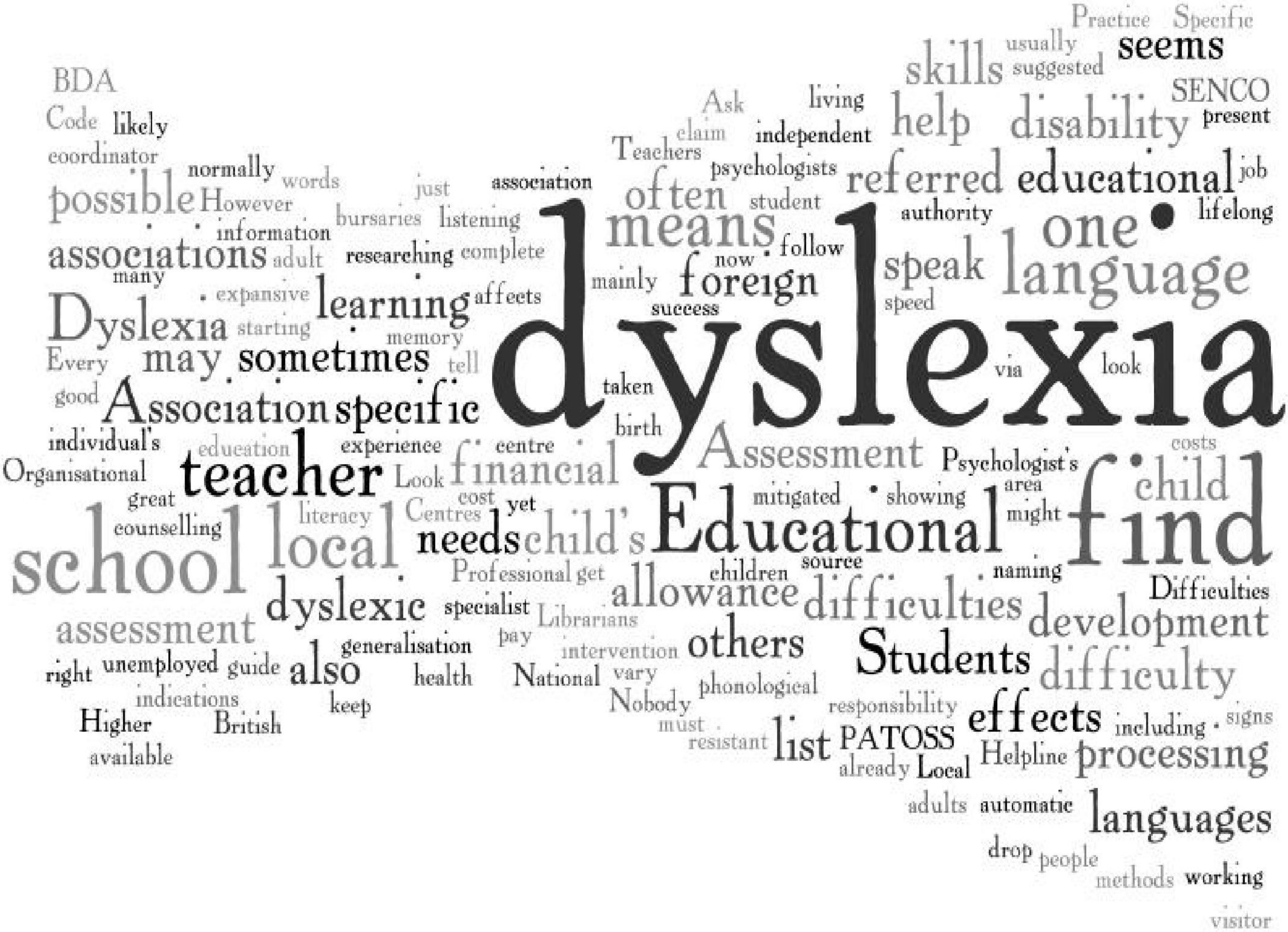

Octávio Noé Teixeira de Oliveira Moura

Avaliação Neuropsicológica na Dislexia de Desenvolvimento

Tese de Doutoramento em Psicologia, especialidade em Neuropsicologia, orientada pelo Professor Doutor Mário Manuel Rodrigues Simões e Professor Doutor Marcelino Arménio Martins Pereira e apresentada à Faculdade de Psicologia e de Ciências da Educação da Universidade de Coimbra

Setembro 2014

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Resumo

A Dislexia de Desenvolvimento (DD) é uma perturbação neurodesenvolvimental que afeta aproximadamente 5% das crianças em idade escolar, sendo caracterizada por um conjunto significativo de dificuldades na leitura e escrita. Estas dificuldades encontram-se tipicamente associadas a alterações em algumas funções neurocognitivas.

O presente trabalho de investigação teve por principal objetivo a avaliação das funções neurocognitivas associadas à DD, estando organizado em quatro estudos empíricos. A amostra foi constituída por 100 crianças (50 crianças com DD e 50 crianças leitoras normais emparelhadas por idade cronológica) com idades compreendidas entre os 8 e os 12 anos (no Estudo 2 foi adicionalmente incluído um subgrupo de crianças leitoras normais emparelhadas por idade de leitura). O protocolo de avaliação neuropsicológica incluiu testes para mensuração do funcionamento intelectual, do processamento fonológico (consciência fonológica, memória fonológica e nomeação rápida), das funções executivas (velocidade de processamento, flexibilidade, planeamento e fluência verbal), da memória de trabalho (verbal, visuoespacial e executiva), para além de medidas de avaliação da leitura e escrita.

As crianças com DD revelaram dificuldades significativas nos perfis cognitivos da WISC-III comummente associados à DD (Bannatyne, FDI, ACID e SCAD), no processamento fonológico, na memória de trabalho (componente verbal e componente executiva) e nas funções executivas (exceto no planeamento), bem como em todas as tarefas de leitura e escrita. A consciência fonológica e a nomeação rápida foram as funções neurocognitivas mais relevantes na discriminação das crianças com DD das crianças leitoras normais, enquanto a memória de trabalho (componente verbal e componente executiva) e a flexibilidade (função executiva) apresentaram uma precisão de diagnóstico moderada. A consciência fonológica foi o mais importante e consistente preditor da precisão da leitura, a nomeação rápida esteve particularmente associada à fluência da leitura e a memória de trabalho (componente verbal e componente executiva) foi um preditor significativo do desempenho da leitura e escrita apenas quando a consciência fonológica e a nomeação rápida não foram estatisticamente controladas.

Palavras-Chave: Dislexia de Desenvolvimento, Processamento Fonológico, Memória de Trabalho, Funções Executivas, Leitura, Escrita, Avaliação Neuropsicológica.

Abstract

Developmental Dyslexia (DD) is a neurodevelopmental disorder, affecting approximately 5% of school-age children. It is characterized by a large number of reading and spelling difficulties. These difficulties are typically associated to deficits in some neurocognitive functions.

The aim of the present study was the assessment of the neurocognitive functions that are often linked to DD, and it is organized in four related empirical studies. The sample consisted of 100 children (50 children with DD and 50 chronological-age-matched controls) between the ages of 8 to 12 years (Study 2 additionally included a subgroup of reading-level-matched controls). The neuropsychological protocol included tests for assessment of the intellectual functioning, phonological processing (phonological awareness, phonological memory and naming speed), executive functions (processing speed, shifting, planning and verbal fluency), working memory (phonological loop, visuospatial sketchpad and central executive), in addition to reading and spelling measures.

The children with DD revealed significant difficulties in the WISC-III cognitive profiles commonly associated to DD (Bannatyne pattern, FDI, ACID and SCAD profiles), phonological processing, working memory (phonological loop and central executive) and in the executive functions (except for planning ability), as well in all reading and spelling tasks. Phonological awareness and naming speed were the most relevant neurocognitive variables to discriminate between children with DD and typical readers, whereas working memory (phonological loop and central executive) and shifting (executive functions) showed a moderate diagnostic accuracy. Phonological awareness was the most relevant and consistent predictor of reading accuracy, naming speed was particularly related to reading fluency, and working memory (phonological loop and central executive) was a significant predictor of reading and spelling performance only when phonological awareness and naming speed were not statistically controlled.

Keywords: Developmental Dyslexia, Phonological Processing, Working Memory, Executive Functions, Reading, Spelling, Neuropsychological Assessment.

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ESTUDO 4

Lista de Acrónimos

INTRODUÇÃO

BANC – Bateria de Avaliação Neuropsicológica de Coimbra

DAE – Dificuldade de Aprendizagem Específica

DD – Dislexia de Desenvolvimento

DSM-5 – Manual de Diagnóstico e Estatística das Perturbações Mentais

WISC – Escala de Inteligência de Wechsler para Crianças

ESTUDO 1

η^2_p – Partial eta-squared

ACID – Pattern of low scores on the Arithmetic, Coding, Information and Digit Span subtests

ADHD – Attention Deficit Hyperactivity Disorder

AUC – Area under the curve

DD – Developmental dyslexia

FD – Freedom from Distractibility

FDI – Freedom from Distractibility Index

FSIQ – Full Scale IQ

LD – Learning disabilities

DSM – Diagnostic and Statistical Manual of Mental Disorders

PIQ – Performance IQ

POI – Perceptual Organization Index

PSI – Processing Speed Index

ROC – Receiver operating characteristic

SCAD – Pattern of low scores on the Symbol Search, Coding, Arithmetic and Digit Span subtests

WISC – Wechsler Intelligence Scale for Children

VCI – Verbal Comprehension Index

VIQ – Verbal IQ

ESTUDO 2

η^2_p – Partial eta-squared

β – Standardized regression coefficient

AUC – Area under the curve

BANC – Bateria de Avaliação Neuropsicológica de Coimbra

CA – Chronological-age-matched controls

DD – Developmental dyslexia

FDS – Forward task from the Digit Span

MANCOVA – Multivariate analyses of covariance

PA – Phonological awareness

PAL-PORT 22 – Oral Reading subtest from the Portuguese version of the Psycholinguistic Assessment of Language

PR^2 – Squared part correlation

R^2 – Total variance

RAN – Rapid automatized naming

RL – Reading-level-matched controls

ROC – Receiver operating characteristic

VSTM – Verbal short-term memory

WISC-III FSIQ – Wechsler Intelligence Scale for Children – Third Edition – Full Scale

ESTUDO 3

η^2_p – Partial eta-squared

ADHD – Attention Deficit Hyperactivity Disorder

ANCOVA – Univariate analyses of covariance

AUC – Area under the curve

BANC – Bateria de Avaliação Neuropsicológica de Coimbra

DD – Developmental dyslexia

EF – Executive functions

FSIQ – Full Scale IQ

GAI – General Ability Index

MANCOVA – Multivariate analyses of covariance

MANOVA – Multivariate analyses of variance

PS – Processing speed

ROC – Receiver operating characteristic

TDC – Typically developing children

TMT – Trail Making Test

ToH – Tower of Hanoi

ToL – Tower of London

VF – Verbal fluency

WCST – Wisconsin Card Sorting Test

WISC – Wechsler Intelligence Scale for Children

ESTUDO 4

η^2_p – Partial eta-squared

β – Standardized regression coefficient

AUC – Area under the curve

B – Unstandardized regression coefficient

BANC – Bateria de Avaliação Neuropsicológica de Coimbra

CE – Central executive

CFI – Comparative fit index

DD – Developmental dyslexia

DS – Digit Span subtest of the WISC-III

FSIQ – Full Scale IQ

MANCOVA – Multivariate analyses of covariance

MANOVA – Multivariate analyses of variance

PAL – Psycholinguistic Assessment of Language

PL – Phonological loop

PR^2 – Squared part correlation

R^2 – Total variance

RCTF – Rey Complex Figure Test

RMSEA – Root mean square error of approximation

ROC – Receiver operating characteristic

SE – Standard error

STM - Short-term memory

t – t-test

TR – Typical readers

VSSP – Visuospatial sketchpad

WISC – Wechsler Intelligence Scale for Children

WM – Working memory

DISCUSSÃO E CONCLUSÃO

AUC – Area under the curve

BANC – Bateria de Avaliação Neuropsicológica de Coimbra

DD – Dislexia de Desenvolvimento

DP – Desvio-padrão

DSM-5 – Manual de Diagnóstico e Estatística das Perturbações Mentais

QIEC – QI Escala Completa

ROC – Receiver operating characteristic

WISC – Escala de Inteligência de Wechsler para Crianças

Introdução

Introdução

A aprendizagem da leitura é uma competência complexa que requer a conversão de símbolos gráficos (grafemas) nos sons (fonemas) correspondentes e envolve um adequado funcionamento de diversas funções neurocognitivas e a ativação de diferentes regiões cerebrais. Para a grande maioria das crianças a aprendizagem da leitura desenvolve-se com relativa naturalidade, contudo para outras esta aprendizagem é particularmente difícil. Entre estas, encontram-se as crianças com Dislexia de Desenvolvimento (DD) que evidenciam alterações específicas em determinadas funções neurocognitivas e um conjunto alargado de dificuldades na leitura e escrita. A DD é atualmente entendida como uma dificuldade de aprendizagem específica de origem neurobiológica, caracterizada por uma dificuldade na precisão e/ou fluência na leitura de palavras e uma fraca competência ortográfica. As dificuldades na leitura resultam de um défice na componente fonológica da linguagem, que são inesperadas em relação às restantes competências cognitivas e às condições educativas proporcionadas (Fletcher, 2009; Lyon, Shaywitz, & Shaywitz, 2003).

A DD é observada em 5% a 10% das crianças (Ramus, 2003; Vellutino, Fletcher, Snowling, & Scanlon, 2004), muito embora alguns estudos apresentem estimativas de prevalência entre os 6% e os 17% dependendo dos critérios de severidade na leitura utilizados (Fletcher, Lyon, Fuchs, & Barnes, 2007). A Associação Americana de Psiquiatria na sua recente revisão do Manual de Diagnóstico e Estatística das Perturbações Mentais (DSM-5; American Psychiatric Association, 2013) estima que 5% a 15% das crianças em idade escolar apresentam uma Perturbação da Aprendizagem Específica. A DD é mais prevalente no género masculino, numa proporção de 1.5:1 a 3:1 rapazes para uma rapariga, podendo atingir valores mais discrepantes em amostras clínicas (Chan, Ho, Tsang, Lee, & Chung, 2007; Hawke, Olson, Willcut, Wadsworth, & DeFries, 2009; Liederman, Kantrowitz, & Flannery, 2005). Em Portugal, um estudo recente de Vale, Sucena e Viana (2011) aponta para uma percentagem de 5.4% das crianças em idade escolar (i.e., aproximadamente uma criança em cada 20), com um rácio de 1.5:1 rapazes para uma rapariga. É também frequente observar-se uma relação comórbida entre a DD e a Perturbação de Hiperatividade com Défice de Atenção (entre 15% a 40% das crianças com DD apresentam uma Perturbação de Hiperatividade com Défice de Atenção; Willcutt, Pennington, Olson, Chhabildas, & Hulslander, 2005) e a Discalculia (entre 15% a 70% das crianças com DD apresentam Discalculia; Willcutt et al., 2013).

Estudos de neuroimagem funcional realçaram a natureza neurobiológica desta perturbação. Investigações levadas a cabo por diversos autores (Pugh et al., 2001; B. A. Shaywitz, Lyon, & Shaywitz, 2006; B. A. Shaywitz et al., 2002) permitiram o mapeamento das áreas cerebrais comprometidas nos indivíduos com DD durante os processos de descodificação da leitura, tendo sido observada uma menor atividade da região parietal-temporal e da região occipital-temporal do hemisfério esquerdo.

Dado o conjunto de evidências empíricas acumulado nas últimas décadas sobre a natureza neurobiológica e neurocognitiva da DD, o DSM-5 (American Psychiatric Association, 2013) incluiu a DD (também designada por *"Specific Learning Disorder with Impairment in Reading"*) no grupo das Perturbações Neurodesenvolvimentais. Segundo o DSM-5, a DD é uma perturbação de origem neurobiológica que estará na base das alterações observadas a nível cognitivo, as quais estarão associadas às diversas manifestações sintomatológicas na leitura e escrita. Para além desta nova classificação, outras alterações nos critérios de diagnóstico foram efetuadas. Assim, o DSM-5 estabelece a necessidade de um desempenho na fluência, precisão e/ou compreensão da leitura substancialmente abaixo do esperado para a idade cronológica do sujeito (o ponto-de-corte deverá ser estabelecido entre -1 a -2.5 desvio-padrão), que interfere significativamente com o rendimento escolar ou atividades da vida quotidiana. O desempenho nas referidas medidas deverá ser confirmado com recurso a provas de referência normalizadas (administradas individualmente) e de uma avaliação clínica compreensiva que deverá incluir a recolha de informação médica, desenvolvimental, escolar e das manifestações sintomatológicas, bem como uma avaliação psicológica/cognitiva. As dificuldades na descodificação da leitura não deverão ser resultantes de dificuldade intelectual, atraso global do desenvolvimento, alterações sensoriais, perturbações neurológicas ou motoras.

Como referido anteriormente, para além das significativas dificuldades na leitura, as crianças com DD tendem a evidenciar défices específicos em algumas funções neurocognitivas (em particular, no processamento fonológico, nas funções executivas e na memória de trabalho). O crescente interesse pelas funções neurocognitivas envolvidas nesta perturbação da aprendizagem tem resultado na publicação de um vastíssimo conjunto de estudos científicos nos diversos sistemas ortográficos. Em Portugal, são ainda residuais os estudos neuropsicológicos publicados com esta população clínica, para além de serem igualmente escassos os instrumentos de avaliação neuropsicológica pediátrica adaptados e validados para a população Portuguesa. Neste contexto, a Bateria de Avaliação Neuropsicológica de Coimbra (BANC; Simões et al., in press) surge como um instrumento relevante no panorama nacional (e no presente estudo), apresentando dados normativos para avaliação de importantes funções neurocognitivas. Os 16 testes incluídos na BANC foram normalizados a partir de uma amostra única de 1104 crianças com idades compreendidas entre os 5 e os 15 anos, estando organizados em 6 domínios para mensuração das funções da memória, da linguagem, da atenção e funções executivas, da orientação, da motricidade e da lateralidade.

A Neuropsicologia é a ciência aplicada que tem por objetivo o estudo da expressão cognitiva e comportamental das disfunções cerebrais (Lezak, Howieson, Bigler, & Tranel, 2012; Riccio, Sullivan, & Cohen, 2010). No âmbito da prática clínica e da investigação, a avaliação neuropsicológica tem vindo a assumir um papel cada vez mais relevante. Tendo como principal objetivo a determinação da integridade estrutural e funcional dos sistemas cerebrais, de modo a permitir um exame cognitivo preciso de uma possível disfunção (Simões, 1997), a avaliação neuropsicológica oferece informações clínicas adicionais e complementares aos tradicionais instrumentos de avaliação (D'Amato, Rothlishberg, & Work, 1999). Benton (1991, p. 507) descreve a avaliação psicológica como "um aperfeiçoamento e alargamento da observação clínica que assenta na descrição mais precisa e fiável dos desempenhos do paciente, através de instrumentos e procedimentos de testes específicos, que suscitam tipos de desempenho que não são acessíveis à observação clínica". Deste modo, sendo a DD uma perturbação de base neurobiológica, a avaliação neuropsicológica das funções da linguagem, dos processos executivos e da memória desempenha um papel central no diagnóstico desta perturbação da aprendizagem específica. Para além da contribuição para uma avaliação compreensiva, o exame neuropsicológico poderá ter um papel igualmente determinante na identificação das áreas comprometidas que deverão ser alvo de intervenção.

Antes de nos centrarmos nos objetivos da presente investigação, iremos fazer uma breve incursão histórica sobre o estudo da DD e abordaremos de forma sucinta as principais funções neurocognitivas que comummente lhe estão associadas e que são alvo de estudo neste trabalho de investigação.

Perspetiva Histórica

O estudo da Dislexia tem cerca de 150 anos e iniciou-se, provavelmente, com a descrição de um paciente adulto sem aparente incapacidade cognitiva mas com uma severa dificuldade na leitura. Esta primeira descrição clínica foi efetuada por Adolph Kussmaul em 1877, tendo sugerido o termo "*word-blindness*" para caracterizar esta severa dificuldade no processamento da leitura[1]. O termo Dislexia foi introduzido pelo médico oftalmologista alemão Rudolf Berlin, em 1887, para se referir a uma forma particular de *"acquired word-blindness"* em adultos. Nas observações que realizou ao longo de 20 anos, Rudolf Berlin descreve seis pacientes que perderam a capacidade de ler após lesão cerebral. Se a lesão fosse generalizada levava a uma completa incapacidade para a leitura de palavras ("*acquired alexia"*), se a lesão fosse focal conduzia a uma grande dificuldade em interpretar símbolos manuscritos ou impressos ("*dyslexia"*) (R. F.

[1] *"A complete text-blindness may exist, although the power of sight, the intellect, and the powers of speech are intact. (…) This morbid inability we will style, in order to have the shortest possible names at our disposition, word-deafness and word-blindness"* (Kussmaul, 1877, p. 770).

Wagner, 1973). A primeira referência histórica da forma desenvolvimental desta perturbação foi descrita em 1896 pelo médico inglês Pringle Morgan, numa publicação no *British Medical Journal*, sobre as severas dificuldades na leitura de um jovem estudante de 14 anos de idade, tendo identificado este caso como *"congenital word-blindness"*[2]. Após esta primeira descrição clínica vários estudos foram publicados, entre os quais se destacam os trabalhos de James Hinshelwood. Das diversas observações realizadas, Hinshelwood (1917) identifica uma maior prevalência de casos no género masculino (10 dos 12 casos reportados) e sugere uma possível predisposição hereditária desta perturbação (seis crianças foram identificados em duas gerações numa mesma família).

Em meados dos anos 20 do século passado, clínicos e investigadores norte-americanos começaram, igualmente, a interessar-se pelo trabalho desenvolvido na Europa. Destes investigadores destaca-se o contributo do médico neurologista Samuel Orton (fundador da atual Associação Internacional de Dislexia), que entendia a leitura como um processo cognitivo complexo que envolvia várias áreas cerebrais (Orton, 1925). Este investigador defendia que a DD era resultante de uma insuficiente dominância cerebral de um hemisfério sobre o outro aquando do processamento da leitura, ao qual deu o nome de estrefossimbolia[3] (i.e., inversão de símbolos). Segundo Orton, esta perturbação teria uma prevalência aproximada de 10% da população escolar (Orton, 1937).

Em 1966 o *National Institute of Neurological Diseases and Blindness* propõe o conceito de Disfunção Cerebral Mínima (*"Minimal Brain Dysfunction"*) que inclui os casos de crianças com um funcionamento intelectual perto da média, na média ou acima da

[2] *"He has always been a bright and intelligent boy, quick at games, and in no way inferior to others his age. His great difficulty has been – and is now – his inability to read. (…) He was what Kussmaul has termed «word blind». (…) The schoolmaster who was taught him for some years says that he would be the smartest lad in the school if the instruction were entirely oral."* (Morgan, 1896, p. 1378).

[3] *"The term congenital word-blindness because of its association with the acquired condition and the implications therefrom, does not seem to be properly descriptive of this disability, and I would therefore like to offer the term 'strephosymbolia' from the Greek words, [strepho], twist, and [symbolon], symbol…"* (Orton, 1925, p. 610).

média, que apresentavam problemas de aprendizagem (leitura, escrita, aritmética, etc.) e do comportamento, entre outros, com uma gravidade de ligeira a severa, associados a desvios funcionais do sistema nervoso central (Clements, 1966). Estes desvios podiam-se manifestar através de diversas combinações de dificuldades da perceção, conceptualização, linguagem, memória, controlo da atenção, impulsividade e coordenação motora. O conceito de Disfunção Cerebral Mínima foi, desde logo, muito criticado uma vez que a grande maioria das crianças passíveis de se enquadrarem neste conceito apenas evidenciavam sinais equívocos de comprometimento neurológico (Birch, 1964; Herbert, 1964; Rie & Rie, 1980). Este conceito foi considerado vago, demasiado amplo (incluía um número muito diversificado de sintomas), com baixo valor preditivo e reduzidas evidências de comprometimento neurológico (Rie & Rie, 1980). O conceito de Disfunção Cerebral Mínima acabou por ser abandonado por falta de suporte científico.

Entre as décadas de 60 e 70, emergiu o conceito de Dificuldade de Aprendizagem Específica (DAE; "*Learning Disability*") que rapidamente se generaliza. Provavelmente, a primeira referência histórica foi proposta por Samuel Kirk (1962) que conceptualizou a DAE como uma perturbação ou atraso no desenvolvimento de uma ou mais áreas académicas (linguagem, leitura, escrita, aritmética ou outras áreas escolares) resultante de alterações psicológicas causadas por uma disfunção cerebral e/ou por um distúrbio emocional ou comportamental (não sendo o resultado de uma deficiência intelectual, privação sensorial ou fatores culturais e educacionais)[4]. Após esta primeira descrição, várias definições e critérios de inclusão/exclusão foram propostos (para uma revisão: Hammill, 1990), sendo, atualmente, uma das problemáticas mais prevalentes de todas aquelas que se inserem no espectro das necessidades educativas especiais. Os principais critérios de inclusão e exclusão utilizados na delimitação das DAE são: (1) origem neurológica, (2) discrepância académica, (3) dificuldades específicas numa ou mais áreas académicas, (4) exclusão de fatores intelectuais, sensoriais, emocionais, culturais, sociais

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[4] *"A learning disability refers to a retardation, disorder, or delayed development in one or more of the processes of speech, language, reading, writing, arithmetic, or other school subject resulting from a psychological handicap caused by a possible cerebral dysfunction and/or emotional or behavioral disturbances. It is not the result of mental retardation, sensory deprivation, or cultural and instructional factors."* (Kirk, 1962, p. 263).

e educativos, (5) condição vitalícia, entre outros (Correia, 2008; Hammill, 1990). De entre as suas diversas formas, a DD é, provavelmente, a mais frequente e a mais estudada.

Processamento Fonológico

Se os défices no processamento visual foram, até finais dos anos 70, a principal referência explicativa da DD (Herman, 1959; Hinshelwood, 1917; Orton, 1925), os défices no processamento neurolinguístico são, nos dias de hoje, entendidos como o principal preditor e o mais importante denominador comum das DAE da leitura (Fletcher, 2009; S. E. Shaywitz, 2003; Snowling, 2000; Vellutino et al., 2004). Vellutino (1979, 1987; Vellutino & Scanlon, 1982) foi dos primeiros a demonstrar a inexistência de alterações significativas nas crianças com DD em medidas de processamento visual quando controlada a influência da componente verbal da linguagem. Nestes estudos experimentais, Vellutino observou que as crianças com DD apresentavam desempenhos similares às crianças leitoras normais em medidas de memória de letras e palavras visualmente similares (por exemplo: 'b' e 'd', '*was*' e '*saw*') quando a tarefa requeria uma resposta escrita, bem como em medidas de reconhecimento visual e de recordação visual de letras e palavras de um sistema alfabético (Hebreu) não familiar para ambos os grupos. Estes resultados permitiram ao autor demonstrar que a DD é mais um sintoma de disfunção durante o armazenamento e recuperação da informação linguística do que consequência de uma deficiência no sistema visual.

Estudos subsequentes de Snowling (1981, 2000; Snowling, Nation, Moxham, Gallagher, & Frith, 1997), Stanovich e Siegel (1994), Torgesen e Wagner (1994; 1997), Shaywitz (1996; 1999), Ramus (2001; 2003), entre vários outros investigadores, vieram fornecer informações adicionais sobre o comprometimento neurolinguístico nos indivíduos com DD, em particular no processamento fonológico. O processamento fonológico é geralmente definido como a perceção, retenção, recuperação e manipulação dos sons da fala no decurso da aquisição, compreensão e produção quer da linguagem oral, quer da linguagem escrita (Catts, Fey, Zhang, & Tomblin, 1999). O processamento fonológico inclui três processos distintos mas relacionados entre si: a consciência fonológica, a codificação fonológica e a recuperação dos códigos fonológicos[5]. Os dois primeiros têm sido avaliados, respetivamente, através de testes de consciência fonológica e de memória fonológica (i.e., memória verbal imediata), enquanto o terceiro processo tem sido avaliado por intermédio de testes de nomeação rápida (i.e., "*rapid automatized naming*") (Albuquerque, 2003; Torgesen et al., 1994; Torgesen et al., 1997; R. K. Wagner & Torgesen, 1987; R. K. Wagner, Torgesen, Laughon, Simmons, & Rashotte, 1993).

Numerosas investigações têm demonstrado de forma consistente que o processamento fonológico é o preditor mais relevante do desenvolvimento da leitura (Caravolas, Lervåg, Defior, Seidlová Málková, & Hulme, 2013; Vaessen et al., 2010; Ziegler et al., 2010), e a variável com maior sensibilidade na identificação de crianças com DD (Landerl et al., 2013), independentemente do nível de opacidade do sistema ortográfico em estudo. De facto, a associação entre processamento fonológico e o desempenho da leitura e escrita em indivíduos com DD e/ou em leitores normais tem sido observada em sistemas ortográficos com elevada opacidade [Inglês (Caravolas et al., 2013; Kirby, Parrila, & Pfeiffer, 2003)], com opacidade intermédia [Francês (Martin et al., 2010); Holandês (Boets et al., 2010); Português (Albuquerque, 2012; Araújo, Pacheco, Faísca, Petersson, & Reis, 2010)], e em sistemas ortográficos mais transparentes [Alemão (Landerl & Wimmer, 2008); Espanhol (Anthony et al., 2006; Jiménez, Rodríguez, & Ramírez, 2009); Italiano (Di Filippo et al., 2005)].

Muito embora estes resultados sejam consistentes nos diversos sistemas alfabéticos, vários estudos têm demonstrado que a influência destes processos fonológicos e o desempenho nas diversas medidas de leitura parecem ser modulados pela opacidade/transparência do sistema ortográfico. Enquanto alguns estudos (Vaessen

[5] A hipótese do Duplo-Défice (Wolf & Bowers, 1999) defende que a nomeação rápida constitui um défice, nos indivíduos com DD, que é independente da consciência fonológica. Segundo este modelo, a nomeação rápida e a consciência fonológica contribuem individualmente para explicar diferentes aspectos da leitura, categorizando os indivíduos com DD de acordo com presença ou ausência de défices nestas duas componentes neurolinguísticas: (1) défices na nomeação rápida com um normal desempenho na consciência fonológica; (2) défices na consciência fonológica com um desempenho normativo na nomeação rápida; e (3) défices nas duas componentes.

et al., 2010; Ziegler et al., 2010) demonstram que a consciência fonológica é o principal preditor universal da leitura (e, de modo mais saliente, nos sistemas linguísticos menos transparentes), outros estudos sugerem que em sistemas ortográficos mais transparentes a nomeação rápida poderá ser o preditor mais significativo do desenvolvimento da leitura (de Jong & van der Leij, 2003) e o mais fiável indicador da presença de DD (Snowling, 2006).

Tem sido ainda observada uma relação entre a consciência fonológica e o processo de descodificação sublexical (ou fonológico), e entre a nomeação rápida e o processamento lexical (ou ortográfico) (Bowers, 1995; Bowers & Wolf, 1993; Manis, Seidenberg, & Doi, 1999). Segundo o modelo de dupla-via (Baron & Strawson, 1976; Coltheart, 1978, 2005) existem duas vias pela qual a leitura é processada: via sublexical (ou fonológica) e via lexical (ou ortográfica). A via sublexical baseia-se no mecanismo de conversão grafema-fonema que agrega um conjunto de regras específicas sobre as relações letra-som, sendo a via utilizada aquando da leitura de pseudopalavras e palavras regulares (via afetada no subtipo de Dislexia Fonológica). A via lexical baseia-se no reconhecimento direto das palavras que o leitor previamente já aprendeu através do acesso ao léxico mental onde se encontram as representações das formas ortográficas das palavras. É a via utilizada aquando da leitura das palavras irregulares e regulares (via afetada no subtipo de Dislexia de Superfície), permitindo uma maior fluência da leitura. De facto, um conjunto vasto de estudos tem posto em evidência a existência de uma forte relação entre a consciência fonológica e o desempenho da leitura nos diversos sistemas alfabéticos (Vaessen et al., 2010; Ziegler et al., 2010), sendo particularmente importante na fase inicial da aprendizagem da leitura (Kirby et al., 2003); enquanto a nomeação rápida surge mais associada à fluência da leitura (Kirby, Georgiou, Martinussen, & Parrila, 2010; Norton & Wolf, 2012) e às suas competências ulteriores (Furnes & Samuelsson, 2010; Vaessen et al., 2010).

Estudos com amostras nacionais têm, igualmente, demonstrado um significativo comprometimento da consciência fonológica e da nomeação rápida em crianças disléxicas, para além de contribuírem de modo independente para explicar a variância em diversas medidas de leitura (Araújo et al., 2011; Araújo et al., 2010; Sucena, Castro, &

Seymour, 2009). Refira-se, que o Português Europeu é considerado um sistema ortográfico de opacidade intermédia, dadas as suas especificidades linguísticas (Fernandes, Ventura, Querido, & Morais, 2008; Seymour, Aro, & Erskine, 2003; Sucena et al., 2009). O desenvolvimento da leitura nas crianças Portuguesas na fase inicial da aprendizagem é consideravelmente mais lento do que o tipicamente observado nos sistemas ortográficos mais transparentes (por exemplo, Alemão, Espanhol, Finlandês, Grego, Italiano), mas mais rápido que o observado no sistema linguístico Inglês, apresentando trajetórias desenvolvimentais bastante similares ao sistema ortográfico Francês. No final do 1º ano de escolaridade a precisão da leitura de palavras é de aproximadamente 74% e de não-palavras de 77%, o que contrasta com o "efeito de teto" observado nos países com ortografias mais transparentes (precisão da leitura de palavras e não-palavras acima dos 92%) e com a baixa precisão nas crianças de língua Inglesa (34% na leitura de palavras e de 29% na leitura de não-palavras) (Seymour et al., 2003). Por outro lado, a correspondência grafema-fonema é claramente não unívoca, por exemplo, as 5 vogais totalizam 18 fonemas (Sucena et al., 2009). Se em termos de opacidade ortográfica o sistema linguístico Português é de dificuldade intermédia, a nível silábico é considerado simples. A estrutura silábica mais frequente é a CV, sendo as palavras com 3 sílabas as mais comuns (33%), seguido das palavras com 4 sílabas (30%) e com 2 sílabas (16%) (Gomes & Castro, 2003; Seymour et al., 2003; Sucena et al., 2009).

De referir, ainda, que alguns autores têm admitido a hipótese do défice fonológico não se encontrar diretamente associado à presença de alterações na consciência fonológica, mas antes a uma maior sensibilidade das crianças com DD na perceção/discriminação de diferenças acústicas irrelevantes do mesmo fonema (alofones) (Noordenbos, Segers, Serniclaes, & Verhoeven, 2013; Serniclaes, Heghe, Mousty, Carré, & Sprenger-Charolles, 2004). Por exemplo, Serniclaes, Sprenger-Charolles, Carré e Demonet (2001) observaram que as crianças com DD manifestavam uma menor perceção categorial[6] comparativamente às crianças leitoras normais, dada a sua maior sensibilidade na discriminação acústica entre estímulos de uma mesma categoria. Este

[6] A percepção categorial corresponde ao grau pela qual as diferenças entre as variantes acústicas do mesmo fonema são menos perceptivas do que as diferenças acústicas entre dois fonemas diferentes.

modelo explicativo tem sido descrito na literatura como a hipótese da perceção alofónica (*"allophonic perception"*).

Outras Teorias Explicativas

Contrastando com a grande maioria dos investigadores que conferem aos défices psicolinguísticos a principal referência explicativa da DD, alguns autores têm sugerido outras bases etiológicas. Das diversas teorias explicativas, aquelas que têm sido mais referidas na literatura são a teoria magnocelular visual, a teoria do processamento auditivo e a teoria cerebelar (para uma revisão crítica sobre estas abordagens teóricas ver: Ramus et al., 2003; Vellutino et al., 2004).

A teoria magnocelular visual (*"visual magnocellular theory"*) enfatiza que para além dos défices na componente fonológica da linguagem algumas crianças com DD apresentam alterações no sistema visual magnocelular (Bellocchi, Muneaux, Bastien-Toniazzo, & Ducrot, 2013; Stein, 2001; Stein & Walsh, 1997). A nível neuroanatómico e funcional o processamento visual faz-se por duas vias: a via magnocelular e a via parvocelular. Durante a leitura, a via parvocelular parece operar durante as fixações do olho e o sistema magnocelular nos movimentos sacádicos. Estudos de Stein (2001; Stein & Walsh, 1997), Livingston (1991), Talcott et al. (1998; 2000), entre outros têm demonstrado que indivíduos com DD apresentam défices na sensibilidade magnocelular, originando alterações na atenção visual, movimento ocular e pesquisa visual. Estas alterações visuais irão interferir no processo de descodificação lexical ou ortográfico (correlacionando-se significativamente com a capacidade de leitura de palavras irregulares).

Por seu lado, a teoria do processamento auditivo (*"rapid auditory processing theory"*) preconiza que a DD assenta etiologicamente num défice primário (o défice fonológico seria secundário a este défice mais primário) na capacidade de percecionar e processar rápidas e subtis variações nos sons (Tallal, 1980). Alguns estudos têm demonstrado que os disléxicos apresentam um baixo desempenho num conjunto de tarefas auditivas, nomeadamente na capacidade de discriminar frequências e intensidades de sons (Ahissar, Protopapas, Reid, & Merzenich, 2000; Amitay, Ben-Yehudah, Banai, & Ahissar, 2002), na avaliação da ordem temporal entre estímulos acústicos (Tallal, 1980) e na resposta neurofisiológica a vários estímulos auditivos (para uma revisão: McArthur & Bishop, 2001; Ramus, 2003). Esta menor capacidade na identificação de rápidas transições de pequenos estímulos auditivos estaria associada a dificuldades nos processos de descodificação sublexical ou fonológica (i.e., na menor capacidade de leitura de não-palavras) (Talcott et al., 2000).

A teoria cerebelar (*"cerebellar theory"*) surge dos estudos iniciais de Levinson (1988) e postula que os disléxicos apresentam ligeiras disfuncionalidades ao nível do cerebelo, que se traduzem num desempenho inferior em diversas tarefas motoras, no equilíbrio, entre outras (Barth et al., 2010; Fawcett, Nicolson, & Dean, 1996; Nicolson et al., 1999). Durante a leitura, o cerebelo parece desempenhar um papel importante na calibração do movimento ocular. Neste contexto, importa acrescentar o esforço de Stein (2001) na tentativa de unificar estas diferentes teorias num único modelo teórico designado de modo abrangente como a teoria magnocelular (*"the magnocellular theory"*).

Funções Executivas

Para além dos défices na componente fonológica da linguagem, as alterações no funcionamento executivo nas crianças com DD têm sido amplamente analisadas na última década. As funções executivas são entendidas como um conjunto de processos metacognitivos, envolvidos na realização de comportamentos complexos, dirigidos para determinado objetivo e capazes de responder de modo adaptativo às diversas exigências e mudanças ambientais (Strauss, Sherman, & Spreen, 2006). Não obstante a sua ampla utilização e importância na avaliação neuropsicológica, a delimitação conceptual e metodológica dos processos executivos tem sido particularmente difícil. Se alguns investigadores vêem o funcionamento executivo numa perspetiva unitária (Barkley, 1997; Sala, Gray, Spinnler, & Trivelli, 1998), outros conceptualizam-no como um sistema multifatorial (Anderson, 2002; Miyake et al., 2000). Por outro lado, o desempenho em testes de funções executivas normalmente requer a utilização de mais do que um processo executivo e está dependente de outros processos cognitivos não executivos (Lehto, Juujärvi, Kooistra, & Pulkkinen, 2003; van der Sluis, de Jong, & van der Leij, 2007).

Na avaliação neuropsicológica o termo de "funções executivas" é utilizado para designar uma ampla variedade de funções cognitivas que envolvem o planeamento, a flexibilidade, a fluência verbal, a inibição, a velocidade de processamento, a atenção dividida, a memória de trabalho, entre outras (para uma revisão: Jurado & Rosselli, 2007; Wasserman & Wasserman, 2013). Alguns investigadores têm levantado a hipótese das funções executivas poderem estar organizadas hierarquicamente. Por exemplo, Miyake et al. (2000) através de modelos de equações estruturais confirmou a separabilidade de três processos executivos básicos – flexibilidade ("*shifting*"), atualização da informação ("*updating*") e inibição ("*inhibition*") – e o seu contributo no desempenho de tarefas executivas mais complexas. Em termos neuroanatómicos, o adequado desempenho das funções executivas está particularmente dependente (mas não limitado) da integridade e maturação do lobo pré-frontal e temporal (Demakis, 2004; Sylvester et al., 2003; Wager & Smith, 2003).

Em geral, as crianças com DD apresentam desempenhos inferiores comparativamente com as crianças leitoras normais em tarefas de velocidade de processamento (Shanahan et al., 2006; Willcutt et al., 2005), de flexibilidade (Helland & Asbjørnsen, 2000) e de fluência verbal (Landerl, Fussenegger, Moll, & Willburger, 2009; Reiter, Tucha, & Lange, 2005). Já relativamente à capacidade de planeamento resultados inconsistentes têm sido reportados na literatura (Condor, Anderson, & Saling, 1995; Marzocchi et al., 2008).

Memória de Trabalho

A memória de trabalho é, igualmente, outra das funções neurocognitivas alvo de um amplo e sistemático estudo na DD. A memória de trabalho é comummente entendida como um sistema de memória de capacidade limitada que envolve o armazenamento temporário e o processamento de informação verbal e visuoespacial. O modelo de memória de trabalho proposto por Baddeley e Hitch (1974) é o mais frequentemente utilizado nos estudos de crianças com DD ou DAE. Este modelo propõe a existência de um sistema executivo (*"central executive"*) responsável pelo controlo e processamento da informação armazenada nos dois sistemas de armazenamento: fonológico *("phonological loop"*) e visuoespacial (*"visuospatial sketchpad"*)[7]. Estudos com as várias edições da Escala de Inteligência de Wechsler para Crianças (WISC) permitiram identificar, de modo relativamente consistente, que as crianças com DD apresentam dificuldades específicas nos subtestes que remetem para a memória de trabalho (em particular, na componente executiva e na componente de armazenamento fonológico): fator Memória de Trabalho da WISC-IV (Clercq-Quaegebeur et al., 2010), fator Resistência à Distração da WISC-III (Thomson, 2003), perfil ACID (Rotsika et al., 2009) e perfil SCAD (Daley & Nagle, 1996; Thomson, 2003).

Estudos com testes mais específicos para mensuração da memória de trabalho em crianças com DD têm sido, na sua grande maioria, publicados com amostras de crianças de língua inglesa. Não são conhecidas investigações de âmbito nacional com crianças disléxicas. De modo consistente, os resultados empíricos obtidos demonstram a existência de défices significativos na componente de armazenamento fonológico (Menghini, Finzi, Carlesimo, & Vicari, 2011; Swanson, Zheng, & Jerman, 2009) e na componente executiva (Savage, Lavers, & Pillay, 2007; Swanson et al., 2009). Relativamente à componente de armazenamento visuoespacial, as crianças com DD tendem a apresentar desempenhos próximos das crianças leitoras normais (Kibby & Cohen, 2008; Schuchardt, Maehler, & Hasselhorn, 2008), exceto quando as tarefas implicam o processamento da informação visuoespacial (Bacon, Parmentier, & Barr, 2013). A memória de trabalho (em particular a componente executiva e de armazenamento fonológico) surge, ainda, como uma variável significativamente preditora

[7] Mais recentemente foi adicionado a este modelo inicial o *"episodic buffer"* (Baddeley, 2000) e o *"hedonic detection system"* (Baddeley, Banse, Huang, & Page, 2012).

do desenvolvimento da leitura (Jerman, Reynolds, & Swanson, 2012; Nevo & Breznitz, 2011) e da escrita (Jongejan, Verhoeven, & Siegel, 2007).

O número de estudos nacionais publicados na área da DD é residual e normalmente centrado na análise dos processos de descodificação da leitura e das alterações neurolinguísticas. Deste modo, a necessidade objetiva de investigações nacionais sobre o funcionamento neuropsicológico das crianças Portuguesas com DD justificam a presente investigação. Como anteriormente referido, o nível de opacidade do sistema ortográfico interfere na manifestação sintomatológica desta perturbação, pelo que se torna igualmente relevante examinar o desempenho das crianças Portuguesas falantes de um sistema ortográfico de opacidade intermédia.

Os resultados desta investigação são apresentados sob a forma de um conjunto articulado de quatro Estudos (um artigo submetido e três artigos publicados em revistas internacionais com revisão por pares e fator de impacto) que procuram dar resposta ao objetivo principal e aos objetivos específicos inicialmente delineados aquando do projeto de Doutoramento em Psicologia na especialidade de Neuropsicologia aprovado pelo Conselho Científico da Faculdade de Psicologia e de Ciências da Educação da Universidade de Coimbra. **O objetivo geral deste estudo consiste na avaliação das funções neurocognitivas associadas à DD**. Pretende-se, especificamente, avaliar o desempenho em medidas de funcionamento intelectual, processamento fonológico, funções executivas e memória de trabalho, de modo a se identificar um possível perfil neuropsicológico com adequada sensibilidade de diagnóstico na avaliação clínica desta perturbação da aprendizagem específica.

O **Estudo 1**, *WISC-III Cognitive Profiles in Children with Developmental Dyslexia: Specific Cognitive Disability and Diagnostic Utility* (Moura, Simões, & Pereira, 2014b), procura comparar o desempenho das crianças com DD relativamente a crianças leitoras normais com a mesma idade cronológica nos diversos subtestes, índices fatoriais e QIs da versão Portuguesa da WISC-III (Wechsler, 2003). São ainda analisados os principais perfis cognitivos comummente associados à DD e às DAE, nomeadamente a discrepância entre o QI Verbal e o QI Realização, o padrão de resultados de Bannatyne, o índice Resistência à Distração e os perfis ACID (resultados inferiores nos subtestes Aritmética, Código, Informação e Memória de Dígitos) e SCAD (resultados inferiores nos subtestes Pesquisa de Símbolos, Código, Aritmética e Memória de Dígitos). Estes perfis cognitivos foram analisados relativamente à sua capacidade de precisão de diagnóstico e ao ponto-de-corte ótimo das suas medidas compósitas.

No **Estudo 2**, *Developmental Dyslexia and Phonological Processing in European Portuguese Orthography* (Moura, Moreno, Pereira, & Simões, *submetido*), pretende-se analisar o desempenho no processamento fonológico (i.e., consciência fonológica, nomeação rápida e memória fonológica) e na leitura (fluência e precisão da leitura de texto; leitura de palavras regulares, irregulares e pseudopalavras) das crianças com DD comparativamente com dois grupos de controlo: (1) crianças leitoras normais emparelhadas por idade cronológica e (2) crianças leitoras normais emparelhadas por idade de leitura. A inclusão de um grupo de controlo emparelhado por idade de leitura é relevante neste tipo de estudo, pois permite analisar se as dificuldades das crianças com DD, no processamento fonológico e nas diferentes medidas de leitura, estão associadas a um défice ou a um atraso no desenvolvimento destas competências. Especificamente, pretende-se determinar a extensão das dificuldades das crianças com DD nestas diversas medidas, estimar a precisão de diagnóstico do processamento fonológico na identificação das crianças com DD e analisar a sua capacidade preditiva no desempenho das diferentes medidas de leitura.

No **Estudo 3**, *Executive Functioning in Children with Developmental Dyslexia* (Moura, Simões, & Pereira, 2014a), analisa-se o desempenho das crianças com DD em algumas funções executivas, nomeadamente na velocidade de processamento, na flexibilidade, no planeamento e na fluência verbal. Na análise inferencial entre as crianças com DD e as crianças leitoras normais com a mesma idade cronológica será ainda analisada a influência do género e da idade. Outro dos objetivos deste estudo passa pela necessidade de estimar a sensibilidade das funções executivas na capacidade de discriminar corretamente as crianças com DD do grupo de controlo.

O **Estudo 4**, *Working Memory in Portuguese Children with Developmental Dyslexia* (Moura, Simões, & Pereira, 2014c), deriva da necessidade de avaliar o contributo da memória de trabalho no diagnóstico da DD e a sua influência no desempenho da leitura e escrita. Representa um desenvolvimento lógico dos estudos anteriores, que incluem algumas medidas para mensuração da componente verbal e da componente executiva da memória de trabalho. Em particular, pretende-se analisar a existência de défices na componente executiva e nas componentes de armazenamento temporário verbal e visuoespacial nas crianças com DD comparativamente com as crianças leitoras normais com a mesma idade cronológica. Pretende-se, ainda, avaliar a validade dessas componentes na capacidade de discriminar as crianças com DD e analisar o seu valor preditivo no desempenho de diversas medidas de leitura e escrita.

No último capítulo desta dissertação serão apresentadas a **Discussão e a Conclusão**, com o objetivo de sistematizar, integrar e analisar os diversos resultados obtidos nos diferentes estudos. Em simultâneo, analisa-se o contributo da avaliação neuropsicológica no estudo da DD e debatem-se as limitações dos diferentes estudos.

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Estudo 1

WISC-III Cognitive Profiles in Children with Developmental Dyslexia: Specific Cognitive Disability and Diagnostic Utility

WISC-III Cognitive Profiles in Children with Developmental Dyslexia: Specific Cognitive Disability and Diagnostic Utility

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Abstract

This study analyzed the usefulness of Wechsler Intelligence Scale for Children (WISC-III) in identifying specific cognitive impairments that are linked to developmental dyslexia (DD) and the diagnostic utility of the most common profiles in a sample of 100 Portuguese children (50 dyslexic and 50 normal readers) between the ages of 8 and 12. Children with DD exhibited significantly lower scores in the Verbal Comprehension Index (except the Vocabulary subtest), Freedom from Distractibility Index (FDI) and Processing Speed Index subtests, with larger effect sizes than normal readers in Information, Arithmetic and Digit Span. The Verbal-Performance IQs discrepancies, Bannatyne pattern and the presence of FDI, ACID and SCAD profiles (full or partial) in the lowest subtests revealed a low diagnostic utility. However, the receiver operating characteristic (ROC) curve and the optimal cut-off score analyses of the composite ACID, FDI and SCAD profile scores showed moderate accuracy in correctly discriminating dyslexic readers from normal ones. These results suggested that in the context of a comprehensive assessment, the WISC-III provides some useful information about the presence of specific cognitive disabilities in DD.

Keyword: Developmental dyslexia, WISC-III, cognitive profiles, children.

Introduction

Developmental dyslexia (DD) is one of the most common learning disabilities (LD), affecting approximately 5% of school-age children (Ramus, 2003) and leading to substantially lower reading performance than expected according to the child's chronological age, intelligence and school grade (American Psychiatric Association, 2000). DD can be conceptualized as a specific LD that is neurobiological in origin and is characterized by difficulties with accurate and/or fluent word recognition as well as poor spelling and decoding abilities (International Dyslexia Association, 2002; Lyon, Shaywitz, & Shaywitz, 2003). Deficits in the phonological domain have consistently been found to be the primary cause of this disorder (see for a review: Fletcher, 2009; Ramus, 2003; Snowling, 2000; Vellutino, Fletcher, Snowling, & Scanlon, 2004), although other cognitive deficits, such as in working memory (Berninger, Raskind, Richards, Abbott, & Stock, 2008; Swanson, 1999, 2011), executive functions (Altemeier, Abbott, & Berninger, 2008; Brosnan et al., 2002; Helland & Asbjørnsen, 2000; Reiter, Tucha, & Lange, 2005), processing speed (Shanahan et al., 2006; Thomson, 2003; Willcutt, Pennington, Olson, Chhabildas, & Hulslander, 2005) and attention (Marzocchi, Ornaghi, & Barboglio, 2009) have also been linked to DD.

According to the Fourth Edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV; American Psychiatric Association, 2000) criteria, intellectual assessment may play an important role in diagnosis because IQ has to be at least normal and there has to be a significant discrepancy between actual reading ability and intellectual ability. Although some authors have argued that intelligence tests are not necessary for the definition of LD (Siegel, 1989, 1992), others support the idea that the discrepancy between achievement and intelligence is important to the concept of LD (Meyen, 1989; Torgesen, 1989). In the context of a comprehensive psychological assessment, the Wechsler Intelligence Scale for Children (WISC, WISC-R, WISC-III and WISC-IV; Wechsler, 1949, 1974, 1991, 2003a) is the most frequently used, not only to exclude intellectual impairments that could explain reading difficulties, but also to analyze specific cognitive deficits that may be useful for diagnosis.

Because specific cognitive deficits are linked to DD, it can be expected that dyslexic children would show weakness in some subtests of the Wechsler scales. The phonological theory postulates that children with DD have a specific impairment in the representation, storage and/or retrieval of speech sounds, and this impairment plays a central and causal role in this disorder (Ramus et al., 2003). The temporary storage of material that has been read is dependent on working memory, and working memory impairments have been related to specific characteristics of children with DD (Beneventi, Tønnessen, Ersland, & Hugdahl, 2010; Fiorello, Hale, & Snyder, 2006; Kibby & Cohen, 2008; Swanson, 1999, 2011). The Digit Span and Arithmetic subtests require processes from the phonological loop and the central executive of Baddeley's (1992, 2002, 2003) working memory model. The forward Digit Span is frequently used as a measure of the phonological loop, whereas the backward Digit Span measures the executive system (Rosenthal, Riccio, Gsanger, & Jarratt, 2006). Several studies have shown that children with DD or other LDs exhibit lower performance in the Digit Span and Arithmetic subtests (Daley & Nagle, 1996; Helland & Asbjornsen, 2004; Mayes, Calhoun, & Crowell, 1998; Rotsika et al., 2009; Thomson, 2003; Ward, Ward, Hatt, Young, & Mollner, 1995) or in the Working Memory Index from WISC-IV (Clercq-Quaegebeur et al., 2010). Coding and Symbol Search are also two subtests in which some children with DD showed impairment (Prifitera & Dersh, 1993; Shanahan et al., 2006; Thomson, 2003; Willcutt et al., 2005), as well the Information and Vocabulary subtests (Daley & Nagle, 1996) because of the relationship between intelligence and reading development known as the "Matthew effect" (Cain & Oakhill, 2011; Stanovich, 1986). As Clercq-Quaegebeur et al. (2010) stated, with less exposure to text, these children fail to build a large lexicon and enrich their general knowledge.

The discrepancy between Verbal IQ (VIQ) and Performance IQ (PIQ) in LD samples has been analyzed in a large number of studies (e.g., Daley & Nagle, 1996; Riccio & Hynd, 2000; Rotsika et al., 2009; Rourke, 1998; Slate, 1995). Although some studies have suggested that a significant VIQ-PIQ difference may be an important indicator of LD (Riccio & Hynd, 2000; Rourke, 1998), others did not find VIQ-PIQ differences to be useful in differentiating children with LDs from other groups of children (Humphries & Bone, 1993; Kavale & Forness, 1984).

As a result of the cognitive impairment observed in those with DD and LDs, many studies have tried to identify specific WISC profiles [e.g., Bannatyne pattern, Developmental Index, Learning Disabilities Index, Successive and Simultaneous Processing, Freedom from Distractibility Index (FDI), a pattern of low scores on the Arithmetic, Coding, Information and Digit Span subtests (ACID) or on the Symbol Search, Coding, Arithmetic and Digit Span subtests (SCAD)], although inconsistent results were obtained. A study about the usefulness of the WISC-III in the context of psychological assessment found that a total of 89% of school psychologists used profile analysis, and almost 70% listed it as among the most beneficial features (Pfeiffer, Reddy, Kletzel, Schmelzer, & Boyer, 2000). In the present study, only the most common profiles were analyzed: Bannatyne pattern, FDI, ACID and SCAD.

Bannatyne (1968) suggested that WISC subtest scores could be re-categorized to identify children with LD. He argued that rather than relying on the traditional VIQ and PIQ, WISC subtest scores could be re-categorized into four composite scores: spatial abilities (Block Design, Object Assembly and Picture Completion), conceptual abilities (Vocabulary, Similarities and Comprehension), sequential abilities (Digit Span, Coding and Arithmetic) and acquired knowledge (Information, Arithmetic and Vocabulary). Bannatyne (1971) reported that disabled readers exhibited a specific pattern: spatial abilities > conceptual abilities > sequential abilities. Subsequent studies found support for Bannatyne's classification system on WISC and WISC-R (Clarizio & Bernard, 1981; Rugel, 1974; M. D. Smith, Coleman, Dokecki, & Davis, 1977), whereas others demonstrated its limited diagnostic validity (D'Angiulli & Siegel, 2003; Henry & Wittman, 1981; Kavale & Forness, 1984; McKay, Neale, & Thompson, 1985; Vance & Singer, 1979). Kaufman (1981) stated that although some studies reported statistically significant mean differences in the composite scores between LD and controls, the proportions of individuals in the LD group displaying the Bannatyne pattern are quite small and their contribution to differential diagnosis is limited. Some studies explored the utility of the Bannatyne pattern with WISC-III in dyslexic and LD samples. The first work, by Prifitera and Dersh (1993), compared the baseline rates of the Bannatyne WISC-III pattern in three groups of children: those with LD, those with Attention Deficit Hyperactivity Disorder (ADHD) and those without disabilities. They found baseline rates of 33% for children with LD, 47% for children with ADHD, and 14% for children without disabilities. Although Ho, Gilger, and Decker (1988) found that this pattern was reliable and specific to their dyslexic twin sample, Smith and Watkins (2004) suggested that the use of the Bannatyne WISC-III pattern is not recommended because they only found a sensitivity of 22.4% and a specificity of 86.1% for the LD group, a sensitivity of 24% and a specificity of 86.1% for the dyslexic group, and 13.9% of false-positives were identified in the normative group.

Freedom from Distractibility (FD) was identified in a factor analysis of WISC-R and includes the Arithmetic, Coding, and Digit Span subtests (Kaufman, 1975; Reynolds & Kaufman, 1990). With the publication of WISC-III, four factors were included: Verbal Comprehension Index (VCI), Perceptual Organization Index (POI), Processing Speed Index (PSI), and FDI. The WISC-III FDI only consists of the Arithmetic and Digit Span subtests because the Coding subtest (which had previously been part of Kaufman's FD) was included with Symbol Search in the PSI. Prifitera, Weiss, and Saklofske (1998) stated that FDI is a misleading name for this construct because it encourages naive interpretations and may be better conceptualized as an index of working memory. A considerable number of studies analyzed the significant mean score differences of FD and FDI between clinical samples and typically developing children, with inconsistent findings (Ackerman, Holloway, Youngdahl, & Dykman, 2001; Anastopoulos, Spisto, & Maher, 1994; Mayes & Calhoun, 2004; Mayes et al., 1998; Prifitera & Dersh, 1993; Slate, 1995; Snow & Sapp, 2000). Thomson (2003) showed that 80% of children with DD had significantly lower mean scores on the FDI and PSI compared to the VCI and POI, whereas Alm and Kaufman (2002) also found that POI > VCI > FD in a sample of dyslexic adults.

The addition of the Information subtest to the FD triad resulted in another WISC profile: the ACID profile. Using WISC-III standardization sample, Prifitera and Dersh (1993) found that the full ACID pattern was quite rare (only 1.1% of the children from the standardization sample showed this profile), although it was more common in the LD (5.1%) and ADHD (12.3%) samples. In a sample of children with LDs, the prevalence of the ACID profile was 4.7% (Ward et al., 1995). In a Greek dyslexic sample, the prevalence was 6.7%, whereas the prevalence of the SCAD profile was 2.4% (Rotsika et al., 2009). Watkins, Kush, and Glutting (1997a) found sensitivities of 4% in the full profile (with a specificity of 99%) and 19% in the partial profile (with a specificity of 94%) in the dyslexic group, whereas the receiver operating characteristic (ROC) curve analysis resulted in an area-under-the-curve (AUC) value of .68. When analyzing group differences of the ACID and FDI profiles, the dyslexic group showed significantly lower scores than the normal reading group (Ackerman et al., 2001).

In analyzing the ACID profile results, Kaufman (1994) notes that the contribution of the Information subtest is minimal and that the differences between the clinical (ADHD and LD) and nonclinical (typically developing children) groups are largely attributable to the subtests comprising the FDI and PSI. He suggested the use of the SCAD profile because it is less vulnerable to contamination from school learning (Information subtest). Ward et al. (1995) also examined the frequency of SCAD profiles in their LD sample and obtained 19.6% true-positives and 16% false-positives. In the Daley and Nagle (1996) LD sample, the full SCAD profile was observed in 2% of the sample (partial profile was observed in 8%), the full ACID profile was observed in 1% of the sample (partial profile was observed in 12%), and the Bannatyne pattern was observed in 26% of the subjects. The mean SCAD and ACID scores were significantly different from the mean scores of the remaining subtests. Other studies showed more diagnostic utility. For instance, Thomson (2003) found that 40% of children with DD displayed a complete ACID profile, and 50% displayed a complete SCAD profile. Considering the subtest-level data, 68% presented the lowest scores on Digit Span and Coding, and 62% presented the lowest scores on Coding, Digit Span, and Symbol Search.

Thus, the results of empirical studies have shown a large cognitive variability and an inconsistency in identifying a specific profile. This variability might be related to the definition of DD used (e.g., discrepancy criterion, reading achievement criterion, response to intervention criterion), sample characteristics (e.g., clinical, school-referred), selection criteria (e.g., cut-off scores, comorbidity), assessment measures (e.g., IQ, reading, spelling, phonological processing), and others. A large body of studies has been conducted on English-speaking samples, but it is also particularly important to analyze the presence of such profiles in samples with native languages other than English (some exceptions: Clercq-Quaegebeur et al., 2010; Filippatou & Livaniou, 2005; Rotsika et al., 2009). For example, it is known that phonological processing, reading fluency and accuracy, and the prevalence of DD subtypes are influenced by specific linguistic characteristics (Boets et al., 2010; Jiménez, Rodríguez, & Ramírez, 2009; Sprenger-Charolles, Colé, Lacert, & Serniclaes, 2000). The present study is an extension of previous studies analyzing the usefulness of WISC-III in identifying the specific cognitive impairments that are associated with DD and the diagnostic utility of the most common profiles. It makes a unique contribution by using a sample of Portuguese children (no similar studies in European Portuguese orthography were found) and performs an optimal cut-off score analysis (the few studies that previously used ROC curve did not compute this type of analysis). The study had the following goals: (i) to analyze the discrepancy between VIQ and PIQ; (ii) to identify characteristic patterns of subtest strengths and weakness in children with DD; and (iii) to analyze the discriminant power of the most common WISC-III profiles through sensitivity-specificity values, ROC curve analysis and optimal cut-off scores.

Method

Participants

Participants were 100 Portuguese children between the ages of 8 and 12 (*M* = 9.81; *SD* = 1.34) in the 3rd to 6th school grades. In the dyslexic group (*N* = 50), 74% were male and 26% were female, with a gender ratio of 2.8 (clinical based sample). A recent population based study found a prevalence of DD in school age Portuguese children of 5.4%, with a gender ratio of 1.5 (Vale, Sucena, & Viana, 2011). This is consistent with the hypothesis that the prevalence of boys with DD is significantly higher in referred or clinical samples than in population samples (e.g., Hawke, Olson, Willcut, Wadsworth, & DeFries, 2009; Rutter et al., 2004; Shaywitz, Shaywitz, Fletcher, & Escobar, 1990; Wadsworth, DeFries, Stevenson, Gilger, & Pennington, 1992). The mean age of the dyslexic group was 9.80 years with a standard deviation of 1.38 years (N_{8y} = 9, N_{9y} = 17, N_{10y} = 8, N_{11y} = 7, N_{12y} = 9). Twenty-six percent of children with DD had school retention, and 36% were participants in special education systems (the Portuguese special education system establishes for children with DD the possibility of individual curriculum adjustment, adjustment in the assessment process, and personalized pedagogical support with a specialized teacher). Ninety-four percent had attended kindergarten, and 30% have relatives with reading difficulties. In the normal reader group (N = 50), 64% were male and 36% were female, with a mean age of 9.82 years and a standard deviation of 1.32 years (N_{8y} = 7, N_{9y} = 19, N_{10y} = 8, N_{11y} = 8, N_{12y} = 8). All normal readers attended kindergarten, only 2% had school retention, and 4% have relatives with reading difficulties. No statistically significant differences were found between groups with regard to gender $\chi^2(1) = 1.169$, $p = .387$, age $\chi^2(4) = 0.487$, $p = .975$ and school grade $\chi^2(3) = 1.776$, $p = .620$.

Criteria for Inclusion. For both groups, only children with the following criteria were included: (i) WISC-III Full Scale IQ (FSIQ) $\geq$ 90; (ii) native speakers of European Portuguese; (iii) at least two years of school attendance; (iv) absence of a visual, hearing or motor impairment; and (v) exclusion of a language impairment, emotional disturbance, dyscalculia, disruptive behavior disorder (ADHD, oppositional defiant disorder and conduct disorder), neurological impairment or other psychiatric disorders. These children were not included in order to ensure that cognitive deficits were not associated with any of these disorders. For the normal reader group, children with special educational needs were also excluded.

In the dyslexic group, only children who were previously diagnosed with DD by a psychologist, child psychiatrist, developmental pediatrician or child neurologist, and simultaneously having a score lower than or equal to the 15th percentile in a reading fluency and accuracy test («O Rei» Assessment Test of the Reading Fluency and Precision; Carvalho & Pereira, 2009) administered during the testing session were included. These cut-off score criteria (WISC-III FSIQ $\geq$ 90 and both reading fluency and accuracy measures $\leq$ 15th percentile) are similar to (and in some cases stricter than) the inclusion criteria used by several other authors (e.g., Ackerman et al., 2001; Frijters et al., 2011; Reiter et al., 2005; Siegel, 1992; Siegel & Ryan, 1989; Stanovich & Siegel, 1994; Swanson, 1999, 2011).

Measure

The WISC-III (Wechsler, 1991) is an individually administered intelligence test, including 13 subtests (M = 10; SD = 3), for children between the ages of 6 to 16 that measures different intellectual abilities and yields three composite IQs scores (M = 100; SD = 15): VIQ, PIQ and FSIQ; and four index scores: VCI, POI, PSI and FDI.

All participants were tested with the Portuguese version of WISC-III (Wechsler, 2003b), which was normed on a representative sample of 1354 children. The factor structure of the Portuguese version of WISC-III, analyzed through an exploratory and confirmatory factor analysis, yielded a three-factor model (VCI, POI and PSI). Thus, in this study, the FDI was analyzed as a profile (sum of the scaled scores of Arithmetic and Digit Span) rather than as an index score. The Mazes subtest was not administered.

Procedures

WISC-III administration was included as part of a broad neuropsychological protocol that also comprised a neuropsychological battery as well as reading and spelling measures. The testing was conducted in two sessions (with an interval of 10 to 15 days), lasting approximately 90 minutes per session, in a clinic or school setting during a weekday. The WISC-III was administered during the first session. All measures were administered by the first author in a standard order. No incentives were offered in exchange for participation.

Data Analysis

Data were analyzed using the Statistical Package for Social Sciences (SPSS 19.0). Independent, paired and one-sample t-tests, repeated measures and multi-factor ANOVA were calculated to investigate the significance of differences in WISC-III IQs, index scores, subscales and profiles between groups. Cohen's *d* or eta squared (η^2) were additionally calculated to determine the effect sizes of these differences. According to Cohen's (1988) criteria, *d* effect sizes are considered to be large if exceeding 0.80, moderate if at 0.50, and small if less than 0.20; whereas for η^2, .01 constitutes a small effect, .06 a medium effect and .14 a large effect.

ROC curve analysis was performed to examine the differential discriminatory power of WISC-III profiles for the diagnosis of DD. ROC curve analysis systematically sweeps across all possible true positive (sensitivity) and false positive (1-specificity) values of a diagnostic test, graphically illustrates the test's full range of diagnostic utility and calculates the AUC, which provides an accuracy index for the test (Fawcett, 2006; McFall & Treat, 1999; Metz, 1978; Watkins et al., 1997a). The more accurately a test is able to discriminate between groups (children with DD vs. normal readers), the more its ROC curve will deviate toward the upper left corner of the graph. The AUC is the average of the true positive rate, taken uniformly over all possible false positive rates (Krzanowski & Hand, 2009) that range between .5 and 1.0. An AUC value of 1.0 is perfectly accurate because the sensitivity is 1.0 when the false positive rate is .0, whereas an AUC value of .5 reflects a completely random classifier. An AUC of .5 to .7 indicates low test accuracy, .7 to .9 indicates moderate accuracy, and .9 to 1.0 indicates high accuracy (Swets, 1988).

Table 1. Percentages of discrepancies between VIQ and PIQ in normal readers and children with DD

Number of Points	Normal Readers			Dyslexic			Daley & Nagle (1996)	Rotsika et al. (2009)
	PIQ>VIQ	VIQ>PIQ	Total	PIQ>VIQ	VIQ>PIQ	Total		
0	4.0		4.0	2.0		2.0	---	4.4
1 – 5	14.0	18.0	32.0	12.2	14.3	26.5	29.5*	33.9
6 – 10	10.0	16.0	26.0	14.3	12.2	26.5	23.1	23.3
11 – 15	4.0	14.0	18.0	6.1	8.2	14.3	20.1	18.3
16 – 20	2.0	10.0	12.0	6.1	4.1	10.2	18.9	13.3
≥ 21	0	8.0	8.0	16.3	4.1	20.4	11.4	6.7

Note. * This percentage value relates to a discrepancy ranging from 0 to 5 points.

Results

IQs and Index Scores

A paired sample t-test showed a statistically significant difference between VIQ and PIQ for normal readers $t(49) = 3.542$, $p = .001$, $d = 0.46$ with VIQ > PIQ; but a non-significant difference for children with DD $t(49) = -1.651$, $p = .105$, $d = 0.32$. The absolute mean Verbal–Performance discrepancy for the dyslexic group was 3.55 (*SD* = 15.05; range = 0–37) and 5.30 (*SD* = 10.57; range = 0–32) for normal readers.

Table 1 shows the percentage of the discrepancies between VIQ and PIQ, compared with the findings of Daley and Nagle (1996) and Rotsika et al. (2009). Thirty-eight percent of normal readers and 44.9% of children with DD had a difference ≥ 11 points. Only 8% (VIQ > PIQ) of normal readers showed a difference ≥ 21 points, in contrast with 20.4% of the dyslexic group (16.3% showed VIQ < PIQ). Thirty percent of normal readers had a PIQ > VIQ discrepancy (and 66% a VIQ > PIQ), whereas among the dyslexic group, the percentage was 55.1% (and 42.9% a VIQ > PIQ).

Statistically significant differences were found between children with DD and normal readers for FSIQ and VIQ (see Table 2). The VIQ scores of normal readers were 12.86 points greater, and for FSIQ they were 9.71 points greater. For PIQ, no significant difference was found. Relative to the WISC-III index scores, children with DD showed significantly lower VCI ($p < .001$) and PSI ($p < .01$) scores.

A two-factor analysis of variance was conducted to explore the impact of group x gender, group x age, and group x school grade on the three IQs and on the three index scores, but no significant differences were found.

Subtests Scores

As shown in Table 2, Vocabulary was the only subtest with no statistically significant difference from the six VIQ subtests. The largest effect sizes were observed in the Information, Arithmetic and Digit Span subtests. The lowest scores for both groups were in the Digit Span subtest, with significant differences between groups in forward and backward span. In the PIQ subtests, significant differences with moderate effect sizes were found in Object Assembly, Coding and Symbol Search. That is, children with DD showed significantly lower scores than normal readers in the subtests included in the WISC profiles (Bannatyne sequential abilities, FDI, ACID and SCAD): the Information, Arithmetic, Digit Span, Coding and Symbol Search subtests (and also in the Similarities, Comprehension and Object Assembly subtests).

A two-factor ANOVA was also conducted to explore the impact of group x gender, group x age, and group x school grade on the 12 WISC-III subtests. No significant differences were found.

Table 2. Mean WISC-III scores and standard deviations for normal readers and children with DD

	Normal Readers	Dyslexic	*t*(98)	*p*	*d*
	M ± *SD*	*M* ± *SD*			
IQs					
FSIQ	108.24 ± 11.64	98.53 ± 8.55	4.721	<.001	0.95
VIQ	109.98 ± 11.20	97.12 ± 10.72	5.831	<.001	1.17
PIQ	104.68 ± 11.91	100.67 ± 11.42	1.707	.091	0.34
Index Scores					
VCI	108.90 ± 11.36	97.71 ± 11.14	4.942	<.001	0.99
POI	104.04 ± 12.48	102.12 ± 11.95	0.780	.437	0.15
PSI	105.98 ± 14.75	97.22 ± 12.07	3.228	<.01	0.65
Subtests					
Information	10.74 ± 2.38	8.18 ± 1.93	5.859	<.001	1.18
Similarities	11.70 ± 2.73	9.22 ± 2.45	4.739	<.001	0.95
Arithmetic	12.10 ± 2.04	9.45 ± 2.11	6.348	<.001	1.17
Vocabulary	11.72 ± 2.14	10.84 ± 2.59	1.847	.068	0.37
Comprehension	11.92 ± 2.53	10.61 ± 2.37	2.648	<.01	0.53
Digit Span (DS)	9.76 ± 2.26	7.63 ± 1.75	5.221	<.001	1.05
DS Forward*	7.36 ± 1.45	6.20 ± 1.13	4.403	<.001	0.89
DS Backward*	4.56 ± 1.34	3.63 ± 0.97	3.929	<.001	0.80
Picture Completion	10.16 ± 2.51	9.43 ± 2.70	1.394	.166	0.28
Picture Arrangement	11.18 ± 2.37	11.78 ± 2.57	-1.197	.234	0.24
Block Design	10.44 ± 2.50	10.47 ± 2.57	-0.058	.954	0.01
Object Assembly	10.90 ± 2.96	9.78 ± 2.32	2.099	<.05	0.42
Coding	11.12 ± 2.70	9.61 ± 2.45	2.900	<.01	0.58
Symbol Search	10.96 ± 3.12	9.31 ± 2.64	2.840	<.01	0.57
Profiles					
Spatial Abilities	31.50 ± 6.10	29.67 ± 5.79	1.526	.130	0.30
Conceptual Abilities	35.34 ± 5.99	30.67 ± 6.04	3.854	<.001	0.77
Sequential Abilities	32.98 ± 4.76	26.69 ± 4.00	7.101	<.001	1.43
Acquired Knowledge	34.56 ± 5.23	28.46 ± 5.20	5.807	<.001	1.16
FDI	21.86 ± 3.30	17.08 ± 2.89	7.654	<.001	1.54
ACID	43.72 ± 6.10	34.87 ± 4.91	7.929	<.001	1.59
SCAD	43.94 ± 7.02	36.00 ± 5.50	6.254	<.001	1.25

Note. * Raw scores are presented for Forward and Backward Digit Span.

Profiles Scores

Statistically significant differences between the dyslexic and normal reader groups were found in six of the seven analyzed WISC-III profiles (see Table 2). Very large effect sizes were observed in the composite score of Bannatyne's sequential abilities, FDI, ACID and SCAD, in which the mean scores of the dyslexic group were 1.43*SD*, 1.54*SD*, 1.59*SD* and 1.25*SD*, respectively, below the mean scores of the group of normal readers. Once again, a two-factor analysis of variance found no significant differences for group x gender, group x age, and group x school grade on these profiles.

Additionally, we were interested in investigating how these profiles operate only in the dyslexic group. A repeated measures analysis of variance revealed significant differences between the Bannatyne's composite scores, $F(3, 47) = 6.358$, $p < .001$, $\eta^2 = .293$. A comparison of main effects (Bonferroni $p < .05$) showed that spatial and conceptual abilities > sequential abilities, and conceptual abilities > acquired knowledge. The means of the FDI ($M = 8.54$; $SD = 1.44$), ACID ($M = 8.71$; $SD = 1.22$) and SCAD ($M = 9.00$; $SD = 1.37$) scaled scores calculated for the dyslexic group were significantly different from the mean of 10.30 ($SD = 1.37$) for the remaining subtests: FDI $t(49) = -8.514$, $p < .001$, $d = 1.25$; ACID $t(49) = -9.006$, $p < .001$, $d = 1.22$ and SCAD $t(49) = -6.614$, $p < .001$, $d = 0.94$.

Discriminant Power of WISC-III Profiles

Following the criteria of Prifitera and Dersh (1993), children were considered to be positive for the full profile when their scores on the four ACID (excluding Symbol Search and Mazes) and SCAD subtests or on the two FDI subtests were less than or equal to the scores on the remaining subtests. For the partial profile, scores on any three of the four ACID and SCAD subtests had to be less than or equal to the scores on the remaining subtests. We were also interested in investigating the presence of FDI in the three and four lowest-scoring subtests, as well of ACID and SCAD in the five and six lowest-scoring subtests.

Table 3. Diagnostic accuracy of specific WISC-III profiles in children with DD versus normal readers

	Sensitivity (True Positive)	Specificity (True Negative)	False Positive	False Negative	Positive Predictive Power	Negative Predictive Power
Bannatyne pattern	.22	.90	.10	.78	.69	.54
FDI						
full profile	.02	.98	.02	.98	.50	.51
in 3 lowest subtests	.18	.94	.06	.82	.75	.54
in 4 lowest subtests	.39	.90	.10	.61	.79	.60
ACID						
full profile	.08	.98	.02	.92	.80	.52
partial profile	.20	.96	.04	.80	.83	.55
in 5 lowest subtests	.22	.98	.02	.78	.92	.56
in 6 lowest subtests	.45	.94	.06	.55	.88	.64
SCAD						
full profile	.00	.98	.02	1.00	.00	.50
partial profile	.08	.92	.08	.92	.50	.51
in 5 lowest subtests	.10	.96	.04	.90	.71	.52
in 6 lowest subtests	.18	.92	.08	.82	.69	.53

Table 3 shows the diagnostic accuracy of WISC-III profiles. The Bannatyne pattern showed a sensitivity of .22 (i.e., 22% of the dyslexic children were correctly diagnosed) and a specificity of. .90 (i.e., 90% of normal readers were classified by the Bannatyne pattern as not having a disability). The number of children displaying the Bannatyne pattern did not differ between the dyslexic and normal reader groups, $\chi^2(1) = 1.986$, p = .159, kappa = .124. The full FDI, ACID and SCAD profiles misclassified the children with DD, only 0% to 8% of whom were properly diagnosed (true positive). The presence of ACID in the lowest six subtests and FDI in the lowest four subtests showed a greater diagnostic utility. For ACID, a sensitivity of .45, a specificity of .94, a positive predictive power of .88

(i.e., 88% of children classified as positive are children with DD) and a negative predictive power of .64 (i.e., 64% of children classified as negative are normal readers) were obtained, whereas FDI revealed a sensitivity of .39, a specificity of .90, a positive predictive power of .79 and a negative predictive power of .60. The number of children displaying ACID in the six lowest-scoring subtests differed between the dyslexic and normal reader groups $\chi^2(1) = 17.830$, $p < .001$, kappa = .391; as did the presence of FDI in the four lowest-scoring subtests $\chi^2(1) = 9.646$, $p < .01$, kappa = .289.

These results suggested that the presence of the full Bannatyne, FDI, ACID and SCAD profiles did not efficiently distinguish between children with and without DD. However, the composite scores of these profiles showed statistically significant differences with large effect sizes between groups (see Table 2). Thus, a ROC curve analysis was performed because it is independent of prevalence rates and cut-off values (McFall & Treat, 1999; C. B. Smith & Watkins, 2004). This analysis was conducted for FDI, ACID and SCAD composite scores; the Bannatyne pattern was excluded because is a dichotomous variable (presence vs. absence).

As shown in Figure 1, the ACID and FDI ROC curves are elevated over the reference line. The AUC value for ACID was .875 ($p < .001$, $SE = .033$, 95%CI = .810–.941), i.e., a randomly selected child with DD will have a lower ACID score than a randomly selected child without DD approximately 87.5% of the time. The AUC values for FDI and SCAD were .862 ($p < .001$, $SE = .036$, 95%CI = .792–.933) and .809 ($p < .001$, $SE = .042$, 95%CI = .727–.891), respectively. As Swets (1988) noted, these AUC values are indicative of a moderate accuracy in discriminating between dyslexic and non-dyslexic children.

Additionally, the Youden index (Youden, 1950) was calculated (J = sensitivity + specificity - 1) to analyze the optimal cut-off scores for FDI, ACID and SCAD (note that all children in the sample have a FSIQ $\geq$ 90 with a mean of 103.43). For FDI, the optimal cut-off score was 17.50 ($J = .552$), yielding a sensitivity of .61 and a specificity of .94; for ACID, the optimal cut-off score was 37.50 ($J = .573$), which yielded a sensitivity of .67 and a specificity of .90; and for SCAD, a cut-off of 41.50 ($J = .437$) yielded a sensitivity of .84 and a specificity of .60.

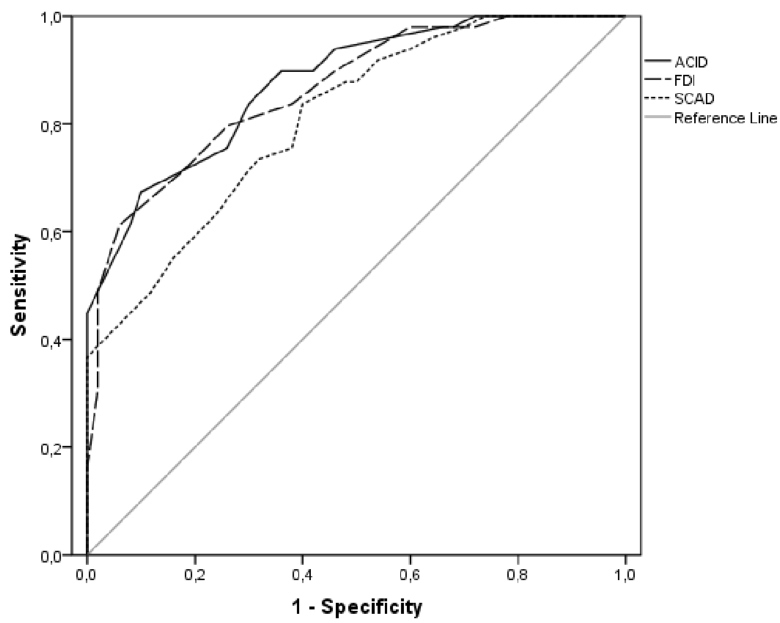

Figure 1. ROC curve comparing true- and false-positive rates among children with DD and normal readers in the FDI, ACID and SCAD profiles

Discussion

In this study, approximately 55% of children with DD had scores such that PIQ > VIQ (30% in normal readers), and 20.4% revealed a VIQ-PIQ discrepancy equal to or above 21 points (compared to 8% in normal readers and 18.1% in the WISC-III Portuguese standardization sample). Non-significant differences were found in the Verbal-Performance discrepancy, and the mean difference was smaller than for normal readers. This finding was also reported by other studies with LD (Pereira & Simões, 2005) and children with DD (Rotsika et al., 2009), and demonstrated the minimal practical value of the VIQ–PIQ discrepancy for differential diagnosis (Kaufman, 1981). The analyses of IQ mean scores between groups revealed that dyslexics had significantly lower scores in FSIQ and VIQ, which is consistent with a large number of studies (e.g., Ackerman et al., 2001; Laasonen, Leppämäki, Tani, & Hokkanen, 2009; Rotsika et al., 2009; Swartz, Gfeller, Hughes, & Searight, 1998). The VIQ comprises subtests that are more strongly associated with school learning, verbal abilities and working memory, and it was therefore expected that these tests would be more difficult for these children. Results from the WISC-III index scores showed that, in addition to VCI, processing speed can also be an additional risk factor, whereas a non-significant difference was found in POI, which may indicate that nonverbal reasoning ability is not compromised in these children with DD. These findings were also reported by previous studies (Ackerman et al., 2001; Prifitera & Dersh, 1993; Thomson, 2003).

At the WISC-III subtest level, inferential analysis showed that the six subtests with the largest effect sizes were Information, Arithmetic, Digit Span, Similarities, Coding and Symbol Search, with the dyslexic children exhibiting significantly lower scores. Significant differences were also found in Comprehension and Object Assembly (the only subtest from POI). Surprisingly, Vocabulary was the subtest with the second-highest scores among children with DD, and no statistical differences were found in comparison with normal readers (although the *p-value* was closer to statistical significance), most likely because some children with DD received a direct intervention from the special education system, may have had adequate cultural opportunities at home and may have revealed some intellectual curiosity for particular topics, thereby minimizing the impact of the low vocabulary knowledge that is common in these children. A similar finding was also reported in samples of dyslexic (Thomson, 2003) and LD children (Mayes et al., 1998). Contrary to the results reported by other studies (Clercq-Quaegebeur et al., 2010; Rotsika et al., 2009; Thomson, 2003), Similarities was one of the lowest subtests among children with DD. Similarities subtest requires greater demands on verbal abstract reasoning skills, that seems to be diminished in our dyslexic group. This unexpected finding was also observed in two studies with Portuguese learning disabled children, which included a subgroup of children with DD (Cardoso, 2007; Pereira & Simões, 2005). It would be particularly relevant clarify in subsequent studies if Similarities is (or it is not) also a "problematic" subtest for Portuguese children with DD.

The six subtests with the lowest scores for the dyslexic group (in order from lowest to highest) were Digit Span, Information, Similarities, Symbol Search, Picture Completion and Arithmetic. As reported by a large number of studies, dyslexics typically exhibit lower performance in Arithmetic, Digit Span, Information, Coding and Symbol

Search (Mayes et al., 1998; Rourke, 1998; Thomson, 2003; Ward et al., 1995). These results seem to suggest that Portuguese children with DD demonstrated difficulties in the same subtests linked to specific cognitive deficits that other international studies have shown are impaired in DD.

Although inferential analysis showed that normal readers outperformed dyslexics in a large number of WISC-III subtests, the scaled scores of the dyslexic group were within norm (the exception was Digit Span), and therefore at risk of being unobserved in clinical assessment. Thus, the analysis of the most common WISC-III profiles linked to DD may provide additional diagnostic information beyond the subtest-level analyses. Relative to the WISC-III re-categorizations, Prifitera and Dersh (1993) stated that the Bannatyne pattern is useful for diagnostic purposes, but our results did not support their conclusion. The Bannatyne pattern was present only in 22% of dyslexic children and in 10% of the normal reader group. Smith and Watkins (2004) also reported similar percentages in their sample of children with DD and LD. Large, significant differences in FDI, ACID and SCAD profiles were also found, with children with DD scoring at least 1.25 *SD* below normal readers. Elwood (1993) stated that the presence of a significant difference alone does not imply that the test can discriminate among subjects with sufficient accuracy for clinical use. We therefore additionally performed an analysis of the discriminant power (sensitivity-specificity values, ROC curve and optimal cut-off scores) of these three profiles.

Although the presence of full or partial FDI, ACID and SCAD profiles was more prevalent among dyslexics than among normal readers, the sensitivity and specificity values revealed a low diagnostic accuracy. However, when we analyzed the mean of the composite scores, moderate accuracy was obtained. A randomly selected child with DD will have a lower FDI, ACID and SCAD score than a randomly select normal reader approximately 86.2%, 87.5% and 80.9% of the time, respectively. These results from the ROC curve analysis revealed a higher diagnostic accuracy than the findings reported by Watkins et al. (1997a; 1997b) in LD samples. One of the particularities of this study was the analysis of optimal cut-off scores for FDI, ACID and SCAD, because previous studies did not perform such analysis. For FDI, a score less than or equal to 17.50 correctly identified 61% of dyslexic children (6% false positives); for ACID, a score less than or equal to 37.50 correctly identified 67% of dyslexic children (10% false positives); and for SCAD, a score less than or equal to 41.50 correctly identified 84% of dyslexic children (40% false positives). The results from the optimal cut-off scores analysis showed greater diagnostic utility than the presence of full or partial profiles. New studies are needed to explore and compare the diagnostic accuracy of these and others cut-off scores in dyslexic samples. Compared to the other profiles, ACID showed a higher discriminant power.

In sum, our findings from a sample of Portuguese children were also consistent with previous studies that found that VIQ-PIQ discrepancies, the Bannatyne pattern and the presence of the FDI, ACID and SCAD profiles in the lowest-scoring subtests do not efficiently distinguish children with DD from those without DD. However, the composite scores of FDI, ACID and SCAD profiles showed greater diagnostic utility and subtest-level analyses may provide useful information beyond the global scores about the presence of specific cognitive impairments in children with DD. In clinical practice, weakness on a specific profile is not a sufficient diagnostic criterion for dyslexia; conversely, the lack of this profile should not exclude the possibility of dyslexia (Clercq-Quaegebeur et al., 2010; Thomson, 2003). As Mayes and Calhoun (2004, p. 566) asserted, "the presence or absence of profile types certainly should not be the basis for making a diagnosis. Profiles are clinically useful because they may alert a clinician to certain diagnostic possibilities and they provide knowledge about the pattern of strengths and weaknesses that characterize certain disorders". IQ tests yield information that is only a component of the DD diagnosis and decision-making process (Prifitera et al., 1998) and need to be viewed in the context of a more comprehensive assessment that must include other tests, such as phonological awareness, rapid naming, working memory, reading and spelling measures, executive functions, and others.

The present study revealed some limitations that needed be addressed in future research. First, the pattern of WISC-III subtests scores found in children with DD was only compared to a control group and did not include other clinical samples (e.g., ADHD). Studies have found that such profiles also have some diagnostic utility in ADHD children (Mayes & Calhoun, 2004; Mayes et al., 1998; Prifitera & Dersh, 1993; Swartz et al., 1998).

Second, the two groups were not matched for WISC-III FSIQ. This additional inclusion criterion would have been a better baseline to compare cognitive profiles differences between groups. Third, we established a strict cut-off score criterion for WISC-III FSIQ (≥ 90) in order to decrease Type I error (false positive). Obviously, such strict criterion increased Type II error (false negative), excluding from the sample some children with DD that had a WISC-III FSIQ lower than 90. Another limitation was the fact that this study did not analyze the effects of socioeconomic status or parental educational attainment.

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Estudo 2

Developmental Dyslexia and Phonological Processing in European Portuguese Orthography

Developmental Dyslexia and Phonological Processing in European Portuguese Orthography

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Abstract

This study analyzed the performance of phonological processing, the diagnostic accuracy and the influence on reading in children who were native speakers of an orthography of intermediate depth. Portuguese children with developmental dyslexia (DD; *N* = 24; aged 10 to 12 years), chronological-age-matched controls (CA; *N* = 24; aged 10 to 12 years) and reading-level-matched controls (RL; *N* = 24; aged 7 to 9 years) were tested on measures of phonological processing (phonological awareness, naming speed and verbal short-term memory) and reading. The results indicated that the children with DD performed significantly poorer in all measures compared with the CA and RL. Phonological awareness and naming speed showed a high accuracy (receiver operating characteristics curve analysis) for discriminating the children with DD from the CA and RL, whereas the presence of abnormally low scores in phonological awareness and naming speed were more frequent in the DD group than in the controls and the normative population. Hierarchical linear regression analyses revealed that phonological awareness was the most important predictor of all reading accuracy measures, whereas naming speed was particularly related to text reading fluency.

Keywords: Developmental dyslexia, phonological awareness, naming speed, verbal short-term memory, reading.

Introduction

There is a strong consensus on the importance of phonological processing for reading development (bidirectional link) and it is widely accepted that the central difficulty in developmental dyslexia (DD) reflects a deficit in the phonological domain (Fletcher, 2009; Ramus, Marshall, Rosen, & van der Lely, 2013; Vellutino, Fletcher, Snowling, & Scanlon, 2004). The phonological domain deficits hypothesis is supported by neuroimaging studies, which have documented the disruption of neural systems for reading in individuals with DD, in particular, the left hemisphere posterior brain systems (Finn et al., 2014; Richlan, Kronbichler, & Wimmer, 2011; Shaywitz, Lyon, & Shaywitz, 2006). Although, the phonological domain is the main factor associated to reading performance, its weight varies as a function of script transparency (Caravolas, Lervåg, Defior, Málková, & Hulme, 2013; Ziegler et al., 2010). Therefore, the present study examined the presence of specific deficits in the phonological processing of children with DD who were native speakers of an orthography of intermediate depth (European Portuguese orthography) and their association with reading fluency and reading accuracy. We also investigated the diagnostic accuracy of phonological processing measures to correctly discriminate between typical readers and children with DD.

Phonological processing is generally defined as the perception, storage, retrieval, and manipulation of the sounds of language during the acquisition, comprehension, and production of both spoken and written codes (Catts, Fey, Zhang, & Tomblin, 1999). Phonological processing includes three interrelated but distinct phonological processes: (1) phonological awareness (PA), (2) phonological recoding in lexical access (also named naming speed, rapid naming or the lexical retrieval of phonological codes), and (3) phonetic recoding to maintain information in working memory [also named phonological memory or verbal short-term memory (VSTM)] (Torgesen, Wagner, & Rashotte, 1994; Wagner & Torgesen, 1987). Originally, these three phonological processes were treated as a single phonological component; however, the double-deficit hypothesis postulates that naming speed constitutes a second core deficit in DD that is independent from a phonological deficit (Wolf & Bowers, 1999, 2000). The double-deficit hypothesis assumes that the naming speed uniquely contributes to the reading performance and that a subgroup of individuals with DD with naming speed problems in the absence of PA problems (and vice versa) should exist. Individuals with a double deficit will show more severe reading problems compared with individuals with a single naming or single phonological deficit because the two problems are independent and additive. Whereas some studies support the double-deficit hypothesis (Araújo, Pacheco, Faísca, Petersson, & Reis, 2010; Sunseth & Greig Bowers, 2002; Wolf, Bowers, & Biddle, 2000), others did not find empirical evidence (Ackerman, Holloway, Youngdahl, & Dykman, 2001; Pennington, Cardoso-Martins, Green, & Lefly, 2001; Vaessen, Gerretsen, & Blomert, 2009; Vukovic & Siegel, 2006).

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 (e.g., deletion, substitution, blending, reversal, segmentation, and other tasks). There is strong evidence of the importance of PA in the acquisition of early reading skills across all alphabetic orthographies. This link appears to be bidirectional. Thus, PA facilitates reading development, and successful reading development improves PA performance (Boets et al., 2010; Perfetti, Beck, Bell, & Hughes, 1987; Wagner, Torgesen, & Rashotte, 1994). Children who are relatively strong in PA before reading instruction begins typically learn to read easier than other children, whereas children who exhibited impairments in PA tend to present significant difficulties in reading achievement (Catts et al., 1999; Nithart et al., 2011; Wagner & Torgesen, 1987). Some of these children are eventually diagnosed with DD during the elementary school grades (Scarborough, 1990). Deficits in PA, relative to chronological-age-matched controls (CA) and/or reading-level-matched controls (RL), have been found in various studies of DD in transparent and opaque orthographies (Boets et al., 2010; Caravolas, Volín, & Hulme, 2005; Martin et al., 2010; Pennington et al., 2001).

Phonological recoding in lexical access refers to the rapid access of phonological information stored in long-term memory, and it is usually assessed by naming speed tests. Denckla and Rudel (1976a, 1976b) found that children with DD are significantly slower in naming a set of well-known visual items (letters, numbers, colors, or objects) than typically developing children, and the authors named these tasks as "Rapid Automatized Naming" (RAN). A wide range of cognitive processes are involved in RAN tasks: integration of visual features and pattern information with stored orthographic representations, integration of visual and orthographic information with stored phonological representations, access and retrieval of phonological labels, attentional processes to the stimulus, processing speed, among others (for a review see: Kirby, Georgiou, Martinussen, & Parrila, 2010; Norton & Wolf, 2012). Several studies have suggested that children with DD have significant difficulties in RAN tasks because these tasks can be viewed as an index of how well children are able to establish the word-specific orthographic representations that underlie reading (Clarke, Hulme, & Snowling, 2005; Ehri, 1995). Even in orthographies that are more regular than English, individuals with DD manifest RAN deficits compared with CA and/or RL, which suggests that the vulnerability extends beyond phonological decoding. These findings have been reported for Dutch (Boets et al., 2010; de Jong & van der Leij, 2003), French (Martin et al., 2010), German (Landerl, 2001), Portuguese (Araújo et al., 2010), Spanish (Jiménez, Rodríguez, & Ramírez, 2009), and other languages. A large number of studies have consistently found that RAN ability is the most relevant predictor of reading fluency across all orthographies in typical and dyslexic readers (Kirby et al., 2010; Norton & Wolf, 2012). Some authors have noted that in transparent orthographies, PA may be a less reliable marker of DD than RAN, most likely because the phonological demands are reduced in transparent orthographies (de Jong & van der Leij, 2003; Snowling, 2006). Indeed, it is expected that children in more transparent orthographies experience less reading decoding (accuracy) problems, due to the more consistent grapheme-phoneme correspondence rules, than their peers of less transparent orthographies, leaving fluency as the most useful reading variable (Davies, Rodríguez-Ferreiro, Suárez, & Cuetos, 2013; Jiménez et al., 2009; Ziegler et al., 2010). On the other hand, some studies have also found that RAN is a better long-term predictor of reading performance (e.g., reading accuracy, word recognition and/or reading comprehension) in transparent (Norwegian and Swedish: Furnes & Samuelsson, 2010) and opaque orthographies (English: Kirby, Parrila, & Pfeiffer, 2003), whereas PA appears to be most strongly related to the early stages of reading development.

The phonetic recoding to maintain information in working memory or VSTM refers to the ability to recode and maintain verbal information in a sound-based representational system. This ability is typically assessed by tasks that require the temporary storage of verbal items, such as digit span, words, pseudowords or nonwords repetition tasks. The temporary storage of material that has been read is dependent on working memory (Baddeley, 2003), which takes into account the storage of items for later retrieval and the demands of the partial storage of information related to several levels of text processing (Swanson, 1999). A large number of studies have found that children with DD perform significantly lower in VSTM tasks than typically developing children, which suggests that they have deficits at least in the phonological loop of Baddeley's working memory model (Everatt, Weeks, & Brooks, 2008; Kibby & Cohen, 2008; Moura, Simões, & Pereira, 2014).

Recent cross-linguistic studies have supported the hypothesis that PA is the best predictor of reading development in transparent and opaque orthographies in typically developing children (Caravolas et al., 2013; Furnes & Samuelsson, 2009; Vaessen et al., 2010; Ziegler et al., 2010). For example, Ziegler et al. (2010) found that PA was the main factor associated with reading accuracy and reading fluency across the five languages studied (Finnish, Hungarian, Dutch, Portuguese and French), and its impact was found to be modulated by the transparency of the orthography (PA is a stronger predictor in less transparent orthographies). The influence of RAN was limited to reading fluency, and VSTM showed some predictive value for reading accuracy only in Finnish and Hungarian orthographies. Note that, Ziegler et al. (2010) used sequential naming of pictured objects and there is evidence that alphanumeric RAN stimuli (e.g., letters or numbers) often lead to higher correlations with reading than do non-alphanumeric RAN stimuli (e.g., colors or objects) (Kirby et al., 2010). In this case, the use of a non-alphanumeric RAN stimulus may explain the atypically (low) relationship between RAN and reading. Similarly, Vaessen et al. (2010) confirmed that cognitive mechanisms underlying reading fluency of different word types were similar across the three alphabetic orthographies studied (Hungarian, Dutch and

Portuguese). The authors also found that the association of reading fluency with PA (but not with RAN or VSTM) was modulated by orthographic complexity and the contribution of PA decreased as a function of grade, whereas the contribution of RAN increased.

The same pattern has also been observed in DD samples. Ackerman and colleagues (2001) found that English-speaking children with DD performed significantly worse than typical readers in the PA and RAN tasks and that PA was the best predictor of reading decoding and word recognition. In a Dutch longitudinal study, Boets et al. (2010) also found that children with DD scored significantly lower than controls in the PA, RAN and VSTM tasks. They further demonstrated through hierarchical regression analyses that PA was more strongly related to reading accuracy and that RAN was more strongly related to reading fluency, whereas VSTM only contributed to a small proportion of the unique variance in reading accuracy. The results from a Portuguese study showed that children with DD scored significantly lower than typically developing children on PA and RAN and that PA predicted reading fluency for both groups, whereas RAN only predicted reading fluency for the DD group (Araújo et al., 2010).

Although the association between phonological processing and reading performance is very well documented in the literature, the diagnostic accuracy of phonological processing measures to correctly discriminate between children with DD and typical developing children is clearly less explored. Recently, Landerl et al. (2013) investigated the relationship between phonological processing and diagnostic accuracy in children with DD and CA (did not include a RL group) speaking six different languages spanning a large range of orthographic complexities (Finnish, Hungarian, German, Dutch, French, and English). They concluded that PA, RAN and VSTM were reliable predictors of DD status (odds ratio of 0.354, 0.356 and 0.694, respectively). They also found that PA and RAN were stronger concurrent predictors in complex (odds ratio of 0.187 and 0.262, respectively) than in less complex orthographies (odds ratio of 0.481 and 0.491, respectively), with an area-under-the-curve (AUC) of the predictive model of .817, .877 and .929 for low, medium and high orthographic complexity languages.

In summary, the extensive body of research with school-age children has shown that: (1) children with DD showed severe impairments in phonological processing; (2) PA and RAN tend to be the strongest predictors of reading in children with DD and typical readers (specific patterns can be observed as a function of the orthographic depth); and (3) PA is the best predictor of reading accuracy, whereas RAN is more related to reading fluency.

The level of orthographic consistency is the key factor determining the rate of reading acquisition across different languages and might influence how DD is manifested. Studying the subcomponents of reading across languages helps researchers to understand what factors are universal and which are language or orthography-specific factors in the reading system (Norton & Wolf, 2012). The few Portuguese studies that have explored the presence of phonological processing deficits in children with DD rarely included a RL group (some exceptions: Araújo et al., 2011; Sucena, Castro, & Seymour, 2009) or investigated the role of VSTM on reading performance (some exceptions: Moura et al., 2014; Silva, Silva, & Martins, 2014). Similarly, few studies have explored the accuracy of phonological processing measures to correctly discriminate between typical (CA and RL) and dyslexic readers (some exceptions: Landerl et al., 2013). Therefore, the present study has three main objectives: (1) to examine the presence of deficits in the phonological domain and in the reading performance of Portuguese-speaking children with DD; (2) to analyze the diagnostic accuracy of phonological processing measures to correctly discriminate between typical readers (CA and RL) and children with DD through a receiver operating characteristics (ROC) curve analysis and an abnormal low scores analysis; and (3) to determine the predictive effect of phonological processing on reading fluency and reading accuracy. Based on the existing literature, we expected that Portuguese children with DD would show significant impairments in all phonological processes and would reveal significant difficulties in reading fluency and accuracy (particularly in the reading of irregular words and pseudowords). We also expected that phonological processing would be an accurate measure for discriminating children with DD from CA and RL. Finally, we expected that PA would be the most significant predictor of reading accuracy in the Portuguese orthography, whereas RAN would be more related to reading fluency.

The European Portuguese orthography is considered to be an intermediate depth (Seymour, Aro, & Erskine, 2003; Sucena et al., 2009). Seymour et al. (2003) examined the beginning of reading acquisition in 13 European orthographies and found that children become fluent and accurate before the end of the first grade. The exceptions to this development pattern were English, French, Danish and Portuguese (the Portuguese and French orthographic code learning trajectories were quite similar). They 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 an opaque orthography (English children read correctly approximately 34% of words and 29% of non-words). They concluded that learning to read in the European Portuguese orthography proceeded less rapidly than in transparent orthographies, such as German, Greek, Italian or Finnish, but more rapidly than English.

Fernandes, Ventura, Querido, and Morais (2008) investigated the initial development of reading and spelling in the European Portuguese orthography and concluded that Portuguese children rely on grapheme–phoneme conversion at the initial stages of literacy acquisition [a regularity effect (i.e., the superiority of regular words over irregular words) was present in both reading and spelling by the middle of the first grade]. By the end of the first grade, the children had acquired some knowledge of the lexical orthographic representation [a lexicality effect (i.e., the superiority of words over pseudowords) was found in spelling]. Several orthographic and phonemic features concur which characterize European Portuguese orthography as an intermediate 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 et al. (2009, p. 794) stated “dyslexia in Portuguese should conform more to the English model than to the German model”. Indeed, previous Portuguese studies found a lexicality effect in typical and dyslexic readers (Araújo, Faísca, Bramão, Petersson, & Reis, 2014; Sucena et al., 2009), and a stronger contribution of PA to reading performance (Araújo et al., 2010; Sucena et al., 2009), which is more consistent with the results from less transparent orthographies. For additional information about the characteristics of the European Portuguese orthography, see: Albuquerque (2012), Fernandes et al. (2008) and Sucena et al. (2009).

Method

Participants

The participants were 72 Portuguese children with a mean age of 10.18 years (*SD* = 1.42). The DD group (*N* = 24; aged 10 to 12 years) included 79% male and 21% female, with a mean age of 11.04 years (*SD* = 0.86). The children with DD were in the 4^{th} to 6^{th} grades, and 36% were included in the special education system. The DD group was compared with two matched control groups: the CA and the RL. In the CA group (*N* = 24; aged 10 to 12 years), 67% were male and 33% were female, with a mean age of 11.00 years (*SD* = 0.83); the children were in the 4^{th} to 6^{th} grades. The CA group was matched for age $\chi^2(2) = 0.125$, $p = .939$, with the DD group, yielding non-significant differences in gender $\chi^2(1) = 0.949$, $p = .330$ and grade $\chi^2(2) = 2.427$, $p = .297$. The RL group (*N* = 24; aged 7 to 9 years) included 58% male and 42% female, with a mean age of 8.49 years (*SD* = 0.58); the children were in the 2^{nd}, 3^{rd} and 4^{th} grades. The RL group ($M = 59.27 \pm 8.95$) was matched on reading text fluency $t(46) = 0.577$, $p = .567$, $d = 0.16$, with the DD group ($M = 56.59 \pm 20.88$), yielding non-significant differences in gender $\chi^2(1) = 2.424$, $p = .119$. The RL group was matched with a reading text fluency measure ("O Rei"; Carvalho & Pereira, 2009) because in less opaque orthographies, the reading text accuracy has tended to reach a ceiling effect after the first years of school attendance (Seymour et al., 2003).

Inclusion criteria. For the three reading groups, only children who met the following criteria were included: (1) Wechsler Intelligence Scale for Children – Third Edition – Full Scale IQ (WISC-III FSIQ) ≥ 90; (2) native speakers of European Portuguese; (3) absence of a visual, hearing or motor handicap; (4) exclusion of a language impairment, emotional disturbance, dyscalculia, disruptive behavior disorder (attention deficit hyperactivity disorder, oppositional defiant disorder and conduct disorder), neurological impairment or other psychiatric disorders. For the CA and RL groups, the children with special educational needs were also excluded.

In the DD group, only children who were previously diagnosed with DD by a psychologist, child psychiatrist, developmental pediatrician or a child neurologist and who simultaneously had a score less than or equal to the 15^{th} percentile in a reading fluency and accuracy test administered during the testing session were included. These cutoff score criteria (WISC-III FSIQ ≥ 90 and both reading fluency and accuracy measures ≤ 15th percentile) are similar to, and in some cases stricter than, the inclusion criteria used in previous studies (e.g., Frijters et al., 2011; Reiter, Tucha, & Lange, 2005; Swanson, 1999, 2011). For the CA and RL groups, only children with a score greater than the 40th percentile on both reading measures were included.

Measures and Procedures

Intellectual Ability. The Portuguese version of the WISC-III (Wechsler, 2003) was administered to measure general intellectual ability. The WISC-III FSIQ scores (*M* = 100; *SD* = 15) were analyzed and used as a covariate in the inferential analysis. The factor structure of the Portuguese version of the WISC-III, analyzed through an exploratory and confirmatory factor analysis, yielded adequate psychometric properties for a two-factor model (Verbal IQ and Performance IQ) and for a three-factor model (Verbal Comprehension, Perceptual Organization and Processing Speed).

Phonological Awareness. The Phonological Awareness subtest of the Coimbra Neuropsychological Assessment Battery (BANC; Simões et al., in press) was used to assess PA and comprises two tasks. In the Deletion task (20 items), the child was asked to delete a particular phoneme on familiar words (e.g., say *sopa* [sopɐ] without the *se* [s]). In the Substitution task (20 items), the child was asked to replace one or more phonemes for other(s) phoneme(s) on familiar words (e.g., say *judo* [ʒudu] but replace the *je* [ʒ] to *xe* [ʃ]). For both PA tasks, the raw scores (number of correct responses) were converted to scaled scores (*M* = 10, *SD* = 3) based on age-specific norms. The reliability of the BANC normative sample for the Deletion task had a Cronbach's alpha = .91 and a test-retest = .83, whereas the Substitution task had a Cronbach's alpha = .90 and a test-retest = .85.

Naming Speed. The RAN (Numbers) task of the BANC was used to examine phonological access to lexical storage. 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 10x5 matrix. The raw scores (amount of time, in seconds, required to complete the task) were converted to scaled scores (*M* = 10, *SD* = 3) based on age-specific norms. The reliability of the BANC normative sample for the RAN task was obtained through test-retest (*r* = .78).

Verbal Short-Term Memory. The Forward task from the Digit Span (FDS) subtest of the WISC-III was used to assess VSTM. This task required that the child correctly repeat a series of digits in the order in which they were read to him/her. One point per trial (raw score) was given for a correct repetition. To control for the influence of age on the results of the FDS, an age-adjusted score was created by regressing the FDS onto age and then saving the unstandardized residual score (the Portuguese version of the WISC-III only provides age-scaled scores for the Digit Span subtest with both forward and backward tasks). The reliability (split-half) of the Digit Span subtest was .80.

Reading Text Fluency and Accuracy. The "O Rei" ("The King"; Carvalho & Pereira, 2009) is a three-minute reading test that measures the reading fluency (the number of correctly read words in one minute) and the reading accuracy (the percentage of correctly read words) of a Portuguese traditional tale for children from 1st to 6th grade. The test-retest from the normative sample was *r* = .94 for reading fluency and *r* = .80 for reading accuracy.

Reading Words. To assess the reading accuracy of individual words, we used the Oral Reading (PAL-PORT 22) subtest from the Portuguese version (Festas, Martins, & Leitão, 2007) of the Psycholinguistic Assessment of Language (PAL; Caplan, 1992). The PAL-PORT 22 comprises 146 words (48 regular, 47 irregular and 51 pseudowords). Based on previous studies that used the PAL-POR 22 with typically developing children, we selected 40 words: 16 regular (8 high-frequency and 8 low-frequency words; e.g., *sardinha* [sɐɾˈδiɲɐ], *rusga* [ˈʀuʒɣɐ]), 16 irregular (8 high-frequency and 8 low-frequency words; e.g., *fluxo* [ˈfluksu], *exotismo* [ezuˈtiʒmu]) and 8 pseudowords (e.g., *lempo* [ˈlẽpu], *glepal* [ɣlɛˈpał]). The percentage of correctly read words was calculated for the regular, irregular and pseudowords. The reliability (Cronbach's alpha) of the PAL-PORT 22 was .75.

The administration of these tests was included as part of a broad neuropsychological research that was also comprised of other measures (e.g., working memory, executive functions and others). Each child completed two individual sessions (separated by an interval of 10 to 15 days), which lasted approximately 90 minutes per session in a clinic or school setting during a weekday. All tests were administered in a fixed order. No incentives were offered in exchange for participation.

Statistical Analyses

Statistical analyses were performed using IBM SPSS Statistics 19. Group differences were analyzed using multivariate analyses of covariance (MANCOVA) with the WISC-III FSIQ as a covariate because significant group differences (CA: 107.25 $\pm$ 12.88; RL: 110.79 $\pm$ 12.47; DD = 96.67 $\pm$ 8.55) were observed, $F(2, 69) = 9.853$, $p < .001$, $\eta^2_p = .22$ (CA = RL > DD). If the multivariate analysis indicated a significant overall difference ($p < .05$), then a univariate test was applied to determine which dependent variables were responsible for the multivariate difference. Post hoc comparisons were conducted with the Bonferroni correction for multiple comparisons. In specific cases, repeated measures ANOVAs were also used. Partial eta-squared (η^2_p) were additionally calculated to determine the effect size of the differences between the groups.

A ROC curve analysis was performed to examine the accuracy of phonological processing measures to discriminate children with DD from CA and RL. A ROC curve analysis systematically sweeps across all possible true positive (sensitivity) and false positive (1-specificity) values of a diagnostic test. That is, sensitivity and specificity are determined for each cut-off point. The ROC curve analysis graphically illustrates the test's full range of diagnostic utility and can be used to calculate the AUC, which provides an accuracy index of the test (Fawcett, 2006). The more accurately a test is able to discriminate between groups, the more its ROC curve will deviate toward the upper left corner of the graph. The AUC is the average of the true positive rate, taken uniformly over all possible false positive rates (Krzanowski & Hand, 2009) that range between .5 and 1.0. An AUC value of 1.0 is perfectly accurate because the sensitivity is 1.0 when the false positive rate is .0, whereas an AUC value of .5 reflects a completely random classifier. An AUC of .5 to .7 indicates a low test accuracy, .7 to .9 a moderate accuracy and .9 to 1.0 a high accuracy (Swets, 1988).

Results

Correlational Analysis

Table 1 shows the Pearson correlation coefficients between general intellectual ability, phonological processing and reading measures. The WISC-III FSIQ showed small to moderate positive correlations with phonological processing and reading measures. Strong correlations were observed between PA tasks and RAN. In general, PA and RAN were highly correlated with reading.

Table 1. Pearson correlation coefficients between general intellectual ability, phonological processing and reading measures

	2	3	4	5	6	7	8	9	10
1. WISC-III FSIQ	.450**	.586**	.133	.343**	.185	.259*	.226	.284*	.284*
2. PA Deletion		.812**	.624**	.422**	.473**	.648**	.733**	.561**	.720**
3. PA Substitution			.558**	.468**	.547**	.629**	.652**	.623**	.648**
4. RAN				.382**	.557**	.656**	.596**	.507**	.536**
5. Forward Digit Span				.410**	.374**	.545**	.458**	.454**	.618**
6. Reading Fluency					.446**	.393**	.356**	.384**	.327**
7. Reading Accuracy						.601**	.587**	.657**	.449**
8. Regular Words							.702**	.694**	.683**
9. Irregular Words								.638**	.648**
10. Pseudowords									.571**

Note. $^{*}p < .05$, $^{**}p < .01$. WISC-III FSIQ = Wechsler Intelligence Scale for Children (Third Edition) Full Scale IQ. PA = phonological awareness. RAN = rapid automatized naming.

Phonological Processing: Group Differences

A MANCOVA was performed with phonological processes as dependent variables, reading group (CA, RL and DD) as fixed factor and WISC-III FSIQ as a covariate. The reading group had a significant main effect, $F(8, 130) = 13.865$, $p < .001$, Wilks' $\Lambda = .29$, $\eta^2_p = .46$. Univariate tests revealed that the children with DD scored significantly lower than the CA and the RL in the PA Deletion, $F(2, 68) = 49.458$, $p < .001$, $\eta^2_p = .59$; PA Substitution, $F(2, 68) = 30.140$, $p < .001$, $\eta^2_p = .47$; RAN, $F(2, 68) = 25.896$, $p < .001$, $\eta^2_p = .43$; and FDS, $F(2, 68) = 8.111$, $p < .01$, $\eta^2_p = .19$ (see Table 2).

Table 2. Means, standard deviations and post hoc comparisons of phonological processing and reading for children with developmental dyslexia and controls

	CA	RL	DD	Post hoc comparisons (Bonferroni)
	M ± *SD*	*M* ± *SD*	*M* ± *SD*	
Phonological Processing				
PA Deletion [a]	10.79 ± 1.86	10.26 ± 2.55	4.42 ± 1.76	CA = RL > DD
PA Substitution [a]	12.00 ± 2.82	9.95 ± 2.99	4.79 ± 2.58	CA > RL > DD
RAN [a]	11.63 ± 2.85	10.54 ± 2.63	6.12 ± 3.12	CA = RL > DD
Forward Digit Span [b]	0.84 ± 1.52	0.06 ± 1.03	-0.91 ± 1.16	CA = RL > DD
Reading Text				
Reading Fluency [c]	100.35 ± 27.10	59.27 ± 8.95	56.59 ± 20.88	CA > RL = DD
Reading Accuracy [d]	98.77 ± 0.75	97.29 ± 1.58	92.62 ± 6.25	CA = RL > DD
Reading Words				
Regular Words [d]	97.65 ± 4.04	88.28 ± 11.84	76.82 ± 13.09	CA > RL > DD
Irregular Words [d]	83.33 ± 10.37	72.91 ± 6.81	61.45 ± 15.27	CA > RL > DD
Pseudowords [d]	88.54 ± 9.69	89.58 ± 13.62	57.81 ± 20.79	CA = RL > DD

Note. [a] age-scaled score. [b] age-adjusted score (unstandardized residual score). [c] number of correctly read words in one minute. [d] percentage of correctly read words. PA = phonological awareness. RAN = rapid automatized naming. CA = chronological-age-matched controls. RL = reading-level-matched controls. DD = children with developmental dyslexia.

Reading: Group Differences

A MANCOVA with reading group (CA, RL and DD) as fixed factor and WISC-III FSIQ as a covariate showed statistically significant differences in reading text, $F(4, 134) = 19.820$, $p < .001$, Wilks' $\Lambda = .40$, $\eta^2_p = .37$, and in reading words, $F(6, 132) = 12.774$, $p < .001$, Wilks' $\Lambda = .40$, $\eta^2_p = .36$. The univariate statistics yielded a significant effect in text reading fluency, $F(2, 68) = 32.773$, $p < .001$, $\eta^2_p = .49$, and accuracy, $F(2, 68) = 13.897$, $p < .001$, $\eta^2_p = .29$, as well as in reading regular words, $F(2, 68) = 20.595$, $p < .001$, $\eta^2_p = .38$, irregular words, $F(2, 68) = 17.911$, $p < .001$, $\eta^2_p = .34$, and pseudowords, $F(2, 68) = 27.335$, $p < .001$, $\eta^2_p = .45$. As shown in Table 2, the CA outperformed the children with DD in all reading measures. Compared with the RL, the children with DD scored significantly lower in text reading accuracy and in reading regular, irregular and pseudowords but a non-significant difference was found in text reading fluency (as expected because this measure was used to match children with DD to RL).

In addition, we performed two repeated measures ANOVAs to analyze the presence of a lexicality effect (regular words > pseudowords) and a regularity effect (regular words > irregular words). A repeated measures ANOVA with lexicality effect (regular vs. pseudoword) as within-subjects factor and reading group (CA vs. RL vs. DD) as between-subjects factor yielded a significant main effect for lexicality, $F(1, 69) = 29.142$, $p < .001$, $\eta^2_p = .29$ and for the interaction between lexicality and reading group, $F(2, 69) = 12.537$, $p < .001$, $\eta^2_p = .26$. This main effect indicates that regular words were read more accurately than pseudowords, whereas the significant interaction occurred because the magnitude of the lexicality effect was stronger for the children with DD (19.01% advantage) than the CA (9.11% advantage) and the RL (-1.3% advantage). For the regularity effect, a repeated measures ANOVA contrasting reading groups (CA vs. RL vs. DD) revealed a significant effect for regularity, $F(1, 69) = 112.533$, $p < .001$, $\eta^2_p = .62$, but the interaction did not reach significance, $F(2, 69) = 0.060$, $p = .942$, $\eta^2_p = .00$. This main effect indicates that regular words were read more accurately than irregular words, whereas the non-significant interaction was because the magnitude of the regularity effect was homogeneous between the groups (CA = 14.32% advantage, RL = 15.37% advantage, and children with DD = 15.37% advantage).

Phonological Processing: Diagnostic Accuracy and Abnormally Low Scores

Although the results from the inferential analyses showed significant group differences in the phonological processing, it does not imply that PA, RAN and FDS tasks can correctly discriminate the children with DD from the CA and RL. Therefore, a ROC curve analysis was performed for the CA versus DD and the RL versus DD separately. The more accurately a task discriminates between the groups, the higher the AUC value. As shown in Table 3, all phonological processing measures were significant variables for discriminating between the subjects with a moderate to high diagnostic accuracy. The PA Deletion task revealed a higher level of accuracy to correctly discriminate the children with DD from the CA (AUC = .980) and the RL (AUC = .957). Thus, a randomly selected child with DD will have a lower score on the PA Deletion task approximately 98.0% and 95.7% of the time compared with a randomly selected child from the CA and the RL groups, respectively.

Table 3. Receiver operating characteristics (ROC) curve analysis

	CA vs. DD		RL vs. DD	
	AUC	*SE*	AUC	*SE*
PA Deletion	.980***	.019	.957***	.028
PA Substitution	.974***	.020	.906***	.042
RAN	.905***	.044	.858***	.053
Forward Digit Span	.831***	.058	.734**	.074

Note. $^{*}p < .05$, $^{**}p < .01$, $^{***}p < .001$. PA = phonological awareness. RAN = rapid automatized naming. CA = chronological-age-matched controls. RL = reading-level-matched controls. DD = children with developmental dyslexia. AUC = area under the curve. *SE* = standard error.

In addition, we computed a pairwise comparison of AUC values in order to analyze the presence of significant differences between PA, RAN and FDS. The comparison was performed using MedCalc 12.7. For the CA versus DD, a significant difference was observed for: PA Deletion > FDS (z = 2.615, p < .01) and PA Substitution > FDS (z = 2.504, p < .05). Similarly, for the RL versus DD, a significant difference was observed for: PA Deletion > FDS (z = 2.865, p < .01) and PA Substitution > FDS (z = 2.049, p < .05).

Analyzing the abnormally low scores in the PA Deletion, PA Substitution and RAN tasks, we found that 41.7% of the children with DD exhibited an age-scaled score < 7 (z < -1), and 16.7% of the children exhibited an age-scaled score < 4 (z < -2) in these three tasks simultaneously. No cases were identified in the CA and RL groups for either cutoff score. To determine the degree of abnormality of these profiles (these three subtests with a z < -1 or with a z < -2) in the normative population, we used the Crawford, Garthwaite and Gault (2007) method and software. Using the BANC standardization sample (N = 1104 children aged 5 to 15 years), we computed the estimated percentage of the healthy population that is expected to exhibit these abnormally low scores. Only 1.87% of the normative population exhibited an age-scaled score < 7, and only 0.02% exhibited an age-scaled score < 4 in these three subtests, which is in contrast to the higher percentage observed in our DD group.

Predictive Effect of Phonological Processing in Reading

To determine the predictive effect of phonological processing on reading ability, a series of hierarchical linear regression analyses were conducted for each of the dependent variable. For PA, a composite score was computed because the PA Deletion and the PA Substitution were highly correlated (r = .812, p < .001). The scores of all measures entered in the regression analysis were converted to z-scores to minimize the possible impact of different variable scaling. The predictive variables were entered in the following order: age (covariate) was entered into the first block, and PA, RAN and FDS were entered into the second block. Table 4 shows the variance (R^2 and ΔR^2) of the regression model, the standardized regression coefficient (β), the t-test and the squared part correlation (pr^2) for each of the four predictor variables (age, PA, RAN and FDS) on each reading task. The pr^2 represents the unique variance of each predictor when the overlapping linear effects of all other predictive variables were statistically removed.

Table 4. Hierarchical linear regression analyses for reading

Dependent Variable	Block	Predictors	R^2	ΔR^2	β	*t-test*	pr^2
Text Reading Fluency	1	Age	.152		.390	3.547**	.152
	2	PA	.712	.560	.352	3.946***	.067
		RAN			.440	5.151***	.114
		FDS			.113	1.486	.009
Text Reading Accuracy	1	Age	.007		.082	0.685	.006
	2	PA	.558	.551	.431	3.901***	.100
		RAN			.405	3.843***	.097
		FDS			.036	0.387	.001
Reading Regular Words	1	Age	.001		.024	0.199	.001
	2	PA	.614	.613	.628	6.084***	.213
		RAN			.287	2.915**	.048
		FDS			.049	0.555	.001
Reading Irregular Words	1	Age	.020		.143	1.210	.020
	2	PA	.519	.499	.533	4.627***	.153
		RAN			.242	2.203*	.034
		FDS			.042	0.424	.001
Reading Pseudowords	1	Age	.068		.260	2.256*	.067
	2	PA	.533	.465	.619	5.449***	.207
		RAN			.140	1.292	.011
		FDS			.017	0.176	.001

Note. $^{*}p < .05$, $^{**}p < .01$, $^{***}p < .001$. R^2 and ΔR^2 = variance explained. β = standardized regression coefficient. pr^2 = squared part correlation, represents the unique variance of each predictor. PA = phonological awareness. RAN = rapid automatized naming. FDS = Forward task from the Digit Span.

For text reading fluency, PA and RAN were significant predictors, with RAN showing the highest unique variance (explained 11.4% of the variance after controlling for age, PA and FDS) and the highest standardized regression coefficient, whereas the regression model explained 71.2% of the total variance. For text reading accuracy, only PA and RAN showed a significant predictive effect, with a unique variance of 10% and 9.7%, respectively, whereas the regression model explained 55.8% of the total variance. A large amount of shared variance was observed.

The results for the three reading words outcomes were very similar. Only PA exhibited significant standardized regression coefficients for all tasks and explained for more than 15% of the unique variance and RAN was a significant predictor for regular and irregular words. Non-significant standardized regression coefficients were found for FDS. The four predictor variables explained between 51.9% (irregular words) and 61.4% (regular words) of the total variance.

Discussion

There is extensive empirical evidence indicating that deficits in phonological processing are among the most prominent characteristics of children with DD, and it is also well-known that the level of orthographic consistency may influence how DD is manifested. The European Portuguese language is considered an orthography of intermediate depth (Sucena et al., 2009); it is more transparent than English, French and Danish, but less regular than Spanish, Italian, Greek, German and Finnish (Seymour et al., 2003).

The first aim of the present study was to investigate the specificity of phonological processing and reading deficits in Portuguese children with DD. The results from the inferential analyses revealed that the children with DD showed significantly lower scores than the CA and the RL in the PA Deletion, PA Substitution, RAN and FDS measures with very large effect sizes. These findings revealed that the ability to perceive and manipulate the sounds of spoken words, the rapid access of phonological information stored in the mental lexicon and the ability to code information phonologically for temporary storage in working memory were significantly impaired in the Portuguese children with DD, which is consistent with other studies from different orthographies (Boets et al., 2010; Everatt et al., 2008; Jiménez et al., 2009; Willburger, Fussenegger, Moll, Wood, & Landerl, 2008).

As expected, the Portuguese-speaking children with DD showed a severe impairment in all reading accuracy measures, suggesting a developmental deficit (CA and RL > DD). The children with DD exhibited specific difficulties in reading pseudowords (57.81% accuracy), which is consistent with studies from less transparent orthographies that have shown significant deficits in the phonological decoding strategy because grapheme-phoneme correspondence rules are considerably more complex. Indeed, studies with German-speaking (Wimmer, Mayringer, & Landerl, 2000) and Spanish-speaking (Davies et al., 2013) children have shown that the nonword reading accuracy in children with DD approaches normal performance, which is in contrast to studies with English-speaking children (for a review, see: Herrmann, Matyas, & Pratt, 2006). A lexicality effect and a regularity effect were also observed, that is, regular words were read more accurately than pseudowords and irregular words, respectively. The dual-route theories (Baron & Strawson, 1976; Coltheart, 1978, 2005) postulate two different ways in which readers can read differently written words: (1) the lexical route (also called the orthographic route) – regular and irregular words can be recognized directly by accessing a representation of their orthographic form in an internal lexicon; and (2) the sublexical route (also called the phonological route) – the reading of regular words and nonwords involves the use of grapheme-phoneme correspondence rules. During word recognition, these two processes work separately and simultaneously: the reading of irregular words requires accessing a lexicon or memory store of previously seen written words (the use of the sublexical route to read an irregular word yields a “regularization error”), the reading of nonwords requires the use of grapheme-phoneme correspondence rules, whereas for regular words, both lexical and sublexical routes generate the correct pronunciation (Castles, 2006; Coltheart, 2005; Cortese & Simpson, 2000). Thus, the reading of irregular words and pseudowords may be less accurate and may have a longer latency time than reading regular words.

When the magnitude of the lexicality and the regularity effects between children with DD and typical readers (CA and RL) were compared, we found that a significant difference occurred for the lexicality effect, but not for the regularity effect. These results may suggest that the phonological decoding strategy is particularly compromised in these Portuguese children with DD. As Ziegler and Goswami (2005, p. 18) stated "phonological rather than orthographic deficits therefore appear to underlie developmental dyslexia in all languages so far studied. Children with dyslexia are not worse than RL children in gaining orthographic access to whole words. Rather, they are worse at computing sublexical phonology". Concerning to regularity effect, mixed results were found in orthographies of intermediate depth. Sprenger-Charolles, Colé, Kipffer-Piquard, Pinton and Billard (2009) also found that the difference between regular and irregular words was not greater for French-speaking children with DD compared to RL. A Portuguese study with children with DD (3rd and 4th grades) showed a developmental delay (CA > DD with RL = DD) in phonological decoding (lexicality effect) and a developmental deficit (CA and RL > DD) in orthographic processing (regularity effect) (Sucena et al., 2009). Note that, in the Sucena et al.'s study a ceiling effect was found for regular words in CA, RL and DD groups (97.3%, 93.4% and 91.8%, respectively), which may explain the difference with our findings. Furthermore, Araújo et al. (2014) found evidence that Portuguese children with DD were not as flexible as CA in switching from phonological decoding (sublexical) strategies to orthographic (lexical) strategies.

Another purpose of the present study was to analyze the accuracy of phonological processing measures in discriminating children with DD from CA and RL. Whereas the presence of a significant impairment in phonological processing measures in the children with DD has been extensively reported in the literature, few studies have explored the accuracy of these measures in differentiating between typical and dyslexic readers. In a recent cross-linguistic study with six different languages, Landerl et al. (2013) found that PA and RAN were strong predictors of DD (predictive power increases with orthographic complexity), while VSTM played a minor role. Our results from the ROC curve analysis also showed a moderate accuracy for VSTM and a high accuracy for PA and RAN in discriminating the children with DD from the CA and the RL. These findings support the relevance of both PA and RAN measures in the diagnostic assessment of DD in an orthography of intermediate depth. Similar to the Landerl et al. (2013) study, earlier assumptions that RAN might be a more reliable marker of DD than PA in less opaque orthographies were not supported by the current study. Taken together, the results from the inferential analysis and the ROC curve analysis showed that PA is the most reliable marker of DD in Portuguese-speaking children, followed by RAN. On the other hand, a higher incidence of abnormally low scores in PA and RAN tasks was observed for the children with DD when compared with the controls and the normative population. These results reinforce the findings from the inferential analysis regarding the significant impairments of PA and naming speed in DD.

The final purpose was to analyze the predictive effect of phonological processing in reading ability. The results showed that PA was the most important predictor for all reading tasks (except for text reading fluency) and RAN was particularly related to text reading fluency. These findings are convergent with previous studies that found that PA is mainly related to decoding accuracy (Boets et al., 2010; Pennington et al., 2001), whereas RAN is an important predictor of reading fluency (Savage & Frederickson, 2005; Torppa, Georgiou, Salmi, Eklund, & Lyytinen, 2012; Vaessen et al., 2009) independent of the transparency of the orthography. In addition, we also found that RAN explained unique variance in the reading of regular and irregular words, but its contribution was not significant for pseudowords. As noted previously, orthographic processing (lexical route) occurs when words are processed as single units rather than as a sequence of grapheme-phoneme correspondence rules. Therefore, because of the greater involvement of orthographic processing in the reading of regular and irregular words, our findings may suggest that RAN is more related to orthographic processing. Indeed, several authors have found that RAN is strongly related to irregular word reading (rather than pseudoword) and reading fluency (rather than accuracy) supporting the hypothesis that RAN is more associated to orthographic processing (Bowers, 1995; Bowers & Ishaik, 2003; Bowers & Newby-Clark, 2002; for a review, see Kirby et al., 2010). Relatively inconsistent findings have been reported regarding the predictive effect of VSTM. As in our study, Ziegler et al. (2010) found that VSTM did not make a unique contribution to reading fluency and accuracy after controlling for PA and RAN in the Dutch, French and Portuguese subsamples. In contrast, some studies of children with DD and/or typical developing children found that VSTM contribute to a small proportion of the unique variance in reading accuracy (Boets et al., 2010) or word reading fluency (Landerl & Wimmer, 2008).

Notwithstanding the relevance of the present study, there are some limitations that should be addressed in future research. First, the inclusion of word reading latency time (or reaction time) measures is important because it has been hypothesized that latency time may be a more critical issue than reading accuracy in less opaque orthographies. Indeed, a ceiling effect was observed in the CA group in some reading accuracy measures, thus the additional inclusion of latency time measures would have been a better baseline to compare reading differences between groups. Second, some authors have suggested that less transparent orthographies would have a higher incidence of phonological dyslexia subtype (Castles & Coltheart, 1993; Jiménez et al., 2009; Sprenger-Charolles, Colé, Lacert, & Serniclaes, 2000). Children with the phonological dyslexia subtype revealed a selective deficit in the sublexical route and showed difficulties in the reading of nonwords (but not in irregular words), whereas children with the surface dyslexia subtype exhibited a selective deficit in the lexical route and showed difficulties in the reading of irregular words (but not in nonwords). Thus, it would also be particularly interesting to analyze the specific psycholinguistic characteristics of the phonological and surface dyslexia subtypes in the European Portuguese orthography and explore their prevalence.

In conclusion, phonological processing deficits were important characteristics of DD in Portuguese children. These results are consistent with the studies that indicated that PA is the most reliable marker of DD and the most important predictor of reading accuracy, whereas RAN was particularly related to text reading fluency, suggesting that the phonological processing role in reading ability may be relatively universal (at least in alphabetic languages).

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Estudo 3

Executive Functioning in Children with Developmental Dyslexia

Executive Functioning in Children with Developmental Dyslexia

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Abstract

The term executive function has been used to describe several higher-order cognitive processes. This study examined the processing speed, shifting, planning, and verbal fluency of a sample of 50 Portuguese children with developmental dyslexia (DD) and 50 typically developing children (TDC; chronological-age-matched controls) between 8 and 12 years of age to evaluate the children’s executive functioning. Compared to TDC, children with DD revealed significant processing speed, shifting, and verbal fluency deficits. After controlling for differences in the general intellectual ability, significant group differences remained for shifting, verbal fluency and marginally for processing speed. No significant differences in planning ability were observed between the groups. No significant interaction of group, gender, and age was found for any of the executive functions measures studied. Word productivity in both semantic and phonemic verbal fluency tasks decreased significantly over the 60 seconds for both groups. Shifting was the only significant predictor of DD in the binary logistic regression analysis and yielded the highest area under the curve value (receiver operating characteristics curve analysis). Therefore, although these findings highlight the presence of specific executive functions deficits in children with DD, they should not be interpreted as indicative of the presence or absence of this learning disorder.

Keywords: Processing speed, planning, shifting, verbal fluency, developmental dyslexia.

Introduction

Developmental dyslexia (DD) is a specific learning disability that is neurobiological in origin and characterized by difficulty with accurate and/or fluent word recognition and by poor spelling and decoding abilities. These traits typically result from a phonological deficit (Lyon, Shaywitz, & Shaywitz, 2003) and are not a consequence of sensory impairments, low intelligence or a lack of educational opportunities (American Psychiatric Association, 2013).

A large number of studies have supported the hypothesis that phonological processing is the most relevant neurocognitive phenotype of DD in opaque and transparent orthographies (Landerl et al., 2013; Ramus, Marshall, Rosen, & van der Lely, 2013). Although deficits are most pronounced in measures of phonological processing, other studies suggest that individuals with DD also have weaknesses in other neurocognitive domains. Traditionally, neuropsychological models of neurodevelopmental disorders have typically proposed that a single primary neurocognitive deficit was sufficient to explain all of the symptoms observed for a disorder. Recently, some researchers have challenged the validity of single-deficit models and suggested the presence of a multiple cognitive deficit model for understanding "complex" neurodevelopmental disorders, such as DD, attention-deficit hyperactivity disorder (ADHD), dyscalculia, and other disorders (Pennington, 2006; Willcutt et al., 2013; Willcutt, Sonuga-Barke, Nigg, & Sergeant, 2008). For instance, Willcutt, Pennington, Olson, Chhabildas, and Hulslander (2005) found evidence of a cognitive overlap between

DD and ADHD, in which both neurodevelopmental disorders were associated with weaknesses on most cognitive measures [more pronounced in measures of processing speed (PS)]. Similarly, shared neuropsychological weaknesses were observed between children with DD and dyscalculia (Willcutt et al., 2013).

Therefore, impairment in executive functions (EF) is ubiquitous across neurodevelopmental disorders, although distinct profiles emerge from various aspects of EF (Willcutt et al., 2008). Many studies have consistently found that children with DD exhibit weaknesses on a range of EF measures (Altemeier, Abbott, & Berninger, 2008; Brosnan et al., 2002; Helland & Asbjørnsen, 2000; Moura, Simões, & Pereira, 2014b; Reiter, Tucha, & Lange, 2005; Varvara, Varuzza, Sorrentino, Vicari, & Menghini, 2014), which are not simply secondary consequence of a deficit in another domain (Willcutt et al., 2008). Nonetheless, the literature has been discordant concerning which executive processes are compromised in DD. Therefore, the present study examined the presence of specific deficits in the executive functioning of children with DD who were native speakers of an orthography of intermediate depth (European Portuguese orthography). We also investigated the diagnostic accuracy of EF measures to correctly discriminate between typically developing children (TDC) and children with DD.

EF is a shorthand description of a complex set of processes associated with the metacognitive capacities that allow an individual to perceive stimuli in his or her environment, respond adaptively, flexibly change direction, anticipate future goals, consider consequences, and respond in an integrated way (Baron, 2004). Studies of brain-damaged patients and neuroimaging studies have located EF in the frontal (particularly the prefrontal cortex) and parietal lobes (Collette, Hogge, Salmon, & Van der Linden, 2006; Demakis, 2004; Wager & Smith, 2003). For example, the ability to maintain verbal information in working memory has been found to rely primarily on the lateral prefrontal cortex (Narayanan et al., 2005), switching ability has been associated with the medial prefrontal cortex and posterior parietal cortex (Collette et al., 2006; Crone, Wendelken, Donohue, & Bunge, 2006), the ability to inhibit responses was found to rely on the right inferior frontal cortex (Aron, Robbins, & Poldrack, 2004), and updating was associated with cerebral activity in the prefrontal (dorsolateral, inferior and cingulate) and parietal (posterior and superior) areas (Collette et al., 2006). Although the frontal and parietal lobes play an important role in the mediation of EF, researchers also agree that the integrity of the entire brain is necessary for efficient executive functioning (Stuss & Alexander, 2000; Tamnes et al., 2010).

So, the current conceptualizations support the idea of a fronto-parietal network supporting executive processes, which is relevant in light of the recent findings about the involvement of frontal and parietal areas in DD (Bloom, Garcia-Barrera, Miller, Miller, & Hynd, 2013; Boets et al., 2013). Reading development requires the coordination of many aspects of cognition; therefore, it is not surprising that early reading skills (Foy & Mann, 2013), reading comprehension (Borella & de Ribaupierre, 2014; Sesma, Mahone, Levine, Eason, & Cutting, 2009) and reading decoding (Altemeier et al., 2008; Bental & Tirosh, 2007) have been associated with specific executive processes, particularly working memory, inhibition and shifting. For example, working memory plays an important role in reading comprehension because it enables readers to process and access text information to build a coherent representation of the text's meaning. Cognitive inhibition has also frequently been considered in reading comprehension to contribute to selecting of relevant items, to enable individuals to form a coherent representation of the text (Borella & de Ribaupierre, 2014).

Neurodevelopmental studies have shown that executive functioning emerges in early childhood, develops significantly throughout childhood and adolescence, and that adult-level performance on the most complex EF tasks does not occur until adolescence or even early adulthood (V. Anderson, Anderson, Northam, Jacobs, & Catroppa, 2001; Best & Miller, 2010; Davidson, Amso, Anderson, & Diamond, 2006). Indeed, executive processes are subject to distinct developmental trajectories. Anderson (2002) found that attentional control appears to emerge in infancy and develops rapidly in early childhood, whereas cognitive flexibility, goal setting, and information processing experience a critical period of development between 7 and 9 years of age, and are relatively mature by 12 years of age. Additionally, working memory capacity has been found to gradually develop throughout childhood and into young-adulthood, shifting attained mature levels during adolescence (Huizinga, Dolan, & van der Molen, 2006), whereas inhibition was found to reach adult-level performance in late childhood or adolescence (Bedard et al., 2002; van den Wildenberg & van der Molen, 2004). These findings about the influence of age on EF task performance have also been supported by neuroimaging studies examining the maturation of frontal lobe (Blakemore & Choudhury, 2006; Tamnes et al., 2010). Studies about the influence of gender differences on EF task performance have reported inconsistent findings. Though some studies have indicated that boys and girls develop executive functioning in similar ways during childhood (Davidson et al., 2006; Marzocchi et al., 2008), others have observed gender differences on specific tasks (V. Anderson et al., 2001; Rosselli, Ardila, Bateman, & Guzman, 2001). These differences may be related to gender-specific differences in brain development (De Bellis et al., 2001; Giedd et al., 1996).

Despite its wide acceptance, conceptually defining EF has been difficult. There is no consensus among researchers about the executive components involved (for a review, see Chan, Shum, Toulopoulou, & Chen, 2008; Jurado & Rosselli, 2007; Wasserman & Wasserman, 2013). Some researchers have conceptualized EF as a single construct (Sala, Gray, Spinnler, & Trivelli, 1998), but others view it as comprising multiple process-related systems (Alexander & Stuss, 2000). As Anderson (2002, p. 73) stated, "this latter framework is probably more accurate given that global executive impairment is rare, specific executive processes are thought to be associated with distinct frontal systems, and executive processes demonstrate variable developmental profiles". Factor analytic-studies have identified multiple EF components. For instance, Welsh, et al. (1991) identified three factors reflecting speeded responding, set maintenance, and planning. Miyake et al. (2000) examined three often-postulated aspects of EF (shifting, inhibition, and updating) through a confirmatory factor analysis and found that, although they are distinguishable, they share some underlying commonality. Anderson (2002) proposed four distinct domains: attentional control, information processing, cognitive flexibility, and goal setting.

Another problem affecting the measurement of executive functioning is the "task impurity problem" (Miyake et al., 2000; van der Sluis, de Jong, & van der Leij, 2007). EF regulates other cognitive processes, and assessing them requires other non-executive cognitive abilities to be considered (e.g., verbal and visual-spatial abilities, motor speed, or attention). Furthermore, executive tasks often require more than one executive function and the intercorrelations among EF tasks are low to moderate (Lehto, Juujärvi, Kooistra, & Pulkkinen, 2003; Miyake et al., 2000).

Despite these methodological issues, there exists a relative agreement in terms of the complexity and importance of executive functioning to human adaptive behavior. EF measures are widely used in clinical neuropsychological assessment and typically include (but are not limited to) PS, planning, shifting, verbal fluency (VF), inhibition, updating, divided attention, and working memory tasks. PS, shifting, planning, and VF tasks are the measures most often used in studies of children with DD (Brosnan et al., 2002; Reiter et al., 2005; Shanahan et al., 2006) and in clinical evaluations. Therefore, these four tasks were used to explore EF deficits in Portuguese-speaking children with DD in the present study.

Processing Speed

PS is the ability to automatically and fluently perform relatively easy or over-learned elementary cognitive tasks, especially when high mental efficiency is required (McGrew, 2009). The Coding and Symbol Search subtests from the Processing Speed Index of the Wechsler Intelligence Scale for Children (WISC) are two of the most common tasks used to measure PS performance among children and adolescents. These subtests also measure visual-motor coordination, scanning ability and visual perception (Kaufman & Lichtenberger, 2000; Martins, Maruta, Freitas, & Mares, 2013).

Several studies have found that children with DD showed deficits on both WISC-III Processing Speed Index subtests (Moura, Simões, & Pereira, 2014a; Thomson, 2003). Shanahan et al. (2006) performed a detailed study that examined the presence of PS deficits in children and adolescents with DD and ADHD using a wide range of PS tasks. The results suggested that, compared to TDC, a general PS deficit exists in both clinical groups but that children with DD showed greater PS deficits than children with ADHD. Likewise, Willcutt et al. (2005) also found that children with DD or ADHD performed worse than TDC on five PS tasks. More recently, Peng, Sha, and Li (2013) also observed that TDC outperformed children with DD on all PS tasks in a sample of Chinese children.

Catts, Gillispie, Leonard, Kail, and Miller (2002) found that PS explained unique variance in reading comprehension and word recognition even when Full Scale IQ (FSIQ) and rapid naming were introduced into the regression model first. Rapid naming, however, did not predict the additional variance in these two reading measures after FSIQ and PS were taken into account. The authors hypothesized that PS deficit may be an extra-phonological factor in some reading disabilities.

Shifting

Shifting has been conceptualized as the ability to flexibly switch between multiple tasks, strategies, or mental sets (Miyake et al., 2000; van der Sluis, de Jong, & van der Leij, 2004; van der Sluis et al., 2007). Miyake et al. (2000) suggested that shifting is a basic underlying component of executive functioning, which is implicated in the performance of more complex executive tasks. A recent meta-analytic study found that shifting was significantly associated with children's performance in both reading and math (Yeniad, Malda, Mesman, van Ijzendoorn, & Pieper, 2013). While some studies have observed that children with DD have difficulty performing tasks that rely on shifting (Helland & Asbjørnsen, 2000; Horowitz-Kraus, 2012; Menghini et al., 2010), others did not find significant differences between children with DD and TDC (Bental & Tirosh, 2007; Reiter et al., 2005).

The Wisconsin Card Sorting Test (WCST) and the Trail Making Test (TMT) – Part B are often used to measure shifting ability. Willcutt and colleagues (2005) found that children with DD scored significantly lower than controls on TMT-B and WCST perseverative errors scores. These main effects did not remain significant after controlling for FSIQ, suggesting that shifting difficulties associated with DD may be explained by group differences in general intelligence. Other studies that used the WCST revealed that individuals with DD committed more perseverative errors (Marzocchi et al., 2008) and more non-perseverative errors (Helland & Asbjørnsen, 2000) and completed fewer categories (Helland & Asbjørnsen, 2000; Menghini et al., 2010) than typically developing individuals. Narhi and colleagues (1997) found that children with DD performed worse on the TMT-B but not on the TMT-A than TDC. They hypothesized that the poorer performance of children with DD on the TMT-B might reflect the difficulty those with DD have in following the alphabetical series. In the studies of both Reiter et al. (2005) and van der Sluis et al. (2004), the results of TMT-B showed non-significant differences between children with DD and chronological-age controls.

Planning

Planning ability is one of the major aspects of executive functioning and has been described as the ability to identify and organize the steps and elements that are required to achieve a goal (Lezak, Howieson, & Loring, 2004). In clinical neuropsychology, planning ability is assessed most often using the Tower of London (ToL) and the Tower of Hanoi (ToH) tests or one of their variants.

Studies testing the planning ability of children with DD have yielded inconsistent findings. Condor, Anderson, and Saling (1995) found that young TDC require significantly fewer trials to reach a successful solution to five-problem variations of the ToH than children with DD, but no significant differences were observed among older children. DD and typical readers did not obtain significantly different scores for number of errors, initial thinking time, or subsequent thinking time in Brosnan et al.'s (2002) study. Reiter et al. (2005) used the ToL to measure differences in planning abilities between children with DD and TDC. They found that the groups did not differ in the number of problems solved but that the planning time was significantly longer in the DD group. Marzocchi et al. (2008), who also used the ToL, did not find significant group differences in total score, planning time, or execution time.

Verbal Fluency

VF tests require participants to retrieve words based on semantic (subjects should produce as many different words as possible within a particular semantic category, e.g., animals, food, names) and phonemic (subjects should produce as many different words as possible that begin with a particular letter, e.g., the letters F, A, or S) criteria within a time constraint (Lezak et al., 2004). VF tests have been used to measure specific aspects of EF, memory, and language. Several neuroimaging studies have suggested that although both semantic and phonemic fluency tasks are associated with frontal and temporal lobe processes, phonemic tasks are more dependent on the frontal lobe and semantic tasks on the temporal lobe (Baldo, Schwartz, Wilkins, & Dronkers, 2006; Birn et al., 2010).

Empirical research has shown that children with DD generate significantly fewer words than TDC in phonemic VF tasks; for semantic VF tasks, however, inconsistent findings have been reported (Landerl, Fussenegger, Moll, & Willburger, 2009; Marzocchi et al., 2008; Reiter et al., 2005). Cohen and colleagues (1999) found that phonemic VF tasks were clinically useful in differentiating two subgroups of children with DD (dysphonetic and dyseidetic) and that the performance of dysphonetic children was significantly lower than that of children with ADHD. Furthermore, semantic VF tasks have been shown to be easier than phonemic VF tasks for TDC (Filippetti & Allegri, 2011; Martins, Vieira, Loureiro, & Santos, 2007; Moura, Simões, & Pereira, 2013; Riva, Nichelli, & Devoti, 2000) and for children with DD (Reiter et al., 2005; Varvara et al., 2014). Researchers have hypothesized that semantic tasks are easier because phonemic tasks depend more on the maturation of the frontal lobe; to retrieve words beginning with a letter, an individual must explore more category subsets than is required to retrieve words within a semantic category (Riva et al., 2000).

Troyer (2000) and Hurks et al. (2004; 2006) argued that the total number of words an individual can generate in 60 seconds does not provide sufficient information about the specific cognitive mechanisms that underlie poor performance on VF tasks. They suggested other scoring methods that measured (i) word productivity as a function of time and/or (ii) systematic organization of information, such as clustering (i.e., the production of two or more words within the same semantic or phonemic subcategory) and switching (i.e., the ability to shift between subcategories). The few studies that have analyzed word productivity as a function of time in children found that word production decreased significantly over time (Filippetti & Allegri, 2011; Hurks, 2012; Hurks et al., 2006; Moura et al., 2013; Takács, Kóbor, Tárnok, & Csépe, 2014). No studies have analyzed children with DD. Using the second alternative scoring method, Troyer et al. (2000; Troyer, Moskovitch, & Winocur, 1997) and other authors (Hurks, 2012; Unsworth, Spillers, & Brewer, 2010) demonstrated that clustering and switching are dissociable components of VF performance. Both skills were equally important in semantic VF tasks, but switching made a greater contribution to phonemic VF than did clustering, possibly because switching is more related to frontal lobe functioning.

Although an increasing number of studies about EF in DD have been published recently, inconsistent findings have been obtained. Therefore, the present study has two main objectives: (i) to examine the presence of specific deficits in the executive functioning of Portuguese-speaking children with DD and (ii) to analyze the ability of four different EF measures (PS, shifting, planning, and VF) to accurately discriminate between children with DD and TDC. Based on the existing literature from different languages spanning a large range of orthographic complexities (e.g., Norwegian: Helland & Asbjørnsen, 2000; Italian: Marzocchi et al., 2008; German: Reiter et al., 2005; English: Willcutt et al., 2005), we expected that Portuguese-speaking children with DD would show significant impairment in the EF measures. We also expected that EF tasks would be accurate measures for distinguished children with DD from TDC. Currently, no studies have analyzed the diagnostic accuracy of EF measures for discriminating between subjects (DD vs. TDC) or have analyzed the executive functioning in Portuguese-speaking children with DD (the European Portuguese orthography is considered to be an intermediate depth). The large body of research about EF deficits in DD has been conducted in English-speaking samples (opaque orthography).

Method

Participants

The participants included 100 Portuguese children between the ages of 8 and 12 (M = 9.81; SD = 1.34) in grades 3 through 6. In the DD group (N = 50), 74% were male and 26% were female, with a mean age of 9.80 years (SD = 1.38). Among the children with DD, 26% had undergone school retention, 36% were included in special education system, and 30% had relatives with reading difficulties. In the TDC group (N = 50), 64% were male and 36% were female, with a mean age of 9.82 years (SD = 1.32). Only 2% had experienced school retention, and 4% had relatives with reading difficulties. The children in the DD and TDC groups were matched for age $\chi^2(4) = 0.487$, $p = .975$, yielding non-significant differences in gender $\chi^2(1) = 1.169$, $p = .387$ and grade $\chi^2(3) = 1.776$, $p = .620$.

Criteria for Inclusion. For both groups, only children who met the following criteria were included in the study: (i) WISC-III FSIQ $\geq$ 90; (ii) native speakers of European Portuguese; (iii) at least two years of school attendance; (iv) absence of a visual, hearing, or motor handicap; (v) never diagnosed with a language impairment, emotional disturbance, dyscalculia, disruptive behavior disorder (ADHD, oppositional defiant disorder, and conduct disorder), neurological impairment, or other psychiatric disorder. Children with special educational needs were excluded from the TDC group.

All subjects attended regular classes in public and private Portuguese schools. Children with DD were recruited for participation through contact with school psychologists and special education teachers, and referrals from the medical, psychological and other educational/clinical professions (e.g., teachers and speech therapists). The TDC group was recruited through contact with teachers, parents and other participants using a snowball sampling strategy. In the DD group, only children who had previously been diagnosed with DD by a psychologist, child psychiatrist, developmental pediatrician, or child neurologist and had received a score lower than or equal to the 15^{th} percentile on a reading fluency and accuracy measure ("O Rei"; Carvalho & Pereira, 2009) administered during the testing session were included. These cut-off criteria (WISC-III FSIQ $\geq$ 90 and reading fluency and accuracy measures $\leq 15^{th}$ percentile) are similar (and in some cases stricter than) the inclusion criteria used by other studies (e.g., Bental & Tirosh, 2007; Frijters et al., 2011; Gooch, Snowling, & Hulme, 2011; Reiter et al., 2005; Swanson, 2011). For the TDC group, only children with a score greater than the 40th percentile on both reading measures were included.

Measures and Procedures

Intellectual Ability. The Portuguese version of the WISC-III (Wechsler, 2003) was administered to measure general intellectual ability. The General Ability Index (GAI) scores were analyzed and used as a covariate in the inferential analysis. The WISC-III GAI is a composite score, which is derived from the four Verbal Comprehension Index subtests and the four Perceptual Organization Index subtests (Prifitera, Weiss, & Saklofske, 1998). We used GAI (rather than FSIQ) because it excludes subtests that are related to EF (i.e., PS and working memory). As suggested by Saklofske, Prifitera, Weiss, Rolfhus, and Zhu (2005), in some special educational cases (e.g., children with learning disability and ADHD), the GAI may be a slightly higher estimate of overall intellectual ability than the FSIQ.

Processing Speed. The Coding and Symbol Search subtests from the WISC-III and the Trail-A test from the Coimbra Neuropsychological Assessment Battery (BANC; Simões et al., in press) were used to measure PS. The Coding (Form B) subtest requires that the child rapidly copy (in two minutes) nine types of symbols, each paired with a number, using a key provided at the top of the page. The Symbol Search (Form B) subtest requires that the child match a specific symbol to an identical target that is displayed among several distracter stimuli. This test also lasts for two minutes. Age-scaled scores (M = 10, SD = 3) from the Portuguese version of the WISC–III (Wechsler, 2003) were used for both tasks. The Trail-A test requires the child to draw a line sequentially connecting 25 encircled numbers (1 through 25) randomly distributed on a sheet of paper (similar to the TMT-A). The raw score of the Trail-A test represents the amount of time (in seconds) taken to complete the task.

Shifting. The Trail-B test from the BANC (Simões et al., in press) was administered to examine participants' shifting ability (similar to the TMT-B). This test requires the child to draw a line connecting 25 circles containing numbers or letters randomly distributed on a sheet of paper, alternating between numbers and letters (1, A, 2, B, etc.). The Trail-B is more complex than the Trail-A because it makes greater demands on an individual's rapid visual scanning and visuospatial sequencing capacities and involves cognitive shifting, flexibility, and divided attention. The raw score of the Trail-B represents the amount of time (in seconds) taken to complete the task.

Planning. The Tower test from the BANC (Simões et al., in press) was used to assess planning and problem solving abilities (similar to the ToL). The test comprises 14 models that the child is asked to reproduce by creating a tower using three balls of different colors (red, blue, and green) and three pegs (large, medium, and small). The child must move the three colored balls to specific positions on the three pegs in a specific number of moves (starting with one move and gradually increasing to five moves). The child has four trials in which to correctly solve each of the 14 models. Three raw scores were analyzed: Correct First Trials (i.e., the total number of models correctly solved on the first trial; range = 0-14), Correct Models (i.e., the total number of models correctly solved; range = 0-14), and Total Trials (i.e., the total number of trials taken to solve the 14 models; range = 14-56).

Verbal Fluency. The Semantic and Phonemic Verbal Fluency test from the BANC (Simões et al., in press) comprises three semantic (Animals, Names, and Food) and three phonemic (letters P, M, and R) tasks. VF tests have been used extensively in neuropsychological assessments to measure executive functioning, executive aspects of language processing, and semantic memory. For each of the semantic and phonemic tasks, the child was asked to generate as many words as possible within a time constraint of 60 seconds. The raw score was the total number of correct words (different forms of the same word were excluded) generated within the time limit for the three semantic or phonemic tasks. Additionally, to analyze word productivity as a function of time, the number of words generated by the child were recorded over four time intervals (0-15 seconds, 16-30 seconds, 31-45 seconds, and 46-60 seconds), as recommended by Hurks et al. (2004; 2006).

The administration of these tasks was part of a broad neuropsychological protocol that also included measures of intelligence, memory, attention, language, reading, and spelling. The children were tested in two sessions separated by a 10- to 15-day interval. The sessions were approximately 90-minutes long and took place in a clinic or school setting during a regular day.

Statistical Analyses

The statistical analyses were performed using IBM SPSS Statistics 19. Group differences were analyzed using a multi-factorial multivariate analysis of variance (MANOVA) and covariance (MANCOVA). Group, gender, and age were included as fixed factors, and the executive functions measures were used as dependent variables. If the multivariate analysis (Pillai's trace) indicated a significant overall difference ($p < .05$), then a univariate test was applied to determine which dependent variables were responsible for the multivariate difference. In specific cases, univariate analysis of covariance (ANCOVA), repeated measures ANOVAs and independent- and paired-samples t-tests were also used. Cohen's *d* or partial eta-squared (η^2_p) was also calculated to determine the effect size of the differences between groups.

A receiver operating characteristics (ROC) curve and binary logistic regression analysis were also performed to examine the accuracy with which EF tasks were able to discriminate between children in the DD and TDC groups. A ROC curve analysis systematically sweeps across all possible true positive (sensitivity) and false positive (1-specificity) values of a diagnostic test and calculates the area under the curve (AUC), which provides an accuracy index of the test (Fawcett, 2006). An AUC of .5 to .7 indicates low test accuracy, .7 to .9 moderate accuracy, and .9 to 1.0 high accuracy (Swets, 1988). For the binary logistic regression analysis, the fit of the model (Hosmer-Lemeshow test, Cox and Snell R^2, and Nagelkerke R^2) and the statistical tests of individual predictors were analyzed (regression coefficient, Wald's χ^2, and odds ratio).

Results

Processing Speed

A 2 X 2 X 5 (group X gender X age) MANOVA was performed and a significant main effect was observed for group, $F(3, 78) = 4.073$, $p = .010$, $\eta^2_p = .135$. The univariate analysis revealed significant effects in Coding, $F(1, 80) = 4.823$, $p = .031$, $\eta^2_p = .057$), Symbol Search, $F(1, 80) = 7.269$, $p = .009$, $\eta^2_p = .083$), and Trail-A, $F(1, 80) = 6.274$, $p = .014$; $\eta^2_p = .073$). Children with DD scored significantly lower than TDC (see Table 1).

No significant group X gender, $F(3, 78) = 0.330$, $p = .804$, $\eta^2_p = .013$, group X age, $F(12, 240) = 0.824$, $p = .625$, $\eta^2_p = .040$, or group X gender X age interactions, $F(9, 240) = 0.604$, $p = .793$, $\eta^2_p = .022$, were found.

Shifting

A 2 X 2 X 5 (group X gender X age) ANCOVA was performed with Trail-B as a dependent variable and Trail-A as a covariate in order to "isolate" the shifting effect on the Trail-B. A significant main effect for group was observed, $F(1, 80) = 10.371$, $p = .002$, $\eta^2_p = .115$. Children with DD took more time than TDC to complete the Trail-B (see Table 1). No significant interactions were observed for group X gender, $F(1, 80) = 0.004$, $p = .953$, $\eta^2_p < .001$, group X age, $F(4, 80) = 0.559$, $p = .693$, $\eta^2_p = .027$, or group X gender X age, $F(3, 80) = 0.149$, $p = .930$, $\eta^2_p = .006$.

Table 1. Means and standard deviations of executive functions for typically developing children and children with developmental dyslexia

	Typically Developing Children								Children with Developmental Dyslexia							
	Total	Gender		Age					Total	Gender		Age				
		Male	Female	8	9	10	11	12		Male	Female	8	9	10	11	12
Processing Speed																
Coding^{ss}	11.12 (2.70)	10.91 (2.64)	11.50 (2.85)	12.57 (1.61)	10.74 (2.42)	12.13 (3.04)	11.00 (3.11)	9.88 (3.09)	9.61 (2.45)	9.33 (2.57)	10.38 (1.98)	9.63 (3.24)	10.12 (2.64)	8.88 (1.45)	10.29 (1.97)	8.78 (2.43)
Symbol Search^{ss}	10.96 (3.12)	10.94 (3.41)	11.00 (2.61)	12.71 (3.63)	10.95 (2.36)	12.00 (3.46)	9.50 (3.50)	9.88 (3.18)	9.31 (2.64)	9.42 (2.41)	9.00 (3.29)	10.50 (2.97)	10.06 (2.30)	8.13 (2.35)	7.57 (3.59)	9.22 (1.56)
Trail-A	37.14 (16.00)	38.06 (17.49)	35.50 (13.26)	47.29 (11.77)	44.26 (19.64)	30.12 (3.39)	26.75 (5.44)	28.75 (11.84)	43.28 (14.39)	44.59 (14.73)	39.54 (13.17)	53.89 (12.98)	43.71 (15.43)	47.50 (14.56)	38.43 (9.48)	31.89 (7.70)
Shifting																
Trail-B	91.12 (31.53)	94.78 (33.14)	84.61 (28.16)	97.86 (11.48)	100.00 (33.59)	84.75 (36.27)	84.13 (29.50)	77.50 (34.46)	120.12 (40.28)	124.32 (42.40)	108.15 (31.95)	149.78 (35.22)	123.24 (44.35)	123.75 (17.10)	124.86 (39.39)	77.67 (16.57)
Planning (Tower)																
Correct 1st Trials	9.76 (1.72)	9.91 (1.87)	9.50 (1.42)	9.71 (1.38)	9.42 (1.50)	8.50 (1.85)	10.88 (1.88)	10.75 (1.28)	9.22 (1.63)	9.24 (1.63)	9.15 (1.72)	9.00 (1.80)	8.76 (1.34)	9.50 (1.85)	9.14 (1.34)	10.11 (1.90)
Correct Models	13.64 (0.56)	13.63 (0.55)	13.67 (0.59)	13.57 (0.78)	13.63 (0.49)	13.50 (0.75)	13.88 (0.35)	13.63 (0.51)	13.54 (0.64)	13.59 (0.55)	13.38 (0.87)	12.89 (0.92)	13.53 (0.51)	14.00 (0.01)	13.57 (0.53)	13.78 (0.44)
Total Trials	20.60 (3.25)	20.31 (3.15)	21.11 (3.44)	20.71 (2.92)	21.42 (2.91)	22.38 (3.88)	18.50 (2.87)	18.88 (2.74)	21.54 (3.07)	21.62 (3.22)	21.31 (2.72)	23.22 (3.52)	21.94 (2.27)	20.63 (3.54)	21.57 (2.87)	19.89 (3.25)
Verbal Fluency																
Semantic	50.72 (10.33)	50.06 (8.87)	51.89 (12.73)	52.00 (7.14)	43.84 (7.84)	57.13 (7.16)	52.75 (11.52)	57.50 (11.30)	44.70 (9.87)	43.86 (10.30)	47.08 (8.46)	38.33 (9.56)	42.82 (8.00)	42.25 (11.42)	51.71 (7.54)	51.33 (8.32)
Phonemic	22.22 (8.39)	22.59 (7.48)	21.56 (10.01)	18.86 (7.92)	18.11 (5.92)	24.25 (8.12)	29.88 (7.75)	25.25 (9.16)	18.12 (6.19)	18.30 (6.00)	17.62 (6.92)	13.67 (4.00)	16.53 (5.49)	18.13 (5.71)	21.86 (5.92)	22.67 (6.34)

Note. ss = age-scaled score (*M* = 10, *SD* = 3). All other scores are raw scores. Standard deviations in parentheses.

As previously noted, Trail-B is a more complex task than Trail-A because it makes greater cognitive demands. To examine this hypothesis, two paired-samples t-tests were performed for each group. The results indicated that TDC, $t(49) = 13.773$, $p < .001$, $d = 2.27$, and children with DD, $t(49) = 15.191$, $p < .001$, $d = 2.54$, take more time to complete the Trail-B than the Trail-A.

Planning

A multi-factorial MANOVA performed on the three Tower scores yielded a non-significant main effect for group, $F(3, 79) = 0.915$, $p = .438$, $\eta^2_p = .034$, and for the group X gender, $F(3, 79) = 2.034$, $p = .116$, $\eta^2_p = .072$, group X age, $F(12, 243) = 1.297$, $p = .221$, $\eta^2_p = .060$, and group X gender X age interactions, $F(9,243) = 0.825$, $p = .593$, $\eta^2_p = .030$ (see Table 1).

Verbal Fluency

The performance scores of TDC and children with DD on Semantic and Phonemic VF tests are shown in Table 1. The scores on the two tasks tapping VF were entered into a MANOVA as dependent variables and group, gender and age as fixed factor. The multivariate main effect of group proved to be significant, $F(2, 80) = 7.975$, $p = .001$, $\eta^2_p = .166$. At the univariate level, significant group differences were observed for Semantic VF, $F(1, 81) = 10.479$, $p = .002$, $\eta^2_p = .115$, and Phonemic VF, $F(1, 81) = 12.579$, $p = .001$, $\eta^2_p = .134$. Children with DD produced significantly fewer words within the 60-second time limit than TDC on both VF tests. No significant interactions were observed for group X gender, $F(2, 80) = 0.516$, $p = .599$, $\eta^2_p = .013$, group X age, $F(8, 162) = 1.525$, $p = .152$, $\eta^2_p = .070$, and group x gender x age, $F(6, 162) = 1.372$, $p = .229$, $\eta^2_p = .048$. For the TDC and DD groups [TDC: $t(49) = 21.033$, $p < .001$, $d = 3.02$; DD: $t(49) = 25.170$, $p < .001$, $d = 3.22$], the higher number of words produced within the time limit were observed on the Semantic VF (see Table 1).

Table 2. Repeated measures ANOVA of verbal fluency over four time intervals

	(1) 0-15s	(2) 16-30s	(3) 31-45s	(4) 46-60s	Repeated Measures ANOVA	Pairwise comparisons*
	M (*SD*)	*M* (*SD*)	*M* (*SD*)	*M* (*SD*)		
Semantic VF						
TDC	24.38 (4.44)	12.36 (3.00)	8.30 (2.60)	5.66 (3.15)	$F(3, 147) = 535.845$ $p < .001$, $\eta^2_p = .916$	1 > 2 > 3 > 4
DD	20.96 (4.12)	10.74 (3.27)	7.84 (3.08)	5.24 (2.42)	$F(3, 147) = 379.214$ $p < .001$, $\eta^2_p = .886$	1 > 2 > 3 > 4
Phonemic VF						
TDC	10.96 (3.30)	4.80 (2.42)	3.44 (2.20)	3.02 (2.36)	$F(3, 147) = 211.141$ $p < .001$, $\eta^2_p = .812$	1 > 2 > 3,4
DD	9.30 (3.11)	3.56 (1.93)	2.86 (1.78)	2.34 (1.61)	$F(3, 147) = 160.869$ $p < .001$, $\eta^2_p = .767$	1 > 2,3,4; 2 > 4

Note. * Bonferroni adjustment for multiple comparisons ($p < .05$); TDC = typically developing children; DD = children with developmental dyslexia; VF = verbal fluency.

To analyze the performance of both groups over four time intervals (0-15 seconds; 16-30 seconds; 31-45 seconds; and 46-60 seconds) on the Semantic VF and Phonemic VF tests, we performed four repeated measures ANOVAs. As shown in Table 2, the number of words produced in each of the four time intervals differed significantly, with word production decreasing over time in both VF measures. The Bonferroni adjustment for multiple comparisons revealed the presence of significant differences among all the time intervals in the Semantic VF task and almost all the time intervals in the Phonemic VF task for both groups. As expected, children tended to produce more words in the first 15 seconds than in the remaining three time intervals. Additional independent-samples t-tests revealed statistically significant differences between the TDC and DD groups in the first two time intervals of both VF tasks, 0-15 seconds: $t_{SVF}(98) = 3.986$, $p < .001$, $d = 0.79$; $t_{PVF}(98) = 2.582$, $p = .011$, $d = 0.51$; 16-30 seconds: $t_{SVF}(98) = 2.576$, $p = .011$, $d = 0.51$; $t_{PVF}(98) = 2.824$, $p = .006$, $d = 0.56$.

Group Differences on Executive Functions after Controlling for WISC-III GAI

The WISC-III GAI scores differed significantly, $t(98) = 3.569$, $p < .001$, $d = 0.71$, between the TDC and the children with DD (sum of the eight age-scaled scores that enter the Verbal Comprehension Index and the Perceptual Organization Index; TDC group: $M = 88.76 \pm 13.02$, and DD group: $M = 80.31 \pm 10.36$). Therefore, we additionally examined whether GAI scores could explain the group differences on EF tasks. A series of 2 X 2 X 5 (group X gender X age) MANCOVAs and ANCOVAs, covarying WISC-III GAI, were conducted on all EF tasks. After controlling for differences in general intellectual ability, the main effect of group remained significant for shifting, $F(1, 79) = 7.616$, $p = .007$, $\eta^2_p = .089$, for VF, $F(2, 78) = 3.901$, $p = .024$, $\eta^2_p = .091$ (univariate analysis: Semantic VF, $p = .022$; Phonemic VF, $p = .022$), and marginally significant for PS, $F(3, 77) = 2.727$, $p = .050$, $\eta^2_p = .096$ (univariate analysis: Coding, $p = .113$; Symbol Search, $p = .020$; Trail-A, $p = .063$). In contrast, none of the interactions or the main effect of group for planning were significant.

ROC Curve and Binary Logistic Regression Analysis

Although the findings presented above report the presence of significant differences in EF between TDC and children with DD (except in the Tower results), it is not certain that these tasks can successfully discriminate between subjects. Therefore, a ROC curve analysis and a binary logistic regression analysis were also performed to determine which EF independently contributed to distinguishing between children with DD and TDC. As shown in Table 3, only the Trail-B test showed moderate accuracy (ROC curve analysis), with an AUC of .730 (i.e., a randomly selected child with DD will take more time to complete the Trail-B than a randomly selected child from the TDC group approximately 73% of the time), while the remaining tasks showed low accuracy.

The goodness-of-fit test of the binary logistic regression analysis yielded a Hosmer-Lemeshow $\chi^2(8) = 5.495$, $p = .704$, suggesting that the model fit the data well. A Cox and Snell $R^2 = .241$ and a Nagelkerke $R^2 = .322$ were also found. This binary logistic regression model of the four EF tasks correctly classified 71.7% of the participants according to their DD diagnosis: 69.4% true-positive (sensitivity), 74% true-negative (specificity), 26% false-positive, and 30.6% false-negative. As shown in Table 3, only the Trail-B score was a significant predictor, with an odds ratio of 1.015 (= $e^{0.015}$). This result indicates that each one-second increase of the Trail-B score increased a child's odds of being in the DD group by 1.5%. For example, an increase of 10 seconds on the Trail-B test increases the odds from 1 to 1.161 (= $e^{10*0.015}$).

Table 3. Receiver operating characteristics curve analysis and binary logistic regression

	ROC Curve Analysis		Binary Logistic Regression Analysis		
	AUC (95% CI)	*SE*	β	Wald's χ^2	Odds Ratio
Processing Speed					
Coding	.670 (.563 – .777)**	.055	-.110	0.931	0.896
Symbol Search	.663 (.556 – .770)**	.055	-.148	1.989	0.863
Trail-A	.651 (.542 – .759)**	.055	.002	0.017	1.002
Shifting					
Trail-B	.730 (.631 – .829)***	.051	.015	3.940*	1.015
Planning (Tower)					
Correct 1st Trials	.592 (.480 – .704)	.057	-.380	1.583	0.684
Correct Models	.539 (.425 – .652)	.058	-.061	0.014	0.940
Total Trials	.594 (.482 – .706)	.057	-.089	0.240	0.915
Verbal Fluency					
Semantic	.660 (.554 – .766)**	.054	-.013	0.192	0.987
Phonemic	.644 (.536 – .753)*	.055	-.047	1.377	0.954

Note: *p < .05, **p < .01, ***p < .001; ROC = receiver operating characteristics; AUC = area under the curve; CI = confidence interval; *SE* = standard error.

Discussion

EF encompasses a set of inter-related processes necessary for goal-directed behavior. These processes develop throughout childhood and adolescence, are largely mediated by the prefrontal and the temporal cortex of the brain, and regulate other cognitive processes. Unsurprisingly, some aspects of EF have been associated with academic achievement (Clair-Thompson & Gathercole, 2006; Thorell, Veleiro, Siu, & Mohammadi, 2012) and reading ability (Foy & Mann, 2013; Sesma et al., 2009) and may therefore play an important role in DD (Altemeier et al., 2008; Booth, Boyle, & Kelly, 2010).

The first main objective of the present study was to analyze the performance of Portuguese TDC and children with DD on EF tasks. As expected, based on previous studies from other orthographies, our findings showed the presence of specific EF deficits in children with DD; the results revealed significant differences on PS, shifting, and VF tasks. Larger effect sizes were observed in analyses of the Trail-B, Semantic and Phonemic VF results, suggesting that children with DD may exhibit more deficits on EF tasks that place greater demands on switching abilities and verbal skills. The finding that DD is associated with slower PS and shifting replicates other studies that used the same measures (Narhi et al., 1997; Willcutt et al., 2005) and with those that used different measures (Boets et al., 2010; Shanahan et al., 2006) that incorporated a verbal component of PS (rapid automatized naming) and shifting (rapid alternating stimulus). Non-significant differences were found for all ToL scores, indicating that planning and problem-solving abilities are not compromised in children with DD. This finding is consistent with previous studies examining children with DD (Brosnan et al., 2002; Marzocchi et al., 2008; Reiter et al., 2005) or reading difficulties (Sikora, Haley, Edwards, & Butler, 2002). This non-significant group difference in planning ability may also be related to the presence of a ceiling effect in two of the three ToL scores (Correct Models score and Total Trials score). No interaction of group and gender and/or age was found for any of the EF tasks.

Because the mean WISC-III GAI scores of TDC and DD were significantly different, we additionally examined whether general intellectual ability could explain group differences on EF tasks. The main effect of group remained significant for shifting, VF and marginally significant for PS. The significant main effect on two of the three PS tasks was eliminated after controlling for WISC-III GAI, suggesting that Coding and Trail-A difficulties associated with DD are explained by group differences in general intellectual ability. Whereas some researchers suggest that general intellectual ability should be statistically controlled in cognitive studies of neurodevelopmental disorders, other researchers propose that this approach is misguided and unjustified (for a review, see Dennis et al., 2009; Willcutt et al., 2013).

A more detailed analysis was performed on the results of the two VF tasks. Despite the existence of statistically significant differences between groups (TDC > children with DD) on both the semantic and phonemic VF tasks (as observed in other studies: Landerl et al., 2009; Marzocchi et al., 2008), both groups scored significantly higher on the semantic than the phonemic VF task. This confirms the results of previous studies (Filippetti & Allegri, 2011; Martins et al., 2007; Reiter et al., 2005), corroborating the consensus that the phonemic VF task is more difficult, possibly because it requires the exploration of more category subsets, relies more on the central executive component of working memory, and it is more dependent on the frontal lobe (Birn et al., 2010). As suggested by Troyer (2000) and Hurks et al. (2004; 2006), the pattern of word production over time is relevant to understanding the specific cognitive mechanisms that underlie poor performance on VF tasks. Our results revealed that there is a significant decrease in the number of words produced among both groups (children with DD and TDC) and on both tasks (semantic and phonemic) as a function of time (over four time intervals), which is congruent with the model of lexical organization proposed by Crowe (1998). This model states that in the first period, a ready pool of frequently used words is available and is automatically active for production (automatic processing), but as time passes, the pool becomes exhausted and the search for new words becomes both more effortful and less productive (controlled processing). Notably, significant group differences were only observed in the first two time intervals (TDC > children with DD), suggesting that poor performance on VF tasks among children with DD was particularly related to deficits in automatic processing. Recently, Takács et al. (2014) also found that TDC and children with ADHD generated the largest number of correct responses during the first two time intervals and that significant group differences were only observed in the first quarter. Similarly, Hurks et al. (2004) also observed that children with ADHD generated fewer words (phonemic VF) in the first 15 seconds than did healthy controls and children with other psychopathologies. The authors suggested that children with ADHD may have a developmental delay in automatic processing of abstract verbal information.

Because the presence of a significant difference alone does not indicate that a test can discriminate among subjects with sufficient accuracy for clinical use, the second main objective of the study was to analyze the accuracy with which the EF measures under study discriminate between children with DD and TDC. The results of the ROC curve analysis yielded low diagnostic accuracy for all the tests except Trail-B. The binary logistic regression model, however, yielded an accuracy rate of 71.7% in classifying children into their correct group (Trail-B was the only significant predictor). No previous studies appear to have analyzed the utility of the different EF processes in diagnosing DD. Although the results highlighted the presence of specific EF deficits in children with DD, they should not be interpreted as indicative of the presence or absence of this learning disorder. As Willcutt et al. (2008, p. 202) stated "EF weakness are neither necessary or sufficient to cause any of the disorders (…), and are instead one important component of the complex neuropsychology of childhood disorders". Indeed, the information obtained from EF measures should only be a component of the neuropsychological evaluation and decision-making process and need to be viewed in the context of a more comprehensive assessment that includes other measures, such as phonological awareness, rapid naming, working memory, reading, and spelling measures.

Notwithstanding the uniqueness of the present study, it had several limitations that should be addressed in future studies. First, some of the EFs were assessed only by one task. Clearly the inclusion of more tasks per component would have increased the construct validity and interpretability of the results. Second, the inclusion of other EF tasks (e.g., inhibition, updating, working memory) would also contribute to a better understanding of executive functioning deficits in children with DD. Third, the performance of children with DD on EF tasks was only compared to a TDC group (chronological-age-matched controls) and did not include other clinical samples or a reading-level-matched control group. The literature has clearly demonstrated that children with ADHD also exhibit deficits in a wide range of EF measures (Frazier, Demaree, & Youngstrom, 2004; Fuggetta, 2006), and that DD and ADHD co-occur more frequently than would be expected by chance (15% to 40% of individuals with DD meet criteria for ADHD) (Willcutt & Pennington, 2000). Furthermore, recent studies proposed a multiple cognitive deficit model of neurodevelopmental disorders and found that DD and ADHD shared neurocognitive deficits (McGrath et al., 2011; Willcutt et al., 2005). Thus, future studies should include ADHD children with and without comorbidity with DD in order to increase the generalizability of the findings.

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Estudo 4

Working Memory in Portuguese Children with Developmental Dyslexia

Working Memory in Portuguese Children with Developmental Dyslexia

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Abstract

A Portuguese sample of 50 children with developmental dyslexia and 50 typical readers who were matched for age (8 to 12 years old) were tested on measures of working memory. Relative to the typical readers, the children with developmental dyslexia performed significantly lower on phonological loop and central executive tasks; however, they exhibited no impairments on visuospatial sketchpad tasks. After controlling for the influence of the phonological loop, the group differences in central executive were no longer significant. The results of a receiver operating characteristics curve analysis and a binary logistic regression analysis suggested that the phonological loop and central executive tasks (but not the visuospatial sketchpad tasks) were relevant variables for identifying children with developmental dyslexia. Hierarchical linear regression analyses showed that the phonological loop and central executive (Backward Digit Span only) tasks were significant predictors of reading and spelling abilities.

Keywords: working memory, short-term memory, developmental dyslexia, reading, spelling

Introduction

Working memory (WM) refers to a limited-capacity memory system that is involved in the temporary storage and processing of verbal and visuospatial information. WM is distinguished from other forms of memory because it reflects both processing and storage capacity (Baddeley, 2000, 2003). Although various models of WM have been developed (e.g., Cowan, 1999; Engle, Kane, & Tuholski, 1999), the framework of WM proposed by Baddeley and Hitch (1974) is the most widely used in studies of children with learning disabilities or other neurodevelopmental disorders. This WM model is conceptualized as a multi-component system comprising a central executive (CE) and two slave systems: the phonological loop (PL) and the visuospatial sketchpad (VSSP). The two slave systems are often referred to as short-term memory (STM), whereas the CE is also referred to as WM (e.g., Just & Carpenter, 1992; Swanson, Zheng, & Jerman, 2009).

The PL is a peripheral slave system specialized for the temporary storage of verbal information. This system comprises a limited phonological store, which can hold memory traces for a few seconds, and an articulatory rehearsal process, which prevents the decay of material stored in the phonological store by successively refreshing memory traces (Baddeley, 2003, 2012). Tasks that measure the PL typically assess the subject's capacity to recall a sequence of verbal items (e.g., digits, letters and words) in the order in which they were presented. The VSSP is a limited-capacity peripheral slave system specialized for the temporary storage of visual and spatial material. Although spatial and visual information was initially considered to be processed by a single VSSP system, subsequent neuropsychological studies have indicated the need to distinguish between visual and spatial STM (Della Sala, Gray, Baddeley, Allamano, & Wilson, 1999; Pickering, Gathercole, Hall, & Lloyd, 2001). Logie and colleagues (1995; Logie & Pearson, 1997) suggested a fractionation of the sketchpad into two subcomponents: a visual cache (temporary visual storage) and an inner scribe (retrieval and a rehearsal mechanisms; analogous to the articulatory rehearsal of the PL). Dynamic (e.g., Corsi Block Test) and static (e.g., Visual Patterns Test) span tasks are typically used to measure spatial and visual memory, respectively. The CE is a supervisory system that is responsible for controlling and manipulating information stored in the two slave systems, and it is often linked to the functioning of the frontal lobes (Baddeley, 1996, 2003). Baddeley (1996) described four functions of the CE: (i) the coordination of multiple tasks; (ii) the capacity to switch between tasks or retrieval strategies; (iii) the capacity to selectively attend to one stimulus while inhibiting others; and (iv) the capacity to retain and manipulate information in long-term memory. Thus, CE tasks (e.g., backward digit span tasks) place greater demands on executive functioning because they require the simultaneous storage and processing of information. Jerman, Reynolds, and Swanson (2012) noted the existence of a considerable overlap between the processes involved in CE and executive functions.

Because a number of phenomena were not addressed by the original three-component model of WM, Baddeley (2000) proposed the inclusion of a fourth component, the episodic buffer, which is controlled by the CE and is responsible for integrating information from a variety of sources. The episodic buffer, a limited-capacity system that provides the temporary storage of information held in a multimodal code, is capable of binding information from both the slave systems and long-term memory into a unitary episodic representation (Baddeley, 2012; Baddeley, Allen, & Hitch, 2011). More recently, in an effort to account for the impact of emotion on cognition, Baddeley (2013; Baddeley, Banse, Huang, & Page, 2012) proposed the existence of a hedonic detection system coupled to WM.

Working Memory Impairments in Developmental Dyslexia

Impairments in WM have been described as one of the major defining characteristics of developmental dyslexia (DD). For years, neurocognitive researchers have observed that children with DD performed extremely poorly on subtests corresponding to the current Working Memory Index of the Wechsler Intelligence Scale for Children (WISC) – Fourth Edition (Clercq-Quaegebeur et al., 2010; Helland & Asbjornsen, 2004). Studies employing more specific measures have also reported that children with DD exhibit strong evidence of WM impairments, particularly in the PL and CE components (Jeffries & Everatt, 2004; Menghini, Finzi, Carlesimo, & Vicari, 2011; Moura, Simões, & Pereira, 2014; Schuchardt, Maehler, & Hasselhorn, 2008).

Almost all studies investigating PL capacity have documented reductions in verbal span in children with DD (Kibby & Cohen, 2008; Menghini et al., 2011; Swanson et al., 2009; Willcutt, Pennington, Olson, Chhabildas, & Hulslander, 2005). Nonetheless, the literature has been discordant concerning which PL subcomponents are compromised. Some researchers have observed that the deficit appeared to be specific to the store mechanism (a reduced phonological similarity effect; i.e., rhyming items are more difficult to remember than non-rhyming items), while the subvocal rehearsal mechanism remained intact. However, others have found that children with DD exhibited less-efficient rehearsal processes (a reduced word length effect; i.e., short words are easier to remember than sequences of long words) or that phonological similarity and word length effects did not differ between children with DD and typical readers (TR) (Kibby, 2009; Pickering, 2004; Steinbrink & Klatte, 2008). Moreover, some researchers have found an association between PL and articulatory/speech rate (i.e., the number of verbal items repeated per second), suggesting that children with DD experience PL impairments due to their slow articulation rates, which cause the PL to function less efficiently (Kibby, 2009; McDougall & Donohoe, 2002). The PL also plays an important role in the development of reading skills. A large number of studies have demonstrated that the PL predicts reading decoding (Hulme, Goetz, Gooch, Adams, & Snowling, 2007; Kibby, 2009; Perez, Majerus, & Poncelet, 2012) and reading comprehension (Goff, Pratt, & Ong, 2005; Swanson & Ashbaker, 2000). Other researchers have found that the PL did not uniquely predict reading after controlling for phonological awareness and naming speed tasks (Parrila, Kirby, & McQuarrie, 2004).

VSSP capacity has been associated with visuospatial reasoning (Kane et al., 2004), spatial orientation (Baddeley, 2002) and arithmetic abilities (Holmes, Adams, & Hamilton, 2008; Sarver et al., 2012), and it appears to be diminished in some children with learning disabilities, such as dyscalculia (Ashkenazi, Rosenberg-Lee, Metcalfe, Swigart, & Menon, 2013; Schuchardt et al., 2008). Research on the relationship between DD and VSSP deficits has yielded mixed results. Although most studies have not found VSSP deficits in individuals with DD (Bacon, Parmentier, & Barr, 2013; Jeffries & Everatt, 2004; Kibby & Cohen, 2008; Schuchardt et al., 2008), others have suggested the presence of significant differences, with individuals with DD performing more poorly than TR (Menghini et al., 2011; Smith-Spark & Fisk, 2007). When visuospatial STM tasks involve CE demands, children with DD tend to exhibit more difficulties. For instance, Bacon et al. (2013) demonstrated that children with DD exhibited no deficits in the forward recall task of the Corsi Block Test but revealed significant impairments in the backward recall task. In addition, studies comparing the VSSP capacities of children with DD and those with other neurodevelopment disorders have found that children with DD performed better than children with comorbid dyslexia/dyscalculia (Landerl, Fussenegger, Moll, & Willburger, 2009) or other learning difficulties (Jeffries & Everatt, 2004) but achieved scores similar to those of children with attention deficit hyperactivity disorder (ADHD) (Willcutt et al., 2005). Among the limited studies that have explored the predictive power of VSSP tasks for reading performance, some have found that VSSP predicts long-term reading achievement (Sarver et al., 2012), reading fluency (Nevo & Breznitz, 2011) and reading comprehension (Goff et al., 2005). However, others found no predictive value of VSSP tasks for reading decoding or reading comprehension (Gathercole, Alloway, Willis, & Adams, 2006; Nevo & Breznitz, 2011).

As described above, the CE is responsible for controlling and processing information stored in STM, which involves the activation of various cognitive processes. Strong empirical evidence supports the presence of significant CE impairments in children with DD (Savage, Lavers, & Pillay, 2007; Swanson et al., 2009). These CE deficits may occur in the presence or absence of significant deficits in the PL or VSSP (Jeffries & Everatt, 2004; Smith-Spark & Fisk, 2007; Swanson, 2012; Swanson & Jerman, 2007), suggesting that the memory deficits associated with DD go beyond the temporary storage of information; information processing is also compromised. However, controlling the influence of PL on CE performance has produced contradictory results. For instance, de Jong (1998) and Smith-Spark and Fisk (2007) found significant differences in the mean scores of individuals with DD and TR on CE tasks, even after PL tasks were controlled through covariance. In contrast, Schuchardt et al. (2008) observed that group differences in CE performance were no longer significant when PL tasks were taken into account. Furthermore, the presence of intact CE functioning in children with DD has also been reported (Kibby & Cohen, 2008; Landerl et al., 2009; van der Sluis, van der Leij, & de Jong, 2005). Numerous studies have found that CE predicted variance in reading decoding (Gathercole et al., 2006; Jerman et al., 2012; Nevo & Breznitz, 2011; Swanson & Ashbaker, 2000), reading comprehension (Sesma, Mahone, Levine, Eason, & Cutting, 2009; Swanson & Jerman, 2007), reading fluency (Berninger et al., 2006; Nevo & Breznitz, 2011; Swanson & Jerman, 2007), and mathematical ability (Andersson, 2008; Jerman et al., 2012). Conversely, some studies have not found a predictive effect of CE on reading accuracy and/or reading comprehension (Berninger et al., 2006; Sesma et al., 2009).

The current study was undertaken to assess the extent to which WM is impaired in Portuguese children with DD. The European Portuguese language is considered an orthography of intermediate depth; more transparent than English and French, but less regular than German, Spanish, Italian or Finnish (Seymour, Aro, & Erskine, 2003; Sucena, Castro, & Seymour, 2009). Seymour et al. (2003) examined the beginning of reading acquisition in 13 European orthographies and found that children become fluent and accurate before the end of first grade. The exceptions to this reading development pattern were English, French, Danish and Portuguese (the Portuguese and French orthographic code learning trajectories were quite similar). Based on the existing literature (the large body of research about WM deficits in DD has been conducted on English-speaking samples), three predictions were made. First, it was expected that Portuguese children with DD who were native speakers of an orthography of intermediate depth would also show significant impairments in PL and CE but exhibit an intact VSSP. Second, it was expected that the PL and CE (but not the VSSP) would be accurate measures for discriminating between subjects (children with DD and TR). Third, as observed in other languages spanning a large range of orthographic complexity, it was hypothesized that only the PL and CE would be significant predictors of reading and spelling in the Portuguese orthography. Few studies have explored the accuracy of WM for discriminating between typical readers and children with DD, and few have analyzed the predictive power of WM for spelling. Furthermore, no published studies have analyzed WM performance among Portuguese children with DD.

Method

Participants

The participants were 100 Portuguese children between the ages of 8 and 12 years (*M* = 9.81; *SD* = 1.34) in the 3rd to 6th school grades. The DD group (*N* = 50) included 74% male and 26% female subjects, with a mean age of 9.80 years (*SD* = 1.38). Among the children with DD, 26% had experienced school retention, 36% were included in special education system, 94% had attended kindergarten, and 30% had relatives with reading difficulties. The TR group (*N* = 50) included 64% male and 36% female subjects, with a mean age of 9.82 years (*SD* = 1.32). All the TR had attended kindergarten, only 2% had experienced school retention, and 4% had relatives with reading difficulties. The children in the DD and TR groups were matched for age $\chi^2(4) = 0.487$, $p = .975$, yielding non-significant differences in gender $\chi^2(1) = 1.169$, $p = .387$ and grade $\chi^2(3) = 1.776$, $p = .620$.

Criteria for Inclusion. For both groups, only children who met the following criteria were included: (i) Wechsler Intelligence Scale for Children – Third Edition (WISC-III) Full Scale IQ (FSIQ) ≥ 90; (ii) native speakers of European Portuguese; (iii) at least two years of school attendance; (iv) the absence of visual, auditory or motor handicaps; and (v) the absence of language impairments, emotional disturbances, dyscalculia, disruptive behavior disorders (ADHD, oppositional defiant disorder or conduct disorder), neurological impairments or other psychiatric disorders. Children with special educational needs were also excluded from the TR group.

In the DD group, only children who had previously been diagnosed with DD by a psychologist, child psychiatrist, developmental pediatrician, or child neurologist and had received a score lower than or equal to the 15th percentile on a reading fluency and accuracy measure ("O Rei"; Carvalho & Pereira, 2009) administered during the testing session were included. These cutoff scores (WISC-III FSIQ ≥ 90 and reading fluency and accuracy measures both ≤ 15th percentile) were similar to (and in some cases stricter than) the inclusion criteria used in previous studies assessing WM deficits in individuals with DD (e.g., Schuchardt et al., 2008; Swanson, 2011, 2012; Tiffin-Richards, Hasselhorn, Woerner, Rothenberger, & Banaschewski, 2008).

Measures and Procedures

Intellectual Ability. The Portuguese version of the WISC-III (Wechsler, 2003), which was normed on a representative sample of 1354 children (aged 6 to 16 years), was administered to measure general intellectual ability. The factor structure of the Portuguese version of WISC-III, analyzed through an exploratory and confirmatory factor analysis, yielded adequate psychometric properties for a two-factor model (Verbal IQ and Performance IQ) and for a three-factor model (Verbal Comprehension, Perceptual Organization and Processing Speed). The reliability of the WISC-III FSIQ was .89 (linear combinations), with a test-retest correlation coefficient of .92 (Wechsler, 2003). The subjects' FSIQ scores (M = 100; SD = 15) were analyzed and used as a covariate in the inferential analysis.

Phonological Loop. The Forward task from the Digit Span[8] subtest of the WISC-III (Forward DS) and the Verbal Learning Test from the Coimbra Neuropsychological Assessment Battery[9] (BANC; Simões et al., in press) were selected to assess verbal STM.

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[8] The reliability of the Digit Span subtest was .80 (split-half), with a test-retest correlation coefficient of .72 (Wechsler, 2003).

[9] The BANC is a comprehensive assessment instrument tapping different functions of children's neuropsychological development, which included 16 tests organized in six main domains: Memory (Verbal Learning Test, Narrative Memory, Memory of Faces, Rey Complex Figure Test, and Corsi Block Test); Language (Phonological Awareness, Instruction Comprehension, and Rapid Naming); Attention and

The Forward DS required that the child correctly recall a series of two to nine digits in the order in which they were presented. One point per trial was given for a correct repetition. In the Verbal Learning Test, a list of 15 unrelated words was read to the child four consecutive times. Following each trial, the child was asked to recall as many words as possible. A new list with 15 words was then presented and recalled once (interference recall). Then, the child was asked to recall the first word list immediately (immediate recall) and after a 20- to 30-minute delay (delayed recall). Finally, a list of 45 words (15 from the first list, 15 from the interference list, and 15 new) was presented, and the child was asked to identify the 15 first-list words (recognition). Because the purpose of the Verbal Learning Test was to measure PL, only the first trial score (i.e., the first time that the child was asked to recall the 15 words) was considered in the subsequent analyses. These tasks are conventional measures used to assess verbal STM (Bora et al., 2008; Jeffries & Everatt, 2004; Kramer, Knee, & Delis, 2000; Schuchardt et al., 2008).

Visuospatial Sketchpad. The Corsi Block Test and the Rey Complex Figure Test (RCFT) were administered to measure VSSP. The Corsi Block Test consists of nine blocks nailed onto a board at random positions. The child was asked to reproduce the sequence (from two to nine blocks) by touching the blocks in the same order as the experimenter. The task ended when the child failed to reproduce both trials at any particular span length. One point per trial was given for a correct reproduction. In the RCFT, the child was instructed to copy the complex figure as accurately as possible and to then reproduce it from memory 3 minutes later (immediate recall) and 20 to 30 minutes later (delayed recall). The Meyers and Meyers (1995) scoring system was used (each of the 18 elements was scored with 2, 1, 0.5 or 0 points according to its presence, accuracy and location). Because the purpose of the RCFT was to measure visuospatial STM, only the immediate recall score was considered. These two tasks are widely used to assess visuospatial STM

Executive Functions (Cancellation, Trail, Semantic Verbal Fluency, Phonemic Verbal Fluency, and Tower); Motricity; Laterality; and Orientation. The BANC (Simões et al., in press) was normed on a representative and stratified sample of 1104 Portuguese children (aged 5 to 15 years) and revealed adequate psychometric properties [e.g., confirmatory factor analysis yielded an adequate model fit with Comparative Fit Index (CFI) = .965 and Root Mean Square Error of Approximation (RMSEA) = .044 for children aged 7 to 9 years; and CFI = .966 and RMSEA = .046 for children aged 10 to 15 years].

(Brunswick, Martin, & Marzano, 2010; Maehler & Schuchardt, 2011; Smith-Spark & Fisk, 2007; Wisniewski, Wendling, Manning, & Steinhoff, 2012).

Central Executive. The Backward task from the Digit Span subtest of the WISC-III (Backward DS) and the Trail from the BANC were chosen to assess the CE component of WM. Backward DS is extensively used as a measure of CE because it assesses the ability to briefly maintain and manipulate information in WM (Jeffries & Everatt, 2004; Schuchardt et al., 2008; Tiffin-Richards et al., 2008). This task required that the child correctly recall a series of two to eight digits in the reverse order. One point per trial was given for correct recall. The Trail (Part A and B), which is similar to the popular Trail Making Test, was chosen because it is another frequently used measure of CE functioning (Andersson, 2008; Baddeley, 1996; McLean & Hitch, 1999). In Trail-A, 25 encircled numbers were randomly distributed on a sheet of paper. The child had to draw a line connecting the numbers sequentially from 1 to 25 as rapidly and accurately as possible. In Trail-B, the child has to draw a line connecting 25 circles with numbers or letters, randomly distributed on a sheet of paper. The child had to draw a line connecting the circles, alternating between numbers and letters (e.g., 1, A, 2, B, etc.), as rapidly and accurately as possible. Thus, Trail-B required that the child focus on both alphabetical and numerical series while simultaneously remembering whether a letter or number should occur next in the series. The raw scores of Trail-A and Trail-B represented the amount of time (in seconds) required to complete the tasks. As suggested by some authors (Andersson, 2008; Drane, Yuspeh, Huthwaite, & Klingler, 2002), to obtain a "purer" measure of shifting, the difference between the Trail-B and Trail-A scores (Trail B-A) was used in the subsequent analyses.

Reading and Spelling Measures. Four measures were used to assess reading and spelling abilities: text reading accuracy, text reading fluency, word reading accuracy, and word spelling accuracy. The "O Rei" ("The King"; Carvalho & Pereira, 2009), an individually administered reading test for children that involves a Portuguese traditional tale, was chosen to measure text reading accuracy (the percentage of correctly read words) and text reading fluency (the number of words read in one minute). To assess word reading and word spelling accuracy, we used the Oral Reading subtest from the Portuguese version (Festas, Martins, & Leitão, 2007) of the Psycholinguistic Assessment of Language (PAL; Caplan, 1992). This subtest comprises 146 words (48 regular, 47 irregular and 51 pseudowords). Based on previous studies of typically developing children, we selected 40 words: 16 regular (8 high-frequency and 8 low-frequency words; e.g., *sardinha* and *delonga*), 16 irregular (8 high-frequency and 8 low-frequency words; e.g., *brinquedo* and *exotismo*) and 8 pseudowords (e.g., *lempo* and *glepal*). This subtest was used in both the reading and spelling tasks, which were separated by an interval of 10 to 15 days.

The administration of these tests was included as part of a broad neuropsychological research that also comprises other measures (e.g., phonological awareness, naming speed, and attention). Each child completed two individual sessions (separated by an interval of 10 to 15 days), lasting approximately 90 minutes per session, in a clinic or school setting during a weekday. All measures were administered by the first author in a fixed order. No incentives were offered in exchange of participation.

Statistical Analyses

The statistical analyses were performed using IBM SPSS Statistics 19. Group differences were analyzed using multivariate analyses of variance (MANOVA) and covariance (MANCOVA) for each WM component. If the multivariate analysis indicated a significant overall difference ($p < .05$), then a univariate test was applied to determine which dependent variables were responsible for the multivariate difference. In specific cases, an independent-samples t-test was also used. Partial eta-squared (η^2_p) or Cohen's *d* was calculated to determine the effect size of the difference between groups.

A receiver operating characteristics (ROC) curve and a binary logistic regression analysis were performed to assess the accuracy of the WM tasks to correctly discriminate between children with DD and TR. A ROC curve analysis systematically sweeps across all possible true-positive (sensitivity) and false-positive (1-specificity) values of a diagnostic test and calculates the area under the curve (AUC), which provides an accuracy index of the test (Fawcett, 2006). An AUC value of .5 to .7 indicates low test accuracy; .7 to .9

indicates moderate accuracy; and .9 to 1.0 indicates high accuracy (Swets, 1988). For the binary logistic regression analysis, the fit of the model (Hosmer-Lemeshow test, Cox and Snell R^2, and Nagelkerke R^2) and the statistical tests of individual predictors were analyzed (regression coefficient, Wald's χ^2, and odds ratio).

To determine the predictive value of WM for reading and spelling abilities, hierarchical linear regression analyses were also conducted. The total variance (R^2) of the regression model, the t-test (*t*), the squared part correlation (pr^2), the standard error (*SE*), and the unstandardized (B) and standardized (β) regression coefficients for each independent variable were calculated.

Results

Group Differences

The WISC-III FSIQ scores differed significantly, $t(98) = 4.721$, $p < .001$, $d = 0.95$, between the TR ($M = 108.24 \pm 11.64$) and the children with DD ($M = 98.53 \pm 8.55$). Therefore, group differences were tested using MANOVA and MANCOVA, with WISC-III FSIQ as a covariate.

For the PL, a MANOVA was performed with Forward DS and Verbal Learning Test (first trial score) as dependent variables and reading group (TR and children with DD) as fixed factor. Reading group had a significant effect, $F(2, 97) = 12.028$, $p < .001$, Wilks' Λ = .800, $\eta^2_p = .200$. The univariate analysis revealed significant effects in both PL tasks, with the TR (Forward DS = 7.36 ± 1.45, Verbal Learning Test = 6.58 ± 1.75) outperforming the children with DD (Forward DS = 6.20 ± 1.13, Verbal Learning Test = 5.59 ± 1.60). Significant differences for PL remained when WISC-III FSIQ was used as a covariate, $F(2, 96) = 7.652$, $p = .001$, Wilks' $\Lambda = .861$, $\eta^2_p = .139$ (see Table 1).

For the VSSP, the scores on the Corsi Block Test and RCFT (immediate recall) were entered into a MANOVA, with reading group as a fixed factor. The multivariate main effect of reading group was not significant, $F(2, 97) = 1.346$, $p = .265$, Wilks' $\Lambda = .973$, η^2_p

= .027 (Corsi Block Test: TR = 7.74 ± 1.93, children with DD = 7.18 ± 1.53; RCFT: TR = 15.86 ± 5.77; children with DD = 15.25 ± 5.67). The result remained non-significant when a MANCOVA controlling for differences in intelligence was performed, $F(2, 96) = 0.558$, $p = .574$, Wilks' $\Lambda = .988$, $\eta^2_p = .012$ (see Table 1).

The scores on the two tasks tapping CE functioning were entered into a MANOVA, and the multivariate main effect of reading group proved to be significant, $F(2, 97) = 11.243$, $p < .001$, Wilks' $\Lambda = .810$, $\eta^2_p = .190$. At the univariate level, significant group differences were observed for Backward DS and Trail B-A: compared with the TR, the children with DD recalled fewer digits in the backward condition (TR = 4.56 ± 1.34, children with DD = 3.63 ± 0.97) and required more time to complete the Trail B-A (TR = 53.98 ± 27.71, children with DD = 77.00 ± 36.11). After controlling for differences in the WISC-III FSIQ, the multivariate main effect of reading group remained significant, $F(2, 96) = 5.852$, $p = .004$, Wilks' $\Lambda = .890$, $\eta^2_p = .110$ (see Table 1).

Table 1. Means, standard deviations and multivariate analyses of variance and covariance

	Typical Readers	Children with DD	MANOVA			MANCOVA (FSIQ)		
	M ± *SD*	*M* ± *SD*	*F*(1, 98)	*p*	η^2_p	*F*(1, 97)	*p*	η^2_p
Phonological Loop								
Forward DS	7.36 ± 1.45	6.20 ± 1.13	19.383	<.001	.167	12.101	.001	.112
Verbal Learning Test	6.58 ± 1.75	5.59 ± 1.60	8.554	.004	.081	5.629	.020	.055
Visuospatial Sketchpad								
Corsi Block Test	7.74 ± 1.93	7.18 ± 1.53	2.570	.112	.026	1.119	.293	.012
Rey Complex Figure	15.86 ± 5.77	15.25 ± 5.67	0.284	.595	.003	0.001	.996	.000
Central Executive								
Backward DS	4.56 ± 1.34	3.63 ± 0.97	15.439	<.001	.137	6.287	.014	.061
Trail B-A	53.98 ± 27.71	77.00 ± 36.11	12.689	.001	.116	8.192	.005	.079

Note: FSIQ = Wechsler Intelligence Scale for Children (Third Edition) Full Scale IQ, DS = Digit Span subtest of the Wechsler Intelligence Scale for Children (Third Edition), DD = developmental dyslexia.

Because CE tasks require both the temporary storage and processing of information, it has been hypothesized that differences in temporary storage systems might underlie group differences in CE. To examine this hypothesis, a MANCOVA was performed with the two CE tasks as dependent variables, reading group as a fixed factor and the two PL tasks as covariates. This type of analysis was not performed for VSSP because a previous inferential analysis did not show significant differences. After controlling for PL tasks, no significant group differences in CE remained, $F(2, 95) = 2.856$, $p = .062$, Wilks' $\Lambda = .943$, $\eta^2_p = .057$ (although the *p*-value was closer to statistical significance), suggesting that the group differences in CE could be accounted for by differences in PL.

Table 2. Receiver operating characteristics (ROC) curve analysis

	AUC (95% CI)	*SE*	*p*
Phonological Loop			
Forward DS	.737 (.639 – .835)	.050	< .001
Verbal Learning Test	.657 (.550 – .763)	.054	.007
Visuospatial Sketchpad			
Corsi Block Test	.581 (.468 – .694)	.058	.165
Rey Complex Figure	.530 (.415 – .646)	.059	.602
Central Executive			
Backward DS	.704 (.602 – .806)	.052	< .001
Trail B-A	.707 (.604 – .810)	.052	< .001

Note: DS = Digit Span subtest of the Wechsler Intelligence Scale for Children (Third Edition), AUC = area under the curve, CI = confidence interval, *SE* = standard error.

Diagnostic Accuracy

The results of previous inferential analyses indicated significant group differences in PL and CE; however, this does not imply that WM tasks can correctly discriminate between children with DD and TR. Therefore, ROC curve and a binary logistic regression analyses were performed.

The results of the ROC curve analysis revealed that only the PL and CE tasks were significant variables for discriminating between subjects. The more accurately a task discriminates between groups, the higher is its AUC value. The AUC values of the Forward DS, Backward DS, and Trail B-A tasks revealed moderate levels of accuracy, indicating that a randomly selected child with DD will receive a lower score than a randomly selected TR approximately 73.7%, 70.4% and 70.7% of the time, respectively (see Table 2).

Table 3. Binary logistic regression analysis

	Sensitivity (%)	Specificity (%)	β	Wald's χ^2	Odds Ratio (95% CI)
Phonological Loop	73.5	64.0			
Forward DS			-.709	10.570**	0.492 (0.321 – 0.755)
Verbal Learning Test			-.257	3.494	0.773 (0.590 – 1.013)
Visuospatial Sketchpad	62.0	48.0			
Corsi Block Test			-.182	2.373	0.833 (0.661 – 1.051)
Rey Complex Figure			-.014	0.151	0.986 (0.919 – 1.058)
Central Executive	57.1	74.0			
Backward DS			-.670	7.475**	0.512 (0.317 – 0.827)
Trail B-A			.019	5.641*	1.019 (1.003 – 1.035)

Note: DS = Digit Span subtest of the Wechsler Intelligence Scale for Children (Third Edition), CI = confidence interval, *p < .05, **p < .01.

An individual binary logistic regression analysis was performed for each WM component. For the PL, the logistic regression model yielded a Hosmer-Lemeshow $\chi^2(8)$ = 7.372, p = .497, suggesting that the model fit the data well. The Cox and Snell R^2 = .206, and the Nagelkerke R^2 = .274. The PL correctly classified 68.7% of the children (sensitivity = 73.5% and specificity = 64%). Only the Forward DS task was a significant predictor, with an odds ratio of 0.492 (i.e., with each one-point increase in the Forward DS score, the odds of being in the DD group decreased from 1 to 0.492). For the VSSP, a Hosmer-Lemeshow $\chi^2(8)$ = 1.854, p = .985; Cox and Snell R^2 = .027; and Nagelkerke R^2 = .036 were obtained. This logistic regression model correctly classified 55% of the children (sensitivity = 62% and specificity = 48%). Neither the Corsi Block Test nor the RCFT (immediate recall) was a significant predictor. For the CE, the goodness-of-fit test yielded a Hosmer-Lemeshow $\chi^2(8)$ = 7.301, p = .504, with a Cox and Snell R^2 = .200 and a Nagelkerke R^2 = .267. This model correctly classified 65.6% of the children (sensitivity = 57.1% and specificity = 74%), and both CE tasks were significant predictors. Each one-point increase in the Backward DS score decreased the odds of being in the DD group by 48.8%, whereas every one-second increase in the Trail B-A score increased the odds of being in the DD group by 1.9% (see Table 3).

Predictive Effect of Working Memory on Reading and Spelling Abilities

Hierarchical linear regression analyses were performed to determine whether the WM tasks were predictive variables for reading and spelling abilities. Four regression models were performed, one for each dependent variable (text reading accuracy, text reading fluency, word reading accuracy, and word spelling accuracy). The predictive variables were entered in the following order: PL tasks were entered into the first block, VSSP tasks were entered next, and CE tasks were entered last. The *B*, *SE*, β and *t* values shown in Table 4 are relative to the last block. The pr^2 value represents the unique variance of each predictor after the overlapping linear effects of all the other predictive variables were statistically removed.

For text reading accuracy, the regression model was statistically significant, $F(6, 92) = 5.364$, $p < .001$, and explained 25.9% of the total variance. After controlling for the PL tasks, the VSSP tasks explained only 1.1% of the variance. The CE tasks explained an additional 7.6% of the variance. Only the Verbal Learning Test (5.1% of unique variance) and the Backward DS (7.3% of unique variance) were significant predictors.

For text reading fluency, the regression model was statistically significant, $F(6, 92) = 15.447$, $p < .001$, and explained 50.2% of the total variance. After controlling for the PL tasks, the VSSP accounted for 1.2% of the variance, whereas the CE tasks uniquely accounted for 15.2% of the variance. The Forward DS, Verbal Learning Test and Backward DS were significant predictors, with unique variances of 2.3%, 5.9% and 15.1%, respectively.

Regressing the word reading accuracy scores on measures of WM yielded a significant model, $F(6, 92) = 6.383$, $p < .001$, which explained 29.4% of the total variance. After controlling for the PL and VSSP tasks, the CE tasks uniquely accounted for 5.7% of the variance. Again, only the Verbal Learning Test (4.8% of unique variance) and the Backward DS (5.5% of unique variance) were significant predictors.

Finally, regressing the word spelling accuracy scores on the WM measures also yielded a significant model, $F(6, 92) = 8.843$, $p < .001$, which explained 36.6% of the total variance. After controlling for the PL, the VSSP tasks explained only 1.8% of the variance, whereas the CE tasks uniquely accounted for 4.5% of the variance. The Forward DS, Verbal Learning Test and Backward DS were significant predictors, with unique variances of 3.8%, 5.3% and 4%, respectively (see Table 4).

Table 4. Hierarchical linear regression analysis

Dependent Variable	Block	Predictors	ΔR^2	*B*	*SE*	β	*t*	pr^2
Text Reading Accuracy	1	Forward DS (PL)	.172	0.403	0.593	.080	0.680	.004
		Verbal Learning Test (PL)		0.970	0.387	.236	2.509*	.051
	2	Corsi Block Test (VSSP)	.011	-0.077	0.387	-.019	-0.200	<.001
		Rey Complex Figure (VSSP)		0.061	0.117	.049	0.524	.002
	3	Backward DS (CE)	.076	1.835	0.610	.322	3.011**	.073
		Trail B-A (CE)		-0.010	0.022	-.046	-0.429	.001
Text Reading Fluency	1	Forward DS (PL)	.338	4.243	2.043	.200	2.077*	.023
		Verbal Learning Test (PL)		4.386	1.333	.254	3.290**	.059
	2	Corsi Block Test (VSSP)	.012	2.188	1.335	.127	1.638	.015
		Rey Complex Figure (VSSP)		-0.148	0.403	-.028	-0.366	.001
	3	Backward DS (CE)	.152	11.105	2.102	.463	5.284***	.151
		Trail B-A (CE)		0.057	0.077	.065	0.742	.003
Word Reading Accuracy	1	Forward DS (PL)	.231	0.902	0.549	.189	1.642	.021
		Verbal Learning Test (PL)		0.899	0.359	.230	2.508*	.048
	2	Corsi Block Test (VSSP)	.006	0.010	0.359	.003	0.027	<.001
		Rey Complex Figure (VSSP)		0.032	0.109	.027	0.295	.001
	3	Backward DS (CE)	.057	1.510	0.565	.279	2.672**	.055
		Trail B-A (CE)		-0.007	0.021	-.037	-0.357	.001
Word Spelling Accuracy	1	Forward DS (PL)	.303	1.386	0.593	.254	2.338*	.038
		Verbal Learning Test (PL)		1.071	0.387	.241	2.768**	.053
	2	Corsi Block Test (VSSP)	.018	0.112	0.387	.025	0.289	.001
		Rey Complex Figure (VSSP)		0.116	0.117	.086	0.990	.007
	3	Backward DS (CE)	.045	1.466	0.610	.238	2.404*	.040
		Trail B-A (CE)		-0.015	0.022	-.066	-0.671	.003

Note: $^*p < .05$, $^{**}p < .01$, $^{***}p < .001$; *B*, *SE*, β and *t* values are relative to the last block; pr^2 represents the unique variance of each predictor; DS = Digit Span subtest of the Wechsler Intelligence Scale for Children (Third Edition); PL = phonological loop; VSSP = visuospatial sketchpad; CE = central executive.

Discussion

WM deficits have been widely studied and identified as one of the major defining characteristics of DD. Whereas the deficits in PL and CE tasks exhibited by children with DD have been reported extensively (Schuchardt et al., 2008; Smith-Spark & Fisk, 2007; Swanson et al., 2009), the findings regarding VSSP have been inconsistent (Bacon et al., 2013; Kibby & Cohen, 2008; Menghini et al., 2011).

Thus, the first purpose of the present study was to investigate the specificity of WM deficits in Portuguese children with DD. Consistent with the published literature and our initial hypothesis, the children with DD performed worse than the TR on PL tasks. This finding, which applied to tasks involving both word list recall and digit span tests, suggests that children with DD experience difficulty when required to perform memory tasks involving verbal material. Similarly, de Jong (1998), and Maehler and Schuchardt (2011) found that children with DD performed significantly lower than typically developing children on word span and forward digit span tasks. The WM deficits exhibited by the children with DD were not confined to the PL, CE impairments were also observed. The TR outperformed the children with DD on both CE tasks, indicating that both the storage and processing of information were compromised in the children with DD. These results are consistent with the findings of a recent meta-analysis (Swanson et al., 2009) that revealed particular deficits in verbal STM and CE measures among children with DD. Consistent with other studies (Jeffries & Everatt, 2004; Schuchardt et al., 2008), we did not find significant differences in VSSP between the groups, suggesting that the WM deficits associated with DD are more specific to the PL and CE components. All the group differences in the WM components remained after general intellectual ability was controlled for, suggesting that the observed variations in PL, VSSP and CE were unrelated to differences in intelligence. Swanson et al. (2009) reported a non-significant moderating effect of intelligence on the magnitude of the effect sizes of STM and CE measures between children with and without DD. Contrary to our findings, van der Sluis et al. (2005) reported that when differences in general intelligence were considered, there were no significant differences in WM between children with DD (with or without arithmetic disability) and TR. This finding indicates that group differences in WM may be attributable to differences in general intelligence. Relationships between WM capacity and intellectual ability have also been documented by studies of typically developing children and young adults (Ackerman, Beier, & Boyle, 2005; Cornoldi, Orsini, Cianci, Giofrè, & Pezzuti, 2013).

Because a multivariate main effect of reading group was observed for both the PL and CE tasks, we examined whether differences in the PL might underlie the group differences in the CE. Indeed, after controlling for the PL tasks, the group differences in CE tasks were no longer significant, suggesting that the most relevant WM deficits in DD may be in PL functioning, rather than in the CE. Schuchardt et al. (2008) also observed that when the influence of PL was controlled for, the differences in measures of CE between dyslexic (with or without comorbid dyslexia/dyscalculia) and non-dyslexic children (TR and children with dyscalculia) were no longer significant.

Another purpose of the present study was to analyze the accuracy of WM measures in discriminating between children with DD and TR. There has been limited research utilizing ROC curve and binary logistic regression analyses to study WM deficits in DD. Shifting ability (Trail B-A) and the capacity to maintain (Forward DS) and manipulate (Backward DS) digits in memory revealed a moderate level of diagnostic accuracy. Binary logistic regression analyses also showed that these three tasks were reliable predictors of DD; the PL and CE tasks correctly predicted group membership for 68.7% and 65.6% of the children, respectively. These findings suggest that PL and CE tasks may be adequate measures to correctly discriminate between children with DD and TR in the Portuguese orthography. Obviously, the information obtained from WM measures should only be a component of the DD clinical diagnosis and need to be viewed in the context of a more comprehensive assessment. Similarly, in a recent cross-linguistic study involving six different languages (Finnish, Hungarian, German, Dutch, French, and English) spanning a large range of orthographic complexity, Landerl and colleagues (2013) found that verbal STM/CE was a significant predictor of DD status, independently of the level of orthographic complexity. All these findings highlight the need for future studies to include such statistical analyses; the presence of significant group differences alone does not imply that WM tasks can discriminate between subjects with sufficient accuracy.

The final purpose of the present study was to analyze the predictive effects of WM tasks on reading and spelling abilities. The relative contribution of each WM task to reading and spelling performance was evaluated using hierarchical linear regression analyses. As expected, the VSSP tasks were not significant predictors for any of the dependent variables under study. Conversely, the Verbal Learning Test (PL) and the Backward DS (CE) were significant predictors for all the reading and spelling measures. Concerning reading, the Backward DS was the most robust predictor for the three reading measures (particularly reading fluency, with 15.1% of unique variance), whereas the Forward DS (PL) contributed to only a small but significant proportion of the unique variance in reading fluency alone. In a sample of first-graders, Nevo and Breznitz (2011) also found that the Backward DS task made the largest contribution to the explanation of unique variance in reading accuracy and in reading fluency, whereas the Forward DS task was a non-significant predictor. Similar to the VSSP tasks, the Trail B-A (CE) did not account for a significant degree of unique variance, suggesting that variance in reading is related to the storage and processing of verbal information rather than to shifting or visuospatial STM capacity. Thus, our findings are consistent with those of previous studies reporting links between WM (PL and CE components) and reading ability (Gathercole et al., 2006; Kibby, 2009; Swanson & Ashbaker, 2000). Other studies of typically developing children (Ziegler et al., 2010) and children with DD (Boets et al., 2010) have found that the PL predicted reading ability even after controlling for other neurocognitive variables (phonological awareness and rapid naming) known to be strong predictors of reading.

Compared to reading, the number of studies exploring the relationship between WM and spelling is clearly limited (some exceptions: Service & Turpeinen, 2001; Steinbrink & Klatte, 2008). As Savage et al. (2007, p. 202) noted, this is surprising because “early spelling might thus be expected to tap central executive and phonological loop resources to a greater degree than in early word reading”. The majority of studies analyzing the predictive value of WM measures for spelling ability have used samples of English-speaking children. For instance, Jongejan, Verhoeven, and Siegel (2007) found that verbal WM was a significant predictor of spelling and explained more unique variance in spelling than in reading. Similar to our reading results, we observed that only simple (Forward DS and Verbal Learning Test) and complex (Backward DS) verbal span tasks were significant predictors of spelling in our sample of Portuguese children. Furthermore, WM (particularly PL and CE tasks) explained more variance in spelling than in reading, suggesting that spelling is more dependent on WM resources than is reading accuracy, even in an orthography that is more transparent than English.

Notwithstanding the relevance of the present study described above, there were at least three limitations that should be addressed in future research. First, only two measures were used to assess each WM component. Certainly, the inclusion of more tasks per component would have increased the construct validity and the interpretability of the results. The inclusion of tasks tapping articulatory rehearsal (PL), static visual span (VSSP), and complex visuospatial span (CE) would be particularly relevant to better elucidate the presence of WM impairments in DD. Second, because there is a well-established, considerable overlap between the cognitive processes involved in CE and executive functions, it would be interesting to investigate the contribution of executive function tasks to the CE score differences between children with DD and TR. Third, because WM capacity is also significant impaired in ADHD (Kasper, Alderson, & Hudec, 2012; Katz, Brown, Roth, & Beers, 2011) and in dyscalculia (Landerl et al., 2009; Schuchardt et al., 2008), it would be particularly interesting to compare WM performance between children with DD and children with those two neurodevelopmental disorders.

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Discussão e Conclusão

Discussão e Conclusão

Neste capítulo, da Discussão e Conclusão da presente dissertação, pretende-se sistematizar, integrar e analisar os principais resultados obtidos nos quatro estudos empíricos anteriormente apresentados. Este conjunto articulado de investigações procurou dar resposta ao objetivo geral e aos objetivos específicos inicialmente delineados. Recorde-se que este trabalho de investigação teve como objetivo geral a avaliação das funções neurocognitivas associadas à Dislexia de Desenvolvimento (DD). Pretendeu-se, especificamente, avaliar o desempenho das crianças com DD em medidas de funcionamento intelectual, processamento fonológico, funções executivas e memória de trabalho, de modo a identificar um possível perfil neuropsicológico com adequada sensibilidade de diagnóstico.

A relevância da avaliação destas funções nas crianças com DD decorre da sua natureza neurobiológica e da necessidade de mensurar as funções neurocognitivas que comummente se encontram prejudicadas nesta perturbação da aprendizagem específica. Neste âmbito, a investigação na área das neurociências tem evidenciado desenvolvimentos importantes nos últimos anos, contribuindo para um maior esclarecimento e conhecimento das áreas e vias cerebrais envolvidas e das funções neurocognitivas implicadas. Não obstante o crescente interesse internacional nesta área, em Portugal são ainda escassos os estudos publicados com esta população clínica específica. Só mais recentemente é que surgiram publicações nacionais na área do processamento fonológico (e.g., Araújo, Faísca, Bramão, Petersson, & Reis, 2014; Araújo et al., 2011; Araújo, Pacheco, Faísca, Petersson, & Reis, 2010; Sucena, Castro, & Seymour, 2009) e nalguns domínios da neuropsicologia (Silva, Silva, & Martins, 2014). Acresce ainda que a DD é uma das perturbações infantis mais prevalentes, em Portugal estima-se que ocorra em cerca de 5.4% das crianças em idade escolar (Vale, Sucena, & Viana, 2011), pelo que claramente se justifica a presente investigação.

A escolha dos critérios de inclusão das crianças com DD foi uma questão particularmente refletida de modo a minimizar a presença de falsos positivos na amostra. Após uma extensa revisão dos critérios de inclusão e exclusão utilizados em estudos similares, optou-se por uma abordagem mais conservadora. Os critérios relativos aos pontos-de-corte e aos instrumentos utilizados nas investigações são bastante díspares, o que pode conduzir a alguns enviesamentos e a dificuldades na comparação de resultados entre estudos. Por exemplo, Frijters et al. (2011) adotaram como critério para a delimitação do grupo com DD um QI ≥ 70 e um desempenho inferior a -1 desvio-padrão (DP) em pelo menos um dos três índices de leitura avaliados, enquanto Swanson (2011) utilizou o critério de QI > 90 e um percentil < 25 (-0.65 DP) em teste de leitura. Por sua vez, Schuchardt, Maehler e Hasselhorn (2008) consideraram como critério de inclusão um QI ≥ 80 e um percentil < 16 (-1 DP) em teste de leitura; enquanto Reiter, Tucha e Lange (2005) definiram um QI > 90 e um desempenho inferior ao percentil 5 (-1.65 DP) ou 16 (-1 DP) num conjunto de testes de leitura.

A Associação Americana de Psiquiatria, na sua recente revisão do Manual de Diagnóstico e Estatística das Perturbações Mentais (DSM-5; American Psychiatric Association, 2013), estabelece como critérios de diagnóstico um desempenho na fluência, na precisão e/ou na compreensão da leitura substancialmente abaixo do que o tipicamente observado em crianças da mesma idade cronológica. O desempenho nestas medidas deverá ser quantificável através de instrumentos estandardizados e psicometricamente validados, aplicados individualmente, com resultados inferiores a -1.5 DP ou percentil < 7 (mas podem assumir outros pontos-de-corte, nomeadamente entre -1 DP a -2.5 DP) comparativamente com o esperado para a idade cronológica da criança, associado a um funcionamento intelectual normativo (QI > 70).

Tal como referido nos quatro estudos que integram a presente investigação, a abordagem utilizada para a delimitação do grupo de crianças com DD teve em conta múltiplos e rigorosos critérios de inclusão e exclusão. Obviamente que a opção por esta abordagem mais conservadora na delimitação do grupo de crianças com DD permitiu a diminuição do erro Tipo I (i.e., falsos positivos), mas inversamente, aumentou a possibilidade de erro Tipo II (i.e., falsos negativos; crianças disléxicas com quadros sintomatológicos mais ligeiros poderão ter sido excluídas da amostra final).

A literatura tem vindo a demonstrar a natureza multifatorial da DD, onde as crianças disléxicas revelam dificuldades em alguns subtestes intelectuais, no processamento fonológico, nas funções executivas e na memória de trabalho. Apesar da relevância destas funções neurocognitivas, o défice no processamento fonológico (em particular na consciência fonológica e na nomeação rápida) surge como a principal característica fenotípica da DD e o preditor mais consistente para explicar a variância da leitura nos diversos sistemas ortográficos.

De seguida, iremos discutir de forma integrada os principais resultados e conclusões obtidos. A discussão dos resultados encontra-se estruturada em função das variáveis neuropsicológicas avaliadas, congruentes com a sequência pela qual foram apresentados os estudos.

Perfis Cognitivos da WISC-III

As escalas de inteligência de Wechsler para crianças (WISC, WISC-R, WISC-III e WISC-IV) são, provavelmente, as provas mais amplamente utilizadas nos estudos que englobam crianças com DD. Seja pela sua utilidade na delimitação do grupo disléxico [os resultados do QI Escala Completa (QIEC) da WISC são utilizados nos critérios de inclusão/exclusão] ou pela relevância de alguns dos seus subtestes na mensuração de funções neurocognitivas específicas comummente deficitárias nas crianças com DD (por exemplo, na memória de trabalho, nas funções executivas e nas habilidades verbais).

Se, inicialmente, a discrepância entre a capacidade intelectual geral e o desempenho na leitura nas crianças com DD foi um fator importante no diagnóstico, estudos posteriores vieram colocar em causa a validade deste critério (Siegel, 1989, 1992). Atualmente, não é frequente a utilização do critério de discrepância nos estudos empíricos, pois aumenta a presença de falsos positivos e falsos negativos na amostra. Por outro lado, diversos estudos têm demonstrado a existência de uma correlação baixa a moderada entre a capacidade intelectual e o desempenho da leitura (Caravolas, Volín, & Hulme, 2005; Compton, Defries, & Olson, 2001; Gathercole, Alloway, Willis, & Adams, 2006), e o seu reduzido poder preditivo quando entram na equação da regressão outras variáveis neurocognitivas (Boets et al., 2010; Caravolas et al., 2005; Nevo & Breznitz, 2011). De facto, os resultados do **Estudo 2** confirmam estes indicadores, a WISC-III QIEC apresenta coeficientes de correlação baixos a moderados com as diversas medidas de leitura e com as funções do processamento fonológico (exceto com prova Consciência Fonológica – Substituição). Por outro lado, mesmo após se controlar através de análise de covariância a capacidade intelectual geral, as crianças com DD exibem défices significativos nas diversas provas de leitura (**Estudo 2**), no processamento fonológico (**Estudo 2**), nas funções executivas (**Estudo 3**) e na memória de trabalho (**Estudo 4**).

Apesar do progressivo abandono do critério de discrepância entre a capacidade intelectual geral e o desempenho da leitura, a análise de perfis cognitivos dos subtestes que compõem a WISC têm sido alvo de um estudo mais sistemático, uma vez que alguns dos subtestes estão associados a funções neurocognitivas de relevante mensuração na DD. Muito embora a análise de perfis cognitivos seja bastante explorada na investigação [mais de 75 perfis cognitivos distintos já foram identificados em estudos com a WISC (para uma revisão crítica, consultar McDermott, Fantuzzo, & Glutting, 1990)] e muito utilizada na avaliação psicológica[10], a sua validade no diagnóstico das perturbações da aprendizagem específicas tem produzido resultados inconsistentes. Como abordado no **Estudo 1**, enquanto alguns estudos demonstram a pouca utilidade dos perfis cognitivos da WISC no diagnóstico de crianças com DD ou com dificuldades de aprendizagem específicas (Rotsika et al., 2009; Watkins, Kush, & Glutting, 1997a, 1997b), outros estudos

[10] Pfeiffer, Reddy, Kletzel, Schmelzer e Boyer (2000) verificaram que a larga maioria dos psicólogos escolares norte-americanos consideravam a WISC-III muito útil no diagnóstico de certas perturbações, 89% utilizavam com regularidade a análise de perfis cognitivos e 70% consideravam-na a característica mais importante da WISC-III.

obtiveram índices de sensibilidade e especificidade bastante satisfatórios (Prifitera & Dersh, 1993; Thomson, 2003). Dos diversos perfis estudados, os que têm evidenciado uma maior validade de diagnóstico na DD são o perfil Bannatyne, o fator Resistência à Distração, e os perfis ACID e SCAD.

Os resultados do **Estudo 1**, apesar de apontarem para diferenças significativas entre as crianças com DD e as crianças leitoras normais nos subtestes que compõem estes perfis cognitivos, confirmam o baixo poder discriminativo dos perfis completos no diagnóstico da DD (por exemplo, o perfil completo ACID apresenta uma sensibilidade de 8%). Contudo, a análise dos perfis parciais e das suas medidas compósitas demonstram uma maior precisão (por exemplo, a presença perfil ACID nos seis subtestes mais baixos revela uma sensibilidade de 45%). Até à data, não são conhecidos estudos que tenham determinado um ponto-de-corte ótimo das medidas compósitas dos perfis ACID, SCAD e do fator Resistência à Distração. Esta análise foi realizada no **Estudo 1** a partir dos resultados das curvas '*receiver operating characteristic'* (ROC) e do cálculo do índice de Youdan (1950). O ponto-de-corte ótimo do perfil ACID situa-se nos 37.50 valores padronizados, classificando corretamente 67% das crianças com DD (sensibilidade) e 90% das crianças leitoras normais (especificidade). Valores de sensibilidade e de especificidade relativamente aproximados são encontrados no fator Resistência à Distração, enquanto o perfil SCAD apresenta um índice de Youden mais baixo comparativamente com estes dois perfis.

Os subtestes da WISC-III que compõem estes perfis remetem para os processos executivos (exceto o subteste Informação do perfil ACID), em particular para a velocidade de processamento e para a memória de trabalho. O **Estudo 3** e o **Estudo 4** analisaram, justamente, o desempenho das crianças com DD num conjunto de testes para mensuração das funções executivas e da memória de trabalho, para além de explorarem o seu valor preditivo no diagnóstico da DD. Os resultados destes dois estudos, que serão discutidos mais à frente, evidenciam, do ponto de vista empírico, a validade destas medidas na discriminação das crianças com DD.

Processamento Fonológico

A importância do processamento fonológico no desenvolvimento da leitura tem sido demonstrada em estudos transversais (Katzir, Schiff, & Kim, 2012; Vaessen et al., 2010; Ziegler et al., 2010), longitudinais (Cardoso-Martins & Pennington, 2004; Furnes & Samuelsson, 2010; Kirby, Parrila, & Pfeiffer, 2003) e de modelos de equações estruturais (Caravolas, Lervåg, Defior, Seidlová Málková, & Hulme, 2013; Caravolas et al., 2005).

A hipótese do défice fonológico na DD tem sido amplamente aceite (Fletcher, 2009; Ramus, Marshall, Rosen, & van der Lely, 2013; Vellutino, Fletcher, Snowling, & Scanlon, 2004) e suportada por estudos de neuroimagem (Boets et al., 2013; Finn et al., 2014). Apesar das funções do processamento fonológico serem consistentemente as variáveis mais preditoras das dificuldades na leitura das crianças com DD (Ackerman, Holloway, Youngdahl, & Dykman, 2001; Boets et al., 2010; Vukovic & Siegel, 2006) e sensíveis no diagnóstico (Landerl et al., 2013), o nível de opacidade/transparência do sistema ortográfico das diferentes línguas pode influenciar o desempenho na leitura e a expressão sintomatológica na DD. Assim, no **Estudo 2** procurou-se, justamente, analisar o desempenho e a associação entre o processamento fonológico e a leitura num grupo de crianças com DD e dois grupos de controlo (por idade cronológica e por idade de leitura). Sendo o Português Europeu um sistema ortográfico de opacidade intermédia (Seymour, Aro, & Erskine, 2003; Sucena et al., 2009), torna-se relevante compreender as alterações psicolinguísticas (eventualmente idiossincráticas) das crianças disléxicas Portuguesas, uma vez que a grande maioria dos estudos publicados é efetuada em sistemas ortográficos transparentes ou de elevada opacidade. Com efeito, o estudo dos fatores neurocognitivos envolvidos no desenvolvimento da leitura e na DD permite aos investigadores compreenderem quais os fatores que são universais e os que são específicos de cada sistema ortográfico.

Os resultados da análise inferencial do **Estudo 2** fornecem evidências sobre os défices na consciência fonológica, na nomeação rápida e na memória verbal imediata das crianças com DD (a magnitude das diferenças entre os grupos é bastante elevada), sendo consistente com o observado em estudos nacionais (Araújo et al., 2010; Sucena et al.,

2009) e internacionais (Boets et al., 2010; Caravolas et al., 2005; Martin et al., 2010). No **Estudo 2** procurou-se, ainda, avaliar a relevância destas medidas no diagnóstico da DD. Surpreendentemente, são em número reduzido os estudos que têm incluído a análise de curvas ROC, a análise de regressão logística ou a análise de resultados anormalmente baixos aquando da avaliação do processamento fonológico em crianças com DD. Uma das exceções é o recente artigo de Landerl et al. (2013) que analisou a capacidade preditora do processamento fonológico no diagnóstico da DD em cinco ortografias de diferentes níveis de opacidade (Inglês, Francês, Holandês, Alemão e Finlandês). Os autores constataram que a consciência fonológica e a nomeação rápida foram relevantes preditores da DD (o poder preditivo aumenta com a maior opacidade do sistema ortográfico), enquanto a memória verbal imediata apresentou um valor preditivo mais reduzido. Os resultados da análise de curvas ROC do **Estudo 2** também evidenciam a elevada precisão no diagnóstico da consciência fonológica e da nomeação rápida na discriminação das crianças com DD, e uma moderada precisão da memória verbal imediata. A elevada frequência de resultados anormalmente baixos nas duas provas de consciência fonológica e na nomeação rápida no grupo de crianças com DD (41.7% para $z < -1$ e de 16.7% para $z < -2$ nos 3 testes analisados) comparativamente com o observado nos grupos de controlo (não foram observados casos) e na população geral (1.87% e 0.02%, respetivamente), reforçam a relevância da mensuração do processamento fonológico na DD.

Alguns investigadores têm levantado a hipótese da nomeação rápida poder ser um marcador mais fiável no diagnóstico da DD (Kirby, Georgiou, Martinussen, & Parrila, 2010; Snowling, 2006) e um preditor mais significativo do desempenho da leitura em sistemas ortográficos mais transparentes comparativamente com a consciência fonológica (de Jong & van der Leij, 2003). Os resultados do **Estudo 2** não suportam esta hipótese pelas seguintes razões: (1) a magnitude do efeito das diferenças entre os grupos é superior nas duas provas de consciência fonológica comparativamente com a nomeação rápida; (2) os valores de '*area under the curve*' (AUC) provenientes da análise das curvas ROC são mais elevados nas provas de consciência fonológica; e (3) os resultados da análise de regressão linear demonstram que a consciência fonológica é o preditor mais relevante da precisão da leitura de texto e de palavras isoladas, estando a nomeação rápida claramente associada à fluência da leitura. De modo similar, estudos translinguísticos que incluíram amostras de crianças leitoras normais Portuguesas demonstraram a importância do processamento fonológico no desenvolvimento da leitura, onde a consciência fonológica surge como o principal preditor "universal" da aprendizagem da leitura (Vaessen et al., 2010; Ziegler et al., 2010). Sucena et al. (2009) também verificaram que a consciência fonológica contribui de modo mais significativo para explicar as dificuldades na leitura de crianças Portuguesas com DD do que a nomeação rápida. Os resultados do **Estudo 2** parecem ainda sugerir, que apesar do Português Europeu ser uma ortografia de opacidade intermédia, poderá estar mais próximo do modelo ortográfico inglês (opaco) que do modelo alemão (transparente), uma vez que ocorre uma maior exigência sobre os processos de descodificação fonológica dada a maior inconsistência das regras de conversão grafema-fonema (Seymour et al., 2003; Sucena et al., 2009).

Outro dado empírico interessante proveniente da análise de regressão linear, está relacionado com o facto da nomeação rápida explicar a variância na leitura de palavras regulares e irregulares, mas não contribuir significativamente para explicar a variância na leitura de pseudopalavras. Segundo o modelo de dupla via, a via lexical está envolvida na leitura de palavras regulares e irregulares, o que parece sugerir que a nomeação rápida estará mais associada com o processamento lexical (ou ortográfico) do que com a descodificação sublexical (ou fonológica). Este resultado tem sido igualmente encontrado em outros estudos (Bowers & Ishaik, 2003; Bowers & Newby-Clark, 2002; Kirby et al., 2010).

Por fim, no **Estudo 2** foi observado um défice no desenvolvimento da leitura (i.e., precisão da leitura significativamente inferior ao obtido pelas crianças leitoras normais com a mesma idade cronológica e com a mesma idade de leitura) e acentuadas dificuldades na descodificação das pseudopalavras por parte das crianças com DD (apenas 58% de precisão). Por outro lado, foi obtido um efeito de lexicalidade e um efeito de regularidade, sendo consistente com o observado em dois outros estudos nacionais (Araújo et al., 2014; Sucena et al., 2009). Contudo, quando se comparou o efeito de lexicalidade e o efeito de regularidade entre os três grupos, apenas foram encontradas diferenças significativas no efeito de lexicalidade, o que parece sugerir um maior défice da estratégia de descodificação sublexical (ou fonológica) comparativamente com o processamento lexical (ou ortográfico) nas crianças Portuguesas com DD. Este padrão de resultados tem sido, sobretudo, encontrado em ortografias de intermédia (Sprenger-Charolles, Colé, Kipffer-Piquard, Pinton, & Billard, 2009; Sucena et al., 2009) ou elevada opacidade (Herrmann, Matyas, & Pratt, 2006). Mais uma vez, este conjunto de indicadores demonstra a importância dos défices fonológicos no desempenho da leitura das crianças disléxicas.

Comparativamente com os subtestes e perfis cognitivos (**Estudo 1**), funcionamento executivo (**Estudo 3**) e memória de trabalho (**Estudo 4**), as funções do processamento fonológico (**Estudo 2**) são as que evidenciam uma maior magnitude do efeito das diferenças entre os grupos, maior capacidade discriminativa do grupo das crianças com DD e maior variância explicada da leitura. Tal como observado noutros sistemas ortográficos com diferentes níveis de opacidade, estes resultados parecem confirmar que os défices no processamento fonológico são a mais importante característica neurocognitiva na DD. Por isso, é manifesta a necessidade de incluir instrumentos de medida específicos, para a mensuração do processamento fonológico, nos protocolos de avaliação neuropsicológica na DD.

Funções Executivas

Dada a existência de dificuldades nos subtestes da WISC-III que remetem para a velocidade de processamento e para a memória de trabalho nas crianças com DD (**Estudo 1**) e dos modelos que sugerem a partilha de múltiplos défices cognitivos entre as principais perturbações neurodesenvolvimentais (Pennington, 2006; Willcutt, Pennington, Olson, Chhabildas, & Hulslander, 2005; Willcutt et al., 2013; Willcutt, Sonuga-Barke, Nigg, & Sergeant, 2008), o estudo do funcionamento executivo, na presente investigação, afigurou-se indispensável para uma compreensão mais exaustiva das alterações neuropsicológicas associadas à DD (**Estudos 3 e 4**).

Nesta sentido, importa assinalar que se encontra relativamente bem documentado na literatura que as crianças com DD revelam défices em várias funções executivas, muito embora os estudos sejam discordantes sobre quais os processos executivos que se encontram comprometidos (Brosnan et al., 2002; Helland & Asbjørnsen, 2000; Reiter et al., 2005; Varvara, Varuzza, Sorrentino, Vicari, & Menghini, 2014).

Os resultados do **Estudo 1** e do **Estudo 3** (que inclui adicionalmente o teste Trail-A) revelam diferenças significativas na velocidade de processamento entre o grupo com DD e os leitores normais. De facto, dificuldades na velocidade de processamento têm sido observadas em crianças com DD quando comparadas com controlos por idade cronológica (Peng, Sha, & Li, 2013; Willcutt et al., 2005) e com crianças com Perturbação de Hiperatividade com Défice de Atenção (Shanahan et al., 2006), o mesmo não ocorrendo quando comparadas com crianças com Discalculia (Willcutt et al., 2013). Num recente estudo com crianças Portuguesas com DD foram, igualmente, observados défices na prova Pesquisa de Símbolos da WISC-III e na prova Trail-A (Silva et al., 2014). Estudos que incorporam provas de velocidade de processamento com uma componente verbal associada (por exemplo, prova de nomeação rápida similar à utilizada no **Estudo 2**) têm observado défices mais significativos do que as provas sem a componente verbal (Moll, Göbel, Gooch, Landerl, & Snowling, 2014; Shanahan et al., 2006). De referir, ainda, que as provas de velocidade de processamento parecem ser medidas mais fiáveis no diagnóstico de DD enquanto medida compósita combinada com provas de memória de trabalho (**Estudo 1**: perfil SCAD com um AUC de .862) do que isoladamente (**Estudo 3**: Código com um AUC de .670 e a Pesquisa de Símbolos com um AUC de .663).

A flexibilidade é um dos mais importantes processos executivos (Miyake et al., 2000) e uma das funções da componente executiva da memória de trabalho (Baddeley, 1996), estando, por isso, incluída no **Estudo 3** e no **Estudo 4**. Os resultados de ambos os estudos reforçam a importância da avaliação da flexibilidade na DD. São observadas diferenças significativas na flexibilidade entre os grupos (**Estudos 3 e 4**) e uma moderada precisão na discriminação das crianças com DD (**Estudos 3 e 4**). Por outro lado, a flexibilidade é a única função executiva relevante para o diagnóstico da DD quando controlado o efeito dos restantes processos executivos (por cada 10 segundos adicionais o risco de ser classificado no grupo disléxico aumenta 16.1%) (**Estudo 3**) e uma das funções da memória de trabalho preditoras do diagnóstico de DD (**Estudo 4**). Apesar destes resultados, a flexibilidade não contribui adicionalmente para explicar a variância no desempenho da leitura e da escrita quando o efeito das restantes componentes da memória de trabalho é estatisticamente controlado (**Estudo 4**).

O planeamento é outro dos mais importantes processos executivos (Welsh, Pennington, & Groisser, 1991). Ao contrário do verificado para a flexibilidade, a revisão da literatura demonstra resultados algo inconsistentes no que à DD diz respeito (Brosnan et al., 2002; Condor, Anderson, & Saling, 1995; Marzocchi et al., 2008; Reiter et al., 2005). No **Estudo 3** não foram encontradas diferenças significativas em nenhuma das tarefas de planeamento entre o grupo com DD e o grupo de controlo, e a capacidade de precisão de diagnóstico foi bastante reduzida. Contudo, futuros estudos deverão explorar melhor esta associação, pois um "efeito de teto" foi observado nos dois grupos no teste da Torre da Bateria de Avaliação Neuropsicológica de Coimbra (BANC; Simões et al., in press). Este "efeito de teto" também se verifica na amostra de normalização da BANC para os diversos grupos etários. Fica, assim, por compreender se a ausência de diferenças entre os grupos no planeamento resulta das características fenotípicas das crianças com DD ou de um erro de medida.

Relativamente à fluência verbal, os resultados encontrados no **Estudo 3** estão em consonância com o tipicamente observado em estudos similares. As crianças com DD produzem menos palavras na tarefa semântica e fonémica em comparação com as crianças leitoras normais. O número de palavras produzidas na tarefa fonémica é significativamente inferior ao obtido na tarefa semântica em ambos os grupos, uma vez que a primeira está mais dependente da maturação do lobo frontal, requer a exploração de mais subcategorias e encontra-se sobretudo associada à estratégia de alternância (Birn et al., 2010; Hurks, 2012; Riva, Nichelli, & Devoti, 2000; Troyer, Moskovitch, & Winocur, 1997). Por outro lado, as diferenças no número de palavras produzidas entre o grupo disléxico e o grupo de controlo são significativas apenas nos 30 segundos iniciais, o que parece remeter para um défice no processamento automático segundo o modelo de organização lexical proposto por Crowe (1998). Não são conhecidas análises temporais em estudos com crianças com DD, mas resultados similares foram encontrados em duas investigações com grupos de crianças com Perturbação de Hiperatividade com Défice de Atenção (Hurks et al., 2004; Takács, Kóbor, Tárnok, & Csépe, 2014).

Todos estes indicadores demonstram a presença de défices específicos no funcionamento executivo, muito embora a sua capacidade de precisão e de predição no diagnóstico de DD (**Estudo 3**) seja inferior à observada no processamento fonológico (**Estudo 2**) e na memória de trabalho (**Estudo 4**). Sendo as dificuldades nas funções executivas uma componente comum às várias perturbações neurodesenvolvimentais, o estudo do funcionamento executivo na DD poderá ser relevante para uma compreensão mais abrangente das alterações neuropsicológicas associadas e da sua relação comórbida com as restantes perturbações.

Memória de Trabalho

O estudo da relação entre a memória de trabalho e o desempenho da leitura e os seus défices na DD (**Estudo 4**) representam um desenvolvimento lógico dos resultados obtidos nos estudos anteriores. Um conjunto muito alargado de investigações tem demonstrado a importância da componente de armazenamento verbal e da componente executiva na DD (para uma revisão: Swanson, Zheng, & Jerman, 2009) e a sua influência na descodificação (Kibby, 2009; Perez, Majerus, & Poncelet, 2012), na fluência (Swanson & Jerman, 2007) e na compreensão da leitura (Goff, Pratt, & Ong, 2005; Swanson, 2011). Os resultados do **Estudo 4** confirmam as dificuldades das crianças disléxicas Portuguesas nestas duas componentes da memória de trabalho, tendo sido observado um desempenho normativo na componente visuoespacial (tal como esperado na revisão da literatura; e.g., Bacon, Parmentier, & Barr, 2013; Kibby & Cohen, 2008; Schuchardt et al., 2008). Os dados obtidos sugerem ainda que as alterações na memória de trabalho nas crianças com DD estão particularmente dependentes da componente de armazenamento verbal, uma vez que controlando estatisticamente esta variável deixam de ser observadas diferenças significativas (p = .062) na componente executiva (similar ao obtido por: Schuchardt et al., 2008).

Muito embora no **Estudo 4** tenham sido incluídas provas adicionais para mensuração da memória verbal imediata (no **Estudo 2** apenas foi incluída a Memória de Dígitos no sentido direto) e da componente executiva (no **Estudo 3** apenas foi incluído o Trail-B) os resultados das análises das curvas ROC e da regressão logística continuam a demonstrar uma moderada precisão e predição destas duas componentes na discriminação das crianças com DD. Tal como no estudo de Landerl et al. (2013), apesar destas duas componentes da memória de trabalho serem medidas fiáveis no diagnóstico da DD, demonstram uma capacidade inferior à observada pela consciência fonológica e nomeação rápida (**Estudo 2**).

Os resultados da análise de regressão linear hierárquica fornecem informações adicionais sobre a influência da memória de trabalho no desempenho da leitura e escrita. Assim, a variância da leitura (precisão na leitura de texto, precisão na leitura de palavras isoladas e fluência da leitura) está unicamente dependente do armazenamento e do processamento verbal (a flexibilidade e a componente de armazenamento visuoespacial não são preditores significativos). Cruzando estes dados com os obtidos no **Estudo 2**, verifica-se que o subteste da Memória de Dígitos no sentido direto é um preditor significativo da fluência da leitura apenas no **Estudo 4**, muito embora esteja significativamente correlacionado com as diversas medidas de leitura (**Estudo 2**). Com efeito, alguns estudos têm documentado que a memória verbal imediata contribui de modo significativo para explicar a variância na leitura (Kibby, 2009; Perez et al., 2012), deixando de ser uma variável preditora quando entram no modelo a consciência fonológica e a nomeação rápida (Boets et al., 2010; de Jong & van der Leij, 1999; Parrila, Kirby, & McQuarrie, 2004). Este resultado sugere que a memória verbal imediata partilha a sua variância com as outras provas do processamento fonológico (Parrila et al., 2004).

A associação entre a memória de trabalho e a escrita está claramente menos explorada na literatura, o que não deixa de ser surpreendente uma vez que a memória de trabalho parece desempenhar um papel mais relevante nos processos de escrita do que na leitura (Jongejan, Verhoeven, & Siegel, 2007; Savage, Lavers, & Pillay, 2007). Mais uma vez, apenas o armazenamento e o processamento verbal surgem como variáveis preditoras da precisão da escrita, reforçando a relevância das dimensões verbais (processamento fonológico e memória de trabalho) na DD e no desempenho da leitura e da escrita. Tal como esperado, a variância explicada na precisão da escrita (36.6%) é superior à observada na precisão da leitura (25.9% e 29.4%), mas inferior à obtida na fluência da leitura (50.2%).

Em suma, os resultados integrados destes quatro estudos demonstram a natureza multifatorial e neurodesenvolvimental da DD, reforçando a necessidade da inclusão de medidas neuropsicológicas específicas em protocolos de avaliação. Para além das alterações nos processos de descodificação sublexical (ou fonológica) e de processamento lexical (ou ortográfico) da leitura, os défices na consciência fonológica, na nomeação rápida e na memória de trabalho (componente de armazenamento fonológico e componente executiva) parecem constituir-se como as medidas mais fiáveis e preditoras da DD.

Relevância e Limites da Investigação. Estudos Futuros.

Em termos dos procedimentos metodológicos desta investigação, salienta-se a criteriosa seleção dos instrumentos utilizados para a mensuração das diversas funções neurocognitivas. Com a exceção da versão Portuguesa da *Psycholinguistic Assessment of Language* (PAL-PORT; Festas, Martins, & Leitão, 2007), todos os instrumentos utilizados estão validados e normalizados para a população Portuguesa, e apresentam adequadas propriedades psicométricas. Por outro lado, para cada uma das funções neurocognitivas estudadas procurámos selecionar os instrumentos mais fiáveis e comummente utilizados na prática clínica e nos estudos empíricos.

Relativamente à amostra, salientamos a rigorosa seleção das crianças com DD e das crianças leitoras normais. Além dos exigentes critérios de inclusão e exclusão para ambos os grupos, foi efetuado um cuidadoso emparelhamento do grupo de crianças disléxicas com o grupo de controlo em função das variáveis género, idade e nível de escolaridade. De realçar que, quer para o grupo disléxico, quer para o grupo de leitores normais, as crianças sem diagnóstico bem definido, em situação de comorbilidade ou que não cumprissem os critérios de inclusão foram excluídos dos estudos, aumentando assim a homogeneidade dos grupos e a validade das conclusões obtidas. De referir, ainda, a inclusão de um grupo de controlo por idade de leitura no **Estudo 2**, procedimento metodológico pouco frequente em estudos nacionais (algumas exceções: Araújo et al., 2011; Sucena et al., 2009).

O número de funções neurocognitivas analisadas é outro dos pontos fortes do presente conjunto de investigações. As análises não se limitaram a explorar as alterações na leitura e no processamento fonológico (**Estudo 2**), mas estenderam os estudos ao funcionamento intelectual e aos perfis cognitivos (**Estudo 1**), às funções executivas (**Estudo 3**) e à memória de trabalho (**Estudo 4**). Não são conhecidos estudos nacionais que tenham procedido a uma análise mais completa das funções neurocognitivas avaliadas no **Estudos 1, 3 e 4**. Para cada um dos estudos, foi efetuada uma análise das diferenças entre os grupos e avaliada a capacidade preditora no diagnóstico da DD dos testes neuropsicológicos aplicados, permitindo, assim, uma comparação mais rigorosa dos resultados.

Salienta-se, ainda, a utilização de procedimentos estatísticos pouco habituais em estudos congéneres, que permitiram uma maior exploração e refinamento dos resultados. São disso exemplo, o cálculo do ponto-de-corte ótimo dos perfis cognitivos através do índice de Youdan (**Estudo 1**), a utilização de curvas ROC (**Estudos 1, 2, 3 e 4**), a análise de resultados anormalmente baixos (**Estudo 2**), a análise de regressão logística (**Estudos 3 e 4**) e a análise de covariância para controlar o possível efeito da capacidade intelectual geral (**Estudos 2, 3 e 4**). Por outro lado, os resultados obtidos da presente investigação são, no nosso entender, relevantes para a literatura. Seja pela inclusão de uma medida de memória verbal imediata (**Estudo 2** e **Estudo 4**) para mensuração da terceira componente do processamento fonológico proposta por Torgesen, Wagner e colaboradores (Torgesen, Wagner, & Rashotte, 1994; Wagner & Torgesen, 1987; Wagner, Torgesen, Laughon, Simmons, & Rashotte, 1993) (mais uma vez, não é conhecida a sua inclusão em estudos nacionais); pela análise da precisão de diagnóstico das funções neurocognitivas (**Estudos 1, 2, 3 e 4**); ou pelo estudo do modelo de organização lexical proposto por Crowe (1998) aquando da avaliação da fluência verbal (**Estudo 3**) (não são conhecidos estudos nacionais e internacionais que tenham realizado esta análise na DD).

Não obstante os pontos fortes anteriormente identificados, este grupo de pesquisas apresenta algumas limitações que importa controlar em futuros estudos. Assim, a ausência de outros grupos clínicos com elevada comorbilidade com a DD (por exemplo, a Perturbação de Hiperatividade com Défice de Atenção e a Discalculia) é uma limitação desta investigação. A sua inclusão acrescentaria validade e interpretabilidade dos resultados, uma vez que permitiria comparar e analisar as alterações neuropsicológicas que são específicas da DD e aquelas que são partilhadas com outras perturbações neurodesenvolvimentais.

A avaliação do tempo de latência da leitura de palavras isoladas seria uma variável importante a incluir nesta investigação. É conhecida a tendência para se observar um "efeito de teto" no índice de precisão da leitura de palavras em ortografias mais transparentes (Jiménez, Rodríguez, & Ramírez, 2009; Sprenger-Charolles, Colé, Lacert, & Serniclaes, 2000; Wolff, 2009), pelo que o uso combinado destas duas medidas (precisão e tempo de latência da leitura) contribuiria para uma avaliação mais fiável dos processos de descodificação sublexical (ou fonológica) e do processamento lexical (ou ortográfico).

Teria sido, igualmente, relevante o estudo da prevalência dos subtipos de DD (Dislexia Fonológica e Dislexia de Superfície) através da metodologia clássica[11] e do método de regressão[12]. Uma vez que estes dois subtipos de DD apresentam padrões de

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[11] A classificação dos subtipos de DD na metodologia clássica é efectuada a partir do cálculo de um ponto-de-corte (-1 DP) para a leitura de palavras irregulares e de um ponto-de-corte (-1 DP) para a leitura de pseudopalavras tendo por referência os resultados do grupo de controlo por idade cronológica e/ou do grupo de controlo por idade de leitura.

[12] A classificação dos subtipos de DD no método de regressão é efectuada a partir do estabelecimento de duas linhas de regressão com intervalo de confiança a 90% ou a 95% (regressão da leitura de palavras

leitura distintos (as crianças com Dislexia Fonológica revelam uma dificuldade seletiva na via sublexical manifestada por dificuldades na leitura de pseudopalavras, enquanto as crianças com Dislexia de Superfície revelam uma dificuldade seletiva na via lexical manifestada por dificuldades na leitura de palavras irregulares), teria sido importante analisar e comparar as características neurolinguísticas destes dois grupos.

Na consecução deste projeto de investigação, várias interrogações e hipóteses foram surgindo, dando oportunidade para o possível desenvolvimento de novos estudos. Algumas destas interrogações darão origem a estudos que já se encontram numa fase avançada, estando outros em fase de preparação. Assim, destacam-se: (1) estudo da prevalência dos subtipos de DD (Dislexia Fonológica e Dislexia de Superfície) e análise do seu funcionamento neurolinguístico; (2) análise do funcionamento executivo e das componentes da memória de trabalho entre as crianças com DD e as crianças com Perturbação de Hiperatividade com Défice de Atenção, alargando assim a validade e a interpretabilidade dos resultados encontrados nos **Estudos 3 e 4**; (3) desenvolver um estudo com uma metodologia longitudinal para determinar as variáveis mais preditoras nos diferentes períodos de desenvolvimento da leitura; (4) o alargamento do estudo do funcionamento neuropsicológico das crianças com DD aos restantes testes incluídos na BANC (Simões et al., in press), examinando a sensibilidade dos diversos testes e das medidas compósitas no diagnóstico desta perturbação da aprendizagem específica; e (5) desenvolvimento de uma bateria/protocolo de avaliação da DD, que incorpore medidas de processamento fonológico, de memória de trabalho e provas de leitura e escrita.

Conclusões Finais

Sendo a DD uma das perturbações neurodesenvolvimentais mais prevalentes (American Psychiatric Association, 2013), torna-se essencial proceder a um estudo mais completo das alterações neuropsicológicas que lhe estão associadas para um rigoroso

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irregulares sobre a leitura de pseudopalavras e a regressão da leitura de pseudopalavras sobre a leitura de palavras irregulares) tendo por referência os resultados do grupo de controlo por idade cronológica e/ou do grupo de controlo por idade de leitura.

diagnóstico e adequada intervenção. Apesar de 5.4% das crianças Portuguesas em idade escolar poderem apresentar esta perturbação (Vale et al., 2011), o número de estudos nacionais publicados nesta área é ainda residual, legitimando as propostas de investigação que apresentamos.

As investigações realizadas pretenderam, de modo articulado, analisar o prejuízo e a sensibilidade no diagnóstico das funções neurocognitivas na DD. Os resultados obtidos confirmam a presença de défices específicos no processamento fonológico, na memória de trabalho e nas funções executivas, demonstrando a natureza multifatorial e neurodesenvolvimental desta perturbação. Todos estes indicadores reforçam a importância da inclusão de medidas neuropsicológicas específicas em protocolos de avaliação na DD, para além dos tradicionais testes de leitura e escrita, para uma definição mais rigorosa do diagnóstico.

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