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Investigating the double-deficit hypothesis of developmental dyslexia in an orthography of intermediate depth

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Abstract
The present study aimed to investigate the double-deficit hypothesis (DDH) in an orthography of intermediate depth. Eighty-five European Portuguese-speaking children with developmental dyslexia, aged 7 to 12, were tested on measures of phonological awareness (PA), naming speed (NS), reading, and spelling. The results indicated that PA and NS were not significantly correlated, and that NS predicts reading fluency (but not reading accuracy and spelling) beyond what is accounted for by PA. Although the majority of the children with developmental dyslexia have double deficit (62.4%), some children have a single phonological deficit (24.7%) or a single NS deficit (8.2%). Children with a double deficit were not more impaired in reading fluency, reading accuracy, and spelling than both single-deficit subtypes. In conclusion, the findings of the present study are partially consistent with the DDH and provide evidence for the multifactorial model of developmental dyslexia. Implications of the DDH for an orthography of intermediate depth are emphasized.

Keywords Developmental dyslexia · Double-deficit hypothesis · Naming speed · Phonological awareness

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Introduction

Developmental dyslexia (DD) can be conceptualized as a specific learning disorder that is neurobiological in origin and characterized by problems with accurate or fluent word recognition, poor decoding, and poor spelling abilities that are not better accounted for by intellectual disabilities, sensory impairments, or inadequate educational instruction (American Psychiatric Association, 2013). Because reading involves multiple linguistic, visual, and attentional processes, it seems unlikely that a single cognitive deficit is sufficient to account for the complexity and heterogeneity of DD (Norton & Wolf, 2012). Phonological awareness (PA) and naming speed (NS) have been the most studied and the most relevant endophenotypes of DD in transparent, intermediate, and opaque orthographies (Catts, McIlraith, Bridges, & Nielsen, 2017; Moura, Moreno, Pereira, & Simões, 2015; Tobia & Marzocchi, 2014).

PA refers to the ability to perceive and manipulate the sounds of spoken words, which is typically measured by tasks that require the ability to discriminate and manipulate syllables or phonemes in words. Children must first become aware of the phonological structure of words, so that they can map those units of sound (i.e., phonemes) onto their corresponding printed letters. Thus, PA is likely to be the strongest predictor of individual differences in reading development (Ziegler et al., 2010) and the most accurate cognitive measure to discriminate between children with DD and typically developing children (Landerl et al., 2013; Moura et al., 2017).

Although strong converging evidence supports the importance of PA for reading development, deficits in PA alone do not account for all cases of DD. For example, recent longitudinal studies have found that the predictive power of PA for reading may have been overestimated (Landerl et al., 2019) and that single PA deficits in kindergarten did not necessarily predict the presence of a DD diagnosis in the second grade (Catts et al., 2017). Thus, NS (which is typically measured by “rapid automatized naming” (RAN) tasks) is another core deficit and perhaps the most universal predictor of reading fluency (Araújo, Reis, Petersson, & Faísca, 2015; Kirby, Georgiou, Martinussen, & Parrila, 2010; Norton & Wolf, 2012). Reading and RAN tasks involve closely related cognitive processes (for a review, see Norton & Wolf, 2012; Wolf & Bowers, 1999), with the RAN tasks reflecting the automaticity of processes which are important for reading. Recently, Georgiou, Ghazyani, and Parrila (2018) found that NS deficits in individuals with DD were due to subtle impairments in lexical access, serial processing, and slower articulation.

Some controversy still exists regarding whether NS should be a subskill related to phonological processing or an independent process (for a review, see Kirby et al., 2010; Norton & Wolf, 2012; Vukovic & Siegel, 2006). The phonological processing deficit hypothesis included NS as a component of a more general construct of phonological processing because it considers that NS depends on the retrieval of phonological codes (Wagner & Torgesen, 1987). On the other hand, the double-deficit hypothesis (DDH) postulates that NS is an independent core deficit in DD that can cause reading difficulties in addition to or in the absence of the phonological processing deficits. That is, the PA and NS deficits are separable sources of reading difficulties, and their combined presence leads to profound reading impairment (Wolf & Bowers, 1999; Wolf, Bowers, & Biddle, 2000).

Studies that have examined the DDH have yielded mixed findings, which is not surprising because the DDH was proposed not to fully explain all reading difficulties, but rather to move the field forward in understanding the heterogeneity of reading and the multiple etiologies of DD (Wolf & Bowers, 1999). This variability may also be attributed to several aspects. First, the large number of studies investigating the DDH used children with DD (Lovett, Steinbach, & Frijters, 2000; Vaessen, Gerretsen, & Blomert, 2009), but others included mixed clinical samples (Heikkilä, Torppa, Aro, Närhi, & Ahonen, 2016) or typically developing children (Papadopoulos, Georgiou, & Kendeou, 2009). Second, different inclusion criteria have been used to diagnose children with DD (e.g., reading accuracy and/or fluency score below the 7th, 15th, or 25th percentile) (cf. Araújo, Pacheco, Faísca, Petersson, & Reis, 2010; Jiménez et al., 2008). Third, studies have used different tasks to measure PA (e.g., deletion, substitution, blending, and segmentation tasks) and NS (e.g., objects, colors, numbers, letters). Fourth, a variety of cutoff scores have been used to classify the DDH subtypes (see Table 1). Fifth, the level of orthographic consistency of the languages in which the DDH was examined is another source of variability in children's reading performance.

The four main assumptions of the double-deficit hypothesis

In their seminal work, Wolf and Bowers (1999) proposed four main assumptions for the DDH. The first main assumption postulates that *PA and NS are not strongly correlated*. In a comprehensive meta-analysis of 35 studies on the relationship between PA and NS, Swanson, Trainin, Necoechea, and Hammill (2003) found an overall correlation coefficient of $r = .38$. More recent studies have reported low to moderate correlation coefficients (Heikkilä et al., 2016; Nelson, 2015; Torppa et al., 2013). Inconsistent results related to the independence of PA and NS have been obtained from factor analytic studies. Nelson (2015) and Swanson et al. (2003) observed that PA and NS tasks load on separate factors, whereas Ackerman, Holloway, Youngdahl, and Dykman (2001) found that they operate on the same factor.

Neuroimaging studies have found that PA and NS may have different neural substrates, which provides additional support for the independence of NS (Cummine, Chouinard, Szepesvari, & Georgiou, 2015; He et al., 2013). For example, Norton et al. (2014) evaluated the DDH using fMRI and found a dissociation between the brain regions that were sensitive to PA (left inferior frontal and inferior parietal regions) and NS (right cerebellar lobule VI).

The second main assumption of the DDH claims that *NS predicts reading performance beyond what is accounted for by PA*. Indeed, cross-sectional and longitudinal studies have found that PA and NS contribute differently to reading. PA has primarily been linked to reading accuracy and spelling, whereas NS has especially been linked to reading fluency (Caravolas, Lervåg, Defior, Málková, & Hulme, 2013; Landerl & Wimmer, 2008; Moll et al., 2014; Moura et al., 2015). Although the majority of the studies with normal and/or dyslexic readers have indicated that NS is a significant predictor and explains unique variance in reading performance beyond that which is explained by PA, some studies have found that its contribution is modest when compared with the contribution of PA (Ackerman et al., 2001; Ziegler et al., 2010).

In addition, cross-linguistic studies have reported that the contribution of PA and NS to reading performance is modulated by the transparency of the orthography (Caravolas et al., 2013; Moll et al., 2014; Vaessen et al., 2010). For example, Ziegler et al. (2010) observed that PA was the main factor associated with reading accuracy and reading fluency across the five alphabetic languages studied (Finnish, Hungarian, Dutch, Portuguese, and French), but its contribution was stronger in less transparent orthographies. Moll et al. (2014) found that NS

Table 1 Percentages of DDH subtypes

	Language	*N*	Age (years)	Cutoff score	Percentages of DDH Subtypes			
					Single PH deficit	Single NS deficit	Double deficit	No deficit
Manis et al. (2000)	English	$85^{(TR)}$	7–8	< 25th perc.	15.3	16.5	9.4	58.8
Vukovic, Wilson, and Nash (2004)	English	$25^{(DD)}$	19–25	≤ − 1 *SD*	16.0	44.0	20.0	20.0
Cronin (2013)	English	$122^{(TR)}$	5	< 33rd perc.	16.4	16.4	16.4	50.8
Norton et al. (2014)	English	$90^{(TR)}$	8–12	< 25th perc.	30.0	11.1	15.6	43.3
Nelson (2015)	English	$149^{(DD)}$	16–24	≤ − 1 *SD*	22.8	24.2	34.2	18.8
Araújo et al. (2010)	Portuguese	$22^{(DD)}$	9.5 ± 1.3	≤ − 1 *SD*	18.2	18.2	50.0	13.6
Vaessen et al. (2009)	Dutch	$162^{(DD)}$	6–12	≤ − 1 *SD*	31.5	10.5	40.7	17.3
Wimmer et al. (2000)	German	$397^{(TR)}$	5–8	< − 1 *SD*	16.6	16.1	6.3	61.0
Escribano (2007)	Spanish	$29^{(DD+TR)}$	8–13	< 25th perc.	34.5	0	34.5	31.0
Jiménez et al. (2008)	Spanish	$89^{(DD)}$	7–12	≤ − 1 *SD*	10.1	5.6	21.4	62.9
Papadopoulos et al. (2009)	Greek	$242^{(TR)}$	6–7	< 20th perc.	13.6	13.6	7.0	65.7
Torppa et al. (2012)	Finnish	$194^{(DD+TR)}$	6.5	< 20th perc.	13.9	14.9	12.9	58.3
Heikkilä et al. (2016)	Finnish	$205^{(DD+CC)}$	8–13	< − 1 *SD*	18.1	23.4	43.9	14.6

DDH double-deficit hypothesis, *NS* naming speed, *PH* phonological, *SD* standard deviation, *Perc* percentile, *TR* typical readers with a wide range of reading abilities, *DD* individuals with developmental dyslexia, *DD* + *TR* individuals with developmental dyslexia and typical readers, *DD* + *CC* individuals with developmental dyslexia and other clinical conditions was the most relevant predictor of reading fluency, whereas PA accounted for more unique variance in reading accuracy. This predictive pattern was comparable across the five orthographies studied (Finnish, Hungarian, German, French, and English), but it tends to be stronger in English. Recently, Landerl et al. (2019) also found that the NS is a universal predictor of reading fluency in five orthographies varying in consistency (Greek, German, Dutch, French, and English). The relationship between PA and reading appears to be complex, interactive, and dependent on the orthographic complexity. Interestingly, studies from non-alphabetic languages (e.g., Chinese) reported similar findings, suggesting that PA and NS are universal correlates of word reading and core deficits in DD (Araújo et al., 2015; Brunswick, McDougall, & Davies, 2010; Georgiou, Parrila, & Liao, 2008; Song, Georgiou, Su, & Hua, 2016).

These predictive patterns are also observed for spelling with PA being the most significant predictor in almost all alphabetic languages (Caravolas, Volín, & Hulme, 2005; Furnes & Samuelsson, 2010, 2011; Landerl & Wimmer, 2008; Moll et al., 2014; Plaza & Cohen, 2004). Moll et al. (2014) found that PA accounted for significant amounts of unique variance in four of the five orthographies studied (English = 8.9%, German = 8.5%, Hungarian = 4.1%, and Finnish = 5.1%; the exception was French 2.2%), whereas NS made a significant contribution to variance in spelling only for the English orthography (16.7%; the highest contribution). Furnes and Samuelsson (2010) observed that PA assessed in kindergarten was the most significant predictor of spelling difficulties at grades 1 and 2 in transparent (Norwegian and Swedish) and opaque (English) orthographies. Albuquerque (2012) also found that PA was the most relevant predictor of spelling in European Portuguese-speaking children (≥ 30% of unique variance), with NS having a modest influence (≤ 8% of unique variance).

Longitudinal studies with children speaking different languages spanning a large range of orthographic complexities have also demonstrated that the contribution of PA and NS is modulated by age, with NS being a better long-term predictor of reading performance and PA being most strongly related to the early stages of reading development (Furnes & Samuelsson, 2010; Landerl & Wimmer, 2008). Catts et al. (2017) reported that children with a PA deficit in kindergarten were five times more likely to have DD in the second grade than children without a PA deficit (47.6% vs. 8.5%, respectively). They also found that children with a single PA deficit in kindergarten showed a higher probability of having DD in the second grade than children with a single NS deficit or a single oral language deficit (36.8% vs. 11.8% vs. 9.1%, respectively), but children with multiple deficits were more likely to have DD than those with single deficits (e.g., 48.1% in children with PA and oral language deficits, and 72.7% in children with PA, NS, and oral language deficits). These results showed that a deficit in PA is a core deficit in DD and that difficulties in NS and oral language may serve as additive factors that increase the probability of DD. Indeed, some studies have found that oral language is another relevant predictor of reading beyond what is accounted for by PA and NS, and that oral language problems are often present in children with DD (Catts et al., 2017; Moll, Loff, & Snowling, 2013; Snowling, Gallagher, & Frith, 2003).

The third main assumption of the DDH asserts the *existence of a subgroup of individuals with DD that has NS deficits in the absence of phonological deficits* (*single NS deficit subtype*). Table 1 shows the percentages of DDH subtypes obtained from 13 studies of alphabetic languages with different levels of orthographic consistency. Single-deficit subtypes (PA or NS) occur in less than 20% in the majority of the studies reported on Table 1, irrespective of the cutoff score used. The double-deficit subtype was the most prevalent in the majority of the studies that investigated the DDH in a sample of individuals with DD. Inversely, the no deficit subtype was the most prevalent among the studies that analyzed typical readers with a wide range of reading abilities. Although NS might be a more reliable marker of DD than PA in transparent orthographies (Kirby et al., 2010), DDH studies from transparent orthographies did not find a higher percentage of individuals with single NS deficits compared with studies from opaque orthographies.

More recently, Norton et al. (2014) found neurofunctional evidence for the DDH subtypes. Through an fMRI study, the authors observed that the double-deficit group showed less activation in the frontoparietal reading network as compared with the single phonological deficit group, who in turn showed less activation than the no deficit group. On the other hand, the double-deficit group showed less cerebellar activation as compared with the single NS deficit group, who in turn showed less activation than the no deficit group.

The fourth main assumption postulates that *children with a double deficit are expected to be more impaired in reading than those with a single deficit* because both deficits contribute independently and additively to reading problems. Papadopoulos et al. (2009) observed that first-graders with a double deficit exhibited greater impairment in reading accuracy and fluency as compared with the single-deficit groups. Other studies have not found support for the claim that the double-deficit subtype shows more reading impairments than the single-deficit subtypes (Nelson, 2015; Vaessen et al., 2009). With respect to spelling, DDH subtypes did not show significant differences (Torppa, Georgiou, Salmi, Eklund, & Lyytinen, 2012; Vaessen et al., 2009; Wimmer, Mayringer, & Landerl, 2000), or the single NS deficit subtype is less impaired than the double deficit or the single phonological deficit subtypes (Heikkilä et al., 2016; Nelson, 2015).

An important issue with implications for this fourth main assumption of the DDH is the possibility that the greater severity of reading impairment found in children with a double deficit could be due in part to a statistical artifact. Schatschneider, Carlson, Francis, Foorman, and Fletcher (2002) demonstrated that categorizing children into four groups based on two correlated continuous variables can lead to distortions in the distributions of mean scores for each group. Thus, children with a double deficit may have more severe reading impairments than the single-deficit groups due to their experiencing more severe PA or NS deficits. Indeed, when the double and single-deficit groups were matched on PA and NS, many of the differences in reading disappeared (Nelson, 2015).

Another hypothesis related to this fourth assumption is that DDH subtypes might reveal a different reading performance pattern given that PA and NS predict different components of the reading process. Many authors have argued that PA is more associated with decoding skills (such as the reading of pseudowords and unfamiliar words), whereas NS is more strongly related to tasks that rely on the recognition of word-specific orthographic patterns (such as the reading of irregular words and the quick recognition of familiar words) (for a review, see Araújo et al., 2015; Kirby et al., 2010). Consistent with this hypothesis, Nelson (2015) found that individuals with a single NS deficit outperformed those with a single PA deficit in pseudoword reading and spelling measures, with a reverse pattern observed for reading fluency.

The present study

This study aimed to investigate the four main assumptions of the DDH in European Portuguese-speaking children with DD. Based on the existing literature, we hypothesized that (i) PA and NS would not be strongly correlated (Heikkilä et al., 2016; Swanson et al., 2003);

(ii) PA and NS would contribute differently to reading performance, the former more related to reading accuracy and spelling, and the latter more associated to reading fluency (Moll et al., 2014; Moura et al., 2015); (iii) the double-deficit group would be the most prevalent DDH subtype (Araújo et al., 2010; Nelson, 2015); (iv) the double-deficit group would not be more impaired in reading and spelling than single-deficit groups (Vaessen et al., 2009).

The present study extends previous research by examining the DDH with European Portuguese orthography, which is placed at the middle of the transparency continuum of orthographies (Seymour, Aro, & Erskine, 2003). The large body of research about DDH has been conducted almost exclusively in transparent and opaque orthographies. The level of orthographic consistency is a key factor determining the rate of reading acquisition across different languages (Seymour et al., 2003; Ziegler et al., 2010) as it may influence how DD is manifested (Brunswick et al., 2010; Landerl et al., 2013), as well as the likelihood of prevalence of the DDH subtypes and the reading and spelling impairments observed in children with a single or double deficit. Thus, extending the study of DDH to an orthography of intermediate depth helps researchers understand which factors are universal and which are orthography-specific.

Compared to previous DDH studies, this study includes spelling measures. The number of DDH studies that included spelling measures is limited (some exceptions: Heikkilä et al., 2016; Torppa et al., 2012). This is surprising because spelling difficulties are another common manifestation of DD and because PA and NS are two of the strongest predictors of spelling ability (Albuquerque, 2012; Moll et al., 2014).

Although Portuguese is little researched, it is commonly listed as the sixth most spoken language in the world, with more than 200 million native speakers (Lewis, Simons, & Fennig, 2015). Several orthographic and phonemic features converge to characterize European Portuguese orthography as intermediate in depth. For example, the use of grapheme-phoneme correspondence rules is particularly difficult (e.g., there are five vowel letters for 18 vocalic phonemes) (Sucena, Castro, & Seymour, 2009). Seymour et al. (2003) found that reading accuracy in most transparent orthographies generally reaches a ceiling effect at the end of the first grade, which contrasts with the reading accuracy found in orthographies of intermediate depth (e.g., Portuguese children read correctly approximately 74% of words and 77% of non-words) or in more opaque orthographies.

Method

Participants

The participants were 85 Portuguese children with DD (55 males and 30 females) aged 7 to 12 ($M = 9.41$, $SD = 1.43$; 7 years $n = 5$, 8 years $n = 18$, 9 years $n = 30$, 10 years $n = 13$, 11 years $n = 7$, 12 years $n = 12$) in the second to sixth grades at school (second grade $n = 8$, 3rd grade $n = 21$, fourth grade $n = 32$, fifth grade $n = 10$, sixth grade $n = 14$). The father's level of education (i.e., years of schooling completed) was 9.34 ($SD = 4.23$), and the mother's was 9.90 ($SD = 4.07$). Twenty-two percent of children had experienced school retention, 35% were included in special education system, and 30% had relatives with reading difficulties.

The participants were either recruited via their contact with psychologists, special education teachers, physicians and speech therapists or were referred to the Community Service Centre of the University of Coimbra by parents given their persistent and severe reading difficulties.

The diagnosis of DD was confirmed by psychologists qualified in specific learning disorders after completing a comprehensive assessment that included a clinical interview, an academic history of reading and spelling difficulties, a review of previous and current school reports and portfolios, and a psychoeducational assessment. Only children with DD who met the following criteria were included: (i) Wechsler Intelligence Scale for Children—Third Edition (WISC-III) Full Scale IQ (FSIQ) ≥ 85; (ii) a score less than or equal to the 15th percentile simultaneously in a reading fluency task and in a reading accuracy task from a Portuguese standardized reading test ("O Rei"; Carvalho & Pereira, 2009); (iii) European Portuguese as the mother tongue; (iv) absence of any visual, hearing, or motor handicap; and (v) exclusion of a language impairment, emotional disturbance, dyscalculia, attention-deficit/hyperactivity disorder, disruptive impulse-control and conduct disorders, neurological impairment, or other psychiatric disorders.

Measures

Intellectual ability Because intellectual ability has been shown to be positively correlated with both PA and NS, its inclusion as a covariate in studies of the impact of PA and NS on reading and spelling has been recommended (Nelson, 2015; Wolf et al., 2002). Thus, the Portuguese version of the WISC-III was administered, and the FSIQ score ($M = 100$; $SD = 15$) was analyzed and used as a covariate in the correlation, regression, and inferential analyses.

Phonological awareness The Phonological Awareness subtest of the Coimbra Neuropsychological Assessment Battery (BANC; Simões et al., 2016) was used to assess PA. It comprised two tasks. In the Deletion task, the child was asked to delete a particular phoneme in familiar words (e.g., say *sopa* [sopɐ] without the *se* [s]). In the Substitution task, the child was asked to replace one or more phonemes for other phoneme(s) in familiar words (e.g., say *judo* [udu] but replace the *je* [ ] to *xe* [ʃ]). The raw score is the sum of the correct responses of the Deletion and Substitution tasks. The reliability of the BANC normative sample for the Deletion and Substitution tasks had a Cronbach's $\alpha = 0.91$ and 0.90, respectively.

Naming speed The RAN-Numbers subtest of the BANC was used to examine NS. The child was asked to name as quickly as possible 50 visual stimuli (numbers 2, 4, 6, 7, and 9) randomly displayed on a card in a 10 × 5 matrix. The raw score represents the amount of time (in seconds) required to complete the task. The reliability of the BANC normative sample for the RAN subtest was obtained through test-retest ($r = .78$).

Reading fluency "O Rei" ("The King"; Carvalho & Pereira, 2009) is a 3-min reading test based on a traditional Portuguese tale that measures the reading fluency and accuracy of children from the first to sixth grade. The raw scores of reading fluency is the number of correctly read words in 1 min. The test-retest from the normative sample was $r = .94$ for reading fluency.

Reading accuracy and spelling To assess the reading accuracy and spelling of individual words, we used the Oral Reading subtest from the Portuguese version of the Psycholinguistic Assessment of Language (PAL-OR; Caplan, 1992). The PAL-OR comprises 146 words (48 regular, 47 irregular and 51 pseudowords). Based on previous studies that used the PAL-OR with children with DD, we selected 40 words: 16 regular (e.g., *sardinha* [sɐˈδiɐ], *rusga*

[ˈʀuɣ ɐ]), 16 irregular (e.g., *fluxo* [ˈfluksu], *exotismo* [ezuˈti mu]), and eight pseudowords (e.g., *lempo* [ˈlẽpu], *glepal* [ɣlɛˈpa]). The PAL-OR was used in both the reading and spelling tasks through a counterbalanced design to reduce the chances of the order of the tasks influence the results. The reading and spelling tasks were separated by an interval of 10 to 15 days. The raw scores of reading accuracy and spelling represent the percentage of words correctly read and spelled, respectively. The reliability (Cronbach's α) of the PAL-OR was 0.75. In the spelling task, 23 children were dropped out due to missing data (11 males and 12 females).

Procedure

Voluntary participation was required of all participants, and the objectives of the study were fully explained to them. Informed parental consent along with child assent was obtained for each evaluation. This research was approved by the Portuguese Data Protection Authority. Each child completed two individual sessions (separated by an interval of 10 to 15 days) which were conducted in either a clinic or school setting, occurring on a weekday. All measures were administered by the authors (psychologists with extensive experience in the psychological assessment of children with reading difficulties and neurodevelopmental disorders) in a fixed order. No incentives were offered in exchange for participation.

Statistical analyses

The statistical analyses were performed using IBM SPSS Statistics 25. Previous to any of these analyses, each dependent variable was assessed for outliers, normal distribution, and homogeneity of variances. No outliers ($SD \geq 3$) were found in the data. All dependent variables revealed skewness and kurtosis values lower than 1, suggesting normal distribution of the data. The Levene's test of homogeneity of variances was not significant ($p > .05$) in all univariate analysis of covariance (ANCOVA).

To control for the influence of the intellectual ability and age in the outcome measures (PA, NS, reading, and spelling), the WISC-III FSIQ and age were entered as covariates in the correlation, regression, and inferential analyses. A partial correlation and a principal components factor analysis with oblique rotation were used to analyze the relation between the measures. The oblique rotation (direct oblimin) was used because it allows factors to be correlated (Costello & Osborne, 2005; Fabrigar, Wegener, MacCallum, & Strahan, 1999; Henson & Roberts, 2006). To determine the predictive value of PA and NS for reading and spelling abilities, hierarchical linear regression analyses were conducted. The total variance (R^2) of the regression model, the standardized regression coefficient (β), the t test (t), and the squared part correlation (pr^2 represents the unique variance of each predictor when the overlapping linear effects of all other predictive variables were statistically removed) for each predictor were calculated. An ANCOVA was also performed for each measure with WISC-III FSIQ and age as covariates. If the initial ANCOVA revealed a significant main effect of DDH subtypes, planned post hoc comparisons were conducted among the groups (single phonological deficit, single NS deficit, and double deficit) with a Bonferroni adjustment for multiple comparisons. Partial eta-squared (η^2_p) was calculated to determine the effect size of the difference between groups.

Results

Correlation between PA and NS

The first main assumption of the DDH postulates that PA and NS are not strongly correlated. A correlation analysis and a factor analysis were conducted to investigate the relation between these two cognitive measures and their association with reading and spelling. Partial correlations between PA, NS, reading, and spelling were computed with FSIQ and age partialled out. A non-significant partial correlation between PA and NS was obtained ($r = -.191$, $p = .083$). According to Cohen's criteria (Cohen, 1988; small r = .100, medium r = .300, and large r = .500), PA was strongly correlated with reading accuracy ($r = .707$, $p < .001$) and spelling ($r = .557$, $p < .001$), whereas NS was more correlated with reading fluency ($r = -.528$, $p < .001$) (see Table 2).

A principal components factor analysis with oblique rotation (direct oblimin) of these variables resulted in two factors with eigenvalues greater than 1.0 that explained 83% of the variance (see Table 2). PA, reading accuracy, and spelling loaded on Factor 1, which accounted for 65.73% of the variance, whereas Factor 2 had high loadings for NS and reading fluency (14.27% of the variance). The correlation between the two factors was $r = -.500$.

Unique contribution of PA and NS to reading and spelling

To investigate the unique contribution of PA and NS to reading and spelling performance (the second main assumption of the DDH), hierarchical linear regression analyses were performed for each of the dependent variables. The predictive variables were entered in the following order: WISC-III FSIQ and age (covariates) were entered into the first block, and NS and PA were entered into the second block (see Table 3).

For reading fluency, NS was the most significant predictor, explaining 12.5% ($pr^2 = .125$) of the variance after controlling for WISC-III FSIQ, age, and PA, whereas PA showed a unique variance of 5.2%. The analysis was rerun to include the WISC-III Processing Speed Index in the regression equation to control for the possible influence of processing speed on reading fluency. NS still made a significant contribution to reading fluency after processing speed, intellectual ability, age, and PA were controlled for ($\beta = -0.456$, t test $= -4.935$, $p < .001$, $pr^2 = .120$).

Table 2 Partial correlation and principal components factor analysis

	Partial correlation (controlling for WISC-III FSIQ and age)				Principal components factor analysis	
	2	3	4	5	Factor 1	Factor 2
1. Phonological awareness	−0.191	0.394***	0.707***	0.557***	0.942	0.109
2. Naming speed		−0.528***	−0.300**	−0.236	0.087	0.984
3. Reading fluency			0.540***	0.430***	0.211	−0.796
4. Reading accuracy				0.688***	0.792	−0.234
5. Spelling					0.849	−0.033

WISC-III FSIQ Wechsler Intelligence Scale for Children (Third Edition)—Full Scale IQ

*$p < .05$; **$p < .01$; ***$p < .001$

Table 3 Hierarchical linear regression analysis

Dependent variable	Block	Predictors	R^2	ΔR^2	β	*t* test	pr^2
Reading fluency	1	WISC-III FSIQ	.411		0.082	1.152	.006
		Age			0.274	3.076**	.044
	2	NS	.628	.217	− 0.460	− 5.186***	.125
		PA			0.250	3.357**	.052
Reading accuracy	1	WISC-III FSIQ	.261		0.044	0.647	.002
		Age			0.201	2.336*	.023
	2	NS	.651	.390	− 0.188	− 2.183*	.020
		PA			0.621	8.606***	.323
Spelling	1	WISC-III FSIQ	.114		0.058	0.538	.003
		Age	.404	.290	0.071	0.522	.003
	2	NS			− 0.161	− 1.190	.015
		PA			0.536	4.716***	.241

R^2 and ΔR^2 variance explained, β standardized regression coefficient, pr^2 squared part correlation, representing the unique variance of each predictor when the overlapping linear effects of all other predictive variables were statistically removed, *WISC-III FSIQ* Wechsler Intelligence Scale for Children (Third Edition)—Full Scale IQ. *NS* naming speed, *PA* phonological awareness

*$p < .05$; **$p < .01$; ***$p < .001$

The results for the reading accuracy and spelling were very similar. PA exhibited a significant predictive effect for both outcomes. It explained 32.3% ($pr^2 = .323$) and 24.1% ($pr^2 = .241$) of the unique variance for reading accuracy and spelling, respectively. NS explained 2% ($p < .05$) of the unique variance for reading accuracy, and it was a non-significant predictor for spelling.

DDH subtypes

The third main assumption of the DDH assumes the existence of a subgroup of individuals with DD that has NS deficits in the absence of phonological deficits. To test this assumption, the DDH subtypes were created using scores from the PA and NS measures. Consistent with previous research (Jiménez et al., 2008; Nelson, 2015; Vaessen et al., 2009), we used a cutoff score of ≤ -1 *SD* (i.e., age-scaled score ≤ 7 derived from BANC's normative data) to establish the DDH groups. Children who scored at or below this cutoff on NS or PA but above the cutoff score on PA or NS were assigned to the *single NS deficit* group (8.2%, $n = 7$) and the *single phonological deficit* group (24.7%, $n = 21$), respectively. Those who scored at or below this cutoff on PA and NS were assigned to the *double-deficit* group (62.4%, $n = 53$). Four children (4.7%) did not show PA and NS deficits.

Performance of DDH subtypes on reading and spelling

The four children who did not show PA and NS deficits were excluded from the following analyses. Before examining the differences between the DDH subtypes in reading and spelling (the fourth main assumption), we compared the DDH subtypes on PA and NS in order to address the potential statistical artifact raised by Schatschneider et al. (2002). As shown in Table 4, children from the double-deficit group did not perform significantly worse on PA than did children from the single phonological deficit group. Similarly, the double-deficit group did not significantly differ on NS from the single NS deficit group.

Table 4 Univariate analysis of covariance

	Single PH deficit		Single NS deficit		Double deficit		ANCOVA (*F*)
	M	*SD*	*M*	*SD*	*M*	*SD*	
Phonological awareness	15.43a	6.02	25.14b	3.53	15.13a	5.31	$F(2, 76) = 17.542$***, η2p = 0.316
Naming speed	27.00a	5.09	38.86b	10.36	35.17b	7.40	$F(2, 76) = 29.401$***, η2p = 0.436
Reading fluency	48.91a	19.25	30.32a,b	15.63	40.86b	19.85	$F(2, 76) = 4.893$*, η2p = 0.114
Reading accuracy	57.73a,b	16.88	65.35a	6.98	56.93b	17.92	$F(2, 76) = 3.333$*, η2p = 0.081
Spelling[a]	42.16a	13.78	50.41a	14.61	42.43a	12.47	$F(2, 53) = 2.642$, η2p = .091

Means with different lowercase letters are significantly different at the $p = .05$ level after Bonferroni adjustment for multiple comparisons

PH phonological, *NS* naming speed, $\eta^2{}_p$ partial eta-squared

*$p < .05$; **$p < .01$; ***$p < .001$

[a] For the spelling task, 23 children were dropped out due to missing data (six children with single phonological deficit, one child with single NS deficit, and 16 children with double deficit).

An ANCOVA was performed for reading fluency, reading accuracy, and spelling, with the DDH subtypes as a fixed factor and WISC-III FSIQ and age as covariates. As shown in Table 4, a significant main effect of DDH subtypes was found for reading fluency ($p = .010$). Planned post hoc comparisons revealed that children with a double deficit performed lower in reading fluency than children with a single phonological deficit (Bonferroni $p = .010$). A significant main effect was also found for reading accuracy ($p = .041$), with the single NS deficit outperformed the double-deficit group (Bonferroni $p = .038$). Non-significant group differences were found for spelling.

Discussion

The present study extended previous research by studying DDH in an orthography of intermediate depth. We examined the four main assumptions of the DDH in Portuguese-speaking children with DD.

Correlation between PA and NS

The DDH postulates that the correlation between PA and NS is modest, and our findings found support for this assumption. Indeed, PA and NS were not significantly correlated ($r = -.191$, $p = .083$), and they loaded on separate factors, which is consistent with other DDH studies from transparent (Torppa et al., 2013; Vaessen et al., 2009) and opaque orthographies (Manis, Doi, & Bhadha, 2000; Nelson, 2015). Swanson et al. (2003) conducted a meta-analysis and found that the overall correlation coefficient between PA and NS was moderate ($r = .38$), with these two measures loading on different factors. Recently, Nelson (2015), through a confirmatory factor analysis, also observed that a two-factor model with PA and NS as independent constructs demonstrated a better fit to the data than the one-factor model in a sample of young adults with DD. These findings suggested that PA and NS involve different cognitive processes, and they contribute differently to reading and spelling. PA seems to be more related to decoding skills and NS more associated with orthographic processing (Bowers & Newby-Clark, 2002; Norton & Wolf, 2012; Pennington, 2009).

As noted by Vaessen et al. (2009), the correlation pattern in a reading-disabled population might be different from that in a normal reading population because the scores on PA, NS, and reading measures suffer from a restricted range relative to typically developing readers. Thus, the correlation between these measures might be lower than that found in a normal reading population (Swanson et al., 2003).

Unique contribution of PA and NS to reading and spelling

The second main assumption of the DDH postulates that NS predicts reading performance beyond what is accounted for by PA. Indeed, a large number of studies have consistently found that NS is the most relevant predictor of reading fluency across all orthographies (for a review, see Norton & Wolf, 2012). NS also contributes to other aspects of reading performance, such as the recognition of word-specific orthographic patterns (e.g., reading of irregular words and the recognition of familiar words) (Araújo et al., 2015; Kirby et al., 2010). The relation between NS and reading performance might be associated with the fact that NS mimics the integration of visual and verbal skills required during efficient word recognition and allows simultaneous processing of multiple stimuli presented in serial fashion (Landerl et al., 2019; Norton & Wolf, 2012; Wolf & Bowers, 1999).

The results from the hierarchical linear regression analyses indicated that NS contributed significantly to reading fluency (12% of the unique variance) over and above PA, processing speed, and intellectual ability. Reading accuracy and spelling were not significantly predicted by NS (only 2% of the unique variance). Congruent with the findings of studies in alphabetic orthographies with different degrees of orthographic complexity (Moll et al., 2014; Ziegler et al., 2010), PA accounted for a sizable amount of unique variance in reading accuracy and spelling (more than 24% of the unique variance). The finding that these literacy measures were not significantly predicted by NS does not necessarily contradict the second main assumption of the DDH because NS is principally related to reading fluency rather than to reading/spelling accuracy (Kirby et al., 2010; Norton & Wolf, 2012). Thus, our findings from an orthography of intermediate depth also found evidence that PA and NS predict different components of the reading and spelling processes.

Some researchers have considered NS to be one facet of general processing speed (Catts, Gillispie, Leonard, Kail, & Miller, 2002), which might explain why NS and reading fluency share a part of the variance. However, other studies found that NS made a unique contribution to reading fluency even after accounting for processing speed (Powell, Stainthorp, Stuart, Garwood, & Quinlan, 2007). Vaessen et al. (2009) in a sample of Dutch-speaking children with DD also found a unique contribution of NS to reading fluency but not to reading accuracy or spelling. They suggested that the unique contribution of NS to reading fluency does not necessarily indicate that NS represents a second core deficit in DD or a general processing speed deficit. They hypothesized that NS is a phonological processing speed task with an important addition of fast matching/integration of visual units to phonological codes that is absent in traditional PA tasks.

DDH subtypes

Although the majority of the children with DD in the present study have double deficit (62.4%), some children have a single phonological deficit (24.7%) or a single NS deficit (8.2%). Thus, our results found some support for the third main assumption of the DDH (i.e., the existence of a subgroup of individuals with DD that has NS deficits in the absence of phonological deficits).

To our knowledge, only one Portuguese study (Araújo et al., 2010) analyzed the prevalence of the DDH subtypes. It found that 18.2% of the children had a single NS deficit, 18.2% had a single phonological deficit, and 50% had double deficit. One possible explanation for this difference might be that we used both reading fluency and reading accuracy measures in the participants' inclusion criteria, whereas Araújo et al. (2010) only used a reading fluency measure. Some studies with children speaking different languages spanning a large range of orthographic complexities have also found that few participants meet the criteria for the single NS deficit subtype: 11.1% of English-speaking children (Norton et al., 2014), 10.5% of Dutch-speaking children (Vaessen et al., 2009), and 14.9% of Finnish-speaking children (Torppa et al., 2012).

As suggested by Wolf and Bowers (1999) and Nelson (2015), individuals with deficits only in reading fluency are more likely to meet the criteria for the single NS deficit subtype than the other DDH subtypes. In orthographically consistent languages, reading fluency constitutes the main reading difficulty for children with DD because word reading typically reaches high accuracy levels during the first years of instruction (Davies, Rodríguez-Ferreiro, Suárez, & Cuetos, 2013; Kirby et al., 2010). Thus, it would be expected that studies from more transparent orthographies would find a higher percentage of individuals with a single NS deficit than studies from less transparent orthographies. Consistent with our findings, the number of individuals with a single NS deficit in transparent and intermediate orthographies is neither higher than that observed in opaque orthographies nor more prevalent than the single phonological deficit subtype.

Performance of DDH subtypes on reading and spelling

The assumption that children with a double deficit experience more severe reading and spelling difficulties because the two deficits (i.e., PA and NS) are independent and additive is not supported in the current study. The double-deficit subtype was not significantly more impaired than both single-deficit subtypes on the reading and spelling tasks, which is consistent with the findings from other studies (Nelson, 2015; Wimmer et al., 2000). Although PA is more strongly related to reading accuracy and NS is more associated with reading fluency, we did not find significant differences between the single phonological deficit and the single NS deficit subtypes in both reading measures. Similar findings are reported by other DDH studies (Heikkilä et al., 2016; Jiménez et al., 2008; Vaessen et al., 2009).

Another topic that can be raised by the present study is whether the predictive patterns found for transparent and opaque orthographies can be generalized to an orthography of intermediate depth. In more transparent orthographies (e.g., Finnish, Greek, Spanish), it is expected that children with DD will experience less PA and reading accuracy problems, owing to the more consistent grapheme-phoneme correspondence rules, leaving NS and reading fluency as the most impaired measures (Araújo et al., 2015; Davies et al., 2013; de Jong & van der Leij, 2003; Kirby et al., 2010). In opaque orthographies (e.g., English), both accuracy and fluency tend to be problematic, with PA and NS being the strongest concurrent predictors of DD (Landerl et al., 2013; Moll et al., 2014; Ziegler et al., 2010). Our findings revealed that children with DD have significant cognitive (PA and NS) and literacy (reading accuracy, reading fluency, and spelling) deficits, with the majority of children showing impairment in all of these measures.

Studies from transparent orthographies also reported that impairments in PA account for reading difficulties only in the first years of schooling, whereas in opaque orthographies, PA is a long-term predictor (Furnes & Samuelsson, 2010; Landerl & Wimmer, 2000, 2008). On the other hand, children with DD that learn to read in a transparent orthography often perform at the ceiling level in reading accuracy after the first phases of reading acquisition (Jiménez et al., 2008; Wimmer, 1993, 1996). We found that PA accounted for significant amounts of unique variance in reading accuracy and spelling in children in the second to sixth grades, and that children with DD exhibited severe reading difficulties (less than 66% of accuracy). Taken together, these findings are closer to the results obtained in less transparent orthographies. Although previous studies with typically developing children have characterized European Portuguese orthography as an intermediate depth (Albuquerque, 2012; Seymour et al., 2003), the cognitive and reading impairments found in children with DD are more consistent with the results observed in opaque orthographies (Araújo et al., 2010; Moura et al., 2015; Sucena et al., 2009).

Notwithstanding the relevance of the present study, there are some limitations that should be addressed in future research. First, children with DD were not recruited from a large representative sample and the single NS deficit group only included seven children, which limits the generalizability of the findings. Second, although the reading accuracy and spelling measures were administered through a counterbalanced design, the use of the same words in both tasks might have influenced the performance due to practice effects. Third, it has been hypothesized that word reading reaction time might be a more critical issue than word reading accuracy in less opaque orthographies (Davies et al., 2013). In this study, we only included a text reading fluency measure. Thus, the additional inclusion of reaction time measures would contribute to a better understanding of the specific reading mechanisms that are compromised in the DDH subtypes. Fourth, a longitudinal methodology would contribute to an examination of the temporal stability of the DDH subtypes and the predictive effect of PA and NS on reading and spelling throughout literacy development. Fifth, despite extensive behavioral research produced in the last years, the neuroanatomical bases of DDH have been little investigated. Future research should also investigate the functional brain correlates of the reading impairments associated to each of the DDH subtypes. Sixth, cross-linguistic studies would be also relevant to understand which DDH assumptions are universal and which are orthography-specific. The methodology implemented in these studies should control for the factors that are referred as the cause of the variability found among DDH studies.

To conclude, the findings of the present study are partially consistent with the DDH in European Portuguese-speaking children with DD. Indeed, we found evidence that PA and NS were not significantly correlated, and that NS predicts reading fluency (but not reading accuracy and spelling) beyond what is accounted for by PA. We also observed that although the majority of the children with DD have double deficit, some children indeed have single PA or single NS deficits. In contrast to the DDH, children with a double deficit were not more impaired in reading and spelling than both single-deficit subtypes.

Consistent with previous research that provide evidence for the multiple-deficit model (e.g., Catts et al., 2017; Pennington et al., 2012; Ring & Black, 2018), our findings from an orthography of intermediate depth suggest that single phonological deficit is neither sufficient to cause DD nor to account for all the observed reading and spelling performance variability in children with DD. Thus, NS should be taken into consideration in the DD diagnosis and reading intervention programs. NS is one of the most relevant endophenotypes of DD (Landerl et al., 2013; Moura et al., 2017) and the strongest predictor of reading fluency in all orthographies (Araújo et al., 2015; Landerl et al., 2019). On the other hand, for those readers that have NS deficits (i.e., single NS deficit or double deficit), an intervention program based uniquely on PA and/or decoding skills may not be sufficient (Jiménez et al., 2008). In these cases, the inclusion of orthographic processing and reading fluency intervention programs may be especially helpful (Hawkins, Hale, Sheeley, & Ling, 2011; Lee & Yoon, 2017; National Reading Panel, 2000).

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