Language-based risk factors in children with developmental dyslexia: A systematic review

by

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I would like to express my sincere gratitude to the following people who supported me in attaining this milestone:

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Abstract

Background: Developmental dyslexia is a wide-ranging and persistent neurobiological disorder classified under specific learning disorders. The literature widely recognises the multifactorial nature of developmental dyslexia, specifically the language-related origin. *Objectives:* The current review systematically identified and synthesised possible language-based risk factors associated with developmental dyslexia in children (age one to three and four to ten years) and determined the level of evidence associated with these risk factors. The review aimed to identify individualised risk factors in children with developmental dyslexia to promote optimal neurodevelopmental outcomes. *Method:* Four relevant databases were searched from January 2002 until December 2018. A hand search of the included articles' reference lists was also performed to identify any relevant publication. The language-based risk factors in children with developmental dyslexia in 48 publications were studied. *Results:* All language components are compromised to some degree in children with developmental dyslexia at specific ages. These results indicate the intricacy of language, but also emphasise that developmental dyslexia should be considered on a continuum and not as an absolute disorder. Only a few studies investigated language-based risk factors in participants five years and younger. Phonological-based risk factors were the most reported language component and comprise of risk factors within phonological awareness, rapid automized naming, and working memory. The second language component entailed morphology. Several inflectional and derivational morphological risk factors, as well as risk factors within syntactic processing, were acknowledged. Compromised receptive and expressive vocabulary knowledge, semantic processing, and fluency were identified as risk factors for the language component of semantics. The final component of language entailed pragmatics. Unfortunately, a limited amount of evidence is available in this regard. However, the evidence available confirmed poor linguistic pragmatics as a risk factor in children with developmental dyslexia. *Conclusion:* These language

components are interrelated and should be viewed holistically as risk factors for developmental dyslexia to promote earlier identification.

Keywords: developmental dyslexia, dyslexia, specific learning disorder, specific reading disorder, language, children, risk factors, indicators, systematic review

Language Editing

Letter from the language editor, Anna M de Wet, to confirm that language editing was performed.

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Kind regards

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List of Abbreviations and Acronyms

APA:	American Psychological Association
ASHA:	American Speech-Language-Hearing Association
CWDD:	Children with developmental dyslexia
DD:	Developmental dyslexia
DSM-5:	The Diagnostic and Statistical Manual of Mental Disorders, 5th Edition
DSR:	Distiller Systematic Review Software
NOS:	Newcastle-Ottawa Scale
PA:	Phonological awareness
PhA:	Phonemic awareness
PRISMA-P:	Preferred Reporting Items for Systematic Reviews and Meta-Analyses Protocols
PROSPERO:	International Prospective Register of Systematic Reviews
RAN:	Rapid automized naming
WM:	Working memory

Formatting

American Psychological Association (APA), 6th edition, referencing style was used to format the research manuscript. Double line spacing is used throughout the manuscript to ensure consistency and adhere to the in-house style of the Annals of Dyslexia where the article has been submitted.

Table of Contents

List of Figures

List of Tables

List of Appendices

Chapter 1: Introduction

"Reading comprehension is one of the most important transversal skills for the achievement of success in both school and society."

(Calet, Pérez-Morenilla, & De los Santos-Roig, 2019, p.1)

Chapter aim:

The aim of Chapter 1 is to provide background knowledge regarding the multifaceted origin and impact of developmental dyslexia on young children. This chapter allows for clarification concerning the definition and classification system of developmental dyslexia. It also describes the importance of systematically reviewing recent evidence relating to the language-based risk factors associated with developmental dyslexia in young children. Chapter 1 concludes with a research question for the current systematic review.

1.1. Developmental Dyslexia

Developmental Dyslexia (DD) originates as a neurodevelopmental impairment with a prevalence ranging from 5% to 17% (FragaGonzález, Karipidis, & Tijms, 2018; Gabel et al., 2016; Ozernov-Palchik et al., 2016a; Stoodley, 2015; Tannock, 2014). This wide-ranging prevalence is due to various definitions (Fletcher, 2009; Taylor, Démonet, & Chaix, 2004). The most accepted definition classifies dyslexia as a persistent reading and spelling disorder despite adequate education, intellectual abilities, sensory skills, and neurological abilities (American Psychiatric Association, 2013; Gabrieli, 2009; Tannock, 2014). Therefore, dyslexia is a specific reading disorder (Shaywitz & Shaywitz, 2005; Taylor et al., 2004). A deficit in decoding prevents fluent and accurate word recognition and/or spelling, impeding literacy development that is crucial to educational achievements (Bruder et al., 2011; Dickman, 2017; Peterson & Pennington, 2012, 2015; Tannock, 2014). Children with developmental dyslexia (CWDD) often experience adverse emotional and behavioural

reactions towards reading (WHO, 2015). Consequently, CWDD presents with less exposure to print knowledge, reduced comprehension, and inferior development of automaticity in contrast with their peers (Gabrieli, 2009; Ozernov-Palchik et al., 2016). As such, children with DD often remain poor readers (Ozernov-Palchik et al., 2016; Raschle, Chang, & Gaab, 2011). Literature denotes the persistent nature of DD as children with reading difficulties in the first grade have a 90% possibility of presenting with reading difficulties in the fourth grade, and a 75% possibility of reading deficits in high school (Catts, Bridges, Little, & Tomblin, 2008; Gabrieli, 2009). Developmental dyslexia is the most common learning disability and occurs across all languages (Mascheretti et al., 2017).

1.2. The Classification of Developmental Dyslexia

The Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) classifies dyslexia within the category of specific learning disorders under the broader category neurodevelopmental disorders (APA, 2013; Handler & Fierson, 2011; Moats & Dakin, 2017; Youman & Mather, 2012). This classification acknowledges the comorbidities among learning disorders that arise from shared risk factors (Jugend, 2014; Peterson & Pennington, 2015; Tannock, 2014). The broadened classification system together with the comorbidities between these disorders mask the specific nature of the underlying deficits of DD (Helland, Plante, & Hugdahl, 2011, Lyytinen et al., 2004; van Alphen et al., 2004) as well as the understanding of the specific risk factors (Margari et al., 2013; Snowling & Hulme, 2012).

Developmental dyslexia is caused by the complex interaction between environmental, genetic, cognitive, and neurological risk factors (Ozernov-Palchik, Yu, Wang, & Gaab, 2016). The neurocognitive influence of dyslexia is multifactorial and results in delayed phonological processing, oral language skills, and reduced processing speed (Helland et al., 2011; Kamza, 2017; Ozernov-Palchik et al., 2016; Peterson & Pennington, 2015). These

aberrant functions correlate with the deviant structure noted in the left hemisphere of children with dyslexia (Peterson & Pennington, 2015). The left hemisphere is mainly responsible for language and reading networks. Accordingly, the risk factors, together with the broadened classification system, indicate the presence of a language-related origin in DD (Catts, 1989; Gabrieli, 2009; Helland et al., 2011).

1.3. Developmental Dyslexia and Language

Language is an intricate field embracing five closely-related components, namely phonology, morphology, syntax, semantics, and pragmatics (Cardillo, Garcia, Mammarella, & Cornoldi, 2018). Phonology is a central contributing language-based risk factor of DD (Daigle, Costerg, Plisson, Ruberto, & Varin, 2016; Dickman, 2017; Vellutino, Fletcher, Snowling, & Scanlon, 2004) and the first language component investigated in the current review. The phonological theory hypothesises that children with dyslexia present with deficits in the representation, storage and/or retrieval of sound properties (Ramus, Marshall, Rosen, & van der Lely, 2003). Mengisidou and Marshall (2019) specified that children with DD performed poorly in all three dimensions of the phonological system. These dimensions include phonological awareness (PA), rapid automized naming (RAN), and working memory (WM) (Mengisidou & Marshall, 2019; Van der Stappen & Reybroeck, 2018).

The first dimension in the phonological system entails PA. Phonological awareness is defined as the “ability to identify and manipulate the sound properties of spoken words”, such as rhyming, alliteration, and syllables (Kilpatrick, 2016, p.13). Phonemic awareness (PhA) is a specific sub-skill classified under the umbrella category PA (Kilpatrick, 2016; Le Roux, Geertsema, Jordaan, & Prinsloo, 2017). Phonemic awareness necessitates the identification and manipulation of phonemes within spoken language, such as the deletion of phonemes

(Kilpatrick, 2016). Both PA and RAN are strong independent predictors of reading difficulties (Van der Stappen & Reybroeck, 2018).

Rapid automized naming correlates with reading ability in CWDD (Krasowicz-Kupis, Borkowska, & Pietras, 2009). Rapid automized naming of objects, colours, digits, and letters requires the retrieval of phonological information from long-term memory (Van der Stappen & Reybroeck, 2018). Thus, RAN is distinct from PA and WM.

Working memory, the final dimension of the phonological system, specifies the temporary, short-term storage of verbal information (Kamza, 2017; Perrachione, Ghosh, Ostrovskaya, Gabrieli, & Kovelman, 2017; Ozernov-Palchik et al., 2016). Thus, WM is specifically related to the phonological system. Deficits in WM are well-established in neurodevelopmental disorders, such as DD (Perrachione et al., 2017). Unfortunately, several controversies regarding the definition and labelling of WM are present in literature (Cowan, 2016, 2017). Consequently, numerous challenges arose during the classification of temporary information related to phonology in CWDD. Recent imaging studies specified that phonological deficits are associated with abnormalities in the cortical structure (associated with the language networks) and cerebral connectivity (Stein, 2018). Weaknesses do not only occur in the phonological aspect of language but are also present in other aspects of language *(*Adlof & Hogan, 2018). Research also concedes weaknesses in morphology, syntax, semantics, and pragmatics *(*Adlof & Hogan, 2018).

Poor phonological abilities affect the development of morphological awareness (Casalis, Colé, & Sopo, 2004). Morphology is the second language component reported in the current systematic review. Less is known about morphological awareness, although it correlates stronger with later literacy skills like reading and writing than phonological or syntactic awareness (Goodwin & Ahn, 2010). Morphological awareness refers to the conscious awareness and ability to manipulate the morphemic structure of words, the smallest

meaningful units of language (Casalis et al., 2004; Goodwin & Ahn, 2010). The awareness of morphemes within words occur at a level of inflection (changes for gender, number or tense), derivation (changes for the grammatical meaning or category) or compounding (Goodwin & Ahn, 2010; Kuo & Anderson, 2006). The literature established an association between morphological awareness and vocabulary and reading comprehension (Vender et al., 2018). A relationship between syntax and morphology also exists. Morphology focuses on the internal structure of words, while syntactic skills entail the combining of these inflected words into phrases and sentence (Teixeira, Schiefer, Carvalho, & Àvila, 2016).

The literature acknowledges a significant correlation between syntactical awareness and reading and writing (Teixeira et al., 2016). Reading experience contributes to syntactic development, signifying the complexity of syntactic deficits (Casalis, Leuwers, & Hilton, 2013). Currently, in literature, the syntactic deficits in DD are widely recognised. However, controversy exists regarding phonological processing giving rise to syntactic deficits. Several authors found that poor syntactic skills are mainly independent of phonological processing problems in children with DD (Antón-Méndez, Cuetos, & Suárez-Coalla, 2018; Gallagher, Frith, Snowling, 2000; Waltzman & Cairns, 2000). Conversely, other authors found that phonological processing difficulties give rise to syntactic deficits in DD (Casalis et al., 2013; Jiménez et al., 2004). The literature indicates a significant correlation between syntactical awareness, reading, and writing (Teixeira et al., 2016). Syntax is not only a contributor to word recognition but also in reading comprehension (Morvay, 2012). To read with comprehension requires the combining and understanding of lexical elements in sentences (Teixeira et al., 2016) as well as the adding of meaning to the decoded text.

Fourthly, children with DD present with difficulties in the retrieval and integration of semantic information (Jednoróg, Marchewka, Tacikowski, & Grabowska, 2010). Neuroimaging studies related to semantics are rare but specify the contribution of semantic

delays in DD (Paz-Alonso et al., 2018). Children with DD present with more difficulty in performing semantic tasks than orthographic and phonological tasks (Penolazzi, Spironelli, & Angrilli, 2008). A reciprocal relationship between semantics and phonology exists in DD. Semantic abilities predict RAN, PA, and word decoding skills (van Rijthoven, Keemans, Segers, & Verhoeven, 2018).

Language use is the final reviewed domain. Pragmatics is defined as the effective and social use of language with communication partners (Koegel, Park, & Koegel, 2013; Ying Sng, Carter, & Stephenson, 2018). Griffiths (2007) indicated a correlation between pragmatic deficits and dyslexia. Cardillo et al. (2018) further stated that children with DD present with noteworthy difficulties in linguistic pragmatics and theory of mind (ToM). An intangible link exists between pragmatics and ToM as children should consider the beliefs and intentions of a communication partner (Cardillo et al., 2018).

1.4. Rationale

Only a limited number of studies related to pragmatics in DD are available. Studies associated with the remaining language components (phonology, morphology, syntax, and semantics) in CWDD are more readily accessible. Still, even these publications do not clarify the extent to which these language-based components are impaired in children with DD (Adlof & Hogan, 2018; Gabrieli, 2009, Snowling & Hulme, 2012). In summary, the nature of the underlying difficulties of language development leading to DD have been masked (Helland et al., 2011, Lyytinen et al., 2004; Margari et al., 2013; Snowling & Hulme, 2012; van Alphen et al., 2004) by a loose definition (Taylor et al., 2004; Tunmer & Greaney, 2010), a broad classification system (Snowling & Hulme, 2012), and little understanding of the few distinguishing features of co-occurring specific learning disorders contributingto the inability

to identify the language-based risk factors of DD (Peterson & Pennington, 2015; Snowling & Hulme, 2012).

A systematic review was employed to obtain consensus on the language-based risk factors for DD. Systematic reviews have become progressively more important in health care (Moher et al., 2015). The use of an explicit, systematic, and reproducing methodology allowed for the identification, appraisal and synthesis of recent evidence (Ganeshkumar & Gopalakrishnan, 2013; Møller & Myles, 2016; Higgins & Green, 2011). The identification and categorisation of the language-based risk factors may lead to earlier identification of DD in young children and promote optimal developmental outcomes (Lange & Thompson, 2006; Zakopoulou et al., 2011). Furthermore, common themes and areas in need of further clarification were identified. The systematic review provided a solid foundation for the identification of unbiased language-based risk factors associated with DD (Higgins & Green, 2011; Pannucci & Wilkins, 2010).

1.5. Research question

The research question posed in the current systematic review was twofold: *What are the language-based risk factors of DD in children (age one to three and four to thirteen years)?* and *What is the strength and level of recent research evidence regarding the language-based risk factors of DD in children (age one to three and four to thirteen years)?*

Chapter 2: Method

"Research is creating new knowledge."

Neil Armstrong

Chapter aim:

The objective of Chapter 2 is to elaborate and provide comprehensive descriptions relating to the method aspects of the systematic review as the journal article only allows for concise descriptions. Chapter 2 expands on the aims of the research study and provides an explicit description regarding the design of the research, the search strategies employed, and the procedure of data analysis. Overall, this chapter acts as an indication of the validity of the research study and allow for possible reduplication in the future.

2.1. Research Aims

The current review necessitated a detailed methodological process of critically appraising recent peer-reviewed publications relating to language-based risk factors in CWDD. The aims of this research project were as follows:

Main aim. To systematically review evidence, over the past 17 years (2002-2018), to identify and describe the language-based risk factors associated with children (age one to three and four to thirteen years) with DD.

Sub-aims. The following subsidiary aims underpinned the primary aim:

- *Sub-aim 1*: To identify all the relevant published evidence, select studies for inclusion, and assess the quality and strength of evidence for these studies.
- *Sub-aim 2*: To synthesise and interpret the findings, and present a balanced and unbiased summary with consideration of flawed evidence levels of the language-based risk factors in CWDD.

2.2. Study Design

A systematic review allows for systematic and explicit methods to identify, select, and critically appraise and synthesise relevant research. The ultimate purpose of this systematic review was to integrate and collate evidence to allow for a meticulous summary of the language-based risk factors in CWDD (Berman & Parker, 2002; Moher et al., 2015). Systematic reviews are essential in health care as high-quality reviews provide trusted evidence and aid the development of evidence-based guidelines (Moher et al., 2015). Furthermore, through employing a systematic review, common themes and areas of clarification, as well as future research, can be identified (Moher et al., 2015). Currently, roughly 170 health care journals endorse the Preferred Reporting Items for Systematic Reviews and Meta-Analysis Protocols (PRISMA-P) guidelines (Moher et al., 2015). These numbers indicate the importance to adhere to these guidelines in a systematic review (Moher et al., 2015; Shamseer et al., 2015).

The current systematic review endorsed the PRISMA-P as a guiding principle to ensure transparent, structured, and complete reporting (Moher et al., 2015). Subsequently, the guidelines allowed for valid and reliable reporting of relevant evidence. The PRISMA-P includes a 26-item checklist (Shamseer et al., 2015). This checklist is organised into three main categories namely, administrative, introduction, and methods (Shamseer et al., 2015). These categories provided structure and quality to the current systematic review by including all the relevant features of the checklist (Appendix A). In accordance with Item 2 on the PRISMA-P checklist, the systematic review was registered with the International Prospective Register of Systematic Reviews (PROSPERO) of the University of York (registration number CRD 42018102613) on 24 July 2018, to avoid any duplication. Registration of the current review aided and facilitated transparency and minimised the risk of bias in the review

process. Ethical approval for the systematic review was obtained from the Research Ethics Committee of the Faculty of Humanities, University of Pretoria, South Africa (Appendix B).

2.3. Data Collection Procedures

2.3.1. Eligibility criteria

In the current systematic review, the electronic searches included publications from 2002 up to 2018. Before 2002, dyslexia was inadequately defined, thus influencing the identification thereof. From 2002 onwards, a meaningful and inclusive definition was documented (Moats & Dakin, 2017). The researcher performed a systematic review, spanning 17 years, ensuring that a complete and detailed search was conducted. The Population, Intervention, Comparison, and Outcome (PICO) synthesis tool was utilised to structure the eligibility criteria (Shamseer et al., 2015) (Item 8 of the PRISMA-P checklist, Appendix A). Studies were identified that pertained to the criteria delineated in Tables 1 and 2.

Study designs. Numerous research designs and methods were eligible for inclusion in the current systematic review since the variability provided an opportunity to evaluate and identify variances within data that conceivably had an impact on early identification of DD (Meline, 2006; Peinemann, Tushabe, & Kleijnen, 2013). The variety of study designs included cohort, observational, case-control, and randomised control trials. However, expert committee reports, clinical experience or consensus, and conference studies were excluded. These publications are classified as the lowest level of evidence, according to the American Speech-Language-Hearing Association (ASHA) evidence rating scale (ASHA, 2004). Combining existing reviews into a new systematic review is also controversial and could introduce a reporting bias (Robinson et al., 2014). Only peer-reviewed publications were included in the systematic review. Lastly, studies with duplicate datasets were excluded as manifold reporting of datasets can lead to multiple publication bias (Mayo-Wilson, Li, Fusco,

& Dickersin, 2017). This criterion ensured that only studies with scientific integrity, such as validity, were included to augment the quality of the systematic review (Larson & Chung, 2012; Moher, 2015).

Participants. Studies were retained if participants were diagnosed with DD. Two age groups were selected for the current review (age one to three and four to thirteen years). The first age group consisted of participants aged one to three years. This age group encompassed the period of optimal neuroplasticity (zero to three years) (Rossetti, 2001). This is an optimal period for intervention, and hence early language-based risk factors in CWDD need to be identified within this age range. The second age cohort, four to thirteen years, embraced studies within the sensitive period of children beginning to read and being exposed to formal schooling (Black et al., 2012). Furthermore, studies related to general reading disorders were excluded from the electronic search method, as dyslexia is not as a result of inadequate intellectual abilities, sensory difficulties, neurological disorders or insufficient education (WHO, 2015). These related reading disorders were, therefore, not compatible with the present study.

Language. Only studies investigating alphabetic languages were included in the current review due to the variances in non-alphabetic orthographies. English is acknowledged as the universal language of science (Morrison et al., 2012). Thus, full-text publications not available in English were excluded. Language bias may have occurred due to an exclusive reliance on English studies (Wang et al., 2015). Table 1 and 2 outline and discuss the inclusion and exclusion criteria applied during the selection of studies.

Table 1: Inclusion Criteria of the current review.

Inclusion Criteria	**Rationale**
Diagnosis of dyslexia	Only studies with diagnosed DD participants were included.
Participants aged one to three and four to thirteen years	The age group one to three years, encompasses the period of optimal neuroplasticity (Rossetti, 2001). The age group four to thirteen years encompasses the sensitive period of children starting to read and being exposed to formal schooling (Black et al., 2012
Published from 2002 and 2018	To ensure a wide-ranging search was conducted in order to obtain relevant literature concerning the research question.
Peer reviewed	To ensure that studies with scientific integrity are selected and to augment the quality of the systematic review (Larson & Chung, 2012; Moher, 2015).

Table 2: Exclusion criteria of the current review

Exclusion criteria	**Rationale**
General reading disorders	DD is not the result of inadequate intellectual abilities, neurological disorders or insufficient education (WHO, 2015).
Review articles	Combining existing reviews into a new systematic review may result in reporting bias (Robinson, 2014
Publication before 2002	Literature before 2002 had not been included as a formal definition for dyslexia was documented in 2002
Expert committee reports, clinical experience and consensus conference articles	Expert committee reports, clinical experience and consensus conference articles are classified on the lowest level of evidence (ASHA, 2004).
Notes, letter to editors, and full texts not available in English	English is acknowledged as the universal language of science (Morrison et al., 2012). Thus, notes, letter to editors, and publications not available in English were excluded.
Studies with duplicate datasets	The reporting of duplicate datasets may result in multiple publication bias (Mayo-Wilson et al, 2017).
Non-alphabetical languages	The orthographic structure of these languages varies completely from alphabetic languages and may influence the associated language aspects

2.3.2. Search Strategy

Comprehensive electronic searches were executed in March 2019, with the last search on 29 March 2019. Four relevant databases, namely PubMed, Scopus, PsychINFO, and CINAHL were carefully examined (Falagas, Pitsouni, Malietzis, & Pappas, 2008; Sampson, McGowan, Cogo, & Horsley, 2006). Four databases were used to balance the sensitivity and specificity of the search and to limit bias and create a suitable search strategy (Leeflang, 2014; Sampson, McGowan, Cogo, & Horsley, 2006). The search strategy was in accordance with Items 9 and 10 of the PRISMA-P checklists (Appendix A) (Moher et al., 2015).

Concept mapping was used to enhance the search strategy. This type of mapping refers to a visual method that acts as a qualitative representation of a complex concept (Wilson, Mandich, & Magalhães, 2016). A combination of key search terms, indexed in the MeSH Terms and Thesaurus, was consistently employed across all four databases. The following key search terms were included: "(language-based risk factors OR phonological risk factors OR early language risk factors OR language risk factors OR signs OR markers OR indicators) AND (young children OR toddlers OR children OR at-risk toddlers OR at-risk young children OR at-risk children) AND (developmental dyslexia OR specific reading disability OR specific reading disorder OR specific spelling disorder OR specific spelling disability OR phonological dyslexia OR dyslexia)". A total of 10 095 hits was obtained through visualising the search terms and identifying the associations between the search concepts (Wilson et al., 2016). Figure 1 specifies the number of hits for each specific database.

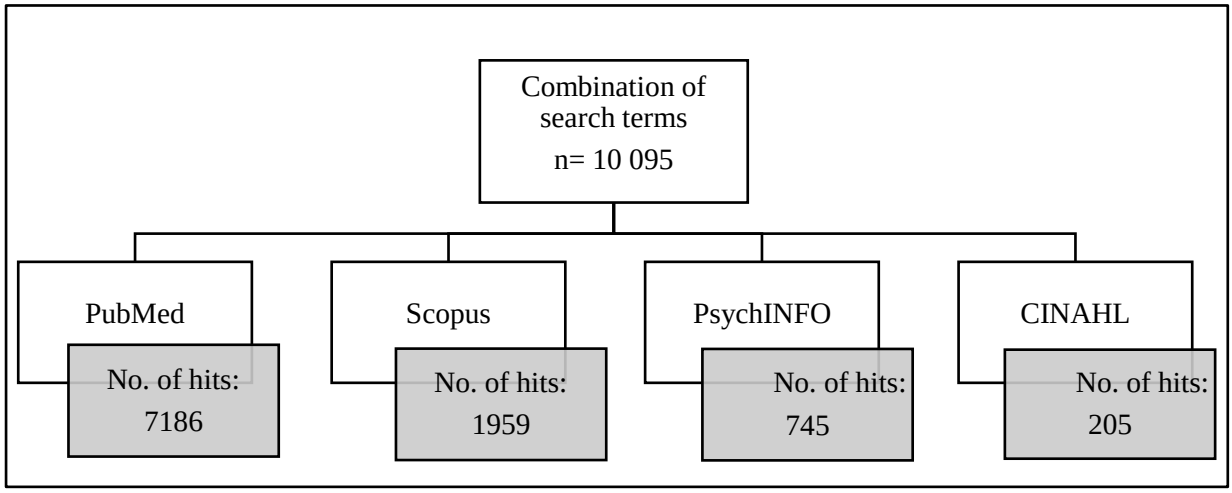

Fig 1: The number of publications obtained from each database.

2.3.3. Study Selection

Throughout the electronic search method, the studies were appraised at two levels of the PRISMA-P statement to determine the final study selection. The four databases were hand scanned by reviewing each study and extracting meaningful data from each study directed by the PRISMA-P guidelines (Moher et al., 2015).

The initial level entailed the screening of titles and abstracts (Moher et al., 2015). First duplicate articles were discarded (n=7439). The titles and abstracts (n=2664) of English studies were reviewed to determine the significance for DD and language-based risk factors, within a 17-year timeframe (2002-2018). Unrelated studies, based on their title and abstract, were excluded at this level (n=2569). Relevant titles and abstracts (n=95) qualified for a full-text review, introducing the second level of the PRISMA-P statement.

On the second level, full-text studies (n=95) were reviewed to determine the appropriateness for inclusion (Moher et al., 2015). The reference lists of studies were hand scanned to identify related articles (n=8). This identification process acted as a secondary literature search. The articles were compared against the comprehensive eligibility criteria (Table 1 and 2). Forty-seven articles were excluded for a number of reasons: The language components were not investigated (n = 18); the participants in the samples were not diagnosed

with DD (n=12); the participants were older than 13 years (n = 9); the study design was a systematic review (n = 1); the same datasets were utilised in other articles (n = 4), and several duplicate articles were discovered (n = 3). A total of 48 articles were accepted for qualitative and quantitative synthesis (Figure 1 in the results section in Chapter 3). To avoid bias and ensure reproducibility, two of the three investigators independently reviewed the full-text studies randomly to determine their compatibility with the eligibility criteria. Excellent agreement between the investigators was attained (kappa=0.875). The third independent investigator resolved three contradictory selections through means of a discussion and determining the final inclusion of studies (n=48). These steps are in accordance with item 11b and 11c of the PRISM-P checklist (Appendix A) (Shamseer et al., 2011).

2.3.4. Data Management

Distiller Systematic Review (DSR), an online software programme (Evidence Partners, 2018), was utilised to combine the results of the searches from the four specified databases (in agreement with the PRISMA checklist item 11a). The Distiller Systematic Review software programme is the most utilised systematic review software as it improves the transparency of a systematic review (Evidence Partners, 2018). This software allowed for collaboration among the investigators during the data management process. The tool permitted screening of the titles and abstracts, elimination of duplicate citations, and full-text study reviews (Evidence Partners, 2018). Furthermore, the DSR programme also allowed for the assessment of the risk of bias and extraction of study characteristics and outcomes.

2.3.5. Data Extraction

Each of the selected forty-eight studies was analysed and relevant data items were extracted. The data was extracted according to the eligibility criteria. Thus, the primary objective of the review was to retain any articles reporting or identifying the language-based risk factors in

CWDD. The data attained were exported into MS Excel document and a summarised table was compiled.

The following data items were included in accordance with Item 12 of the PRISMA-P checklist (Appendix A): the authors, year of publication, method, design (qualitative, quantitative or mixed methods), the number of participants (sample size of children diagnosed with DD and the control groups), age-group (mean age of the group with DD), method, level of evidence, and the major findings related to the language-based risk factors of participants identified in the studies.

The included studies' levels and strength of evidence were critically appraised through employing the American Speech-Language-Hearing Association (ASHA) level of evidence rating scale (ASHA, 2004). Table 3 depicts these outcomes. The ASHA evidence scale is widely accepted in the subject field and ensures that the highest-quality evidence is selected for final inclusion (ASHA, 2004). The scale comprises of four levels - the highest being level I, and the lowest level IV. Two of the investigators (DR and SG) independently rated the selected studies according to the rating scale. The investigators appraised the extracted data to ensure the accuracy, consistency, and clarity of the data. No inconsistent ratings were obtained. Table 3 outlines the levels of evidence according to the ASHA evidence rating scale.

Table 3: Levels of Evidence (ASHA, 2004).

Level	Description
I	A well-designed meta-analysis of >1 randomised controlled trial.
Ib	A well-designed randomised controlled study.
IIa	A well-designed controlled study without randomisation. Selected when there were control and experimental groups but procedures were not randomised
IIb	A well-designed quasi-experimental study. Selected when within-participant control measures were discussed.
III	A well-designed non-experimental study, i.e., correlational and case studies. Selected when the within-participant design was not discussed
IV	Expert committee report, consensus conference, clinical experience of respected authorities.

2.3.6. Risk of Bias

The assessment of the risk of bias is critical, as it is equivalent to internal validity (Bero et al., 2018). To ensure a valid systematic review, publication bias should be avoided (Mlinarić, Horvat, & Šupak Smolčić, 2017; Moher et al., 2015). The avoidance of publication bias is included in the PRISMA checklist (item 14, Appendix A). Any absence of relevant evidence may result in publication bias (Müller et al., 2013). The literature specifies that studies with positive outcomes are preferentially published or accompanied by full-text publication (Knobloch, Yoon, & Vogt, 2011). Incomplete reporting regarding published studies not published yet may result in partial data (Moher et al., 2015). The primary objective of a systematic review involves the assimilation of all relevant information (Müller et al., 2013; Mlinarić et al., 2017).

Four databases using multiple search terms were utilised to obtain an optimal number of relevant articles to minimise the risk of publication bias (White & Schimdt, 2005). Additionally, the risk of bias within the articles was considered. Publication bias in a systematic review occurs mostly during the selection process. A transparent process is thus necessary to avoid such bias (Knobloch et al., 2011). For this transparency, the PRISMA-P statement was adhered to (Knobloch et al., 2011; Moher et al., 2015). Each article selected for the review was individually analysed to evaluate the risk of further biases.

The Newcastle-Ottawa Scale (NOS), a star-system tool, was used to assess the quality of the non-randomised studies included in the current review (Wells et al., 2014). This scale is recommended by the Cochrane Collaboration (Higgins & Green, 2011). The NOS entails three broad categories: study group selection, group comparability, and the exposure and outcome of the cohort or case-control studies (Stang, 2010; Wells et al., 2014). Two independent investigators (DR and SG) appraised the quality of the cohort (n=46), and case-control (n=2) studies, according to the NOS. Each study was assessed according to the "star-system". The number of stars indicates the level of evidence achieved. A star was awarded to high-quality characteristics within the defined categories. Thus, the higher the number of stars allocated, the greater the level of evidence (Wells et al., 2014). The content validity and inter-rater reliability of the NOS have been established (Wells et al., 2014). Tables 4 and 5 outline the categories and the high-quality characteristics for which stars were awarded.

Table 4: Criteria for the assessment of the risk of bias for the cross-sectional studies; adapted from the Newcastle-Ottawa Scale.

Selection

1) *Representativeness of the exposed cohort: (maximum 1 star)*

   Truly representative of the average child with DD in the community. *

   Somewhat representative of the average child with DD in the community *

   No description of the derivation of the cohort

2) *Selection of the non-exposed cohort: (maximum 1 star)*

   Drawn from the same community as the children with DD.*

   Drawn from a different source.

   No description of the derivation of the non-exposed cohort.

3) *Ascertainment of exposure: (maximum 1 star)*

   Secure record (e.g. clinical records). *

   Structured interview.

   No description.

4) *Demonstration that outcome of interest was not present at start of study: (maximum 1 star)*

   Yes. *

   No.

Comparability

1) *Comparability of cohorts on the basis of the design or analysis: (maximum two stars)*

   Study controls for diagnosis of DD. *

   Study controls for cognitive impairments, educational deprivations and sensory impairments. *

Outcome

1) *Assessment of outcome: (maximum 1 star)*

   Independent assessment by trained allied and health-care professionals. *

   Record linkage.

   No description.

2) *Adequacy of follow up of cohorts: (maximum 1 star)*

   Complete follow-up - all subjects accounted for. *

   Subjects lost to follow up unlikely to introduce bias - small number lost - <20 % and a description of those lost. *

   Follow up rate < 80% and no description of those lost

No description of those lost.

Note. NOS= Newcastle -Ottawa Scale; DD= developmental dyslexia. Maximum of 8 stars* were awarded to the cross-sectional studies

Table 5: Criteria for the assessment of the risk of bias for the case-control studies; adapted from the Newcastle-Ottawa-Scale.

Selection

1. *Is the case definition adequate? (maximum of 1 star)*
   - Yes, independent validation. *
   - Yes, e.g. record linkage
   - No description
2. *Representativeness of the cases: (maximum of 1 star)*
   - Consecutive representative series of cases. *
   - Potential for selection biases or not indicated.
3. *Selection of controls: (maximum of 1-star *)*
   - Community controls*
   - Hospital controls
   - No description.
4. *Definition of controls: (maximum of 1 star)*
   - No history of DD. *
   - No description of source.

Comparability

1. *Comparability of the cases and controls on the basis of the design or analysis: (maximum of 2 stars)*
   - Study controls for children with diagnosed DD. *
   - Study controls for the exclusion of cognitive impairments, educational deprivations and sensory impairments. *

Exposure

1. *Ascertainment of exposure: (maximum of 1 star)*
   - Secure records (clinical records). *
   - Medical reports only.
   - No description.
2. *Same method of ascertainment for cases and controls: (maximum of 1 star)*
   - Yes. *
   - No.

Note. NOS= Newcastle-Ottawa Scale; DD= developmental dyslexia. Maximum of 8 stars* were awarded to the case-control studies.

2.4. Reliability, Validity, and Trustworthiness

Consideration was given to the validity, reliability, and trustworthiness of the systematic review. Leung (2015) states that validity in qualitative research can be defined as the 'appropriateness of the tools, processes, and data (Leung, 2015, p.325). In quantitative research, trustworthiness refers to the level of confidence of the data, interpretation, and methods employed to ensure the quality of a study (Polit & Beck, 2014). For the current

systematic review, trustworthiness necessitated credibility, dependability, confirmability, and transferability. Reliability is a concept often used in most types of research (Golafshani, 2003). The notion refers to the "precise replicability of the processes and the results" (Leung, 2015, p. 325). The integrity of the data obtained was ensured by:

- Following and adhering to the PRISMA Statement Checklist (Moher et al., 2015). These steps ensured that a complete systematic review was obtained as it outlines all the steps required to fulfil a systematic review study.
- Including a multifaceted approach by employing four electronic databases in collaboration with several search terms to categorise literature (excluding review studies and notes) from peer-reviewed research studies.
- Analysing and synthesising relevant publications according to the inclusion criteria as well as to obtain consensus between the three reviewers, when necessary, to ensure that reliable and valid data was obtained.
- Determining the level and strength of recent evidence through independent and random ratings by two investigators (DR and SG). Consensus was reached between the two investigators. The potential bias was discussed between the three investigators (DR, SG, and MLR) and the selection bias was prevented through reporting on all results, and not only the desired data (Moher et al., 2015)

2.5. Data Analysis

Once the information was obtained and critically appraised, descriptive statistics were employed to analyse the results of the data and classify the language-based risk factors associated with DD. Descriptive statistics, which consisted of the means, standard deviations, frequencies and percentages, were calculated using Microsoft Excel. These statistics were employed in the systematic review to generate a firm conclusion of the risk factors commonly

found to be significant, as well as the risk factors identified in only a few studies (Leedy & Ormrod, 2015).

Thematic analysis was also utilised to extract, organise, and synthesise data from quantitative and mixed (qualitative and quantitative) methods. Thematic analysis was done in adherence to the five intricate language components - phonology, morphology, syntax, semantics, and pragmatics. This qualitative analysis was used to interpret both implicit and explicit data related to the language-based risk factors in CWDD (Guest, MacQueen, & Namey, 2012). As such, the information obtained in the systematic review was analysed qualitatively as well as quantitatively (Gough, 2015).

Chapter 3: Research Article

Chapter aim:

Chapter 3 contains the research article based on the current research manuscript. The article has been submitted to the "Annals of Dyslexia", an internationally accredit journal with an impact factor of 2.171, and is currently in review (Appendix C). The journal specified the APA (6th edition) as a formatting guide, which is in accordance to the formatting and structuring of the dissertation.

Language-based risk factors in children with developmental dyslexia: A systematic review

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Introduction

The Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) classifies Developmental Dyslexia (DD) within the category-specific learning disorders under the broader category neurodevelopmental disorders (APA, 2013; Moats & Dakin, 2017). This classification acknowledges the comorbidities among learning disorders that arise with shared risk factors (Peterson & Pennington, 2015; Tannock, 2014). The broadened classification system and shared risk factors indicate the presence of a language-related origin in DD (Adlof & Hogan, 2018; Gabrieli, 2009; Helland, Plante, & Hugdahl, 2011).

Language is an intricate field embracing five related components, namely phonology, morphology, syntax, semantics, and pragmatics (Adlof & Hogan, 2018). Phonology is a central contributing language-based risk factor (Adlof & Hogan, 2018) and our first field of investigation in relation to DD. Mengisidou and Marshall (2019) specified that children with DD (CWDD) performed poorly in phonological awareness (PA), working memory (WM), and rapid automized naming (RAN) (Mengisidou & Marshall, 2019). Both PA and RAN are strong independent predictors of reading difficulties (Van der Stappen & Reybroeck, 2018). Rapid automized naming tasks require the retrieval of phonological information from long-term memory (Van der Stappen & Reybroeck, 2018). Therefore, RAN is independent from PA and WM. Working memory specifies the short-term storage of verbal information (Perrachione, Ghosh, Ostrovskaya, Gabrieli, & Kovelman, 2017). Deficits in WM are well-established in neurodevelopmental disorders such as DD (Perrachione et al., 2017). Due to several controversies regarding the definition and labelling of WM, challenges arose during the classification of temporary phonological information. However, we know that poor phonological abilities affect the development of morphological awareness (Casalis, Colé, & Sopo, 2004), our second included component.

Although morphological awareness correlates stronger with later literacy skills, less is known about it than phonological or syntactic awareness. The awareness of morphemes within words occurs at a level of inflection, derivational or compounding (Goodwin & Ahn, 2010). An association of morphology with syntax also exists. Morphology focuses on the internal structure of words, while syntactic skills, our third component, entail the combining of these inflected words into phrases and sentences.

Literature acknowledges a significant correlation between syntactical awareness and reading and writing (Teixeira, Schiefer, Carvalho, & Àvila, 2016). Reading experience contributes to syntactic development, signifying the complexity of syntactic deficits (Casalis, Leuwers, & Hilton, 2013). Controversy exists regarding phonological processing giving rise to syntactic deficits (Antón-Méndez, Cuetos, & Suárez-Coalla, 2018; Casalis, et al., 2013). Syntax contributes to word recognition and reading comprehension (Morvay, 2012). Reading with comprehension requires the combining and understanding of words in sentences (Teixeira et al., 2016) as well as adding meaning to the decoded text.

The fourth language component investigated involved semantics as CWDD present with difficulties in the retrieval and integration of semantic information (Jednoróg, Marchewka, Tacikowski, & Grabowska, 2010). Neuroimaging studies specify the contribution of semantic delays in DD (Paz-Alonso et al., 2018). Regarding DD, a reciprocal relationship between semantics and phonology exists. Semantic abilities predict RAN, PA, and word decoding skills (van Rijthoven, Keemans, Segers, & Verhoeven, 2018).

Finally, we reviewed the use of these language components. An intangible link exists between pragmatics and theory of mind (ToM) as children should evaluate the intentions of a communication partner (Cardillo, Garcia, Mammarella, & Cornoldi; 2018). These authors specified that CWDD present with difficulties in linguistic pragmatics and ToM. Few studies

related to pragmatics in DD are available. Studies associating the other components and DD are more readily accessible. Still, these do not clarify the extent to which language-based components are impaired in CWDD (Adlof & Hogan, 2018; Gabrieli, 2009, Snowling & Hulme, 2012).

In summary, the nature of underlying difficulties of language development leading to DD has been masked by a loose definition (Tunmer & Greaney, 2010), a broad classification system (Snowling & Hulme, 2012), and little understanding of the few distinguishing features of co-occurring specific learning disorders contributing to the inability to conclusively identify the language-based risk factors of DD (Peterson & Pennington, 2015; Snowling & Hulme, 2012).

To obtain a consensus on the possible language-based risk factors for DD, we employed a systematic review to identify, appraise, and synthesise recent evidence (Møller & Myles, 2016). The research question posed in this systematic review is twofold: *What are the language-based risk factors of DD in children (age 1 to 3 and 4 to 13 years), and what is the strength and level of recent research evidence regarding the language-based risk factors of DD in this population?*

Method

Study Design

The Preferred Reporting Items for Systematic Reviews and Meta-Analysis Protocol (PRISMA-P) guidelines directed this review (Moher et al., 2015) and it was registered with the International Prospective Register of Systematic Reviews (PROSPERO) of the University of York (registration number CRD 42018102613) on 24 July 2018.

Eligibility Criteria

We identified studies according to these inclusion criteria: a) Peer reviewed, empirical articles in English as the universal language of science (Morrison et al., 2012); b) studies published between 2002 and 2018. Before 2002, DD was inadequately defined (Moats & Dakin, 2017); c) studies with participants diagnosed with DD; d) studies investigating alphabetic languages; e) studies with an age sample ranging from 1 to 13 years. Children aged 1 to 3 years encompass the period of optimal neuroplasticity (Rossetti, 2001). The age sample ranging from 4 to 13 years allowed for the inclusion of the first stages of reading and exposure to formal schooling (Black et al., 2012). f) Studies with various research designs and methods were included. However, we excluded expert committee reports, clinical experience, and consensus conference articles as the American Speech-Language-Hearing Association (ASHA) (2004) classifies these as the lowest level of evidence (ASHA, 2004). Combining existing reviews into a new systematic review could introduce a reporting bias, thus review articles were excluded (Robinson et al., 2014). g) Studies with duplicate datasets were not included as multiple reporting of duplicate datasets can lead to multiple publication biased results (Mayo-Wilson, Li, Fusco, & Dickersin, 2017). Finally, we excluded studies on reading disorders related to inadequate intellectual abilities, sensory difficulties, neurological disorders or insufficient education as DD is not a result of these factors (WHO, 2015).

Search Strategy

We executed comprehensive electronic searches across four databases: PubMed, Scopus, PsychINFO, and CINAHL. A combination of key search terms, indexed in the MeSH Terms and Thesaurus, was employed across all four databases. The search terms included: "(language-based risk factors OR phonological risk factors OR early language risk factors OR language risk factors OR signs OR markers OR indicators) AND (young children OR toddlers OR children OR at-risk toddlers OR at-risk young children OR at-risk children) AND

(developmental dyslexia OR specific reading disability OR specific reading disorder OR specific spelling disorder OR specific spelling disability OR phonological dyslexia OR dyslexia)". A total of 10 095 hits was obtained during the final search on 29 March 2019. An online software programme Distiller Systematic Review (DSR) (Evidence Partners, 2008) permitted synthesis of searches, identification of duplications, screening of titles and abstracts, as well as full-text article reviews. The eligibility of the studies was ascertained by screening the titles and abstracts, to permit the exclusion of unrelated studies and to discard duplicate studies (n=7439). The remaining articles (n=95) were reviewed in full. The reference lists of the included articles were hand scanned to identify related articles (n=8). This identification process acted as a secondary literature search. To avoid bias and ensure reproducibility, two of the three independent investigators reviewed the studies, determining their compatibility with the eligibility criteria (Higgins & Green, 2011). We attained excellent investigator agreement (kappa=0.875). Three contradictory selections were resolved after discussion with a third independent investigator to determine the final inclusion of articles (n=48) (Figure 1 in the results section).

Data Extraction

Each article was analysed for: authors, year of publication, design (qualitative, quantitative or mixed methods), method, number of participants (sample size of CWDD and control groups), age group (mean-age of the group with DD), level of evidence, and major findings specific to the language-based risk factors of participants identified in the articles. We applied the ASHA level of evidence rating scale (ASHA, 2004) to determine the strength and level of evidence.

Risk of Bias Assessment

The assessment of risk of bias is critical, as it is equivalent to internal validity (Bero et al., 2018). Two independent investigators appraised the quality of the cohort (n=46) and

case-control (n=2) studies according to the Newcastle-Ottawa Scale (NOS). This scale appraised the risk of bias according to selection, comparability, and exposure and outcome of the cohort or case-control studies (Wells et al., 2000). Each study was assessed according to the 'star-system'. A star was awarded to high quality characteristics within the defined categories.

Data Analysis

Descriptive statistics, consisting of means, standard deviations, frequencies and percentages, were calculated using Microsoft Excel. Thematic analysis was used to organise and synthesise data from the articles to explain the findings qualitatively (Leedy & Ormrod, 2015).

Results

Study Selection

Overall, 95 full-text articles were scrutinised conferring with the predetermined eligibility criteria. Forty-seven articles were excluded. Forty-eight articles were accepted for qualitative and quantitative synthesis. Figure 1 depicts the selection process.

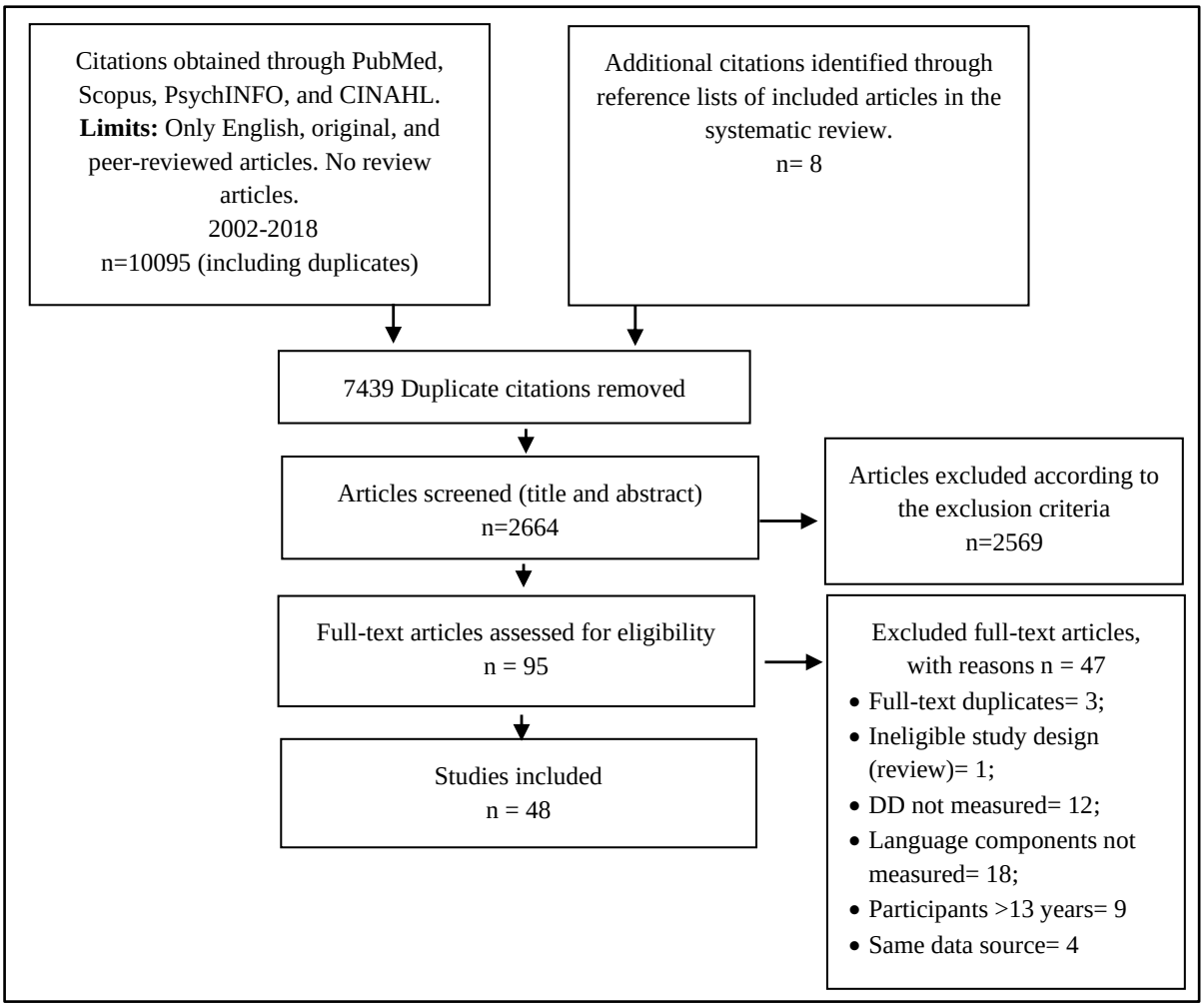

Fig 1. The Process of Identifying Articles for Review, adapted from the PRISMA-P statement (Moher et al., 2015).

Risk of bias results

Forty-six cross-sectional and two case-control studies were assessed and graded by two independent investigators. Perfect agreement was obtained (kappa=1) (Table 1). For the analysis, studies obtaining six stars indicated "low risk of bias" and fewer than six stars were considered as "high risk of bias". Forty-four (91.67%) articles received more than six stars and were classified as "low risk of bias". Results from the ASHA levels of evidence rating scale also indicated an acceptable level of quality. Two studies obtained a level III-rating due to case-control design (ASHA, 2004), while forty-six studies obtained an IIb-rating due to a cross-sectional and longitudinal design (ASHA, 2004).

Table 1: Study scores of the adapted Newcastle-Ottawa Scale.

Reference	Level of Evidence (ASHA, 2004)	Adapted NOS		
Cross-sectional studies		Selection (max. of 4*)	Comparability (max. of 2*)	Outcomes (max. of 2*)
Antón-Méndez et al. (2018)	IIb	***	**	**
Araújo et al. (2011)	IIb	**	**	**
Backes et al. (2002)	IIb	***	**	**
Berthiaume and Daigle (2014)	IIb	***	**	**
Bexkens, van den Wildenberg, and Tijms (2014)	IIb	***	**	**
Bloom, Garcia-Barrera, Miller, Miller, and Hynd (2013)	IIb	****	**	**
Boets et al. (2010)	IIb	***	**	**
Burani, Marcolini, De Luca, and Zoccolotti (2008)	IIb	*	**	**
Cantiani, Lorusso, Perego, Molteni, and Guasti (2015)	IIb	***	**	**
Cardillo et al. (2018)	IIb	**	**	**

Reference	Level of Evidence (ASHA, 2004)	Adapted NOS		
Casalis, Colé, and Sopo (2004)	IIb	***	**	**
Casalis, Leuwers, and Hilton (2013)	IIb	**	**	**
Dandache, Wouters, and Ghesquière (2014)	IIb	***	**	**
De Groot, Van den Bos, Van der Meulen, and Minnaert (2017)	IIb	***	**	**
Di Filippo et al. (2006)	IIb	***	**	**
Eisenmajer, Ross, and Pratt (2005)	IIb	***	**	**
Farquharson, Centanni, Franzluebbers, and Hogan (2014)	IIb	***	**	**
Germano and Capellini (2011)	IIb	****	**	**
Helland, Posserud, Helland, Heimann, and Lundervold (2016)	IIb	****	**	**
Jednoróg et al. (2010)	IIb	***	**	**
Jednoróg, Gawron, Marchewka, Heim, and Grabowska (2014)	IIb	***	**	**
Jiménez et al. (2004)	IIb	***	**	**
Krasowicz-Kupis, Pietras, and Burkowski (2009)	IIb	***	**	**

Reference	Level of Evidence (ASHA, 2004)	Adapted NOS		
Landerl et al. (2013)	IIb	***	**	**
Moura, Moreno, Pereira, and Simões (2014)	IIb	***	**	**
Moura, Pereira, Alfaiate, Fernandes, Fernandes, and Nogueira (2017)	IIb	****	**	**
Nithart et al. (2009)	IIb	***	**	**
Penolazzi, Spironelli, and Angrilli (2008)	IIb	***	**	**
Plaza, Cohen, and Chevrie-Muller (2002)	IIb	*	**	**
Puolakanaho et al. (2007)	IIb	**	**	**
Quaglino et al. (2008)	IIb	*	**	**
Ramus, Marshall, Rosen, and Van der Lely (2013)	IIb	****	**	**
Rispens and Been (2007)	IIb	***	**	**
Rispens, Roeleven, and Koster (2004)	IIb	***	**	**
Sabisch, Hahne, Glass, von Suchodoletz, and Friederici (2006)	IIb	***	**	**

Reference	Level of Evidence (ASHA, 2004)	Adapted NOS		
Schulz et al. (2008)	IIb	*	**	**
Schulz et al. (2009)	IIb	**	**	**
St-Pierre and Béland (2010)	IIb	***	**	**
Talli, Sprenger-Charolles, and Stavrakaki (2016)	IIb	***	**	**
Thompson et al. (2015)	IIb	****	**	**
Tunmer and Chapman (2006)	IIb	***	**	**
Van Bergen et al. (2014)	IIb	****	**	**
Van Rijthoven, Kleemans, Segers, and Verhoeven (2018)	IIb	**	**	**
Vender, Mantione, Savazzi, Delfitto, and Melloni (2017)	IIb	***	**	**
Wachinger et al. (2018)	IIb	***	**	**
Wise, Sevcik, Morris, Lovett, and Wolf (2007)	IIb	**	**	**

Reference	Level of Evidence (ASHA, 2004)	Adapted NOS		
Case-control studies		Selection (max. of 4*)	Comparability (max. of 2*)	Exposure (max. of 2*)
Nergård-Nilssen (2006)	III	****	**	**
Sprenger-Charolis, Colé, Kipffer-Piquard, Pinton, and Billard (2009)	III	****	**	**

Study Characteristics

One study (2.1%) considered English, French, Dutch, German, and Finish. Of the remaining studies, French and English were considered by eight (16.7%) studies respectively for each language. Nine (18.8%) studies considered Dutch. Six (12.5%) studies considered Italian and four (8.3%) German and Portuguese. Three (6.3%) studies considered Polish, two (4.2%) Finnish, whilst one study (2.1%) was done in Greek, Norwegian, and Spanish respectively.

The settings from which the subjects were recruited varied from schools to specialised clinics. Twenty-six (54.2%) studies were conducted at schools. Two (4.2%) studies were done in special education centres, one (2.1%) in a school and special education centre, two (4.2%) at dyslexia centres and six (12.5%) did not specify the setting. Of the remaining studies, three (6.3%) were conducted at hospitals, two (4.2%) at neuropsychiatric medical facilities, and five (10.4 %) at clinics.

A total of 12 576 subjects were included in our qualitative synthesis of which 2 985 subjects were diagnosed with DD. The mean sample size involved 141 participants. The mean age of the DD subjects was 9 years. Thirty-seven studies (79.2%) employed cross-sectional designs, six (12.5%) longitudinal designs, one (2.1%) a cross-linguistic cross-sectional design, one (2.1%) a retrospective longitudinal case study, one (2.1%) a multiple case study, and one (2.1%) a longitudinal cohort study.

The 48 articles were synthesised according to the language-based risk factors investigated in CWDD (Table 2 and 3). Among these, 37 (77.1%) focused on phonological risk factors, 17 (35.4%) reported on semantic risk factors, while nine (18.8%) investigated syntactic and morphological risk factors, respectively. Only two studies (4.2%) focused on pragmatics risk factors in CWDD. A detailed discussion of each language component follows after Table 2 and 3.

Table 2: Identified language components.

Language components	Number of articles	Percentage articles	Mean age (in years)	SD of age (in years)
Phonology	37	77.1	9.216	1.923
Morphology	9	18.8	9.973	1.012
Syntax	9	18.8	9.944	1.054
Semantics	17	35.4	9.292	2.257
Pragmatics	2	4.2	8.570	0.806

Table 3: Characteristics of the included studies.

Study						Participants		Language-based risk factors
Authors	Year	Design	Method	Language	Context	Sample size	Mean age	Language-based risk factors
Antón-Méndez et al.	2018	Quantitative	Cross-sectional	Spanish	School	48	10.58	Syntax • Syntactic processing
Araújo et al.	2011	Quantitative	Cross-sectional	Portuguese	Clinic	87	9.40	Phonology • RAN
Backes et al.	2002	Quantitative	Cross-sectional	Dutch	Clinic	16	11.50	Phonology • Phonological processing Semantics • Semantic processing

Note. Sample size includes CWDD and control groups. Mean age only includes CWDD. RAN= rapid automized naming; PA= phonological awareness; WM= working memory

Berthiaume and Daigle	2014	Quantitative	Cross-sectional	French	School	82	10.10	Morphology • Inflectional morphology
Bexkens et al.	2014	Quantitative	Cross-sectional	Dutch	Dyslexia centre	117	10.90	Phonology • Phonological processing • RAN • PA
Bloom et al.	2013	Quantitative	Cross-sectional	English	Neuro-psychiatric medical facility	55	10.00	Phonology • Phonological processing
Boets et al.	2010	Quantitative	Longitudinal study	Dutch	School	62	5.30	Phonology • RAN • PA • WM

Note. Sample size includes CWDD and control groups. Mean age only includes CWDD. RAN= rapid automized naming; PA= phonological awareness; WM= working memory

Burani et al.	2008	Quantitative	Cross-sectional	Italian	Not specified	68	11.30	Morphology • Inflectional morphology
Cantiani et al.	2015	Quantitative	Cross-sectional	Italian	School	32	10.09	Phonology • Phonological processing Morphology • Inflectional and derivational morphology
Cardillo et al.	2018	Quantitative	Cross-sectional	Italian	Clinic	63	9.14	Pragmatics • Linguistic pragmatics

Note. Sample size includes CWDD and control groups. Mean age only includes CWDD. RAN= rapid automized naming; PA= phonological awareness; WM= working memory

Casalis et al.	2013	Quantitative	Cross-sectional	French	Hospital	74	10.60	Phonology • Phonological processing Morphology • Inflectional morphology Syntax • Syntactic processing
Casalis et al.	2004	Quantitative	Cross-sectional	French	School	66	10.10	Phonology • Phonological processing • PA Morphology • Derivational morphology Syntax • Syntactic processing

Note. Sample size includes CWDD and control groups. Mean age only includes CWDD. RAN= rapid automized naming; PA= phonological awareness; WM= working memory

Dandache et al.	2014	Quantitative	Longitudinal	Dutch	School	57	T1: 5.00 T2: 6.60 T3: 8.60 T4: 11.60	Phonology • RAN • PA
De Groot et al.	2017	Quantitative	Cross-sectional	Dutch	School	1229	9.14	Phonology • RAN • PA
Di Filippo et al.	2006	Quantitative	Cross-sectional	Italian	School	143	10.20	Phonology • RAN
Eisenmajer et al.	2005	Quantitative	Cross-sectional	English	Clinic	69	9.60	Phonology • Phonological processing
Farquharson et al.	2014	Quantitative	Cross-sectional	English	School	51	7.95	Phonology • PA

Note. Sample size includes CWDD and control groups. Mean age only includes CWDD. RAN= rapid automized naming; PA= phonological awareness; WM= working memory

Germano and Capellini	2011	Quantitative	Cross-sectional	Portuguese	School	94	10.00	Phonology • PA
Helland et al.	2016	Quantitative	Cross-sectional	Finnish	School	5672	8.00	Phonology • Phonological processing Pragmatics • Linguistic pragmatics
Jednoróg et al.	2014	Quantitative	Cross-sectional	Polish	Dyslexia centre	81	10.26	Phonology • RAN
Jednoróg et al.	2010	Quantitative	Cross-sectional	Polish	School	36	11.70	Phonology • Phonological processing Semantics • Semantic processing
Jiménez et al.	2004	Quantitative	Cross-sectional	English	School	97	9.15	Syntax • Syntactic processing

Note. Sample size includes CWDD and control groups. Mean age only includes CWDD. RAN= rapid automized naming; PA= phonological awareness; WM= working memory

Krasowicz-Kupis et al.	2009	Quantitative	Cross-sectional	Polish	School	63	9.11	Phonology • RAN • WM Semantics • Vocabulary knowledge
Landerl et al.	2013	Quantitative	Cross-Linguistic cross-sectional Study	English, French, Dutch, German and Finnish	School	2252	9.69	Phonology • RAN • PA • WM

Note. Sample size includes CWDD and control groups. Mean age only includes CWDD. RAN= rapid automized naming; PA= phonological awareness; WM= working memory

Moura et al.	2017	Quantitative	Cross-sectional	Portuguese	School	66	9.00	Phonology • RAN • PA • WM Semantics • Semantic fluency
Moura et al.	2014	Quantitative	Cross-sectional	Portuguese	Special education centre	72	11.04	Phonology • Phonological processing • RAN • PA
Nergård-Nilssen	2006	Mixed (qualitative and quantitative)	Retrospective longitudinal case-study	Norwegian	School	27	T1: 2.00 T2: 2.50 T3: 3.00 T4: 4.00 T5: 5.00	Phonology • PA

Note. Sample size includes CWDD and control groups. Mean age only includes CWDD. RAN= rapid automized naming; PA= phonological awareness; WM= working memory

							T6: 6.00 T7: 7.00 T8: 7.50 T9: 10.30	
Nithart et al.	2009	Quantitative	Cross-sectional	French	Hospital	34	11.20	Phonology • Phonological processing • PA
Penolazzi et al.	2008	Quantitative	Cross-sectional	Italian	Neuro-psychiatric medial facility	42	10.12	Phonology • Phonological processing Semantics • Semantic processing
Plaza et al.	2002	Quantitative	Cross-sectional	French	Not specified	78	10.40	Phonology • WM Syntax • Syntactic processing

Note. Sample size includes CWDD and control groups. Mean age only includes CWDD. RAN= rapid automized naming; PA= phonological awareness; WM= working memory

								Semantics • Semantic fluency
Puolakanaho et al.	2007	Quantitative	Longitudinal	Finnish	Not specified	198	T1: 3.50 T2: 4.50 T3: 5.50	Phonology • RAN • PA
Quaglino et al.	2008	Quantitative	Cross-sectional	French	Not specified	18	10.90	Phonology • Phonological processing
Ramus et al.	2013	Quantitative	Cross-sectional	English	Special education centre and school	86	10.82	Phonology • Phonological processing Syntax • Syntactic processing Semantics • Vocabulary knowledge

Note. Sample size includes CWDD and control groups. Mean age only includes CWDD. RAN= rapid automized naming; PA= phonological awareness; WM= working memory

Rispens & Been	2007	Quantitative	Cross-sectional	Dutch	Not specified	34	8.66	Phonology • PA • WM Morphology • Inflectional morphology Syntax • Syntactic processing
Rispens et al.	2004	Quantitative	Cross-sectional	Dutch	School	61	8.09	Morphology • Inflectional morphology Syntax • Syntactic processing
Sabisch et al.	2006	Quantitative	Cross-sectional	School	German	32	11.10	Phonology • PA Syntax • Syntactic processing

Note. Sample size includes CWDD and control groups. Mean age only includes CWDD. RAN= rapid automized naming; PA= phonological awareness; WM= working memory

								Semantics • Semantic processing
Schulz et al.	2009	Quantitative	Cross-sectional	German	Not specified	57	11.60	Semantics • Semantic processing
Schulz et al.	2008	Quantitative	Cross-sectional	German	School	52	12.20	Semantics • Semantic processing
Sprenger-Charolles et al.	2009	Quantitative	Multi Case Study	French	Special education centre	116	9.25	Phonology • Phonological processing
St-Pierre and Béland	2010	Mixed (quantitative and qualitative)	Cross-sectional	French	School	30	10.82	Phonology • PA • WM Morphology • Inflectional morphology

Note. Sample size includes CWDD and control groups. Mean age only includes CWDD. RAN= rapid automized naming; PA= phonological awareness; WM= working memory

Talli et al.	2016	Quantitative	Cross-sectional	Greek	Hospital	75	7.57	Phonology • PA • WM Semantics • Vocabulary knowledge
Thompson et al.	2015	Quantitative	Longitudinal	English	School	162	T1: 3.50 T2: 4.50 T3: 5.50 T4: 6.50	Phonology • RAN • PA Semantics • Vocabulary knowledge
Tunmer and Chapman	2006	Quantitative	Longitudinal Cohort Study	English	School	93	T1: 5.08 T2: 5.41 T3: 6.41 T4: 6.80 T5: 7.50	Phonology • Phonological processing Semantics • Vocabulary knowledge

Note. Sample size includes CWDD and control groups. Mean age only includes CWDD. RAN= rapid automized naming; PA= phonological awareness; WM= working memory

Van Bergen et al.	2014	Quantitative	Longitudinal	Dutch	School	212	T1: 3.92 T2: 4.43 T3: 8.00	Semantics • Vocabulary knowledge
Van Rijthoven et al.	2018	Quantitative	Cross-sectional	Dutch	Clinic	55	8.55	Semantics • Vocabulary knowledge
Vender et al.	2017	Quantitative	Cross-sectional	Italian	School and special clinical speech centres	52	10.00	Phonology • PA • WM Morphology • Inflectional morphology Semantics • Vocabulary knowledge
Wachinger et al.	2018	Quantitative	Longitudinal	German	School	31	T1: 6.90 T2: 7.80 T3: 8.20	Phonology • Phonological processing

Note. Sample size includes CWDD and control groups. Mean age only includes CWDD. RAN= rapid automized naming; PA= phonological awareness; WM= working memory

Wise et al.	2007	Quantitative	Cross-sectional	English	School	279	7.60	Semantics • Vocabulary knowledge

Note. Sample size includes CWDD and control groups. Mean age only includes CWDD. RAN= rapid automized naming; PA= phonological awareness; WM= working memory

Phonology. Early phonologically-based risk factors, specifically PA, letter knowledge, and executive skills, were identified as significant predictors of DD upon school entry (Thompson et al., 2015). Puolakanaho et al. (2007) found that letter knowledge and RAN were the best predictors at 3.5 and 5.5 years, while at 4.5 years letter knowledge and PA were the best predictors. St-Pierre and Béland (2010) exposed positive correlations between PA, WM, and reading ability in DD. Bloom et al. (2013) and Bexkens et al. (2014) agree that RAN and phonological processing predict reading and spelling abilities.

Ten studies identified deficits associated with phonological processing (PA, WM, and phonological retrieval) in CWDD (Cantiani et al, 2015; Casalis et al., 2004, 2013; Eisenmajer et al., 2005; Helland et al., 2016; Moura et al., 2014, Nithart et al., 2009; Ramus et al., 2013; Sprenger-Charolles et al., 2009; Tunmer & Chapman, 2006). Two studies found that deficits within these sub-lexical skills increased as the complexity increased (Cantiani et al., 2015; Sprenger-Charolles et al., 2009). Six studies confirmed the phonological deficit hypothesis (Backes et al., 2002; Bloom et al., 2013; Jednoróg et al. 2014; Penolazzi et al., 2008; Wachinger et al., 2018; Quanglino et al., 2008). Wachinger et al. (2018) determined that neurophysiological differences were mainly present in the temporoparietal region in DD. Backes et al. (2002) found reduced activation within the temporal and prefrontal cortex in CWDD (Jednoróg et al., 2010, 2014). Brain areas associated with visuospatial processing instead of specialised language areas were activated in DD. Bloom et al. (2013) found an association between DD and a significantly leftward asymmetry of the planum temporale.

Phonological processing skills significantly correlated with RAN abilities (Bexkens et al., 2014). CWDD presented with significant deficits in RAN (digits, letters, objects, and colours) (de Groot et al., 2017; Di Filippo et al., 2006; Krasowicz-Kupis et al., 2009). Bexkens et al. (2014) stressed the multifaceted nature of RAN. Phonological processing (including PhA) (Bexkens et al., 2014), processing speed, and interference control contributed to RAN abilities

in DD (Krasowicz-Kupis et al., 2009). A significantly slower performance of RAN components was identified in CWDD. This may be indicative of a generalised impairment in the speed of accessing the mental lexicon (Krasowicz-Kupis et al., 2009), influencing reading fluency (Moura et al., 2014). Krasowicz-Kupis et al. (2009) also reported a significant correlation between RAN performance and cognitive interference control. RAN of colours and objects placed greater demands on inhibition control of CWDD. These authors reported that phonological skills and processing speed are the connection between naming speed and reading abilities. RAN significantly correlated with text comprehension. Landerl et al. (2013) revealed that RAN is a strong risk factor for various orthographic complexities. Nevertheless, the impact of RAN and phoneme deletion in complex orthographies was stronger. The correlation between RAN and reading ability was found to be dependent on a child's reading level and age (Araújo et al., 2011), with RAN deficits increasing as CWDD developed (Di Filippo et al., 2006). Boets et al. (2010) found that RAN solely predicted reading speed and accuracy after two years of reading instruction.

In contrast, two studies found that RAN and PA skills were significant factors pertaining to reading accuracy and speed in CWDD up until sixth grade (Dandache et al., 2014; Moura et al., 2014). Three studies identified PA and PhA as risk factors of DD (Germano & Capellini, 2011; Nithart et al., 2009; Talli et al., 2016). One study reported that PA is the best predictor of word and pseudo-word reading abilities in DD (Vender et al., 2017). Farquharson et al. (2014) noted the following: deviant and immature phonological representations influencing CWDD, lexical organisation skills resulting in continued deficits in segmentation, intra-word discrimination and inter-word organisation within reading (Nergård-Nilssen, 2006). Cassalis et al. (2004) confirmed a phonological segmentation impairment in DD. Sabisch et al. (2006) found that CWDD present with impairments in the processing of both segmental and suprasegmental information. Rispens and Been (2007) identified that the population presents

with deficits during the deletion of consonants in the initial word position. PhA, specifically phoneme deletion, showed a strong correlation with DD (Landerl et al., 2013). Moura et al. (2014, 2017) established that PA was the most reliable cognitive predictor of DD, followed by naming speed deficits, and WM (Moura et al., 2014, 2017).

The final aspect studied within the phonological system documented impaired WM in CWDD (Talli et al., 2016). Farquharson et al. (2014) specifically reported that CWDD present with difficulties in repeating multi-syllabic words due to a disturbance in the storing, retrieving, encoding, and processing of phonological information resulting in the inability to develop strong phonological representations. Rispens and Been (2007) also reported pseudo-word repetition difficulties. Vender et al. (2017) concur, finding that WM acts as a risk factor for pseudo-word reading accuracy. One study found that WM deficits were present until third grade, but were absent in sixth grade, due to compensatory strategies employed by CWDD (Dandache et al., 2014). Deficits within the phonological system are interconnected and act as risk factors for DD from pre-literacy until sixth grade.

Morphology. Currently, inflectional morphology is the main literature focus in CWDD. Rispens et al. (2004) found significant inflectional morphology deficits in DD. Three studies (Rispens et al., 2004; Rispens & Been, 2007; St-Pierre & Béland, 2010) found delayed inflectional markings; however, inflected verbs were more delayed than adjectives. Casalis et al. (2013) concur, reporting a reduced understanding and use of inflectional markers on adjectives in both reading and listening modalities. CWDD performed better in listening modalities when compared to written, due to ineffective morphological awareness abilities. Vender et al. (2017) agree that they have insufficient abilities to apply inflectional rules during written pseudo-word pluralisation tasks. Higher error rates occurred as the morphological complexity for words and pseudo-words that included nouns, verbs and determiners increased. CWDD were found to be more impaired in inflecting and applying common rules to pseudo-

words than words, indicating deficits within morphological awareness. Nevertheless, Burani et al. (2008) discovered increased accuracy when pseudo-words composed of a root and derivational suffix were involved. Berthiaume and Daigle (2014) identified deficits within derivational morphology during decomposition and plausibility tasks during writing. Cantiani et al. (2015) found that this behavioural component correlates with deviant electrophysiological patterns due to the enhancement of the N400 component. CWDD present with some degree of morphological knowledge used as a compensatory strategy during reading (Casalis et al., 2004). Berthiaume and Daigle (2014) and Vender et al. (2017) indicated that morphological skills significantly correlate with reading proficiency in CWDD. All studies indicated that CWDD have less sophisticated morphological skills. Compromised inflectional and derivational morphological abilities are a risk factor for DD.

Syntax. Syntactic risk factors included syntactic processing deficits. Jiménez et al. (2004) found that impairment within syntactic processing is characterised by phonological processing abilities in CWDD. Deficits are exaggerated during gender and number agreement tasks due to increased phonological demands. Plaza et al. (2002) concur that WM influences syntactic processing. A syntactic processing deficit was noted during sentence completion tasks. Casalis et al. (2004, 2013) confirmed syntactic comprehension delays within the oral and written modality. Comprehension of the written modality specifically, was significantly impaired as greater demands were placed on WM. An event-related potentials study (Sabisch et al., 2006) supports the explanation that syntactic processing is inhibited by phonological deficits. Their population presented with a delayed left lateralised anterior negativity pattern, indicating an alteration in hemispheric distribution during syntactic negativity and delayed automatized processes of phrase structure building. During auditory sentence comprehension, unimpaired controlled processes of syntactic reanalyses (P600) were noted. Rispens and Been (2007) concur that WM is the basis of deficits in morphosyntactic skills in DD. These authors

found poor pseudo-word repetition and grammatical judgement performances in CWDD at the age of 8 years. These six studies support the processing limitation hypothesis of poor syntactic comprehension in dyslexia being a result of overload in WM during sentence processing as opposed to a result of a delay in reading ability (Rispens et al., 2004).

Still, controversy seems to exist regarding the nature of syntactic deficits in DD. Antón-Méndez et al. (2018) specified that syntactic deficits in DD are not a result of phonological deficits, irrespective of the modality. Ramus et al. (2013) oppose the view that syntactic processing is impaired, but classified poor syntactic performance as a delay and not a deficit in CWDD. These children are susceptible to syntactic errors not due to atypical syntactic representations, but because of less robust syntactic representations.

Semantics. The semantic component included semantic processing, vocabulary, and semantic fluency risk factors in CWDD. Four studies (Jednoróg et al., 2010; Schulz et al., 2008, 2009; Sabisch et al., 2006) focused on semantic processing of congruent and incongruent words at a neurological level. Semantic processing was identified as a developmental delay (Schulz et al., 2009). Penolazzi et al. (2008) accept that semantic processing difficulties are present in DD due to a right posterior activation resulting in an altered delta EEG band during semantic processing tasks. Backes et al. (2002) identified substantial bilateral inferior prefrontal activation during semantic processing tasks - semantic categorisation. CWDD performed significantly slower and less accurate on these tasks. Van Bergen et al. (2014) established that nonverbal IQ aspects, such as semantic categorisation, are unique risk factors of reading abilities as CWDD at age 8 presented with relatively poor verbal and nonverbal IQ at age 4 years.

Vender et al. (2017) found no correlation between receptive vocabulary or nonverbal intelligence and reading abilities in DD. However, Van Bergen et al. (2014) and Wise et al. (2007) found that verbal abilities are uniquely related to later reading. Van Bergen et al. (2014)

recognised that CWDD at 8 years presented with early oral language deficits, including weak vocabulary, poor expressive syntax and WM at 4.5 years. Tunmer and Chapman (2006) concur that significant differences relating to oral language deficits, specifically receptive vocabulary, between CWDD and younger controls exist. Thompson et al. (2015) agree that receptive and expressive vocabulary skills were significant risk factors of DD at age 5.5 through 7 years. Van Rijthoven et al. (2018) revealed that CWDD's semantic abilities act as risk factors of word and pseudo-word decoding abilities by means of phonological abilities (RAN and WM). Vocabulary knowledge directs the development of pre-reading competence (Wise et al., 2007). Two studies (Moura et al., 2017; Plaza et al., 2002) reported significant semantic fluency delays in CWDD. The studies widely acknowledge that delayed semantic processing development in CWDD as well as semantic fluency and vocabulary knowledge influence pre-reading development and act as risk factors for DD.

Pragmatics. Cardillo et al. (2018) and Helland et al. (2016) identified pragmatic risk factors in CWDD. Both studies investigated different aspects of linguistic pragmatics. The former reported impairment specifically related to conversational skills, while the latter found weaker inferential processing related to linguistic pragmatics in CWDD with associated language difficulties. Significant group effects were found when CWDD needed to extract linguistic information within context through inferential processes. CWDD also performed significantly worse during the comprehension of figurative language compared to typical developing children. Limited ToM (specifically social perception abilities) was noted in CWDD, when language was compromised. Both studies confirmed the existence of pragmatic risk factors in DD.

Table 4: Identified language-based risk factors.

Language components	Risk factors	Number of articles	Percentage of articles	Mean age (in years)	SD of age (in years)
Phonology	Phonological processing	17	35.4%	9.982	1.475
	RAN	13	27.1%	8.576	2.239
	PA	19	39.6%	8.693	2.243
	WM	9	18.8%	8.950	1.680
Morphology	Inflectional	6	12.5%	9.710	1.094
	Derivational	4	8.3%	10.398	0.602
Syntax	Syntactic processing	9	18.8%	9.944	1.054
Semantics	Semantic processing	6	12.5%	11.370	0.708
	Semantic fluency	2	4.2%	9.700	0.990
	Vocabulary	9	18.8%	7.816	2.004
Pragmatics	Linguistic pragmatics	2	4.2%	8.570	0.806

Discussion

We used an explicit, reproducing method to diminish bias that allowed for systematic presentation of our findings (Higgins & Green, 2011). Up to date, no systematic review considered language-based risk factors associated with DD. Novel and robust language-based risk factors, on a behavioural and neurological level, were identified in DD during the preliterate, early, and later stages of literacy.

Phonology was the language risk factor reported the most. RAN, PA and WM were identified as risk factors for DD. These risk factors became significant predictors at school entry. Six studies found that RAN abilities are strong risk factors as significant delays in RAN abilities were reported, especially during the naming of objects and colours. RAN abilities were reported to be a stronger risk factor in opaque orthographies. Two studies found that during the preliterate and early reading stage RAN and PA are significant risk factors for reading accuracy and speed, while RAN became the only predictor during later literacy development. Dandache et al. (2014), however, reported that PA remains a strong predictor up until sixth grade. Thirteen studies found that CWDD present with impairments in PA and PhA. These difficulties stem from impaired phonological representations and the impaired ability to reorganise lexical organisations. Working memory impairments were investigated in five studies. Repetition tasks exceeding five-syllable items, pseudo-words and low-frequency words, digits and sentence repetition were the most challenging. The behavioural phonological-based risk factors were in accordance with the neurological observations. Letter knowledge and RAN were risk factors of DD at age 3.5, 4.5, and 5.5 years. Letter knowledge and PA acted as risk factors for DD at 4.5 years. These risk factors were reliable cognitive predictors and sensitive enough for the clinical diagnosis of DD (Moura et al., 2014, 2017).

Morphological knowledge is not independent of phonological skills (Casalis et al., 2004). Five studies found delayed inflectional morphological abilities in CWDD - in both the comprehension and use of inflectional markers on nouns, verbs, and adjectives of words and pseudo-words - due to poor morphological awareness. CWDD read pseudo-words composed of a root and suffix faster and more accurate than simple pseudo-words due to the benefit of the morphological structure (Angelelli, Marinelli, & Burani, 2014). More distinctively, they rely on morphemic parsing because of underdeveloped whole-word lexical processing. CWDD are more sensitive to inflectional morphology than derivational morphology (Cantiani et al., 2015). Consequently, they present with impaired derivational morphology. This population also presents with significant error effects during the reading of pseudo-words compared to skilled readers and younger children. These behavioural observations were in accordance with electrophysiological research, indicating the enhancement of the N400 component that is due to the retrieval of explicit rules to compensate for the difficulty in creating implicit rules for dealing with inflectional morphology. Thus, both inflectional and derivational morphology were identified as risk factors for DD.

Syntax is the third component of language included in the review. Controversy exists regarding the limitation processing hypothesis. Eight of the nine studies confirmed this hypothesis as a cause for impaired syntactic abilities in DD. Syntactic impairments during oral and written modalities stem from phonological processing deficits. CWDD performed better within the spoken modality as a result of reduced demands. An electrophysiological study concurred with these behavioural results as delayed left lateralised anterior negativity patterns were noted altering the hemispheric distribution. In contrast, Antón-Méndez et al. (2018) indicated that a syntactic processing disorder is not due to a phonological processing deficit. Ramus et al. (2013), however, found that CWDD present with a delay and not a syntactic deficit. The majority of research, however, concur that syntactic processing deficits are risk

factors for DD, but more detailed and specific risk markers within syntax need to be determined through future research.

The second most investigated language-based risk factor entailed semantics. Vocabulary, influencing a child's pre-reading competence, was established as a significant risk factor at the age of 5.5 until 7 years for DD. CWDD also presented with delayed semantic fluency. Semantic fluency is an indicator of executive functioning and linguistic abilities (Oriá, Costa, Lima, Patrick, & Guerrant, 2009). Semantic processing difficulties were identified in CWDD.

Finally, two studies investigated pragmatic abilities. Children with DD present with difficulties in linguistic pragmatics. Helland et al. (2016) found deficits within conversational skills while Cardillo et al. (2018) found difficulties within inferential processing regarding metaphors and the implicit comprehension of meaning and contexts. Interestingly, Cardillo et al. (2018) also reported poor ToM abilities in CWDD as it was closely related to pragmatics. Language and social skills are the basis for pragmatic abilities (Cardillo et al., 2018) although pragmatic abilities are more extensive than these two risk factors. Currently, literature reports on the investigation of two pragmatic aspects and indicates that poor conversational abilities and linguistic pragmatics, including ToM, act as risk-factors for DD.

A credible level of evidence was obtained about language-based risk factors in CWDD. The studies included were well-designed and mainly achieved a rating of IIb on the ASHA level of evidence rating scale (ASHA, 2004). No study attained a rating level of IIa or above.

Limitations

In the current systematic review, language bias may have occurred as only articles written in English were included. Mainly cross-sectional study designs were obtained, which impeded the identification of causality between the risk factors.

Directions for Future Research and Conclusion

Over the last decade, a significant number of studies have investigated phonological risk factors in CWDD, while less evidence is available concerning the broader language abilities. Our review obtained credible evidence that CWDD does not only present with PA, RAN and WM risk factors but we also identified deficits within inflectional and derivational morphology, syntactic processing, semantic processing, semantic fluency, vocabulary knowledge and linguistic pragmatics. It is important to pinpoint the onset of these language risk factors as reading experience influence language development. Language starts to develop within the first year of a child's life; however, only a limited number of studies investigated children younger than 6 years. Future research should employ retrospective and longitudinal study designs to allow for more comprehensive and explicit language-based risk factors in CWDD. This will enable the in-depth examination of risk factors within pragmatics, semantics, syntax, and morphology.

Contributors

All the authors contributed to the article.

Conflict of Interest

The authors declared no competing interests.

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Chapter 4: Discussion and Conclusion

"Dyslexia is a different brain organisation that needs different teaching methods. It is never the fault of the child, but rather the responsibility of us who teach to find methods that work for that child."

Dr Maryanne Wolf

Chapter aim:

The final chapter aims to synthesise and discuss the evidence related to the aims of the current systematic review. Additionally, the objective of this chapter is to discuss the strengths and limitations of the current review, as well as the implication of the findings. Finally, areas in need of future research were identified, and a conclusion was reached.

The current systematic review ensured a balanced and systematic presentation of the findings by employing an explicit, reproducing methodology that also diminished bias (Higgins & Green, 2011). The main objective of this review was to identify the language-based risk factors in alphabetic orthographies in CWDD as well as to determine the strength and level of evidence. Up to date, no systematic review investigated language-based risk factors in CWDD. Therefore, the risk factors associated with phonology, morphology, syntax, semantics, and pragmatics needed to be comprehensively appraised and synthesised. A wide variety of orthographic languages were investigated across 18 countries. Also, several contexts, including schools, special education institutions, dyslexia centres, and medical facilities, were explored. This allowed for a wide variety of participants from several contexts which may permit generalisation to a larger population. Novel and robust language-based risk factors in CWDD were identified, on a behavioural and neurological level, in the current review during the preliterate, early, and later stages of literacy. The behavioural and

neurological relationships were more intricate than anticipated due to the complex nature of DD.

4.1. Language-Based Risk Factors

Reduced phonological ability is a well-established risk factor of DD and was the most reported language-based risk factor in the current systematic review. All three dimensions of the phonological system - RAN, PA, and WM - are compromised in CWDD and were identified as risk factors for DD. Snowling and Melby-Lervåg (2016) concur with these results with the addition that CWDD present with significant difficulties in letter knowledge. These risk factors became significant predictors at school entry. Phonological processing significantly correlates with RAN as an independent skill (Bexkens, van den Wildenberg, & Tijms, 2014). Six studies found that poor RAN abilities are a strong risk factor for DD in children, especially the naming of objects and colours. Children with DD found low-frequency word and pseudoword repetition tasks the most difficult (Snowling & Melby-Lervåg, 2016), followed by digits and sentence repetition (Adlof & Hogan, 2018). The reason for this is that CWDD have difficulty in retrieving phonological codes, to encode the segmental phonological representations, and to plan articulatory movements. Rapid automized naming abilities were reported to be a stronger risk factor in opaque orthographies. The association between reading ability and the RAN components are dependent on a child's reading level and age (Araújo et al., 2011). Two studies reported that during the preliterate and early reading stage RAN, and PA became significant risk factors for reading accuracy and speed. The degree of RAN deficits became greater as children with DD developed (Di Filippo et al., 2006), thus RAN became the only predictor during later literacy development. Nevertheless, PA remains a strong predictor up until sixth grade (Dandache, Wouters, & Ghesquière, 2014). Thirteen studies found that CWDD present with impairments in PA and

the subcategory PhA. These difficulties originate from impaired phonological representations as well as the impaired ability to reorganise lexical organisations. Working memory impairments were investigated in five studies. Children with DD presented with difficulties in repetition tasks exceeding five-syllable items, pseudowords and low-frequency words, digits and sentence repetition. Working memory deficits in CWDD were present until third grade but not at sixth grade, due to the development of a compensating mechanism (Dandache et al., 2014). Vender et al. (2018) found that WM acts as a predictor for pseudoword reading accuracy. The behavioural phonological-based risk factors were in accordance with the neurological observations. Letter knowledge and RAN were risk factors of DD at age 3.5, 4.5, and 5.5 years. While letter knowledge and PA acted as risk factors for DD at 4.5 years. Moura, Moreno, Pereira, and Simões (2014) and Moura et al. (2017) specified that these risk factors were reliable cognitive predictors and sensitive enough for clinical diagnosis of DD.

Morphological knowledge is not independent of phonological skills (Casalis et al., 2004) as deficits within phonology inhibit the development of morphological awareness. Currently, in literature, inflectional morphology is the focus due to its crucial role in language acquisition from an early age. Five studies found that CWDD present with delayed inflectional morphological abilities. The studies indicated that CWDD specifically present with difficulties in both the understanding and use of inflected nouns, verbs, and adjectives on words and especially pseudowords, due to poor morphological awareness (Vender et al., 2018). These children read pseudowords composed of a root and suffix faster and more accurate than simple pseudowords due to the benefit of the morphological structure (Angelelli, Marinelli, & Burani, 2014). More specifically, they rely on morphemic parsing because of underdeveloped whole-word lexical processing. Morphemic parsing acts as a compensatory strategy to aid this population with limited reading ability in processing linguistic stimuli (Burani, Marcolini, De Luca, & Zoccolotti 2008). Inflectional morphology

develops relatively early, while derivational morphology continues to develop throughout school (Kuo & Anderson, 2006). Children with DD are more sensitive to inflectional morphology than derivational morphology (Cantiani, Lorusso, Perego, Molteni, & Guasti, 2015). Consequently, they present with impaired derivational morphology during decomposition and plausibility tasks. This population also presents with significant error effects during the reading of pseudowords compared to skilled readers and younger children. These behavioural observations were established during electrophysiological research, indicating the enhancement of the N400 component is due to the retrieval of explicit rules to compensate for the difficulty in creating implicit rules for dealing with inflectional morphology. Snowling and Melby-Lervåg (2016) confirmed that morphological abilities are predictors of reading. In the current systematic review, inflectional morphology, specifically, inflected verbs, nouns, and determiners as well as derivational morphology specifically, decomposition and plausibility derivations, were identified as risk factors for DD.

The systematic review investigated syntax as the third language-based component. Currently, in literature, controversy exists regarding the limitation processing hypothesis that poor syntactic comprehension in dyslexia is a result of overload in working memory during sentence processing. Eight of the nine studies confirmed this hypothesis as a cause for impaired syntactic abilities in DD. Phonological processing deficits give rise to syntactic impairments within the oral and written modalities (Jiménez et al., 2004). Children with DD performed better within the spoken modality as a result of reduced demands. These children found number and gender agreement tasks difficult due to these being more phonologically complex, while function word and grammatical structure tasks were less difficult due to the contextual information (Jiménez et al., 2004). An electrophysiological study concurred with the limitation processing hypothesis as delayed left lateralised anterior negativity patterns were noted altering the hemispheric distribution. In contrast, Antón-Méndez et al. (2018)

indicated that a syntactic processing disorder is not due to a phonological processing deficit. Other authors agreed that poor syntactic skills are mainly independent of phonological processing difficulties in DD (Antón-Méndez et al., 2018). Regardless of the afore-mentioned, Ramus et al. (2013) found that CWDD present with a delay and not a syntactic deficit. Adlof and Hogan (2018) performed a systematic review and also identified that CWDD present with significantly delayed syntactic processing compared to typical developing children. However, although CWDD present with a delay in syntactic processing, it is not fully explained by a reading delay but is correlated with reading level. The majority of research, nevertheless, concur that syntactic processing deficits are risk factors for DD, but more detailed and specific risk markers within syntax need to be determined within future research.

The second most investigated language-based risk factor entailed semantics. A delay in vocabulary was found in CWDD (Adlof & Hogan, 2018; Snowling & Melby-Lervåg, 2016). Vocabulary was found to be a significant risk factor at the age of 5.5 until seven years in CWDD. Vocabulary knowledge influences a child's pre-reading competence (Wise, Sevcik, Morris, Lovett, & Wolf., 2007). Snowling and Melby-Lervåg (2016) also recognised the association between reading and receptive and expressive vocabulary. The receptive vocabulary was stronger related to pre-reading skills, while expressive vocabulary knowledge significantly predicted real word identification abilities in CWDD. Van Setten, Hakvoort, van der Leij, Maurits, and Maassen (2018) concur that vocabulary knowledge significantly correlates with reading fluency and reading comprehension. The second aspect found to be delayed in CWDD involved semantic fluency. Oriá, Costa, Lima, Patrick, and Guerrant (2009) established that semantic fluency is an indicator of executive functioning and linguistic abilities. Overall, CWDD present with semantic processing difficulties. These children found semantic tasks more difficult than orthographic and phonological tasks

(Penolazzi et al., 2008). Semantic abilities act as risk factors for word and pseudoword decoding abilities using phonological abilities (RAN and WM) in DD. Therefore, by improving semantic abilities in children with DD, their phonological skills will improve, which will encourage reading development (van Rijthoven et al., 2018). These behavioural findings were supported by neurological findings. Deficits within vocabulary, semantic fluency, and semantic processing were identified as risk factors in DD.

Pragmatic abilities were the final language component reviewed in the present study. Two studies established that CWDD presented with difficulties in linguistic pragmatics (Cardillo et al., 2018; Helland et al., 2016). These studies investigated different aspects of linguistic pragmatics. Helland et al. (2016) found deficits within conversational skills while the second study (Cardillo et al., 2018) found difficulties within inferential processing specifically metaphors and the implicit comprehension of meaning and contexts. Interestingly, Cardillo et al. (2018) also reported poor ToM abilities in CWDD as these are closely related to linguistic pragmatics. Language and social skills are the foundation for pragmatic abilities (Cardillo et al., 2018). However, pragmatic abilities are more extensive than these two established risk factors. Pragmatics includes speech acts, linguistic and conversational codes, conversational abilities and speech parameters, inferences, and referential communication (Cardillo et al., 2018). This systematic review identified that poor conversational abilities, inferential processing, as well as ToM, act as risk-factors for DD.

4.2. Level of Evidence

A credible level of evidence was obtained, with regard to language-based risk factors in CWDD. The studies included were well-designed, and two (4,17%) studies achieved a level III on the ASHA level of evidence rating scale (ASHA, 2004). Moreover, 46 (95,83%) studies achieved a rating of IIb. These publications did not utilise randomisation. However,

they encompassed within-subject comparisons which allowed for reduced error variances. No study attained a rating level of IIa or above. Randomised studies may advance research in DD. Nevertheless, the majority of studies obtained an evidence rating of IIb, reflecting a high level of evidence. Furthermore, a total of forty-four (91.67%) articles were classified as "low risk of bias" indicative of a good level of quality.

4.3. Strengths and Limitations

Numerous strengths were identified for the current systematic review. The first strength entails the adherence to the PRISMA-P guidelines as it allowed for valid and reliable reporting of evidence and limited possible bias. Furthermore, the eligibility criteria also acted as a strength as it necessitated that children in the various publications were required to be diagnosed with DD. These specific diagnoses eliminated reading disorders related to inadequate intellectual abilities, sensory difficulties, neurological disorders or insufficient education as DD is not a result of these factors. The timeframe of the current review only allowed for children diagnosed with the widely recognised definition of DD to be included.

The current systematic review also presented with some limitations that may contribute to bias. A limited variation of study designs was included in the current review. Mainly cross-sectional study designs were attained, which obstructed the identification of causality between the risk factors. Articles investigating non-alphabetic languages were excluded due to the variance in the orthographic structure, which may have resulted in valuable publications being excluded. Additionally, only studies available in English were included, thus language bias may have occurred. Finally, heterogeneity may have occurred in the current systematic review. The included studies used a wide variety of terminology to define and describe concepts. Several studies used several terms to describe and label

working memory and phonological processing. These variances may act as a limitation due to unclear reporting of concepts, influencing the ability to draw interferences.

4.4. Clinical Implications

The current systematic review may clarify the language-based risk factors in CWDD as well as the level of evidence for current health care providers. The literature identified significant disparities between early and late identification of DD (Catts et al., 2008; Peart, 2013). Earlier identification supports optimal developmental outcomes which can minimise reading failure (Gabrieli, 2009). Therefore, identifying the specific language-based risk factors is important as it may enable clinicians to identify and ensure appropriate interventions in this population. Overall, the identification and categorisation of these risk factors may contribute to the development of a preventative intervention to reduce the persistent nature of DD. Snowling (2012) explicitly stated that early identification and timely and appropriate intervention is a realistic aim of health-care professionals and policy-makers.

4.5. Recommendations for Future Research

Significant advances have been made over the last decade in the identification of specific language-based risk factors such as phonological factors. Yet, evidence related to pragmatics, syntax, and morphology as risk factors in CWDD, is scant. This lack of evidence is ironic as language is an intricate field, and linguistic components are closely related. Thus, future research should comprehensively investigate each language component. Forthcoming research should also focus on providing more detailed associations between language-based risk factors and DD. Risk factors within pragmatics, semantics, syntax, and morphology should be examined more comprehensively. Furthermore, as language starts to develop within the first year of life, it is alarming that only seven articles were obtained for children younger than six years old. Future research should focus on early language-based risk factors

contributing to DD. Retrospective and longitudinal study designs should be employed as future research designs to allow for more comprehensive and explicit language-based risk factors in children with DD. Finally, future systematic reviews and meta-analysis should aim to combine the multifaceted nature of DD to allow for holistic guidelines on the services provided.

4.6. Conclusion

Over the last decade, a significant number of studies has explored phonological risk factors in CWDD, while less evidence is available concerning the broader language abilities (morphology, syntax, semantics, and pragmatics). The current systematic review obtained credible evidence (primarily an IIb rating on the ASHA evidence rating scale) that CWDD does not only present with PA (8.69 years), RAN (8.58 years), and WM (8.95 years) risk factors but also acknowledged specific deficits at explicit mean ages. These risk factors involve: inflectional (9.71 years) and derivational (10.40 years) morphology, syntactic processing (9.94 years), semantic processing (11.37 years), semantic fluency (9.70 years), vocabulary knowledge (7.82 years) and linguistic pragmatics (8.57 years). It is important to pinpoint the onset of these language risk factors, as reading experience, further influences language development. Language starts to develop within the first year of a child's life; however, only a limited number of studies' participants' sample comprised of participants five years and younger. In summary, the evidence within the review confirmed that the language components are interrelated and that various degrees of deficits and delays are present in CWDD. Tager-Flushberg (2016) stated that the ultimate goal within neurodevelopmental disorders should be to develop preventative intervention according to each child's individualised cluster of risk markers to maximise their potential.

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Appendices

Appendix A: Evidence of the adapted PRISMA-P Checklist in the review (Shamseer et al., 2015)

Section		Checklist item	Reported in article
Administration			
Identification	1a	Identify the report as a protocol of a systematic review	Title page
Update	1b	If the protocol is for an update of a previous systematic review, identify as such	N/A
Registration	2	If registered, provide the name of the registry (e.g., PROSPERO) and registration number in the Abstract	Chapter 2: Method Study design [2.2]
Contact	3a	Provide name, institutional affiliation, and e-mail address of all protocol authors; provide physical mailing address of corresponding author	Chapter 3: Article Title page
Contributions	3b	Describe contributions of protocol authors and identify the guarantor of the review	Chapter 3: Article
Amendments	4	If the protocol represents an amendment of a previously completed or published protocol, identify as such and list changes; otherwise, state plan for documenting important protocol amendments	N/A
Sources	5a	Indicate sources of financial or other support for the review	N/A

Sponsor	5b	Provide name for the review funder and/or sponsor	N/A
Role of sponsor/funder	5c	Describe roles of funder(s), sponsor(s), and/or institution(s), if any, in developing the protocol	N/A
Introduction			
Rationale	6	Describe the rationale for the review in the context of what is already known	Chapter 1: Introduction Rationale [1.4]
Objectives	7	Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO)	Chapter 1: Introduction Research question [1.5]
Method			
Eligibility criteria	8	Specify the study characteristics (e.g., PICO, study design, setting, time frame) and report characteristics (e.g., years considered, language, publication status) to be used as criteria for eligibility for the review	Chapter 2: Method Eligibility criteria [2.3.1]

Information sources	9	Describe all intended information sources (e.g., electronic databases, contact with study authors, trial registers, or other grey literature sources) with planned dates of coverage	Chapter 2: Method Search strategy [2.3.2]
Search strategy	10	Present draft of search strategy to be used for at least one electronic database, including planned limits, such that it could be repeated	Chapter 2: Method Search strategy [2.3.2]
Data management	11a	Describe the mechanism(s) that will be used to manage records and data throughout the review	Chapter 2: Method Data management [2.3.4]
Selection process	11b	State the process that will be used for selecting studies (e.g., two independent reviewers) through each phase of the review (i.e., screening, eligibility, and inclusion in meta-analysis)	Chapter 2: Method Study selection [2.3.3]
Data collection process	11c	Describe planned method of extracting data from reports (e.g., piloting forms, done independently, in duplicate), any processes for obtaining and confirming data from investigators	Chapter 2: Method Study selection [2.3.3]

Data items	12	List and define all variables for which data will be sought (e.g., PICO items, funding sources), any pre-planned data assumptions and simplifications	Chapter 2: Method Eligibility criteria [2.3.1]; data extraction [2.3.5]
Outcomes and prioritization	13	List and define all outcomes for which data will be sought, including prioritization of main and additional outcomes, with rationale	Chapter 2: Method Research aims [2.1]; data analysis [2.5]
Risk of bias in individual studies	14	Describe anticipated methods for assessing risk of bias of individual studies, including whether this will be done at the outcome or study level, or both; state how this information will be used in data synthesis	Chapter 2: Method Risk of bias [2.3.6]
Synthesis	15a	Describe criteria under which study data will be quantitatively synthesized	Chapter 2: Method Data analysis [2.5]

	15b	If data are appropriate for quantitative synthesis, describe planned summary measures, methods of handling data, and methods of combining data from studies, including any planned exploration of consistency (e.g., I^2, Kendall's tau)	N/A
	15c	Describe any proposed additional analyses (e.g., sensitivity or subgroup analyses, meta-regression)	N/A
	15d	If quantitative synthesis is not appropriate, describe the type of summary planned	Chapter 2: Method Data analysis [2.5]
Meta-bias(es)	16	Specify any planned assessment of meta-bias(es) (e.g., publication bias across studies, selective reporting within studies)	Chapter 2: Method Risk of bias [2.3.6]
Confidence in cumulative evidence	17	Describe how the strength of the body of evidence will be assessed	Chapter 2: Method Data extraction [2.3.5]; reliability, validity, and

			trustworthiness [2.4]

Appendix B:

Ethical clearance form – Faculty of Humanities

UNIVERSITEIT VAN PRETORIA
UNIVERSITY OF PRETORIA
YUNIBESITHI YA PRETORIA

Faculty of Humanities
Research Ethics Committee

30 November 2018

Dear Ms Roesch

Project: Language-based risk factors in children with Development Dyslexia: A systematic review
Researcher: D Roesch
Supervisors: Dr S Geertsema and Dr M le Roux
Department: Speech-Language Pathology and Audiology
Reference number: 15082599 (GW20181104HS)

Thank you for the application that was submitted for ethical consideration.

The Research Ethics Committee notes that this is a literature-based study and no human subjects are involved. The application has been approved on 30 November 2018 along these guidelines, data collection may therefore commence.

Please note that this approval is based on the assumption that the research will be carried out along the lines laid out in the proposal. However, should the actual research depart significantly from the proposed research, a new research proposal and application for ethical clearance will have to be submitted for approval.

The Committee requests you to convey this approval to the researcher.

We wish you success with the project.

Sincerely

Prof Maxi Schoeman
Deputy Dean: Postgraduate Studies and Ethics
Faculty of Humanities
UNIVERSITY OF PRETORIA
e-mail:tracey.andrew@up.ac.za

CC: Dr S Geertsema (Supervisor) and Dr M le Roux (Co-supervisor)
Dr J van der Linde (HoD)

Fakulteit Geesteswetenskappe
Lefapha la Bomaseko

Research Ethics Committee Members: Prof MME Schoeman (Deputy Dean); Prof KL Harris; Mr A Bless; Dr L Blokland; Dr K Booyens; Dr A-M de Boer; Ms A dos Santos; Dr R Fasselt; Ms KT Govinder Andrew; Dr E Johnson; Dr W Kelleher; Mr A Mohamed; Dr C Puttergill; Dr D Reyburn; Dr M Sosa; Prof E Taljard; Prof V Thebe; Ms B Tsebe; Ms D Mokalapa

Appendix C:

Proof of submission – The Annals of Dyslexia

Gmail Darike Roesch <darikeroesch@gmail.com>

ANDY-D-19-00061 - Submission Confirmation

1 message

Annals of Dyslexia <em@editorialmanager.com> Fri, Nov 1, 2019 at 4:49 PM
Reply-To: Annals of Dyslexia <madelyn.marciano@springer.com>
To: Darike Roesch <darikeroesch@gmail.com>

Dear Ms Roesch:

Thank you for submitting your manuscript, "Language-based risk factors in children with developmental dyslexia: A systematic review", to Annals of Dyslexia.

The submission id is: ANDY-D-19-00061
Please refer to this number in any future correspondence.

During the review process, you can keep track of the status of your manuscript.

Your username is: Darike Roesch
If you forgot your password, you can click the 'Send Login Details' link on the EM Login page at https://www.editorialmanager.com/andy/.

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With kind regards,

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