



MONASH University

Identifying the language and cognitive deficits contributing to phonological skill impairment in a sample of children with learning disabilities

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Thesis including published works declaration

I hereby declare that this thesis contains no material which has been accepted for the award of any other degree or diploma at any university or equivalent institution and that, to the best of my knowledge and belief, this thesis contains no material previously published or written by another person, except where due reference is made in the text of the thesis.

This thesis includes three original submitted publications. The core theme of the thesis is an examination of specific topics related to learning disabilities. The ideas, development and writing up of all the papers in the thesis were the principal responsibility of myself, the student, working within the School of Psychological Sciences under the supervision of Dr Renee Testa, Ms Nola Ross, Dr Caroline Nadebaum and Prof Jennie Ponsford.

The inclusion of co-authors reflects the fact that the work came from active collaboration between researchers and acknowledges input into team-based research.

In the case of Chapters 3 to 5, my contribution to the work involved the following:

Thesis Chapter	Publication Title	Status <i>(published, in press, accepted or returned for revision, submitted)</i>	Nature and % of student contribution	Co-author name(s) Nature and % of Co-author's contribution*	Co-author(s), Monash student Y/N*
3	Structural analysis of phonological skills and related cognitive skills in children referred to a learning difficulties clinic	Submitted	70% contribution by candidate. This included formulation of research design and question, data analysis and manuscript presentation.	1) Renee Testa, formulation of research design and question, data analysis and input into manuscript 20% 2) Caroline Nadebaum, input into manuscript 4% 3) Nola Ross, formulation of research design and question, and input into manuscript 6%	No No No
4	The nature of phonological skill impairments in children with specific language impairment and reading disability	Submitted	70% contribution by candidate. This included formulation of research design and question, data analysis and manuscript presentation.	1) Renee Testa, formulation of research design and question, data analysis and input into manuscript 20% 2) Caroline Nadebaum, input into manuscript 4% 3) Nola Ross, formulation of research design and question, and input into manuscript 6%	No No No
5	Characteristics of children	Submitted	70% contribution by candidate. This included	1) Renee Testa, formulation of research design and question, data	No

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	referred to a multidisciplinary learning difficulties clinic for assessment		formulation of research design and question, data analysis and manuscript presentation.	analysis and input into manuscript 20%	No
				2) Caroline Nadebaum, input into manuscript 4%	
				3) Nola Ross, formulation of research design and question, and input into manuscript 6%	No

I have renumbered sections of submitted or published papers in order to generate a consistent presentation within the thesis.

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Date: 17.04.2019

The undersigned hereby certify that the above declaration correctly reflects the nature and extent of the student's and co-authors' contributions to this work. In instances where I am not the responsible author I have consulted with the responsible author to agree on the respective contributions of the authors.

Main Supervisor signature:

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List of Common Abbreviations

ANOVA	Analysis of variance
CVC	Consonant-vowel-consonant
CELF	Clinical evaluation of language fundamentals
CCVC	Consonant-consonant-vowel-consonant
CFI	Comparative fit index
fMRI	Functional magnetic resonance imaging
FSIQ	Full-scale intelligence quotient
GFI	Goodness of fit index
Ph	Phonological
PIQ	Performance intelligence quotient
RD-only	Reading disability only
RD+SLI	Reading disability and specific language impairment
RMSEA	Root-mean-square error
SD	Standard deviation
SE	Standard error
SLI-only	Specific language impairment only
SPAT	Sutherland phonological awareness test
SRMR	Standardised Root Mean Square Residual
TLI	Tucker-Lewis index
VIQ	Verbal intelligence quotient
WIAT	Wechsler individual achievement test
WISC	Wechsler intelligence scale for children
WRAML	Wide range assessment of memory and learning

Thesis Abstract

Phonological skills underpin the development of early language and have repeatedly been linked to development of reading skills in children. Conversely, poor phonological skills have been hypothesised to be the main contributing factor for impaired reading and language development. Phonological skills have been comprehensively studied, but a number of questions remain unanswered. Firstly, it is unclear whether phonological skills are best represented as a single cognitive construct, or alternatively, as multiple separate constructs. There is evidence to suggest that distinct higher- and lower-level phonological skills constructs can be identified, which are differentiated by cognitive load. Questions also remain as to whether the nature of phonological impairments differ between children with reading disability and specific language impairment.

In addition to poor phonological skills, children with reading disability and other learning disabilities can also display a range of other non-academic impairments which contribute to learning disabilities (including cognitive, language, motor, behavioural and emotional difficulties). Multiple disciplinary assessment clinics can be well suited to provide a thorough evaluation of these children but research examining the characteristics of children who are referred to these clinics is limited.

To address these literature gaps, data was retrospectively analysed from 326 primary school aged children who had undergone multidisciplinary assessment at a learning difficulties clinic between 1996 and 2008. Confirmatory factor analysis, using data from 121 children, showed that phonological skills were best represented by two separate phonological factors. The factors were distinguished by their cognitive load (low vs high). Comparison of the phonological impairments between children with reading disability only (RD-only; N=27), specific language impairment only (SLI-only; N=17) and reading disability and specific language impairment (RD+SLI; N=54), revealed that children with a RD+SLI had more prevalent and severe phonological impairments than those with reading disability only or specific language impairment only. There were no differences in prevalence, severity or pattern of phonological impairments

between children in the single deficit groups (i.e. RD-only and SLI-only), suggesting that phonological impairments may represent a shared cognitive risk factor for these two neurodevelopmental disorders. Descriptive statistics were used to characterise academic and non-academic impairments in the entire sample of children (N=326). Approximately 65% of children met criteria for multiple learning disabilities. A range of cognitive, language, motor, behavioural and emotional impairments were also experienced by these children.

Taken together, the results highlighted cognitive load as an important factor to consider when examining the structure of phonological skills and when assessing children with suspected reading and/or language impairments. Comprehensive assessment of these children should aim to include tasks with varied cognitive load requirements. The findings also emphasised the complex assessment and diagnostic needs of children with suspected learning disabilities, and suggested that the multiple disciplinary learning difficulties clinic was well placed to support those needs.

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Thesis Overview

Learning disability is a heterogeneous term used to describe children who have significant difficulties in the acquisition of literacy and numeracy skills (Louden et al., 2000). Difficulties can occur in a variety of academic domains, but this thesis will focus on significant and unexpected difficulties in word decoding (reading disability), spelling (spelling disability) and/or mathematics (math disability). Identification and subsequent management of learning disabilities is an important issue for teachers, parents and professionals working in this area because of the potential negative outcomes associated with these disabilities. Learning disabilities not only negatively affect classroom learning, but can also be associated with social difficulties (Nowicki, 2003) and emotional and behavioural disorders (Willcutt et al., 2013). Adults with learning disabilities can experience mental health issues (Klassen, Tze, & Hannok, 2011) and other negative outcomes including reduced employment opportunities, lower wages and increased rates of substance use disorders (Beitchman, Wilson, Douglas, Young, & Adlaf, 2001; Geary, 2011). Societal impacts of learning disabilities are widespread. Those who do not obtain functional levels of literacy experience higher levels of poverty, poor health, dependence of social welfare and increased crime participation. Learning disabilities are also associated with increased unemployment, reduced business productivity, lower technology skills capacity and lost wealth creation opportunities for individuals and businesses. These issues emphasise the importance of research aimed at improving diagnosis and outcomes for individuals with learning disabilities.

This thesis aimed to address several questions unanswered by past research by providing an in-depth examination of specific topics related to learning disabilities. This included investigating the structure of phonological skills and how these skills are related to

reading disability and specific language impairment, as well as a broad examination of the type of academic and other (non-academic) impairments experienced by children referred for learning disability assessment. The research included in this thesis aims to contribute to our theoretical knowledge of phonological skills, reading disabilities and learning disabilities more broadly. It also aimed to provide information which can improve the assessment and diagnosis of learning disabilities.

The following literature review consists of two chapters. In Chapter 1, the review primarily focuses upon phonological skills, which are commonly impaired in children with learning disabilities (particularly reading disability). This chapter describes how phonological skills have typically been defined and measured, and provides a review of how phonological skills are thought to develop throughout childhood. The structure of phonological skills is then reviewed in detail, focusing on whether phonological skills are best represented as one construct or multiple constructs. This is a topic of much debate and is the focus of Chapter 3 in this thesis. Within this discussion cognitive load is identified as an important variable. To explore the relationship between phonological skills and cognitive load, a review of short-term memory and working memory is undertaken. These skills contribute to successful performance on phonological tasks and understanding the relationship between phonological skills, short-term memory and working memory facilitates knowledge of how cognitive load and phonological skills are related. Chapter 1 next explores how phonological skills are impacted in children with reading disability and specific language impairment, which is a primary focus of Chapter 4. Emphasis is placed on studies investigating this issue using comparisons of reading disability only (RD-only), specific language impairment only (SLI-only) and reading disability *and* specific language impairment (RD+SLI) groups. Chapter 2 of the review focuses more broadly on learning disabilities. It aims to review the types of difficulties that children with learning disabilities face (including academic, cognitive,

language, motor impairments and emotional and behavioural difficulties). It will also discuss how the broad nature of these difficulties influences the type of assessment process that is best suited to assess these children.

Chapter 1 – Phonological Skills

1.1 Overview of Phonological Skills

1.1.1 Definition.

Phonological skills are defined as an individual's ability to be sensitive to the speech sound structure of their language (Muter, 2005). This includes an awareness of speech segments within words (e.g. syllables, onset-rime, phonemes) and the ability to recognise, isolate and manipulate these speech segments (Anthony et al., 2002). The neural basis of phonological skills is lateralised to the left hemisphere, encompassing both posterior brain structures (superior temporal gyrus) and anterior brain structures (inferior frontal gyrus) (e.g. Burton, LoCasto, Krebs-Noble, & Gullapalli, 2005; Burton, Small, & Blumstein, 2000). Phonological skills are related to both reading and language abilities. Adequate understanding of these relationships is dependent upon knowledge of phonological skills themselves, including how they are measured, their development over time, structure, and relationship to other cognitive abilities.

1.1.2 Measurement.

Phonological tasks differ from one another in a number of ways. They can focus on different size sound segments; some tasks require identification or manipulation of syllables while others focus on phonemes. The position of the sound to be identified or manipulated can also vary; initial sounds, medial sounds and/or final sounds can be targeted in any given task. Finally, tasks differ in the type of stimuli they use; some tasks require instructions and stimuli to be provided orally, while others provide both oral and visual stimuli. Selection of the most appropriate task to administer should include consideration of 1) whether the task/s are appropriate for the age and stage of development of those being assessed and 2) what the purpose of the assessment is (e.g. clinical versus research purposes) (Neilson, 1999).

One common type of phonological task is that requiring an understanding of rhyme. This can include rhyme detection and production tasks, which require identifying or producing word/s that rhyme with a target word. There is debate as to whether rhyme tasks are qualitatively distinct and separable from other phonological tasks. Detailed discussion of this issue is provided in the structure of phonological skills section (1.1.4) of this review. Other phonological tasks are designed to measure a child's awareness of syllables or phonemes. These can include counting tasks (i.e. "tap out the number of sounds in the word cake"); identification tasks (i.e. "what sound does ball start/end with?"); segmentation tasks (i.e. what sounds do you hear in the word hat?"); blending tasks (i.e. what does /c/ /a/ /t/ make?"); and deletion tasks (e.g. what is star without the /s/?").

1.1.3 Development.

The developmental trajectory of phonological skills is contentious. One area of debate is whether phonological skills development begins with an awareness and understanding of large phonological units, such as words, onset-rime and syllables, and progresses until the smallest unit, the phoneme, is reached. According to this hypothesis, initially a child becomes aware that streams of speech are comprised of individual words. The child then begins to understand that each word is made up of a series of syllables, allowing them to master tasks such as syllable counting. Awareness and understanding of onset-rimes is thought to develop next, aiding children in accurate completion of rhyming tasks. Finally, around the age that Australian children enter school (Muter, Hulme, Snowling, & Stevenson, 2004), explicit awareness of phonemes begins to develop. At the level of the phoneme, the ability to blend phonemes generally develops prior to the ability to segment phonemes. More complex tasks of phoneme manipulation (e.g. phoneme deletion tasks) develop even later and are usually beyond the reach of children prior to the end of first grade (Adams, 1992; Farrall, 2012). This view proposes that developmental trajectory is relatively predictable, but there is individual

variation in the age each skill begins to develop and the time point at which each skill reaches maturity (Ziegler & Goswami, 2005).

Others argue against a standard progression from large to small linguistic units through development. This hypothesis is supported by evidence suggesting that awareness of syllables may pre-date awareness of words in infants. Babies aged 8-months old were found to segment words from speech streams using knowledge about syllables and their likelihood of following one another (Saffran, Aslin, & Newport, 1996). There is also evidence that awareness of phonemes can, under certain conditions, develop before awareness of onset-rimes (Duncan, Seymour, & Hill, 1997). One key idea underlying these findings is that external factors can influence the way phonological skills develop. Gombert's theory of metalinguistic development suggests that conscious awareness of different speech segments (i.e. metalinguistic awareness) only emerges when an external linguistic demand is placed upon earlier developed implicit knowledge (Gombert, 1992). An example of such a demand is learning to read, which explicitly draws a child's attention to letters and phonemes through teaching and activities. This teaching would support increased awareness and ability to manipulate phonemes, therefore supporting development of phoneme awareness prior to development of explicit awareness of other larger linguistic units such as onset-rimes (that are not the focus of early reading instruction). This is in contradiction to the 'large to small unit progression' theories of phonological development.

Phonological skill development is underpinned by the development of early language skills (e.g. speech perception and production) and development of implicit phonological knowledge (e.g. phonological representations). Development of language begins in early infancy. With regards to speech production, newborn babies produce pre-linguistic vocalisations such as crying. Canonical babbling (i.e. strings of alternative consonants and vowels) begins by approximately six months of age, where infants imitate the speech sounds

of those around them (Nathani, Ertmer, & Stark, 2006; Oller, 2000). By 12 months of age most children can produce a small number of single words that, while often pronounced imperfectly, correspond to words the child has been exposed to. Simultaneously, development of speech perception skills occurs. Infants can innately perceive and distinguish virtually every speech sound used in every language produced by humans. This ability declines over the first year of a child's life, as they simultaneously tune in to speech sounds used in their native language and begin to lose the ability to perceive speech sounds that are not used in speech around them (Hollich, 2010). Within this first year, babies also develop the ability to discriminate individual words within speech (around 5-6 months of age) and develop the ability to begin to store the sound patterns for words (around 8 months of age) (Owens, 2008).

Ongoing growth of a child's vocabulary and accurate perception of speech sounds supports the development of implicit phonological knowledge. This includes the development of phonological representations, which are sound based codes of words stored in long term memory (Claessen, Heath, Fletcher, Hogben, & Leitao, 2009; Sutherland & Gillon, 2007). Phonological representations initially comprise relatively large, holistic acoustic segments, which become more refined (Fowler, 1991; Walley, 1993). They eventually reach a level whereby they approximate the size of a phoneme. Development of well specified, accurate phonological representations is thought to support the development of strong phonological skills (Claessen et al., 2009; Gillon, 2015). This is illustrated in a Danish study by Elbro, Borstrom, and Petersen (1998) who showed that distinctness of phonological representations measured in the first year of school uniquely contributed to phonological skill performance and reading skills in Grade 2 children (after controlling for early phoneme awareness, articulation and productive vocabulary).

1.1.4 Structure.

The structure of phonological skills is not yet well defined; it is unclear whether these abilities are best represented as a single cognitive construct (Adams, 1992; Anthony & Francis, 2005; Schatschneider, Francis, Foorman, Fletcher, & Mehta, 1999; Stahl & Murray, 1994) or are representative of distinct and separable cognitive constructs (Høien, Lundberg, Stanovich, & Bjaalid, 1995; Oakhill & Kyle, 2000; Ramus, Marshall, Rosen, & van der Lely, 2013; Wagner, Torgesen, & Rashotte, 1994; Yopp, 1988). Research has suggested several ways that phonological abilities may be separable – 1) in terms of the size of phonological units being manipulated (e.g. syllable versus phonemes), 2) in terms of separation between rhyme skills and other types of phonological skills, or 3) in terms of cognitive load. Cognitive load is a multidimensional construct which represents the load that performing a particular task imposes on an individual's cognitive system (Paas & Van Merriënboer, 1994). The following section provides examples of each of these proposals in turn.

Drawing upon knowledge of the development of phonological skills as discussed earlier, the ability to recognise, isolate and manipulate phonemes, syllables and rhymes have been found to form part of a single phonological ability construct. Anthony et al. (2002) used confirmatory factor analysis to show that syllable, rhyme, onset-rime and phoneme tasks were best represented by a single underlying phonological ability in preschoolers. These results were consistent with several earlier studies suggesting that phonological skills are a single cognitive ability which can be measured using a variety of rhyme, syllable and phoneme based tasks (Anthony & Lonigan, 2004; Stahl & Murray, 1994; Wagner & Torgesen, 1987). In contrast, additional findings in the literature suggest phonological skills are best represented as distinct cognitive abilities. Using a cohort of Norwegian preschoolers, Høien et al. (1995) identified three separate components using a principal component

analysis: a rhyme factor, a syllable factor and a phoneme factor. They also showed that each factor was a separate and independent predictor of early word decoding ability.

Rhyme skills may also form a cognitive construct that is qualitatively distinct from other phonological skills. In structural analyses, rhyme tasks often (though not always) load onto a separate factor to other phonological tasks (Beach & Young, 1997; Runge & Watkins, 2006; Yopp, 1988). There is evidence that this is more likely to occur in primary school aged children than pre-school aged children (Anthony & Lonigan, 2004). The age-related differences may arise because rhyme ability separates from other, more advanced, phonological skills as a part of typical development. Alternatively, ceiling effects on phonological tasks can also impact the observed relationship between rhyme and other phonological constructs, and may therefore be responsible for the observed separation (see Anthony & Lonigan, 2004 for further discussion of these alternative hypotheses). Rhyming abilities and other phonological skills have also been shown to be differentially related to literacy abilities. Muter et al (1998) found that rhyming skills were related to vocabulary and letter recognition but not to reading (word recognition) skills. In contrast, other phoneme level skills were related to word recognition in their sample of kindergarten children. Rhyme and phoneme skills were also found to be weakly correlated. These results suggested that rhyme and other phonological skills represent separate constructs.

The structure of phonological skills can also be examined from a cognitive load perspective. Wolff and Gustafsson (2015) recently emphasised the importance of considering the relationship between cognitive load and the structure of phonological skills. They found that cognitive load was more important than linguistic factors in accounting for the relationships among phonological tasks. Understanding the nature of the relationship between cognitive load and phonological skills is important and can be considered in the context of age-related changes in phonological skills. The age-related developmental trajectory of

phonological skills demonstrates that there are early developing phonological skills, such as phoneme identification and blending, which are less cognitively demanding than other, later developing phonological skills such as phoneme deletion (Farrall, 2012). The differences in how cognitively taxing or demanding these tasks are relate to differences in their cognitive load. Examining how cognitive load relates to these early- and later- developing phonological skills can help us understand how these skills are related to other cognitive abilities (e.g. short-term memory and working memory) as well as academic abilities (e.g. reading). It may be that earlier developing, lower-level phonological skills are related to reading abilities in a different way than later developing, higher-level phonological skills. There has been limited research comprehensively examining this topic. As such, the relationship between cognitive load and phonological skills was selected as a primary focus of this literature review and the research presented in Chapter 3.

Perhaps the most comprehensive example of how phonological skills and cognitive load are related comes from an early study (Yopp, 1988), which aimed to examine the validity and reliability of a range of phonological tests in pre-school children. Using factor analysis, Yopp identified two separate (but strongly correlated) phonological factors, onto which ten phonological tasks loaded. The ‘lower-level’ factor included tasks that required the participant to perform one cognitive operation (e.g. phoneme blending, segmentation, phoneme counting and sound isolation tasks). The ‘higher-level’ factor included two phoneme and/or syllable deletion tasks that required participants to isolate and delete a sound in a word before recalling and blending the remaining sounds to produce a new word. These required higher levels of attention and cognitive effort. Cognitive load would therefore be greater for these tasks than for the lower-level phonological tasks.

A more recent study also emphasised the importance of considering cognitive load in the context of phonological skills (Ramus et al., 2013). The study used two analytic

techniques (principle components analysis and confirmatory factor analysis) to examine the structure of phonological skills in the context of a broader study investigating phonological deficits in reading disability and specific language impairment. Like Yopp (1988), Ramus et al. (2013) identified distinct higher- and lower-level phonological constructs, with more cognitively complex and demanding tasks (i.e. those with higher levels of cognitive load) loading onto the higher-level factor. Examination of the higher-level phonological factor in the Ramus et al. study showed that it was multifaceted. Tasks loading onto the higher-level factor included a digit span task (both digit span forward and backward components, and which are generally considered to measure short-term memory and/or working memory; see section 1.2.1 for a more detailed discussion), a rhyme task, a rapid digit naming task, and a spoonerism task (with two types of items – those requiring exchanging the sounds of words, e.g. “sad cat” = “cad sat”; and those requiring replacement of an initial phoneme with a new phoneme, e.g. cot with a /g/ = “got”). Given heterogeneity of the tasks included, it is difficult to define the main skill underpinning this factor.

There were three key methodological differences which may help explain differences in findings between the Yopp and Ramus et al. studies. Firstly, Ramus et al. included several tasks thought to correspond to phonological representations (e.g. articulation and non-word repetition tasks). These tasks loaded onto the lower-level construct, which was ultimately defined as a phonological representation construct rather than phonological skills per se. The Ramus et al. (2013) study also included a smaller number of phonological tasks in comparison to Yopp (1988), in part due to differences in the primary aims of each study. Inclusion of a broad variety of phonological tasks which differ in their cognitive load is important to maximise the likelihood of observing separation between higher and lower-level cognitive skills in structural analyses. Lastly, the nature of participants varied. Ramus (2013) included children with neurodevelopmental disorders (reading disability and specific

language impairment) and matched controls, while Yopp (1988) included children recruited directly from primary schools without consideration of whether reading, language or other developmental conditions were present. There is evidence to suggest that the relationships between cognitive constructs can differ between neurotypical individuals and those with developmental disorders (e.g. Giofre & Cornoldi, 2015; Numminen et al., 2000). Taken together, methodological differences mean that findings in the two studies cannot be directly compared. Both studies, however, provide evidence that phonological skills may represent distinct and separable constructs in children.

In contrast to the abovementioned studies, others have utilised exploratory factor analysis to show that phonological skills are best conceptualised as a single cognitive construct (e.g. Runge & Watkins, 2006; Stahl & Murray, 1994). Most of these studies measure phonological skills in pre-school children or those who are in their first years of schooling, and they typically have not investigated the structure of phonological skills in a learning disability population. No identified study focused explicitly and comprehensively on the relationship between phonological skills and cognitive load in a broad range of primary school aged children with learning disabilities.

The limited amount of research described in the literature, combined with inconsistent results in studies that have been published, emphasises the need for further investigation into the relationship between cognitive load and phonological skills. Examining how cognitive load relates to lower-level (early developing) and high level (later developing) phonological skills can help us understand how these skills are related to other academic abilities, such as reading. Earlier developing phonological skills may be related to reading abilities in a different way than later developing, higher-level phonological skills. Furthermore, lower-level and higher-level phonological skills may be differentially impaired in children with

reading disabilities and other neurodevelopmental disorders, such as specific language impairment.

One way to progress our understanding of the relationship between cognitive load and phonological skills is to examine the cognitive skills, such as short-term memory and working memory, which contribute to performance on phonological tasks. Short-term memory and working memory have been shown to be related to phonological skills but also differ in their respective levels of cognitive load. The following section of this review aimed to provide a general introduction to short-term memory and working memory (including their development and how they are usually measured) before examining the relationship between the two. Consideration as to how short-term memory and working memory are related to phonological skills is also discussed.

1.2 Short-Term Memory and Working Memory

1.2.1 Definition and measurement.

Short-term memory is defined as the passive storage of small amounts of verbal or non-verbal information over a short period of time (seconds) to reproduce the information in a sequential or untransformed way (Swanson, Zheng, & Jerman, 2009). Short-term memory is most commonly measured through administration of digit, block, letter, word or non-word span tasks. In both verbal and non-verbal (visuospatial) domains, these tasks involve presentation of lists or sequences of stimuli, which increase in length across trials. The individual must repeat back the stimuli sequentially and untransformed.

Working memory is defined as the ability to attend to and maintain verbal information that has been just experienced (or has been retrieved from long term memory) while manipulation of that, or other, information occurs (D'Esposito, 2008; S. Gathercole & Alloway, 2006). The working memory system can maintain and manipulate verbal, visual or spatial information, although verbal working memory will be the focus of this review because

verbal working memory abilities have been found to be related to phonological skills in neuroimaging and empirical research studies (see section 1.2.4 for a detailed discussion).

Working memory tasks are more cognitively demanding (and therefore place higher levels of cognitive load on an individual) than short-term memory tasks. In contrast to the simple repetition required in short-term memory tasks, working memory tasks require encoding, maintenance and manipulation of information to produce an appropriate response. For example, the digit span backwards task (a commonly used working memory measure) requires participants to attend to a string of digits, hold those digits in mind, and manipulate the digits so that they can be recalled backwards. Other types of working memory tasks include dual processing tasks such as complex memory span tasks, item recognition tasks and n-back tasks.

Perhaps the most influential and well-researched model describing the short-term memory and working memory networks is Baddeley's model (Baddeley, 2000). According to this model, short-term memory performance is supported by at least two individual systems - the phonological loop and the visuospatial sketchpad (Baddeley & Hitch, 1974). The phonological loop is responsible for the temporary storage of verbal information. It consists of two separable elements; 1) a short-term store, which holds verbal information in phonological form, and 2) a sub vocal rehearsal process. The rehearsal process allows phonological information to be sub vocally rehearsed to prevent decay and facilitates the "recoding" of visually encoded information into phonological form so as it can be stored in the phonological loop (e.g. Baddeley, 2000, 2002; Baddeley & Hitch, 1974; Salame & Baddeley, 1982). The visuospatial sketchpad is proposed to be responsible for temporary storage of visuospatial information. It is also limited in capacity and is thought to include a rehearsal process along with its storage capabilities (Logie, 1989). Baddeley's working memory model also incorporates a central executive (thought to act as a control system with

limited attentional capacity), and the episodic buffer (thought to be involved in integrating information from long term memory and the STM systems; Baddeley, 2002).

The structure of short-term memory and working memory (as described by Baddeley) is supported by empirical research. Verbal and visuospatial short-term memory tasks generally load onto separate factors in structural analyses, supporting the separation of the phonological loop and visuospatial sketchpad. Similarly, working memory tasks typically load onto a separate factor to both verbal and visuospatial short-term memory tasks, which provides support for a central executive or higher level component to the working memory system (Alloway, Gathercole, & Pickering, 2006).

1.2.2 Development of short-term memory and working memory.

Visual and verbal short-term memory are in place from a young age. Children as young as six months of age can imitate speech sounds, suggesting that once they hear these sounds they can hold them in short-term memory prior to reproducing them. By age three to four years sub-vocal rehearsal processes are available for use and a typically developing child can generally hold up to three digits or letters in verbal short-term memory (Dehn, 2008). This verbal span increases in a linear manner until reaching approximately seven digits at the age of 13-15 years (Conklin, Luciana, Hooper, & Yarger, 2007) and the remains stable at least until 50 years of age (Siegel, 1994).

Research investigating the development of visuospatial short-term memory is comparatively scarce but consistently shows a steady increase in span through childhood (Orsini et al., 1987; Pickering, Gathercole, Hall, & Lloyd, 2001; Pickering, Gathercole, & Peaker, 1998). One early study found that mean block span scores grew from four blocks at seven years old to approximately five-and-a-half blocks by 15 years of age (Isaacs & Vargha-Khadem, 1989). There is additional evidence that visual and spatial components of visuospatial short-term memory may develop at different rates (e.g. Logie & Pearson, 1997).

The general structure of working memory (i.e. two short-term memory stores and a higher level component responsible for manipulation of information) seems to be in place from a fairly young age (Alloway et al., 2006; S. Gathercole, S. J. Pickering, B. Ambridge, & H. Wearing, 2004). However, mean level of performance on tasks of working memory increases steadily throughout childhood and into adolescence (Alloway et al., 2006; S. Gathercole et al., 2004; Siegel, 1994). The developmental growth of working memory is likely to be impacted by a number of factors that interact with each other, including improved processing speed (for discussion see Hitch, 2006) and increased in storage capacity (e.g. Bayliss, Jarrold, Baddeley, Gunn, & Leigh, 2005; Bayliss, Jarrold, Gunn, & Baddeley, 2003), controlled attention, use of strategies (Palmer, 2000) and general operating efficiency (Case, Kurland, & Goldberg, 1982).

1.2.3 The relationship between short-term memory and working memory.

Mirroring the debate within the phonological skills literature, questions remain as to how to best conceptualise the relationship between short-term memory and working memory. Empirically, short-term memory and working memory tasks are significantly correlated with one another (e.g. S. Gathercole, Alloway, Willis, & Adams, 2006; S. E. Gathercole, S. J. Pickering, B. Ambridge, & H. Wearing, 2004). Factor analytic and structural equation modelling studies suggest that short-term memory and working memory are separate but related cognitive constructs in both adults and children (e.g. Alloway et al., 2006; Engle, Tuholski, Laughlin, & Conway, 1999; S. E. Gathercole & Pickering, 2000; S. E. Gathercole et al., 2004; Kail & Hall, 2001; Kane et al., 2004; Swanson, 2008). Neuroimaging literature also provides further evidence of a relationship between the two. Digit span forward and backward tasks, for example, have been found to rely on a largely overlapping neural network that includes the dorsolateral prefrontal cortex, the inferior parietal lobule and the anterior cingulate cortex (Gerton et al., 2004).

Knowledge of the precise nature of this relationship is incomplete however, with research in this area complicated by several issues. Firstly, it can be difficult to compare studies across age groups because the strength of the relationship between short-term memory and working memory may change throughout development. In one identified study, correlations between verbal short-term memory and verbal working memory tasks increased with age, from 0.73 in the youngest group (aged six-seven) to over 0.90 in the two older groups (10-12 and 13-15 years old). The results are suggestive of a stronger relationship between verbal short-term memory and verbal working memory in older children than their younger counterparts. The authors suggest that this may be a result of increased processing efficiency, whereby working memory tasks are less cognitively demanding (and therefore more like short-term memory tasks) in older children (S. E. Gathercole et al., 2004). Alloway et al. (2006) found that correlations between visuospatial short-term memory tasks and working memory tasks were stronger in young children suggesting that, when performing visuospatial short-term memory tasks, these children drew more heavily on working memory than older children do. Further research is needed, however, to clarify how the strength of relationships between short-term memory and working memory change across development.

Understanding the relationship between short-term memory and working memory is further hindered by the multifactorial nature of tasks and the set of multiple underlying skills that are needed to complete them. The developmental trajectories of short-term memory, working memory and other related skills, such as attention, follow different developmental pathways. At each age a child will have a different level of capacity in each skill. A task designed to measure a specific cognitive ability will therefore tap into each required underlying skill in a different way depending on the age and development of the individual completing it (Engle et al., 1999). This means that what may be considered a working memory task for one individual can be a short-term memory task for another. For example, St

Clair-Thompson (2010) conducted confirmatory factor analysis with separate groups of adults and primary school aged children. She found that in adults, digit span forwards, backwards and word span loaded onto a short-term memory factor. In contrast, in children, digit span backwards was more strongly associated with complex span tasks, loading onto a working memory factor. It was proposed that the digit span backwards task was more cognitively demanding and therefore required higher levels of cognitive load for children than for adults. These results are consistent with several earlier studies investigating whether backward span tasks are best conceptualised as a short-term memory or working memory task. In children, these tasks generally load onto a working memory factor (Alloway et al., 2006; Alloway, Gathercole, Willis, & Adams, 2004; S. E. Gathercole et al., 2004)

Despite the issues listed above, several hypothesised models have aimed to describe the relationship between short-term memory and working memory. The most influential conceptualise short-term memory as a component of working memory (e.g. Baddeley, 2000; Colom, Shih, Flores-Mendoza, & Quiroga, 2006; Cowan, 2008; Engle, Cantor, & Carullo, 1992; Engle et al., 1999). They propose that working memory is a multicomponent system consisting of both a short-term memory component/s and a higher level control/attentional system.

In summary, evidence suggests that short-term memory and working memory are separable but highly related cognitive abilities in childhood. Understanding the specific nature of the relationship between the two constructs is challenging because 1) The strength of the relationship changes across age and 2) The cognitive skill/s measured by a task can vary depending on age and level of cognitive development. This is related to differences in the developmental trajectories of cognitive skills (e.g. short-term memory and working memory), whereby what may be considered a short-term memory task for one individual could be a working memory task for another (who is younger or has less mature cognitive

skills). Despite these challenges, it is important to acknowledge the separation of short-term memory and working memory as it raises the possibility that the two constructs may be differentially associated with other academic and cognitive skills, including phonological skills.

1.2.4 The relationship between short-term memory, working memory and phonological skills.

Knowledge of how phonological skills, short-term memory and working memory are related is important to understand how cognitive load relates to the structure of phonological skills. The discussion in this section focused primarily on how short-term memory and working memory have been shown to be related to phonological skills in research to date, as well as providing a brief summary of hypotheses put forward to explain the aetiology of these relationships. Following this will be a discussion regarding issues that contribute to the complexity in understanding findings within this area.

With regards to short-term memory, the majority of relevant research has investigated the relationship between verbal short-term memory tasks and phonological skills. One hypothesis suggests that verbal short-term memory and phonological tasks tap a common phonological resource, and performance on these tasks are limited by the quality of a child's underlying phonological representations (Bowey, 1996). Some evidence supports this hypothesis by finding that performance on short-term memory and phonological tasks can be explained by a single underlying latent construct (Bowey, 1996; S. E. Gathercole & Willis, 1991; Wagner, Torgesen, Laughon, Simmons, & Rashotte, 1993). Yet there is also contrasting evidence that short-term memory and phonological tasks load onto separate cognitive constructs in structural analyses (Alloway et al., 2004; Dufva, Niemi, & Voeten, 2001), suggesting that short-term memory and phonological skills are distinct cognitive abilities. Further supporting this hypothesis, short-term memory and phonological skills have

been found to differentially contribute to reading skills (Nithart, Demont, Metz-Lutz, Majerus, & Poncelet, 2011; Parrila, Kirby, & McQuarrie, 2004).

There is also evidence of a relationship between working memory and phonological skills. Burton et al. (2000) examined phonological skills using different speech discrimination tasks with different working memory demands (i.e. requiring either single or multi-step phonological judgements) in an fMRI study. They found that the superior-temporal regions within the left hemisphere (and to a lesser degree in the right hemisphere) were activated during each type of task, indicating overlap between the neural networks underlying working memory and phonological skills. Frontal activation, however, was only evident on tasks with high working memory demands (i.e. those with multi-step phonological judgement requirements). Their results demonstrate that the neural networks recruited for these tasks can be differentiated by the working memory demands of the phonological task being completed.

In the context of the research described in this section, several questions remain. Firstly, does the multifactorial nature of short-term memory, working memory and phonological tasks underlie the relationship between these constructs? Short-term memory, working memory and phonological tasks do not necessarily measure a single cognitive ability. Instead, other underlying skills, such as attention and processing speed, are likely to contribute to performance. Overlap between these underlying skills could therefore be mediating the relationship between short-term memory, working memory and phonological skills. An additional question is how are higher- and lower-level phonological skills related to short-term memory and working memory? The research discussed in this review suggests that higher- and lower-level phonological skills may be differentially related to short-term memory and working memory, with working memory more strongly related to the higher-level phonological skill construct than lower-level phonological skills. Working memory tasks and higher-level phonological tasks require significantly greater cognitive effort for

successful completion, and both are therefore considered to recruit greater resources for information processing and assume greater cognitive load.

1.2.5 Cognitive load and phonological skills summary.

Understanding how cognitive load is related to the structure of phonological skills represents a significant gap within the literature to date. Examining how cognitive load relates to early developing (lower-level) and later developing (higher-level) phonological skills can help us understand how these skills are related to other cognitive abilities, such as short-term memory and working memory, along with academic abilities such as reading. Of the few studies which have addressed this topic, two in particular suggest that phonological skills are best represented by distinct constructs that are differentiated by cognitive load (high versus low). The following segment of this review continues to examine phonological skills in the context of learning disabilities, focusing on children with reading disability and specific language impairment. It provides definitions and descriptions of both reading disability and specific language impairment along with a summary of how phonological skills are impacted in these disorders.

1.3 Phonological Skills in Reading Disability and Specific Language Impairment

1.3.1 Definition and aetiology of reading disability and specific language impairment.

Reading disability and specific language impairment are neurodevelopmental disorders which are each thought to occur in approximately 5-10% of children (Shaywitz, Shaywitz, Fletcher, & Escobar, 1990; Tomblin et al., 1997). Diagnosis of a reading disability is made when a child has a significant, unexpected difficulty learning to decode words (Nithart et al., 2009), which results in slow, effortful and often inaccurate reading. Diagnosis of a reading disability is made following the onset of formalised reading instruction. Reading

disabilities are often associated with co-occurring academic and non-academic impairments (see Chapter 2 of this review for a detailed description). Impaired phonological skills are frequently found in children with reading disabilities as compared to their same aged peers (Fletcher et al., 1994; Kudo, Lussier, & Swanson, 2015; Landerl, Fussenegger, Moll, & Willburger, 2009; Melby-Lervag, Lyster, & Hulme, 2012; Swan & Goswami, 1997). These impairments are evident across numerous phonological tasks, including phoneme deletion tasks (Fletcher et al., 1994; Landerl et al., 2009; Larkin & Snowling, 2008), spoonerisms tasks (Ramus et al., 2013), segmentation tasks (Everatt, Weeks, & Brooks, 2007; Swan & Goswami, 1997) and rhyme tasks (Swan & Goswami, 1997).

Phonological impairments are hypothesised to be a primary underlying cause of reading difficulties in most children with reading disabilities (Ramus, Pidgeon, & Frith, 2003), but these alone are not necessary *or* sufficient to cause poor reading. Instead, reading disability is likely the result of interactions between a number of cognitive risk and protective factors. This is hypothesised to include imprecise or deficient phonological representations. Deficient representations can negatively impact on a child's ability to acquire adequate phonological skills, which in turn leads to difficulties learning to read accurately and fluently (Fowler, 1991; Peterson & Pennington, 2012; Stanovich & Siegel, 1994; Sutherland & Gillon, 2007). Poor phonological representations have been found in children with reading disabilities (e.g. Boada & Pennington, 2006). Alternatively, phonological representations may be intact in children with reading disabilities, but they may have difficulty accessing these representations efficiently (Ramus & Szenkovits, 2008).

Specific language impairment is diagnosed in those who demonstrate impaired oral language skills that cannot be attributed to other factors such as hearing loss or neurological damage (Leonard, 2014). The nature of the language deficits demonstrated by children with specific language impairment is variable. While one child may perform more poorly on

expressive language tasks than on receptive language tasks, another may show the reverse pattern of impairments. Others can exhibit comparable deficits in expressive and receptive language skills. Impairments can also span several different components of language, including grammar, syntax, morphology and phonological skills (Bishop & Snowling, 2004; Briscoe, Bishop, & Norbury, 2001; Joanisse & Seidenberg, 1998; Snowling, Bishop, & Stothard, 2000).

The aetiology of specific language impairment is unclear. Hypotheses typically suggest that either linguistic (e.g. syntactic or grammatical impairments) or non-linguistic (e.g. rapid temporal processing) deficits are responsible for the language impairments seen in specific language impairment. In the context of this review, the most relevant hypothesis is the phonological hypothesis, which states that poor phonological representations are the foundation of impaired language acquisition in children with specific language impairment (Corriveau, Pasquini, & Goswami, 2007). Phonological representations are important in development of aspects of language, including syntax, morphology and vocabulary (Chiat, 2001; Gray, 2005; Joanisse & Seidenberg, 1998). Therefore, poor phonological representations may impede development of language. The phonological hypothesis represents an overlap between reading disability and specific language impairment, with both hypothesised to be caused by underlying phonological impairments.

1.3.2 The nature of phonological skill impairments in reading disability and specific language impairment.

Despite extensive research examining phonological skills in reading disability and specific language impairment, questions remain as to whether the prevalence, severity and pattern of phonological skill impairments are comparable between the two disorders. This is partly due to inconsistencies in the definitions of reading disability and specific language impairment, and the quantitative criteria (i.e. cut-off scores) used to allocate children to

reading or language impaired groups can also be variable. Differences are also evident in the type of reading or language tasks used to identify children with reading disability or specific language impairment. Classification into a specific language impairment group can be based on impairments in a both expressive *and* receptive language scores (Nithart et al., 2009) or impairments in either expressive *or* receptive language scores (Conti-Ramsden & Durkin, 2007). Reading tests (Eisenmajer, Ross, & Pratt, 2005; Ramus et al., 2013) and phonological tasks (Nithart et al., 2009; Rispens & Been, 2007) also vary between studies. Because the structure of phonological skills remains unclear, use of different phonological tasks leads to questions regarding whether the selected tasks are measuring the same construct or not. Another major issue is that studies have typically measured reading *or* language abilities in their cohorts rather than measuring both abilities. Given that reading disability and specific language impairment frequently co-occur, this can create heterogeneous groups of children, making it difficult to ascertain whether the observed phonological deficits are specific to reading disability or specific language impairment or are present in both.

A number of studies have overcome this issue by measuring a child's reading and language skills before allocating them according to their abilities in both domains. This gives three groups of children with language and/or reading impairments – specific language impairment only (SLI-only), reading disability only (RD-only), and reading disability *and* specific language impairment (RD+SLI). A summary of study design and results for six key studies utilising this methodology is provided in Table 1.1. Examination of the design and methodology of the six studies identified some of the issues discussed previously.

Differences between the studies are evident in the age of participants; diagnostic criterion for specific language impairment and reading disability; cut off scores to define phonological skill impairments; and tasks used to measure reading, language and phonological skills.

When comparing these six key studies, groups of children with a reading disability (i.e. RD-only and RD+SLI groups) demonstrate impaired phonological skills in all but one study (Eisenmajer et al., 2005). Poor phonological skills in groups of children with a reading disability is consistent with a large body of past research findings. The phonological impairments are also evident in children with reading disability *without* specific language impairment, suggesting that a reading disability in and of itself is associated with poor phonological skills. The findings in Eisenmajer et al. (2005) were somewhat difficult to interpret. The RD-only group in this study did not show impaired phonological skills as compared to a ‘no specific learning disability’ control group. This control group had been referred to a learning difficulties clinic, but upon assessment did not meet criteria for any other clinical group. They demonstrated age appropriate scores on tasks of non-verbal intellectual functioning, phonological skills, language, reading and math. However, the RD-only group’s phonological skills also did not differ from the RD+SLI group or another ‘general learning disability’ group, both whose phonological skills were clearly impaired. The general learning disability group also demonstrated impaired language and reading skills, with non-verbal intellectual functioning in the low average range or below (i.e. 80 or less). The RD-only group’s unexpected result was most likely due to analytic issues (e.g. Small sample sizes leading to lack of statistical power), leading to the authors themselves to suggest that “the findings are inconclusive” (p.1113).

In contrast to the generally consistent findings of the reading disability groups, results for the SLI-only groups are mixed. Three of the six key studies found impaired phonological skills in the SLI-only group (Fraser, Goswami, & Conti-Ramsden, 2010; Ramus et al., 2013; Snowling et al., 2019), and two found no impairments in the SLI-only group compared to controls (Eisenmajer et al., 2005; G. McArthur & Castles, 2013). In the remaining study children with SLI-only had mild phonological skill difficulties in Kindergarten (first year of

school) and Grade 2, but not in Grade 4 and Year 8 (Catts, Adlof, Hogan, & Weismer, 2005). Determining the reasons for the inconsistent results is challenging. One potential contributor is greater variability in the phonological skills of children with SLI-only as compared to those with RD-only. In Ramus et al. (2013), nearly 87% (18 of 21) of children in the RD-only group met criteria for impaired phonological skills, while in the SLI-only group, approximately 60% (8 of 13) showed impairment. Ramus et al. did not examine the underlying cause of the increased variability in the SLI-only group. The heterogeneous nature of children with specific language impairment might contribute; as discussed previously, definitions of specific language impairment vary and children can be classified using variable criterion. While some children with specific language impairment may have phonological impairments, for others, non-phonological language or cognitive impairments could lead to poor language skills. Groups of children with specific language impairment would then demonstrate increased variability in phonological skills (as was identified in Ramus et al.), which may underpin the inconsistencies in SLI-only groups in the six key studies summarised in Table 1.

Table 1.1

A summary of key research studies comparing phonological skills in children with RD-only, SLI-only and RD+SLI

	Age range	N (group)	Inclusion criteria RD and SLI	Phonological tests and criteria for impairment	Phonological skills impaired?			Magnitude of phonological skills deficit
					RD-only	SLI-only	RD+SLI	
Snowling, 2019¹	T1=3.5 years T2=4.5 years T3- T5=5.5 -8 years	<u>RD only=21</u> <u>SLI only=38</u> <u>RD+SLI=29</u>	RD: <u>Composite score</u> (average of age standardised <u>SWRT and WIAT-II</u>) <u>Z<-1.5</u> SLI: <u>Composite score</u> (average on <u>CELF-4 expressive vocabulary & formulated sentences;</u> <u>TROG-2</u>) <u>Z=<-1</u>	T2 - Syllable and alliteration matching tasks. T2&T3 – Phoneme isolation task T3-T5 – Phoneme deletion task (YARC) Groupwise comparison with an	✓	✓	✓	<u>T2&T3:</u> (RD only = SLI only = RD+SLI) < Control <u>T4&T5:</u> RD only = SLI only RD only = RD+SLI RD+SLI<SLI only

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				age-matched control group				
Ramus, 2013	Clinical groups: 8-12 years Control group: 5-12 years	<u>13 (SLI- only); 21 (RD- only); 30 (RD+SLI)</u>	<u>RD or SLI</u> diagnosis; attendance at a special school/unit for children with RD or SLI <i>and</i> $Z \leq -1.5$ on a single word reading subtest (RD; WORD) or on one or more of the language tests administered (SLI; TROG-2, CELF-3 Sentence repetition, Test of word finding). *All children minimum score of 80 and average combined	Composite phonological skills score calculated from PhAB subtests (rhyme; spoonerisms; rapid digit naming) and a digit span task. Individual children considered impaired if scored more than 1.5SD below the mean Z score of the control group. Also conducted groupwise	✓	✓	✓	$RD+SLI \leq (RD-only = SLI-only)$

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			score of 85 ($Z \geq -1$) on two nonverbal cognition tests.	comparison with (non age-matched) control group.				
Eisenmajer 2005	7-12 years	25 (SLI- only); 21 (RD- only); 57 (RD+ SLI)	<u>RD</u> : WIAT Reading $Z < -1$ <u>SLI</u> : Total Language Score on the CELF $Z < -1$ * Both groups PIQ score > 80 .	Groupwise comparison with a ‘no learning disability’ group using SPAT total score.	X	X	✓	RD+SLI \leq SLI-only RD-only = SLI-only RD-only = RD+SLI
Fraser, 2010	9-11 years	16 (SLI- only); 14 (RD- only); 21 (RD+SLI)	<u>RD</u> : Std reading score < 85 on one or more of: BAS-II single word reading ability, TOWRE sight word efficiency/ phonological coding efficiency. <u>SLI</u> : $Z < 85$ on at least two CELF-III subtests	Phoneme deletion; rhyme oddity; rhyme fluency Groupwise comparison with chronological age matched control group.	✓	✓	✓	RD+SLI \leq (SLI-only = RD-only) ²

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			(formulated sentences, sentence assembly, concepts and directions, semantic relations).					
Catts, 2005	Kinder ³ – Year 8	<u>43 (SLI-only); 21 (RD-only); 18 (RD+SLI)</u>	<p><u>RD</u>: Classified using Grade 4 data. At least 1SD below the mean on a composite word recognition measure and actual word recognition score more than 1SD below predicted word recognition score.</p> <p><u>SLI</u>: Classified using Kindergarten data. $Z \leq -1.25$ on at least two of five</p>	<p>Syllable/Phoneme deletion (Kinder, grade 2, grade 4)</p> <p>Phoneme deletion (Year 8)</p> <p>Groupwise comparison with control group who had normal kindergarten language scores and normal reading achievement in Grade 4.</p>	✓	✓ ⁴	✓	<p><u>Kinder</u>: (RD-only=SLI-only=RD+SLI) < Control</p> <p><u>Grade 2</u>: (RD-only=RD+SLI) < SLI-only < Control</p>
					✓	X	✓	<p><u>Grade 4 and Year 8</u>: (RD-only = RD+SLI) < (SLI-only = control)</p>

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			language composites (vocabulary, grammar, narration, receptive language, expressive language), nonverbal IQ >85					
McArther & Castles (2013)	7-12 years	<u>4-5 (SLI-only); 41-43 (RD-only); 17-19 (RD+SLI)⁵</u>	<p><u>RD</u>: Z<-1 on at least one of: letter-sound reading; whole word reading.</p> <p><u>SLI</u>: Z<-1 on at least one of: receptive language, recalling sentences⁶</p> <p>* Both groups nonverbal IQ > 85.</p>	<p>PhAB alliteration with pictures</p> <p>RD-only and RD+SLI: Groupwise comparison with 1) an age related normative mean score and 2) the other group's mean.</p> <p>SLI-only: Individual scores compared to 1) age based normative mean score and 2)</p>	✓	X	✓	<p>RD < SLI</p> <p>RD+SLI<RD⁷</p>

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				the mean scores of RD-only and RD+SLI. If most individuals significantly different, SLI-group deemed to differ from that mean.				
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BAS-II=British abilities scale II; CELF=Clinical evaluation of language fundamentals; PhAB=Phonological Assessment Battery, PIQ=Performance IQ; TOWRE=Test of word reading efficiency; TROG-2=Test for the reception of grammar 2; WIAT=Wechsler individual achievement test; WORD=Wechsler objective reading dimensions; York Assessment of Reading for Comprehension (YARC)

- ¹ Longitudinal study with five time points (T1-T5); SLI was termed developmental language disorder (DLD) in this paper. SLI is used here for comparison purposes.
- ² RD+SLI group performed more poorly than RD-only and SLI-only groups on phoneme deletion and rhyme oddity tasks, but not the rhyme fluency task.
- ³ Kindergarten refers to the first year of formal schooling.
- ⁴ While SLI-only group performed more poorly than the control group in both kindergarten and Grade 2, the group’s standard scores on the syllable/phoneme deletion task were approximately 90 (kindergarten) and 100 (Grade 2).

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⁵. 73 children were allocated to RD-only, SLI-only and RD+SLI groups. Group n varied depending on which of four different analyses was examined.

⁶. Different combinations of reading and language scores were used to create groups for four different analyses – 1) receptive-expressive language+ letter-sound reading, 2) receptive-expressive language+whole-word reading, 3) receptive language+letter-sound reading, and 4) receptive language+whole-word reading‘

⁷. RD+SLI group performed more poorly than RD-only group on two of the four analyses (both receptive-expressive language group analyses).

Within the six studies summarised in Table 1.1, the magnitude of phonological impairments was generally larger in the RD+SLI group than in either the SLI-only (Catts et al., 2005; Eisenmajer et al., 2005; Fraser et al., 2010; G. McArthur & Castles, 2013; Ramus et al., 2013; Snowling et al., 2019), and RD-only (Fraser et al., 2010; G. McArthur & Castles, 2013; Ramus et al., 2013) groups with three exceptions (Catts et al., 2005; Eisenmajer et al., 2005; Snowling et al., 2019). These three studies did not show statistically significant differences when comparing RD+SLI and RD-only, but did show a trend towards poorer performance by the RD+SLI group. Comparison of the magnitude of impairments in the single deficit groups is complicated by the equivocal phonological skill results in children with SLI-only. Of the four studies showing impaired phonological skills in children with SLI-only, three found no difference in magnitude between RD-only and SLI-only groups (Fraser et al., 2010; Ramus et al., 2013; Snowling et al., 2019). In contrast, the fourth identified greater magnitude of phonological impairments in the RD-only group in Grade 2 children, but no difference in kindergarten children (Catts et al., 2005).

The pattern of phonological impairments with regards to higher- and lower-level phonological skills is unknown, in part because of limited research investigating the relationship between cognitive load and phonological skills. There is also continued debate as to whether distinct higher- and lower-level phonological skills can even be identified, though several studies now support this proposition (Lee, Ross, Nadebaum, & Testa, under review; Ramus et al., 2013; Yopp, 1988). Of the six key studies, just one includes discussion of higher and lower-level phonological skills, finding different patterns of performance between the RD-only and SLI-only groups. The SLI-only group were equally impaired on higher and lower-level phonological skill composite scores, while the RD-only group performed more poorly on the higher-level than the lower-level composite score (Ramus et al., 2013). Given that Ramus et al. utilised composite scores to investigate pattern of

performance, it was not clear whether individual phonological tasks are differentially impacted in reading disability and specific language impairment.

In summary, results from six key studies showed that children with a reading disability demonstrate impaired phonological skills regardless of whether a co-occurring specific language impairment is present. It was not clear whether children with SLI-only experience phonological impairments. The magnitude of phonological impairments tended to be greater in the RD+SLI group in comparison to either the RD-only and SLI-only (single deficit) groups, but results were equivocal when comparing the two single deficit groups to one another. Preliminary findings suggest that the pattern of phonological impairments may differ between children with RD-only and SLI-only. Taken together, while these results demonstrated some consistency (i.e. poor phonological skills in children with reading disability and more severe deficits in the combined RD+SLI group) there were also a number of discrepancies, suggesting that further well-designed studies would be beneficial.

Chapter 2 – Academic and Non-Academic Impairments in Learning Disabilities

2.1 Overview of Learning Disabilities

2.1.1 Definition and comorbidity.

Chapter 2 aimed to review the types of impairments with which children with learning disabilities are challenged with, including impairments of academic, cognitive, language and motor functioning. Emotional and behavioural difficulties will also be discussed. Discussion of the broad nature of these difficulties, and how this influences the type of assessment process best suited to assess these children is also reviewed.

Approximately 4-10% of school aged children experience a learning disability (Lagae, 2008; Louden et al., 2000), with 5-17.5% of children meeting criteria for a reading disability (Shaywitz, 1998) and 5-8% meeting criteria for a math disability (Geary, 2004; Geary & Hoard, 2005). The prevalence of spelling disability is less clear, as relevant studies generally incorporate a combined group of children with both reading disability and spelling disability (sometimes labelled ‘dyslexic’) rather than spelling disability only.

Importantly, while reading disabilities, spelling disabilities, and math disabilities can present in isolation, it is more common to meet criteria for multiple learning disabilities (Landerl & Moll, 2010). The aetiology of co-morbid learning disabilities is not well defined, but may be a result of shared genetic, neurobiological or cognitive risk factors (Landerl & Moll, 2010; Moll, Gobel, Gooch, Landerl, & Snowling, 2016; Simmons & Singleton, 2008; Trzaskowski et al., 2013; Willcutt et al., 2013). Recent genetic studies have provided evidence that academic skills are impacted by generalist genes. These are defined as genes that have an effect on multiple academic or cognitive abilities (e.g. affecting both reading and math abilities) (Haworth et al., 2009; Kovas & Plomin, 2007; Plomin & Kovas, 2005; Trzaskowski et al., 2013). Under this hypothesis, learning disabilities commonly co-occur as

the result of generalist genes, while dissociation between learning disabilities is due to independent, non-shared environmental influences and/or the involvement of specialist genes which influence some academic abilities but not others. Overlap between cognitive risk factors may also contribute to the co-occurrence between learning disabilities. Multiple cognitive deficit models propose that neurodevelopmental disorders (including learning disabilities) result from a combination of shared underlying etiological and cognitive risk factors (Pennington, 2006). Phonological skills have been hypothesised as a potential underlying cause of both reading disability and spelling disability.

The co-occurrence between learning disabilities is important for clinicians and educational staff to be aware of. It demonstrates the need for a comprehensive assessment of academic functioning across multiple domains in any child who displays learning difficulties in the classroom. Research has also identified other (non-academic) domains that can be negatively impacted in children with learning disabilities. Impairments are often evident in aspects of cognitive and language functioning as well as in fine and gross motor skills. Poor emotional and/or behavioural functioning have also been identified. The following section of this review examines each of these domains in turn.

2.1.2 Cognitive Functioning.

A variety of cognitive impairments have been identified in children with reading disability, spelling disability and/or math disability, including impairments in aspects of intellectual functioning, attention, processing speed, rapid automatized naming, short-term memory and working memory. These cognitive impairments have the potential to impact a child's ability to learn (e.g. Alloway & Alloway, 2010), which means that a thorough cognitive assessment forms an important part of a learning disability assessment.

Identification of the pattern of cognitive impairments in children with different types of learning disabilities is complex. Although a complete analysis of each cognitive domain is

beyond the scope of this review, a summary of several common types of cognitive functioning impairments (i.e. intellectual functioning, attention, processing speed, rapid automatized naming, short-term memory, working memory and executive functioning) in children with learning disabilities is provided.

2.1.2.1 Intellectual functioning.

Children with learning disabilities have generally been found to have verbal and non-verbal intellectual functioning skills within age appropriate ranges (Cornoldi, Giofre, Orsini, & Pezzuti, 2014; De Clerq-Quaegebeur et al., 2010; Poletti, 2014; Styck & Watkins, 2014; Willcutt et al., 2013). However, they often perform below expected levels on tasks that are designed to measure working memory, such as the Working Memory Index of the Wechsler Intelligence Scale for Children tests (De Clerq-Quaegebeur et al., 2010; Poletti, 2014; Styck & Watkins, 2014).

2.1.2.2 Attention.

Poor attention can impact academic achievement in a number of ways; it can affect classroom engagement and participation (leading to missed learning opportunities); motivation; organization and planning abilities; and a student's ability to complete homework tasks accurately and effectively. Poor attention can also negatively impact other cognitive abilities (e.g. executive functioning skills), which in turn impact learning (e.g. Sayal, Washbrook, & Propper, 2015).

Longitudinal research shows that attentional skills predict later reading and mathematical skill in children and adolescents (Breslau et al., 2009; Duncan et al., 2007; Horn & Packard, 1985; Rabiner, Coie, & group, 2000). Teachers and parents often report that children with learning disabilities display poorer attentional skills than their typically developing peers (Kempe, Gustafson, & Samuelsson, 2011; Maughan, Pickles, Hagell, & Rutter, 1996; Shalev, Auerbach, & Gross-Tsur, 1995). Increased rates of attention deficit

hyperactivity disorder have also been found in children with learning disabilities compared to same age peers (Carroll, Maughan, Goodman, & Meltzer, 2005; DuPaul, Gormley, & Laracy, 2013; Willcutt & Pennington, 2000). Children with learning disabilities *and* attention deficit hyperactivity disorder have been found to not only have more significant attentional deficits but also more severe learning difficulties than those children with learning disabilities alone (Mayes, Calhoun, & Crowell, 2000). However, children with learning disabilities who do not meet criteria for attention deficit hyperactivity disorder also demonstrate poorer attention than their typically developing peers (Mayes et al., 2000) suggesting that the relationship between inattentiveness and learning disabilities is not mediated entirely by the presence of co-occurring attention deficit hyperactivity disorder.

2.1.2.3 Processing speed.

Processing speed is found to be impaired in groups of children with reading disability (Catts, Gillispie, Leonard, Kail, & Miller, 2002; Shanahan et al., 2006; Willcutt, Pennington, Olson, Chhabildas, & Hulslander, 2005), and math disability (Geary, Hoard, Byrd-Craven, Nugent, & Numtee, 2007) in comparison to their same aged peers. Processing speed has been found to be a potential shared cognitive weakness in reading disability and math disability (Willcutt et al., 2013), as well as in reading disability and attention deficit hyperactivity disorder (McGrath et al., 2011; Shanahan et al., 2006; Willcutt et al., 2010).

2.1.2.4 Short-term memory.

Groups of children with literacy difficulties, including reading disability and/or spelling disability, are generally found to demonstrate poor verbal short-term memory skills compared to their peers (Dawes, Leitao, Claessen, & Nayton, 2015; De Weerd, Desoete, & Roeyers, 2013; Fischbach, Konen, Rietz, & Hasselhorn, 2014; Jeffries & Everatt, 2004; Maehler & Schuchardt, 2016; Schuchardt, Maehler, & Hasselhorn, 2008). These deficits are pervasive, and effect sizes for the impairments range from moderate to high (-.39 to -1.10;

Swanson, Zheng, et al., 2009). In contrast, in children with mathematic disabilities, verbal short-term memory abilities are often age appropriate (e.g. Landerl et al., 2009; Swanson & Jerman, 2006), but visuospatial short-term memory impairments are generally found (Geary, 2011; Maehler & Schuchardt, 2016; Schuchardt et al., 2008).

2.1.2.5 Working Memory.

Poor working memory skills have been identified in groups children with reading disabilities (Dawes et al., 2015; De Weerd et al., 2013; Fischbach et al., 2014; S. Gathercole et al., 2006; Schuchardt et al., 2008; Swanson & Ashbaker, 2000), particularly within the verbal domain (Beneventi, Tonnessen, Ersland, & Hugdahl, 2010; Kudo et al., 2015; Landerl et al., 2009; Peng & Fuchs, 2016; Swanson, Zheng, et al., 2009). Evidence also suggests that verbal working memory is impaired in children with math disabilities (Geary, 2011; Peng & Fuchs, 2016; Raghubar, Barnes, & Hecht, 2010; Swanson & Jerman, 2006).

Non-verbal working memory impairments can also be marked in children with math disabilities (Andersson & Ostergren, 2012; McDonald & Berg, 2017; Swanson, Jerman, & Zheng, 2009; Szucs, Devine, Soltesz, Nobes, & Gabriel, 2013; but see De Weerd et al, 2013 for contrasting results). Non-verbal working memory can act as a mental workspace for mathematical operations (when information stored and used to complete tasks is visuospatial, or if visuospatial strategies are used). In children with math disability, the ineffective or inefficient use of this workspace can therefore be associated with poor performance on math tasks (Szucs et al., 2013). Evidence for non-verbal working memory impairments in children with reading and spelling disabilities is inconsistent. Some studies have identified impairments (Fischbach et al., 2014; Willcutt et al., 2005), while others have not (Marzocchi et al., 2008; Willcutt et al., 2005).

2.1.2.6 Rapid Automated Naming.

Rapid automatized naming skills have an established relationship with learning; they predict reading abilities (Furnes & Samuelsson, 2011; Kirby, Parrila, & Pfeiffer, 2003; Parrila et al., 2004; Schatschneider, Fletcher, Francis, Carlson, & Foorman, 2004), spelling abilities (Furnes & Samuelsson, 2011; Savage, Pillay, & Melidona, 2008; Strattman & Williams Hodson, 2005), and mathematic abilities (Koponen et al., 2016). Accordingly, children with reading disability consistently show impaired performance on rapid automatized naming tasks (Denckla & Rudel, 1976; Shanahan et al., 2006; Willburger, Fussenegger, Moll, Wood, & Landerl, 2008). Rapid automatized naming deficits have also been proposed as a core deficit in children with reading disability, either as one component of a broader phonological deficit (e.g. Wagner et al., 1993) or as a cognitive deficit independent of phonological skills (Wolf et al., 2002). Recent evidence suggests that those with math disability also display rapid automatized naming impairments (e.g. Mazzocco & Grimm, 2013), but these impairments may be limited to rapid naming of numerical information such as quantities (Willburger et al., 2008).

In summary, children with learning disabilities can experience an array of cognitive impairments, but the aetiology and precise pattern of these impairments is not yet fully understood. Further research would be beneficial as deficits in cognitive abilities have the potential to impact academic performance (Alloway & Alloway, 2010). Moreover, understanding the pattern of an individual's impairments can guide choice regarding which intervention may be most beneficial to them in the classroom (Fiorello, Hale, & Snyder, 2006).

2.1.3 Language functioning.

The relationship between language and reading skills has been the focus of an extensive body of research. As described in Chapter 1, this research emphasises a

relationship between phonological skills and learning to read (Blaiklock, 2004; Dally, 2006; Ellis, 1990; Hulme, Goetz, Gooch, Adams, & Snowling, 2007; Muter et al., 2004; Oakhill & Kyle, 2000; Wagner et al., 1993; Windfuhr & Snowling, 2001). It also demonstrates that children with reading disabilities experience phonological impairments in relation to their same aged peers (Kudo et al., 2015; Landerl et al., 2009; Larkin & Snowling, 2008; Melby-Lervag et al., 2012). Impairments in phonological skills are hypothesised to be a 'core' deficit in both reading disability and specific language impairment. Approximately 55% of children with reading disabilities also meet criteria for a co-occurring specific language impairment (G. M. McArthur, Hogben, Edwards, Heath, & Mengler, 2000). This indicates that non-phonological language impairments, such as expressive and/or receptive language deficits, are also common in children with reading disabilities. Taken together, the findings suggest that any child presenting with reading difficulties should also have a comprehensive language assessment to quantify any co-occurring phonological and non-phonological language impairments.

The relationship between language and mathematics is less defined. Growth in certain mathematic abilities (e.g. arithmetic, fact retrieval) seem to be influenced by phonological skills (Fuchs et al., 2005; Hecht, Torgesen, Wagner, & Rashotte, 2001; Leather & Henry, 1994; Vukovic, 2012). However, the relationship between math and phonological abilities may be mediated by reading skills. Children with math disability and co-occurring reading disability have been found to demonstrate poor phonological abilities while those without co-occurring reading disability had age appropriate phonological abilities (e.g. Landerl et al., 2009).

2.1.4 Motor skills.

Some children with learning disabilities also demonstrate co-occurring motor impairments (Iversen, Berg, Ellertsen, & Tonnessen, 2005; Ramus, Pidgeon, et al., 2003;

Stoodley, Fawcett, Nicolson, & Stein, 2005). Poor balance and postural instability are often identified (Iversen et al., 2005; Kasselimis, Margarity, & Vlachos, 2008; Ramus, Pidgeon, et al., 2003; Rochelle & Talcott, 2006; Stoodley et al., 2005), while other gross motor deficits, such as impaired object control skills (such as the ability to catch, throw or kick a ball) and locomotor skills (such as running, jumping, hopping) (Westendorp, Hartman, Houwen, Smith, & Visscher, 2011) have also been found. Fine motor impairments include poor manual dexterity (Iversen et al., 2005; Ramus, Pidgeon, et al., 2003; Vuijk, Hartman, Mombarg, Scherder, & Visscher, 2011), and impaired motor sequencing and co-ordination (Marchand-Krynski, Morin-Moncet, Belanger, Beauchamp, & Leonard, 2017).

Knowledge of the relationship between learning disabilities and motor impairments is limited. Impaired cerebellar functioning has been proposed to underlie both learning and motor difficulties; the cerebellum has been found to contribute to both motor skills and cognitive functioning (Nicolson & Fawcett, 2005). Alternatively, motor deficits may be related to academic skills via the influence of a third (moderator) variable, such as presence of another neurodevelopmental disorder, comorbid language impairments, and/or differences in level of intellectual functioning (Brookman, McDonald, McDonald, & Bishop, 2013; Rochelle & Talcott, 2006)

2.1.5 Emotional and behavioural functioning.

Children with learning disabilities demonstrate more externalising behaviours than their same aged peers, as well having an increased incidence of externalising disorders such as oppositional defiant disorder and conduct disorder (Willcutt & Pennington, 2000).

Attention deficit hyperactivity disorder also frequently co-occurs with learning disabilities, and attention deficit hyperactivity disorder symptomatology is thought to mediate the association between learning disabilities and externalising disorders (Barriga et al., 2002; Carroll et al., 2005; Willcutt & Pennington, 2000). Internalising symptoms, including

symptoms of anxiety and depression, are also more common in children with learning disabilities (Mugnaini, Lassi, La Malfa, & Albertini, 2009; Nelson & Harwood, 2011; Willcutt & Pennington, 2000). Research has also suggested increased rates of anxiety and depressive disorders in these children (Goldston et al., 2007; Willcutt & Pennington, 2000; Willcutt et al., 2013), but others have found that symptoms of anxiety and depression are generally sub-clinical in nature in children with learning disabilities (Nelson & Harwood, 2011). Risk factors for co-occurring emotional difficulties in children with learning disabilities include (but are not limited to) severity of academic impairments, presence of multiple learning disabilities, low intellectual functioning and female gender (Mugnaini et al., 2009).

Taken together, past research suggests that children with learning disabilities often experience co-occurring difficulties which span academic, cognitive, language, motor, emotional and behavioural domains. The next section follows on from this discussion, detailing how learning disabilities are assessed and diagnosed in Australia, focusing primarily on diagnostic frameworks that allow thorough assessment of the multifaceted difficulties discussed above.

2.2 Diagnosis of Learning Disabilities

Diagnosis of learning disabilities is complicated for a number of reasons. Firstly, there are inconsistencies in how children with learning disabilities are identified. Low achievement methods (i.e. academic score below a pre-determined cut off), IQ-achievement discrepancy methods (i.e. a significant discrepancy between intellectual ability and academic achievement scores), and response to intervention methods (i.e. diagnosis following an inadequate response to instruction during intervention) have been used variably by learning disability researchers and clinicians (Fletcher, 2012). Precisely what constitutes specific sub-types of

learning disabilities also remains a topic of debate. For example, there are differences between the sub-types of learning disabilities identified in two major diagnostic manuals – Diagnostic and Statistical Manual of Mental Disorders (DSM-V) and International Statistical Classification of Diseases and Related Health Problems (ICD-10), which gives rise to both theoretical and clinical issues.

In Australia, there are several alternative frameworks designed to assess children with suspected learning disabilities. One pathway involves children being administered a cognitive and/or language assessment by a psychologist/speech pathologist employed by their school or department of education. This type of assessment is limited in scope and may not consider potential co-occurring impairments a child may demonstrate. Alternate models of assessment include paediatrician-led assessment or assessment through a multidisciplinary assessment team (Mittal, Sciberras, Sewell, & Efron, 2014). Referral pathways within these models vary depending on the service involved, but can include referral via a general practitioner, school, allied health professional or self-referral by a child's family. Within a paediatrician-led assessment model, the paediatrician works individually and will see a child over several sessions to allow time for a thorough assessment. They may assume multiple roles during the assessment and on-going care phase, including excluding or managing medical or behavioural comorbidities, acting as a child advocate to ensure appropriate community services are involved, working on health education with parents and school staff, co-ordinating a comprehensive assessment process, and/or monitoring longitudinal progress (Oberklaid, 1984). Most paediatricians refer to colleagues (e.g. psychologists and/or speech pathologists) for further evaluation where necessary, which essentially creates a 'de-facto' multidisciplinary team (Roberts, Price, & Oberklaid, 2012).

Multidisciplinary assessment refers to assessments conducted within a setting with various medical/allied health staff working together, but staying within the boundaries of

their individual fields (Choi & Pak, 2006). This allows a comprehensive evaluation to be conducted. Multifaceted evaluations such as these involve the gathering of social, developmental, medical and educational information (via interviews and/or questionnaires) from parents and teachers. Parent and teacher questionnaires completed in the context of learning disability assessment allow efficient gathering of large amounts of information from multiple sources. Information gathered in this way can often also be standardised using normative data (Oberklaid, 1984). The child undergoing assessment is also asked to complete a series of psychometric tests. Broad psychometric testing helps ensure that the variety of domains impacted in children with learning disabilities are assessed, including academic achievement, cognitive functioning, language functioning, motor skills, and social-emotional and behavioural components (Handler & Fierson, 2011).

Given the complexities involved in assessing children with suspected learning disabilities, multidisciplinary assessment can be well suited to provide a thorough and efficient evaluation. It also contributes to accurate diagnosis of neurodevelopmental disorders, particularly when diagnosing uncommon disabilities and in determining co-occurring conditions (Hendriksen et al., 2007; Mittal et al., 2014). Multidisciplinary assessments allow each clinician to provide their own expertise and perspective on an issue (Choi & Pak, 2006), which can improve understanding of complex problems that a single discipline alone may have difficulty adequately addressing. A recent Australian based study found that parents also noted benefits of multidisciplinary assessment clinics for learning and behavioural problems (Mittal et al., 2014). Parents of children referred to these clinics were more likely than parents of children referred for paediatrician-led assessment to report that they better understood their child's difficulties, that their communication with the school improved, and that their child's school experience improved following the assessment.

One issue associated with multidisciplinary assessment clinics is that they can be difficult to access. These clinics are relatively scarce within the public health system. In part, this is likely due to them being costly to run and the need for them to be staffed by health professionals with expertise in this type of assessment and care. Private multidisciplinary clinics are also available, but the cost of attending these clinics is prohibitive for some families.

In summary, clinicians involved in diagnosis of learning disabilities face a number of challenges. There are several well-established methods of assessment and diagnosis available in Australia, with multidisciplinary clinics (though difficult to access for some families) particularly well placed to provide a thorough, efficient and accurate assessment in the context of suspected learning disabilities.

2.3 Thesis Rationale, Aims and Hypotheses

2.3.1 Summary and rationale.

Learning disabilities are the focus of an abundance of literature but further research is warranted in a number of areas. Clarification is needed regarding the nature of the relationship between cognitive load and phonological skills structure. There is some evidence to suggest that cognitive load may differentiate between distinct phonological constructs. It is also unclear as to whether the prevalence, magnitude and pattern of phonological impairments is the same or different in two neurodevelopmental disorders, reading disability and specific language impairment. A broad discussion of the characteristics of impairments commonly experienced by groups of children with learning disabilities is also needed. These impairments can span a variety of domains (e.g. academic, cognitive, language, motor, emotional and behavioural domains), which have typically been investigated individually rather than described broadly. In sum, this review identified a

number of gaps within the learning disabilities literature, and these gaps formed the basis of the following aims and hypotheses.

2.3.2 Aims and hypotheses.

The specific aims and related hypotheses for this thesis were as follows:

1. To investigate the structure of phonological skills in children referred to a learning difficulties clinic and to understand how these skills are related to short-term memory and verbal working memory.

- i. To determine whether phonological skills were best conceptualised as a single cognitive construct or as multiple distinct constructs.

It was hypothesised that phonological skills would be best conceptualised as two distinct, but related, cognitive constructs which would be differentiated by cognitive load. That is, we expected separation of higher- and lower-level phonological skills constructs.

- ii. If multiple constructs were identified, to determine whether these were differentially related to short-term memory and verbal working memory.

It was hypothesised that the higher-level phonological skill construct would be more strongly related to verbal working memory than it would to short-term memory (which is associated with tasks that do not place high load on cognitive systems).

2. To examine the nature of phonological impairments in children with reading disabilities (RD) and/or specific language impairment (SLI).

- i. To determine the prevalence of phonological impairments in groups of children with RD-only, SLI-only and RD+SLI.

It was hypothesised that all groups would show impaired phonological skills as demonstrated by a mean phonological skill score within the impaired range on a standardised phonological skill test battery.

- ii. To determine the magnitude of phonological impairments in each of the three groups.

It was hypothesised that the magnitude of phonological impairments would be greater in the RD+SLI group, indicating more severe phonological deficits. We tentatively expected no differences in the magnitude of phonological impairments between the RD-only and SLI-only groups.

- iii. To determine the proportion of children in each of the clinical groups who display phonological impairments.

It was hypothesised that a lower proportion of the SLI-only group would demonstrate phonological impairments than the RD-only group.

- iv. To describe the pattern of phonological impairments in each of the three groups using a standardised phonological skills assessment battery.

It was expected that the patterns of performance would be different for the RD-only, SLI-only and RD+SLI groups as indicated by non-parallel profiles using profile analysis.

3. To provide a broad overview of the characteristics of children referred to a multidisciplinary learning difficulties clinic for assessment. In a cohort of primary school aged children, we aimed to determine:

- i. The pattern and nature of academic impairments in the areas of reading, spelling and mathematics.
- ii. The pattern and nature of non-academic difficulties across cognitive, language, motor, emotional and behavioural domains.
- iii. Potential risk factors for non-academic difficulties in a sub-group of children who met criteria for at least one learning disability.

Chapter 3 - Structural Analysis of Phonological Skills and Related Cognitive Skills in Children Referred to a Learning Difficulties Clinic

Preamble

Chapter 3 contains a manuscript submitted for publication in the *Journal of Educational Psychology* on 18/04/2019. It has been inserted into this thesis in the format required for manuscript submission. The manuscript comprises a structural analysis of phonological skills, short-term memory and working memory in a cohort of primary school aged children referred to a learning difficulties clinic.

Abstract

Opposing theories question whether phonological skills are best represented as a single cognitive construct or, alternatively, are representative of multiple distinct constructs. Cognitive load has been identified as an important variable when considering these theories. There is some evidence that distinct higher- and lower-level phonological skills constructs can be identified which differentially recruit working memory networks and are therefore differentiated by cognitive load. This study aimed to investigate whether phonological skills were best conceptualised as a single cognitive construct or as multiple distinct constructs. If multiple constructs could be identified, we were also interested in whether these were differentially related to short-term memory and verbal working memory, and how cognitive load impacted upon these relationships. To examine the structure of phonological skills, short-term memory and working memory in primary aged children, data was retrospectively analysed for 121 children who were referred to a learning difficulties clinic. Children had completed a selection of phonological and memory tasks. Confirmatory factor analysis showed that, as hypothesised, phonological skills were best conceptualised as two closely related but distinct factors which were distinguished by their cognitive load (high versus low). The results confirm the importance of considering cognitive load when examining structure of phonological skills.

Keywords: Phonological skills, short-term memory, verbal working memory, cognitive load, phonological awareness

3.1 Introduction

3.1.1 Phonological skills in childhood.

A child's ability to be consciously aware of the sound segments within words is underpinned by the development of early language skills, including the development of speech perception and production. Precursors to speech production begin in early infancy, with pre-linguistic vocalisations such as crying. Canonical babbling (i.e. strings of alternative consonants and vowels) begins in the second half of the first year and by 12 months, word production is generally beginning to develop (Nathani, Ertmer, & Stark, 2006; Oller, 2000). By this age, children can distinguish most speech sounds within their native language (Hollich, 2010). Ongoing growth of speech perception and production supports the development of phonological representations, which are the sound based codes of words stored in long term memory (Claessen & Leitao, 2012; Sutherland & Gillon, 2007). Other phonological skills also begin to develop, following a relatively predictable trajectory from an awareness and understanding of large phonological units (e.g. words) through progressively smaller phonological units, until the smallest unit (the phoneme, such as /wh/ in when; /h/ in his, /oo/ in foot) is reached (Ziegler & Goswami, 2005). This occurs around the age that Australian children enter school (Muter, Hulme, Snowling, & Stevenson, 2004). As phoneme-level skills develop, children can perform tasks that require them to isolate a beginning or end phoneme within a word, blend phonemes together to form a word, and segment phonemes within a word. The ability to complete more cognitively demanding phoneme manipulation tasks (e.g. phoneme deletion tasks) develops later and in parallel with other cognitive skills, such as working memory. Mastery of these complex tasks generally occurs after the end of first grade (Adams, 1992; Farrall, 2012).

3.1.2 Structure of phonological skills

The structure of phonological skills is not yet well understood. Opposing theories question whether different phonological skills are best represented as a single cognitive construct (e.g. Adams, 1992; Anthony & Francis, 2005; Schatschneider, Francis, Foorman, Fletcher, & Mehta, 1999; Stahl & Murray, 1994) or, alternatively, are representative of multiple, distinct cognitive constructs (e.g. Høien, Lundberg, Stanovich, & Bjaalid, 1995; Oakhill & Kyle, 2000; Ramus, Marshall, Rosen, & van der Lely, 2013; Wagner, Torgesen, & Rashotte, 1994; Yopp, 1988). For instance, the ability to recognise, isolate and manipulate different sized phonological units may represent distinct phonological constructs (Høien et al., 1995), or alternatively may form one part of a unitary phonological construct (Anthony & Lonigan, 2004; Stahl & Murray, 1994). Similarly, some research has found rhyme skills (e.g. the ability to detect or produce rhyming words) are so closely related to other aspects of phonological skills that they combine to form part of a single phonological construct (Anthony & Lonigan, 2004). Conflicting evidence suggests, however, that rhyme skills are separable and distinct from other phonological skills. When compared to other phonological skills, rhyming abilities have been found to be differentially related to reading and spelling (Muter, 1998), and rhyming tasks often load onto a separate factor in structural analyses (Beach & Young, 1997; Runge & Watkins, 2006; Yopp, 1988).

Another way to examine the structure of phonological skills is to look at the developmental trajectory of phonological skills and to consider whether cognitive load may differentiate between different types of phonological skills. Cognitive load is a multidimensional construct which represents the load that performing a task imposes on an individual's cognitive system (Paas & Van Merriënboer, 1994). The age-related developmental trajectory of phonological skills emphasises that there are early developing phonological skills, such as phoneme identification and blending, which are less cognitively

demanding than other, later developing phonological skills such as phoneme deletion (Farrall, 2012). The differences in how cognitively taxing or demanding these tasks are related to differences in their cognitive load. Understanding how cognitive load relates to these early- and later- developing phonological skills can help us understand how these skills are related to other cognitive abilities (e.g. short-term memory and working memory) as well as academic abilities (e.g. reading).

Past research investigating the relationship between cognitive load and phonological skills is limited. Two identified studies found distinct lower- and higher-level phonological constructs. Overall, results showed that tasks loading onto the higher-level phonological construct required greater cognitive effort and attention than those loading onto the lower-level construct. In a community based group of children, the lower-level phonological construct comprised of tests that required completion of a single cognitive operation (e.g. isolate a sound) prior to providing a response. The higher-level construct included tests that required multiple cognitive operations, where participants needed to hold information in their mind while simultaneously using or manipulating that information (Yopp, 1988). Yopp (1988) also found that the lower and higher-level constructs were highly correlated but differentially contributed to reading skills, whilst Ramus et al. (2013) identified higher and lower-level constructs that were differentially impaired in children with reading disabilities compared to those with a specific language impairment.

3.1.3 The relationship between phonological skills and memory skills

One way to better understand the relationship between cognitive load and phonological skills is to consider the cognitive skills that contribute to performance on phonological tasks. A number of authors have emphasised a relationship between phonological skills, short-term memory and verbal working memory (e.g. Alloway, Gathercole, Willis, & Adams, 2004; Leather & Henry, 1994; Yopp, 1988). Short-term

memory refers to the storage of small amounts of information over a short period of time (seconds) in order to recall the information in an untransformed way (Swanson, Zheng, & Jerman, 2009). Typically this is measured by asking the individual to serially recall a series of digits, words or locations. While nonword repetition tasks can also be conceptualised as a short-term memory measure (Alloway, Gathercole, & Pickering, 2006; Alloway et al., 2004), recall of novel phonological information (e.g. nonwords) may rely upon different (phonological) memory processes than immediate recall of familiar information such as digits or words (e.g. Briscoe, Bishop, & Norbury, 2001). Verbal working memory is the ability to attend to and maintain verbal information that has just been experienced (or retrieved from long-term memory) whilst manipulation of that, or other, information occurs (D'Esposito, 2008; S. Gathercole & Alloway, 2006). Verbal working memory tasks include backward span tasks, where individuals recall a series of digits, words, or letters in reverse order, and complex span tasks with dual processing requirements. Both short-term memory and verbal working memory tasks require holding and maintaining information in mind for a short period of time. Verbal working memory tasks have an additional cognitive load as they require simultaneous processing, such as the manipulation and integration of relevant information. Cognitive load is therefore greater when completing more complex (higher-level) verbal working memory tasks than when completing (lower-level) short-term memory tasks.

Similar to the structure of phonological skills, there are questions regarding how to best conceptualise the relationship between short-term memory and working memory (see Aben, Stapert, & Blokland, 2012 for a review of this topic). The most influential models have conceptualised working memory as a multicomponent system consisting of a short-term memory component (or components) and a higher-level attentional control system (e.g. Baddeley, 2000; Cowan, 2008; Engle, Tuholski, Laughlin, & Conway, 1999). Empirical

findings in children and adults are relatively consistent. Structural analyses show that short-term memory and working memory load onto separate factors in factor analyses (Alloway et al., 2006; Alloway et al., 2004; S. E. Gathercole, Pickering, Ambridge, & Wearing, 2004) and are differentially related to reading abilities in children (Kail & Hall, 2001). Taken together, the findings suggest that short-term memory and working memory are separate, but related, cognitive abilities.

Empirical research findings also suggest that the strength of the relationship between short-term memory and working memory changes with age. S. E. Gathercole et al. (2004) found that verbal short-term memory and verbal working memory became less differentiated across age in four to 15 year olds. These changes likely relate to differences in the developmental trajectories of these cognitive skills. The specific nature of age-related changes may also depend on the material being presented, attended to, and used (i.e. verbal vs non-verbal material), with Alloway et al. (2006) finding increased differentiation between visuospatial short-term memory and working memory over time.

The relationships between phonological skills, short-term memory and working memory have also been examined. It has been hypothesised that both short-term memory and phonological constructs may share a common underlying phonological resource but also tap into distinct cognitive mechanisms (e.g. S. E. Gathercole & Willis, 1991). This suggests that they are related but separable from one another. This hypothesis is supported by structural analyses findings which show that short-term memory and phonological tasks load onto separate cognitive constructs (Alloway et al., 2004; Dufva, Niemi, & Voeten, 2001), as well as evidence showing that short-term memory and phonological skill differentially contribute to reading skills (Nithart, Demont, Metz-Lutz, Majerus, & Poncelet, 2011; Parrila, Kirby, & McQuarrie, 2004).

The relationship between working memory and phonological skills is evident in neuroimaging studies which show that different neural networks are recruited depending on the working memory demands of the phonological tasks being completed. Frontal regions of the brain, which are frequently considered to form part of verbal working memory neural networks are recruited when individuals are required to perform effortful, multiple step sequencing of phonological information (i.e. selectively attend, segment and hold a consonant from one word to compare it with an initial consonant from a second word). In contrast, when no segmentation is required, these regions are not engaged (Burton, Small, & Blumstein, 2000). Further evidence that working memory demands can differ between phonological tasks comes via examination of the requirements of the tasks themselves. Tasks requiring the deletion of a phoneme from within a word are likely to place significant demands on working memory systems as they require both maintenance and manipulation of phonemes to produce an accurate response. Other phonological tasks, such as simple CVC phoneme blending tasks, are easier and less cognitively demanding for children to complete (Anthony, Lonigan, Driscoll, Phillips, & Burgess, 2003; Yopp, 1988). Although brief maintenance and manipulation (i.e. blending) of phonemes is required in these tasks, they are likely to be significantly less demanding of working memory systems than deletion tasks are.

Taken together, the aforementioned studies provide some understanding about the relationships between phonological skills, short-term memory and verbal working memory, although the precise nature of these relationships needs to be further examined. A cognitive load framework may be a useful and informative way to investigate these relationships. Within this framework, it can be determined whether higher and lower-level phonological constructs, which differentially recruit working memory neural networks, can be differentiated.

3.1.4 The present study

This study investigated the structure of phonological skills in a cohort of children referred to a learning difficulties clinic. It examined whether phonological skills were best conceptualised as a single cognitive construct or as multiple distinct constructs; how phonological skills were related to short-term memory and verbal working memory; and how cognitive load impacted upon these relationships. Investigation into these questions can inform and guide clinical practice; if distinct higher- and lower-level phonological constructs are able to be identified, clinicians can ensure both process levels are considered in a learning difficulty assessment and interventions. We hypothesised that phonological skills would be best conceptualised as two distinct, but related cognitive constructs that would be differentiated by cognitive load (i.e. high- and low-level phonological skills). Further, tasks associated with the higher-level phonological construct and verbal working memory constructs would be assumed to be associated with high cognitive load. As such, we expected the higher-level phonological construct to be more strongly related to verbal working memory than it would to short-term memory (which is associated with tasks that do not place high load on cognitive systems).

3.2 Method

3.2.1 Participants.

Participants were selected from a cohort of primary school aged children (N=326) who represented consecutive referrals to the Learning Difficulties Clinic, Sunshine Hospital, between 1996 and 2008. The Learning Difficulties Clinic specialised in multidisciplinary assessment of primary school aged children with suspected learning difficulties. Referrals to the clinic were made by paediatricians or schools and were not accepted if the child had any

known intellectual disability, neurological or psychiatric condition, or significant emotional or behavioral difficulties which would preclude a valid assessment.

Children were selected for this study if they had completed the Sutherland Phonological Awareness Test (SPAT; Neilson, 1995) *and* the Wechsler Intelligence Scale for Children, Third edition (WISC-III; Wechsler, 1991). The final included sample consisted of 121 children with 87 (72%) males and 34 (28%) females and a mean age of 8.76 (range six-12) years. Twenty-four (20%) of children were in grade 1 at the time of their assessment, 32 (26%) grade 2, 26 (22%) grade 3, 19 (16%) grade 4, 17 (14%) grade 5 and 3 (3%) grade 6. Formal intelligence testing had not previously been completed for most of these children, and subsequent formal assessment revealed seven children whose full scale IQ fell within the Extremely Low range (≤ 70). Mean full scale IQ for the group was 86.7 ($SD=11$). A number of children experienced literacy impairments, with 86 (71%) falling more than one grade below the expected level in reading and 80 (66%) falling more than one grade below the expected level in spelling (reading and spelling data for one individual was missing). Sixty-six (55%) demonstrated phonological impairments.

3.2.2 Materials.

3.2.2.1 *Phonological skills.*

In order to measure higher and lower-level phonological skills, children completed 11 subtests from the SPAT (Neilson, 1995). The score range for each subtest was 0-4, and administration included an example item, practice item and four test items. Subtests were designated as lower or higher-level tasks based upon consideration of the age of participants, examination of the cognitive skills presumed to be required to successfully complete each task, and evidence from past empirical research. Phoneme counting, identification, blending and segmentation tasks, along with rhyme tasks have been found to be easier and less cognitively demanding than phoneme deletion tasks (Stanovich, Cunningham, & Cramer,

1984; Yopp, 1988). Accordingly, subtests measuring these skills were designated as lower-level phonological subtests. Phoneme deletion tasks are generally more cognitively demanding (e.g. Yopp, 1988), but the level of difficulty of deletion tasks varies depending on the linguistic complexity of their items. Tasks requiring deletion of a phoneme from a consonant blend (e.g. remove the /p/ from play) are more challenging than tasks requiring deletion of an individual phoneme from a CVC word (e.g. remove the /c/ from cat; Stahl & Murray, 1994; Treiman, 1992). As such, tasks requiring deletion of a phoneme from a consonant blend were designated higher-level phonological subtests, while the other deletion task was considered to be less cognitively demanding for primary school aged children.

3.2.2.1.1 Lower-level phonological subtests.

Using the above rationale, nine subtests, which focused on either syllables, onset-rimes or phonemes, were selected to measure lower-level phonological skills. The *syllable counting* subtest required the child to tap out the number of syllables in a word (e.g. helicopter) provided orally by the examiner. The *rhyme detection* subtest required the child to pick one of two pictures that rhymed with a simple picture provided to them. Oral descriptions of all three pictures were provided by the examiner (e.g. “This one is ‘pig’. You have to choose the picture that rhymes with ‘pig’ ‘dig’ or ‘cup’?”). In the *rhyme production* subtest the examiner provided two words that rhymed, and the child was required to make up a word that also rhymed (e.g. cat, fat,?). Non-words provided by children in this subtest were scored as correct. The *onset identification* and *final phoneme identification* subtests required the child to identify the sound at the beginning or end of a word respectively (e.g. “What sound does sun begin with?” or “What is the last sound you hear in the word boot?”). In the *segmentation 1* and *segmentation 2* subtests the examiner orally presented a word, and the child was required to segment it into its corresponding phonemes. In addition, the child was presented with a stimulus sheet containing five numbered boxes, and was asked

to tap one box for each sound they produced. To score a point for a correct response, the child needed to pronounce all phonemes correctly as well as tapping the correct number of boxes. The *blending* subtest required the child to blend together a number of sounds provided by the examiner. The *initial phoneme deletion* subtest required the child to delete the first sound of the word and say the word that was left (e.g. “Meat. Take off /m/”).

3.2.2.1.2 Higher-level phonological subtests.

Two subtests were selected as higher-level phonological skills tasks. Both tasks consisted of items beginning with consonant blends (CCVC words). In the *deletion of first phoneme* task, the child was asked to take the first sound out of a word and say the word that was left (e.g. “Take off the /p/ from ‘play’”). In the *deletion of second phoneme* task, the child was to remove the second sound from a word and say the word that was left (e.g. “Take the /m/ out of ‘smack’”).

3.2.2.2 Short-term memory and verbal working memory.

The digit span subtest from the WISC-III (Wechsler, 1991) was administered as a measure of verbal short-term memory (digit span forward) and verbal working memory (digit span backward) abilities. This subtest consisted of two parts. In the ‘forward’ component, the child was asked to repeat sequences of digits of increasing length, whilst in the ‘backwards’ component, the child listened to sequences of digits of increasing length and repeated the sequence backwards.

The sentence memory subtest from the Wide Range Assessment of Memory and Learning (WRAML; Sheslow & Adams, 1990) or WRAML, Second Edition (WRAML-2; Sheslow & Adams, 2003) was administered as a measure of short-term memory for words in context. In this subtest, individuals must repeat a series of sentences (varying from 2-26 words) one at a time. Raw scores from the WRAML and WRAML-2 were combined for analyses as items from the two versions were considered equivalent. The ability to access the

phonological form of a sentence in short-term memory has been identified as a strong contributor to performance on sentence memory tasks (Willis & Gathercole, 2001), though various other speech and language abilities are also thought to support accurate performance on these tasks (Klem et al., 2015; Polisenska, Chiat, & Roy, 2015)

A verbal fluency sorting task was also included as a verbal working memory task. Individuals generated as many animals as possible in one minute, in size order from smallest to largest (which placed demands on working memory).

A visuospatial working memory task was not included because at the time of data collection, no such task was readily available at the Learning Difficulties Clinic. A measure of visuospatial short-term memory, block span, was included because we aimed to broadly examine short-term memory and working memory where appropriate tasks were available. On the block span task children repeated sequences of increasing length across a nine-position spatial array. Two trials at each span length were administered and the total score was recorded as the total number of trials correct.

3.2.3 Procedure.

This project was reviewed and approved by Western Health (HREC/11/WH/79) and Monash University (2012000503) HREC's. At the time of assessment a parent/guardian provided consent for data to be used in future research studies. Each child assessed in the clinic underwent a two-day assessment which included various psychometric tests and gathering of a detailed background history from each child's parent/guardian. Results were then entered into a database, and relevant data was retrospectively accessed for the purposes of this study.

3.2.4 Analyses.

Two-tailed Pearson correlation analyses were used to examine zero-order relationships among age, memory and phonological task performances. In order to adjust for

any effects of age, linear regression analyses were performed and standardised residuals were calculated for all memory and phonological tasks that were significantly correlated with age. For these variables, standardised residuals (rather than raw scores) were used for subsequent primary analyses. For variables not significantly correlated with age, raw scores were used for subsequent primary analyses. In order to examine the impact of age on the results, follow up analyses were also conducted using raw scores for all variables to allow comparison with age adjusted results.

Confirmatory factor analysis, conducted using AMOS (Arbuckle, 2013), was used to examine how phonological skills, short-term memory and verbal working memory were related. Five measurement models were created to define the relationship between the observed variables and unobserved latent constructs. These models differed along three dimensions. Firstly, whether phonological skills were considered as a unitary construct or separate (lower and higher-level) constructs; Secondly, whether memory skills were considered as unitary or separate (short-term memory and verbal working memory) constructs; and finally, whether the phonological skills *and* memory skills were considered to be lower- and higher-level cognitive skills.

Model fit was initially assessed using the chi-squared statistic. Small and non-significant chi-squared values are considered indicative of good fit (Hair, Black, Babin, & Anderson, 2014). Further fit statistics were also examined: root-mean-square error (RMSEA), standardised root mean square residual (SRMR), comparative fit index (CFI), goodness of fit index (GFI) and the Tucker-Lewis index (TLI). RMSEA values of ≤ 0.07 are considered acceptable (Steiger, 2007), while for SRMR values over 0.1 indicate a poor fit and ≤ 0.08 are considered acceptable (Hair et al., 2014; Hu & Bentler, 1999). For the remaining indices (CFI, GFI and TLI), values > 0.9 are considered to demonstrate good fit (Hair et al., 2014).

3.3 Results

Sixteen variables were considered for inclusion in this study; 11 SPAT subtests, digit span forwards and backwards, block span, animal fluency (sort), and sentence memory. Descriptive statistics for each of these variables are summarised in Table 3.1. Each variable was analysed for the percentage of missing data and data was found to be missing completely at random, Little's MCAR test $p=.075$. Four variables had between 1% and 14% of data missing. In accordance with Hair, Black, Babin and Anderson (2014), the available data were judged appropriate for imputation using regression replacement, and this was completed using SPSS Missing Value Analysis.

Each variable was examined for the presence of univariate outliers and violations of normality. In accordance with Field (2009) and Tabachnick and Fidell (2013) a visual inspection of histograms was conducted and a unimodal distribution was found for all variables. However, for three variables (SPAT rhyme detection, SPAT onset identification and SPAT final phoneme identification), skewness and kurtosis values were above the acceptable limit of ± 2 (George & Mallery, 2010), suggesting extreme non-normal distributions. At least 74% of participants scored at ceiling on each of these three variables and the variables were therefore excluded from further analyses. Within the remaining variables, one item had a skewness of 2.08, associated with a particularly extreme outlier. This was corrected by winsorizing the value to one unit larger than the next most extreme score for that variable (Tabachnick & Fidell, 2013). One variable (digit span forwards) had acceptable skewness but kurtosis >5 . It was decided that transformation of this variable was unnecessary given that underestimates of variance that are associated with positive kurtosis disappear with samples of >100 cases (Tabachnick & Fidell, 2013).

Table 3.1

Descriptive statistics for phonological, short-term memory and working memory tasks (N=121).

Variable	Mean	SD
Digit span forwards	4.97	1.26
Digit span backwards	2.78	1.16
Sentence memory	6.28	2.29
Block span	5.47	1.69
Animal fluency (sort)	4.05	2.90
SPAT syllable counting	3.05	1.12
SPAT rhyme detection ^a	3.66	0.76
SPAT rhyme production	2.97	1.37
SPAT onset identification ^a	3.77	0.70
SPAT final phoneme identification ^a	3.45	1.13
SPAT segmentation 1	3.01	1.27
SPAT blending	3.27	1.30
SPAT initial phoneme deletion	2.30	1.66
SPAT segmentation 2	2.11	1.57
SPAT deletion first phoneme	1.60	1.57
SPAT deletion second phoneme	1.24	1.57

^aThese subtests were not included in subsequent analyses due to severely non-normal distributions

3.3.1 Correlational analyses.

Two-tailed Pearson correlation analyses among all variables were computed using raw scores for each short-term memory, verbal working memory and phonological task (see Table 3.2). No intercorrelation exceeded 0.7, suggesting that multicollinearity was not an issue (Field, 2009). Intercorrelations between six of the phonological tasks were significant at a more conservative level ($\alpha=0.01$) to adjust for multiple comparisons, ranging in magnitude from small (0.27) to large (0.62). However, intercorrelations between the syllable counting subtest and other phonological tasks were relatively lower, ranging from small (0.10) to moderate (0.31). Intercorrelations between short-term memory tasks ranged from small (0.10) to moderate (0.29), while the intercorrelation between the two working memory tasks was moderate (0.30).

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Table 3.2

Correlations between age, phonological skills, short-term memory and working memory (raw) scores

	1	2	3	4	5	6	7	8	9	10	11	12	13	14
1. Age														
2. DSF	.29**													
3. DSB	.42**	.31**												
4. Sentence memory	-.17	.24**	.15											
5. Block span	.49**	.29**	.36**	.10										
6. Fluency (sort)	.35**	.14	.30**	-.05	.28**									
7. Syllable count	.23*	-.08	.16	.12	.13	.17								
8. Rhyme prod	.39**	.14	.31**	.09	.28**	.21*	.18							
9. Segment 1	.32**	.30**	.35**	.13	.41**	.13	.21*	.31**						
10. Blending	.29**	.23*	.44**	.08	.21*	.16	.12	.57**	.58**					
11. Initial ph deletion	.39**	.29**	.40**	.09	.30**	.21*	.31**	.39**	.62**	.62**				
12. Segment 2	.28**	.20*	.37**	.15	.20*	.10	.10	.41**	.57**	.55**	.56**			

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13. Deletion 1st ph	.22*	.30*	.35**	.19*	.20*	.01	.16	.27**	.52**	.41**	.56**	.52**	
14. Deletion	.44**	.32*	.39**	.15	.16	.22*	.24**	.32**	.36**	.34**	.56**	.49**	.61**
2nd ph													

Ph=phoneme

** indicates statistical significance at $\alpha = .01$, * indicates statistical significance at $\alpha = .05$

3.3.2 Confirmatory factor analysis.

To adjust for age, standardised residuals were used in the primary confirmatory factor analyses for all but one task (sentence memory). Raw scores were used for sentence memory as it was not significantly correlated with age. Statistics and goodness-of-fit indices for five tested measurement models are presented in Table 3.3. For all models, the paths between latent constructs were left free to covary (represented by bidirectional arrows) because there was insufficient evidence regarding direction of causality in the relevant literature.

Table 3.3

Goodness-of-fit statistics for measurement models (using standardised residuals)

	Chi- square	Degrees of freedom	P	RMSE A	SRM R	CFI	GFI	TLI
Model 1 (2 factors: Memory and phonological skills)	118.63	64	<.001	0.08	0.07	0.85	0.88	0.82
Model 2 (2 factors: Higher level and lower-level phonological skills)	114.80	64	<.001	0.08	0.08	0.86	0.88	0.83
Model 3 (3 factors: Memory, higher-level phonological skills, and lower-level phonological skills)	99.84	62	.002	0.07	0.07	0.90	0.89	0.87

Model 3a* (3 factors: Memory, higher-level phonological skills and lower-level phonological skills)	74.98	51	.016	0.06	0.06	0.93	0.91	0.91
Model 4 (3 factors: STM, verbal WM, and phonological skills)	125.30	63	<.001	0.09	0.08	0.83	0.87	0.79
Model 5 (4 factors: STM, verbal WM, higher-level phonological skills and lower-level phonological skills)	105.16	60	<.001	0.08	0.08	0.87	0.89	0.84

RMSEA=Root-mean-square error; SRMR=Standardised Root Mean Square residual;

CFI=Comparative fit index; GFI=Goodness of fit index; TLI=Tucker-Lewis index

* Model 3a and Model 5a do not include the rhyme production task.

3.3.2.1 Two-factor models.

Two two-factor models were tested. Model One distinguished between a memory construct (associated with all short-term memory and verbal working memory tasks) and a phonological construct (associated with all phonological tasks). Model Two distinguished between a higher-level cognitive construct (working memory tasks and higher-level phonological tasks) and a lower-level cognitive construct (short-term memory and lower-level phonological tasks). Neither of the two-factor models provided a good fit for the data (see Table 3). In both cases the SRMR was acceptable (Model One=0.07 and Model

Two=0.08), but chi-squared value was significant, RMSEA was 0.08, and all other goodness-of-fit statistics were below 0.90.

3.3.2.2. Three-factor models

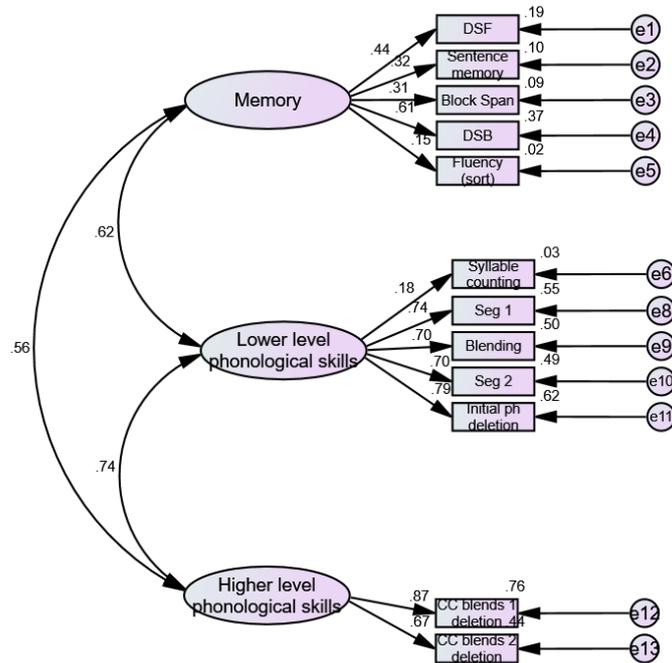
Two three-factor models were tested. Model Three distinguished between a higher-level phonological construct (associated with two higher-level phonological deletion tasks), a lower-level phonological construct (associated with all other phonological tasks) and a memory construct (associated with all short-term memory and working memory tasks). Model Four distinguished between separate short-term memory and verbal working memory constructs, as well as an overall phonological construct. Neither model provided a good fit for the data (see Table 3.3).

3.3.2.3 Four-factor model.

Model Five distinguished between higher and lower-level phonological constructs and two memory constructs (i.e. short-term memory and verbal working memory). Statistics and goodness-of-fit indices for Model Five were all considered inadequate except for SRMR (0.08).

Given that none of the initial five models provided an adequate fit for the data, re-examination of included variables was undertaken and it was decided that models would be re-evaluated after the removal of one variable (rhyme production). This variable was excluded for theoretical reasons, as there is evidence that rhyme tasks may tap into different phonological processes than the other phonological tasks (Beach & Young, 1997; Muter, 1998; Runge & Watkins, 2006; Yopp, 1988). Each of the five models were re-tested after excluding rhyme production; Model 3a (see Figure 3.1) was considered a good fit for the data (see Table 3.3). While the chi-square was significant ($p=.016$), RMSEA value (.06) and SRMR values (.06) were adequate and all goodness-of-fit statistics were greater than .90.

Figure 3.1. *Model 3a* - Three factor model distinguishing between higher- and lower-level phonological constructs and a memory construct.



In order to examine whether age impacted the results, follow up analyses were then conducted using raw scores for each variable. When compared to the primary analyses (which used standardised residuals), confirmatory factor analyses using raw scores resulted in a similar pattern of results. Model 3a once again provided an adequate fit to the data, while Model 5a was also considered adequate (see Table 3.4). Comparison of these two models showed that Model 3a would be considered to fit slightly better than Model 5a. The fit statistics were very similar between the two, but Model 3a had three more degrees of freedom

(Hair, 2014). As such, in the context of both the primary (standardised residual) results and raw score results, Model 3a was selected for further examination.

Table 3.4.

Goodness-of-fit statistics for the two measurement models which provided an adequate fit for the data (using raw scores).

	Chi-sq.	Degrees of freedom	P	RMSEA	SRMR	CFI	GFI	TLI
Model 3a* (3 factors: Memory, higher-level phonological skills and lower-level phonological skills)	80.06	51	.006	0.07	0.06	0.93	0.90	0.91
Model 5a* (4 factors: STM, verbal WM, higher-level phonological skills and lower-level phonological skills)	78.86	49	.004	0.07	0.06	0.93	0.91	0.90

RMSEA=Root-mean-square error; SRMR=standardised root mean residual;

CFI=Comparative fit index; GFI=Goodness of fit index; TLI=Tucker-Lewis index;

STM=Short-term memory; WM=Working memory * Model 3a and 5a do not include the rhyme production task.

Using standardised residuals, correlations between factors in Model 3a ranged from 0.56 to 0.74, while correlations between factors ranged from 0.62 to 0.81 using raw scores. (see Table 3.5).

Table 3.5.

Correlations between factors from Model 3a: Standardised residuals (raw scores)*

	Lower-level ph. skills	Higher-level ph. skills	Memory
Lower-level ph. skills			
Higher-level ph. skills	0.74 (0.81)		
Memory	0.62 (0.69)	0.56 (0.62)	

* Standardised residuals were used for all tasks except for sentence memory in this analysis.

3.4 Discussion

The purpose of this study was to investigate the structure of phonological skills, short-term memory and verbal working memory in primary school aged children referred to an Australian Learning Difficulties Clinic. A majority of children included in the study demonstrated reading, spelling and/or phonological impairments. Confirmatory factor analysis results were similar using both raw scores and age adjusted scores. As hypothesised, phonological skills were best conceptualised as two distinct, but closely related factors that were distinguished by their level of cognitive load (high versus low). The first phonological factor included syllable counting, in addition to simple phoneme blending, phoneme deletion

(initial sound) and phoneme segmentation tasks. This factor, which we labeled ‘phonological awareness’, was conceptualised as an earlier developing, lower-level phonological construct. The associated tasks necessitated an understanding of and ability to identify phonemes within a word, but were considered to have a relatively low cognitive load, and were less demanding than the other tasks. The second phonological factor included two phoneme deletion tasks that were considered part of a later developing, higher-level construct, termed ‘higher-level’ phonological awareness. These tasks required children to be aware of and understand the sounds of their language, and concurrently maintain, manipulate and reintegrate those sounds to produce an accurate response. Because of the additional cognitive processes required, they were considered to have a higher cognitive load than the first construct identified.

Confirmatory factor analysis results for short-term memory and verbal working memory revealed that the best fitting model incorporated three-factors – a memory factor (which included both the short-term memory and working memory variables) as well as phonological awareness and ‘higher-level’ phonological awareness factors. This was unexpected given relatively consistent findings that short-term memory and working memory are separate, but related, cognitive abilities in school aged children (Alloway et al., 2006; S. E. Gathercole et al., 2004; Kail & Hall, 2001; Swanson, 2008). Several factors may have contributed to this unexpected result, including 1) Inclusion of short-term memory and/or verbal working memory tasks that were not sensitive enough; and 2) A wide age range of children included in the study (see detailed discussion below for consideration of age and development issues in research such as this).

Overall, our results closely align with an early study conducted by Yopp (1988), who examined the structure of phonological skills in a sample of children. Despite differences in study design and methodology (e.g. participant age, criteria for inclusion and tasks used), both studies identified separate phonological awareness and ‘higher-level’ phonological

awareness factors that were strongly correlated but differentiated by cognitive load. This result argues against phonological skills being conceptualised as a unitary factor.

In part, our results also supported Ramus et al.'s (2013) study, which highlighted the important role that cognitive load plays in the differentiation of phonological skills. Ramus et al. identified distinct higher and lower-level phonological factors in a mixed cohort of five- to 12-year-old children (typically developing and those with impaired language and/or reading abilities). In contrast to our results, the lower-level factor in Ramus et al.'s study was represented by tasks measuring phonological representations, while their higher-level factor was multifaceted and included a limited number of phonological tasks. It was represented by four tasks (phoneme substitution, rhyme detection, digit span, and rapid naming), which we would consider to assess short-term and working memory and phonological skills. The multifaceted nature of the higher-level factor in Ramus et al. (2013) makes it difficult to compare with our phonological awareness and 'higher-level' phonological awareness constructs, but is nevertheless useful to compare given the qualitative differences in load that were identified.

Another important difference between the two studies was the limited number of phonological awareness and 'higher-level' phonological awareness tasks included by Ramus. Seeing separation of phonological awareness and 'higher-level' phonological awareness constructs was unlikely without inclusion of a wider variety of phonological tasks. Limited tasks could also help explain discrepancies between our findings and those who have identified a unitary phonological construct. A number of published studies have included a small number of phonological tasks (Hagiliassis, Pratt, & Johnston, 2006; Muter, 1998; Rohl & Pratt, 1995), and in some cases all included tasks would be defined as only assessing phonological awareness (e.g. Vloedgraven & Verhoeven, 2007), meaning that identifying a

separation of phonological awareness and ‘higher-level’ phonological awareness factors would not be possible.

Consideration of age and level of cognitive development

Research examining cognition in childhood must consider the impact of age and cognitive development. The developmental trajectories of cognitive skills, such as short-term memory, working memory, phonological awareness and ‘higher-level’ phonological awareness, each follow a different age-related path. At each age, a child will present with differing levels of capacity in each skill. The less mature the skill, the more cognitively demanding a related task is likely to be. As a result, the cognitive load of a task will vary depending on the age of a child, and possibly development of other related skills.

Neuroimaging studies have supported this notion. Adults and children show similar patterns of brain activation on verbal working memory tasks, but the extent to which adults and children rely on various areas in response to increasing cognitive load is different (Vogan, Morgan, Powell, Smith, & Taylor, 2016). This was also explored by St Claire-Thompson (2010), who found that digit span backwards required significant cognitive effort and attentional resources and hence loaded onto a verbal working memory factor in children. In contrast, in adults the same task was less cognitively demanding and called upon primarily short-term memory resources (possibly due to factors such as improved attentional control, more efficient working memory processing systems, more mature brain networks, and/or better strategy use in adults). It is therefore possible that the same phonological task may represent a lower-level phonological awareness task in older children with more mature skills, while in younger children with less developed cognitive skills, the same task may have higher, more complex processing demands.

Level of cognitive development also needs to be considered alongside age. Children with developmental deficits may have less mature or well-developed skills despite their age.

For instance, in a group of children with reading disabilities or other neurodevelopmental disorders, children who are older and expected to have well developed short-term memory capacity may find lower-level tasks cognitively demanding and effortful. What would be considered a lower-level task for a typically developing child, may therefore actually be analogous to a higher-level task for a child with a neurodevelopmental disorder.

In the context of our results, age and cognitive development are important because this study included children aged six to 12 years old. Across this age range, there may be differences in the strength of relationships between cognitive skills such as short-term memory, working memory and phonological skill (e.g. S. E. Gathercole et al., 2004). The design of this study did not allow us to confirm age-related differences in these relationships (e.g. by examining sub-groups with more restricted age ranges), but potential differences could help to explain why two different models with different memory structure both provided an adequate fit to the data using raw scores.

3.4.2 Strengths and limitations.

The strengths of this study lie both in the nature of the study itself and the study design. Firstly, it is one of few recent studies to comprehensively consider cognitive load when examining the structure of phonological skills. The results clearly demonstrated that cognitive load is an important contributing factor to the structure of phonological skills in children suspected of learning disabilities. Further, the inclusion of a wide variety of phonological tasks (which varied in their cognitive load) has been lacking in many other studies in this area. This design feature maximised the likelihood of differentiating between phonological awareness and ‘higher-level’ phonological awareness constructs in this cohort. Tasks for this study were selected primarily on the basis of clinical assessment requirements rather than for theoretical or research-based reasons. Inclusion of more sensitive short-term memory and/or verbal working memory tasks may have improved construct differentiation.

The nature of the cohort was also important as it allowed an examination of how phonological, short-term memory and verbal working memory skills were related in children with learning disabilities. The clinical nature of the cohort, however, also limits generalisability of the results from this study as the structure of cognitive skills can differ between children with typical and atypical development. Age was identified as an important variable to consider when conducting research of this type. Though adjusting for age did not change the pattern of confirmatory factor analysis results, it did alter the strength and nature of the relationships between phonological awareness, ‘higher-level’ phonological awareness, short-term memory and verbal working memory.

3.4.3 Conclusion.

This study identified distinct phonological awareness and ‘higher-level’ phonological awareness factors that were differentiated by cognitive load. The strength and nature of the relationships between factors were impacted by age, particularly for short-term memory and verbal working memory. These findings have both theoretical and clinical implications. Theoretically, cognitive load has been somewhat neglected within the phonological literature to date, and this study highlights the importance of rectifying this. Understanding the relationships between phonological awareness, ‘higher-level’ phonological awareness, short-term memory and verbal working memory in children with learning disabilities allows development of more precise theoretical models, which can inform accurate diagnosis and treatment. Moreover, understanding that there are distinct types of phonological skills would assist clinicians to select appropriate tasks to assess both phonological awareness and processing skills. Interventions can then be tailored to the level and type of phonological deficit demonstrated by the child, thereby maximising the likelihood of positive outcomes. Longitudinal studies would be well suited to examine how age and development impact upon the cognitive load of different phonological tasks. It also remains unclear as to whether there

are differences in the structure of phonological skills in children with learning disabilities as compared to those without learning disabilities.

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Chapter 4 - The Nature of Phonological Impairments in Children with Specific Language Impairment (SLI) and Reading Disabilities (RD)

Preamble

Findings described in Chapter 3 provide evidence of distinct higher- and lower-level phonological skill constructs that are differentiated by cognitive load. Chapter 4 continues to investigate phonological skills, but shifts focus to describe the prevalence, severity and pattern of phonological impairments in three groups of primary school aged children (RD-only, SLI-only and RD+SLI). As outlined in the literature review, phonological impairments have been identified in children with reading disabilities and those with specific language impairment, and this chapter sought to clarify the nature of these impairments. Though not the primary focus of this chapter, examining the pattern of performance across a variety of higher- and lower-level phonological tasks was considered important given results described in Chapter 3.

This chapter contains a manuscript submitted to the Scientific Studies of Reading journal on 18/04/2019. The manuscript was inserted into Chapter 4 in its entirety and as such is formatted as per the journal's requirements.

Abstract

The nature of phonological impairments in children with reading disabilities and specific language impairments are not fully understood. We investigated the prevalence, severity and pattern of phonological impairments in school-aged children with reading disability-only, specific language impairment-only, and reading disability *and* specific language impairment. Children with both disorders generally had more prevalent and severe phonological impairments than those with a single disorder. Comparison of the single disorder groups revealed no differences in severity, prevalence, or pattern of phonological impairments. Findings suggest that phonological impairments in these neurodevelopmental disorders are similar and may represent a shared cognitive risk factor.

Keywords: Reading disability; specific language impairment; phonological skills, phonological impairment

4.1 Introduction

This article aimed to examine the nature of phonological impairments in children with reading disability and/or specific language impairment. Reading disability and specific language impairment are neurodevelopmental disorders which occur in approximately 5-10% of children (Shaywitz, Shaywitz, Fletcher, & Escobar, 1990; Tomblin et al., 1997). Reading disability is diagnosed when a child has significant, unexpected difficulty learning to read (Nithart et al., 2009), while specific language impairment is diagnosed in those who demonstrate impaired oral language (receptive, expressive or mixed) that cannot be attributed to other factors such as hearing loss or neurological injury (Leonard, 2014). Research suggests that specific language impairment and reading disability are best conceptualized as distinct disorders (Bishop & Snowling, 2004; Catts, Adlof, Hogan, & Weismer, 2005), although some commonalities are proposed. At a functional level, diagnoses of reading disability and specific language impairment often co-occur (Bishop & Snowling, 2004; Ramus, Marshall, Rosen, & van der Lely, 2013; Snowling et al., 2019). Australian statistics suggest that approximately 55% of children with a reading disability also meet criteria for a specific language impairment, while 51% of children with a specific language impairment also have a reading disability (G. M. McArthur, Hogben, Edwards, Heath, & Mengler, 2000). A multiple deficit framework is one way to consider this overlap. Multiple deficit models propose that neurodevelopmental disorders, such as reading disability and specific language impairment, arise from the interactions between multiple etiological risk and protective factors, which can be genetic and/or environmental. Risk factors combine to negatively impact on the development of multiple cognitive abilities, which in turn interact with one another to give rise to the behavioural manifestation of a disorder (Pennington, 2006; Peterson & Pennington, 2012). Etiological factors can be shared between

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different disorders, giving rise to a higher than expected level of co-occurrence between them. For instance, shared genetic factors have been suggested as one underlying reason for the frequent co-occurrence between reading disability and specific language impairment (e.g. Newbury et al., 2011). In research, this co-occurrence can lead to recruitment of heterogeneous groups of children if language *and* reading skills are not considered. This heterogeneity then makes it difficult to ascertain whether observed impairments (e.g. cognitive or language impairments) are specific to reading disability or specific language impairment or are impaired in both.

The overlap between reading disability and specific language impairment is also evident at a cognitive level, with phonological skills often impaired in children with reading disability and those with specific language impairment. Phonological skills are defined as an individual's ability to be sensitive to the speech sound structure of their language (Muter, 2005). This includes an awareness of phonological information, such as speech segments within words (e.g. syllables, onset-rime, phonemes), and the ability to process phonological material, such as the capacity to recognize, isolate and manipulate these speech segments (Anthony et al., 2002). Phonological skills are operationalized using a wide variety of tasks, including tasks of rhyme detection or production; syllable or phoneme identification; and phoneme blending, segmentation, or deletion. Phonological tasks differ in their level of cognitive load (Yopp, 1988), with higher-level phonological tasks (such as those requiring deletion of a phoneme from within a word) requiring more cognitive effort than lower-level phonological tasks (such as syllable identification tasks). One recent study suggests that the pattern of impairments with respect to higher- and lower-level skills may vary between children with specific language impairment and those with reading disability (Ramus et al., 2013).

4.1.1 Phonological skills in reading disability and specific language impairment.

When compared to their same aged peers and/or reading level controls, children with reading disability have consistently demonstrated phonological impairments (Kudo, Lussier, & Swanson, 2015; Larkin & Snowling, 2008; Melby-Lervag, Lyster, & Hulme, 2012; Stothard & Hulme, 1995). The identified phonological deficit likely results from imprecise or deficient phonological representations (i.e. the sound-based codes these children have developed and stored in long term memory is degraded in some way). Poor phonological representations negatively impact the ability to acquire adequate phonological skills, which can in turn lead to impaired reading (Fowler, 1991; Peterson & Pennington, 2012; Stanovich & Siegel, 1994; Sutherland & Gillon, 2007). Alternatively, phonological representations may be intact in children with reading disability, but they have difficulty accessing the representations efficiently (Ramus & Szenkovits, 2008). Phonological impairments are thought to be a primary underlying cause of reading difficulties in most children with reading disabilities (Ramus, Pidgeon, & Frith, 2003). However, consistent with the multiple deficit models discussed earlier, these impairments are not necessary or sufficient to cause reading difficulties in *all* children. Rapid automatized naming deficits and slow processing speed have been proposed as additional cognitive risk factors for reading disability (Moll, Gobel, Gooch, Landerl, & Snowling, 2016), while protective factors may include strong oral language and rapid automatized naming abilities, good reading instruction and/or well-tailored interventions, and the ability to be highly engaged and focused on tasks (see Catts, 2017 for a recent review)

Impaired phonological skills are also found in groups of children with specific language impairment (Bishop & Snowling, 2004; Briscoe, Bishop, & Norbury, 2001; Snowling, Bishop, & Stothard, 2000). As with reading disability, poor phonological representations and inadequate

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phonological skills have been hypothesised to underlie the impaired language acquisition in children with specific language impairment (Corriveau, Pasquini, & Goswami, 2007). Alternate theories propose that these language impairments are the result of linguistic (e.g. syntactic impairments) or non-linguistic deficits. These include slowed processing speed, procedural learning deficits and rapid temporal processing deficits (see Leonard, 2014 for a detailed discussion).

Knowledge of whether the prevalence, severity and pattern of phonological impairments are the same in reading disability and specific language impairment is limited, partly due to methodological differences within relevant investigations. Definitions of specific language impairment and reading disability (and therefore criteria used to recruit children to clinical groups) vary considerably between studies (G. McArthur & Castles, 2013), and there is a lack of consistency in the tasks used to assess phonological skills, which can make comparison between studies difficult. Studies have also recruited children with a reading disability or specific language impairment without determining their language and reading skill, which results in heterogeneous groups of children of unknown skill deficit level. To address this issue, a small number of studies have recruited more homogenous groups of children by measuring both language abilities and reading skills (Catts et al., 2005; Eisenmajer, Ross, & Pratt, 2005; Fraser, Goswami, & Conti-Ramsden, 2010; G. McArthur & Castles, 2013; Ramus et al., 2013; Snowling et al., 2019). Each compared an age matched control group to two ‘single deficit’ groups – a reading disability only group (RD-only) and a specific language impairment only group (SLI-only), as well as a ‘double deficit’ group which included children with both reading disability and specific language impairment (RD+SLI group).

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Table 1 provides a summary of methodology and key findings for each of these studies with regards to phonological impairments. Comparison of results from these studies revealed that with the exception of Eisenmajer et al. (2005) (who considered their findings inconclusive due to a lack of statistical power), children in the RD-only and RD+SLI groups showed impaired phonological skills.

Table 4.1

A summary of key research studies comparing phonological skills in children with RD-only, SLI-only and RD+SLI

	Age range	N (group)	Inclusion criteria RD and SLI	Phonological tests and criteria for impairment	Phonological skills impaired?			Magnitude of phonological skills deficit
					RD-only	SLI-only	RD+SLI	
Snowling, 2019¹	T1=3.5 years T2=4.5 years T3- T5=5.5-8 years	<u>RD only=21</u> <u>SLI only=38</u> <u>RD+SLI=29</u>	RD: <u>Composite score</u> <u>(average of age standardised SWRT and WIAT-II) Z<-1.5</u> SLI: <u>Composite score</u> <u>(average on CELF-4 expressive vocabulary & formulated sentences;</u> <u>TROG-2) Z=<-1</u>	T2 - Syllable and alliteration matching tasks. T2&T3 – Phoneme isolation task T3-T5 – Phoneme deletion task (YARC)	✓	✓	✓	<u>T2&T3:</u> (RD only = SLI only = RD+SLI) < Control <u>T4&T5:</u> RD only = SLI only RD only = RD+SLI RD+SLI<SLI only

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				Groupwise comparison with an age-matched control group				
Ramus, 2013	Clinical groups: 8-12 years Control group: 5-12 years	<u>13 (SLI-only); 21 (RD-only); 30 (RD+SLI)</u>	<u>RD or SLI</u> diagnosis; attendance at a special school/unit for children with RD or SLI <i>and</i> $Z \leq -1.5$ on a single word reading subtest (RD; WORD) or on one or more of the language tests administered (SLI; TROG-2, CELF-3 Sentence repetition, Test of word finding). *All children minimum score of 80 and average combined	Composite phonological skills score calculated from PhAB subtests (rhyme; spoonerisms; rapid digit naming) and a digit span task. Individual children considered impaired if scored more than 1.5SD below the mean Z score of the control group. Also conducted groupwise	✓	✓	✓	RD+SLI \leq (RD-only = SLI-only)

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			score of 85 ($Z \geq -1$) on two nonverbal cognition tests.	comparison with (non age-matched) control group.				
Eisenmajer 2005	7-12 years	25 (SLI-only); 21 (RD-only); 57 (RD+SLI)	<u>RD</u> : WIAT Reading $Z < -1$ <u>SLI</u> : Total Language Score on the CELF $Z < -1$ * Both groups PIQ score > 80 .	Groupwise comparison with a 'no learning disability' group using SPAT total score.	X	X	✓	RD+SLI \leq SLI-only RD-only = SLI-only RD-only = RD+SLI
Fraser, 2010	9-11 years	16 (SLI-only); 14 (RD-only); 21 (RD+SLI)	<u>RD</u> : Std reading score < 85 on one or more of: BAS-II single word reading ability, TOWRE sight word efficiency/ phonological coding efficiency. <u>SLI</u> : $Z < 85$ on at least two CELF-III subtests	Phoneme deletion; rhyme oddity; rhyme fluency Groupwise comparison with chronological age matched control group.	✓	✓	✓	RD+SLI \leq (SLI-only = RD-only) ²

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			(formulated sentences, sentence assembly, concepts and directions, semantic relations).					
Catts, 2005	Kinder ³ – Year 8	43 (SLI-only); 21 (RD-only); 18 (RD+SLI)	RD: Classified using Grade 4 data. At least 1SD below the mean on a composite word recognition measure and actual word recognition score more than 1SD below predicted word recognition score.	Syllable/Phoneme deletion (Kinder, grade 2, grade 4) Phoneme deletion (Year 8)	✓	✓ ⁴	✓	<u>Kinder:</u> (RD-only=SLI-only=RD+SLI) < Control <u>Grade 2:</u> (RD-only=RD+SLI) < SLI-only < Control
			SLI: Classified using Kindergarten data. $Z \leq -1.25$ on at least two of five language composites	Groupwise comparison with control group who had normal kindergarten language scores and normal reading achievement in Grade 4.	✓	X	✓	<u>Grade 4 and Year 8:</u> (RD-only = RD+SLI) < (SLI-only = control)

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			(vocabulary, grammar, narration, receptive language, expressive language), nonverbal IQ >85					
McArther & Castles (2013)	7-12 years	4-5 (SLI-only); 41-43 (RD-only); 17-19 (RD+SLI) ⁵	<p><u>RD</u>: Z<-1 on at least one of: letter-sound reading; whole word reading.</p> <p><u>SLI</u>: Z<-1 on at least one of: receptive language, recalling sentences⁶</p> <p>* Both groups nonverbal IQ > 85.</p>	<p>PhAB alliteration with pictures</p> <p>RD-only and RD+SLI: Groupwise comparison with 1) an age related normative mean score and 2) the other group's mean.</p> <p>SLI-only: Individual scores compared to 1) age based normative mean score and 2) the mean scores of RD-only</p>	✓	X	✓	<p>RD < SLI</p> <p>RD+SLI<RD⁷</p>

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				and RD+SLI. If most individuals significantly different, SLI-group deemed to differ from that mean.				
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BAS-II=British abilities scale II; CELF=Clinical evaluation of language fundamentals; PhAB=Phonological Assessment Battery, PIQ=Performance IQ; TOWRE=Test of word reading efficiency; TROG-2=Test for the reception of grammar 2; WIAT=Wechsler individual achievement test; WORD=Wechsler objective reading dimensions; York Assessment of Reading for Comprehension (YARC)

- ¹ Longitudinal study with five time points (T1-T5); SLI was termed developmental language disorder (DLD) in this paper. SLI is used here for comparison purposes.
- ² RD+SLI group performed more poorly than RD-only and SLI-only groups on phoneme deletion and rhyme oddity tasks, but not the rhyme fluency task.
- ³ Kindergarten refers to the first year of formal schooling.
- ⁴ While SLI-only group performed more poorly than the control group in both kindergarten and Grade 2, the group’s standard scores on the syllable/phoneme deletion task were approximately 90 (kindergarten) and 100 (Grade 2).
- ⁵ 73 children were allocated to RD-only, SLI-only and RD+SLI groups. Group n varied depending on which of four different analyses was examined.

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⁶ Different combinations of reading and language scores were used to create groups for four different analyses – 1) receptive-expressive language+ letter-sound reading, 2) receptive-expressive language+whole-word reading, 3) receptive language+letter-sound reading, and 4) receptive language+whole-word reading⁶

⁷ RD+SLI group performed more poorly than RD-only group on two of the four analyses (both receptive-expressive language group analyses).

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This demonstrates that phonological impairments, including poor ability to understand and manipulate segments of speech, occur in children with reading disabilities even without co-occurring language impairments. In contrast, while three of the six key studies identified phonological impairments in the SLI-only group (Fraser et al., 2010; Ramus et al., 2013; Snowling et al., 2019), another two studies found that this group was not impaired on phonological tasks (Eisenmajer et al., 2005; G. McArthur & Castles, 2013). In the remaining study children with specific language impairment had mild phonological difficulties in Kindergarten (first year of school) and Grade 2, but not in Grade 4 and Year 8 (Catts et al., 2005). These inconsistent findings were likely affected by several factors. Firstly, a recent study found that the prevalence of phonological impairments in a group of children with specific language impairment was lower than in a group of children with reading disability (Ramus et al., 2013). This finding suggests that phonological abilities in children with specific language impairment could be more variable than those in reading disability, leading to increased heterogeneity (and therefore more varied results) in specific language impairment groups. In addition, the definition and classification of specific language impairment and the measurement of language and phonological skills vary across these key studies. All of the studies used standardized language tests to measure language functioning, but the combination of subtests used was different in each of them. Further, some of the studies used subtest scores independently (e.g. requiring poor performance on one or more of the subtests administered) (Fraser et al., 2010; G. McArthur & Castles, 2013; Ramus et al., 2013), while others used composite or total language scores to determine impairment (Catts et al., 2005; Eisenmajer et al., 2005; Snowling et al., 2019). The proportion of children with different types of language deficits, such as expressive versus receptive language, may also have varied across studies.

Inconsistencies such as these make it difficult to compare results across studies and may contribute to the varied results for the SLI-only group.

Differences in the magnitude of phonological impairment was also found. The RD+SLI group generally exhibited significantly larger deficits than the SLI-only group (Catts et al., 2005; Eisenmajer et al., 2005; Fraser et al., 2010; G. McArthur & Castles, 2013; Ramus et al., 2013; Snowling et al., 2019) and RD-only group (Fraser et al., 2010; G. McArthur & Castles, 2013; Ramus et al., 2013). Comparisons of the RD-only and SLI-only groups are inconsistent. Of the four studies which found impaired phonological skills in children with SLI-only, three found that the magnitude of impairments was not significantly different in groups of RD-only and SLI-only (Fraser et al., 2010; Ramus et al., 2013; Snowling et al., 2019). In contrast, the fourth identified greater magnitude of phonological impairments in the RD-only group in Grade 2 children, but no difference in kindergarten children (Catts et al., 2005). The inconsistencies indicate that additional investigation is required to better understand the magnitude of phonological deficits in children with RD-only and SLI-only.

The pattern of phonological impairments with regards to higher- and lower-level phonological skills is unknown, partly due to limited research investigating the relationship between cognitive load and phonological skills. There is continued debate as to whether distinct lower- and higher-level phonological skills can be identified, though several studies now support this proposition (Lee, Ross, Nadebaum & Testa, under review; Ramus et al., 2013; Yopp, 1988). Of the five key studies described above, just one (Ramus et al., 2013) included discussion of lower- and higher-level phonological skills (in the context of composite scores). Higher-level skills were represented by phonological (e.g. rhyme and spoonerisms tasks) and other cognitive skill tasks (e.g. digit span), while lower-level skills were represented by tasks thought to measure

phonological representations (e.g. non-word discrimination). The study found different patterns of performance between the RD-only and SLI-only groups; the SLI-only group were equally impaired on lower- and higher-level phonological composites, while the RD-only group performed more poorly on the higher-level than the lower-level composites (Ramus et al., 2013). Given that Ramus et al. utilized composite scores to investigate pattern of performance, it is not clear whether individual phonological tasks are differentially impacted in reading disability and specific language impairment.

4.1.2 The present study.

This study aimed to further investigate the prevalence, severity and pattern of phonological impairments in groups of primary school aged children with RD-only, SLI-only, or RD+SLI. We expected each of the clinical groups to demonstrate impaired phonological skills, as indicated by a mean phonological skill score within the impaired range. The magnitude of the phonological impairment was predicted to be higher (more severe) in the RD+SLI group than both the RD-only and SLI-only groups. We tentatively hypothesized no difference in severity of phonological impairments between the RD-only and SLI-only groups, while noting the inconsistent results of past research in this area. Following a recent finding of increased diversity of phonological skills within groups of children with specific language impairment as compared to groups of children with reading disability (Ramus et al., 2013), we expected a lower proportion of children in the SLI-only group would demonstrate phonological impairments compared to the RD-only group. Understanding the pattern of performance on a variety of higher- and lower-level phonological tasks was also of interest and was explored using profile analysis. Although this analysis was primarily exploratory in nature, it was expected that the

profiles of the RD-only, SLI-only and RD+SLI groups would be significantly different from each other (i.e. not parallel).

4.2 Method

4.2.1 Participants.

Participants were selected from a group of primary school aged children referred to and assessed at a learning difficulties clinic between 1996 and 2008. The children were English speaking and ranged in age from six to 12 years old (grades 1 to 6). No child had any known neurological or psychiatric condition or significant emotional/behavioural difficulties. Language skills were assessed using The Clinical Evaluation of Language Fundamentals, Revised (CELF-R; Semel, Wiig, & Secord, 1987) or The Clinical Evaluation of Language Fundamentals-3 (CELF-3; Semel, Wiig, & Secord, 1995). Children who obtained a receptive or expressive language score less than or equal to 85 (i.e., at least one standard deviation below the mean) on either version of the Clinical Evaluation of Language Fundamentals were considered to have a language impairment and were placed in the SLI-only group or RD+SLI group (depending on whether they also met criteria for a reading disability). The word reading subtest of the Wechsler Individual Achievement Test (WIAT; Wechsler, 1993) was used to measure single word reading skills. Participants were classed as having a reading disability if they performed more than one grade level below expected on this subtest. Only children with valid scores on both the WIAT single word reading skill subtest and the CELF receptive or expressive language subtests were included in this study (N=123).

4.2.2 Materials.

Non-verbal intellectual functioning was assessed using the Performance IQ score from the Wechsler Intelligence Scale for Children, Third edition (Wechsler, 1991). Phonological skills were assessed using the Sutherland Phonological Awareness Test (SPAT; Neilson, 1995). This standardised test battery consists of 13 subtests as follows: The *syllable counting* subtest required each child to identify, through tapping, the number of syllables in a word (e.g. helicopter) which was provided orally by the examiner. In the *rhyme detection* subtest each child was provided with a simple picture and asked to pick another picture (out of two) that rhymed it. Oral descriptions of all three pictures were provided by the examiner (e.g. “This one is ‘pig’. You have to choose the picture that rhymes with ‘pig’.....’dig’ or ‘cup’?”). In the *rhyme production* subtest each child listened to two words that rhymed, and was then asked to make up a word that also rhymed (e.g. cat, fat,.....?). The *onset identification* subtest and *final phoneme identification* subtests required each child to identify the sound at the beginning or end of a word respectively (e.g. “What sound does sun begin with?” or “What is the last sound you hear in the word boot?”). In the *segmentation 1* and *segmentation 2* subtests the child listened to a word provided orally by the examiner, and was then asked to segment it into its corresponding phonemes. In addition, the child was presented with a stimulus sheet containing five numbered boxes, and was asked to tap one box for each sound they produce. To score a point for a correct response, the child had to pronounce all phonemes correctly as well as tap the correct number of boxes. The *blending* subtest required the blending together a number of sounds provided by the examiner. In the *initial phoneme deletion*, *deletion of first phoneme*, and *deletion of second phoneme* subtests, the child was asked to delete a sound from a word and say the word that was left. These three subtests differed from each other with regards to the position (e.g. initial or second phoneme)

and the type of phoneme (e.g. a consonant cluster or not) that was to be deleted. Each of the above subtests included an example item, practice item and four test items. Score range was 0-4 and the subtests represented a mix of lower- and higher-level phonological tasks (Citation removed for blinding purposes). In the *non-word reading* and *non-word spelling* subtests the child was provided with nonsense words (in written form for reading, and orally for spelling) that they were asked to read or spell. Score range for these subtests was 0-7.

Raw scores were calculated for each SPAT subtest and combined to form a total SPAT raw score. Using normative data (where available), a grade adjusted Z score was then created. A child's phonological skills were considered to be impaired if the grade adjusted Z score was less than -1. For children in Grade 4-6 normative data was not provided in the SPAT manual. For these older children (who represented a significant proportion of children assessed at the learning difficulties clinic) there was no alternative phonological skills test battery available with appropriate normative data. As such, older children also completed the SPAT, and normative data for Grade 3 children was used for children in Grade 4-6. If these older children scored more than one standard deviation below the mean score for Grade 3 children, they were classed as having impaired phonological skills.

4.2.3 Procedure.

This project was reviewed and approved by Western Health (HREC/11/WH/79) and Monash University (2012000503) HRECs. At the time of assessment, each parent/guardian provided consent for data to be used for future research. Once accepted into the clinic each child underwent an audiology assessment and was individually assessed by a multidisciplinary team of allied health professionals (clinical neuropsychologist, speech pathologist and occupational therapist) over a two-day period. Relevant psychometric measures were administered and scored

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in accordance with the test manuals. Each child's parent/guardian was interviewed by a social worker (to confirm and further explore any identified difficulties) and a number of questionnaires were provided to each child's parent/guardian and school. Following the assessment, all results were entered into a database and relevant data was retrospectively accessed for the purposes of this study.

4.2.4 Statistical analyses.

A chi-square test was used to compare the proportion of children in each group who met criteria for a phonological impairment. Using grade adjusted Z-scores, a between-subject ANOVA was then used to compare the phonological skills of each of the three groups, with Tukey post-hoc tests used to examine pairwise comparisons. Z-scores were also used in a profile analysis which examined the performance of the three groups on individual subtests of the SPAT.

4.3 Results

Of the 123 children with valid scores on the WIAT single word reading and CELF receptive and expressive language subtests, 16 children did not meet criteria for a reading disability or a specific language impairment, and as such these children were excluded from the analyses. In addition, a further nine children were excluded from the analyses as they did not complete the SPAT during their assessment. Six children had non-verbal intellectual functioning scores below 70. A decision was made to include these children in the analyses based on our aim to include a cohort of children that accurately represent typical referrals to the Learning Difficulties Clinic. As such, 98 children were allocated to a relevant clinical group (RD-only=27; SLI-only=17; RD+SLI=54) and were subsequently included in all analyses.

Table 4.2 shows the mean scores for age, academic, cognitive and language measures for the three clinical groups. One-way between subject ANOVAs found significant group differences on all measures ($p=.02$ for PIQ, $p=.01$ for age, and $p<.01$ for all other measures). Tukey's post-hoc analyses demonstrated that the SLI-only group was significantly younger than the RD-only group ($p=.007$). For FSIQ and PIQ, the RD-only group scored significantly higher than the SLI-only group ($p=.01$ and $p=.04$ respectively), while for VIQ the difference between these groups approached significance ($p=.053$) with the RD-only group again scoring higher. The RD-only group also scored significantly higher than the RD+SLI group on FSIQ and VIQ ($p=.001$ for both). Expressive, receptive and overall language scores were also significantly higher in the RD-only group in comparison to SLI-only ($p<.001$) and RD+SLI ($p<.001$) groups. For WIAT reading scores, the SLI-only group scored significantly higher than both RD-only ($p=.01$) and RD+SLI ($p<.001$) groups, while the RD-only group scored significantly higher than the RD+SLI group ($p=.002$). Phonological skills, as measured by the SPAT grade adjusted Z-scores, were significantly lower in the RD+SLI group in comparison to the RD-only ($p<.001$) and SLI-only ($p=.005$) group.

The mean grade adjusted SPAT Z-scores showed that the RD+SLI group, on average, performed more than 2 standard deviations below the grade adjusted mean, which falls clearly into the impaired range (defined as 1SD below the mean). In contrast, the RD-only and SLI-only groups did not show phonological impairments as per this definition, performing almost one standard deviation below the grade adjusted normative mean. Two subsequent analyses were conducted in order to determine whether mild phonological impairments were present in these groups. Single group t-tests showed that both of these groups performed more poorly than the SPAT normative group, $t(26)=3.41$, $p=.002$ and $t(16)=2.82$, $p=.012$ respectively. Next, all Grade

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4-6 children (n=35) were temporarily removed from the analyses to examine whether the lack of SPAT normative data for these older children impacted results. It was thought that the conservative criterion used to designate Grade 4-6 children as impaired/not impaired may have led to a proportion of these children being labelled unimpaired despite some degree of phonological impairment. Indeed, removal of Grade 4-6 children from within the RD-only and SLI-only groups resulted in both groups' average phonological score shifting to within the impaired range ($Z=-1.01$ and -1.06 respectively).

Table 4.2

Mean (SD) demographic, academic, intellectual, phonological and language scores for clinical groups

Mean (SD)	RD-only (n=27)	SLI-only (n=17)	RD+SLI (n=54)
Age*	9.34 (1.37)	8.14 (1.21)	8.91 (1.23)
FSIQ**	92.26 (11.13)	82.94 (12.20)	83.69 (8.64)
VIQ**	93.11 (11.45)	85.71 (11.57)	83.98 (8.92)
PIQ*	93.22 (12.69)	83.35 (16.56)	86.30 (11.14)
Total language score**	96.74 (8.87)	71.18 (9.80)	73.33 (8.24)
Receptive language score**	97.19 (8.68)	71.94 (13.48)	76.63 (10.87)
Expressive language score**	97.59 (11.21)	73.53 (10.39)	73.63 (7.91)

WIAT Reading score**	84.04 (5.71)	90.88 (10.80)	77.89 (6.94)
Overall SPAT grade adjusted Z-score**	-0.82 (1.25)	-0.88 (1.29)	-2.23 (1.68)

FSIQ=Full-scale IQ; VIQ=Verbal IQ; PIQ=Performance IQ; WIAT=Wechsler individual

achievement test; SPAT=Sutherland phonological awareness test; * p<.05, **p<.01

As seen in Figure 4.1, 37% of children with RD-only and almost half of children (47.1%) with SLI-only had impaired phonological skills, compared to approximately three-quarters (75.9%) of those in the RD+SLI group. A chi-square test of independence found a significant difference in the prevalence of impaired phonological skills across the three groups ($\chi^2(2)=12.85$, $p=.002$). Post-hoc pairwise chi-square tests performed with a Bonferroni adjusted alpha=.017 showed that the RD+SLI group had significantly higher prevalence of phonological impairment than the RD-only group ($p=.001$) but a trend only was evident in comparison to the SLI-only group ($p=.025$). There was no significant difference in prevalence between the RD-only and SLI-only groups ($p=.51$).

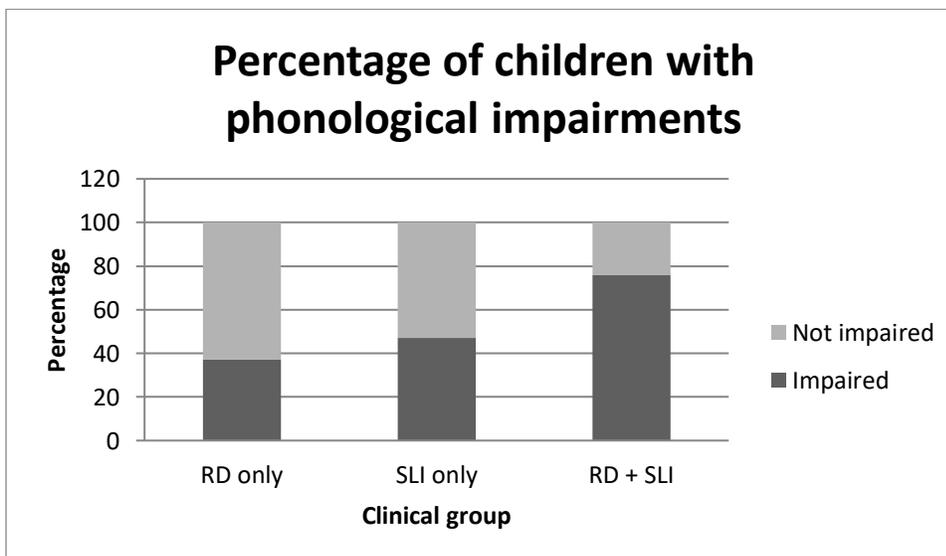


Figure 4.1. Percentage of children with phonological impairments.

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To examine the pattern of performance on individual subtests of the SPAT, a profile analysis was conducted. Eleven SPAT subtests were considered for inclusion in this analysis (syllable counting, rhyme detection, rhyme production, onset identification, final phoneme identification, segmentation 1, blending, initial phoneme deletion, segmentation 2, deletion of first phoneme, and deletion of second phoneme). Each variable was examined for univariate outliers and violations of normality. Skewness and kurtosis values were examined and five variables (rhyme detection, rhyme production, final phoneme identification, identification of onset, blending) were identified to have very extreme skewness and kurtosis values in one or more clinical groups (George & Mallery, 2010), with values ranging from two to 26.6. Due to almost all participants within some groups scoring the same value, transformation would not have improved the approximation of normality in any of these variables. These variables were therefore excluded from the analysis. Within the retained variables, two cases were identified as extreme univariate outliers, with Z-scores of -3.69. However, given that the Z-scores equate to an acceptable raw score on this subtest (one out of four) and re-running the analysis without these cases did not alter results, a decision was made to retain them in the analysis. No multivariate outliers were detected using Mahalanobis distance and no Cook's distance values were greater than one. A visual inspection of the bivariate scatterplots suggested approximate linear relationships among the dependent variables. While the assumption of variance-covariance was met, the assumption of sphericity was violated and to adjust Huynh-Feldt adjusted results were reported.

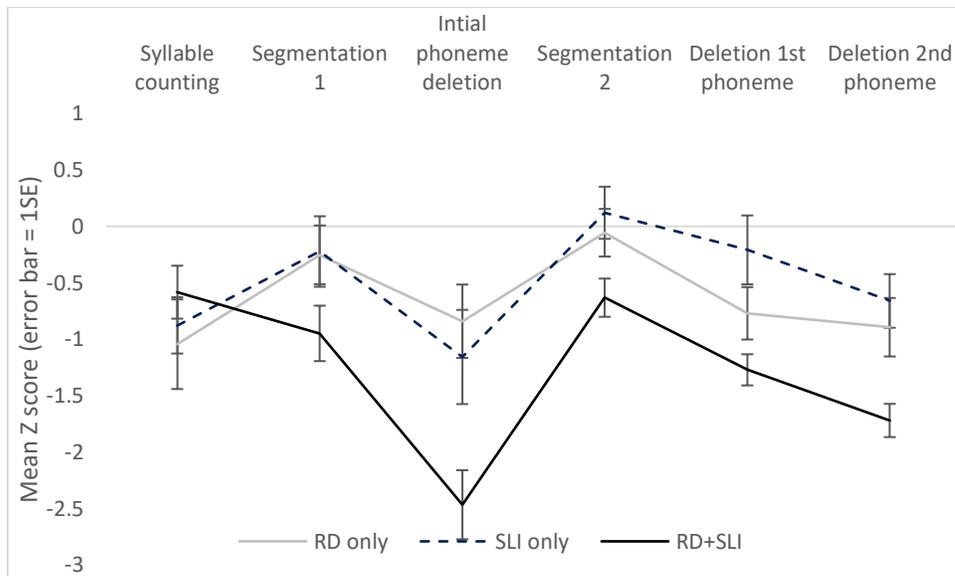


Figure 4.2. Mean (SE) Z-scores for each clinical group.

Profile analysis (see Figure 4.2) was conducted using a mixed model ANOVA, which found an overall significant interaction effect, $F(7.65,363.24)=3.12, p=.002$, demonstrating non-parallelism. To examine which groups displayed differences in their pattern of performance a series of three post-hoc profile analyses were conducted, applying a Bonferroni adjusted p-value of .017. A significant interaction was only identified between the RD-only group and the RD+SLI group (.001), with no other significant interaction effects evident.

4.4 Discussion

The aim of this study was to investigate the prevalence, severity and pattern of phonological impairments in primary school aged children with RD-only, SLI-only, and RD+SLI. We firstly hypothesized that each of the clinical groups would demonstrate impaired phonological skills. This hypothesis was partially supported. The performance of the RD+SLI group on phonological tasks fell well within the impaired range, with these children also more

likely to demonstrate phonological impairments than the RD-only group. A non-significant trend was found for the SLI-only group. Impairments in children with RD+SLI were significantly more severe than both the RD-only and SLI-only groups. These findings are consistent with a growing body of research demonstrating that when compared to children with a single deficit (i.e. RD-only or SLI-only), children with both reading and language impairments have more prevalent and severe phonological impairments. Along with substantial phonological deficits, the RD+SLI group displayed poorer reading skills than either of the single deficit groups. The association between more severe phonological impairments and increased reading difficulties is consistent with research showing a significant correlation between reading and phonological skills (Melby-Lervag et al., 2012).

The RD-only and SLI-only groups also exhibited mild phonological impairments, performing more poorly on phonological tasks than the SPAT normative group of children. However, their performance was slightly higher than our predetermined cut off for impairment. Removing Grade 4 to Grade 6 children from the RD-only and SLI-only groups resulted in both groups phonological skill mean scores moving into the impaired range. This suggests that the lack of normative data for children in Grade 4 to Grade 6 and the subsequent use of Grade 3 norms for these children resulted in our impairment criteria not quite being met.

Group comparisons revealed no difference in the severity of phonological impairments in the SLI-only and RD-only groups, which supported our hypothesis and was consistent with results from most previous research studies (e.g. Fraser et al., 2010; Ramus et al., 2013; Snowling et al., 2019). Comparisons between the RD-only and SLI-only groups also revealed no difference in the prevalence of phonological impairments. This is in contrast to Ramus et al. (2013), who was the only key study to provide the prevalence data for phonological skill

impairments in children with SLI-only as compared to RD-only. They found a lower prevalence of phonological impairments in children with SLI-only as compared to those with RD-only.

Profile analysis aimed to extend the above findings by comparing the performance of the groups on a selection of phonological tasks. This analysis was largely exploratory in nature. It was included in the context of results from Ramus (2013), which showed that children with RD-only and those with SLI-only showed different patterns of performance on higher- and lower-level phonological composite scores. We were interested in examining whether the groups had different patterns of performance across a variety of lower and higher-level phonological tasks. Profile analysis demonstrated that there were no significant differences between profiles of the RD-only and SLI-only groups. The findings suggest that the two single deficit groups have similar patterns of performance across the phonological subtests included in the analysis. In contrast, the profiles of the RD+SLI and RD-only groups differed from one another, indicating that the overall pattern of performance across the six subtests was different between these two groups. A visual inspection of the results suggested that this difference may be driven by performance on the syllable counting subtest, with the RD+SLI group not performing more poorly than the RD-only group on this task. The syllable counting test was the only one of the six subtests which examined phonological abilities at the level of the syllable rather than the phoneme. These results suggest that in children with RD, having an additional language impairment does not further impair performance on this syllable-based phonological task.

Methodological differences may help explain the differences in results between our study and Ramus et al. (2013). Firstly, Ramus et al. used composite scores to identify different patterns of performance across lower- and higher-level phonological abilities. The profile analysis in this study used individual subtest scores from a variety of individual phonological tasks to examine

patterns of performance across lower- and higher-level abilities. Furthermore, the current study utilized tasks designed to measure the ability to process phonological material, including the capacity to recognize, isolate and manipulate speech segments. Ramus et al. calculated a phonological skills composite score which was multifactorial in nature, using phonological tasks as well as tasks thought to measure short-term memory and working memory (digit span). The nature of the phonological skills composite score in Ramus et al. (2013) suggests that this score is likely to be measuring something different than the phonological skills examined in this study, which makes comparison between the studies difficult.

The findings in this study can be considered in the context of multiple deficit models of neurodevelopmental disorders. For reading disabilities, these models propose that the combination of genetic and environmental etiological risk and protective factors impact the development of cognitive abilities. Reading impairments then result from the interactions between multiple cognitive risk factors, rather than from a phonological impairment alone (Peterson & Pennington, 2012). Multiple deficit models also suggest that risk factors can be shared between multiple neurodevelopmental disorders. These shared risk factors can explain higher than expected levels of comorbidity between particular disorders. Findings from a number of studies have identified patterns of cognitive and genetic risk factors that are shared between reading disability, specific language impairment and speech sound disorders (see Pennington & Bishop, 2009 for a review). The similarities in the severity and pattern of phonological impairments of the RD-only and SLI-only groups in this study is consistent with the hypothesis that poor phonological skills may be a shared cognitive risk factor between the two disorders.

Our results also emphasize that knowledge of the nature of phonological impairments in groups of children is not sufficient in and of itself to explain patterns of reading and language

abilities. For instance, presence of a phonological impairment was not sufficient to cause a reading disability as children in the SLI-only group also showed phonological impairments. Furthermore, SLI-only and RD-only groups, who demonstrated similar phonological impairments, obviously (by design) had significantly different patterns of reading and language abilities. These findings provide further support for multiple deficit hypotheses, suggesting that other, non-phonological, cognitive impairments may interact with poor phonological skills to impact upon the trajectory of reading and language abilities in children. A potential contributor is rapid automatized naming ability. Bishop et al. (2009) found that rapid automatized naming ability affected the developmental outcome of children who had poor phonological skills at age four. Poor phonological skills and rapid naming led to a specific language impairment and reading disability, whilst poor phonological skills within intact rapid naming lead to a reading disability only. Their findings emphasize the importance of assessing phonological skills *and* other, related, cognitive skills in any child suspected of having literacy or language disorders. Assessment of phonological skills alone is unlikely to provide a thorough understanding of the cognitive risk and protective factors that are contributing to an individual's presentation.

4.4.1 Strengths and limitations.

The strengths of this paper lie in its design, which ensured that both language and reading skills were assessed in order to create groups of children with reading disability (with and without language impairments) and specific language impairments (with and without reading deficits). This type of methodology is critical as it allows comparison of more homogenous groups of children. In addition, the design allowed comparison of groups not only in terms of prevalence and severity of phonological impairments, but also in terms of the pattern of performance across a variety of phonological tasks. Inclusion of an extensive phonological skills

test battery made up of both higher- and lower-level phonological subtests was a further strength of the study. However, the test battery administered also had limited normative data available (covering Prep-Grade 3 only), which likely impacted upon the observed magnitude of phonological impairments in each of the groups. A further limitation of the SPAT (as used in this population) was that ceiling effects were identified in a number of subtests. This led to the exclusion of these subtests, which limited the scope of the profile analysis and subsequent discussion regarding the pattern of phonological impairments in reading disability and specific language impairment. The design of the study means that a control group was not included in this study.

4.4.2 Future research.

Future research in this area would benefit from consensus of definitions and diagnostic criterion for reading disability and specific language impairment. Given the conflicting results described earlier, research to clarify the prevalence and pattern of phonological impairments in children with RD-only and/or SLI-only would be beneficial. Inclusion of a broad array of higher- and lower-level phonological tasks is critical to further elucidate prevalence estimates. Researchers should work to ensure that phonological tasks selected for inclusion have adequate normative data, and take steps to minimize issues such as ceiling effects. Testing of multiple deficit models using multivariate analysis may be beneficial in explaining the co-occurrence between reading disability and specific language impairment in terms of shared and unique cognitive (e.g. rapid automatized naming, short-term memory, working memory) and other (e.g. genetic) risk and protective factors. In addition, longitudinal research would be useful to examine whether the prevalence, severity and pattern of phonological impairments in reading disability and specific language impairment vary across time.

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**Chapter 5 - Characteristics of Children Referred to a Multidisciplinary Learning
Difficulties Clinic for Assessment**

Preamble

Chapters 3 and 4 described an in-depth investigation of the structure of phonological skills and how these skills are impacted upon in reading disability and specific language impairment. Chapter 5 focuses on learning disabilities more broadly. It follows on from research described within the literature review which highlights the complex array of difficulties experienced by children with learning disabilities. These difficulties are broad in nature and can span multiple academic and non-academic domains.

This chapter aimed to characterise the academic, cognitive, language, motor, emotional and behavioural difficulties experienced by children referred to an Australian Learning Difficulties Clinic for assessment by a multidisciplinary team of allied health professionals. Chapter 5 incorporates a manuscript submitted to the Journal of Learning Disabilities on 18/04/2019. It has been inserted into this thesis in its entirety and in the format required for manuscript submission.

Abstract

Accurate assessment of children with suspected learning disabilities is a challenging process. In children with learning disabilities, academic impairments often span multiple academic subjects and the comorbidity of cognitive, language, motor, emotional and behavioural difficulties is significant. Multidisciplinary assessment can provide a detailed and useful evaluation of these children by enabling the assessment of multiple domains. This study conducted a retrospective analysis of clinical information gathered from 326 primary school aged children referred to an Australian Learning Difficulties Clinic. Each child underwent multidisciplinary assessment conducted by a neuropsychologist, speech pathologist, occupational therapist and social worker. Results identified a range of academic, cognitive, language, motor, behavioural and emotional impairments experienced by these children. While approximately 65% of children met criteria for multiple learning disabilities, almost one in five children did not meet criteria for any learning disability at the time of assessment. In children that did meet criteria for at least one learning disability, environmental (e.g. family functioning) and non-environmental (e.g. younger age, lower IQ) risk factors increased the likelihood of them experiencing comorbid language, motor, behaviour and emotional impairments. The findings suggest that the multidisciplinary assessment process supported the complex diagnostic requirements of children referred to the Learning Difficulties Clinic.

Keywords: Learning disabilities, multidisciplinary, phonological skills, language, intellectual functioning, comorbidity

5.1 Introduction

Learning disability is a heterogeneous term used to describe children with significant difficulties in the acquisition of literacy and numeracy skills (Louden et al., 2000).

Approximately 4-10% of school aged children experience a learning disability (Lagae, 2008; Louden et al., 2000), with reading disabilities being the most common diagnosis (Shaywitz, 1998). It is common for children with an impairment in one academic domain to also have impairments in other academic domains (Landerl & Moll, 2010), though Australian data describing the comorbidity between learning disabilities is lacking. Additional impairments in non-academic areas (such as cognitive, language, motor, emotional and/or behavioural difficulties) are also observed.

5.1.1 Non-academic impairments in children with learning disabilities.

Aspects of cognitive functioning, including short-term and working memory, visuospatial abilities, and executive functioning have been found to be impaired in children with learning disabilities (Brosnan et al., 2002; Geary, 2011; Geiger et al., 2008; Rourke, 1993; Willcutt et al., 2013), although verbal and nonverbal intellectual functioning skills are generally age appropriate (De Clerq-Quaegebeur et al., 2010; Giofre & Cornoldi, 2015; Styck & Watkins, 2014). Language abilities, including phonological skills, are also often delayed in children with learning disabilities (McArthur, Hogben, Edwards, Heath, & Mengler, 2000). For example, many children who meet criteria for a reading disability also display language impairments and vice versa (Mattis, 1978; McArthur et al., 2000). Children with reading (Melby-Lervag, Lyster, & Hulme, 2012) and language disabilities (Fraser, Goswami, & Conti-Ramsden, 2010) also often experience comorbid phonological skill deficits.

Impairments in motor domains are also reported; fine and gross motor skills in children with learning disabilities can be significantly poorer than in their typically developing peers (Vuijk, Hartman, Mombarg, Scherder, & Visscher, 2011; Westendorp,

Hartman, Houwen, Smith, & Visscher, 2011). Similarly, emotional and behavioural difficulties are frequently seen in children with learning disabilities (Mugnaini, Lassi, La Malfa, & Albertini, 2009; Shalev, Auerbach, & Gross-Tsur, 1995). In a study of 1013 children and adolescents, Willcutt, Petrill et al. (2013) found that those with a reading and/or a math disability had more internalising and externalising psychiatric symptoms (e.g. attention deficit hyperactivity disorders, conduct disorder, depression and/or generalised anxiety disorder) than those with no reading or math disability.

5.1.2 Assessment of learning disabilities

In Australia, there are several frameworks in which learning disability assessments are undertaken, including paediatrician-led assessment or assessment via a multidisciplinary assessment team (Mittal, Sciberras, Sewell, & Efron, 2014). Within a paediatrician-led assessment model, the paediatrician may exclude or manage medical or behavioural comorbidities, advocate to ensure appropriate community services are involved, provide health education to parents and school staff, co-ordinate a comprehensive assessment process, and/or monitor longitudinal progress (Oberklaid, 1984). Paediatricians can also refer to individual colleagues (e.g. psychologists and/or speech pathologists) or for multidisciplinary assessment for further evaluation where necessary.

Multidisciplinary assessments involve a variety of medical/allied health practitioners within a setting working together to complete an assessment. This type of assessment can facilitate an understanding of complex problems that may not be adequately addressed by a single discipline alone and provide a comprehensive and accurate evaluation where different clinicians can each provide their own expertise and perspective on an issue (Choi & Pak, 2006; Hendriksen et al., 2007). Multidisciplinary assessments include the gathering of information (via interviews and/or questionnaires) from parents and teachers, and the obtaining of social, developmental, medical and educational histories. The child is asked to

complete psychometric tests, which may incorporate measures of academic achievement, cognitive functioning, language functioning, motor skills, and social-emotional and behavioural components (Handler & Fierson, 2011).

Empirical data describing the characteristics of children referred to multidisciplinary clinics is limited (Hendriksen et al., 2007). By concentrating on only one or two types of difficulties experienced by children with learning disabilities, past research has tended to provide a narrow view of the prevalence and nature of non-academic difficulties in these children. Moreover, prevalence of comorbidities between learning disabilities themselves is poorly understood. The current paper aimed to address literature gaps by characterising the academic, cognitive, language, emotional, behavioural, and motor characteristics of a cohort of children referred to a multidisciplinary Australian Learning Difficulties Clinic for assessment. In addition, this paper aimed to identify potential risk factors for non-academic difficulties; specifically, language impairments, motor impairments, emotional difficulties and behavioural difficulties, in children who met criteria for at least one learning disability diagnosis.

5.2 Method

5.2.1 Service description

The Learning Difficulties Clinic was a multidisciplinary clinic specialising in the assessment of primary school aged children with suspected learning difficulties. The clinic was located at Sunshine Hospital which is a publically funded hospital servicing the western suburbs of Melbourne. Entry into the clinic required a referral by a paediatrician or school. Referrals were not accepted if the child had any known neurological or psychiatric condition, intellectual disability, or significant emotional or behavioural difficulties that would preclude valid assessment of learning disabilities.

5.2.2 Participants and general procedure

This project was reviewed and approved by Western Health (HREC/11/WH/79) and Monash University (2012000503) HREC's. At the time of assessment, parents/guardians provided consent for clinical information and results to be used in future research studies. Participants were 326 children representing consecutive referrals to the Learning Difficulties Clinic between 1996 and 2008. Clinical information gathered from all children assessed during this period was retrospectively analysed in this study. After being accepted into the clinic each child's parent/guardian completed a number of questionnaires, which asked for information regarding family history, birth, development and health history, school history, and details of the child's current academic, social, emotional, behavioural and cognitive functioning. The child's school was also provided with relevant questionnaires, which were generally completed by the classroom teacher. Each child underwent an audiology examination and was individually assessed by a clinical neuropsychologist, speech pathologist and occupational therapist using psychometric tests administered and scored in accordance with the test manuals. A social worker interviewed each child's parent/guardian to confirm and further explore any identified difficulties. Following the assessment, all team members and a school representative(s) met, discussed results, diagnoses and recommendations for home and school, and each child's parent/guardian was provided feedback.

5.2.3 Materials

During the 12-year data collection period, revised versions of several psychometric tests were introduced. Unless otherwise stated, children completed the test versions that were current at the time of their assessment. The following details describe the psychometric tests used.

The Wechsler Individual Achievement Test (WIAT: Wechsler, 1993) and the Wechsler Individual Achievement Test, Second Edition (WIAT-II: Wechsler, 2002) were used to measure reading, spelling and mathematical abilities. Participants were classed as having a specific learning disability (in maths, spelling and/or reading) if they performed more than one grade level below expected on the relevant WIAT/WIAT-II subtest (numerical operations, spelling, and word decoding respectively).

The Wechsler Preschool and Primary Scale of Intelligence, Revised (WPPSI-R: Wechsler, 1989), the Wechsler Intelligence Scale for Children, Third edition (WISC-III: Wechsler, 1991), Fourth edition (WISC-IV: Wechsler, 2003), or WISC-IV Australian Standardised Edition (Wechsler, 2005) were used as measures of intellectual functioning. Where possible, Verbal Comprehension, Perceptual Reasoning/Perceptual Organisation, Working Memory/Freedom from Distractibility, Processing Speed and Full Scale IQ scores were calculated. Age-adjusted normative scores on each scale have a mean of 100 and standard deviation of 15. For each scale, approximately 2% of the standardisation sample scored 2 standard deviations below the mean (i.e. below 70), falling within the 'Extremely Low' range.

The Clinical Evaluation of Language Fundamentals (CELF) was used to measure language skills. Participants completed the CELF-R (Semel, Wiig, & Secord, 1987), CELF-3 (Semel, Wiig, & Secord, 1995), CELF-4 (Semel, Wiig, & Secord, 2003), or CELF-4 Australian Version (Semel, Wiig, & Secord, 2006). The CELF evaluates both expressive and receptive language ability, as well as providing an overall language score. Age-adjusted normative scores on each scale have a mean of 100 and standard deviation of 15. Children who obtained a receptive or expressive score ≤ 85 (i.e., at least one standard deviation below the mean) on any version of the CELF were considered to have a receptive or expressive language impairment respectively. Using this criteria, 16% of the standardisation sample

would be classed as demonstrating a receptive language impairment using receptive language scores and 16% would be classed as demonstrating an expressive language impairment using expressive language scores. A language impairment was classed as Mild if the CELF score fell between 78 and 85 (inclusive), Moderate if between 71 and 77 (inclusive), and Severe if 70 or below (Semel et al., 2006).

The Sutherland Phonological Awareness Test (SPAT; Neilson, 1995) or the Sutherland Phonological Awareness Test – Revised (SPAT-R; Neilson, 2003) were used to assess phonological skills in 216 children. Subtests measured rhyme detection and production, onset and final phoneme identification, blending, segmentation and deletion, as well as reading and spelling abilities. The SPAT/SPAT-R manuals provide normative data for children in the first four years of primary school. As such, children in Grade Prep to Grade Three were classified as having a phonological impairment if their score on the SPAT/SPAT-R fell more than one standard deviation below the mean for their grade level. Within the standardisation sample, 16% of children would be classified as having impaired phonological skills using this criteria. For those children in higher grades (Four to Six) where normative data was not provided, a conservative approach was adopted whereby normative data for Grade Three children was used. So, if children in Grades Four to Six scored more than one standard deviation below the mean score for Grade Three children, they were classed as having impaired phonological abilities. Lack of normative data for these grades meant that the proportion of a standardisation sample that would be classified as having impaired phonological skills is unknown.

For a subset of children (N=110) phonological skills were assessed using the Phonological Assessment Battery (PhAB) (Frederickson, Frith, & Reason, 1997), which includes measures of alliteration, rhyme, naming speed, fluency, spoonerisms, picture and digit naming and nonword reading. Raw scores obtained on the PhAB can be converted into

age-adjusted standard scores with a mean of 100 and standard deviation of 15. Children who were assessed using the PhAB were classified as having a phonological impairment if their total score fell at least one standard deviation below the mean (i.e. if their standard score was less than or equal to 85). As such, 16% of children within the PhAB standardisation sample would be considered impaired using this criteria.

A proportion of the children completed selected subtests from the Bruininks-Oseretsky Test of Motor Proficiency (Bruininks, 1978). Fine motor skills were measured using the upper limb speed and dexterity subtest, which requires children to complete eight items that measure hand and finger dexterity, hand speed, and arm speed. Gross motor skills were measured using the bilateral co-ordination subtest, which requires children to complete eight items that measure the ability to sequence precise movements and simultaneously coordinate movements on both sides of the body. Subtest raw scores on this test can be converted to standardised scores with a mean of 15 and standard deviation of 5. Children were classified as having a fine or gross motor impairment if their standard score on the relevant subtest fell more than one standard deviation below the mean (i.e., <10).

The Movement Assessment Battery for Children (MABC: Hendersen & Sugden, 1992) and the Movement Assessment Battery for Children-2 (MABC-2: Hendersen, Sugden, & Barnett, 2007) were also used to measure fine and gross motor ability. Age-adjusted standard scores ($M=10$, $SD=3$) were calculated for individual subtests. Gross motor skills were considered to be impaired if a child scored more than one standard deviation below the mean on the balance or aiming and catching component standard scores. Fine motor skills were considered to be impaired if a child scored more than one standard deviation below the mean on the manual dexterity component standard score. For both the Bruininks-Oseretsky Test of Motor Proficiency and MABC/MABC-2, 16% of children in the standardisation

sample would be classed as having a fine- and/or gross-motor impairment using the criteria employed in this study.

Emotional and behavioural difficulties were identified using teacher and parent questionnaires in conjunction with a thorough developmental history interview conducted by a social worker. Following the interview, the social worker met with the rest of the assessment team and a clinical decision was made as to whether emotional and behavioural difficulties were present or absent.

5.2.4 Data analysis

In the event of missing data, listwise deletion was used such that all analyses only used validly collected data. Group means were calculated for test performance and other continuous variables and frequencies were calculated for categorical data. To examine risk factors for comorbidity, a number of smaller groups were created using the classification criteria described previously. Firstly, children who met criteria for one or more learning disability were selected from the initial cohort. From this group, smaller groups of children with/without expressive/receptive language impairments; with/without motor impairments; with/without emotional difficulties and with/without behavioural difficulties were compared. T-tests were used for continuous variables (i.e. age and IQ), and chi-square tests of independence for categorical variables (i.e. gender, social history, family dysfunction, dual language home).

5.3 Results

A total of 326 children were included in the initial part of this study, with 235 males (72.1%) and 91 females (27.9%). Age ranged from five to 12 years (mean(SD)=8.8 years (1.36)), with 0.6% of children in their first year of school (Prep), 15.3% in Grade One, 25.2% in Grade Two, 27.3% in Grade Three, 16.9% in Grade Four, 13.8% in Grade Five, and 0.9%

in Grade Six. Two or more schools had been attended by 22.4% of children (70/312) and 18.1% (54/298) had repeated a Grade. Most children (189/306; 61.8%) had a close family member (e.g. parent) affected by learning difficulties. One-third (104/315; 33.0%) of children were part of a separated or blended family. Family functioning (e.g. dysfunction due to significant stress in the home or a recent negative life event) was felt to negatively impact 22.9% (72/314) of children, while home life was thought to be causing significant disruption to the broader family unit for 19.1% (60/314) of children.

5.3.1 Academic functioning.

On average, reading, spelling and math scores for the group fell within the Low Average range (Table 5.1), while the prevalence of reading (199/307; 64.8%), spelling (198/307; 64.5%) and maths (181/294, 61.6%) disabilities were similar.

Table 5.1

Mean (SD) for measures of intellectual, academic and language functioning for the learning disability sample

Domain	Mean (SD)
Academic functioning	
Reading (N=312)	85.55 (12.11)
Spelling (N=312)	83.65 (11.38)
Math (N=297)	86.75 (11.67)
Intellectual functioning	
Verbal comprehension index (N=313)	89.23 (12.08)
Perceptual reasoning/ Perceptual organisation index (N=261)	93.86 (14.64)
Working memory index (N=312)	85.84 (11.54)

Processing speed index (N=286)	90.24 (14.04)
Language functioning	
Total language score (N=273)	82.08 (13.08)
Receptive language (N=281)	85.06 (13.65)
Expressive language (N=281)	82.04 (14.05)

The prevalence of comorbidities between learning disability diagnoses was examined in children with valid scores for the three academic measures ($N=292$; see Table 5.2). Of these children, a small minority met criteria for a single learning disability diagnosis (7.9% math disability only, 4.5% spelling disability only and 4.5% reading disability only), 20.3% of children met criteria for two learning disability diagnoses, and 44.5% met criteria for comorbid math, spelling *and* reading disabilities. Fifty-four children of these children (18.5%) did not meet criteria for any learning disability. An independent sample t-test showed that children with no learning disability diagnosis ($M=7.83$, $SD=1.16$) were significantly younger than children who met criteria for at least one learning disability diagnosis ($M=9.04$, $SD=1.30$), $t(290)=-6.30$, $p<.001$. Further, 68.5% of this group fell at least half a grade below their expected level in one (or more) academic domain. In addition, many children with no learning disability diagnosis experienced non-academic difficulties (e.g. receptive language impairment, 14/47, 29.8%; expressive language impairment, 21/47, 44.7%; impaired phonological skills, 27/46, 58.7%; fine motor, 14/39, 35.9%, or gross motor impairments, 11/24, 45.8; past/current emotional difficulties, 18/53, 34.0%; or past/current behavioural difficulties, 20/53, 37.7%).

Table 5.2

Comorbidities between learning disability diagnoses for participants with valid scores on all academic measures

Learning disability status (N=292)	n (%)
Math disability only	23 (7.9)
Spelling disability only	13 (4.5)
Reading disability only	13 (4.5)
Math + spelling disability	13 (4.5)
Math + reading disability	14 (4.8)
Spelling + reading disability	32 (11.0)
Math+ spelling + reading disability	130 (44.5)
No learning disability	54 (18.5)

5.3.2 Intellectual functioning.

Mean Verbal Comprehension Index and Working Memory/Freedom from Distractibility Index scores fell within the Low Average range (Table 5.1), whereas Perceptual Reasoning Index/Perceptual Organisation Index and Processing Speed Index scores fell within the Average range. Full scale IQ for seventeen children (5.2%) scored within the Extremely Low range (<70).

5.3.3 Language functioning and phonological skills.

Almost two-thirds (178/281; 63.3%) of children met criteria for expressive language impairment (29.2% mild, 14.6% moderate and 19.6% severe impairment), while just under half (133/281, 47.3%) met criteria for receptive language impairment (21.0% mild, 12.5% impairment and 13.9% severe impairment). Overall, nearly two-thirds (189/289; 65.4%) of children displayed impaired phonological skills. In children with valid reading, language and

phonological skills scores ($N=248$), we found that the proportion of children who experienced phonological impairments differed depending on their language and reading abilities, $X^2(1)=13.23, p<.001$. A phonological impairment was evident in 69% of children with a language impairment and/or a reading disability compared with 39% of children with age-appropriate reading and language abilities. Further, children with both a reading disability and language impairment were significantly more likely to experience phonological impairments than children with a reading disability only or a language impairment only, $X^2(3)=27.72, p<.001$.

5.3.4 Motor skills, and emotional and behavioural functioning.

One-third (69/210; 32.9%) of children met criteria for a gross motor impairment, with a similar number experiencing impaired fine motor skills (93/247; 37.7%). Past or current emotional difficulties were identified for 126 ($N=289$; 43.6%) children, and 121 ($N=296$; 40.9%) had experienced past or current behavioural difficulties.

5.3.5 Risk factors for comorbid difficulties.

In 238 children with a learning disability diagnosis, risk factors for comorbid language, motor, emotional and behavioural difficulties were assessed (see Table 5.3). Emotional difficulties were more likely in children: with a family history of learning disabilities ($X^2(2)=9.06, p=.011$), who had repeated a Grade ($X^2(1)=4.80, p=.028$), who formed part of a separated or blended family ($X^2(2)=10.25, p=.006$), or who had experienced family disruption/dysfunction ($X^2(2)=21.89, p<.001$). Behavioural difficulties were more likely in children: who had experienced family disruption/dysfunction ($X^2(2)=15.25, p<.001$) or had a familial learning disability history ($X^2(2)=9.31, p=.010$). Language impairment was more likely in children who resided in a dual language home ($X^2(1)=6.03, p=.014$) or had lower intellectual functioning abilities ($t(199)=3.76, p<.001$). Motor impairment was more likely in

children who were younger ($t(157)=3.10, p=.002$) or had lower intellectual functioning abilities ($t(151)=2.37, p=.019$).

Table 5.3

Risk factors for comorbid language, motor, emotional and behavioural difficulties in children with a learning disability diagnosis (N=238)

	No LI	+ LI	No MI	+ MI	No ED	+ ED	No BD	+ BD
	N(%)	N(%)	N(%)	N(%)	N(%)	N(%)	N(%)	N(%)
Gender: <i>n</i> (%)								
Male	38 (75)	112 (72)	52 (67)	57 (70)	91 (73)	70 (74)	100 (74)	63 (73)
Female	13 (25)	43 (28)	26 (33)	24 (30)	34 (27)	25 (26)	36 (26)	23 (27)
Age: mean (SD)	8.9 (1.4)	9.1 (1.3)	8.7 (1.3)**	9.3 (1.2)	8.9 (1.3)	9.2 (1.3)	9.0 (1.2)	9.0 (1.4)
FSIQ: mean (SD)	92.7 (8.8)**	86.4 (10.6)	88.3 (11.6)**	84.2 (10.2)	88.4 (11.4)	86.5 (11.1)	87.6 (11.6)	87.6 (10.8)
Family history LD: <i>n</i> (%)								
Close family	33 (72)	98 (66)	43 (57)	50 (66)	71 (58)*	68 (76)	75 (57)*	64 (77)
Extended family	7 (15)	23 (16)	19 (25)	8 (11)	26 (21)	7 (8)	27 (21)	8 (10)
None	6 (13)	26 (18)	14 (18)	18 (24)	26 (21)	15 (17)	30 (23)	11 (13)
Mother's education: <i>n</i> (%)								
Primary	0 (0)	4 (3)	2 (3)	0 (0)	2 (2)	1 (1)	2 (2)	2 (2)
Lower secondary	16 (33)	37 (26)	23 (32)	17 (23)	32 (27)	22 (24)	36 (28)	20 (24)
Upper secondary	23 (47)	67 (47)	30 (41)	38 (51)	54 (46)	51 (55)	64 (49)	41 (50)
Post-secondary	6 (12)	26 (18)	12 (16)	15 (20)	24 (20)	9 (10)	22 (17)	11 (13)
University	4 (8)	8 (6)	6 (8)	5 (7)	6 (5)	10 (11)	7 (5)	8 (10)
Social history: <i>n</i> (%)								
Intact family	35 (69)	88 (60)	53 (68)	47 (61)	89 (71)**	47 (50)	84 (62)	51 (59)
Separated family	11 (22)	45 (30)	16 (21)	21 (27)	26 (21)	34 (36)	38 (28)	25 (29)
Blended family	5 (10)	15 (10)	9 (12)	10 (13)	10 (8)	13 (14)	13 (10)	10 (12)

Family dysfunction:

n (%)

None 20 (39) 82 (56) 38 (49) 43 (56) 84 (68)** 34 (36) 85(63)** 33 (39)

Family function 12 (24) 35 (24) 24 (31) 15 (20) 18 (14) 31 (33) 21 (16) 30 (35)

impacts

Family disruption 19 (37) 30 (20) 15 (20) 19 (25) 22 (18) 29 (31) 28 (21) 23 (27)

Dual language home

(yes): *n* (%) 2 (4)* 22 (15) 12 (15) 10 (13) 17 (14) 10 (11) 14 (10) 12 (14)

Repeated a grade

(yes): *n* (%) 8 (17) 28 (20) 12 (15) 15 (20) 14 (12)* 21 (23) 18 (14) 18 (23)

BD = Behavioural difficulty (past or current); ED = Emotional difficulty (past or current); LI = Language Impairment (Expressive or Receptive language score <85); MI = Motor Impairment (gross or fine motor)

* *p* < .05; ** *p* < .01

5.4 Discussion

This paper aimed to examine the characteristics of school aged children referred to a multidisciplinary Australian Learning Difficulties Clinic for assessment. The co-occurrence of academic, cognitive, language, motor, emotional and behavioural difficulties in children with learning disabilities is significant. Multidisciplinary assessment can provide a detailed evaluation of these difficulties through the assessment of multiple domains.

Consistent with past research, demographic information revealed that males were more likely to be referred to the clinic than females (e.g. Hendriksen et al., 2007). A large proportion of children had a close or extended family member who had experienced learning difficulties (DeFries, Stevenson, Gillis, & Wadsworth, 1991; Plomin & Kovas, 2005; Shastry, 2007; Snowling, et al., 2019; Stevenson, Graham, Fredman, & McLoughlin, 1987). This familial aggregation is important to consider as it allows for potential identification and monitoring of ‘at risk’ children early in life. This would facilitate programs for early

intervention, which have been shown to improve outcomes for children with learning disabilities (Berninger & Armtmann, 2003; Snowling & Hulme, 2011).

5.4.1 Academic functioning.

Within our cohort, almost two-thirds of children met criteria for multiple learning disability diagnoses. In contrast, single learning disability diagnoses (i.e. reading disability only, spelling disability only or math disability only) were identified in a small minority. A high rate of comorbidity between learning disabilities is consistent with previous epidemiological research (e.g. Landerl & Moll, 2010). It confirms the need for a full and comprehensive assessment in all children referred for suspected learning difficulties in order to understand each child's cognitive and academic profile. Findings also revealed that the prevalence of reading, spelling and math disabilities were relatively equal in our sample, which is in keeping with evidence of equal prevalence of math and reading disabilities in clinically referred samples (e.g. Mayes & Calhoun, 2007).

Almost one in five children in this study did not meet our criteria for any learning disability. This was of interest given that referrals to the Learning Difficulties Clinic were only made when children were experiencing difficulties in the classroom and learning disabilities were suspected. Although not reaching diagnostic level, almost 70% of the children were at least half a grade below their expected grade level in one (or more) academic domains. These same children also presented with deficits in language, phonology, and fine and gross motor development. These children may also have been experiencing academic difficulties that fell outside of the scope of the assessment measures utilised in this study. For example, reading comprehension deficits, handwriting impairments, or written expression deficits would not have been identified using this study's methodology. The younger age of this group is also relevant as academic difficulties may not be readily identified in young children using the measures utilised in this study. For example, early items on the

WIAT/WIAT II word reading subtest do not place a strong emphasis on word reading skills per se, instead focusing on skills such as letter identification and rhyming. As such, young children who test at age appropriate levels are at risk and may still go on to show significant reading difficulties in the future. Longitudinal research would be beneficial to confirm whether these children go on to develop learning disabilities and to identify any early identifiable risk factors. If learning disabilities do develop, thorough assessment at a young age and regular follow up assessments would allow progress to be monitored and would provide opportunities to benefit from early intervention (Foorman & Breier, 2003)

5.4.2 Intellectual functioning

Our results showed that, on average, non-verbal and verbal intellectual abilities along with processing speed fell within or very close to the average range, while working memory abilities fell within the low average range. The findings were considered to be broadly consistent with past research showing that verbal and non-verbal intellectual skills tend to be age appropriate in children with learning disabilities, while Working Memory tends to be the lowest of the four Index scores, falling in the Low Average range or lower (De Clercq-Quaegebeur et al., 2010; Giofre & Cornoldi, 2015; Styck & Watkins, 2014). Working memory abilities are related to general academic achievement (Alloway & Alloway, 2010; Gathercole & Pickering, 2000) and to other cognitive abilities that are important for learning, including processing speed, attention, short-term memory and phonological skills. Impairments in these cognitive domains have been found in children with learning disabilities (e.g. Kudo, Lussier, & Swanson, 2015; Willcutt, Sonuga-Barke, Nigg, & Sergeant, 2008), but tests of intellectual functioning alone are not sufficient to assess and identify these impairments. A comprehensive, specialised learning disabilities assessment is therefore better suited to provide an accurate picture of the nature and pattern of cognitive impairments in this population.

5.4.3 Language functioning and phonological skills.

Expressive language impairments were detected in more than 60% of children with valid data, while almost half of the cohort met criteria for receptive language impairment. These figures are much higher than population prevalence estimates of language impairments/disabilities. However, this is unsurprising given that this cohort was referred for academic difficulties and that learning disabilities (particularly reading and spelling disabilities) are frequently comorbid with language impairments in children (Eisenmajer, Ross, & Pratt, 2005; McArthur et al., 2000). Phonological impairments, which were found in approximately two-thirds of the overall cohort, were more common in children with a reading disability and/or language impairment than those without these deficits, highlighting the importance of including a phonological skills assessment in any child suspected of having reading and/or language difficulties. Further, phonological impairments were more common in children with reading disability *and* language impairment. This is in line with previous research suggesting that children with language impairments *and* reading disability are more likely to experience phonological impairments, and that these impairments are generally of greater magnitude than those in children with reading disability or language impairment alone (Bishop, McDonald, Bird, & Hayiou-Thomas, 2009; Eisenmajer et al., 2005; Fraser et al., 2010; Ramus, Marshall, Rosen, & van der Lely, 2013).

5.4.4 Motor skills.

With approximately one-third of the cohort with valid data meeting criteria for gross motor impairment and a similar proportion with fine motor impairment, our results were consistent with past research showing that motor impairments are more prevalent in children with academic difficulties than in the general population (Brookman, McDonald, McDonald, & Bishop, 2013; Lingam, Hunt, Golding, Jongmans, & Emond, 2009; Vuijk et al., 2011). This is relevant because motor impairments have the potential to impact upon children's level

of functioning both at home and school (Jackman & Stagnitti, 2007; Wang, Tseng, Wilson, & Hu, 2009). Poor gross motor skills can be associated with trouble sitting still in the classroom or difficulties playing sport or other games at the same level as their peers. Fine motor impairments can also negatively impact upon classroom activities, including cutting with scissors, colouring in and handwriting, along with self-care activities such as opening lunch boxes and tying shoe laces. This indicates an important role for occupational therapists who can provide a thorough assessment of fine and gross motor abilities along with recommendations for intervention.

5.4.5 Emotional and behavioural functioning.

Results in this domain were consistent with research showing significant levels of internalising and externalising symptomatology in children with learning disabilities (Faraone et al., 1993; Mugnaini et al., 2009; Ritter, 1989; Sanson, Prior, & Smart, 1996; Shalev et al., 1995; Stott, 1981; Westman, Ownby, & Smith, 1987; Willcutt & Pennington, 2000). Almost half of the children in this study had reportedly experienced past or current emotional and/or behavioural difficulties, such as anxiety, low mood, poor emotional regulation and over activity. Emotional and behavioural difficulties have the potential to not only impact on a child's emotional wellbeing, but also affect their learning capacity. This highlights the importance of practitioners considering the mental health status of any children suspected of learning disabilities.

5.4.6 Co-occurring difficulties in children with learning disabilities.

We identified environmental factors (e.g. within family functioning and social history domains) that were present at increased rates in children with learning disabilities who had co-occurring language, motor, emotional or behavioural difficulties (compared to children with learning disabilities without the corresponding difficulty). Differences between these groups were also evident in non-environmental factors (e.g. age, level of intellectual

functioning). The family environment was particularly important, as being in a separated or blended family was associated with an increased likelihood of emotional difficulties, and family disruption/dysfunction was associated with an increased likelihood of both emotional and behavioural difficulties. Lower intellectual functioning was a risk factor for language and motor impairment, and motor impairment was also more likely in younger children. This suggests that children presenting with these risk factors and an accompanying learning disability should be referred for further multidisciplinary assessment. This would ensure the multiple contributors to their academic difficulties are identified and addressed.

5.4.7 Strengths and limitations.

An important strength of this study was its broad nature. We aimed to provide a comprehensive description of the difficulties experienced by children suspected of learning disabilities, and the multidisciplinary nature of the Learning Difficulties Clinic was well placed to address this. Inclusion of consecutive referrals over a 12 year period gave rise to a good sample size which accurately reflected the typical referrals provided to the Learning Difficulties Clinic over that time and provided for a thorough description of the type of difficulties experienced by these children. One limitation was that the nature of the clinic did not allow for inclusion of a control group. As such, the study could not statistically test whether the difficulties examined were more frequently experienced by children with learning disabilities than those in their typically developing peers. However, while this would have been a useful addition to the study, it did not impact the primary aim of the investigation, which was to describe the difficulties experienced by children referred to the Learning Difficulties Clinic. It is also acknowledged that this study examined a clinically-referred cohort, which may not be representative of children with learning disabilities within the general population. Clinically referred cohorts can be associated with referral biases (e.g. Angold, Costello, & Arkanli, 1999). For example, difficulties arising from the co-occurrence

between two disorders may be what prompts referral to a specialist clinic. This leads to inflation of the estimation of comorbidity within these cohorts. The results of this study can therefore be generalised to other clinically referred learning disability populations but care should be taken before generalising the results more broadly.

5.4.8 Conclusion.

Overall, the results highlighted that children referred to a Learning Difficulties Clinic experience a variety of academic, cognitive, language and phonological skills, motor, behavioural, and emotional impairments. Impairments in only one academic domain (i.e. reading disability only, spelling disability only or maths disability only) were uncommon. In children with learning disabilities, a number of risk factors (both environmental and non-environmental) were identified that increased the likelihood of experiencing comorbid language, motor, emotional or behavioural difficulties. The multidisciplinary assessment process in place at the Learning Difficulties Clinic supported the complex diagnostic requirements within this population, delivering the framework for accurate diagnoses and management recommendations across multiple domains. Further research should examine how impairments in each domain interact with one another. In addition, investigation into whether multidisciplinary assessment and intervention is associated with improvements in treatment efficacy for children with learning disabilities would be beneficial.

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Chapter 6 – General Discussion

The research described within this thesis aimed to a) provide an investigation into the structure of phonological skills, short-term memory and working memory in children with suspected learning disabilities; b) to explore the pattern and nature of phonological skill abilities in sub-groups of children identified with reading disability (RD-only), specific language impairment (SLI-only) and a comorbid group of reading disability and specific language impairment (RD+SLI), and c) broadly examine and describe the types of academic and non-academic impairments experienced by children referred to a multidisciplinary learning difficulties clinic for assessment. A thorough review of relevant literature was provided earlier; this general discussion will provide a brief review of our salient results and discuss how our findings relate to past research and theory. Strengths and limitations, along with ideas for future research will also be provided.

6.1 Summary of Findings

6.1.1 The structure of phonological skills, short-term memory and verbal working memory.

Confirmatory factor analysis was used to examine whether phonological skills were best conceptualised as a single cognitive construct or as multiple distinct constructs; how phonological skills were related to short-term memory and verbal working memory; and how cognitive load impacted upon these relationships. Two sets of analyses were conducted – one using raw scores and the other using age-adjusted scores for variables that were significantly correlated with age. The pattern of results were similar whether or not age was adjusted for, with a three factor model selected as the best fitting model in both types of analyses. As hypothesised, this model identified two distinct, but closely related phonological skills factors

which were differentiated by cognitive load (low versus high). Tasks loading onto the first phonological factor included syllable counting, phoneme blending, segmentation, and deletion (initial sounds) tasks. This construct was conceptualised as an earlier developing, lower-level construct, termed phonological awareness. The associated tasks were considered to be relatively cognitively undemanding for primary school children and thus were low in cognitive load. In contrast, two cognitively demanding phoneme deletion tasks loaded onto a second phonological construct, which we described as ‘higher-level’ phonological awareness. This construct was considered to mature later in childhood. The associated tasks required the children to not only have an awareness and understanding of the sounds of their language, but also explicitly and concurrently maintain, manipulate and integrate those sounds in order to produce an accurate response. Results also revealed that a single memory construct (incorporating both short-term and verbal working memory variables) provided an adequate fit to the data.

6.1.2 Phonological skills in reading disability and specific language impairment.

Comparison of RD-only, SLI-only and RD+SLI groups found that children with RD+SLI displayed phonological impairments which were more prevalent than phonological impairments in the RD-only group. A non-significant trend was found for the SLI-only group. Impairments in children with RD+SLI were also significantly more severe than both the RD-only and SLI-only groups. Children in the RD-only and SLI-only groups displayed mild phonological impairments. They performed more poorly than children in the SPAT normative group, but performed slightly higher than our predetermined cut-off for phonological impairment. Removing Grade 4 to Grade 6 children from the RD-only and SLI-only groups resulted in phonological skills being impaired in both groups. This suggests that the lack of normative data for children in Grade 4 to Grade 6 and the subsequent use of Grade 3 norms for these children resulted in an impairment not being found (rather than a lack of

deficit). There were no significant differences between RD-only and SLI-only groups in prevalence or severity of phonological impairments. We had hypothesised a lower prevalence of phonological impairments in the SLI-only group as compared to the RD-only group based upon a finding that groups of children with SLI-only have increased diversity in their phonological functioning as compared to those with RD-only (Ramus et al., 2013).

Profile analysis compared each group's overall pattern of performance on a selection of higher and lower-level phonological tasks (i.e. syllable counting, two phoneme segmentation tasks and three phoneme deletion tasks). The analysis was considered largely exploratory given a scarcity of past research in this area. Group comparisons revealed that, across the six tasks, the RD-only group had a significantly different profile from the RD+SLI group (i.e. the pattern of performance of the two groups across the six tasks were not parallel to one another). This difference seemed to be driven by differences in performance on the syllable counting task, which was the only syllable level task amongst the six examined. There were no other significant differences identified. These results further emphasise the similarities between the RD-only and SLI-only groups by suggesting that they have similar patterns of performance across the phonological tasks included in the analysis.

6.1.3 Characteristics of children referred to a learning difficulties clinic.

6.1.3.1 Demographics and academic functioning.

Males were more likely to be referred to the clinic than females, which is consistent with past research in this area (Hendriksen et al., 2007). A large proportion of children had a close or extended family member who had experienced learning difficulties. Prevalences of reading disability, spelling disability and math disability were relatively equal and almost two-thirds of children met criteria for multiple learning disability diagnoses. This highlights the frequent co-occurrence between different learning disabilities. Despite being referred due to concerns about their learning, almost one in five children did not meet criterion for any

learning disability diagnosis. Closer examination of this group of children suggested that they were experiencing sub-clinical academic difficulties that placed them at significant risk of a learning disability diagnosis as they progressed through primary school and academic expectations increase.

6.1.3.2 Non-academic functioning.

Mean intellectual functioning index scores all fell close to the intersection of low average and average ranges. Language and phonological impairments were common, with expressive language impairments evident in approximately 60% of children with relevant data, while receptive language impairments were identified in almost half of the cohort. Overall, approximately two-thirds of the children demonstrated phonological impairments. Further examination demonstrated that children who met criteria for a reading disability and/or a language impairment were more likely to demonstrate poor phonological skills than those with age appropriate language and reading skills. Those who met criteria for both reading disability *and* language impairment were more likely to demonstrate phonological impairments than those with a single deficit (i.e. reading disability only or language impairment only). Prevalence of fine and gross motor impairments were relatively equal with about one-third of the cohort who had relevant data experiencing fine motor difficulties and one-third experiencing gross motor difficulties. Almost half of the children had experienced emotional and/or behavioural difficulties in the past or at the time of assessment.

Risk factors for comorbid language, motor, emotional and behavioural difficulties in children with a learning disability diagnosis were also examined. Results showed an increased probability of emotional difficulties if the child formed part of a separated or blended family and an increased likelihood of both emotional and behavioural difficulties if parents reported the presence of family disruption or dysfunction. Lower intellectual

functioning was a risk factor for language and motor impairments, whilst motor impairment was also more likely in younger children.

6.2 Relationship to Past Research and Theoretical Implications

6.2.1 Cognitive load and phonological skills.

The findings presented in Chapter 3 of this thesis align with two research studies which both identified separate higher- and lower-level phonological factors in school aged children (Ramus et al., 2013; Yopp, 1988). In both studies, tasks loading onto the higher-level phonological construct required greater cognitive effort and attention than those loading onto the lower-level construct. Our results partially supported Ramus et al. (2013) who highlighted the important role that cognitive load plays in differentiating different types of phonological skills. However, direct comparison between the two studies was difficult due to differences in the nature of the phonological constructs identified. Ramus et al. (2013) identified a lower-level phonological factor, which was conceptualised as phonological representations, while their higher-level phonological factor was multifaceted. It was associated with four tasks (phoneme substitution, rhyme task, digit span and rapid automatized naming) that we considered to assess phonological skills, short-term memory and working memory domains. The heterogeneity of tasks loading onto the higher-level factor and the limited number of phonological awareness and processing tasks in this study make direct comparison with our results difficult. The findings in Ramus et al. (2013) do, however, support the hypothesis that phonological skills are not a unitary construct and highlight the important role that cognitive load plays in understanding this issue.

In contrast to our results, other researchers have found that phonological skills best represent a unitary construct (e.g. Adams, 1992; Schatschneider et al., 1999; Stahl & Murray, 1994). Inconsistencies in methodology and analytic techniques probably contribute to the

discrepancies between our findings and those who identified a unitary construct. In particular, some studies have included limited phonological tasks, with ‘higher-level’ phonological awareness tasks sometimes omitted completely. This makes it less likely that separation of phonological awareness and ‘higher-level’ phonological awareness would occur in any structure analysis, increasing the likelihood of finding a unitary phonological skills factor. Differences between studies in the age and cognitive development of participants is another factor that may impact upon results. Cognitive skills, such as short-term memory, working memory and phonological skills mature at different rates and their developmental trajectories therefore differ. As such, there may be differences in the strength of relationships between these cognitive skills at different points in time (S. E. Gathercole et al., 2004). This adds to the complexities associated with comparing studies involving children of different ages.

Relatedly, differences in age and cognitive development can also make it challenging to estimate the cognitive load of a task. This can be seen through examination of the different developmental trajectories of cognitive skills, such as short-term memory, working memory, phonological awareness and ‘higher-level’ phonological awareness. Because the age-related trajectory for differs for each cognitive domain, each age group will have different mastery of each domain, changing in part the demands of each phonological awareness or ‘higher-level’ phonological awareness task. So, a phonological task may be relatively cognitively undemanding and represent a phonological awareness task in older children for whom the associated cognitive skills are well developed, but the same task could require higher levels of cognitive load (and hence represent a ‘higher-level’ phonological awareness task) in younger individuals whose cognitive skills are less mature and well developed. Similarly, children who have cognitive deficits or delays in these domains will also differ. What would be considered a lower-level task for a typically developing child, may therefore actually be akin to a higher-level task for a child with a neurodevelopmental disorder. Furthermore, a

child who has participated in an intervention may be more adept at a related task and the cognitive load of that task could be less than expected for this child.

The complexities associated with estimating cognitive load may also partly explain one observed difference between findings in Chapter 3 and those obtained by Yopp (1988). Comparison of the deletion tasks in these studies show that the two deletion tasks in Yopp (1988) were both considered higher-level tasks, while the three SPAT deletion tasks in this thesis (see Chapter 3) were spread over both phonological awareness (initial phoneme deletion) and ‘higher-level’ phonological awareness (1st phoneme deletion; 2nd phoneme deletion) constructs. The differences again highlight the challenges of accurately estimating cognitive load of these tasks and most likely relate to differences in 1) the age and/or cognitive development of participants as described above, and 2) item characteristics within each task. Briefly, with regards to item characteristics, the position of the target phoneme and the linguistic complexity of items can affect the overall level of cognitive load of a task. For example, tasks which require deletion of the first phoneme within a consonant cluster (e.g. “Say play without /p/”) or from within the middle of a word are more demanding than initial phoneme or final phoneme deletion tasks where the phoneme is not a part of a consonant cluster (Pufpaff, 2009). Examination of deletion tasks in Yopp (1988) showed that they included items which were heterogeneous in the position of their target phoneme and linguistic complexity. This makes estimation of cognitive load for each task difficult. In contrast, our research included phoneme deletion tasks which were quite homogenous in their task requirements and item characteristics. Tasks such as these increase the likelihood of accurately estimating cognitive load.

In summary, the results described in this thesis add to our understanding of the structure of phonological skills through the identification of distinct phonological awareness and ‘higher-level’ phonological awareness constructs which differ in their level of cognitive

load. There are theoretical and clinical implications of this finding. Theoretically, phonological skills are closely related to reading and language development and are commonly impaired in children with difficulties in language and literacy domains (e.g. those with reading disability and/or specific language impairment). It may be, however, that phonological awareness and ‘higher-level’ phonological awareness are differentially related to reading and language or differentially impaired in the related developmental disorders. Further research is clearly needed. The refinement of theoretical models describing the relationships between phonological awareness, ‘higher-level’ phonological awareness, language and literacy can in turn be useful to inform assessment and intervention practices. Assessment of phonological skills by classroom teachers or clinicians could incorporate both phonological awareness and ‘higher-level’ phonological awareness tasks. Moreover, understanding the pattern of strengths and weaknesses across both phonological awareness and ‘higher-level’ phonological awareness would aid the development of appropriate interventions. Intervention and classroom teaching strategies are likely to differ depending on whether impairments are identified in the areas of ‘higher-level’ phonological awareness only or both phonological awareness and ‘higher-level’ phonological awareness.

6.2.2 The relationship between phonological skills, reading disability and specific language impairment.

The results presented in Chapter 4 of this thesis were compared to six previous studies which utilised similar methodology to our own. Our findings revealed that RD-only and SLI-only groups demonstrated mild phonological impairments, scoring slightly higher than our cut-off for phonological impairments. We had expected the RD-only group to demonstrate a more severe phonological impairment and therefore to meet our criteria given that given that five of the six key studies had found impaired phonological skills in children with RD-only (Catts et al., 2005; Fraser et al., 2010; G. McArthur & Castles, 2013; Ramus et al., 2013;

Snowling et al., 2019). A lack of age appropriate normative data for the phonological test battery used (i.e. the SPAT) lead to an overestimate of phonological skill functioning in our cohort and was considered a likely contributor to this unexpected finding.

Comparisons of RD-only and SLI-only groups showed no significant differences in prevalence, severity or pattern of phonological impairments. This is consistent with our hypothesis that there would be no difference in the severity of impairments between these groups, as had been found in three of four relevant studies (Fraser et al., 2010; Ramus et al., 2013; Snowling et al., 2019). Inconsistent with our results, we had also hypothesised lower prevalence of impairments in the SLI-only group and we had proposed that the pattern of performance may be different for these groups. These predictions were largely based upon findings from Ramus et al. (2013) and was made tentatively due to differences in the design and methodology of our study in comparison to theirs. In particular (as described earlier) the multifactorial nature of the higher-level phonological construct made direct comparison with our results difficult. In addition, Ramus et al. (2013) used composite phonological scores for comparisons so did not test whether performance on individual phonological tasks may be differentially impacted in the RD-only and SLI-only groups. Our results suggest that the overall pattern of performance across a number of phonological awareness and ‘higher-level’ phonological awareness skills tasks does not differ.

Another key finding in our study was that the double deficit RD+SLI group had more severe phonological deficits than the RD-only and SLI-only groups, and more often showed phonological impairments than the RD-only group (with a trend evident for the SLI-only group). More severe deficits for the double deficit group had been identified consistently in past research when compared to both SLI-only (Catts et al., 2005; Eisenmajer et al., 2005; Fraser et al., 2010; G. McArthur & Castles, 2013; Ramus et al., 2013; Snowling et al., 2019)

and RD-only (Fraser et al., 2010; G. McArthur & Castles, 2013; Ramus et al., 2013) groups of children.

The implications of these research findings are multifaceted. Clinically, mild deficits were noted in both RD-only and SLI-only groups, while more severe deficits were evident in the double deficit group. The co-occurrence of language, reading and phonological impairments emphasises the need for children suspected of reading or language impairments to undertake a comprehensive phonological assessment along with assessment of both reading and language skills. Ceiling effects were evident on a number of the tasks considered for inclusion in this study, which emphasizes the importance of selecting phonological tasks that are appropriate to a child's age and level of cognitive development. Developmental trajectories of phonological skills indicate earlier and later developing phonological abilities (see Chapter 3), and this should be taken into consideration when choosing tasks. Tasks that are far too easy for a child or beyond the child's cognitive capacity will lead to ceiling or floor effects in research and will provide limited clinical information during clinical assessments. Ensuring that phonological tasks have adequate range and normative data will also aid clinical assessment and interpretation. Accurate identification of each child's particular areas of difficulty will facilitate selection of appropriate interventions and classroom teaching strategies.

In the context of research into models of reading disability and specific language impairment, the results support multiple deficit models of neurodevelopmental disorders. These models propose that the combination of etiological risk and protective factors impact the development of cognitive abilities. Reading and/or language impairments then result from the interactions between multiple cognitive risk factors (Peterson & Pennington, 2012). Both reading disability and specific language impairment have been linked with poor phonological skills (Bishop & Snowling, 2004; Melby-Lervag et al., 2012). Furthermore, difficulty

acquiring adequate phonological skills has been hypothesised as a possible foundation for impaired reading and language acquisition in reading disability and specific language impairment respectively (Corriveau et al., 2007; Ramus, Rosen, et al., 2003).

However, multiple deficit models of reading disability also propose that phonological impairments are not necessary or sufficient to cause reading difficulties in children (Pennington, 2006; Peterson & Pennington, 2012). Our results support this; phonological impairments were found in children with SLI-only and there were children with reading disabilities that did not have phonological impairments. Another aspect of our results provides further evidence that phonological skills are not sufficient to explain the presentation of reading and language impairments in children. In our groups, children with RD-only and SLI-only had comparable phonological impairments in severity and pattern. Despite these similarities, they had different patterns of reading and language abilities (i.e. adequate language and poor reading in the RD-only group, but poor language and adequate reading in the SLI-only group).

6.2.3 Multidisciplinary assessment of children with suspected learning disabilities.

The findings described in Chapter 5 demonstrate that children referred for suspected learning disabilities experience a wide variety of difficulties and impairments that could potentially impact their day-to-day functioning. The results were largely consistent with a variety of past research studies which have identified language, cognitive, motor, emotional and behavioural difficulties in children who experience learning disabilities (e.g. G. M. McArthur et al., 2000; Mugnaini et al., 2009; Vuijk et al., 2011; Willcutt et al., 2013). Several relevant studies suggest that a multidisciplinary assessment framework, such as that at the Learning Difficulties Clinic, have a number of benefits. They are a positive experience for the child being assessed; can be helpful to improve parents' understanding of their child's

strengths and weaknesses and may help parents communicate better with the school (if the school is involved in the initial assessment process; Oberklaid, 1984). Other advantages for families may include improved convenience, with fewer appointments to attend. This may improve compliance and willingness to attend appointments and therefore improves diagnostic procedures for children. The involvement of multiple practitioners from complementary disciplines also maximises the likelihood of a thorough assessment of academic difficulties and co-occurring impairments. There are also benefits arising from combining the knowledge, expertise and clinical experience of practitioners across different disciplines. Multidisciplinary clinics also dilute systematic individual diagnostic bias that may be evident when practitioners work independently to assess children with suspected learning disabilities.

Benefits of multidisciplinary frameworks have also been found in the context of other neurodevelopmental disorders or medical conditions commonly diagnosed in childhood. Relevant studies emphasise the challenges in diagnosis of neurodevelopmental disorders and suggest that multidisciplinary assessment is well placed to address these complexities. For instance, Efron & Sciberras (2010) found that a proportion of children referred to one multidisciplinary clinic did not meet diagnostic criteria for attention-deficit hyperactivity disorder, despite being referred with symptoms of this disorder. Instead, these children were either provided with an alternate diagnosis (including anxiety, depression, oppositional defiant disorder, learning disability, autism spectrum disorder and speech or language impairment) *or* met criteria for no diagnosis. Similar findings were described in Hendriksen et al. (2007), whereby over one quarter of the final diagnoses provided by the multidisciplinary team did not correspond to the referral question from monodisciplinary healthcare services. Taken together, these results mirror our findings whereby a proportion of children referred due to academic difficulties in the classroom failed to meet our criteria for

reading disability, spelling disability or math disability. We proposed that a proportion of these children may go on to develop a learning disability as they progress through an increasingly academically challenging school system. Alternatively, they may in fact meet diagnostic criteria for an alternate neurodevelopmental or psychiatric condition.

Taken together, the above results show that behaviours of concern or difficulties noticed by teachers, parents and/or practitioners are not easily understood without a comprehensive assessment process. Signs of various psychiatric and neurodevelopmental disorders, including learning disabilities, can mirror or overlap with one another, making accurate diagnosis challenging. Further complicating the diagnostic process, children may meet criteria for more than one disorder or condition, leaving practitioners to make a number of diagnostic decisions regarding the likely primary and secondary diagnoses applicable in each case. Findings in Efron & Sciberras (2010) suggest that multidisciplinary assessment can effectively identify a range of problems and help inform targeted interventions in children referred with symptoms of ADHD. Similarly, the findings described in Chapter 5 suggest that multidisciplinary assessment could be well placed to identify a variety of difficulties or impairments in children with suspected learning disabilities. Assessment conducted in this setting provides a broad assessment conducted by a variety of relevant practitioners. In turn, this allows consideration of various potential differential diagnoses that may not be measurable by a single practitioner alone. However, multidisciplinary clinics are expensive to run and can be difficult to access for some families (e.g. due to the relative rarity of these clinics in the public health system or due to the cost of multiple disciplinary assessments in the private system). These drawbacks need to be considered alongside the benefits described above.

The richness of information gathered during multidisciplinary assessments also has the potential to improve ongoing management of these children. The presence of co-

occurring difficulties can limit an individual's ability to respond positively to an intervention program. For example, the presence of attention deficits, poor rapid naming skills, weak verbal abilities and behaviour difficulties have been associated with poor outcomes in children with reading disability undergoing reading intervention programs (Alexander & Slinger-Constant, 2004). Moreover, the presence of co-occurring academic impairments can change the type and intensity of interventions required for remediation (Fuchs, Fuchs, & Compton, 2012). Provision of accurate diagnoses and mapping of an individual's strengths and weaknesses across domains are therefore important when designing appropriate interventions.

6.3 Strengths and Limitations

Given the range of difficulties experienced by children with learning disabilities, access to results of a large number of multidisciplinary learning assessments was an important strength of the current research. The nature of the clinic allowed broad, integrated assessments to be conducted across several complimentary allied health disciplines and also included information gathered from multiple sources (i.e. from home and school). In turn, this provided data spanning a variety of domains. The nature of the clinic also contributed to several limitations within this thesis. Children had been referred to the clinic for assessment of suspected learning disabilities. Clinically referred samples can result in referral bias and as such the results should not be generalised to non-clinical populations or groups of children with other neurodevelopmental disorders. Combined with the retrospective design of the research, the nature of the clinic also meant that no control group was included in any of our studies. However, most of the tasks administered had standardised normative data available which acted as a sort of 'pseudo control group' via creation of standardised scores which

were used in most analyses (see below for more information regarding the SPAT, which was an exception).

Furthermore, although a wide variety of cognitive, language and motor assessments were administered to each child, test selection for this study was constrained by the clinical decision making of staff and test availability rather than tests being selected to match our specific research aims and study design. This presented us with several complications.

Firstly, in order to maintain high clinical assessment standards and meet the goals of the clinic's staff, some tests were replaced with different tests during the period of data collection. For example, to measure phonological skills, a new test battery the Phonological Skills Assessment Battery, was introduced for a short period of time replacing the SPAT. In addition, new versions of several tests were introduced (e.g. the WISC-IV replaced the WISC-III). These changes affected sample size of the cohorts described in Chapters 3 and 4 because in some cases it was not possible to combine and analyse data from different tests. An additional complication related to the short-term memory and working memory tasks administered. Selection and inclusion of more sensitive tests in these domains may have improved the clarity of short-term memory and working memory results in Chapter 3.

The phonological test battery (SPAT) had the advantage of including tasks which spanned both phonological awareness and 'higher-level' phonological awareness. This was an important strength of this research. The battery also had several limitations in the context of the current research. Firstly, with just four items, the score range of each subtest included in this study was restricted, which contributed to ceiling effects being evident on a number of subtests. This led to exclusion of several subtests from the profile analysis described in Chapter 4. Secondly, normative data provided by the publisher of this test is limited to children in Grades Prep to Three. The conservative methodology we adopted meant that children in Grade Four to Six therefore were compared to Grade Three normative data. This

did impact upon some of our results (e.g. magnitude of phonological impairments in children with reading disability and specific language impairment), but it did not affect the overall pattern of results obtained.

Inclusion of 326 children representing successive referrals to the learning disabilities clinic over 12 years (described in Chapter 5) was another strength of the study. Sample sizes in the previous two Chapters however were more circumscribed, in part due to the changes to tests administered at the Learning Difficulties Clinic as described previously. Chapter 4 also focused only upon those children with reading and/or language impairments, which lead to the exclusion of some of the initial cohort. However, though larger group sizes would have been preferred, the sizes were considered adequate for the analyses conducted and ensured methodological issues were more closely controlled.

The broad age range of the children assessed at the learning difficulties clinic was another potential limitation. Each of the studies included in this thesis sought to examine results independent of age by using age (or grade) standardised scores where possible, but sample sizes were not large enough to separately examine groups of children using more circumscribed age ranges. It is possible that some of the results obtained would differ in children of different ages and stages of development. For example, the relationships between phonological skills, verbal working memory and short-term memory may be different in children in early primary school as compared to those in late primary school. Similarly, the pattern of phonological impairments in children with reading disability and specific language impairment could potentially vary with age. The design and methodology of the studies in this thesis did not allow examination of these potential differences.

6.4 Future Directions

6.4.1 The structure of phonological skills.

Our results suggest separation of phonological awareness and ‘higher-level’ phonological awareness in a cohort of children with suspected learning disabilities, and similar results were found by Yopp (1988) in a community based sample of young children. However, it is not clear whether this separation can be generalised to other groups. It would be beneficial to replicate our results in other cohorts of children with learning disabilities or developmental disorders, along with community based samples. Longitudinal research in this area would allow investigation into whether the structure of phonological skills varies across age. In paediatric populations longitudinal research is particularly beneficial given the significant cognitive, language and other developmental changes that take place throughout childhood and adolescence. Recruitment of relatively homogenous groups and systematic variations in age may improve clarity of results and provide information as to how changes in these variables impact the level of cognitive load required for task completion and how age impacts upon the structure of phonological skills themselves.

The development and use of well-designed phonological tasks and test batteries is also important. These batteries need to adequately measure both phonological awareness and ‘higher-level’ phonological awareness. They also need to have adequate normative data across a wide age range so that both pre-school and school aged children can be effectively assessed. Potential floor and ceiling effects can be minimised by ensuring a reasonable score range within each task and by choosing a task that is appropriate for the age and level of development of the child/ren being assessed.

If it is possible to confirm separation of phonological awareness and ‘higher-level’ phonological awareness in future research studies, it would be helpful to examine how these different phonological skills are related to other cognitive and academic abilities. For

instance, at this stage it is not clear as to whether they are differentially related to language and reading development, or differentially impacted in language or reading disabilities.

We also do not know which phonological tasks are the best measures of phonological awareness and ‘higher-level’ phonological awareness. In part, this is complicated by inconsistent definitions of terms such as phonological skills and phonological awareness within existing literature. Research examining this question will also be complicated by the fact that the best measures of these constructs are likely to vary with age and development. If clinicians understand which tasks provide accurate measurement of phonological awareness and processing skills, they can ensure that both types of phonological skills are assessed and subsequently target their interventions on the basis of the results.

6.4.2 Phonological skills in reading disability and specific language impairment.

Additional studies with similar methodology to that described in Chapter 4 would be beneficial in improving our understanding of cognitive and other impairments experienced by children with specific language impairment and reading disability. In the case of phonological skills, it would be useful to clarify whether phonological awareness and ‘higher-level’ phonological awareness skills are differentially impacted in these developmental disorders.

Future research is also needed to clarify the potential genetic and environmental factors (both risk and protective) which are unique or shared between reading disability and specific language impairment. Inclusion of multiple cognitive factors would be beneficial. Such research would be bolstered by using clear, consistent definitions of reading disability and specific language impairment and recruitment of groups which are as homogenous as possible on relevant variables (e.g. magnitude of reading and language impairment, type of language impairment, intellectual functioning, and age).

Additional longitudinal research is needed to investigate whether the relationships between phonological abilities, reading disability and specific language impairment change over time. Emphasizing the importance of further research in this area, a newly published study recently found that the phonological impairments in children with specific language impairment (but no comorbid reading disability) decrease over time, in contrast to the phonological impairments of those with reading disabilities (Snowling et al., 2019).

6.4.3 Multifaceted nature of difficulties experienced by children with learning disabilities.

Our research highlighted the broad spectrum of difficulties experienced by children with suspected learning disabilities. We also considered how the co-occurrence of two or more impairments may interact with one another. Further research is needed to understand these interactions better. Empirical research examining how a thorough understanding of an individual's strengths and weaknesses can lead to improvement in targeted interventions and in turn, whether these targeted interventions lead to improved outcomes in children with learning disabilities.

Despite identification of a number of potential benefits of multidisciplinary assessment of children with suspected learning disabilities, well-designed empirical research in this area is still lacking. As such, it is not yet clear which model is best suited to assess and manage these children. Comparison of multidisciplinary assessment models and alternate models of assessment and identification of children with learning disabilities (e.g. response to intervention models) would therefore be beneficial. Contrasts would allow comparison of the outcomes of these assessment models, as well comparing and contrasting other factors such as cost, waiting times for assessment and diagnoses, staff and family satisfaction, and confidence in the assessment. Moreover, although we feel that multidisciplinary assessment models are well placed to provide a thorough and accurate assessment of children with

suspected learning disabilities, it is not yet clear as to whether there are benefits with regards to academic and other outcomes for children and their families.

Comparison of differently structured clinics would also be useful. The clinic described in this thesis included allied health practitioners working within a learning disabilities clinic, but paediatric multidisciplinary assessment clinics could include practitioners from other disciplines. It is unclear as to which combination of disciplines would be best suited to maximise skill and knowledge for the assessment while keeping costs and assessment time to a minimum. The importance of research of this sort lies in the fact that multidisciplinary assessments can be time consuming and costly. Understanding how best to structure these assessments, including which practitioners to involve, hopefully allows services to maximise the benefits of their assessment service while reducing potential limitations of the model.

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