $\alpha = 0.946$).

Rapid automatized naming (RAN)

Based on the classical paradigm by Denckla and Rudel (1976), a rapid-automatized-naming task with objects was designed with five different stimuli (*apple*, *bed*, *shoe*, *spoon*, *glass*) presented in two blocks (15 items per block). Participants were instructed to name the stimuli as quickly and accurately as possible. The number of correctly named items per second was computed as a measure of the rapid automatized naming. The correlation between the performance on the two blocks of stimuli was used as a reliability index ($r = 0.704$).

Vocabulary (VOC)

The vocabulary subtest of the WISC-III was used (Wechsler, 2006). Children were asked to explain the meaning of a maximum of 30 words (e.g., *What is a hat?*). Each item could have a maximum of two points and a minimum of zero (up to a maximum total of 60 points). After four consecutive errors, the examiner considered the task completed.

Digit span (DS)

The digit span task from the WISC-III (Wechsler, 2006) was used to evaluate verbal short-term memory. On each trial, a sequence of digits was read aloud to the child and the task was to repeat the sequence in exactly the same order (forward DS) or in the inverse order (backward DS). There were two trials per sequence length (sequence lengths from two to eight digits), and each correct response corresponded to one point (up to a maximum of 30 points).

Nonverbal Intelligence (NVIQ)

Nonverbal IQ was assessed using Raven Coloured Progressive Matrices (Raven, Raven, & Court, 2009). This test is composed of three series of twelve items each, for a maximum score of 36 points.

All measures were transformed into standardized z scores according to age/grade norms adapted to the Portuguese school population. Standardized scores for the 3DM tasks (RWR, PWR, SP, PD, RAN) were computed with reference to a large-scale study with Portuguese students ($n = 820$, grades 1st–4th; Reis et al., 2011). The z scores for the 5th grade children were estimated through polynomial regression procedures (Van Breukelen & Vlaeyen, 2005), with the number of months of formal reading instruction as the predictor. For the remaining measures (VOC, DS, NVIQ), Portuguese published norms were used. The tasks were applied in a controlled environment in several individual sessions with a duration no longer than 30 min each (usually three sessions were required, one each day). All children were assessed after school board and parental informed consent was obtained, in compliance with the Helsinki Declaration. Tasks from the 3DM battery were displayed on a computer screen via Presentation Software (version 11.0; www.neurobs.com), and a headset was used during all the tasks.

Cluster analysis

Cluster analysis was chosen as the statistical approach to subtype children with dyslexia. Because the objective was to differentiate (profile) children with dyslexia into different cognitive subtypes, only cognitive variables (PD, RAN, VOC, DS, and NVIQ) were used in the analysis. Reading and spelling measures were considered the characterization variables, with the intent of describing a posteriori the clusters found.

All variables were expressed as *z* scores, and Mahalanobis D^2 distances were computed to confirm the nonexistence of multivariate outliers in the sample (*p* values associated with D^2 were >0.02 for all participants). Multicollinearity was also evaluated, and the collinearity statistics were acceptable for all variables (variance inflation factor <5, according to Hair, Anderson, Tatham, & Black, 1992; see Table 1). For the hierarchical clustering analysis, squared Euclidian distance and Ward's agglomerative clustering method were chosen because of their statistical advantages, as denoted in similar studies (Crews & D'Amato, 2009; Milligan & Cooper, 1987). King, Giess, and Lombardino (2007) highlight the problems hierarchical clustering methods may bring, particularly the difficulty in determining the optimal number of clusters and evaluating the stability of the obtained solution. To avoid these hazards, the gap statistic was used to estimate the true number of clusters in the sample (Tibshirani, Walther, & Hastie, 2001).

The gap statistic method involves generating 1, 2, …, k cluster solutions from the original data matrix and computing a goodness-of-fit index for each solution, W_k (the within-cluster sum-of-squares, a measure of within-cluster dispersion, was computed and then averaged across clusters to evaluate the global quality of each solution). If W_k is plotted as a function of k, then a monotonically decreasing function is obtained. Typically, W_k decreases noticeably when the number of clusters increases (because initial clusters are too generic and become less dispersed when fractioned into smaller clusters). However, when the number of clusters in the solution becomes excessive and surpasses the "true" number of clusters in the population, W_k changes tend to diminish and become irrelevant (indicating that natural subgroups begin to break up). Thus, the estimation of the ideal number of clusters can be detected as an 'elbow' in a plot of W_k as a function of k. The gap statistic, *GS*, formalizes the detection of this elbow, and to objectively distinguish real changes in W_k from noise decrements observed when coherent clusters begin to split, it compares the W_k plot computed from the original data with a W^*_k plot based on null reference simulated data (uniformly random data matrixes with no cluster structure). The use of null simulated data also allows the computing of a bootstrapped standard error for the gap statistic. In the present study, null reference data were obtained through 100 random replications, twice the minimum number of 50 replications suggested by Tibshirani, Walther, and Hastie (2001).

Results

The hierarchical cluster analysis of the cognitive measures resulted in the dendrogram depicted in Fig. 1, in which two well-separated clusters can be identified. The gap statistic (*GS*) is maximized for a two-cluster solution (the value for *GS* and its bootstrapped standard error were $GS = 0.30 \pm 0.07$ for the one-cluster solution, $GS = 0.39 \pm 0.07$ for the two-cluster solution, $GS = 0.36 \pm 0.07$

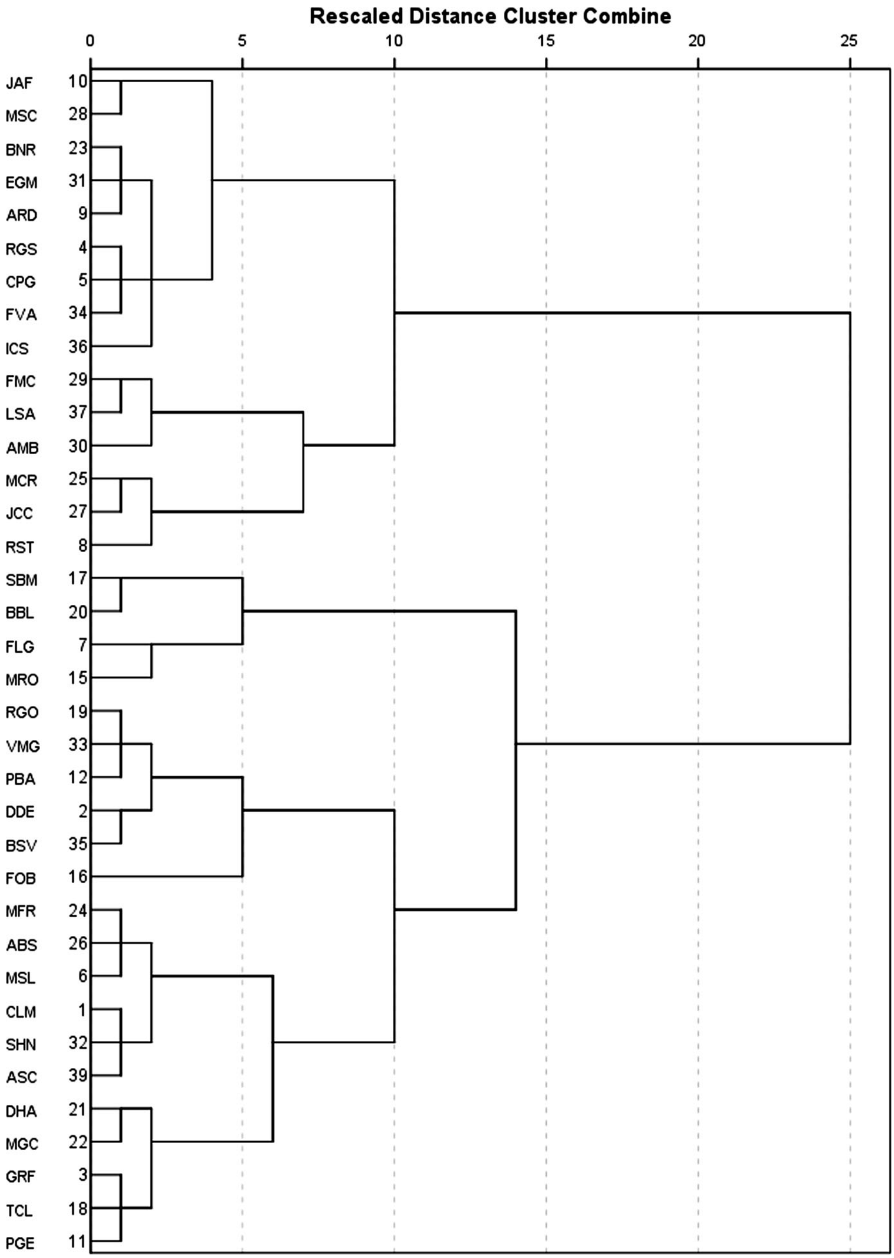

Fig. 1 Dendrogram based on Ward's algorithm illustrating the two clusters solution for the sample of 36 Portuguese children with dyslexia

for the three-cluster solution, $GS = 0.34 \pm 0.08$ for the four-cluster solution, $GS = 0.36 \pm 0.08$ for the five-cluster solution and $GS = 0.37 \pm 0.08$ for the six-cluster solution), confirming the visual inspection of the dendrogram.

Table 2 Mean profiles cognitive and alphabetic measures (z scores) by cluster (mean ± SD)

Tasks	Cluster 1 ($n = 21$)	Cluster 2 ($n = 15$)	Cohen's d	Mann–Whitney U test (p value)
PD	−1.17 ± 0.571	−1.60 ± 0.500	0.803	.023
RAN	−1.32 ± 0.736	−0.44 ± 0.908	−1.070	.004
DS	−0.54 ± 0.572	−1.78 ± 0.586	2.141	.000
VOC	−0.14 ± 0.910	−0.69 ± 0.672	0.665	.062
NVIQ	0.60 ± 0.778	0.20 ± 0.584	0.568	.096
RWR				
RWR (speed)	−2.00 ± 0.400	−1.81 ± 0.239	−0.554	.042
RWR (accuracy)	−2.15 ± 0.648	−1.62 ± 1.279	−0.556	.357
PWR				
PWR (speed)	−1.89 ± 0.503	−1.64 ± 0.444	−0.509	.077
PWR (accuracy)	−1.46 ± 0.859	−1.34 ± 0.982	−0.136	.825
SP	−1.41 ± 0.622	−1.62 ± 0.771	0.304	.340

Real word reading (RWR-speed: correct items/second; RWR-accuracy: percentage of correct items), pseudoword reading (PWR-speed: correct items/second; PWR-accuracy: percentage of correct items), spelling (SP: percentage of accuracy), phoneme deletion (PD: percentage of accuracy), rapid automatized naming (RAN: correct items/second), digit span (DS: number of repeated series), vocabulary (VOC), and non verbal intelligence (NVIQ: percentage of accuracy)

Cluster 1 contained 21 children (58.3 %), and Cluster 2 contained 15 children (41.7 %). There were no differences between clusters related to age ($p = .63$), sex ($p = .64$), or grade ($p = .41$). The cognitive profile of each cluster, based on the participants' performance on the five assessed variables, can be observed in Table 2.

Compared with Cluster 2, the participants from Cluster 1 presented better performance on phoneme deletion, digit span, vocabulary and nonverbal IQ but not on rapid automatized naming. In contrast, participants from Cluster 2 underperformed on the variables most related to phonological processing (phoneme deletion and digit span), whereas their rapid automatized naming and vocabulary scores came close to the normal range. Although the differences between clusters had a moderate-to-large magnitude (Cohen's $d > 0.5$), only the differences for phoneme deletion, rapid automatized naming and digit span reached statistical significance (Mann–Whitney U test). To better characterize the two profiles, we also calculated the mean differences for alphabetic measures (not included in the clustering analysis). In general, Cluster 1 showed a worse performance on all measures except the spelling task. However, the only significant difference between clusters was on word reading speed ($p = .04$) (see Figure 2 in the online supplementary materials).

Discussion and Conclusions

Several studies have highlighted the existence of heterogeneity at the level of symptoms in dyslexia (see, for example, Menghini et al., 2010; Pennington et al., 2012). It has also been suggested that the orthography the child is learning contributes to this heterogeneity (e.g., Bergmann & Wimmer, 2008). Within this context, this study aimed to characterize the distinct profiles present in Portuguese children with dyslexia who were assessed on various cognitive abilities related to reading. The cluster analysis revealed two different cognitive profiles, although there were almost no significant differences between groups regarding alphabetic outcomes (word and pseudoword reading accuracy and spelling). When compared with Cluster 2, participants belonging to Cluster 1 showed better results on all cognitive measures (although the only significant differences were for phoneme deletion and digit span), except for a significant performance decrease in rapid automatized naming. Therefore, in the Cluster 1 profile, the poor performances on phoneme deletion and rapid automatized naming stand out compared with the remaining cognitive variables (short-term verbal memory, vocabulary and nonverbal IQ), which approach the normal range. In contrast, the Cluster 2 participants underperformed on the variables most related to phonological processing (phoneme deletion and digit span), whereas rapid automatized naming, vocabulary scores and nonverbal IQ came close to the normal range. Overall, the clusters identified in our sample of children with dyslexia are in partial agreement with the double-deficit hypothesis (Wolf & Bowers, 1999). Cluster 1 (58 % of the participants) showed weak phonological processing and rapid naming (corresponding to the double-deficit subtype), whereas Cluster 2 (42 %) had significantly impaired phonological processing but normal rapid-naming skills (corresponding to the single-phonological deficit subtype). The percentage of children in Cluster 1 was similar to that found in previous studies for the double-deficit subtype (e.g., Araújo et al., 2010; King, Giess, & Lombardino, 2007; Lovett, Steinbach, & Frijters, 2000; O'Brien, Wolf, & Lovett, 2012).

Another argument that supports considering Cluster 1 a double-deficit profile follows from the fact that this subgroup achieved systematically worse reading scores than Cluster 2. In fact, the double-deficit hypothesis assumes that there is a cumulative effect of rapid naming and phonological difficulties in this subtype (Wolf & Bowers, 1999), which leaves these subjects without an effective compensatory strategy for reading development (i.e., grapheme–phoneme decoding for phonological measures or orthographic identification for rapid-naming measures). However, despite the observed moderate size differences between clusters in alphabetic outcomes (Cohen's $d \approx 0.5$), only the word reading speed difference reached significance. For this measure, Cluster 2 outperformed Cluster 1, an expected result considering the well-established relation between fluent reading and rapid automatized naming (preserved in Cluster 2). Pseudoword reading speed shows a similar result, although only marginally significant ($p = .077$). The fact that different cognitive profiles do not clearly translate into reliably different alphabetic outcomes is a relevant issue in the context of dyslexia profiling, but the present results should be taken with some caution due to a potential lack of power in our sample for detecting moderate differences as statistically significant (group sizes provide only 30 % power to detect medium effect sizes such as Cohen's $d = 0.5$).

Our results did not confirm a third subtype predicted by the double-deficit hypothesis, with impaired naming performance and intact phonological skills (the single naming-deficit). One possible reason for this finding could be related to the

Portuguese orthography, which, according to Seymour et al. (2003), has a medium opacity. A medium or non-consistent orthography, in which letters can have multiple pronunciations, places heavier demands on phonological decoding during reading acquisition, particularly for children struggling with reading. Thus, phonological difficulties represent an important risk factor for developing reading problems in Portuguese children, at least during the first years of reading development. It may be the case that the single-naming deficit will have a relevant negative impact only on later phases, when fluency problems interfere more seriously with reading comprehension. Within this context, the most salient symptom that highlights the presence of reading deficits is the existence of phonological decoding problems. Consequently, a child who masters the mapping from orthography to phonology while exhibiting a low reading rate is less likely to be identified as having reading problems and will therefore have less chance of being included in a sample like ours, where participants were preselected by experts. Although this argument may explain the absence of a cluster characterized by a single-naming deficit, in our opinion, clarification of this issue will require larger samples with a wider range of ages as well as studies with longitudinal design. Nevertheless, Araújo et al. (2010), using theory-driven criteria to identify double-deficit subtypes, found four children with a single-naming deficit (18 %) in a sample of twenty-two Portuguese children with dyslexia. On closer examination of the individual scores in our sample, we identified two participants who fulfilled the criteria for a single-naming deficit (rapid automatized naming score 1 SD or more below the average of the participants' school grade and normal phoneme deletion score). With only two cases, however, which differed on the remaining variables (non-verbal IQ and vocabulary), the cluster algorithm did not group the cases together, preventing the formation of a single-naming-deficit cluster.

In addition to rapid automatized naming and phoneme deletion differences, the two clusters observed in the sample differed on digit span performance. Cluster 2 showed an unexpectedly low performance compared with Cluster 1, which scored close to normal (Cohen's $d = 2.1$). This dissociation between groups on verbal short-term memory might explain why Cluster 1 was significantly better at phoneme deletion than Cluster 2 (Cohen's $d = 0.80$), although both groups presented a phoneme deletion deficit. The better performance of Cluster 1 on phoneme deletion seems to be explained by results from previous studies suggesting that phonological processing is highly dependent on efficient operation on phonological codes in memory (Beneventi, Tønnessen, Ersland, & Hugdahl, 2010; Gathercole, & Baddeley, 1993). The role of verbal short-term memory in dyslexia and its relation to the phonological deficit was recently readdressed by Ramus and Szenkovits (2008) within the context of the phonological access hypothesis. According to the authors, the phonological representations of individuals with dyslexia may be intact, and their poor performance on phonological tasks might instead reflect a limitation in the phonological access mechanisms. Such mechanisms comprise storing, accessing and retrieving phonological representations (as required for verbal short-term memory tasks), conscious access to phonological representations (as required for phoneme deletion tasks) or multiple fast access to phonological representations (as required for rapid naming tasks). Our results seem to be consistent with the framework proposed by Ramus and Szenkovits (2008), which suggests that multiple mechanisms may underlie the phonological deficit in dyslexia. While Cluster 2's phonological problems may be directly attributed to a deficit on verbal short-term memory, Cluster 1 showed a phonological deficit despite having reasonably good verbal short-term memory skills. This dissociation might be explained by the specific demands of the phoneme deletion task, which, in addition to involving the verbal short-term memory system, requires a special type of access, namely, conscious access to the phonological representations (Ramus & Szenkovits, 2008). However, this conscious access to phonology seems insufficient in explaining the participants' reading problems because both clusters show additional deficits, including verbal short-term memory (Cluster 2) and rapid naming (Cluster 1). Thus, the cognitive profile of our clusters seems to expose the complex nature of the phonological processing problems in dyslexia, which may result from multiple underlying causes.

Overall, our results are consistent with the view that dyslexia is a heterogeneous phenomenon, here characterized in terms of different cognitive profiles. In general, children of both clusters revealed phonological processing deficits, although these deficits were accompanied by other impairments that may have contributed to their reading difficulties. Thus, the present work does not clearly support either a more stringent version of the single-phonological-deficit perspective on dyslexia (e.g., Ramus et al., 2003) or a pure version of the double-deficit hypothesis (Wolf & Bowers, 1999).

Therefore, our approach seems to cluster children with single as well as multiple deficits for whom phonology could or could not have been the core deficit, emphasizing the multiple-deficit nature of dyslexia (Pennington, 2006). Recently, Pennington et al. (2012), using a regression-based approach to test single- and multiple-deficit models for the prediction and diagnosis of dyslexia, found roughly equal proportions of participants who fit each model, and they concluded that a hybrid model provided the best overall fit with the data. If a hybrid model is better suited to explaining the cognitive profiles of Portuguese children with dyslexia, as our data suggest, this needs to be confirmed by future studies.

The present research used a quantitative bottom-up approach to identify cognitive profiles, namely, an agglomerative hierarchical cluster analysis. Some authors have highlighted the complexity of clustering algorithms (Blashfield, 1976), but this methodological option offers several advantages. First, hierarchical cluster analysis is a data-driven approach and is therefore not influenced by previous assumptions concerning the number or cognitive nature of the subtypes. Second, the use of the gap statistic allowed for optimizing the choice of the number of clusters, minimizing the risk of detecting spurious clusters and increasing the reliability of the analysis (Crews & D'Amato, 2009; Hair et al., 1992; Tibshirani et al., 2001). In addition, reading and spelling measures did not enter the clustering algorithm, thus minimizing the risk of obtaining simple severity profiles and maximizing the identification of qualitatively dissimilar profiles. Nonetheless, the selection of the variables to include in the clustering algorithm can bias the final cluster solution. In our case, the inclusion of phonological awareness and rapid automatized naming measures may have led to the identification of profiles that distinguished themselves precisely on those variables, possibly favoring the identification of profiles coherent with the double-deficit hypothesis. This caveat does not invalidate the existence of such profiles among the Portuguese children with dyslexia, but it also does not rule out that other relevant cognitive profiles may exist in this population.

In sum, the present study confirms the assumption of several previous investigations that children with dyslexia have heterogeneous cognitive characteristics and, consequently, distinct cognitive impairments, despite a phonological processing deficit (assessed here by a phoneme deletion task) being one of the most prominent features. Due to this heterogeneity, prevention and remediation programs should be specifically targeted toward children's particular deficit patterns. These programs might be more effective in terms of outcome if they take the diversity of performance deficits into account.

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